

**Motor development in typically developing children and children with
autism spectrum disorders**

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Motor development in typically developing children and children with autism

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Motor skill impairments in typically developing children and children with autism spectrum disorders

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Abstract

By Maja Jasmin Knudsen

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Background: Research into early identifying features and associated symptoms of autism has shown high prevalence rates of motor skill deficits in children with autism spectrum disorders (ASD). This research has shown evidence of deficiencies in a wide range of motor skills in children with autism, but there is little agreement in the field on whether motor skill impairments are a defining feature of autism or not. This study explored differences in motor development on parental reports of motor skill at 18 months and 36 months, and individual differences in motor development among 3-4 year old children on a standardized measure of fine motor skills (Mullen Scales of Early Learning, fine motor section, or MSEL). The design of the study included comparisons between the ASD group, children with mental retardation without autism (MR), and typically developing children (TD). The relationship between levels of cognitive functioning (IQ) and fine motor skills was also explored. The current study is based on data from the Norwegian Mor & Barn Study (MoBa) and the Autism Birth Cohort study (ABC study).

Methods: The present study is divided into two sub-studies based on: 1) parental reports of motor skills at 18 and 36 months and 2) the MSEL fine motor section and IQ based on the Stanford-Binet V. The results from the parental reports in sub-study 1 were compared between the ASD and TD group at each point in time. The results from the measures in sub-study 2 were compared between the HF-ASD, TD, LF-ASD and MR groups. The relationship between IQ and fine motor skills was also examined closely.

Results: Mean scores on both parental reports of motor skill and a standardized measure of fine motor skill (MSEL) were significantly lower for children with autism than typically developing children. Mean scores on the MSEL were *not* different between the ASD group and the MR group. Levels of cognitive functioning were strongly associated with performance on the MSEL across groups. Mean standardized scores on parental reports of motor skill were not significantly different at 18 months and 36 months, indicating that

children with delays in motor development at 18 months continue to be delayed at 36 months as well.

Conclusion: Children with ASDs are a highly varied group, but as a whole they are more likely to have deficits and/or delays in motor development compared to typically developing children of the same age. The variation in motor development appears strongly associated with intellectual functioning, but the difference in fine motor skill between the typically developing children and the children with ASD continues to be significant after the effects of IQ have been controlled for. These findings are discussed in relation to methodological factors, interventions and causal theories.

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1 Introduction

1.1 From Autism to the Autism Spectrum

Over the past few decades, there has been a steady rise in the amount of research on Autism Spectrum Disorders (henceforth referred to as ASD). As defined by the Diagnostic and Statistical Manual IV (DSM-IV: American Psychiatric Association, 2000) and the International Statistical Classification of Diseases and Related Health Problems 10 (ICD-10), autism is a pervasive developmental disorder characterized by qualitative impairment in social interaction and communication, and restricted, repetitive or stereotyped behavior (American Psychiatric Association, 2000). Although many children are not diagnosed until much later, some even in adulthood, onset of the disorder must be prior to 36 months of age with delays or abnormal functioning in one or more of the following domains: social interaction, symbolic or imaginative play and language used in social communication.

The term ASD refers not only to Autistic disorder, but also to other diseases such as Asperger's syndrome and Pervasive Developmental Disorder Not Otherwise Specified (PDD-NOS). The most severe of these is Autistic disorder, which is referred to as "childhood autism" in the ICD-10. Whereas children with autistic disorder generally show quite severe impairments in all three of the core domains, children with Asperger syndrome are often more highly functioning with average to above average intellectual functioning and language skills within the range of typical development. Children in the PDD-NOS group, which also includes children with the diagnosis "atypical autism" from the ICD-10, present an even more diverse group. These children do not meet the criteria for autistic disorder or Asperger's disorder because their symptoms present atypically, their symptoms are considered sub-threshold and/or age of onset occurs after 36 months of age (American Psychiatric Association, 2000). The classifications denoting the subgroups within the autism spectrum are likely to change with the recent publication of the DSM-V, which gathers these diagnoses into a single "umbrella disorder" to further emphasize the idea of an autism *spectrum* (American Psychiatric Association, 2013). It is thought that this approach is more suitable for capturing the broad range of severities and symptoms, and that it will lead to earlier identification and more flexible diagnosis.

1.1.1 Associated Symptoms

The main factors contributing to the diversity within the ASD group have been identified as language skills and level of cognitive functioning (Carr, 2006; Engeland & Buitelaar, 2008). However, there are many other factors contributing to the difficulties faced by children with autism. In addition to the three cardinal clinical features, there is a range of other symptoms closely associated with autism (see table 1 on p. 3) (Carr, 2006; Engeland & Buitelaar, 2009; Ventola & Tsatsanis, 2011). Amongst these associated symptoms, perceptual peculiarities and difficulties have perhaps garnered the most interest. According to some estimates, sensory abnormalities are found in approximately 90% of individuals with autism (Baker, Lane, Angley, & Young, 2008; Engeland & Buitelaar, 2009; Ventola & Tsatsanis, 2011). These perceptual peculiarities often come in the form of hypo- or hypersensitivity to auditory, visual, tactile and temperature stimuli. This hypo- or hypersensitivity is expressed in a variety of ways, such as visual fascination, aversion to certain foods and textures, failure to react to pain, fascination with certain smells or extreme reactions to everyday noises (Ventola & Tsatsanis, 2011).

Motor impairments are often associated with autism (Fournier, Hass, Naik, Lodha, & Cauraugh, 2010). Although motor development issues have previously been described, they have not received as much focus as other associated difficulties, such as perceptual issues, emotional control or impairments in Theory of Mind. In their meta-analysis, Fournier, Hass, Naik, Lodha & Cauraugh (2010) examined research on motor coordination impairments and ASD. They reviewed a total of 41 different studies, focusing on seven outcome measures concerning ASD and motor control impairments: a) movement time/reaction time b) movement accuracy/error c) adaptation rate d) gait velocity e) center of pressure excursion f) balance stability and g) standard motor control scales, such as the Vineland Advanced Behavior Scales (VABS), the Physical and Neurological Examination for Soft Signs (PANESS), the Peabody Developmental Motor Scales (PDMS) and Bruininks-Oseretsky Test of Motor Proficiency (BOTMP). The authors concluded that the studies reviewed provide evidence that there are substantial motor coordination deficits in the ASD group in comparisons with typically developing children in a range of motor abilities. These abilities include arm movements, gait, balance, postural stability and movement preparation/planning. They also reported significant deficits in both upper and lower extremity motor

performances, but point out that motor deficiencies appear more prevalent when the evaluations are based on postural control and mobility, deficiencies that could indicate an

Table 1: Associated symptoms and features of autism, adapted from Carr (2006) and Ventola & Tsatsanis (2011)

<i>Domain</i>	<i>Feature</i>
Physical	<ul style="list-style-type: none"> • Epilepsy • Enuresis and encopresis • Gastrointestinal symptoms, such as gluten intolerance • Syndromes such as Fragile X and Tuberous Sclerosis • Motor impairments
Affect	<ul style="list-style-type: none"> • Impaired ability to regulate emotional responses • Fears and phobias (common in younger individuals) • Impaired understanding of emotions in others
Behavior	<ul style="list-style-type: none"> • Restricted interests • Dependence on routine and rituals • Resistance to change • Self-injurious behavior such as head-banging or biting • Absence of imaginative play • Other forms of challenging behavior
Perception	<ul style="list-style-type: none"> • Hypo- and/or hypersensitivity to perceptual stimuli • Extreme fascination with certain types of stimuli
Cognitive	<ul style="list-style-type: none"> • Up to 70% have an IQ below 70 (Engeland & Buitelaar, 2008) • Difficulties with social problem solving and Theory of Mind issues
Interpersonal adjustment	<ul style="list-style-type: none"> • Impaired ability to empathize with others, possibly related to Theory of Mind issues • Poorer understanding of rules governing social interaction • Impaired social conversation • Impaired reciprocity in social interactions

immature postural system (Fournier et al., 2010). It should, however, be noted that this particular meta-analysis was not able to control for IQ due to a paucity of studies that provide specific information regarding IQ scores of the individuals included. Other studies report hypotonia, motor apraxia, toe-walking, gross-motor delay (Ming, Brimacombe, & Wagner, 2007) and lower overall motor skills (C. Hilton et al., 2007) as motor impairments frequently found in autism. There are also frequent reports of deficiencies in fine motor skills, such as handwriting (Fuentes, Mostofsky, & Bastian, 2010), reach-to-grasp movements (Mari, Castiello, Marks, Marraffa, & Prior, 2003) and object manipulation (Jasmin et al., 2009).

1.2 Motor development in typically developing children

Motor development refers to the child's increasing control over their own bodies, and the actions that allow them to interact more and more with their surroundings. Hence, motor development is often defined as a transactional process, wherein the biology of the individual, the conditions of the environment and requirements inherent in the movement task interact and influence each other to bring about changes in motor behavior (Gallahue, Ozmun, & Goodway, 2012). In many ways, motor development can be considered a prerequisite for the development of both cooperation and autonomy (Tetzchner, 2012). Motor development is often divided into fine- and gross motor development. Gross motor development refers to development of *locomotor* skills, which are skills that allow the child to move around with increasing independence, and manipulation of objects by for instance kicking, throwing, catching or striking the objects. On the other hand, fine motor development involves using the small muscles in the hands and wrists to manipulate objects, such as grasping something, buttoning a shirt, stacking blocks or holding a pencil in a pincer grip (Gallahue et al., 2012).

In typically developing children, motor development is a very broad concept. There are milestones children are expected to reach within certain ages, but these ages should only be considered a rough guideline. Some of the milestones of gross and fine motor development in the first few years of life are summarized in table 2. In typically developing children, motor skills mostly develop in an organized manner, following a cephalocaudal trend, although there is great individual variation. Here, the infant first learns to control his or her head. Next there is increased control of the arms and upper body/trunk, which is then followed by increased control of the legs. There is also a trend for control of the central parts of the body to develop first, followed by the distal parts of the body, such as hands and feet –

a proximodistal developmental direction. This largely matches the growth patterns of human infants, which follows the same head-to-tail and inwards-outwards sequence (Gallahue et al., 2012). Most of these “motor milestones” build upon one another. Consequently, before the children can learn to walk, they must learn to stand, and before they learn to catch, they must learn how to grasp. However, if a child is late in reaching some milestones, it does not automatically mean they will be late in reaching other milestones. Some children completely skip certain milestones, such as learning to walk without first learning to crawl.

There are several factors that together lead to the acquisition of each new motor skill. These include 1) central nervous system development 2) the body’s movement capacities (such as muscle development in infancy) 3) the child’s own goals (such as getting hold of a cookie or toy) and 4) environmental support for the skill (such as visual stimuli to promote grasping and access to hand-holds to help them stand or walk) (Gallahue et al., 2012).

Acquiring motor skills is a complex process for the growing child, and most newly acquired skills need practice and refining before becoming a part of the child’s repertoire. Typically developing children practice and refine these motor skills on their own as they explore their surroundings. For instance, toddlers learning to walk will engage in up to six hours of “practice” per day to perfect this skill, repeating the necessary motions more than thousands of times. This, in turn, promotes new and stronger synapses in the motor areas of the brain, which will promote further motor development (Berk, 2009). These patterns of typical development continue as the child grows from infancy to childhood, and then into adolescence.

All of these skills contribute to the child’s social development during infancy, as each new motor skill achieved generally elicits a new social response from the child’s caretakers (Campos et al., 2000). If the child acquires a new motor skill, then this should provide new possibilities to learn, develop and engage with the environment in novel ways. The child’s increased independence and repertoire of skills allow them to be increasingly active participants in the social environment through play, for instance, and their environment will react accordingly. Whereas caretakers are a source of comfort and support, they now also take on the role of being a hindrance to the child (Gallahue et al., 2012; Karasik, Tamis-LeMonda, Adolph, & Dimitropolou, 2008; Tetzchner, 2012). Examples of the latter include denying the child access to a steep staircase the child wants to climb, or scolding the child for doing something dangerous. This adds a new dimension to the child-caretaker relationship, and as the child’s ability to act independently increases, he or she will also seek out emotional information via social referencing, for instance by looking back towards his or her

Table 2: Motor milestones from infancy through early childhood, adapted from Berk (2009).

Motor Development During First Two Years of Life		Gross Motor Development During Early Childhood (2-5 years)	
<i>Motor Skill</i>	<i>Average Age Achieved</i>	<i>Motor skill</i>	<i>Approximate Average Age Achieved</i>
	<i>Age Range in Which 90 Percent of Infants</i>		
Grasps cube	3 months, three weeks	Walks more rhythmically, fast walk turns into run	24-36 months
Sits alone	7 months	Jumps, hops, throws and catches with rigid upper body	24-36 months
Crawls	7 months	Walks up stairs with alternating feet, and down stairs, leading with one foot	36-48 months
Plays pat-a-cake	8 months	Jumps and hops, flexing upper body	36-48 months
Stands alone	11 months	Throws and catches with some upper-body involvement, catches by trapping object against chest	36-48 months
Walks alone	11 months, 3 weeks	Walks down stairs with alternating feet	48-60 months
Builds tower of two cubes	11 months, 3 weeks	Runs more smoothly	48-60 months
Scribbles	14 months	Skips with one foot	48-60 months
Walks up stairs with help	16 months	Throws ball with increased body rotation and transfer of weight, catches thrown object with hands	48-60 months

mother in a novel situation to see if the mother deems it safe or not, or looking at his or her peers to discern whether to join a play situation or not (Campos et al., 2000). As such, motor development, cognitive development and social development tend to go hand in hand (Campos et al., 2000; Davis, Pitchford, & Limback, 2011). They are not necessarily dependent upon one another, but there is evidence that motor development can facilitate development in other domains as well, such as socioemotional development (Anderson et al., 2013; Campos et al., 2000).

1.3 Motor development in children with ASD

Some studies indicate that there are early motor abnormalities in both children with ASD and their siblings, the latter group often considered at risk for ASD (Bolton, Golding, Emond, & Steer, 2012; Mari et al., 2003; Mieres, Kirby, Armstrong, Murphy, & Grossman, 2012). Other studies have found that difficulties with both gross and fine motor skills are common symptoms in children within the autism spectrum, with reported prevalence rates for significant motor delays compared to normative data of up to 50-73% (Provost, Lopez, & Heimerl, 2006). Despite many studies reporting motor impairments in children with ASD in a variety of areas (Dewey, Cantell, & Crawford, 2007; Dziuk et al., 2007; Fournier et al., 2010; Fuentes, Mostofsky, & Bastian, 2009; Jasmin et al., 2009; Kopp, Beckung, & Gillberg, 2010; Ming et al., 2007; Provost, Lopez, & Heimerl, 2007; Staples & Reid, 2010), it is still not known how these impairments potentially interact with the core symptoms of autism.

Motor abnormalities are not considered a core feature of autism spectrum disorders according to the DSM-IV-TR or the ICD-10 criteria. This is perhaps due to few studies reporting findings that support motor function as a feature distinguishing the ASD-group from unaffected individuals and other diagnostic groups (Provost, Lopez, et al., 2007). Another suggested reason is that children with autism often start to walk at the same time as typically developing children, and generally achieve functional mobility while still having qualitative motor impairments (Jasmin et al., 2009). In other words, they reach motor milestones within the normal age-range, but may, for instance, be clumsier, less coordinated and have less skilled movements. Jasmin et al. further suggest that there is a lack of distinction between quantitative and qualitative measures of motor skill that may explain why the literature on motor development in ASD remains ambiguous, and why few interventions have been developed to target motor skills in this group. Given the high reported prevalence of motor issues in the ASD group and knowing the importance of early detection and early

intervention in ASD (Stenberg, 2012), it seems that further inquiry into motor abnormalities in ASD could be a useful avenue for research.

Within the autism group, motor development seems to vary greatly. Some individuals have no motor impairments whereas other children require physical therapy to help them develop. Nevertheless, there seems to be a preponderance of cases with impaired motor abilities (Fournier et al., 2010; Green et al., 2009; Ming et al., 2007; Provost, Heimerl, & Lopez, 2007) even when taking into consideration the variation in normal motor development. It appears that children with ASD might develop motor skills more slowly, and with less refined actions than typically developing children, though there are still many uncertainties regarding this. According to Canitano (2011), the most common difficulties in motor development in the ASD group are related to sequencing movements, hypotonia, postural control problems, clumsiness, and poor coordination, which is substantiated by evidence from several studies (Fournier et al., 2010; Fuentes et al., 2009; Green et al., 2009; Jasmin et al., 2009; Matson, Matson, & Beighley, 2011; Ming et al., 2007; Ozonoff et al., 2008). One particular study by Ming et al., which examines the prevalence of motor issues in a relatively large sample of children with autism (N = 154), found that hypotonia, defined as reduced resistance during passive movement in the limbs and increased joint mobility to passive stretching, and apraxia, which they define as an impairment in the ability to execute skilled movements and gestures, were common motor impairments in children aged 2-6 years old (Ming et al., 2007). Upon examining the children using retrospective chart reviews, clinical intake forms completed by caregivers and physical examinations conducted by a child neurologist, they found that hypotonia and apraxia were present in respectively 63% and 41% of children between 2-6 years. However, the prevalence was significantly lower in the older segment of their sample, with hypotonia present in 38% of children between 7-18 years of age, and apraxia present in 27%. Only 9% of the children in this sample had gross motor delays, and all of these children had reached the target milestones by the time of inclusion in the study. The authors conclude that while the prevalence of motor impairments is high, there appears to be a lower incidence amongst older children, and these motor impairments do not seem to interfere with the acquisition of basic locomotor skills (Ming et al., 2007). This possible improvement in motor function over time is also tentatively supported by Fournier et al. (2010), but since these studies utilize cross-sectional rather than longitudinal designs, it is difficult to make a clear conclusion about the effects of age on motor development within the ASD group.

Based on the available evidence, it seems as though motor impairments in autism are qualitative in nature – motor development milestones may or may not be reached, within or without normal age brackets, but the skills that are acquired may be qualitatively impaired. An example of this type of qualitative impairment was found by Fuentes et al. upon examining handwriting skills in children (2009) and adolescents (2010) with ASD. They used a case-control design comparing children and adolescents with and without ASD on the Minnesota Handwriting Assessment, and found that while the children with autism are capable of writing by hand, they have specific handwriting impairments consisting of poor letter formation, and that these impairments continue on into adolescence. Fuentes et al. also found that handwriting deficits were predicted by overall motor skills in children, whereas perceptual reasoning was the main predictor for adolescents, perhaps reflecting the ability to learn how to compensate for the motor impairments.

Canitano (2011) notes that dyspraxia, which refers to difficulties with the execution of voluntary movements, is often reported for children in the ASD group, which is supported by other evidence as well (Dziuk et al., 2007). These difficulties with executing voluntary movements have been highlighted by several studies investigating motor skills. Mari et al. (2003) examined the reach-to-grasp movement in 20 children with ASD without additional movement disorders (such as tremors or cerebral palsy) by comparing them with 20 sex-and-age matched children with no neurological or skeletomotor dysfunctions, and IQ within the normal range, on a kinetic measure of reaching and grasping specific objects. They found several differences in the reach-to-grasp movement pattern in children with autism as compared to the control group, such as a generalized slowness in the movement pattern, and suggest that these differences arise from a dysfunctioning ability to initiate, switch, efficiently perform or continue ongoing action – in other words executing goal-directed, voluntary movements. Dyspraxia also includes difficulties with purposeful oral movements, which can give rise to problems with speech and/or facial expressions. Furthermore, in the ASD group it appears that dyspraxia has been observed in particular during imitation tasks (Canitano, 2011).

1.3.1 Intellectual development and motor skill development

High- and Low-Functioning autism

Evidence shows that there is a significant overlap between ASDs and intellectual disability. However, children with autism are a highly heterogenous group, and there is great variation

in levels of intellectual functioning among individuals with ASD. The spectrum includes individuals with severe mental retardation at one end, to very high functioning individuals at the upper extremes of the IQ scales on the other. The group of individuals affected by autism spectrum disorders present quite heterogeneously, but autistic symptomatology often varies with levels of functioning and intellectual ability (Lecavalier, Snow, & Norris, 2011). In literature and clinical practice, individuals with autism are often divided into two categories based on their levels of cognitive functioning: High-functioning ASD (IQ > 70) and low-functioning ASD (IQ < 70). These will henceforth be referred to as HF-ASD and LF-ASD. Individuals with HF-ASD may function with minimal assistance in their day-to-day lives, and some display giftedness in one or more areas (Carr, 2006). Individuals with LF-ASD, on the other hand, are more likely to have co-morbid diagnoses such as epilepsy or severe language impairments. Some estimates stipulate that approximately 75% of children with autism have IQs below 70, with an asymmetrical profile favoring the non-verbal or performance domain on standardized intelligence tests (Carr, 2006). These children will likely require high levels of support throughout childhood and adolescence, continuing into adulthood, and will probably be unable to live independently.

Motor impairments are relatively common in children with intellectual disability (IQ <70) as part of a general developmental delay (Einfeld & Emerson, 2008). This gives rise to the question of whether motor issues in the autism group can be considered the same, or whether they are something unique to the ASD group itself. Some observations indicate that motor skill deficiencies are still prevalent in the ASD group, even when the effects of IQ have been controlled for (Kopp et al., 2010; Staples & Reid, 2010). Others have concluded differently, for instance when comparing a group of 19 children with ASD between 21 and 41 months of age with an age-and-gender matched group of children with developmental delay (DD), and controlling for mental development (Provost et al., 2006). They found that while motor development in the ASD group tends to be atypical, it does not clearly distinguish them from the DD group, meaning that motor impairments in the ASD group may not be related to autism in and of itself. Other findings point to the possibility of a link between intellectual development and motor development in the ASD group (Green et al., 2009; Mari et al., 2003). Findings from these studies further imply that the motor impairments observed in ASD may possibly stem from a more serious neurological impairment that also contributes to intellectual disability (Green et al., 2009), such as a bilateral brain dysfunction (Hauck & Dewey, 2001). A study by Kopp et al. (2010) reported a significant correlation between motor impairments and lower abilities in Daily Living Skills as measured with the Vineland

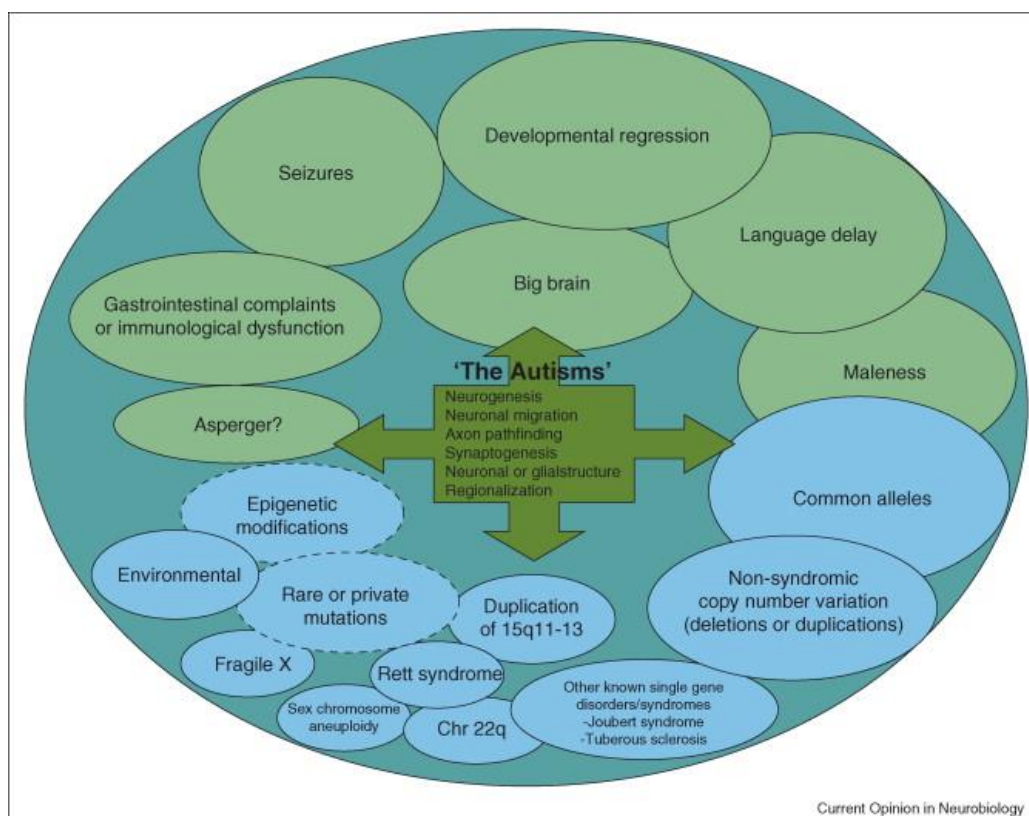
Adaptive Behavior Scales (VABS). They point out that motor problems continue to contribute to poorer daily living skills even when the effects of IQ have been taken into account. This highlights the possible benefits of developing interventions and support systems for individuals with autism that take motor impairments into account.

1.4 Theoretical explanations

1.4.1 Changes in neuroanatomy

Research into motor impairment and motor development in ASD has thus far provided few possible explanations for the association between autism and motor difficulties. There are, however, some theories. A common factor for many of the theories that are applicable to motor development in ASD, is the idea of an underlying neurobiological brain mechanism (Casanova, 2007; Coskun et al., 2009; Stanfield et al., 2008). Changes in regional and functional brain anatomy in a wide variety of brain regions have been reported, but there appears to be little consistency in these findings across individuals and studies. Studies utilizing magnetic resonance imaging have indeed found consistent evidence for increased brain volume in younger children with autism, as well as a reduction in size of the corpus callosum (Stanfield et al., 2008), but these findings do not adequately explain the motor

Figure 1: “The possible etiological features (genetic and environmental heterogeneity; blue circles) and the clinical or syndromic heterogeneity (green circles) within autism spectrum disorder” (Geschwind & Levitt, 2007)



difficulties experienced by many children with ASD. Etiologically speaking, there is a wealth of different theories and hypotheses that attempt to explain autism, illustrated in figure 1 by Geschwind & Levitt (2007) on p. 11. Amongst these theories, there are some that, albeit controversial, also attempt to account for the motor impairments observed in children with ASD in addition to the core symptoms.

1.4.2 Mirror neurons

One of the theories concerning motor development in children with ASD revolves around mirror neurons. This is a controversial theory, with conflicting findings, but there is evidence that suggests we are born with systems of mirror neurons, which are highly specialized cells, in the motor areas of the cerebral cortex. Although there is currently uncertainty regarding the actual role of mirror neurons in human behavior and development, there has been speculation that dysfunction in mirror neuron systems (MNS) may play a role in autism (Fan, Decety, Yang, Liu, & Cheng, 2010; Iacoboni & Dapretto, 2006; Oberman et al., 2005; Rizzolatti & Fabbri-Destro, 2010; Rizzolatti, Fabbri-Destro, & Cattaneo, 2009). Research into mirror neurons has led to speculation that these systems play a role in language abilities, theory of mind skills, empathy and the ability to understand the actions and motivations of others (Gazzaniga, Ivry, & Mangun, 2009; Sommerville & Decety, 2006), all of which are abilities closely tied to autistic symptomatology. In regard to autism, the mirror neuron literature often cites that difficulties with imitation tasks are very common amongst children with autism. Difficulties with imitation tasks are considered to be part of the core difficulties faced by children with autism, and are heavily implicated to be a result of dysfunction of mirror neuron systems. Learning through observing and copying/imitating actions is an integral part of an infant's ability to learn everything from social responses and facial expressions to motor skills (Meltzoff, 2007a, 2007b). There is also research indicating that there are actual anatomical differences in the cortical regions related to MNS in individuals with autism when compared with typically developing individuals (Hadjikhani, Joseph, Snyder, & Tager-Flusberg, 2006). However, the evidence for systems of mirror neurons playing a role in autism is conflicting at best, and does not fully explain the association between motor impairments and autism, and the mirror neuron hypothesis has been criticized (e.g. Enticott et al., 2013; Fan et al., 2010; Jacob, 2009).

1.4.3 Developmental disconnection

A recently proposed etiological explanation is the connectivity/disconnection hypothesis, which posits that ASD may arise from disconnections between brain regions involved in higher-order associations in humans (Courchesne & Pierce, 2005; Geschwind & Levitt, 2007). This hypothesis emphasizes that the disconnections in question are not primarily disruptions of previously connected brain regions, but a failure in the development of these connections that might be caused by a wide range of etiologies. Suggested etiologies include malfunctions in histogenic events vital to the establishment of basic connectivity and columnar organization of the cortex, such as prenatal neuronal migration and axon pathfinding, and postnatal events such as synaptic pruning, development of the dendrites and synaptogenesis. In order to account for the heterogeneity of deficits and symptoms observed in autism, Geschwind & Levitt (2007) propose that the key disconnections could be between cortical areas in the frontal lobe and temporal lobe. This is supported by evidence from fMRI studies indicating reduced or abnormal frontal cortical neural activity and early frontal maldevelopment, which has also been linked to the increase in brain volume in younger children with autism (Courchesne & Pierce, 2005). According to Courchesne & Pierce these findings could indicate impairment in the fundamental frontal function of integrating information from systems across the cerebrum. They propose that connectivity within the frontal cortex is functional, albeit excessive, disorganized and poorly selective, whereas connectivity between the frontal cortex and other areas of the brain remains reduced, unsynchronized and poorly responsive. This has also been suggested by others (e.g. Shalom, 2009). As of yet, however, there is only limited evidence supporting this hypothesis, and the cellular basis for dysfunctional circuitry within the brain and potential mechanisms resulting in poor connectivity remains poorly understood.

In summary, it appears that motor skill impairments are neither ubiquitous nor uniform in individuals with autism spectrum disorders. Evidence is contradictory, and still rife with speculation in terms of etiology. Although there is still uncertainty regarding the causes of motor skill impairments in autism, there is evidence that these impairments, when present, may also impair their daily living skills (DLS) (Jasmin et al., 2009). Research investigating individual differences in motor development over time, and possible associations with levels of cognitive functioning could therefore be of value to help broaden the view of parents, health professionals and others working to develop means of early

detection and interventions for children with autism. Based on the available evidence, it would appear that young children with autism spectrum disorders are likely to have poor performance on a range of motor tasks when compared with typically developing controls. These tasks include, but are not limited to, handwriting tasks, motor imitation tasks, tasks that challenge balance and posture, fine motor dexterity tasks and tasks that include more complex planning of motor actions.

1.5 The present study

The aim of the present study is to examine the variability in motor development among children with ASD, as many children with ASDs have motor impairments, though it is not characteristic for the group as a whole. The present study is divided into two sub-studies. The first of these (S1) compares a group of children with ASD to a group of typically developing children (TD) matched for age (year of birth), gender and parental education and age on a survey-based parental report of motor skill. By comparing children with autism to a group of typically developing children on a questionnaire-based measure of motor development at 18 months and 36 months, one can explore between-group differences in motor development. The second sub-study (S2) compares a group of children with ASD with a group of typically developing children and a group of children with mental retardation (MR) on a standardized measure of fine motor skill. By also comparing high-functioning and low-functioning children with autism with typically developing children (TD) and children with mental retardation (MR) on a standardized measure of fine motor skill, one can further elucidate this relationship by examining both between- and within-group differences. Based on previous research in the field, these relationships will be examined with a working hypothesis postulating that children with ASDs will have poorer scores on measures of motor skills than a control group of typically developing children, with factors other than ASDs possibly mediating the relationship. Together, these two sub-studies examine some of the factors that may influence motor development in the ASD group.

Research questions:

- 1) Does the variation in motor skills in the ASD group differ from the variation in a typically developing group?
- 2) Are children with delayed or atypical motor development at 18 months also impaired at 36-48 months?

- 3) Does level of cognitive functioning correlate with levels of motor development?
- 4) What, if anything, distinguishes the ASD children *with* motor skill impairments from those without?

2 Methods

This study is a part of the Autism Birth Cohort Study (ABC Study), a subsidiary of the Norwegian Mother and Child Study (MoBa). The MoBa study is a nationwide population-based cohort study. The MoBa study began recruiting participants via the Medical Birth Registry of Norway in 1999 by sending out invitations to all pregnant women in Norway who were scheduled for an ultrasound between the 17th – 19th week of pregnancy. More than 100000 women were recruited, and both biological data and questionnaires were collected periodically from pregnancy throughout birth and beyond. The goal of the MoBa study is to continue following these children into adulthood.

The ABC-study identified children who screened positive for autism traits based on the MoBa questionnaire sent out when the children were 36 months of age. This particular questionnaire contains elements from standardized diagnostic instruments used to identify children with possible developmental disorders. Any child with symptoms that might indicate an autism spectrum disorder was then invited to the ABC study for further assessment. A part of the participants were recruited through one of three alternative mechanisms: 1) referrals from health care providers, 2) direct request from the parents if they were already part of the MoBa study or 3) children who did not screen positive on the 36 month MoBa questionnaire, but later received an autism diagnosis, which was made available through the Autism Database which is linked to the Norwegian Patient Registry (NPR).

The comparison group was selected by randomly drafting MoBa participants who are matched to potential cases by birth date. Within the ABC study, diagnoses of autism spectrum disorders were made by a child psychiatrist and/or psychologist based on the Autism Diagnostic Interview – Revised (ADI-R), the Autism Diagnostic Observation Schedule (ADOS), clinical judgement and the diagnostic criteria for ASDs from the DSM-IV. The assessment of the children includes clinical observation of the children as well as information gathered from multiple informants, such as parents and pre-school teachers. As such, the ABC study employs early identification of ASD cases with a broad range of symptom manifestations and severity levels.

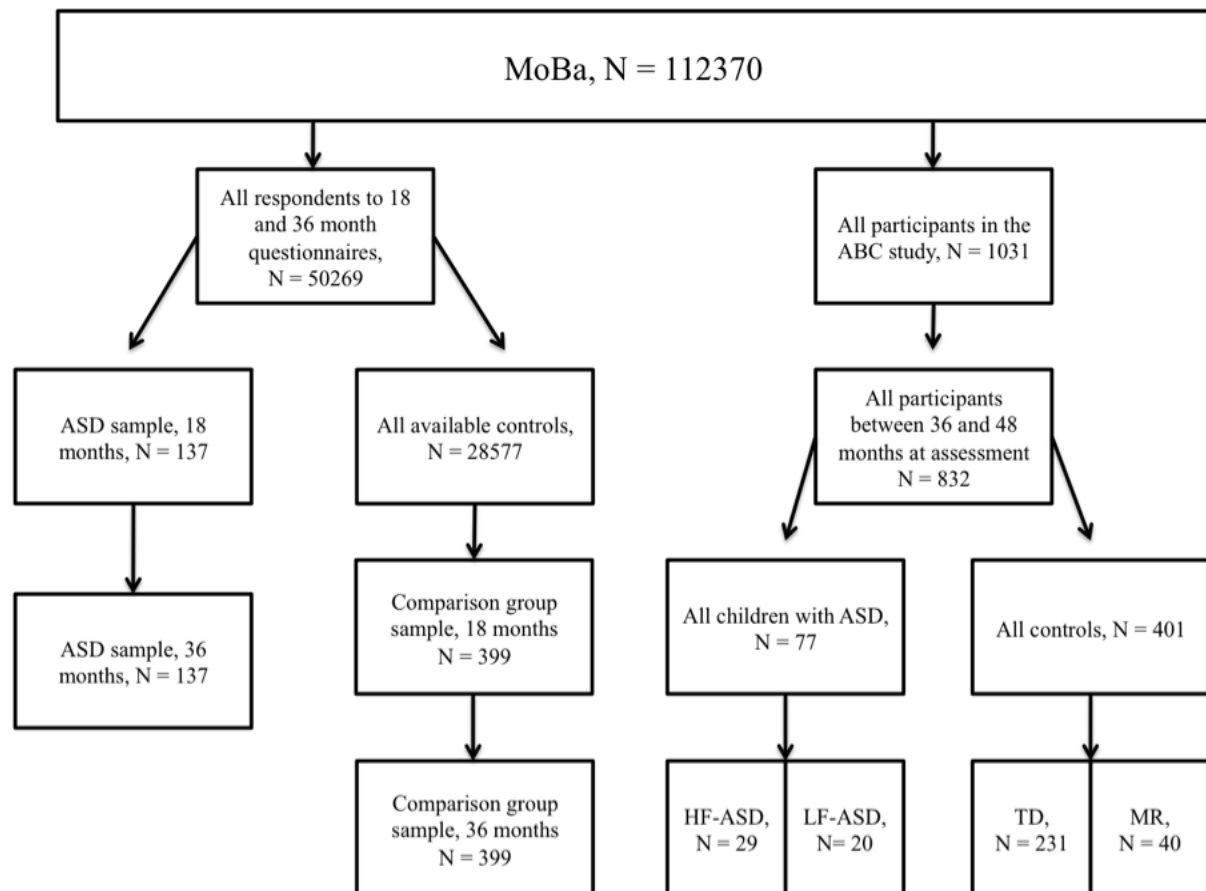
2.1 Participants

All participants included in the present study were originally recruited to the MoBa study and/or the ABC study. The total number of children included in the S1 analyses is 137, of

which 114 are males. In the MoBa study, the number of children with a diagnosis of autism in the Norwegian Patient Registry was higher, but only 137 of these had answered all of the relevant items in both of the forms in question. In a number of cases where both questionnaires had been filled out, the parents had not answered one or more of the relevant items on either one or both questionnaires. These children were excluded from the present study. By utilizing the Random Sample function in SPSS, a comparison group (N = 399) matched on gender, age, parental age and parental education was drawn from the totality of children included in the MoBa study after filtering out children with known diagnoses and recent referrals to educational or mental health services. The comparison group is therefore considered to consist of a group of typically developing children. The male-female ratio in this sample is relatively close to the 4:1 gender ratio most often reported in prevalence studies (Newschaffer et al., 2007), with 17% females in this case.

In the S2 analyses, based on the assessment from the ABC study, there were 77 children in the appropriate age group (36-48 months at time of assessment) diagnosed as having ASD, 60 males and 17 females. The male/female ratio in this group is therefore fairly close to the oft-reported 4:1 ratio, with 22.07% females in this case. The ASD group was divided into two groups based on level of cognitive functioning: high functioning autism (IQ > 70) and low functioning autism (IQ < 70) based on their scores on the Stanford-Binet V. To preserve statistical power, the groups were not further subdivided into diagnostic sub-groups. A comparison group (N = 271) of individuals matched to the ASD group on age, gender, parental age and parental education was randomly drawn. The comparison group was then divided into typically developing children and children diagnosed as having mental retardation (IQ < 70) outside of autism by the clinicians of the ABC study to match the division of the ASD group into HF-ASD and LF-ASD. Children who were older than 48 months upon time of assessment were excluded from analyses, because the majority of the participants in the ABC study were between 36 and 48 months of age at the time of clinical assessment and different instruments were used to assess the older children, which would have complicated the analysis greatly.

Figure 2: Model of the flow of participants from the original MoBa study to the groups being compared in the present study.



2.2 Instruments

The instruments used to assess the children participating in the ABC study include, but are not limited to, Stanford-Binet V (SB5), the Mullen Scales of Early Learning (MSEL), the Autism Diagnostic Observation Schedule (ADOS), the Autism Diagnostic Interview Revised version (ADI-R), and the Vineland Adaptive Behavior Scales (VABS). In addition to the clinical examinations that are at the core of the ABC Study, the parents of the child also complete a number of questionnaires concerning the mother and child’s health, the family’s socio-economic conditions and the child’s development at regular intervals from early on in the pregnancy, to the child is born and beyond as part of the MoBa study. None of these instruments focus on the motor skills of the child, but several of them have components dealing with motor skills and motor skill development. All the relevant instruments will each be given a brief presentation.

2.2.1 The MoBa Questionnaires

The MoBa questionnaires, completed at 18 and 36 months of age by the parent(s) of the child participating in the study, query about health, dietary intake, socio-economic status, child development and behavior, and the psychosocial and emotional status of the mother, father and child (Stoltenberg et al., 2010). The 18-month questionnaire consists of 111 items and includes the Early Screening of Autistic Traits and the Modified Checklist for Autism in Toddlers (M-CHAT). The 36-month questionnaire consists of 75 items, and includes the Social Communication Questionnaire (SCQ) and selected M-CHAT items.

This study will utilize data composed of several items from these forms relating to motor skills. There are six “sub-items” from the 18-month questionnaire (questions 4-9 of item 32). In the 36-month questionnaire, there are four “sub-items” (question 1-4 of item 17). The motor items from these questionnaires are roughly equivalent to motor development milestones most children reach by the ages of 18 months and 36 months respectively, and can be found in table 3 on p. 20.

Since the number of items dealing with motor skills on the two questionnaires is unequal, and the questionnaires do not distinguish clearly between fine motor and gross motor skills, an index score representing total motor skill for each case on each of the two forms was created. This was accomplished by assigning a number to each of the three possible replies to the items, 1 for “Yes”, 2 for “Sometimes” and 3 for “Not yet”, and then compounding these numbers into a composite score. This means that the higher a child’s composite score is, the poorer their motor skills are likely to be. These composite scores were then converted to standard scores in order to facilitate comparative analyses. These composite scores are a fairly rough estimate of a child’s motor skill, based on only a few questions answered by the child’s parent(s), but this approach enables a longitudinal view of the data. Coupling these longitudinal data with the data from the clinical assessments will give a much more complete picture than either set of data on its own.

Table 3: Summary of items from the 18-month and 36-month MoBa questionnaires

Items from 18 month MoBa questionnaire	
<i>Item no.</i>	<i>Full-text</i>
Q32.4	Does the child move around by walking instead of crawling on its hands and knees?
Q32.5	Does the child walk steadily, without falling over often?
Q32.6	Can the child walk down stairs if you hold him/her by the hand?
Q32.7	Can the child throw a small ball with a forward motion of the arm?
Q32.8	Can the child stack blocks or other small toys (approx. 3cm in size) atop one another?
Q32.9	Can the child turn the pages of a book on his/her own, turning more than one page at a time?
Items from 36 month MoBa questionnaire	
<i>Item no.</i>	<i>Full-text</i>
Q17.1	Without support, can the child kick a ball by swinging the leg in a forward motion?
Q17.2	Can the child catch a big ball with both hands?
Q17.3	When the child is drawing, does he/she hold the pencil/crayon between the fingers and thumb like adults do?
Q17.4	Can the child button and unbutton one or more buttons?

2.2.2 The Mullen Scales of Early Learning

The Mullen Scales of Early Learning (MSEL) (Mullen, 1995) is a battery designed to assess cognitive abilities in children aged 0-68 months. It consists of 124 items distributed over five

scales: gross motor functions; fine motor functions; visual reception; expressive language skills and receptive language skills. Each of these five scales consists of tasks that are performed by the child, or scored via interview with the child's parent(s), and each item is scored on a scale of 1-5. There are scores for each of the five scales, as well as a composite score based on all 124 items – The Early Learning Composite score. The Early Learning Composite score does *not* include scores from the gross motor scale.

The ABC-study utilized the full MSEL for a number of years during data collection, but it was later decided to use an abbreviated version due to practical constraints. The abbreviated version of the Mullen scales assesses fine motor skills, but not gross motor skills. The child is asked to stack blocks vertically, to string beads, to cut paper with a pair of scissors, copy shapes and letters onto a piece of paper and to touch fingers in a specific pattern in the fine motor portion of assessment. The children are scored as having either “Strong”, “Medium” or “Weak” skills, if they are able to complete the task, as “Cannot do”, if they are incapable of completing the task, or as “Does not cooperate” if they refuse to do so.

The child's gross motor functioning was roughly assessed by the attending physician via another set of tasks based on the gross motor subscale of the MSEL. In these tasks, the child is asked to stand up from a prone position, climb a set of small stairs, stand and then jump on one leg, run a short distance and balance/walk on a straight line. Given the child's cooperation, this allows the physician to note any major motor impairments or signs of clumsiness/coordination issues, but the exercises are not scored on a formal scale like the fine motor portion of the assessment and will therefore be omitted from analysis.

2.2.3 Stanford-Binet V

The fifth edition of the Stanford-Binet Intelligence Scales was utilized by the ABC-study to assess intelligence. The SB5 is a standardized cognitive test suitable for administration to children from 24 months and up. It consists of a verbal and a non-verbal domain, and is divided into five cognitive factors which can be defined as follows: *Fluid reasoning* is the ability to solve previously un-encountered, or novel, problems, be it with visual, verbal or printed material. *Knowledge* is the totality of general information an individual possesses, accumulated over time from their experiences at home, in school, at work or otherwise in their environment. *Quantitative Reasoning* is the ability to deal with numerical problems, either in the form of number concepts or word problems (Roid & Pomplun, 2005). *Visual-*

Spatial Processing is described as “the ability to see relationships among figural objects, describe or recognize spatial orientation, identify the ‘whole’ among a diverse set of parts, and generally see patterns in a visual material”. *Working Memory* is described as the ability to hold information in one’s memory and manipulating this information at the same time.

The SB5 battery consists of a verbal and a non-verbal section. The testing itself begins with a non-verbal routing subtest, followed by a verbal subtest. In each of these subtests, the first task is a so-called “teaching task” where the child gets feedback on whether or not their answer is correct. The scores on these two routing subtests determine which level of the next subtests the test administrator moves on to (Roid & Pomplun, 2005). Once all appropriate subtests have been completed, the tests are scored in one of two provided scoring systems; either norm-referenced or criterion-referenced. In the ABC-study, the SB5 was scored according to the norm-referenced system, yielding scores for non-verbal IQ, verbal IQ and full-scale IQ. In some cases where it was not possible to complete the full battery, the abbreviated IQ was calculated. ABIQ is based on scores from the two routing subtests.

2.3 Procedure

Upon being asked to participate in the MoBa study, parents were informed that they might be contacted concerning taking part in other, related studies as a result of their participation. If they had consented to this, letters of information concerning the ABC study were then issued twice to families identified as possible participants. If there was no reply to the second letter, it was assumed that they did not wish to participate and no further attempts were made to recruit them.

As part of the protocol for the ABC-study, all children were assessed with a standardized instrument to assess levels of cognitive functioning (SB5 for children below 5 years of age, the Wechsler Abbreviated Scales of Intelligence for older children, the Mullen Scales of Early Learning for non-verbal children deemed unable to complete the SB5) and either the full or abbreviated MSEL, which includes a measure of fine motor skill. Assessments of fine motor skills (MSEL and MSEL Abbreviated) and levels of cognitive functioning (SB5) in were conducted by clinical psychologists trained in the administration of these tests.

Upon arrival to the ABC study facilities, the attending clinician(s) met the families. The assessment would typically begin with the SB5, or the full MSEL if the child were non-verbal and/or unable to complete the routing part of the SB5. Where both parents were

present for the assessment, the father would usually accompany the child during administration of the SB5 whereas a research assistant would interview the mother with the ADI-R and VABS at the same time in a separate room. Depending on whether both parents were present or not, the attending child physician would perform an anamnestic interview with the child's mother after administration of the SB5 regarding the child's developmental history and the parents' concerns. During this interview, the child would be looked after by one of the research assistants in the play-room at the facility. The attending child physician would then perform a physical examination of the child, which includes testing the child's reflexes, strength, and visual acuity, as well as looking for physical signs of syndromes. Following the physical assessment, there would be a lunch break after which a nurse would arrive to take a sample of blood from the child. Finally, the attending psychologist assessed the child with the ADOS. Once all assessments were completed, the attending clinicians and research assistant(s) would discuss the results from their parts of the assessment, and, where appropriate, formulate a diagnosis. Prior to this discussion, every person administering some part of the assessment would be unaware of the results from the other parts so as to not be influenced. At the end of the day, the parent(s) were given thorough information about the results from the assessment. In some instances, there were twins being assessed simultaneously, and in these cases the order of assessment would be slightly different than normal to allow the children to have a parent present at all times if needed.

Assessment took place in a quiet observation room with as few distractions for the child as possible, with the child seated across from the clinician at a desk. The child's mother and/or father were present for support for most of the children. They were asked not to intervene in the assessment process unless requested by the clinician conducting the assessment. Each observation room was equipped with two discrete cameras to record the testing. All assessments took place over the course of 1-1.5 days, with each testing session typically lasting between 15 and 45 minutes depending on the endurance of the individual child. Each clinician was regularly observed during testing, and the assessment scored by one or more of the other clinicians to maintain reliability.

2.4 Ethical considerations

Before agreeing to participate in the MoBa study, parents who were asked to participate were informed about the main purpose of the study, and that they might be asked to participate in other, related studies as a result of their participation in the MoBa. They were informed that

their answers to the MoBa questionnaires might, in some cases, indicate health issues with their child that could lead to requests to partake in, for instance, the ABC study and asked if they consented to this. Upon being asked to participate in the ABC, parents of the children were provided with forms informing them of the purpose of the main study, as well as the possibility that the data could be appropriated for future sub-studies. They were also informed that they could leave both the MoBa study and the ABC study at any point, without providing a reason. Parents were also informed during assessment at the ABC clinic that the assessments would be filmed and/or recorded on tape where video was not available, and given the option to opt out if they were uncomfortable with being recorded.

When the clinical assessments of the ABC study were conducted, there were several ethical issues to take into consideration. Since possible participants were identified through four different mechanisms, not all children identified as having signs of autism during the screening process had diagnoses prior to or after assessment. Therefore it was deemed important to have a clear procedure of what to do when a child fulfilled the criteria for ASD or other diagnosis. At the end of each assessment, the parent or parents of the child were given thorough information about the results of the assessment from two of the attending clinicians. The clinicians would talk the parents through each step of the assessment, continuously clarifying what formed the basis for their conclusion, regardless of what that conclusion was. All parents were given this information by the clinicians regardless of whether a diagnosis had been made or not. If the child met the diagnostic criteria for an ASD, and did not have one prior to assessment, the parents were given a folder with relevant information about autism, support groups for autistic children and their parents, and various relevant resources. During the post-assessment debriefing, the clinicians would advise the parents on the next appropriate step, which in most cases would be referral to health professionals for official diagnoses and further follow-up. Because receiving a diagnosis from healthcare specialists can be quite difficult for the families involved, the parents were also informed that they were welcome to contact the ABC study clinicians by phone or e-mail if they required further support or had any questions upon returning home. The same type of procedure was followed with the parents of children who meet the criteria for other diagnoses, such as ADHD, specific language impairment or developmental delay. These procedures were followed to help ensure the parents and children were given the appropriate information and support following their departure from the ABC study.

2.5 Analysis

2.5.1 Sub-study 1 - The MoBa questionnaire data

All analyses were conducted in SPSS version 20 and 21. Because of skewedness in the longitudinal data from both the ASD group and the control group, a Mann-Whitney U test was conducted to explore between-group differences on total score at 18 months and 36 months separately. Total scores on the measures of motor skill were converted to standard scores to facilitate analysis over time, since different measures were used at 18 months and 36 months. A series of Chi-square tests for independence were conducted to explore between-group differences on individual items from both questionnaires. The Wilcoxon Signed Rank test was conducted to explore change over time. A Mann-Whitney U test was also conducted to explore potential gender differences within the ASD group in parental reports of motor skill.

An alpha level of .05 was used for all statistical tests apart from where the Bonferroni adjustment was applied to control for Type 1 errors. Missing cases were excluded pairwise.

2.5.2 Sub-study 2 - The ABC assessment data

To compare means and variance between HF-ASD, LF-ASD, MR and TD groups in the clinical data, an independent samples t-test and one-way between-groups analysis of variance (one-way between-group ANOVA) were conducted. Because the data was found to violate assumptions of normality, a Kruskal-Wallis Test followed by a series of Mann-Whitney U Tests were conducted as a non-parametric alternative to the one-way between-group ANOVA. Spearman's Rho correlations were used to explore associations between levels of cognitive functioning (IQ) and motor skill.

A Mann-Whitney U test was conducted to explore possible gender differences within the ASD group on the MSEL fine motor section. A Chi-square test for independence was also conducted to explore potential gender differences when the group had been divided in two based on normal versus impaired motor development.

An alpha level of .05 was used for all statistical tests apart from where the Bonferroni adjustment was applied to control for Type 1 errors. Missing cases were excluded pairwise.

3 Results

3.1 Differences between the ASD group and the typically developing group

A statistically significant difference in the standardized scores from the parental reports of motor skills between children with ASD at 18 months ($Md = 7$) and typically developing children of the same age ($Md = 6$) was found using a Mann-Whitney U test. There was also a significant difference in parental reports of motor skills between children with ASD at 36 months ($Md = 7$) and typically developing children ($Md = 4$) (see table 4 on p. 26 for results and table 5 on p. 27 for statistics for the standardized scores). In both cases, the TD group's scores were significantly lower than the ASD group's scores, indicating that the typically developing children have reached more motor milestones than children with ASD of the same age.

Table 4: Results from between-group analysis of total scores using Mann-Whitney U tests on 18- and 36-month MoBa Questionnaires

	<i>18 months</i>		<i>36 months</i>	
	<i>TD</i>	<i>ASD</i>	<i>TD</i>	<i>ASD</i>
<i>N</i>	399	137	399	137
<i>Md</i>	6	7	4	7
<i>U</i>	16220	16220	8882	8882
<i>z</i>	-8.1	-8.1	-12.6	-12.6
<i>r</i>	.35	.35	.55	.55
<i>p</i>	.000	.000	.000	.000

There was also a greater deal of variation in the ASD group scores than in the TD group scores. On the 18-month questionnaire, the lowest possible score was 6, which indicates that all motor milestones have been reached. The highest possible score was 18, indicating that none of the motor milestones in questions have been reached. On the 36-month questionnaire the minimum score was 4 and the maximum score was 12. On both questionnaires there was a tendency for the typically developing group's scores to cluster at the lower end of the scale, indicating few delays in motor development, whereas the ASD group's scores were distributed across the entire scale, indicating more delays in attainment of motor milestones, as can be seen in fig. 3.1 and 3.2 on p. 27 and 28.

Table 5: Statistics for the standardized scores from the 18- and 36-month MoBa questionnaires

	<i>N</i>	<i>Min.</i>	<i>Max.</i>	<i>Range</i>	<i>Mean</i>	<i>SD</i>
<i>ASD 18mo</i>	137	-.48	5.06	5.54	.70	1.60
<i>TD 18mo</i>	399	-.48	3.68	4.15	-.23	.47
<i>ASD 36mo</i>	137	-.70	3.83	4.54	1.05	1.29
<i>TD 36mo</i>	399	-.70	2.13	2.83	-.36	.50

Figure 3.1: Percentage of children who received each possible total score on the items from the 18-month MoBa questionnaires in the ASD group and the typically developing comparison group, minimum score = 6, maximum score = 18.

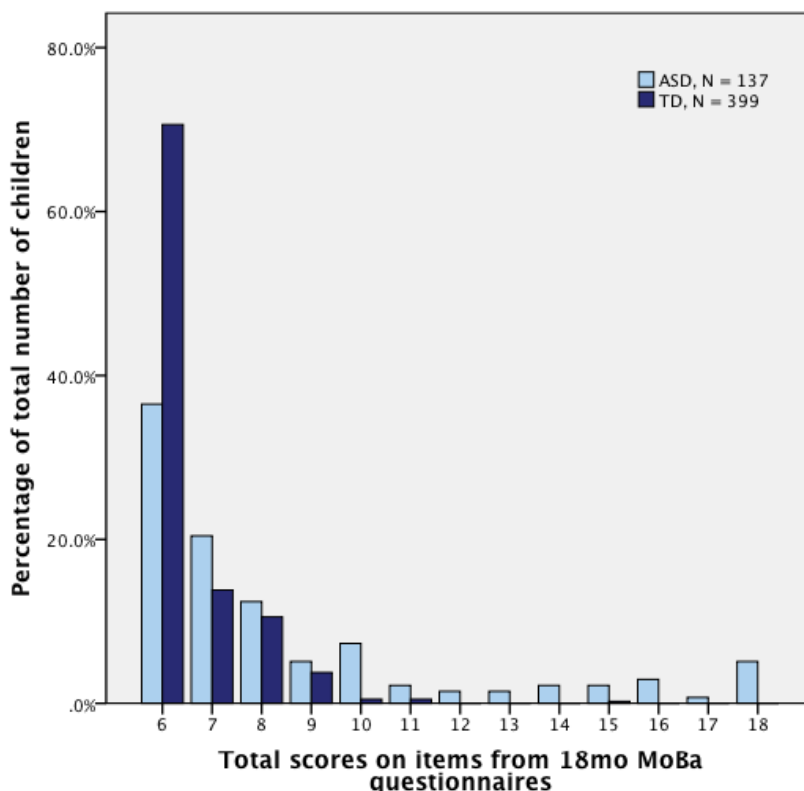
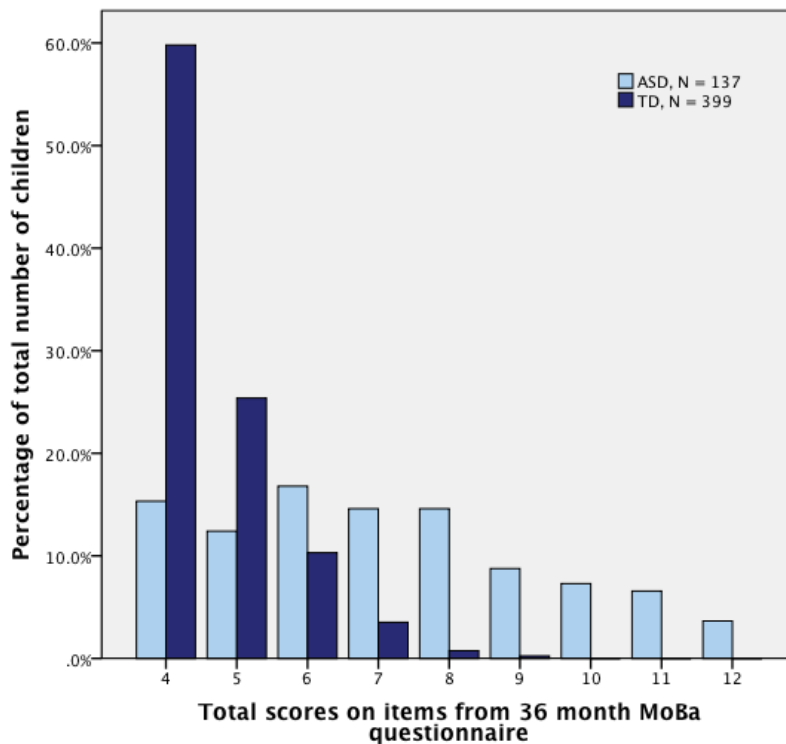


Figure 3.2: Percentage of children who received each possible total score on the items from the 36-month MoBa questionnaires in the ASD group and the typically developing comparison group, minimum score = 4, maximum score = 12.



To explore associations between diagnoses of autism and individual motor skills/milestones described in the 18-month and 36-month questionnaires, a series of Chi-square tests for independence were conducted. The Chi-square tests for independence indicated significant associations between a diagnosis of ASD and parental reports of motor skills at both 18 months and 36 months of age, with the ASD group scores being significantly higher than the typically developing children’s scores on all six motor milestones from the 18 months questionnaire and all four motor milestones from the 36 month questionnaire. Associations between individual skills and ASD did, however, differ, and the strongest association, based on effect size, was found with item Q32.9 “*Can the child turn the pages of a book on his/her own, turning more than one page at a time?*” The weakest associations were found with item Q32.4 “*Does the child move around by walking instead of crawling?*” and item Q32.5 “*Does the child walk steadily, without falling over often?*” from the 18-month questionnaires. The results from the Chi-square tests for independence are presented in table 5 on page 29.

Table 6: Results from Chi-square tests for independence on associations between autism and scores on parental reports of individual motor skills¹

	Item	<i>n</i>	<i>df</i>	χ^2	<i>p</i>	Effect Size: Cramer's <i>V</i>
<i>Items from 18 month MoBa questionnaire</i>	Q32.4 Move by walking instead of crawling	535	2	40.15	.000	.274
	Q32.5 Walk steadily, without falling over often	535	2	21.66	.000	.201
	Q32.6 Walk down stairs if held by the hand	535	2	70.59	.000	.363
	Q32.7 Throw a small ball with a forward motion of the arm	535	2	84.84	.000	.398
	Q32.8 Stack blocks or other small toys	535	2	71.72	.000	.366
	Q32.9 Turn the pages of a book on his/her own, turning more than one page at a time	535	2	322.78	.000	.777
<i>Items from 36 month MoBa questionnaire</i>	Q17.1 Without support, kick a ball by swinging the leg in a forward motion	535	2	68.88	.000	.359
	Q17.2 Catch a big ball with both hands	535	2	77.04	.000	.379
	Q17.3 Hold the pencil/crayon between the fingers and thumb like adults do when drawing	535	2	62.91	.000	.343
	Q17.4 Button and unbutton one or more buttons?	535	2	38.39	.000	.268

Analyses conducted on the sample of children drawn from those assessed in the ABC Study, reported in section 3.3 on page X, confirm these findings. These results clearly indicate a statistically significant difference between the performances of children with autism and the typically developing controls on standardized measures of fine motor skills in addition to parental reports of motor skills, with mean T scores on the MSEL fine motor section significantly higher in the typically developing group than in the autism group, both as a whole and when subdivided into high-functioning ASD and low-functioning ASD.

¹ For full-text items, see table 3 on p. 20

In sum, the children with ASD performed significantly poorer than the comparison group of typically developing children matched on age, gender, parental age and parental education on both parental reports of motor skill, and a standardized measure of motor skill. Although there are many children within the ASD group who lie within the normal range on measures of motor skill, the variation within the group is much higher than in the comparison group.

3.2 Development of motor skills over time

No statistically significant changes in standardized scores from parental reports of motor skills between 18 ($Md = -.478$) and 36 months ($Md = -.142$), were found using the Wilcoxon Signed Ranks test, $z = -.324$, $p = .746$. The median standardized score on the parental reports of motor development increased from 18 months to 36 months but not significantly so, indicating that children who were delayed in reaching motor milestones at 18 months were also delayed at 36 months.

3.3 Relationship between IQ and fine motor skill

In order to explore the impact of level of cognitive functioning on fine motor skill, as measured by the Stanford-Binet V and the Mullen Scales of Early Learning respectively, a one-way between-groups analysis of variance was conducted. Subjects were divided into four groups according to level of cognitive functioning and whether they have an autism diagnosis or not: high-functioning ASD ($IQ > 70$, $n = 17$), low-functioning ASD ($IQ < 70$, $n = 19$), typically developing children ($IQ > 70$, $n = 192$) and children with mental retardation, but no other diagnoses ($IQ < 70$, $n = 36$). In addition, there was a fifth group of children with ASD where IQ was not known ($n = 24$), henceforth referred to as the “uncertain IQ” group. Because preliminary analyses showed that the data violated assumptions of equality of variances, results from the Brown-Forsythe test are reported instead of the ANOVA (Pallant, 2007), and there was a statistically significant difference in MSEL fine motor scores for the four groups being compared. Post-hoc comparisons using the Tukey HSD test (see table 6 on p. 31 for p values) indicated that the mean score for the HF-ASD group ($M = 34.6$) was significantly higher than the LF-ASD group ($M = 25.2$), and the MR group ($M = 20.2$). The mean score for the HF-ASD group was also significantly lower than the mean score for the TD group ($M = 51.9$). The LF-ASD group mean score did *not* differ significantly from that of the MR group or the “uncertain IQ” group ($p = .890$). Mean IQ scores and mean MSEL

scores for each of the five groups as well as the ASD group as a whole can be found in table 8 on p. 32, and mean MSEL scores from the SB5 for each of the four groups are also available in fig. 4 on p. 26.

Table 7: Results from between-group comparisons of the four groups on the Tukey HSD test. Significant results are marked with *.

	TD	HF-ASD	MR	LF-ASD
TD	-	.000*	.000*	.000*
HF-ASD		-	.002*	.006*
MR			-	.002*
LF-ASD				-

Preliminary analyses showed that the data violated assumptions of normality and therefore a Kruskal-Wallis Test was also conducted as a non-parametric alternative to the one-way between-groups ANOVA in order to confirm the results. The Kruskal-Wallis Test revealed a statistically significant difference in MSEL fine motor scores across the groups ($\chi^2 (n = 288) = 172.519, p = .000$).

Table 8: Results from between-group comparisons of the four groups using Mann-Whitney U tests. Significant results with the Bonferroni adjustment applied are marked with *.

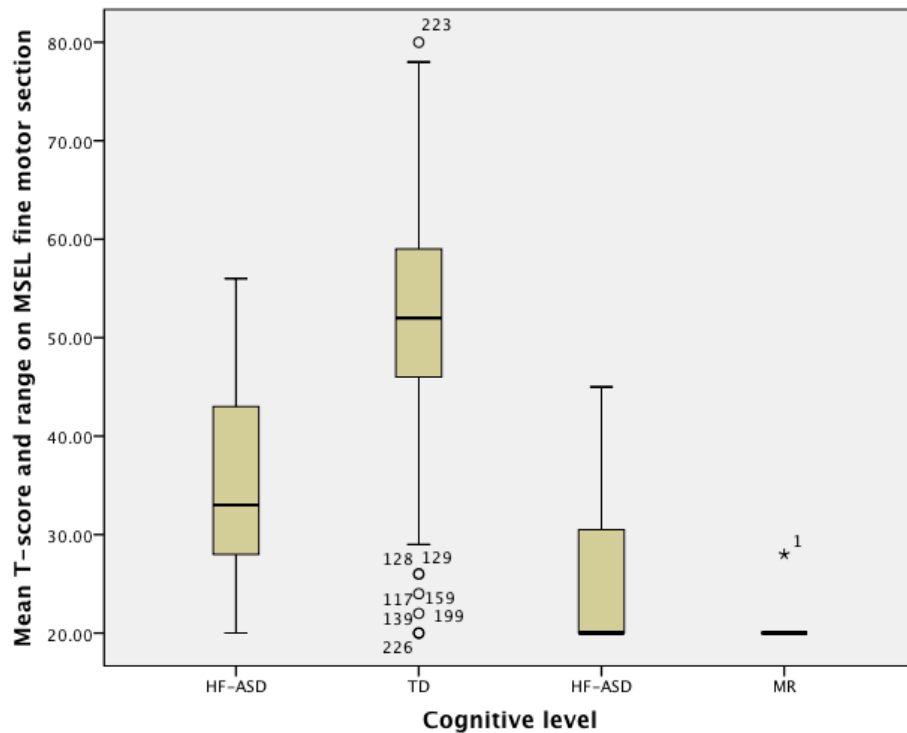
	TD	HF-ASD	MR	LF-ASD
TD	-	.000*	.000*	.000*
HF-ASD		-	.000*	.044*
MR			-	.429
LF-ASD				-

The TD group and the HF-ASD group recorded a higher median score ($Md = 52$, $Md = 33$ respectively) than the other groups, which all recorded median values of 20. To further explore which groups are statistically significantly different from one another, several Mann-Whitney U tests were conducted with a Bonferroni adjustment applied to the alpha values in order to control for Type 1 errors. Since four comparisons were planned, alpha level was adjusted to $p < .0125$ (see table 7 on p. 31 for p values). There were several statistically significant differences in MSEL fine motor scores between the groups being compared. The HF-ASD group scored significantly higher than the LF-ASD group and the MR group, but significantly lower than the TD group. The LF-ASD group MSEL fine motor scores were significantly higher than the scores of the MR group, but there was no significant differences found between the LF-ASD group and the “uncertain IQ” group ($p = .234$), suggesting that the “uncertain IQ” group could be similar to the LF-ASD group.

Table 9: Mean scores from the MSEL fine motor section and SB5, standard deviation and range for each of the five groups being compared, and the ASD group as a whole.

<i>Group</i>	<i>IQ</i>				<i>MSEL Fine Motor</i>			
	<i>N</i>	<i>Mean score</i>	<i>Std. Dev.</i>	<i>Range</i>	<i>N</i>	<i>Mean T-score</i>	<i>Std. Dev.</i>	<i>Range</i>
TD	230	104.95	11.232	63-132	192	51.932	11.52	20-80
HF-ASD	29	90.5172	13.31	71-118	17	34.65	11.01	20-56
LF-ASD	20	59.00	6.74	50-70	19	25.16	8.52	20-45
MR	21	61.4	71.2	50-70	36	20.2	1.3	20-28
ASD	49	77.7	19.1	50-118	60	26.7	9.7	20-56
IQ unknown	28	-	-	-	24	22.29	5.39	20-39

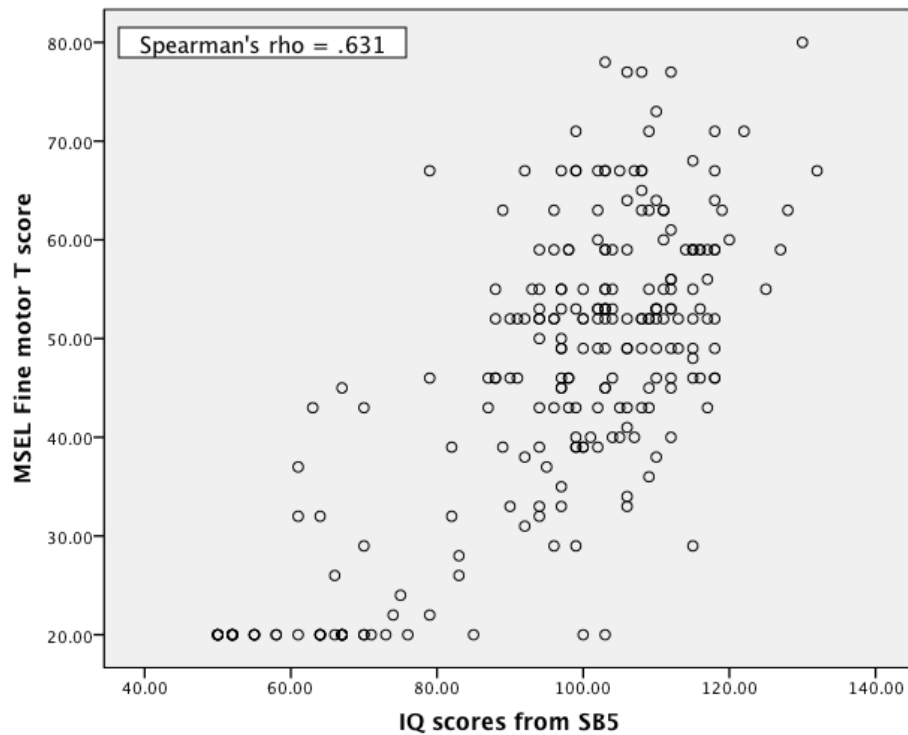
Figure 4: Mean T scores and range on the MSEL fine motor section for each of the four groups being compared



The relationship between IQ (as measured by the SB5) and fine motor skill (as measured by the MSEL) was investigated using Spearman's Rho. Preliminary analyses were performed to ensure no violations of assumptions of linearity and homoscedasticity. A violation of the assumption of normality was already known. There was a strong positive correlation between the two variables ($\rho = .631, n = 247, p < .0005$) with higher levels of cognitive functioning associated with stronger fine motor skills (see fig. 5 on p. 29).

In summary, the results show that children with autism perform significantly poorer than the typically developing controls on measures of fine motor skills, but when dividing the ASD group into HF-ASD and LF-ASD, the LF-ASD group shows no significant difference from a group of controls with mental retardation. However, despite scoring significantly lower than the TD group, the HF-ASD group also scored significantly *higher* than the LF-ASD group. This indicates that there are motor impairments in the ASD group even when IQ has been taken into account, which in turns indicates that motor impairments might be of importance. Further analyses indicated that IQ and motor skills are highly correlated.

Figure 5: Relationship between T scores on the MSEL fine motor section and IQ scores from the SB5 for the ASD group and TD group as a whole



3.4 Differences in motor development within the ASD group

Based on the results from previous analyses, showing a strong indication that IQ and motor skills are related on between-group measures, a Mann-Whitney U test was conducted to explore whether the same association exists *within* the ASD group, between children with and without motor skill deficits. In order to explore possible differences within the ASD group between the children without motor skill impairments and the children with motor skill impairments, the children were divided into two groups depending on their scores on the MSEL fine motor section, or their performance on the abbreviated MSEL, depending on which test was administered. If the child scored equivalent to a t-score of 40 or higher, or had a majority of 1 and/or 2 scores on the abbreviated MSEL, they were placed in the “normal motor development” group. If they scored equivalent to a t-score below 40, or had a majority of 3 and/or 4 scores on the abbreviated MSEL, they were placed in the “impaired motor

development” group. The T-score cut-off points utilized are based on commonly used cut-off points, which consider T-scores of ± 1 standard deviation within the average score of 50 to be within normal range. The Mann-Whitney U test indicated that the IQ scores (as measured by the SB5) of children with autism who have motor skill deficits ($Md = 68.50, n = 34$) were significantly lower than the IQ scores of children with autism who are considered to have motor skills within the normal range ($Md = 97.00, n = 11$), ($U = 68, z = -3.146, p = .001, r = .47$).

3.5 Gender differences

No statistically significant differences between boys ($Md = 7, n = 114$) and girls ($Md = 8, n = 23$) on parental reports of motor skill at 18 months ($U = 1055, z = -1.520, p = .128, r = .13$) were found using a Mann-Whitney U test. There were also no statistically significant differences between boys ($Md = 7, n = 114$) and girls ($Md = 7, n = 23$) on parental reports of motor skill at 36 months, $U = 1230, z = -.468, p = .640, r = .04$.

When examining scores on the MSEL fine motor section, no significant differences in the fine motor skill levels of boys ($Md = 20, n = 44$) and girls ($Md = 20, n = 16$), ($U = 255.5, z = -1.823, p = .068, r = .24$) were found using a Mann-Whitney U test. To confirm these results, a Chi-square test for independence (with Yates’ Continuity Correction) was conducted. This test also indicated no significant association between gender and motor skills, ($\chi^2 (1, n = 67) = 1.279, p = .270, \phi = .183$). Because one of the cells had an expected frequency of less than five, p -value from Fisher’s Exact Test is reported in lieu of the $\chi^2 p$ -value.

4 Discussion

4.1 Differences between the ASD group and the typically developing group

The aim of this study was to investigate variability in motor development among children with autism spectrum disorders (ASD). The results confirm that children with ASD are a very heterogeneous group, with motor skills varying from extremely poor to within normal range. As hypothesized, there is a high prevalence rate of motor skill impairments within the ASD group when compared with typically developing children. The differences between the ASD group and the comparison group are evident on both parental reports of motor skills at both 18 and 36 months of age, and a standardized measure of fine motor skill at 36 months of age. The results were confirmed using both parametric and non-parametric statistical analyses.

Based on the MSEL fine motor section, approximately 71% of the children in the ASD group performed more than one standard deviation below average, indicating that they have motor skills impairments. These findings are in line with previous epidemiological studies of motor skill impairments in children with autism, with reported prevalence rates of up to 73% (Ming et al., 2007; Provost et al., 2006). In the comparison group of typically developing children, on the other hand, only 11.7% of the children performed more than one standard deviation below the average, which is a substantial difference. However, in the comparison group composed of children with mental retardation outside of ASD, every single child included performed more than one standard deviation below average. A relatively large proportion of the children in the MR group also had at least one additional diagnosis (27.5%), such as expressive language disorder, ADHD, phonological disorder or developmental coordination disorder (DCD). In comparison, approximately 18% of the children in the ASD group received additional diagnoses, with the most common diagnoses being ADHD (6.49%), DCD (5.19%) or specific language disorders (5.19%). This discrepancy between the percentages of children receiving additional diagnoses between the two groups could be part of why prevalence rates of motor impairments were higher in the MR than in the ASD group. There are several studies linking motor impairments to both ADHD and language skills, (Alcock & Krawczyk, 2010; Kopp et al., 2010; Martin, Piek, Baynam, Levy, & Hay, 2010), which provide reason to expect higher rates of motor skill impairments in children with lower levels of cognitive functioning comorbid with language skill impairments and/or ADHD.

On the parental reports, the between-group differences manifest as delays in the achievement of both gross- and fine motor milestones, and children with ASD perform more poorly than the typically developing children across the range of individual skills being measured. These results are as expected based on other studies examining motor skills in autism, and add to the body of evidence suggesting motor skills impairments could be an important associated symptom of autism. Deficits and delays in motor skills appear to be one of a range of symptoms distinguishing children with ASD from typically developing children of the same age, but it does not necessarily distinguish them from other groups of children, such as children with developmental delay, mental retardation or other developmental disorders.

4.2 Relationship between levels of cognitive functioning (IQ) and fine motor skills

In the present study, cognitive functioning was strongly related to fine motor skills across all groups being compared. When dividing the ASD group into HF-ASD and LF-ASD, and the comparison group into typically developing children (TD) and children with mental retardation outside of autism (MR), there was a strong association between performance on the MSEL and IQ scores from the SB5. This is relatively unsurprising given evidence from previous studies, which in many cases has indicated that there is such a relationship. However, evidence from previous studies concerning the relationship between levels of cognitive functioning and motor skills has been contradictory. Some studies have indicated that children with ASD have poorer motor skills than typically developing comparison groups even when the effects of IQ have been accounted for, such as a study of fundamental movement skills in 25 children with autism aged 9-12 years (Staples & Reid, 2010). This particular study found that, when compared with a group of children matched on mental age, the children with ASD had more impaired movement skills than would be expected based on their cognitive level (Staples & Reid, 2010). Other studies, such as a study designed to assess motor delay in children aged 21-41 months with ASD, developmental delay or developmental concerns, reported no differences between children with ASD and children with DD (Provost, Lopez, et al., 2007), suggesting that motor impairments in autism are part of a more general developmental delay which also encompasses lower levels of cognitive functioning. There is a significant body of evidence supporting a close correlation between motor skills and levels

of cognitive functioning (e.g. Davis et al., 2011; Green et al., 2009; C. L. Hilton, Zhang, Wilte, Klohr, & Constantino, 2012).

In the current study, it is important to note that even though there *was* a strong correlation between IQ and fine motor skills in this sample, IQ does not fully explain the variation in motor skills in the ASD group. Motor skill impairments were also present in the HF-ASD group, albeit to a lesser degree than in the LF-ASD group, and the difference in fine motor skills between the TD group and the HF-ASD group *was* significant. Variation in levels of cognitive functioning explains some of this relationship, but not all of it. While the TD group's mean IQ score was approximately one standard deviation higher than the HF-ASD group's mean IQ (TD mean IQ = 104, HF-ASD mean IQ = 90), the gap between the two groups' mean T-score on the MSEL was even larger – approximately 1.5 standard deviations apart. This indicates that there are factors in addition to IQ influencing motor development in children with autism, which has also been indicated by a range of previous studies (e.g. Dziuk et al., 2007; Fuentes et al., 2009; Kopp et al., 2010). Given the fact that the data the current study is based upon is from a larger study not originally designed to assess motor skills in particular, further research specifically designed to assess the relationship between motor skills and levels of cognitive functioning is required.

4.3 Development of motor skills over time

When comparing the ASD group with the TD group at both 18 and 36 months of age, it was found that the ASD group scored significantly lower than the TD group on all individual motor skills assessed on a parental report of motor skills. The items from the parental reports in question are based on motor milestones generally achieved at 18 and 36 months respectively. This includes gross motor skills, such as walking unaided, and fine motor skills, such as proper pencil grip and buttoning buttons. In addition to the ASD group being more delayed in their attainment of these milestones than the TD group, it was also found that the children who were delayed in their attainment of motor milestones at 18 months of age were also delayed at 36 months of age. These results were found when examining the children in this sample as a whole after converting the total scores on the parental reports into standardized scores. As a whole, based on standard scores, the children were slightly less delayed at 36 months than at 18 months, but not significantly so. It is possible that this apparent increase in motor skills is due more to the nature of the measure being utilized than the actual skills of the children, since there were fewer items on the 36-month questionnaire

than on the 18-month questionnaire, as well as the fact that only a few skills were evaluated in general. This provides only a very rough measure of motor development without properly distinguishing between gross motor and fine motor skills. A prospective study that examined motor skills in infants considered at risk for developing ASD (e.g. siblings of children with an autism diagnosis) over time (7-36 months of age) using the Mullen Scales of Early Learning and Vineland Adaptive Behavior Scales, found that the gap in fine motor skills between typically developing children and children with autism (at-risk children who were later formally diagnosed) generally increases with age, whereas the gap in gross motor skills tends to *decrease* (Leonard, Elsabbagh, & Hill, 2013). A study by Ming, Brimacombe & Wagner (2007) provides further evidence for a decrease in gross motor abnormalities such as apraxia and hypotonia with age, having found a significant reduction in the prevalence of these issues in a sample of 154 children with autism as the children grew older.

Leonard, Elsabbagh & Hill (2013) also found that correlations between parental reports of motor skills and standardized measures of fine motor skills were generally low with infants, but became stronger as the children got older and the demands on their fine motor control become more stringent. Leonard et al. suggested that it is easier for a trained professional to spot difficulties in fine motor skills in toddlers than it is for parents, and that parental reports of motor skills may, as such, not be an entirely reliable measure. With this in mind, it is important to emphasize that parental reports of motor skills showed stability over time in the current study. However, one must be careful in interpreting these results since there are no standardized measures of motor skill at 18 months available to substantiate the results, and it has not been possible to control for IQ. Still, given the fact that the clinically assessed ASD group had significantly lower scores than the TD group on the MSEL fine motor section at 36 months, it appears likely that delays in motor development continue as the children age, though longitudinal studies with more stringent assessment protocols would be required to elucidate this relationship further.

4.4 Differences in motor development within the ASD group

In order to explore potential differences between the children in the ASD group who were classified as having motor skill impairments (71%) and those who were classified as having motor skills within the normal range, exploratory analyses were conducted. Since earlier

analyses indicated that IQ and motor skills are strongly correlated, the purpose of these analyses was to determine whether this relationship also existed *within* the ASD group. As hypothesized, the results showed that the children in this group with motor skills within the normal range were much more likely to have higher levels of cognitive functioning, whereas the children with poorer motor skills were also likely to have lower levels of cognitive functioning.

An exhaustive review of the literature available has yielded few previous studies examining potential differences between children with ASD who have motor impairments and those who do not. There is, however, evidence from some studies that may indicate future directions of research that could elucidate this relationship further. For instance, a study of neural correlates underlying the disruption of basic motor execution by Mostofsky et al. (2009) which compared a group of 13 individuals between the ages of 8 and 12 years with high-functioning autism to a group of 13 age- and sex-matched typically developing peers found differences in connectivity and cerebellar activity during motor tasks. In this study, the TD group showed greater activity in the expected areas of the cerebellum – those linked to controlling routine movements such as walking. The children with HF-ASD, on the other hand, showed increased activity in the supplementary motor areas of the brain. They also demonstrated decreased connectivity across the motor execution networks in comparison with the TD group. Mostofsky et al. propose that these different activation patterns during motor tasks may represent “difficulty shifting motor execution from cortical regions associated with effortful control to regions associated with habitual execution”. Further, they also suggest that the decreased connectivity may be a reflection of “poor coordination within the circuit necessary for automating patterned motor behaviour” (Mostofsky et al., 2009).

These findings provide evidence that there may be certain brain substrates underlying the motor deficits reported in ASD. Given the fact that Mostofsky et al. only examined high-functioning children with autism, and no comparisons were made between children with and without motor impairments, there are no specific answers to be found concerning the differences between children with ASD with and without motor impairments. There is also the possibility that low levels of cognitive functioning in the LF-ASD groups disguise motor impairments specific to children with autism. Their findings do, however, indicate a potentially useful venue for further research, and several other studies support the possibility of such connectivity issues. The study by Geschwind and Levitt (2007) offers one possible explanation in what they call developmental disconnection. This falls into line with a study by Courchesne & Pierce (2005) that focuses on macroscopic and microscopic evidence of

local over-connectivity and long-distance disconnection within the brain. However, even though evidence from these types of studies offers a tantalizing glimpse into possible etiological models that not only account for the core features of autism, but also for the reported motor impairments and other associated symptoms. Is there a possibility that these differences in activation and connectivity patterns between children with HF-ASD and typically developing children also exist between children with ASD with and without motor impairments?

4.5 Gender differences

In the current study, the ratio between the genders was close to the oft-reported 4:1 ratio in both the clinical sample and the questionnaire sample, and there were no differences between boys and girls on the MSEL fine motor section. Gender differences in autism spectrum disorders, and the idea that gender plays a role in autism, are currently accepted, but there is still uncertainty concerning what that role might be. According to Kopp, Beckung & Gillberg (2010), there are few studies examining motor development in girls with ASD, though work by the authors indicates high prevalence rates of motor difficulties amongst girls as well as boys. The few available studies generally examine small groups, and may, as such, not be representative for the ASD group as a whole. This is an issue for the current study as well. In addition, there is a possibility that many of the girls included in the current study's sample are girls with less severe autism than what is common in the autism population as a whole. Since families participating in the ABC study had to travel to Oslo and, in most cases, stay overnight to partake in the study, this could have prevented families with more severely afflicted children from participating, as traveling, changes and new situations in many cases pose a significant challenge to these families. On the other hand, there were no differences between boys and girls on parental reports of motor skills from the MoBa questionnaires at 18 or 36 months. As the questionnaires were to be filled out at home, they are unlikely to be affected by these kinds of issues. It therefore seems safe to conclude that there are no significant differences between boys and girls with ASD in terms of motor skills in the current study, although there is a possibility that such differences may manifest later in life.

Results are varied regarding the differences in motor skills in boys and girls with autism, albeit there is some evidence that gender plays a role in motor skills. In their study of gender differences in ASD, Penn and colleagues (2007) found that girls with ASD have better motor skills and general adaptive function than boys with ASD, as measured by the VABS.

Other studies have found that boys with ASD perform better than girls in terms of both language skills, motor skills and social development (Nichols, Moravcik, & Tetenbaum, 2009). However, a study of developmental coordination disorder (DCD) and other motor control problems in girls with ASDs and/or ADHD (Kopp et al., 2010) found that the rates of DCD and other motor control deficits were less common in girls with ASD than in other studies examining predominately male groups of children. Based on these results, the authors proposed that there could be actual sex-differences, which may favor girls in terms of motor skills when controlling for IQ. No such differences between the genders were found in the current study.

4.6 Methodological strengths and limitations

4.6.1 Strengths of the current study

In most research on autism, a major limitation lies in the small sample sizes available. In their 2010 meta-analysis Fournier et al. summarize much of the recent research studying motor development in children with autism. The average number of participants in each ASD sample is approximately 19 based on the studies cited, with a large proportion of the studies cited having far fewer participants than this. In the current study, sample size is comparatively large, with 137 children included in the sample based on the questionnaires, and 77 children included in the sample based on the clinical assessments. In addition, comparison groups of typically developing children are approximately 3 times the size of the ASD groups, providing a solid basis for comparison. With such large groups, results from the analysis are much more likely to be robust, thereby reducing the likelihood of making type I or type II errors. It is also more likely that the samples are representative of the population from which they were drawn, evidenced by the variation present in demographical variables.

4.6.2 Limitations of the current study

The main limitation of the present study is that neither the MoBa nor ABC studies were designed to assess motor skills specifically. The available data is therefore very limited, without standardized measures of gross motor skill and only one standardized measure of fine motor skill, which means only limited conclusions may be drawn. Another limitation is that results were compared to a control group consisting of typically developing children. Children with mental retardation are also known to have motor difficulties, and a large portion of the ASD group in this study had cognitive difficulties. Unfortunately it was only

possible to include IQ in the analyses based on the clinical data. It was not possible to control for the effects of IQ in the analyses based on data from the questionnaires without drastically reducing sample size by including only the few children who had completed all relevant items from both of the questionnaires *and* participated in the ABC study, thereby sacrificing statistical power. Possible future studies could consider comparisons with a control group of children with mental retardation/cognitive difficulties across all measures being studied. It may also be prudent to consider comparisons with a control group of children with language impairments as well, since research has indicated a correlation between language impairments and motor skill impairments (Alcock & Krawczyk, 2010; Chuang et al., 2011). It was unfortunately not possible to control for the effects of language in the present study due to a lack of standardized measures of language skill.

A further limitation lies in the fact that there are no available Norwegian norms for the SB5, used to assess levels of cognitive functioning in the ABC study. The norms utilized are based on American populations and might not be wholly appropriate for Norwegian children. Results must therefore be interpreted accordingly.

It should also be mentioned that the sample in the current study might not be entirely representative of neither the population in general nor the clinical population. Participants in the MoBa study generally have a higher socio-economic status than the population average, with higher incomes and longer educations (Stoltenberg et al., 2010). In addition, there is reason to believe that many children from the more severely affected end of the autism spectrum may not have been able to participate in the ABC study, since traveling to attend would have been too difficult for their families.

4.6.3 Concluding remarks

The main findings of the current study, supported by the results from both sub-study 1 and sub-study 2, add to the body of evidence that motor deficits are prevalent amongst individuals with autism. On both parental reports and standardized tests of motor skills, individuals with autism perform more poorly than typically developing children, but not more so than children with mental retardation outside of autism. The second main finding of this study is that variations in motor skills in the ASD group are closely associated with variations in IQ. When dividing the ASD group into individuals with high-functioning autism and low-functioning autism based on IQ, it was found that while the LF-ASD group did not differ from the MR group (both of which have $IQ < 70$) on a standardized measure of motor skill,

the HF-ASD group did differ significantly from the TD group, as well as from the MR group and the LF-ASD group. The HF-ASD group scored significantly higher than the MR and LF-ASD groups, but still significantly lower than the TD group. This could indicate that delays and deficits in motor development amongst children with autism is a phenomenon that distinguishes them from other groups of children. Since IQ accounts for some of the variation in motor skill in the ASD group, but not all of it, one potential hypothesis is that the lower levels of cognitive functioning somehow disguise motor impairments specific to children with ASD in the lower functioning group. This, in turn, indicates that motor impairments might not just be part of a more general developmental delay, but something unique to the autism group itself. This possibility is particularly important to keep in mind, given the fact variations in levels of cognitive functioning do not fully explain the variations in fine motor skills, and there are likely to be other factors in play, which should be examined more thoroughly in future studies.

Therefore, knowing the high proportion of individuals with lower levels of cognitive functioning in the autism group (26% of the sample in the current study had a confirmed IQ < 70 in the current study, while 36% did not complete the SB5 and are also likely to have IQs in the lower range) and the high prevalence rates of motor development issues, it seems important to take motor skills into account when developing interventions for children with ASDs even if motor issues are not a clearly distinguishing feature for the group as a whole. Considering that motor difficulties may contribute to exacerbating other core symptoms by limiting interaction with the physical and social world during critical developmental periods, this is especially important (Anderson et al., 2013; Campos et al., 2000; Fournier et al., 2010). Interventions that also target the development of motor skills may contribute to the individual child's increasing independence, and could, as has been indicated in previous studies, contribute to lowering the threshold for engaging socially with other children (Bhat, Landa, & Galloway, 2011). Because of the individual variability within the ASD group, it would be prudent to include systematic assessments of motor abilities as a routine investigation when assessing a child with autism.

5 References

- Alcock, K. J., & Krawczyk, K. (2010). Individual differences in language development: relationship with motor skill at 21 months. *Developmental Science*, *13*(5), 677–691.
- Anderson, D. I., Campos, J. J., Witherington, D., Dahl, A., Rivera, M., He, M., . . . Barbu-Roth, M. A. (2013). The role of locomotion in psychological development. *Frontiers in Psychology*, *4*, 1–17.
- American Psychiatric Association (2000). *Diagnostic and Statistical Manual of Mental Disorders* (4th Edition, Text Revision ed.). Washington, DC: American Psychiatric Association.
- American Psychiatric Association (2013). Autism Spectrum Disorder Fact Sheet Retrieved 09/02/13, 2013, from <http://www.dsm5.org/Pages/Default.aspx>
- American Psychiatric Association (2013). *Diagnostic and Statistical Manual of Mental Disorders* (5th edition). Washington, DC: American Psychiatric Association
- Baker, A. E., Lane, A., Angley, M. T., & Young, R. L. (2008). The relationship between sensory processing patterns and behavioural responsiveness in autistic disorder: a pilot study. *Journal of Autism and Developmental Disorders*, *38*(5), 867–875.
- Bhat, A. N., Landa, R. J., & Galloway, J. C. (2011). Current perspectives on motor functioning in infants, children, and adults with autism spectrum disorders. *Journal of Physical Therapy*, *91*(7), 1116–1129.
- Bolton, P. F., Golding, J., Emond, A., & Steer, C. D. (2012). Autism spectrum disorder and autistic traits in the Avon Longitudinal Study of Parents and Children: precursors and early signs. *Journal of the American Academy of Child & Adolescent Psychiatry*, *51*(3), 249–260 e225.
- Campos, J. J., Anderson, D. I., Barbu-Roth, M. A., Hubbard, E. M., Hertenstein, M. J., & Witherington, D. (2000). Travel Broadens the Mind. *Infancy*, *1*(2), 149–219.
- Canitano, R. (2011). Neurological Assessment. In E. Hollander, A. Kolevzon & J. T. Coyle (Eds.), *Textbook of Autism Spectrum Disorders* (1st ed. ed., pp. 67–74). London: American Psychiatric Publishing.
- Carr, A. (2006). *The Handbook of Child and Adolescent Clinical Psychology* (2nd ed.). London, England: Routledge.
- Casanova, M. F. (2007). The neuropathology of autism. *Brain Pathology*, *17*(4), 422–433.
- Chuang, Y. C., Hsu, C. Y., Chiu, N. C., Lin, S. P., Tzang, R. F., & Yang, C. C. (2011). Other impairment associated with developmental language delay in preschool-aged children. *Journal of Child Neurology*, *26*(6), 714–717.
- Coskun, M. A., Varghese, L., Reddoch, S., Castillo, E. M., Pearson, D. A., Loveland, K. A., . . . Sheth, B. R. (2009). How somatic cortical maps differ in autistic and typical brains. *Neuroreport*, *20*(2), 175–179.
- Courchesne, E., & Pierce, K. (2005). Why the frontal cortex in autism might be talking only to itself: local over-connectivity but long-distance disconnection. *Current Opinion in Neurobiology*, *15*(2), 225–230.
- Davis, E. E., Pitchford, N. J., & Limback, E. (2011). The interrelation between cognitive and motor development in typically developing children aged 4-11 years is underpinned by visual processing and fine manual control. *British Journal of Psychology*, *102*(3), 569–584.

- Dewey, D., Cantell, M., & Crawford, S. G. (2007). Motor and gestural performance in children with autism spectrum disorders, developmental coordination disorder, and/or attention deficit hyperactivity disorder. *Journal of International Neuropsychological Society*, 13(2), 246–256.
- Dziuk, M. A., Gidley Larson, J. C., Apostu, A., Mahone, E. M., Denckla, M. B., & Mostofsky, S. H. (2007). Dyspraxia in autism: association with motor, social, and communicative deficits. *Developmental Medicine & Child Neurology*, 49(10), 734–739.
- Einfeld, S., & Emerson, E. (2008). Intellectual Disability. In M. Rutter, D. Bishop, D. Pine, S. Scott, J. Stevenson, E. Taylor & A. Thapar (Eds.), *Rutter's Child and Adolescent Psychiatry* (5th ed.): Blackwell Publishing.
- Engeland, H. V., & Buitelaar, J. K. (2008). Autism Spectrum Disorders. In M. Rutter, D. Bishop, D. Pine, S. Scott, S. Stevenson & E. Taylor (Eds.), *Rutter's Child and Adolescent Psychiatry* (pp. 759–783). Oxford: Blackwell Publishing.
- Engeland, H. v., & Buitelaar, J. K. (2009). Autism Spectrum Disorders. In M. Rutter, D. Bishop, D. Pine, S. Scott, J. Stevenson, E. Taylor & A. Thapar (Eds.), *Rutter's Child and Adolescent Psychiatry* (5th ed., pp. 759–781). Malden, Massachusetts: Wiley-Blackwell.
- Enticott, P. G., Kennedey, H. A., Rinehart, N. J., Bradshaw, J. L., Tonge, B. J., Daskalakis, Z. J., & Fitzgerald, P. B. (2013). Interpersonal motor resonance in autism spectrum disorder: evidence against a global "mirror system" deficit. *Frontiers in Human Neuroscience*, 7(218), 1–8.
- Fan, Y. T., Decety, J., Yang, C. Y., Liu, J. L., & Cheng, Y. (2010). Unbroken mirror neurons in autism spectrum disorders. *Journal of Child Psychology and Psychiatry*, 51(9), 981–988.
- Fournier, K. A., Hass, C. J., Naik, S. K., Lodha, N., & Cauraugh, J. H. (2010). Motor coordination in autism spectrum disorders: a synthesis and meta-analysis. *Journal of Autism and Developmental Disorders*, 40(10), 1227–1240.
- Fuentes, C. T., Mostofsky, S. H., & Bastian, A. J. (2009). Children with autism show specific handwriting impairments. *Neurology*, 73(19), 1532–1537.
- Fuentes, C. T., Mostofsky, S. H., & Bastian, A. J. (2010). Perceptual reasoning predicts handwriting impairments in adolescents with autism. *Neurology*, 75(20), 1825–1829.
- Gallahue, D. L., Ozmun, J. C., & Goodway, J. D. (2012). *Understanding Motor Development: Infants, Children, Adolescents, Adults* (Seventh Edition ed.). New York: McGraw Hill.
- Gazzaniga, M. S., Ivry, R. B., & Mangun, G. R. (2009). *Cognitive Neuroscience: The Biology of the Mind* (3rd ed. ed.). New York: W. W. Norton & Company.
- Geschwind, D. H., & Levitt, P. (2007). Autism spectrum disorders: developmental disconnection syndromes. *Current Opinion in Neurobiology*, 17(1), 103–111.
- Green, D., Charman, T., Pickles, A., Chandler, S., Loucas, T., Simonoff, E., & Baird, G. (2009). Impairment in movement skills of children with autistic spectrum disorders. *Developmental Medicine & Child Neurology*, 51(4), 311–316.
- Hadjikhani, N., Joseph, R. M., Snyder, J., & Tager-Flusberg, H. (2006). Anatomical differences in the mirror neuron system and social cognition network in autism. *Cerebral Cortex*, 16(9), 1276–1282.
- Hauck, J. A., & Dewey, D. (2001). Hand preference and motor functioning in children with autism. *Journal of Autism and Developmental Disorders*, 31(3), 265–277.

- Hilton, C., Wente, L., LaVesser, P., Ito, M., Reed, C., & Herzberg, G. (2007). Relationship between motor skill impairment and severity in children with Asperger syndrome. *Research in Autism Spectrum Disorders, 1*(4), 339–349.
- Hilton, C. L., Zhang, Y., Whilte, M. R., Klohr, C. L., & Constantino, J. (2012). Motor impairment in sibling pairs concordant and discordant for autism spectrum disorders. *Autism, 16*(4), 430–441.
- Iacoboni, M., & Dapretto, M. (2006). The mirror neuron system and the consequences of its dysfunction. *Nature Reviews Neuroscience, 7*(12), 942–951.
- Jacob, P. (2009). The tuning-fork model of human cognition: A critique. *Consciousness and Cognition, 18*, 229–243.
- Jasmin, E., Couture, M., McKinley, P., Reid, G., Fombonne, E., & Gisel, E. (2009). Sensori-motor and daily living skills of preschool children with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 39*(2), 231–241.
- Karasik, L. B., Tamis-LeMonda, C. S., Adolph, K. E., & Dimitropoulou, K. A. (2008). How Mothers Encourage and Discourage Infants' Motor Actions. *Infancy, 13*(4), 366–392.
- Kopp, S., Beckung, E., & Gillberg, C. (2010). Developmental coordination disorder and other motor control problems in girls with autism spectrum disorder and/or attention-deficit/hyperactivity disorder. *Research in Developmental Disabilities, 31*(2), 350–361.
- Lecavalier, L., Snow, A. V., & Norris, M. (2011). Autism Spectrum Disorders and Intellectual Disability. In J. L. Matson & P. Sturmey (Eds.), *International Handbook of Autism and Pervasive Developmental Disorders* (pp. 37–52). New York: Springer.
- Leonard, H. C., Elsabbagh, M., & Hill, E. L. (2013). Early and persistent motor difficulties in infants at-risk for developing autism spectrum disorder: A prospective study. *European Journal of Developmental Psychology.*
- Mari, M., Castiello, U., Marks, D., Marraffa, C., & Prior, M. (2003). The reach-to-grasp movement in children with autism spectrum disorder. *Philosophical Transactions of the Royal Society London B: Biological Sciences, 358*(1430), 393–403.
- Martin, C. N., Piek, J., Baynam, G., Levy, F., & Hay, D. (2010). An examination of the relationship between movement problems and four common developmental disorders. *Human Movement Science, 20*, 799–808.
- Matson, M. L., Matson, J. L., & Beighley, J. S. (2011). Comorbidity of physical and motor problems in children with autism. *Research in Developmental Disabilities, 32*(6), 2304–2308.
- Meltzoff, A. N. (2007a). The 'like me' framework for recognizing and becoming an intentional agent. *Acta Psychologica (Amsterdam), 124*(1), 26–43.
- Meltzoff, A. N. (2007b). 'Like me': a foundation for social cognition. *Developmental Science, 10*(1), 126–134.
- Mieres, A. C., Kirby, R. S., Armstrong, K. H., Murphy, T. K., & Grossman, L. (2012). Autism spectrum disorder: an emerging opportunity for physical therapy. *Pediatric Physical Therapy, 24*(1), 31–37.
- Ming, X., Brimacombe, M., & Wagner, G. C. (2007). Prevalence of motor impairment in autism spectrum disorders. *Brain Development, 29*(9), 565–570.
- Mostofsky, S. H., Powell, S. K., Simmonds, D. J., Goldberg, M. C., Caffo, B., & Pekar, J. J. (2009). Decreased connectivity and cerebellar activity in autism during motor task performance. *Brain: A journal of neurology, 132*(Pt 9), 2413–2425.
- Mullen, E. M. (1995). *Mullen Scales of Early Learning*. Oxford: NCS Pearson.

- Newschaffer, C. J., Croen, L. A., Daniels, J., Giarelli, E., Grether, J. K., Levy, S. E., . . . Windham, G. C. (2007). The epidemiology of autism spectrum disorders. *Annual Review of Public Health, 28*, 235–258.
- Nichols, S., Moravcik, G. M., & Tetenbaum, S. P. (2009). *Girls Growing Up on the Autism Spectrum: What Parents and Professionals Should Know About the Pre-Teen and Teenage Years*. London: Jessica Kingsley Publishers.
- Oberman, L. M., Hubbard, E. M., McCleery, J. P., Altschuler, E. L., Ramachandran, V. S., & Pineda, J. A. (2005). EEG evidence for mirror neuron dysfunction in autism spectrum disorders. *Brain Research and Cognitive Brain Research, 24*(2), 190–198.
- Ozonoff, S., Young, G. S., Goldring, S., Greiss-Hess, L., Herrera, A. M., Steele, J., . . . Rogers, S. J. (2008). Gross motor development, movement abnormalities, and early identification of autism. *Journal of Autism and Developmental Disorders, 38*(4), 644–656.
- Pallant, J. (2007). *SPSS Survival Manual: A Step-by-Step Guide to Data Analysis using SPSS version 15* (3rd ed.). Maidenhead, Berkshire: McGraw-Hill Education.
- Provost, B., Heimerl, S., & Lopez, B. R. (2007). Levels of gross and fine motor development in young children with autism spectrum disorder. *Physical and Occupational Therapy in Pediatrics, 27*(3), 21–36.
- Provost, B., Lopez, B. R., & Heimerl, S. (2006). A Comparison of Motor Delays in Young Children: Autism Spectrum Disorder, Developmental Delay and Developmental Concerns. *Journal of Autism and Developmental Disorders, 37*, 321–328.
- Provost, B., Lopez, B. R., & Heimerl, S. (2007). A comparison of motor delays in young children: autism spectrum disorder, developmental delay, and developmental concerns. *Journal of Autism and Developmental Disorders, 37*(2), 321–328.
- Rizzolatti, G., & Fabbri-Destro, M. (2010). Mirror neurons: from discovery to autism. *Experimental Brain Research, 200*(3-4), 223–237.
- Rizzolatti, G., Fabbri-Destro, M., & Cattaneo, L. (2009). Mirror neurons and their clinical relevance. *Nature Clinical Practice Neurology, 5*(1), 24–34.
- Roid, G. H., & Pomplun, M. (2005). Interpreting the Stanford-Binet Intelligence Scales, Fifth Edition. In D. P. Flanagan & P. L. Harrison (Eds.), *Contemporary Intellectual Assessment* (2nd ed., pp. 325–343). New York: The Guilford Press.
- Shalom, D. B. (2009). The medial prefrontal cortex and integration in autism. *Neuroscientist, 15*(6), 589–598.
- Sommerville, J. A., & Decety, J. (2006). Weaving the fabric of social interaction: articulating developmental psychology and cognitive neuroscience in the domain of motor cognition. *Psychonomic Bulletin & Review, 13*(2), 179–200.
- Stanfield, A. C., McIntosh, A. M., Spencer, M. D., Philip, R., Gaur, S., & Lawrie, S. M. (2008). Towards a neuroanatomy of autism: A systematic review and meta-analysis of structural magnetic resonance imaging studies. *European Psychiatry, 23*(4), 289–299.
- Staples, K. L., & Reid, G. (2010). Fundamental movement skills and autism spectrum disorders. *Journal of Autism and Developmental Disorders, 40*(2), 209–217.
- Stenberg, N. (2012). Identifisering av autismspekterforstyrrelser: Tidlige tegn og differensialdiagnostiske utfordringer. *Autisme i dag, 39*(1), 3–10.
- Stoltenberg, C., Schjøberg, S., Bresnahan, M., Hornig, M., Dirtz, H., Dahl, S., . . . Lipkin, W. (2010). The Autism Birth Cohort: a paradigm for gene-environment-timing research. *Molecular Psychiatry, 15*, 676–680.
- Tetzchner, S. v. (2012). *Utviklingspsykologi* (2nd ed.). Oslo: Gyldendal Akademisk.

Ventola, P., & Tsatsanis, K. D. (2011). Associated Symptoms. In E. Hollander, A. Kolevzon & T. C. Joseph (Eds.), *Textbook of Autism Spectrum Disorders* (pp. 189–200). London, England: American Psychiatric Publishing

