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The co-occurrence of motor and language impairments in children evaluated for autism spectrum disorder. An explorative study from Norway.

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ABSTRACT

Background: Current research suggest that motor and language impairments are common and closely related in infants with autism spectrum disorder (ASD). In older children, less is known about how these impairments are related to each other.
Aims: The current study explored the co-occurrence and potential impact of motor and language impairments in a sample of school-aged children evaluated for ASD by Norwegian specialist health services.
Methods: Besides clinical evaluation for ASD, all participants (N = 20, mean age 10.7 (SD = 3.4) years) underwent a standardized test of motor performance (MABC-2), parent report measures of current motor (DCDQ'07), language (CCC-2), and social (SRS) skills, and a caregiver interview on everyday functioning, providing an overall impairment score (DD-CGAS).
Results: The majority (85%) had motor and/or structural language deficits in addition to their social impairment. All children identified with motor impairment on both measures (39%) also had structural language deficits. Better motor performance was strongly correlated with better structural language skills (r = .618, p = .006).
Conclusions: Our findings suggest that co-occurring motor and structural language deficits should be anticipated and assessed when evaluating children for ASD. These deficits may need specific interventions that complement those targeting social skills deficits and other ASD core symptoms.

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1. Introduction

Autism spectrum disorder (ASD) is a common and highly heterogenous neurodevelopmental disorder (NDD). Assigning an ASD diagnosis requires persistent deficits in social communication and interaction, alongside atypical and restricted patterns of behavior sufficiently severe to cause functional impairment (American Psychiatric Association, 2013). Yet, there is considerable variation in the clinical presentation of children with ASD. The core social deficiency of ASD is now considered a continuous trait with no natural cut-off between ASD and subthreshold autistic traits (Happé and Frith, 2020).

The revised Diagnostic and Statistical Manual of Mental Disorders (DSM-5) (American Psychiatric Association, 2013) has recognized the complex presentations of ASD. Acknowledging the need to interpret core symptoms within a broader developmental context, cognitive and language level specifiers to be noted alongside the diagnosis have been included (American Psychiatric Association, 2013). It is now recommended that common comorbidities are investigated and treated in children with ASD (Fuentes, Hervás, & Howlin, 2020; Hyman, Levy, & Myers, 2020). Furthermore, that evaluation includes assessment of potential needs beyond diagnosis, to avoid children with subthreshold symptoms but significant impairment missing out on vital services (Thapar, Cooper, & Rutter, 2017). While motor and language impairments (beyond not having a functional language) are common in children with autistic symptoms, current evidence suggest they are often poorly recognized and triaged behind core ASD symptoms in both evaluation and treatment planning (Bhat, 2020; Boucher, 2012; Licari et al., 2019; Suren et al., 2019). Thus, many children evaluated for suspected ASD potentially miss out on available interventions. Motor and language impairments often present at an early age, prior to formal diagnosis (Hyman et al., 2020). Early fine- and gross motor skills have been linked to concurrent and future communication in infants with ASD (West, 2019), including expressive language (LeBarton & Landa, 2019). Whether the same co-occurrence and close relationship between motor and language impairments seen in infants with ASD are also present in school-aged children evaluated for ASD is currently unknown but of great interest as it could inform potential targets for earlier identification and intervention for affected children.

1.1. *Motor and language impairments in children with autistic symptoms*

Although not universal or specific to the disorder, motor deficits are common in children with ASD, even within the first year of life (West, 2019), and across the range of autistic symptoms and cognitive abilities (Fournier, Hass, Naik, Lodha, & Cauraugh, 2010; Licari et al., 2019; Reindal et al., 2020). Possible deficits include delayed attainment of motor milestones, deviant muscle tone, balance, gait, fine and gross motor coordination (Fournier et al., 2010; West, 2019), some of which may be captured by parent report or standardized assessment of motor skills. Depending on age, criteria, and measures applied, as many as 25–90% of children with ASD may have co-occurring developmental coordination disorder (DCD) (Kopp, Beckung, & Gillberg, 2010; Miller et al., 2021). Still, motor deficits were recognized by clinicians at a low rate (1.34%) relative to their prevalence (35.4%)(Licari et al., 2019), indicating the need for more knowledge.

Social communication difficulties are a core diagnostic feature of ASD, albeit with wide variation in functional language (American Psychiatric Association, 2013). Comprehension and expression of language *form* and *content* (structural language skills), as well as appropriate *use* of verbal and nonverbal language in social contexts (pragmatic language skills) may all be affected alone or in combination to cause *language impairments* (Baird & Norbury, 2016). Although of importance for specifying language in ASD, the extent and role of structural language deficits has received less attention than the more prominent pragmatic difficulties (Boucher, 2012; Reindal et al., 2021). However, their reported variability with deficits often presenting early and being associated with persisting impairments, render them a potential target of early identification and intervention for subgroups within the autism spectrum (Boucher, 2012; Reindal et al., 2021).

1.2. *The relationship between motor, language, and social communication impairments*

Longitudinal data suggest that early motor deficits may be a risk factor for later motor difficulties, but also for the development of language and social communication difficulties related to ASD (LeBarton & Landa, 2019; Leonard, Bedford et al., 2014). Developing motor skills enables the infant to interact with other people and their surroundings, and are considered to assist the development of language and communication (West, 2019). Early motor disruptions could therefore have downstream effects that further compromise language and social development in children with ASD. In a recent meta-analysis West (2019) aggregated data from 890 infants with ASD (age 6.0–42.9 months) across nine studies. A significant association between motor and language/communication skills was found ($r = .35$, $p < .001$), that held for both fine and gross motor skills. In school-aged children with ASD, motor and language impairments have mostly been studied separately, not addressing their potential co-occurrence or additive impact. An exception is the cross-syndrome study by McPhillips, Finlay, Bejerot, and Hanley (2014), where an association between motor performance (standardized assessment) and general communication skills (teacher report) among children with ASD (n = 28; mean age 9 years 11 months) was reported. More recently, Bhat (2021) analyzed parent reported motor skills from 13,887 children with ASD in the SPARK study. An increasing risk for motor impairment was found with greater language, social communication, cognitive, and functional impairments. However, none of these studies investigated language deficits beyond general communication or language functioning.

1.3. *Theoretical and clinical importance of co-occurring motor and language impairments*

Despite efforts to improve earlier diagnosis, many children are school-aged when they receive their ASD diagnosis (Lord, Elsabbagh, Baird, & Veenstra-Vanderweele, 2018). Levy et al. (2010) found that children with ASD and co-occurring diagnoses were diagnosed later, indicating that the ASD was masked by other problems. Compared to a decade ago many individuals referred for ASD assessment are now more language-abled, display milder symptoms, and are diagnosed with ASD based on fewer symptoms (Arvidsson, Gillberg, Lichtenstein, & Lundström, 2018; Avlund, Thomsen, Schendel, Jørgensen, & Clausen, 2021). However, perceived impairment has increased, and most in individuals with autistic symptoms that previously were considered subthreshold (Lundström et al., 2021), suggesting that overall impairment might reflect overlooked co-occurring problems that may be better predictors of support need than the ASD diagnosis alone (Gillberg & Fernell, 2014).

Functional impairment refers to the extent to which a diagnosed condition results in limitations in daily life and is found to predict future adolescent problems (Costello, Angold, & Keeler, 1999) and adult outcomes (Copeland, Wolke, Shanahan, & Costello, 2015). In children with ASD, motor impairments may affect participation in leisure activities, sports, or interactive play "beyond the effect of their social skills alone" (Hyman et al., 2020, p.27). Language impairments may further limit social learning opportunities. In school-aged children with ASD or high autistic traits, co-occurring language and motor difficulties have been linked to reduced daily living skills, participation in physical education and out-of-school activities, overall impairment and contact with services (Bhat, 2021; Hilton, Crouch, & Israel, 2008; Kopp, Beckung, & Gillberg, 2010; Licari et al., 2019; Posserud, Hysing, Helland, Gillberg, & Lundervold, 2018). However, few studies have investigated the co-occurrence of motor and language impairments, their relationship to each other and to overall functioning in school-aged children evaluated for ASD.

1.4. *Aims of the current study*

In this exploratory study we assessed the co-occurrence of motor and language impairments in a sample of school-aged children evaluated for ASD by specialist health services. We further explored relationships between motor, language, social, and overall functional impairment, regardless of meeting the diagnostic criteria for ASD or having subthreshold autistic symptoms. The following objectives were addressed:

1. To explore the co-occurrence of motor and language impairments, in particular structural language deficits, as measured by parent report and standardized assessment of motor skills.
2. To explore the relationship between motor, structural language, and social skills.
3. To assess overall functional impairment and participation, and explore potential relationships with motor, structural language, and social skills.

2. Materials and methods

2.1. *Study design and participants*

The study has a cross-sectional design including children referred for evaluation of ASD at four outpatient clinics, providing public specialist child and adolescent mental health services (CAMHS) in Mid-Norway.

We invited children participating in an ongoing large multi-site study on NDDs in Norway, in which children are eligible for enrollment if a suspicion of ASD has been raised by local or specialist health services (BUPgen, see Reindal et al., 2020). Children aged 6–18 years with available information on ASD diagnostic status were eligible for participation in the present study. Medical records were reviewed to ensure that the participants did not have moderate or severe intellectual impairment, severe sensory, neurological, or muscular impairments that could interfere with motor testing. As one of the assessments required that the child could speak in at least simple sentences, all participants were verbal. Children and their caregivers also had to be sufficiently fluent in Norwegian language. A total of 20 children and adolescents with mean age 10.7 (SD = 3.4, range 6–17) years at inclusion were eligible, of which 15 had been diagnosed with ASD. For simplicity, we use the term 'children' or 'school-aged children' to refer to the whole group. Retrospective data on clinical assessments, parent-reported history, and supplementary parent-reported measures, as well as data from the additional assessment in the present study were collected.

Written informed consent was obtained from all parents and participants (when appropriate due to age) before inclusion in the study. The study was approved by the Norwegian Regional Committee for Medical and Health Research Ethics South East (REK#2016/1954; REK#2012/1967), and the Norwegian Data Inspectorate. The study was performed in accordance with the ethical standards of the 1964 Declaration of Helsinki and its later amendments.

2.2. *Measures*

2.2.1. *Diagnoses*

All diagnoses were assigned by Norwegian specialist health services, using the *International Classification of Diseases, 10th Revision* (ICD-10) criteria (World Health Organization, 1992). All participants had completed either the Autism Diagnostic Interview-Revised (ADI-R; n = 1) (Rutter, Le Couteur, & Lord, 2003), the Autism Diagnostic Observation Schedule (ADOS; n = 2) (Lord, Rutter, DiLavore, & Risi, 1999), or both (n = 17) as part of their clinical evaluation. NDDs were grouped according to ICD-10 codes into the following categories: ASD (F84), intellectual disability (F70–79), attention-deficit/hyperactivity disorder (ADHD) (F90), communication disorder (F80), specific learning disorder (F81 and F83), motor disorder (F82: DCD and F95: tic disorders), other NDD (F88, F89 and F94). The presence of previous or currently active epilepsy was also included in the total number of NDDs.

2.2.2. Motor skills

The Developmental Coordination Disorder Questionnaire 2007 (DCDQ'07) (Wilson et al., 2009) was used to ascertain everyday motor skills, as reported by parents. The DCDQ'07 is a 15-item questionnaire to screen for DCD and to confirm the functional consequences of a motor deficit in clinical and research settings (Wilson et al., 2009). Raw scores for three subscales (*control during movement, fine motor/handwriting,* and *general coordination*) are summarized into a total score, with possible values from 15 to 75. The original version has a high internal consistency (Cronbach's alpha = .89) and concurrent validity with the original Movement Assessment Battery for Children (MABC; $r = -.55$) (Wilson et al., 2009). For the present study an unpublished prefinal Norwegian version of the DCDQ'07 (Wilson et al., 2009); Norwegian cross cultural adaptation by V. Johannesen, H. A. Lillehaug, N. R. Nielsen, G. Skard & S. van Zuiden, 2012), was used with the recommended age-appropriate cut-offs to indicate the presence of motor difficulties (Wilson et al., 2009). Cronbach alpha was .86 (DCDQ'07 total).

The Movement Assessment Battery for Children-2 (MABC-2) (Henderson, Sugden, & Barnett, 2007) is a standardized assessment of fine and gross motor skills frequently used to identify children with motor difficulties for clinical or research purposes. Eight individual test items grouped into three categories (*manual dexterity*, *aiming and catching*, and *balance*) are given a raw score and a standard score, that translate to a component score. From the three categories, a total test score is derived and an overall percentile in that child's age band. While total test score ≤ 5th percentile is considered to represent a definite motor problem requiring motor intervention, scores between the 5th and the 15th percentile suggest a borderline degree of motor difficulties (Henderson et al., 2007).

The MABC-2 was administered by either the fourth (n = 16) or the first author (n = 4), both trained in the assessment. The 15th percentile on the MABC-2 and the appropriate cut-off for the child's age on the DCDQ'07 was used to identify *'motor deficits'* or *'motor difficulties'* (these terms are used interchangeably). *'Motor impairment'* refers to being identified with *'motor deficits'* on both measures. Notably, the MABC-2 protocol, as described in Liu and Breslin (2013), was modified by showing a picture of each task to the child and minimizing the verbal instructions to emphasize visual supports.

2.2.3. Language skills

Language skills were assessed using *the Children's Communication Checklist Second Edition (CCC-2)* (Bishop, 2003; Norwegian version: Bishop, 2011), completed by parents. This checklist consists of 70 items to screen for the presence and profile of language deficits in children who can speak in at least simple sentences. Items are grouped into 10 subscales (A-J) that measure different aspects of communication: *language structure* (A-D), *pragmatic language skills* (E-H), and two scales measuring *social aspects* (I, J). The Cronbach alpha in the present sample was .97 (total alpha, based on raw scores), comparable to previous reports (e.g., Helland, Biringer, Helland, & Heimann, 2009). We report the General Communication Composite (GCC), an overall measure of communication skills (sum A-H), with a suggested cut-off < 55 to identify *'language impairment'* (Bishop, 2003, 2011). Further, we used the Structural Language Score (sum A-D) and the General Pragmatics Score (sum E-H) (see Reindal et al., 2021) as continuous measures of structural and pragmatic language skills. *'Structural language deficits'* were defined as having a score ≤ 5th percentile on two or more of the structural subscales, comprising 'speech' (A), 'syntax' (B), 'semantics' (C), and 'coherence' (D).

2.2.4. Social and cognitive skills

The Social Responsiveness Scale (SRS) (Constantino & Gruber, 2005) was collected to reflect parent reported current (last 6 months) social impairment. This 65-item questionnaire ascertains autistic symptoms across the spectrum of difficulties, with higher scores indicating greater social impairment (Constantino et al., 2003). SRS raw total score was applied as a dimensional trait variable, for which the Cronbach alpha was .95. To assess clinical-level social impairment we converted raw scores to T scores ($M = 50$, $SD = 10$), according to the SRS manual, finding that all participants had *T*-score ≥ 60 (clinical range).

All children had completed formal testing of cognitive abilities with age-appropriate Wechsler scales as part of their clinical evaluation. Standard scores for nonverbal and verbal IQ were available for 16 children, for one child only verbal IQ was available, and for three others the IQ scores were not available. Mean age at assessment (n = 19) was 8.8 ($SD = 2.6$) years.

2.2.5. Functional impairment and participation

The Developmental Disability-Children's Global Assessment Scale (DD-CGAS) (Wagner et al., 2007) was rated to provide a measure of overall functional impairment during the previous month. The DD-CGAS is a revised version of the Children's Global Assessment Scale (CGAS) (Shaffer et al., 1983). In the DD-CGAS, text revisions are introduced to enable a more targeted functional assessment of children with NDDs such as ASD. The instrument focuses on four domains: *self-care, communication, social behavior,* and *school functioning* (Wagner et al., 2007). Scores range from 1 (most impaired) to 100 (superior functioning), with scores < 70 indicating clinically relevant atypical functioning (Wagner et al., 2007). The DD-CGAS has been translated to Swedish, with good inter-rater reliability in ASD cases (Choque Olsson & Bolte, 2014). Convergent validity with measures of adaptive functioning and autistic symptom severity have been reported for the original version (Wagner et al., 2007).

The DD-CGAS was translated into Norwegian for this study, after permission from the original author. Individual DD-CGAS scores were assigned by the same rater (first author), based on all available information at inclusion, including a semi-structured interview with the caregiver(s). During this interview, caregivers were asked to compare their child's functioning and participation, as well as necessary environmental accommodations and level of support, to same-aged peers across functional domains. As part of the DD-CGAS rating, the level of impairment across four domains (self-care, communication, social behavior, and school functioning) was classified as 'not present', 'slight', 'moderate', 'severe', or 'extreme', while considering the child's behavior across environments, and the accommodations necessary to support the child. The DD-CGAS score was chosen to best reflect overall impairment across domains. Supplementary information on participation was collected from the *Child Behavior Checklist/6–18* (Achenbach & Rescorla, 2001), completed by caregiver(s) at inclusion.

2.3. Procedure

After consent to participate, an appointment for inclusion was made and report forms were sent to the caregiver(s) for completion prior to the assessment. Each child was assessed in one session. Both caregivers were invited to complete report forms, while ensuring that the same caregiver(s) participating in the interview completed a set of forms for each participant. In the following, only data reported by the interviewed caregiver (85.0% mother, 5.0% father, 10.0% both parents) are included.

2.4. Statistical analyses

Descriptive statistics are presented as n (%) and mean *(SD)*. First, we report the proportion of children with scores to indicate motor (DCDQ'07, MABC-2) and language (CCC-2) impairment or deficits, as well as their co-occurrence. Second, scatter plots and correlations were used to explore the relationship between motor, structural language, and social skills. For these analyses symptoms were not dichotomized, but the total scores on the respective skill measures were used as dimensional trait variables. Lastly, we report functional impairment by the mean DD-CGAS, the proportion of children with a moderate to severe level of impairment across functional domains, and overall impairment in the clinical range. Relationships with motor, structural language, and social skills were explored. Spearman's rank correlation was used to analyze the relationship between the different measures, because of non-normality of some of the continuous variables. The magnitude of effect sizes was interpreted as small (r = .10 to .29), medium (r = .30 to .49), or large (r = .50 to 1.0) (Cohen, 1992). Two-sided p values < .05 were regarded as statistically significant. IBM SPSS 27/28 was used for statistical analyses.

3. Results

3.1. Participant characteristics

The sample included 20 children (6 girls), born between 2000 and 2013, with a mean (*M*) age of 10.7 (SD = 3.4) years. Of these, 15 children (75.0%) were diagnosed with ASD, while 5 children did not receive an ASD diagnosis (non-ASD). ASD subtypes included childhood autism (26.7%), atypical autism (6.7%), Asperger syndrome (40.0%) and pervasive developmental disorder not otherwise specified (26.7%). Mean age at ASD diagnosis was 10.2 (SD = 3.3) years. All children in the non-ASD group were diagnosed with one or more NDDs, most commonly ADHD (n = 4). Within the whole sample, frequent NDDs beyond ASD were ADHD (n = 9), tic disorders (n = 3), and epilepsy (n = 3). Average cognitive abilities were in the normal range (Table 1).

Most parents were Norwegian or European in origin (97.5%). All participants were followed-up by municipal services and/or specialist health services. All participants attended mainstream schools, albeit with 85.0% receiving special adaptations (e.g., support teaching, own curriculum, daily schedule, social skills training, own assistant).

Table 1
Participant characteristics.

	n	%	Mean	*SD*
Age at inclusion (years)	20		10.7	3.4
SRS raw total	20		88.3	29.3
DCDQ'07 total	20		48.0	11.3
MABC-2 total	18		8.1	2.7
Manual Dexterity	19		8.4	2.4
Aiming & Catching	19		7.5	3.5
Balance	18		9.1	3.2
CCC-2				
General Communication Composite (GCC)	20		36.1	21.5
Structural Language Score	20		18.6	12.4
General Pragmatics Score	20		17.5	9.8
Nonverbal IQ	16		101.3	15.6
Verbal IQ	17		93.4	16.7
DD-CGAS	20		61.1	8.4
Comorbidity				
≥ 2 NDDs	10	50.0		
≥ 1 psychiatric disorder	5	25.0		
≥ 1 somatic disorder	9	45.0		
Current medication	15	75.0		

CCC-2 = Children's Communication Checklist Second Edition; DCDQ'07 = Developmental Coordination Disorder Questionnaire 2007; DD-CGAS = Developmental Disability-Children's Global Assessment Scale; IQ = intelligence quotient; MABC-2 = Movement Assessment Battery for Children-2; NDD = neurodevelopmental disorder; *SD* = standard deviation; SRS = Social Responsiveness Scale.

3.2. Co-occurring motor and/or language impairment

Most children (80.0%) were rated by their caregivers as having motor deficits on the DCDQ'07. Limitations in all aspects of motor functioning (*control during movement, fine motor/handwriting, general coordination)* were reported. On standardized assessment with the MABC-2, seven of 18 children with valid results (38.9%) had total scores indicating motor deficits. Although all subdomains were affected, composite scores indicated most difficulties with *manual dexterity* and *aiming & catching* (36.8% for each subdomain). Due to a technical error on one task, total test score could not be calculated for one child, and MABC-2 results for another child were not valid because of intercurrent illness.

The distribution of DCDQ'07 and MABC-2 total scores are shown in Fig. 1. All cases identified with motor deficits on the MABC-2 (n = 7) were also captured by the DCDQ'07. The DCDQ'07 total was positively correlated with the MABC-2 total score, with Spearman's rho = .211, although not significant and with a small effect size.

The proportion of children with scores below cut-off to indicate language impairment on the CCC-2 was also large (75.0%) (Table 2). A smaller proportion (55.0%) had structural language deficits, i.e., they had subscale scores at or below the 5th percentile on two or more of the structural subscales ('speech', 'syntax', 'semantics', 'coherence'). Deficits were observed across all subscales, albeit with 'syntax' being relatively spared. Taken together, co-occurring motor and structural language deficits were common, with all but three children (85.0%) having deficits in one or both developmental domains (Table 2). All children identified with *motor impairment* also had structural language deficits. The three children with no co-occurring deficits had all been diagnosed with ASD.

3.3. The relationship between motor, structural language, and social skills

Motor performance and structural language skills varied both among children diagnosed with ASD and children with subthreshold autistic symptoms (non-ASD) (Fig. 2). Within the whole sample, a strong, positive correlation was found between MABC-2 total and the Structural Language Score (Spearman's rho = .618, p = .006) (Table 3), indicating that better motor performance on standardized assessment was associated with more advanced structural language. A strong, negative association was also found between the MABC-2 and the SRS total (Spearman's rho = –.521, p = .027), as well as between the Structural Language Score and the SRS total (Spearman's rho = −.691, p < .001). Thus, better motor and structural language skills were both associated with less social impairment, as reported by parents on the SRS.

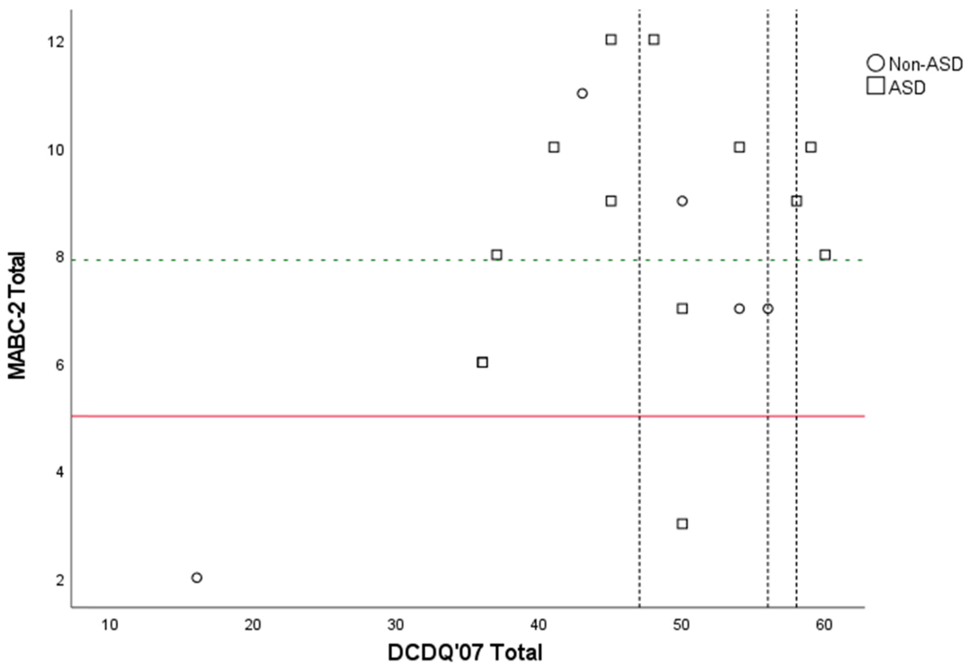

Fig. 1. Distribution of DCDQ'07 and MABC-2 total scores (n = 18), by diagnostic group. Horizontal lines indicate the MABC-2 cut-off for motor difficulties (at or below the 15th percentile; green dashed line) and more definite motor problems (at or below the 5th percentile; red solid line). Dashed vertical lines indicate the DCDQ'07 cut-off for motor difficulties at ages 5:0–7:11 (<47), 8:0–9:11 (<56) and 10:0 and older (<58). 2 participants are not represented on this graph as they did not have valid MABC-2 total scores. 2 children had identical scores on both measures and appear as a single point in the distribution. ASD = autism spectrum disorder; DCDQ'07 = Developmental Coordination Disorder Questionnaire 2007; MABC-2 = Movement Assessment Battery for Children-2.

Table 2
Frequencies and percentages for classifications of functional motor and language performance.

Measure	Classification	Whole sample *n* (%)
DCDQ'07 (n = 20)	Motor deficits*	16 (80.0)
MABC-2 (n = 18)	Motor deficits*	7 (38.9)
MABC-2 and DCDQ'07 (n = 18)	Motor impairment**	7 (38.9)
CCC-2 (n = 20)	Language impairment (GCC<55)	15 (75.0)
	Structural language deficits***	11 (55.0)
MABC-2 and DCDQ'07 and CCC-2 (n = 18 to 20)	Motor impairment** *and* structural language deficits***	7 (38.9)
	Motor deficits* *and* structural language deficits**	3 (15.0)
	Motor deficits* *only*	6 (30.0)
	Structural language deficits*** *only*	1 (5.0)
	None	3 (15.0)

*Total scores below cut-off to indicate motor deficits on either the DCDQ'07 or the MABC-2.
**Total scores below cut-off to indicate motor deficits on both the DCDQ'07 and the MABC-2.
*** ≤5th percentile on two or more structural subscales on the CCC-2 (A-D).
CCC-2 = Children's Communication Checklist Second Edition; DCDQ'07 = Developmental Coordination Disorder Questionnaire 2007; MABC-2 = Movement Assessment Battery for Children-2

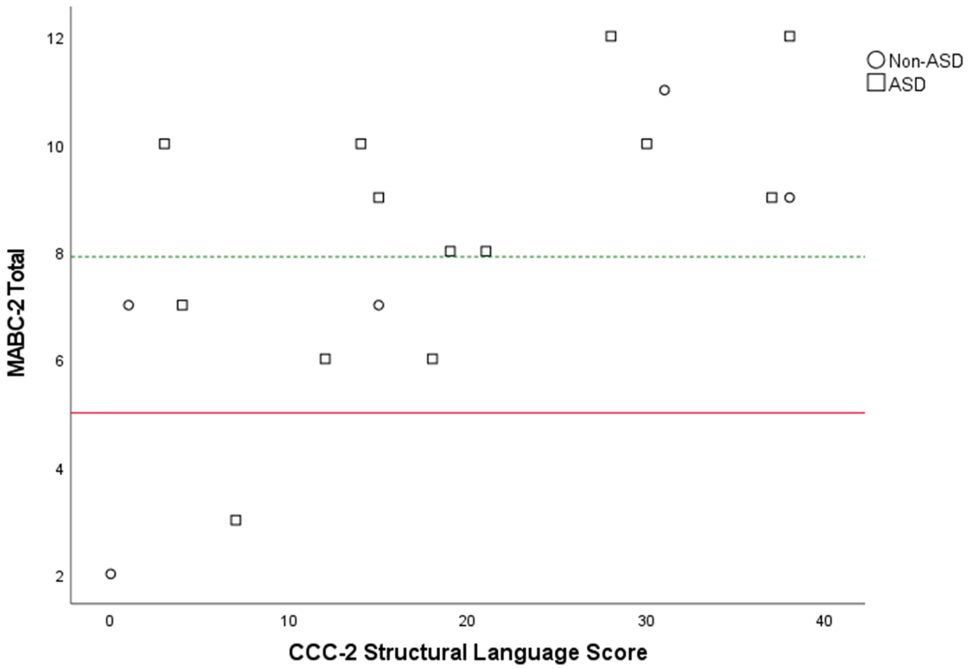

Fig. 2. The relationship between structural language and motor skills, illustrated by the distribution of scores on the Children's Communication Checklist-Second Edition (CCC-2 Structural Language Score) and the Movement Assessment Battery for Children-2 (MABC-2 Total), by diagnostic group. Horizontal lines indicate the MABC-2 cut-off for motor difficulties (at or below the 15th percentile; green dashed line) and more definite motor problems (at or below the 5th percentile; red solid line). ASD = autism spectrum disorder.

3.4. Functional impairment and participation

DD-CGAS scores ranged from 43 to 73, implying considerable variations in overall functioning during the last month. Two children had scores indicating overall functioning consistent with that of same-aged peers (DD-CGAS>70). The rest (90.0%) had scores in the clinical range, although mostly varying within the "upper half" of the scale. The DD-CGAS was negatively correlated with SRS total score, with a large effect size, albeit not reaching statistical significance (Table 3). Notably, all children with childhood autism (n = 4) had DD-CGAS ≤ 61, while children with other ASD subtypes (n = 11) had DD-CGAS ≥ 61. In the non-ASD group, DD-CGAS ranged from 44 to 68. To assess whether DD-CGAS scores were associated with core ASD symptoms, we performed an additional correlation analysis between DD-CGAS and ADI-R verbal total among individuals with available scores (n = 16). A strong negative correlation was found (Spearman's rho= −.657, 95% CI [−.873 to −.224], *p* = .006), suggesting that less prominent core ASD symptoms were associated with better overall functioning.

None of the participants presented with an extreme level of impairment regarding *self-care, communication, social behavior,* or *school*

Table 3
Nonparametric correlations between functional impairment, structural language, motor, and social skills.

	n	Mean	*SD*	Spearman's *rho* / 95% *CI* / *p*-value				
				1	2	3	4	5
1. DD-CGAS	20	61.1	8.4	1				
2. CCC2-SLS	20	18.6	12.4	.276 (−.204 to .648) $p = .24$	1			
3. DCDQ'07	20	48.0	11.3	.061 (−.404 to .501) $p = .80$	.238 (−.241 to .625) $p = .31$	1		
4. MABC-2 total	18	8.1	2.7	-.015 (−.490 to .467) $p = .95$	.618 (.198 to .846) $p = .006$	.211 (−.297 to .627) $p = .40$	1	
5. SRS total	20	88.3	29.3	-.369 (−.705 to .101) $p = .11$	-.691 (−.871 to −.345) $p < .001$	-.525 (−.790 to −.094) $p = .017$	-.521 (−.800 to −.057) $p = .027$	1

CCC-2 = Children's Communication Checklist Second Edition; *CI* = confidence interval; DD-CGAS = Developmental Disability-Children's Global Assessment Scale; DCDQ'07 = Developmental Coordination Disorder Questionnaire 2007; MABC-2 = Movement Assessment Battery for Children-2; *SD* = standard deviation; SRS = Social Responsiveness Scale.

functioning. The most affected functional domain was social behavior, where level of impairment was moderate (90.0%) or severe (10.0%) for all participants. School functioning was moderately or severely affected for 13 children (65.0%), and communication was slightly (30.0%) to moderately (70.0%) affected for all participants. Twelve children (60.0%) participated in ordinary physical education (PE), while three did not participate at all, and five (25.0%) participated with some level of accommodation or alternative activity. Nine children (45.0%) participated in one or two organized leisure activities. Five children (25.0%) did not have any close friends.

4. Discussion

In this study we explored the co-occurrence of motor and language impairments, as well as their potential relationship to each other, current social, and functional impairment in a clinical sample of school-aged children evaluated for suspected ASD.

4.1. Extent of motor impairments

The majority (80%) of participating children had deficits on one or both measures of motor performance, a proportion close to reports from a recent large study using the DCDQ among children with ASD and normal range cognitive abilities (Bhat, 2020). The larger proportion with motor deficits on parent report (80%) compared to standardized assessment (39%) is plausible, as these measures capture different aspects of motor ability (Wilson et al., 2009). The DCDQ'07 was designed to screen for possible motor difficulties and is more likely to over-identify than to miss such deficits (Wilson et al., 2009). Contrary to parent report based on observations over time, standardized test results represent a "snap-shot" of motor performance, which may be impacted by other non-motor factors (Licari et al., 2019). The well-structured, one-to-one assessment setting may also allow some children to perform better than when faced with the demands of everyday life.

Although recommended to confirm more definite motor impairments or a diagnosis of DCD (Wilson et al., 2009), only a few studies have combined the MABC-2 and the DCDQ'07 when investigating motor skills in ASD. Comparable to the present study, Hirata et al. (2015) found that while 47% of 19 children with ASD (7–15 years) had motor deficits on the MABC-2, all were identified with motor deficits on the DCDQ'07. Methodological differences likely have contributed to some discrepancies observed. The present sample comprised children with a broader spectrum of autistic symptoms. While mean MABC-2 total was comparable to Hirata et al. (2015), mean DCDQ'07 total was higher, indicating that parents in the present sample reported their children to have less motor difficulties. As there are no Norwegian norms for the DCDQ'07, our results should be interpreted with caution. The distribution of scores may differ between cultures. Furthermore, we applied a modified MABC-2 protocol (Liu & Breslin, 2013), which may have elicited better motor performance in our study.

4.2. Extent of language impairments and co-occurrence with motor impairments

The extent of language impairment as measured by the CCC-2 was substantial (75%), and consistent with previous results among preschool and school-aged children with ASD, ADHD, and subthreshold autistic symptoms (Helland, Biringer, Helland, & Heimann, 2012; Reindal et al., 2021). More than half the sample had difficulties with structural language (e.g., language sounds, articulation, grammar, understanding the meaning of words). While pragmatic difficulties (the appropriate use of language in social contexts) are closely related to the core social communication impairment in ASD, structural language deficits have traditionally been considered a characteristic of specific language impairment (developmental language disorder). Although being less closely related to core ASD symptoms, such deficits are also commonly reported in children with ASD and subthreshold autistic symptoms (Boucher, 2012; Reindal et al., 2021). Notably, CCC-2 is not a diagnostic tool. Rather this checklist was developed to screen for language impairment in clinical and community contexts, as well as to identify structural and pragmatic language deficits that may be difficult to elicit in a test situation (Bishop, 2011). In line with previous findings, neither motor nor language impairments were universal among children with

ASD (Boucher, 2012; West, 2019).

Nevertheless, the most striking finding was the common co-occurrence of structural language *and* motor deficits in the present sample. While motor deficits were reported in isolation for some children, structural language deficits were mostly found in children also identified with motor deficits, and in *all* children identified with *motor impairment*. These findings are consistent with recent reports from the SPARK study that the risk of motor impairment increased with increasing language impairment (Bhat, 2021).

4.3. The relationship between motor, structural language, and social skills

Our finding that better motor performance on standardized assessment was strongly associated with better structural language skills as reported by parents is consistent with the observed association between motor performance and general communication skills (teacher report) among school-aged children with ASD reported by McPhillips et al. (2014). Notably, both composites used in the present analyses (MABC-2 total and CCC-2 Structural Language Score) include various aspects of motor and structural language skills. Although a strong correlation was found, it is likely that some aspects of motor performance (e.g., fine- or gross motor skills) bear a stronger significance on structural language skills, and vice versa. While the correlation between parent reported motor skills (DCDQ'07 total) and structural language skills in the present sample did not reach statistical significance (spearman's rho .238, $p = .31$, Table 3), the effect size of this correlation is of the same magnitude as the significant relationship found between the DCDQ total and language function in the SPARK study (Bhat, 2021). Taken together, these findings highlight not only the pervasiveness of motor impairments, but also the close association with language impairments in school-aged children with a broad spectrum of autistic symptoms.

Strong associations between social impairment (SRS total) and both measures of motor skills were also found, suggesting a clear relationship between these factors. These results build on and extend previous reports among children with ASD (e.g., Bhat, 2021; Ohara, Kanejima, Kitamura, & Izawa, 2019). While reasons for the apparent relationship between motor skills and social skills currently remain unclear, a commonly suggested mechanism includes shared neural correlates between these skill domains (Ohara et al., 2019). West (2019) suggested that motor and communicative ability may also have overlapping neural correlates, which could disrupt both domains. Several postmortem and brain imaging studies have consistently identified the cerebellum as one of the most abnormal brain regions associated with ASD (see Wang, Kloth, & Badura, 2014 for a review). The cerebellum is considered to play an important role not only for motor coordination and movement control but also for higher functions such as cognition and language/communication, both of which are linked to an individual's social interactions (Ohara et al., 2019; Wang et al., 2014). Another possible mechanism has been suggested through the cascading effects of early motor deficits on other developmental domains (e.g., Leonard & Hill, 2014). Importantly, these potential mechanisms are not mutually exclusive. Even if motor and social communication skills are both affected by atypical neural development, the resulting motor deficits may further impact a developing child's social interaction and experiences, with potential down-stream effects on other developmental skills and overall functioning. Much remains to be understood about the developmental consequences of early motor deficits, as well as their role as potential intervention targets for cross-domain impact (Hudry, Chetcuti, & Hocking, 2020). Prospective longitudinal studies of at-risk infants tracking developmental skills across several domains, as well as employing randomized controlled trials to test the utility of specific motor interventions whilst also testing hypotheses about their causal role has been suggested as a way forward (Hudry et al., 2020).

4.4. Functional impairment and participation

While current guidelines generally converge on a set of well-established tools for assessing core ASD symptoms, evidence-based assessment tools addressing functional impairment are limited (Choque Olsson & Bolte, 2014; Winters, Collett, & Myers, 2005). We used the DD-CGAS, allowing us to synthesize the child's level of functioning across multiple domains (Winters et al., 2005), independent of main or co-occurring diagnoses. DD-CGAS scores mostly varied within the "upper range", as expected in a sample of verbal children without severe cognitive disabilities. Consistent with the fact that all participants were evaluated for ASD, social impairment was most affected, albeit with limitations seen across all functional domains. Klin et al. (2007) reported a similar profile using the Vineland Adaptive Behavior Scales (Sparrow, Balla, & Cichetti, 1984), a more general standardized measure of adaptive functioning which is found to correlate with the DD-CGAS (Wagner et al., 2007). Thus, although being less resource-demanding, DD-CGAS is considered to capture a related construct. Contrary to recent results from the SPARK study (Bhat, 2021), we found no significant association between motor and language skills and overall impairment, as measured by the DD-CGAS. Limited sample size and range of functioning, as well as contextual and methodic factors may have contributed to this result. Together with previous findings by Wagner et al. (2007) our results indicate that the DD-CGAS may align better with measures of core ASD symptoms, such as the ADI-R, instead of co-occurring language and motor difficulties.

Our finding that several children did not participate in organized sports/leisure activities and had no close friends are consistent with a previous study by Hilton et al. (2008) among children with ASD. In their study, physical activities showed the greatest differences, both in terms of the number of activities and the frequency of participation. The authors point to the potential importance of motor skills for participation, and of motor skills interventions for children with ASD (Hilton et al., 2008). Similar concerns have been expressed by Kopp et al. (2010). While participation was limited, our results highlight school as *the* arena where many children *do* participate. Thus, well-tailored physical education for children with NDDs may represent a potential intervention to promote both motor skills and social skills training. Tailored efforts to integrate children with social and other functional impairments in out-of-school activities may also be beneficial, acknowledging each child's individual capacities, and modifying the demands of school and daily life to a level the child can cope with.

4.5. Strengths and limitations

The present study was cross-sectional, small, and exploratory in nature. Therefore, it can only be used to illuminate potential relationships, not to make any causal inferences. Nevertheless, we consider the provision of a detailed developmental skill profile of school-aged children evaluated for suspected ASD a major strength of our study. By using validated instruments, complementary parent reports, and a standardized measure of motor skills we provide valuable information that is difficult to accomplish in larger-scale studies. Still, the relationships found here may differ in larger, more diverse samples. The relatively large number of girls may be considered a strength. Nevertheless, it is possible that the larger proportion of girls may be due to a selection bias, where individuals with co-occurring motor and language impairments were more prone to participate. Referral bias may also have influenced the reported extent of co-occurring deficits and functional impairment. However, a previous Norwegian study on children with high ASD traits found that co-occurring problems were also common in a population-based sample (Posserud et al., 2018). While some of the applied measures are validated and well-established in other countries, the Norwegian norm base is limited. Thus, we underscore the exploratory nature of our results, which should be replicated in larger samples, and compared to same-aged, typically developing children to confirm their relevance.

4.6. Clinical implications

Although preliminary, our results suggest that co-occurring difficulties beyond the core social impairment should be anticipated and planned for when evaluating children for ASD, considering more specific motor and language assessments and interventions. Where available and indicated, guided interventions from physical and/or speech-language therapists may prove useful (Fuentes et al., 2020). Motor interventions may focus on building strength, coordination, or acquisition of adaptive skills such as handwriting, safer mobility and play (Hyman et al., 2020). Acknowledging the child's difficulties, it may be wise to encourage participation in activities based on the child's area of interest or competence, and to ensure structured settings with available support to promote mastering. Whether specific interventions delivered to children presenting with early motor deficits could also mitigate downstream effects on social and language skills should be addressed in future studies (Hudry et al., 2020).

5. Conclusion

Results from the present study suggest that co-occurring motor and structural language deficits are common and closely related in school-aged children referred for evaluation of ASD. The extent of this co-occurrence, as well as the potential role and timing of specific interventions targeting motor and language skills in children with autistic symptoms should be addressed in future studies. Meanwhile, assessment should be broad to tailor interventions to the child's profile of strengths and difficulties and adjust demands to the child's level of functioning.

CRediT authorship contribution statement

Lise Reindal: Conceptualization, Methodology, Investigation, Formal analysis, Writing-Original Draft, Funding acquisition. **Terje Nærland:** Conceptualization, Methodology, Writing-Review & Editing, Supervision, Funding acquisition. **Anne Mari Sund:** Conceptualization, Methodology, Writing-Review & Editing, Supervision, Project administration. **Birgit Avseth Glimsdal:** Investigation, Writing-Review & Editing. **Ole A. Andreassen:** Conceptualization, Methodology, Writing-Review & Editing, Supervision, Funding acquisition. **Bernhard Weidle:** Conceptualization, Methodology, Writing-Review & Editing, Supervision, Project administration.

What this paper adds?

Co-occurring motor and language impairments are common and closely related in infants with autism spectrum disorder (ASD), with potential downstream effects on other developmental domains. Thus, they represent potential targets for earlier identification and intervention for subgroups of children. In school-aged children with ASD, motor and language deficits have mostly been studied separately, not considering their potential co-occurrence and additive impact on overall functioning. This study provides new information suggesting that the co-occurrence of motor and structural language deficits is common also in school-aged children evaluated for ASD, with the majority having deficits in one or both domains when assessed with a combination of parent report measures and a standardized test of motor performance. Furthermore, motor, and structural language deficits seem to be closely related, with potential impact on symptom presentation, overall functioning, and service needs. The extent of this co-occurrence, as well as the potential role and timing of specific interventions targeting motor and language skills in children with autistic symptoms should be addressed in future studies. Meanwhile, assessments should be broad and consider co-occurring motor and language impairments when evaluating children for ASD, so that interventions can be tailored to the child's profile of strengths and difficulties and demands adjusted to the child's level of functioning.

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