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A further study of relations between motor impairment and social communication, cognitive, language, functional impairments, and repetitive behavior severity in children with ASD using the SPARK study dataset

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Abstract

Motor impairments are pervasive and persistent in children with Autism Spectrum Disorder (ASD) throughout childhood and adolescence. Based on recent studies examining motor impairments in children with ASD between 5 and 15 years (i.e. SPARK study sample), 87–88% of this population is at-risk for a motor impairment, these problems persisted until 15 years, and related to their core (social communication skills and repetitive behaviors) and comorbid (language, cognitive, and functional) impairments. Persistent motor impairments extending into adolescence/adulthood could negatively impact their independent daily living skills, physical fitness/activity levels, and physical/mental health. While multiple studies have examined relations between motor dimensions and core/comorbid impairments in young children with ASD, few studies have examined such relations in school-age children/adolescents with ASD. This paper conducts a further multidimensional study of which motor domains (i.e., gross-motor including visuo-motor or multilimb coordination/planning, fine-motor or general coordination skills) best distinguish subgroups of school-age children/adolescents with ASD and help predict core and comorbid impairments after accounting for age and sex. Visuomotor, fine-motor and certain general coordination skills were better at explaining variations in / predicting social communication impairments whereas fine-motor skills were slightly better at explaining variations in / predicting repetitive behavior severity. Multi-limb coordination/planning and fine-motor skills explained variations in /predicted cognitive delays whereas visuomotor and fine-motor skills explained variations in and better predicted language delays. All 3 motor dimensions explained variations in / predicted functional delays. This study provides further evidence for inclusion of motor impairments within the ASD definition (criteria or specifiers).

Conflict of Interest Disclosure

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Lay Summary

Gross-motor skills were related to social communication and functional delays of children with ASD (visuomotor skills related to language delays and multilimb coordination/planning skills related to cognitive delays). Fine-motor skills were related to repetitive behavior severity, language, cognitive, and functional delays in ASD. Diagnosticians should recommend systematic motor screening, further evaluations, and treatments for children at-risk for and diagnosed with ASD. Motor advocacy and enhanced public/clinical community awareness is needed to fulfill the unmet motor needs of children with ASD.

Introduction

With 1 in 44 children diagnosed with Autism Spectrum Disorder (ASD), it is one of the most common pediatric developmental disorders (Maenner et al., 2021). There is an enormous financial and psychological burden on families caring for children with ASD with average annual medical, non-medical, and productivity costs in the US ranging from ~268 to 461 billion (Leigh & Du, 2015). Parents of children with ASD struggle to receive an early diagnosis and comprehensive assessments for their child in order to gain access to various evidence-based early interventions for the multiple core and comorbid problems that occur in the early years (Shaw et al., 2020; Landa, 2008). Together, these issues make it urgent for clinical researchers to recognize and bring to forefront issues of under-diagnosis and under-treatment so that healthcare can be improved to provide appropriate assessments and interventions early on in life with the hope of improved future outcomes.

ASD is traditionally considered a social communication disorder; but recently has been recognized by many as a complex, multisystem disorder with multiple core and comorbid impairments (Elsabbagh & Johnson, 2016; Srinivasan & Bhat, 2013). Children with ASD present with social communication impairments and repetitive behaviors that together form the defining criteria for ASD (American Psychiatric Association, 2013). Apart from core impairments, children with ASD also present with cognitive and language impairments that are captured within the ASD definition through the use of "specifiers", for example, a child may receive a diagnosis of "ASD with a language/intellectual impairment" (American Psychiatric Association, 2013). Sensory processing issues were more recently included within ASD criteria under the repetitive behaviors category. However, motor issues in ASD are still not recognized within the ASD defining criteria or specifiers. In this study, we examine variations in, predictive validity of, and associations between various motor dimensions (e.g., visuomotor, balance/postural control, complex motor coordination, etc.) and the core/comorbid impairments across the spectrum of children and adolescents with ASD. These findings will shed light on whether motor issues should be generally recognized as part of ASD specifiers or if certain motor issues should be part of the core impairments of ASD. It would also help to hone in on the types of motor skills that should be evaluated and treated when providing services to children and adolescents with ASD.

Recent findings from population-based studies such as the SPARK study in the United States and the West Australian Autism Register study in Australia confirm motor impairments in 79–88% of children with ASD (Licari et al., 2019; Bhat, 2020). These numbers align

with smaller studies that also report similar high proportions of motor impairment in their ASD samples (Green et al., 2009; Miller et al., 2021). Moreover, the first author's recent publications based on the SPARK cohort confirm that motor impairments in children with ASD persist between 5 and 15 years and increase with increasing severity of core impairments in social communication skills and repetitive behavior severity as well as comorbid, cognitive, language, and functional impairments (N=13,887) (Bhat, 2020, 2021). Persistent motor impairments extending into adolescence and adulthood not only negatively impact the ASD population's independent daily living skills but also their physical fitness/activity levels and overall physical/mental health and well-being (Srinivasan, Pescatello, & Bhat, 2014, Bremer & Cairney, 2020, Ketcheson, Hauk, & Ulrich, 2017). The present study is a further extension of our earlier work to highlight how different motor domains/dimensions: gross-motor (i.e., visuo-motor or multi-limb coordination/planning), fine-motor, and general coordination skills (i.e., sport practice, motor speed, competence or clumsiness, and fatigability) help distinguish subgroups of children with ASD based on aforementioned core and comorbid impairments. Additionally, how are the different types of motor impairment associated with and predictive of other system impairments in school-age children with ASD?

There is copious literature on the range of motor impairments reported in children and adolescents with ASD that distinguish them from children with other / no diagnoses as reviewed earlier in Bhat, 2020 and 2021. Motor impairments in ASD include gross-motor problems in static/dynamic balance, motor coordination (i.e., visuomotor and multilimb/ bilateral coordination), dyspraxia/motor planning problems, as well as problems with fine-motor problems in precision/integration and hand dexterity, etc. (Dewey, Cantell, & Crawford, 2007; Bhat, Galloway, & Landa, 2011, Shield et al., 2017; Bhat et al., 2018; Kaur et al., 2013, 2018; Jansiewicz et al., 2006; Fleury, Kushki, Tanel, Anagnostou & Chau, 2013; Ament et al., 2014; Kushki, Chau, Anagnostou, 2011; Fournier, Hass, Naik, Lodha, & Cauraugh, 2010). Definitions for various motor terms are provided in the supplementary materials. Moreover, there is mounting evidence for how motor impairments in infants at-risk for and children with ASD are associated with / predictive of core and comorbid impairments in ASD including social communication, receptive/expressive language, cognitive, and functional impairments as well as repetitive behavior severity (Macdonald, Lord, & Ulrich, 2013a, 2013b, 2014; Bhat et al., 2018; Srinivasan & Bhat, 2016; Shield et al., 2017; Kaur, Srinivasan & Bhat, 2015, 2018; LeBarton & Landa, 2019; Licari et al., 2019; Bhat, 2021; Radonovich, Fournier, & Hass, 2013). Both, fine- and gross-motor skills of children with ASD between 14–33 months (based on the Mullen Scales of Early Learning) predicted their concurrent autism severity using the diagnostic tool, Autism Diagnostic Observation Schedule (ADOS) (Macdonald et al., 2013a). While motor impairments in ASD are present across the ASD spectrum the prevalence and severity of motor impairment is greater in children with greater cognitive impairments (Green et al., 2009; Kaur et al., 2018; Licari et al., 2019; Bhat, 2021; Ketcheson, Hauk, & Ulrich, 2021). Additionally, greater motor impairment in children with ASD is linked to lower functional independence (Macdonald et al., 2013b; Licari et al., 2019; Bhat, 2021) as well as higher rates of repetitive behaviors (Radonovich et al., 2013; Bhat, 2021). When comparing fine and gross-motor performance in young children with ASD between 13 and 33 months, the

fine-motor subtest of the Mullen scales predicted concurrent overall adaptive functioning, social, and communication functioning, and daily living skills performance whereas grossmotor skills predicted concurrent *daily living* skills performance only on the Vineland Adaptive Behavioral Scales (VABS) measure (Macdonald et al., 2013b). In contrast, in even younger infants at-risk for ASD between 7 to 36 months, early gross-motor but not fine-motor trajectories predicted future expressive language development (Leonard, Bedford, Pickles, & Hill, 2015). Overall, motor impairments generally differentiate children with ASD from children with no/other diagnoses and relate to core and comorbid impairments of ASD. However, majority of the aforementioned studies have been conducted in young children at-risk for ASD or those who were newly diagnosed with ASD. Few studies have reported relationships between fine- and gross-motor impairments and core / comorbid impairments in school-age children and adolescents with ASD; which will be the focus of the present study.

While many studies have reported associations between motor and other system impairments in children with ASD, even fewer studies have conducted comprehensive, multidimensional motor assessments to relate a variety of motor domains to the core and comorbid impairments of young and older children with ASD. Children with ASD have consistently shown associations between *imitation/praxis performance and autism severity* (based on ADOS scores) even after controlling for basic motor skill (Dewey et al., 2007; Dziuk et al., 2007; Dowell, Mahone, & Mostofsky, 2009; Stieglitz Ham et al., 2011; Macneil & Mostofsky, 2012; Kaur et al., 2018). Children with ASD also showed specific impairments in catching and balance skills that increased the risk of an ASD outcome indicating that visuomotor skills were particularly affected in children with ASD compared to those with Attention Deficit Hyperactivity Disorder (ADHD, Ament et al., 2014). Similarly, children with ASD had greater manual dexterity problems (requires hand-eye coordination) compared to children with Specific Language Impairment (SLI, McPhillips et al., 2014). The most consistent finding so far has been the relationship between early fine motor performance and concurrent and future language development and the ability to predict future language delays (Bhat et al., 2012; Bedford et al., 2015; Choi, Leech, Tager-Flusberg, & Nelson, 2019; Bal et al., 2020). Bedford et al. (2015) found that gross-motor development at 2 years of age was predictive of receptive and expressive language development in children with ASD between 2 and 9 years of age. Choi et al. (2019) and Bal et al. (2020) found that fine motor skills of children with ASD during early infancy and childhood (6 months to 2 years) were predictive of their future *expressive language* skills at 3 or 19 years of age even after controlling for visual reception skills. Using the Peabody Developmental Motor Scale (PDMS), a multidimensional motor tool, LeBarton & Landa (2019) found that postural control and reach-grasp performance (visuomotor skills) at 6 months predicted expressive language in toddlers at 30–36 months and ASD outcomes at 24–36 months. Using the PDMS measure, Wu et al. found that early motor performance between 24–42 months (including stationary, locomotion, object manipulation, grasping, and visuomotor integration) was predictive of *concurrent receptive and expressive language delay* in newly diagnosed children with ASD (Wu, Tsao, Huang, Yang, & Li, 2021). Overall, evidence suggests that fine-motor skills are related to and predictive of language delays and ASD outcomes and gross-motor skills, specifically visuomotor, fine-motor, and balance/postural

control skills maybe related to the core impairments of children with ASD. However, the aforementioned studies involved small, biased samples with limited ranges of functioning, cognitive, or verbal capacities. Additionally, many studies examining relations between multiple motor dimensions and core/comorbid impairments involve young children at-risk for and newly diagnosed with ASD. Hence, the present study will extend the past work to study such associations/predictive relations between various motor dimensions and core/ comorbid impairments in a broad and variable spectrum of school-age children / adolescents with ASD.

The SPARK study is a large population-based study engaging a large group of parents of school-age children and adolescents with ASD within the US using the Developmental Coordination Disorder Questionnaire (DCD-Q, Schoemaker et al., 2006), a motor screener, the Social Communication Questionnaire - Lifetime (SCQ, Berument et al., 1999), a social communication delay screener, and the Repetitive Behaviors Scale (RBS-R, Lam & Aman, 2006), a measure of various repetitive behaviors and restricted interests. Additionally, parents provided information on the child's current language, functional, and cognitive abilities compared to same-age peers. This rich dataset includes basic demographic information, birth history, and diagnostic information as well. The present study examined the SPARK study dataset, version 3 to analyze the DCD-Q motor measure and its subscales and items. The standard domain / subscale scores of DCD-Q (control during movement/gross-motor, fine-motor, and general coordination) were compared across multiple subgroups of children with ASD and effect sizes were calculated for differences between subgroups based on SCQ, RBS-R, and parent-reported current abilities (cognitive, language, and functional delay) data. In terms of hypothesis, DCD-Q total scores (i.e., risk for motor impairment) and subscale scores (fine-motor, gross-motor, and general coordination scores) will be lower in children with ASD with higher SCQ and RBS-R scores as well as greater language, cognitive, and functional impairments. Internal consistency of the DCD-Q items will be high and will confirm its validity in screening motor problems of school-age children with ASD using the DCD-Q. New DCDQ subdomains will be obtained through factor analysis (FA). These new subdomains will also be examined for differences across various subgroups. Gross-motor performance (i.e., visuo-motor coordination) will relate more/be more predictive of core social impairments of ASD and fine-motor performance will relate more/be predictive of repetitive behaviors as well as general cognitive and language delays.

Methods

SPARK Study Procedures and Data Access

The SPARK research team recruited families with one or more children with ASD through 21 clinical sites across the US using a diverse social media strategy (Feliciano et al., 2018). Families who volunteered for the study completed several online questionnaires on the SPARK website [\(https://sparkforautism.org/registration/account_information/\)](https://sparkforautism.org/registration/account_information/). They also received information on local research studies they could participate in. The first author's lab signed up with the SPARK study to utilize their participant match resource after receiving approval for ongoing studies from the University of Delaware (UD)'s Human Subjects

Review Board. UD also signed an authorization agreement with the Simons Foundation; after which the first author was given access to version 3 of the SPARK study database.

SPARK Forms and Measures

The SPARK database comprises of multiple parent questionnaires such as the basic medical screening form, individual data form, and background history form. The basic medical screening form includes demographic information, birth history, professional diagnosis of ASD and other disorders, as well as other general medical conditions. The individual data form provides details about the child's ASD diagnosis, whether a professional provided the diagnoses, whether there is a cognitive impairment, whether there is an Individualized Education Plan (IEP) for the child, and whether the child receives ASD services. The background history form lists the various intervention services received by the child as well as data on cognitive, language, and functional age level of each participant (i.e., above, at, slightly below, or significantly below same-age peers). Table 1 summarizes the type of SPARK study data used for our analysis. Apart from participant information, data from the following 3 parent questionnaires were analyzed: a) the Developmental Coordination Disorder Questionnaire (DCD-Q, Schoemaker et al., 2006), b) the Social Communication Questionnaire – Lifetime (SCQ, Berument et al., 1999), and c) the Repetitive Behaviors Scale – Revised (RBS-R, Lam & Aman, 2006).

Inclusion/Exclusion Criteria

Out of the total 150,064 individuals in the SPARK database, which include children with ASD and their family members, parents of 16,705 children with ASD completed the DCDQ form; hence that was our base sample. The final sample included 13,887 children with ASD was obtained after applying various filters already reported in the earlier companion paper, Bhat, 2021. Compared to the sample analyzed in Bhat, 2020, the current study sample includes participants with cognitive /intellectual impairment because one of our questions focuses on how multiple motor dimensions differ in children with ASD as a function of cognitive abilities.

DCD-Q

The DCD-Q is a 15-item parent questionnaire used to screen for gross- and finemotor performance during everyday functions/play within the child's natural environment (Schoemaker et al., 2006). The questionnaire includes various motor skills such as ball skills (e.g., hitting or catching a ball), complex body coordination skills (e.g., jumping, running, etc.), fine motor skills (e.g., writing, cutting, etc.), and general motor abilities (e.g., quickness, clumsiness, fatigability, etc.). These skills are categorized into three subscales: control during movement, fine motor coordination, and general coordination. The total final score is the sum of the individual subscale scores. Definite motor impairment or suspect DCD (<10th percentile) is determined based on the final score cutoffs which differ for different age groups. For example, these cutoffs include a score < 47 for children between 5 years to < 8 years, a score below 56 for children between 8 years to < 10 years, and a score < 58 for children between 10–15 years. Based on these criteria, an assignment of risk for DCD (1 = Yes, $0 = No$) is provided for each participant. A DCD diagnosis is typically confirmed with a follow-up, standardized motor assessment, and clinical judgment

of a trained movement clinician. Note that the positive predictive value of the DCD-Q with a clinical motor assessment such as the Movement ABC is 80–92% (i.e., 80–92% of the children who are at-risk for a motor impairment using the DCD-Q are likely to perform poorly on the M-ABC motor measure) (Green et al., 2009; van Damme, Vancampfort, Thoen, Sanchez, Biesen, 2021). The risk for DCD/motor impairment in this sample has already been reported in previous papers - Bhat 2020, 2021. This paper focuses on the various DCD-Q subdomain scores and the effect sizes for differences between subgroups based on various core and comorbid impairments as well as associations between DCD-Q

subdomain scores and other impairments.

SCQ

The SCQ is a common 40-item parent questionnaire (Yes/No format) to screen for autistic traits in children above 4 years of age with a mental age of at least 2 years (Berument et al., 1999). It is based on the popular diagnostic interview, the Autism Diagnostic Interview-Revised (ADI-R). The SCQ has two versions – Lifetime (used to support a diagnosis) and - Current (used to support an evaluation of current difficulties). The Lifetime version provides a total SCQ score. If the total score is >12, it indicates a social communication delay and higher likelihood to be on the autism spectrum. The cut-off of 12 is a research recommended, sensitive cut-off and was implemented in this study (Lee et al., 2010; Daniels et al., 2012; Zwaigenbaum et al., 2015; Marvin et al., 2017). The total SCQ score was also used as a measure of social communication impairment.

RBS-R

The RBS-R is a 43-item parent report measure to characterize the repetitive behaviors of children with ASDs. It has high internal consistency and medium reliability (Lam & Aman, 2006). Each item/question is scored on a 4-point scale: 0 (no such behavior), 1 (mild problem), 2 (moderate problem), and 3 (severe problem), therefore the total score ranges between 0 and 129 with higher score indicating more repetitive behavior. It has six subscales on the child's stereotyped (I), self-injurious (II), compulsive behaviors (III), ritualistic (IV), sameness behaviors (V), and restricted interests (VI). The total RBS score was use as a measure of repetitive behavior severity.

Subgrouping Analysis

As reported in Bhat, 2021, the sample was divided into 5 subgroups/categories (C) based on scoring ranges defined for SCQ scores, RBS-R scores, and DCD-Q scores using each measure's sample mean and standard deviations (see grouping details in Bhat, 2021 and Table S1 in supplementary materials). Subgrouping categories included very low (C1), low (C2), high (C3), very high (C4), and extremely high (C5) for both social communication impairments (SCI) or repetitive behavior (RB) severity. Missing data for scores was only 0.6% for the RBS-R scores, but no data was missing for the DCD-Q and SCQ scores because presence of valid DCD-Q and SCQ scores was a key inclusion criterion.

Data extracted from parent reported levels of cognitive, functional, and language delays are presented in Table S1. As reported in Bhat, 2021, the parent-reported outcome data were divided into 3 subgroups based on the reported level of cognitive, functional, and language

delay compared to peers (i.e., at or above (C1), slightly below (C2), or significantly below (C3) peers). ~5% cognitive, ~3% functional, and ~3% language delay information was missing for the present sample.

Demographic Information

Key demographic information for this sample is a presented in Table 1. The sample had \sim 80% males and \sim 20% females. In terms of race, \sim 76% were White, \sim 10% were multiracial, and ~5% were African American. In terms of ethnicity, ~16% were Hispanic and ~81% were non-Hispanic. The sample was generally evenly distributed in terms of annual household income. Note that certain items were not reported, hence those participants were labeled as having missing data (up to ~5% of the sample). As reported in Bhat, 2021 and also shown in Table S1, 88.2% of children had a risk for motor impairment based on their DCD-Q performance. Many children held formal comorbid diagnoses – specifically, \sim 41% had ADHD, ~17% had motor delay or DCD, ~22% had learning disability, ~15% had cognitive impairment, and ~44% had language disorders. In terms of services received, ~87% received ASD services, ~84% had an IEP for ASD, ~64% received behavioral or developmental services, $\sim 83\%$ received speech and language therapy services, $\sim 81\%$ received OT services, ~46% received social skills training, only ~34% received PT services, and only ~14% received recreational therapies.

Statistical Analysis

Statistical analyses were conducted using JMP Pro 15.0 (JMP, Inc). The original factor analysis (FA) of the DCD-Q measure by Rivard et al. (2012) provided 3 standard subdomains (CDM – Control During Movement, FM – Fine Motor, and GC – General Coordination). However, to identify unique patterns of motor impairment in children with ASD, an FA of the SPARK ASD sample was conducted using the maximum likelihood estimation and Varimax orthogonal rotation method. This method determines where each DCD-Q question/item most likely belongs in terms of a factor/subdomain. Additional DCD-Q subdomains were identified compared to the subdomains determined from the original FA (Rivard et al., 2012).

Internal consistency (reliability) or degree of homogeneity among the 15 DCD-Q questions/ items was also evaluated by calculating Cronbach's alpha for the whole sample. The criterion for sufficient homogeneity among the items was set to 0.7 (Bland and Altman, 1997). If deletion of an item results in significantly higher alpha, that item is problematic for the measure and perhaps could be removed to consolidate the tool. On the other hand, if deletion of an item results in significantly lower alpha, that item is very important for the measure, and hence must be preserved.

Spearman rank correlations were calculated between the DCD-Q subdomains and subgroup assignments based on SCQ, RBS-R scores (1 to 5) or language, cognitive, and functional delay levels (1 to 3). Ordinal logistic regression analyses were used to predict subgroup assignment (i.e., subgroup 1 to 5 based on SCI and RB severity or subgroup 1 to 3 based on language, cognitive, and functional delays). For each regression model, age and sex effects were also included and accounted for. Four different models were analyzed for each type of

subgroup assignment based on SCI, RB severity and the 3 delay types (20 models in total) using total scores, original subdomain scores, FA-based subdomain scores, and item-level DCD-Q scores as predictors. The Wald's chi-squared test values are reported for predictors that significantly contributed to a given model.

One-way Analysis of Variances (ANOVAs) comparing DCD-Q scores across subdomains (standard and FA-based) between categories of SCI and RB severity as well as language, cognitive, and functional delays were conducted. Statistically significant differences are being reported based on significant Tukey post-hoc testing along with confidence intervals that do not include a zero value.

For all analyses, statistical significance was modified based on p-value thresholds set after Bonferroni corrections. The magnitude of group differences between the extreme categories (C1 vs. C5 or C1 vs. C3) based on SCI, RB, Cognitive delay, Functional delay, or Language delay was calculated using the Cohen's d effect size estimate (small effect: <0.5, medium: >0.5 but <0.8, large: ≥0.8). This helped in understanding which DCD-Q items / standard subdomains / FA-based subdomains / most explained the variation in the SPARK ASD sample based on core (SCI / RB variations) or general/comorbid (cognitive, language, functional) impairments.

Results

Factor Analysis

Based on factor analyses of the DCD-Q conducted in the general population (Rivard et al., 2012), DCD-Q items were grouped into three standard subdomains – Control During Movement (CDM – Items/Questions Q01 to Q06), Fine Motor (FM – Q07 to Q10), and General Coordination (GC – Q11 to Q15). In the present study, FA of the SPARK ASD sample was carried out with 3 to 7 factors. A 5-factor solution was chosen because it explained an optimal amount of variance (58%, Table 2). With 3 or 4 factors, more items belonged to each of the factors, but the explained variance was lower $(-52\% \text{ and } -55\%$, respectively). With 6 or 7 factors, only slightly more variance was explained as compared to 5 factors (59.5% and 60%, respectively), but none of the DCD-Q items belonged to these extra factors. Therefore, the 5-factor solution was considered ideal.

Items Q01 to Q03 (Throw, Catches, Hits) loaded cleanly on Factor 2, which was termed Control During Movement 1 (CDM1) or visuomotor items. Items Q04 to Q06 (Jumps, Runs, Plans) loaded cleanly on Factor 3, which was termed Control During Movement 2 (CDM2) or multilimb coordination/planning items. Items Q07 to Q10 (Writes fast, Writes legibly, Effort pressure, Cuts) loaded cleanly on Factor 1, which were the Fine Motor (FM) items in the original DCD-Q as well. Items Q11 to Q12 (Likes sports, Learning new) loaded cleanly on Factor 5, which was termed General Coordination 1 (GC1). Items Q14 to Q15 (Bull in a china shop, Fatigues easily) loaded cleanly on Factor 4, which was termed General Coordination 2 (GC2). Item Q13 (Quick competent) showed factorial complexity, with about equal factor loadings on Factors 1 (FM), 4 (GC1), and 5 (GC2); hence for simplicity, it was assigned to GC1. Overall, Q01-Q03 / visuomotor items and Q04-Q06 / multilimb coordination/planning items belonged to two *distinct* subdomains, indicating that they might

be able to capture distinct information in the SPARK ASD sample. Correlations among items are shown in Table S2 and support this observation, where Q01-Q03 correlate more with each other compared to Q04-Q06, and vice versa.

Visuomotor, multilimb coordination/planning, fine-motor, and general coordination scores worsen with and explain variations in core impairments (SCI and RB severity)

Bhat (2021) reported the variation in DCD-Q total score and standard subdomain scores (CDM, FM, and GC) as a function of core impairments (SCI and RB severity, shown in Table S3–S6 of supplementary materials). Similar analysis is now extended to the FA-based subdomains (CDM1, CDM2, FM, GC1, and GC2). Note that the standard FM subdomain is identical to the FA-based FM subdomain. As shown in Figure 1, the new FA-based subdomain DCD-Q scores (CDM1, CDM2, GC1, and GC2) worsened across subgroups with increasing SCI and RB severity (p-values < 0.001 , Tables S7–S10). Certain subgroup comparisons were an exception to these findings (1 CDM1-SCI, 1 CDM1-RB severity, 1 CDM2-SCI, 1 CDM2-RB severity, 1 GC1-SCI, 2 for GC1-RB severity, 2 for GC2-SCI, and 1 for GC2-RB severity, see details in the footnotes of Tables S7–S10).

Bhat (2021) had also reported DCD-Q vs. impairment category correlations for DCD-Q total scores only. Additional correlations between SCQ / RBS-R scores and DCD-Q subdomain scores (standard and FA-based) as well as DCD-Q item-level correlations highlight which subdomains / items best correlate with core impairments in ASD (Table 3). The standard subdomain CDM and the FA-based subdomains CDM1/visuomotor and CDM2/multilimb coordination/planning moderately correlated with SCI ($r = -0.24$ to -0.28 , ps < 0.0001); however, they correlated less with RB severity ($r = -0.17$ to -0.19 , ps < 0.0001). Instead, the standard FM and GC subdomains moderately correlated with RB severity ($r = -0.22$ to -0.23 , ps < 0.0001).

Ordinal logistic regression analyses were used to predict subgroup assignment based on SCI and RB severity using age, sex, and DCD-Q total, original subdomain, FA-based subdomain, and item-level scores as predictors. Wald chi-squared test shows the contributions of significant predictors in Table 4. For the SCI categories model using standard subdomain score predictors, in the order of most to least importance, child's age, CDM, FM, and GC DCD-Q scores were significant contributors to the model. For the SCI categories model using FA-based subdomain score predictors, in the order of most to least importance, child's age, FM, CDM1, CDM2, and GC1 DCD-Q scores were significant contributors to the model. For the SCI categories model using item score predictors, child's age as well as 9 out of 15 items including Q06/plans, Q03/hits ball, and Q09/writing effort were significant contributors to the model. For the RB categories model, using standard subdomain score predictors, in the order of most to least importance, FM, GC, child's age, and CDM DCD-Q scores were significant contributors to the model. For the RB categories model using FA-based subdomain score predictors, in the order of most to least importance, FM, GC2, child's age, GC1, and CDM2 DCD-Q scores were significant contributors to the model. For the RB categories model using item score predictors, child's age, and 10 out of 15 items including Q09/writing effort, Q13/quick competent, and Q14/bull in china shop were significant contributors to the model.Effect sizes for differences across categories based on

SCI and RB severity are reported in Table 5 (C1 vs. C5), Table S11 (for all category pairs), and are also plotted in Figures 2 and 3 for visual comparisons. When examining effect sizes for DCD-Q total scores for extreme categories (C1 vs. C5) based on SCI and RB severity, the differences had large effects. Additionally, effect sizes for the standard subdomain scores across extreme categories were large for the following pairs: (CDM – SCI, GC – SCI, CDM – RB, FM – RB, GC – RB) and moderate for FM – SCI along with no small effects. When examining effect sizes for the FA-based subdomain scores across extreme categories, large effects were seen for the following pairs: (CDM1 – SCI, CDM2 – SCI, GC1 – SCI, FM – RB); moderate effects seen for the following pairs: (FM – SCI, GC2 – SCI, CDM1 – RB, CDM2 – RB, GC1 – RB, GC2 - RB), and no small effects were seen.

Similar effect size analyses were also conducted for the 15 individual items as shown in Table 5 (C1 vs. C5), Table S11 (for all category pairs) and are also plotted in Figures S1 and S2 for visual comparisons. When examining effect sizes for individual items across extreme categories, large effects were seen for the following pairs: Q01 (throws), Q04 (jumps), Q06 (plans), and Q11 (likes sports) – SCI, Q09 (writing effort) and Q10 (cuts) – RB. Small effects were seen for Q08 (write legibly) and Q15 (fatigues easily) – SCI and Q11 (likes sports) – RB. Moderate effects were seen for all the remaining item – SCI/RB impairment pairs. Overall, 3 gross-motor items (Q01/throw, Q04/jumps, Q06/plans) and 1 general coordination (GC/GC1) item (Q11/likes sports) explained SCI variations with large effects whereas two fine-motor items explained RB variations with large effects (Q09/ writing effort, Q10/cuts). Together, these findings revealed that gross-motor, followed by fine-motor, and certain general coordination skills were associated with, predictive of and better explained SCI variations and fine-motor and certain general coordination skills were slightly more associated with, predictive of, and better explained variations in RB severity.

Visuomotor, multilimb coordination/planning, fine-motor, and general coordination scores worsen with and explain variations in general/comorbid impairments (cognitive, language, and functional delays)

Bhat (2021) reported the variation in DCD-Q total score and standard subdomain scores (CDM, FM, and GC) as a function of comorbid impairments (cognitive, language, and functional delays, Tables S3–S6). Similar analysis is now extended to the FA-based subdomains (CDM1, CDM2, FM, GC1, and GC2). Note that the standard FM subdomain is identical to the FA-based FM subdomain. As shown in Figure 1, the new FA-based subdomain DCD-Q scores (CDM1, CDM2, GC1, and GC2) worsened with increasing cognitive, language and functional delay (p-values < 0.0021, Tables S7–S10). There were 4 exceptions to this trend (GC1-language delay, 1 for GC2-cognitive delay, and 2 for GC2-language delay, see details in the footnotes of Tables S9–S10).

Bhat (2021) reported DCD-Q vs. impairment category correlations for DCD-Q total scores only. Additional correlations between cognitive, language and functional delay categories and DCD-Q subdomain scores (standard and FA-based) and DCD-Q items were conducted to highlight which subdomains / items best correlate with general/comorbid impairments (Table 3). The standard subdomain CDM and the FA-based subdomains CDM1/visuomotor and CDM2/multilimb coordination/planning moderately correlated with cognitive delay $(r =$

 -0.22 to -0.29 , $ps < 0.0001$) and functional delay ($r = -0.32$ to -0.36 , $ps < 0.0001$). CDM and CDM1 moderately correlated with language delay ($r = -0.22$ to -0.24 , $p < 0.0001$); however, CDM2 correlated less with language delay ($r = -0.16$, $p < 0.0001$). The FM subdomain is moderately associated with all 3 general/comorbid impairments ($r = -0.34$ to −0.40, ps < 0.0001). The standard GC subdomain only moderately correlated with functional delay ($r = -0.33$, $p < 0.0001$) and less with cognitive or language delays ($r = -0.05$ to -0.17 , p_s < 0.0001). GC1 subdomain moderately correlated with functional delay (r = −0.33, p < 0.0001) and cognitive delay ($r = -0.22$, $p < 0.0001$) but less with language delay ($r = -0.12$, $p < 0.0001$). GC2 items correlated less with the general/comorbid impairments ($r = 0.03$ to −0.16, cognitive $p < 0.0001$, language $p = 0.0023$, functional $p < 0.0001$).

Ordinal logistic regression analyses were used to predict subgroup assignment based on cognitive, language, and functional delays using age, sex, and DCD-Q total, original subdomain, FA-based subdomain, and item-level scores as predictors. Wald chi-squared test shows the contributions of significant predictors in Table 4. For the cognitive delay model using standard subdomain score predictors, in the order of most to least importance, FM, CDM, age, sex and GC DCD-Q scores were significant contributors to the model. For the cognitive delay model using FA-based subdomain score predictors, in the order of most to least importance, FM, CDM2, age, sex, and GC2, and GC1 DCD-Q scores were significant contributors to the model. For the cognitive delay model using item score predictors, child's age and sex as well as 10 out of 15 items including Q06/plans, Q07/writes fast, and Q10/cuts were significant contributors to the model. For the language delay model using standard subdomain score predictors, in the order of most to least importance, FM, age, GC, and CDM DCD-Q scores were significant contributors to the model. For the language delay model using FA-based subdomain score predictors, in the order of most to least importance, FM, age, GC2, CDM1, GC1, and lastly CDM2 DCD-Q scores were significant contributors to the model. For the language delay model using item score predictors, child's age as well as 12 out of 15 items including Q06/plans, Q07/writes fast, and Q10/cuts were significant contributors to the model. For the functional delay model using standard subdomain score predictors, in the order of most to least importance FM, CDM, and GC DCD-Q scores were significant contributors to the model. Notably, age was not a contributor to this model. For the functional delay model using FA-based subdomain score predictors, in the order of most to least importance, child's age, FM, GC1, CDM2, CDM1, and GC2 DCD-Q scores were significant contributors to the model. For the functional delay model using item score predictors, child's age, sex as well as 9 out of 15 items including Q13/quick and competent, Q07/writes fast, and Q10/cuts were significant contributors to the model.Effect sizes for differences across extreme categories based on cognitive, language, and functional delays are reported in Table 5 (C1 vs. C3), Table S11 (all category pairs) and are also plotted in Figures 2 and 3 for visual comparisons. Effects sizes for DCD-Q total scores across extreme categories were large for cognitive and functional delays, but moderate for language delay. Effect sizes for the standard subdomain scores across extreme categories were large for the following pairs: (FM – Cognitive, FM – Language, CDM – Functional, FM – Functional, and GC - Functional); moderate for the following pairs: (CDM – Cognitive and CDM – Language), and small for the following pairs: (GC – Cognitive and GC – Language). When examining effect sizes for FA-based subdomains across extreme categories, large

effects were observed for the following pairs: (FM – Cognitive, FM – Language, CDM1 – Functional, CDM2 – Functional, FM – Functional, and GC1 - Functional); moderate effects for the following pairs: (CDM1 – Cognitive, CDM2 – Cognitive, GC1 – Cognitive, and CDM1 – Language), and small effects were seen for the following pairs: (GC2 – Cognitive, CDM2 – Language, GC1 – Language, GC2 – Language, and GC2 – Functional).

For individual DCD-Q items, large effects were observed for the following pairs: (Q06, Q07, Q08, Q10 – Cognitive, Q10 – Language, Q03, Q06, Q07, Q08, Q09, Q10, Q12, Q13 – Functional), moderate effects for the following pairs: (Q03, Q04, Q09 – Cognitive, Q03, Q06, Q07, Q08, Q09 – Language, Q01, Q02, Q04, Q5, Q11 – Functional), and small effects were seen for the following pairs: (Q01, Q02, Q05, Q11, Q12, Q13, Q14, Q15 – Cognitive, Q01, Q02, Q04, Q05, Q11, Q12, Q13, Q14, Q15 – Language, Q14, Q15 – Functional). Overall, fine-motor skills (Q07-Q10/related to writing/cutting) were best associated with, predictive of, and better explained variations in cognitive and language delays. Additionally, multilimb coordination/planning skills (Q03/hits ball, Q04/jumps, and Q06/plans) were also better associated with, predictive of, and better explained variations in cognitive delays. All three types of motor skills (CDM, FM, and GC) were associated with, predictive of, and better explained variations in functional delays (except Q14 and Q15). Finally, GC/GC2 items such as Q14 (Bull in china shop) and Q15 (Fatigues easily) did not correlate well with the 3 comorbid impairments.

Internal Consistency of the DCD-Q

For the entire DCD-Q items set, Cronbach's alpha coefficient was 0.894 (significantly higher than the criterion of 0.7). Deletion of items Q14 or Q15 resulted in slightly higher alpha (0.895 and 0.897, respectively). Deletion of any of the other items (Q01 to Q13) resulted in slightly lower alpha (0.884 to 0.888). This indicates that Q14 and Q15 are slightly less important compared to the other items, but none of the items are problematic for the measure, and removal of no one item consolidated the total DCD-Q score.

For the standard sub-domains, Cronbach's alpha coefficients were as follows: 0.858 for CDM, 0.879 for FM and 0.692 for GC. Deletion of items from CDM resulted in minor changes in alpha (range = 0.823 to 0.850 , most important item = Q01 throws ball), indicating that the CDM items are fairly homogeneous. Deletion of items from FM resulted in minor changes in alpha values (range $= 0.827$ to 0.861, most important item $= Q08$ prints letters), indicating that the FM items are fairly homogeneous. Deletion of items from GC resulted in moderate changes in alpha (range $= 0.599$ to 0.673, most important item $= Q12$ learns new skills).

For the FA-based sub-domains, Cronbach's alpha coefficients were as follows: 0.863 for CDM1, 0.789 for CDM2, 0.879 for FM, 0.691 for GC1, and 0.522 for GC2. The very low value for GC2 indicates that its items (Q14 to Q15) are not homogeneous. Deletion of items from CDM1 resulted in moderate changes in alpha (range = 0.763 to 0.830, most important item = Q02 catches ball). Deletion of items from CDM2 resulted in large changes in alpha (range $= 0.640$ to 0.800, most important item $= Q04$ jumps over obstacles). Deletion of items from FM resulted in minor changes in alpha values (range = 0.827 to 0.861, most important item = Q08 prints letters), indicating that FM items are fairly homogeneous. Deletion of

items from GC1 resulted in large changes in alpha (range = 0.485 to 0.681, most important item = Q12 learns new skills). Finally, deletion of items from GC2 could not be tested as there are only two questions.

Discussion

The present study is a further analysis of the Developmental Coordination Disorder Questionnaire (DCD-Q) measure obtained from one of the largest ASD cohorts in the US, the SPARK study sample of children with ASD between 5 and 15 years. The earlier companion paper, Bhat (2021), examined the risk of motor impairment and changes in DCD-Q total scores across subcategories of children with ASD with increasing levels of core and comorbid impairments (social communication, repetitive behavior severity, as well as cognitive, language, functional impairments). This paper conducts a multidimensional assessment of motor abilities: visuomotor (CDM1), multilimb coordination/planning (CDM2), fine-motor, and general coordination (GC1 or GC2) to identify which motor impairments are associated with, predictive of, and better explain variations in core and/or comorbid impairments in school-age children with ASD. We conducted factor analysis to identify unique motor impairments observed in the SPARK study sample. Gross-motor including visuomotor (CDM1), multilimb coordination/planning (CDM2), fine-motor (FM), and certain general coordination skills (GC1) were associated with, predictive of, and better explained the variations in social communication impairment in ASD. In contrast, fine-motor (FM) and certain general coordination (GC) skills were slightly better associated with, predictive of, and better explained the variations in repetitive behavior severity in ASD compared to gross-motor skills. In terms of comorbid impairments, gross-motor skills, especially, multilimb coordination/planning (particularly, Q06/plans) and fine-motor skills were associated with, predictive of, and better explained variations in cognitive impairments. Fine-motor and gross, visuomotor skills were most associated with and best explained variations in language delay. Both, fine- and gross-motor (visuomotor and multi-limb coordination/planning) skills and certain general coordination skills (Q11/likes sports, Q12/ learns new skills, and Q13/quick/competent) best explained variations in functional delay.

Gross- and fine-motor skills are related to both core impairments in ASD

In the previous paper (Bhat, 2021), general motor performance (using total DCD-Q scores) correlated equally with social communication impairments (using SCQ total scores) as well as repetitive behavior severity (using total RBS-R scores) (Bhat, 2021). However, the present study attempted to distinguish gross- and fine-motor relations with core ASD impairments. Gross-motor (visuomotor (CDM1) and multilimb coordination/ planning (CDM2)), fine-motor, and general coordination skills (liking sports, learning new motor skills, and being quick/competent (GC1)) were similarly associated with social communication impairments in the SPARK ASD sample (r=−0.24 to −0.26). Gross-motor and certain general coordination skills were better at explaining variations in social communication impairment (effect size = 0.86–1.02) compared to fine-motor skills (effect size $= 0.76$). While all three motor skills (CDM, FM, and GC) contributed to the social communication impairment models, gross-motor skills made stronger contributions when predicting social communication impairments after accounting for age and sex effects.

Motor delays are one of the earliest developmental markers noted in children who eventually develop ASD or related language delays (Landa & Garrett-Mayer, 2006; Bhat et al., 2012; Flanagan et al., 2012; Sacrey et al., 2015). These delays in turn affect a child's ability to explore their environment (objects during play) as well as their ability to communicate and interact with caregivers during early learning contexts (Kaur et al., 2015; Bhat et al., 2006, 2007; Srinivasan & Bhat, 2016, 2019, 2020; Iverson et al., 2018a, 2018b). Early social communication acts of head turning, physical approach through walking, reaching, showing, giving, and pointing require motor coordination and postural control (head and trunk control) and are known to be affected in young children with ASD (Nickel et al., 2013; Kaur et al., 2015; Srinivasan & Bhat, 2016). The gap in fine- and gross-motor development increases with development with motor performance in school-age children with ASD being at the level of peers half their age (Lloyd, Macdonald, & Lord, 2013, Staples & Reid, 2010).

In preschool and elementary school years, children spend a lot of time on the playground engaging in small and large group activities to connect with other children, play together, make friends, and build relationships (Bhat et al., 2011). Poor early motor skills and a growing motor gap leads to a lower sense of self-worth and poor perceived competence in children with motor problems (Piek, Baynam, & Barrett, 2006). This could further escalate the lack of social connectedness in children with ASD due to reduced movement play and lack of opportunity to build friendships. These findings join other smaller studies on relations between motor skills and social communication performance in children with ASD using standardized assessments of motor and social communication/social cognitive development as well as ASD severity (Macdonald et al., 2013a; Licari et al., 2019; Hellendoorn et al., 2015).

In contrast to gross-motor skills, fine-motor skills were only slightly better associated with $(r = -0.23)$ and better explained variations (effect size = 0.88) in repetitive behavior severity than gross-motor skills ($r = -0.17$, effect size = 0.71–0.77). Once again, while all three motor skills (CDM, FM, and GC) contributed to the repetitive behavior severity models, fine-motor skills made strongest contributions when predicting repetitive behavior severity after accounting for age and sex effects. Apart from Bhat, 2021, there are only a handful of studies reporting associations between motor performance and repetitive behaviors in individuals with ASD (Bhat, 2021). Uljarevic et al. found that early motor milestone attainment (mainly standing and toe walking) predicted repetitive sensory mannerisms and insistence on sameness scores using the Social Responsiveness Scale in a large sample of school-aged children between 2 to 18 years (Uljarevic, Hedley, Alvares, Varcin, Whitehouse, 2019). In contrast, Ravizza et al. found that stereotyped movements on the RBS-R measure was more associated with fine motor performance on a rhythmic finger tapping task and not a spatial attention task in adolescents with ASD (Ravizza, Solomon, Ivry, & Carter, 2013). Shared underlying processes were said to be impaired in adolescents with ASD leading to impairments in motor control as well as presence of repetitive behaviors. Overall, both fine and gross-motor skills seemed to correlate /predict / explain variations in core ASD impairments in social communication and repetitive behavior severity. These findings provide supportive evidence for why motor challenges should be among the core symptoms of ASD.

Fine-motor and gross-motor, multilimb coordination/planning skills are related to cognitive impairments in children with ASD

Fine-motor (r=−0.39, effect size = 0.99) and gross-motor skills (multi-limb coordination/ planning (CDM2), r=−0.29, effect size = 0.77) were more associated with, predictive of, and explained cognitive variations in children with ASD. Motor impairments are greater in magnitude and/or prevalence in children with ASD with cognitive impairment compared to those without (Green et al., 2009; Licari et al., 2019; Bhat, 2021; Ketcheson et al., 2021). It is important to note that motor impairments are observed across the autism spectrum regardless of IQ (Jansiewicz et al., 2006); however, children with accompanying cognitive impairment have more motor difficulties. Kaur et al. (2018) reported impaired fine-motor performance (fine manual and manual dexterity) on the Bruininks-Oseretsky test of Motor Proficiency as well as more imitation-based praxis errors (mirroring, overflow, and total) when copying complex gross-motor actions during praxis subtests of the Sensory Integration and Praxis test, in children with ASD with low IQ compared to children with ASD with high IQ. Additionally, while performing multilimb actions of clapping, marching, and drumming, children with ASD with low IQ had greater movement variability compared to those with high IQ. Both, fine-motor and multilimb coordination/planning skills involve complex action sequences requiring significant multitasking / executive functioning / cognitive abilities. For this reason, it is not surprising that these skills were more related to the cognitive variations in children with ASD.

Fine-motor and gross-motor (visuomotor) skills are related to language delays in children with ASD

Fine-motor (r=−0.34, effect size = 0.8) and gross-motor skills (visuomotor (CDM1), r=−0.24, effect size = 0.57) were more associated with, predictive of, and explained language variations in children with ASD. The relationship between fine-motor and language development in young and older children with and without ASD is probably the most well-reported in the literature (Bedford et al., 2015; Iverson, 2018; Choi et al., 2018; LeBarton & Landa, 2019; Gonzalez et al., 2019; Bal et al., 2020; Shield et al., 2018; Bhat et al., 2018; Gernsbacher et al., 2008). Early motor skills such as reaching, sitting, crawling, walking, etc. provide opportunities to explore new spaces and to interact with objects and caregivers, which in turn affords new perceptual information (e.g., early limb movements improve awareness of limb/bodily constraints, vertical height during walking offers a new perspective of surroundings), access to distal spaces and awareness of objects and people in it, more opportunities / instances to communicate/learn as well as more complex linguistic input from caregivers (Bhat et al., 2006, 2007; Gonzalez et al., 2019; Iverson, 2018b). While typically developing infants show a surge in rhythmic arm movements before the onset of babbling, such co-occurrence was not seen in infants at-risk for ASD (Iverson, 2018b). Early object interactions such as manual shaking/transfers, looking, and mouthing in an upright sitting position provides opportunities for caregivers to initiate communication bids and label the name and actions of objects to promote verbal and nonverbal communication in their infants. Infants at-risk for ASD or those who go onto develop ASD later are known to have different, reduced or less variable object exploration behaviors in the first year of life compared to infants without ASD (Kaur et al., 2015; Iverson et al., 2019). Additionally, infants at-risk for ASD and those who go onto develop ASD produce fewer early (showing,

giving, requesting, yes/no using head movements) and late (symbolic play-based gestures) gestures compared to infants without ASD even as early as 9 or 14 months and these differences persisted until 3 years (Iverson et al., 2018a; Leezenbaum, Campbell, Butler, & Iverson, 2014). Together, these altered language learning contexts early on in life may reduce the number of opportunities for caregiver labeling / scaffolding to facilitate early vocabulary and subsequent language development.

In contrast to the burgeoning literature on motor-language relations, there are fewer studies on how visuomotor skills (e.g., ball and balance skills) may be specifically impaired in children with ASD compared to other diagnoses such as ADHD and specific language impairment (SLI) (Dowell et al., 2009; Bhat et al., 2010; 2018; Whyatt & Craig, 2012; Hellendoorn et al., 2015; Ament et al., 2014; McPhillips et al., 2014; Macdonald et al., 2014). McPhillips et al. (2014) reported greater associations between visuomotor and language impairments in children with ASD than those with specific language impairments. More recently, Wu et al. (2021) reported associations between object manipulation and visuo-motor integration performance on the Peabody Developmental Motor Scale with receptive/expressive language development in children newly diagnosed with ASD. Shared perceptuo-motor requirements, developmental processes, and common underlying neural processes between visuomotor and language development, such as early caregiver observation and imitation skills as well as shared neural networks (explained later in more detail), provide a basis for the associations between these two developing systems.

Gross- and fine-motor skills have profound impact on functional independence of children with ASD

In the present study, visuomotor (CDM1), multilimb coordination/planning (CDM2), and fine motor skills moderately correlated with, predicted, and well-explained variations in parent-reported functional delays (r=−0.32 to −0.40, effect sizes = $0.91-1.22$). There are few reports on relations between motor skills and daily functioning in young and older children with ASD (Macdonald et al., 2013b; Bhat, 2021; Licari et al., 2019; Jasmin et al., 2009). The Bhat, 2021 paper reported moderate associations between general motor performance and parent-reported functional delays in the SPARK ASD sample. Licari et al. (2019) reported that motor skill performance correlated with the daily living skills domain on the Vineland Adaptive Behavioral Scales (VABS) in a large Australian sample of children with ASD between 2 to 6 years and yet few children were being screened and treated for their motor problems. Macdonald et al. (2013b) found that fine motor skills of young children with ASD (14 to 49 months) were more predictive of all adaptive domains on the VABS whereas gross motor skills were more predictive of the daily living skills only. Jasmin et al. (2009) also reported similar associations between motor and daily living skills in preschoolers with ASD using 2 different functional measures - Functional Independence Measure (Wee FIM) and the VABS and the Peabody Developmental Motor Scales (PDMS) in preschoolers with ASD. Multiple motor dimensions including locomotion, object manipulation, grasping, and visuomotor integration correlated with the self-care domain of the WeeFIM and/or the personal/daily living skills domain of the VABS. Poor motor performance / perceived motor competence as well as dyspraxia or difficulty performing complex motor sequences in children with ASD may affect their ability to perform a variety

of daily physical functions (fine and gross-motor) leading to negative cascading effects on their social, communication, and cognitive functioning.

Certain General Coordination Skills Linked to Core and Functional Impairments in ASD

General coordination skills (GC1) of liking sports participation, being able to learn new motor skills easily, and being quick/competent at various daily motor functions were moderately associated with, contributed to, and/or explained variations in core social communication skills as well as comorbid cognitive and functional impairments. The last 2 general coordination skills $(GC2 = \text{bull in china shop and fatigues easily})$ weakly correlated with core / comorbid impairments. This could be because they were the only items stated in reverse order to reduce respondent bias whereas the earlier 13 items were not. To be clear, Q01 to Q13 DCD-Q items are stated as follows "Your child catches a ball or hits a ball accurately, etc." with better scores being a 5. On the other hand, the last 2 items (Q14 and Q15) are stated in reverse, "Your child would never be described as clumsy or does not fatigue easily" with better scores also being a 5. The wording describing the last 2 items could be confusing and may be contributing to their unreliability/ineffectiveness. It is possible that the skills stated in the last 2 questions are still valuable and important for describing motor problems in children. From an intervention perspective, children with ASD may be limited in their sports/physical activity participation, have challenges acquiring new motor skills, and are slower and need more time to complete complex (multistep) motor functions.

Neural Framework for Motor-Other System Relations in ASD

School-age children with ASD presented with visuomotor (CDM1), multilimb coordination/ planning (CDM2), fine-motor and general coordination (GC1) impairments that were associated with their core as well as comorbid impairments. These findings fit with the framework of disrupted connectivity in individuals with ASD including structural and functional abnormalities within primary motor cortices, corticospinal tracts, interhemispheric connectivity, as well as connectivity between various cortical and subcortical regions including cortico-cerebellar, cortico-striatal, and thalamo-cortical connectivity (Turner, Frost, McKilroy, Liesenbardt, & Mueller, 2006; Courchesne et al., 2007; Just, Keller, Malavi, Kana, & Varma, 2012; Frazier, Keshavan, Minshew, & Hardan, 2012; Nair et al., 2013; Maximo, Cadena, & Kana, 2014; Carper, Solders, Treiber, Fishman, & Mueller, 2015; Vasa, Mostofsky, & Ewen, 2016; Chen et al., 2018; Floris & Howells, 2018). Specific brain abnormalities attributed to visuospatial/visuomotor (includes fine/gross-motor) disturbances include reduced/aberrant long-range fronto-occipital, frontoparietal, cortico-subcortical functional connectivity (Villalobos et al., 2005; Just et al., 2012; Maximo et al., 2014; Nebel et al., 2016; Wang et al., 2019; Lidstone et al., 2021) along with increased local connectivity in frontal, temporal, parietal, and occipital cortices as well as overconnectivity in certain cortico-cortical and cortico-subcortical pathways (Maximo et al., 2014; Wang et al., 2019; Lidstone et al., 2021). Nebel et al. (2016) reported greater temporal asynchrony between motor (primary motor and ventral premotor, and parietal operculum) and visual cortices (Brodmann's areas 17–19) in individuals with ASD implying aberrant functional connectivity between these regions. Additionally, visuo-motor asynchrony in individuals with ASD was associated with gestural imitation performance on

the Florida Apraxia Battery as well as social communication performance on the Social Responsiveness Scale (SRS). Both, Wang et al. (2019) and Lidstone et al. (2021) have implicated increased cortico-cerebellar (including cerebellar-occipital and cerebellar-inferior parietal) connectivity as the basis for visuomotor disturbances in individuals with ASD. The aforementioned brain regions are not only important for visuomotor functions but also for imitation-based learning and social communication development and offer plausible explanations for motor - social communication relations in ASD.

Additionally, multilimb coordination/planning impairments (including fine/gross-motor) could also be explained by executive functioning (EF) problems that not only affect cognitive processing but also complex, social communication and perceptuo-motor skills. EF skills include inhibition, working memory, motor planning, and task switching/organization that are often impaired in children with ASD and could directly contribute to their difficulty performing complex, multistep motor skills/gestures also known as dyspraxia (Lai et al., 2017). Individuals with ASD have aberrant connectivity in the EF / frontoparietal networks including prefrontal/frontal cortices (include superior, middle, and inferior frontal and medial prefrontal gyri), posterior parietal cortex, and anterior cingulate cortex, and insula which is also associated with poor performance on EF tasks as well as repetitive behavior severity (Dajani & Uddin, 2015; May & Kana, 2020). Wang et al. (2019) reported reduced functional connectivity between cerebellar-frontal/prefrontal and cerebellar-temporal cortices in individuals with ASD; which may also contribute to EF / perceptuo-motor difficulties. Taken together, connectivity disruptions found in the various aforementioned brain networks may contribute to the multisystem impairments (including motor issues) in ASD.

Implications for future DSM criteria

The overall implications of Bhat, 2020, 2021 and the present study are that motor impairments in children with ASD are pervasive (seen in 87–88% of the sample), persistent (did not change between 5 and 15 years) and associated with, predictive of, and/or explain variations in core and comorbid impairments in ASD. Gross-motor (visuomotor, multilimb coordination/planning) and fine-motor skills are uniquely affected in ASD and relate to various other system impairments. Motor impairments are also very well-explained by the popular disrupted connectivity framework of ASD. These multidimensional motor findings in the SPARK ASD sample join the multiple recent calls (Licari et al., 2019; van Damme et al., 2021; Miller et al., 2021) for inclusion of motor impairments within the definition of ASD either within the criteria (relations to social communication skills or repetitive behaviors) or specifiers (relations to cognitive and language impairments). If the motor system is represented within the ASD definition, it would strongly encourage diagnosticians (physicians, pediatricians, psychologists, and psychiatrists) to conduct early motor screening and diagnostic referrals to movement clinicians (OTs and PTs) to address the early motor delays and the increasing motor gaps in childhood, as well as the negative impacts of developmental dyspraxia on overall child development and functioning in adolescence and adulthood. Identifying motor problems early on through motor screening could make available interventions that prevent the cascading negative impacts on children's concurrent

and future cognitive, social communication, language, and functional development, physical fitness/activity levels, social participation, as well as physical/mental health and well-being.

Implications for Motor Screening, Assessment, and Intervention

Often motor evaluations and services (specifically, PT) are not recommended to children with ASD because most children acquire major motor milestones despite some delays. More strategic efforts will be needed to change the mindset of clinicians, educators, and researchers for them to often recommend and practice motor screening, assessments, and interventions to address the more complex and persistent fine and gross motor problems associated with developmental dyspraxia/DCD. Screening tools such as the Ages-Stages Questionnaire (ASQ), DCD-Q and Movement-ABC checklist provide quick tools to screen for motor delays in young infants and children (van Damme et al., 2021; Miller et al., 2021). In particular, the psychometrics of the DCD-Q using the internally consistent and validated 5-subdomain structure might work better for motor screening in children with ASD. If a risk for motor impairment is identified, then the next step would be to complete an age-appropriate, full motor assessment such as the Alberta Infant Motor Scale (AIMS) for infants, Peabody Developmental Motor Scale (PDMS) for infants and toddlers, or the Movement-ABC (MABC) or Test of Gross Motor Development (TGMD) for school-age children. This would determine the extent and nature of motor impairments which in concert with family/child's goals for motor functioning could help determine the next steps for providing effective motor interventions.

Research conducted in the first author's lab has evaluated effects of various creative movement (music and movement, yoga, dance) and physical activity interventions that were truly multisystem in nature due to key ingredients such as gross-motor coordination/balance training, interpersonal synchrony/turn-taking, as well as social interactions including, listening, singing/conversations, and leading/following (Srinivasan et al., 2015a, 2015b, 2016a, 2016b; Kaur & Bhat, 2019, 2021). Our recent systematic review found strong evidence for the positive effects of music and movement on the social communication, cognitive-behavioral, and perceptuo-motor skills of children with ASD (Amonkar, Su, Srinivasan, & Bhat, 2021). Such interventions may also facilitate changes in functional activation and connectivity to normalize brain functioning in ASD using recently identified neurobiomarkers (Su et al., 2020a, 2020b, 2021, 2022; Sharda et al., 2018). Our current work has found reduced superior temporal and inferior frontal activation in children with ASD compared to age-matched controls, during naturalistic motor tasks (Bhat et al., 2017, 2019; Su et al., 2020a, 2020b, 2021). These biomarkers are currently being utilized to assess changes in neural response following 8-week long, multisystem, movement-based interventions (Su, Srinivasan, Cleffi, & Bhat, 2021; Srinivasan, Su, Cleffi, & Bhat, 2021; Bhat, Su, Cleffi, & Srinivasan, 2021; Cleffi, Su, Srinivasan, & Bhat, 2022). Taken together, there is a need to diversify autism interventions through inclusion of evidence-based, multisystem approaches that target perceptuo-motor, social communication, and cognitive skills of children.

Finally, on the clinical front, a lot more remains to be done to increase community awareness about the under-diagnosis (only 15% have DCD co-diagnosis in the SPARK

ASD sample) and under-treatment (PT in 32% and recreational therapies in 13% in the SPARK ASD sample) of motor problems in ASD. ABA therapists, special educators, as well as movement clinicians (OTs, PTs, adaptive physical educators) must utilize evidencebased movement interventions (age-appropriate motor skills training, physical activity/ exercise, sports participation, or creative movement such as yoga and music and movement individually or in small groups) to make interventions more embodied (i.e., inclusive of movement) and socially embedded/relevant to promote motor, cognitive, and social communication development. Even early on in life, when receiving early intervention (EI), it will be important to identify the mild to moderate motor delays in infants/preschoolers at-risk for ASD and address them through parent-mediated interventions and/or IDEA-based EI services as many parents/clinicians might make the error of "waiting" when in fact, early delays can be easily addressed to avoid more significant motor deficits in the future. On the research front, there is a clear dearth of motor intervention studies in individuals with ASD across the lifespan to facilitate motor functioning/physical activity and to study its cascading effects on other developmental domains/functioning, physical/mental health, as well as its underlying neural mechanisms.

Limitations

This study relied on parent report questionnaires and parent reports of children's abilities which could be influenced by reporting biases such as the Horn effect i.e., parents of children with greater impairments may have rated their child worse. However, parent reports are a feasible method to obtain information from a large sample to help elucidate the population scale patterns of a disorder. Moreover, clinical assessments using large-sized samples would be important to further confirm the pattern of motor impairment reported here. Even though this study used a large sized-sample the SPARK study seems biased towards recruiting white $({\sim}76\%)$ families at this time. However, the income and age distributions were fairly even across the sample. Last but not the least, it would be valuable to study motor performance across the lifespan in individuals with ASD by adding more objective, clinical motor / functional assessments to the SPARK study battery (e.g., PDMS, MABC, TGMD, etc.).

Conclusion

The present study examined the SPARK ASD sample of children between 5 and 15 years to conduct a multidimensional motor assessment using factor analysis, subgrouping, and correlational/effect size analyses. Visuomotor, multilimb coordination/planning, fine motor, and general coordination impairments were uniquely distinguishing motor problems of children with ASD. Both, gross-and fine-motor skills as well as certain general coordination skills were associated with, predictive of, and explained variations in the core impairments of children with ASD as well as their comorbid impairments in cognition, language and functioning. The comprehensive analysis of motor impairments in a large ASD cohort across 3 studies (Bhat, 2020, 2021, and this paper) provide robust evidence for the inclusion of motor impairments within the definition of ASD, either under diagnostic criteria or specifiers. Given the growing calls to address motor problems in the ASD population, diagnosticians should begin recommending systematic motor screening, further evaluations, and treatments for children at-risk for and newly diagnosed with ASD. Parents, clinicians,

and educators in early intervention and school systems should be made aware of the need to address motor delays/gaps in their children and should not be complacent with a "wait and see" strategy or inaction. Motor advocacy and enhanced public/clinical community awareness is urgently needed to begin moving the needle in the right direction for children and families who clearly have unmet motor needs that they too are potentially unaware of.

Refer to supplementary information for list of acronyms and abbreviations, list of motor definitions, Figures S1–S2, and Tables S1–S11.

Supplementary Material

Refer to Web version on PubMed Central for supplementary material.

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Figure 1:

DCD-Q total, standard subdomain (CDM, FM, GC), and factor analysis-based subdomain (CDM1, CDM2, FM, GC1, GC2) scores as a function of SCI, RB severity, cognitive, language, and functional impairment categories (C1 to C5) in children with ASD. Note that FM subplot is the same for the standard subdomains and the factor analysis-based subdomains. The total and standard subdomain subplots are taken from Bhat, 2021. CDM: Control During Movement, FM: Fine Motor, GC: General Coordination.

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Figure 2:

Effect sizes for DCD-Q total score, standard subdomains (CDM, FM, GC), and factor analysis-based subdomains (CDM1, CDM2, FM, GC1, GC2). Horizontal dashed lines are added to illustrate high effect sizes (0.8 or -0.8). CDM: Control During Movement, FM: Fine Motor, GC: General Coordination.

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Figure 3:

Radar chart showing the effect sizes for DCD-Q total score and factor analysis-based subdomains (CDM1, CDM2, FM, GC1, GC2) to compare effects across impairments/delays. CDM: Control During Movement, FM: Fine Motor, GC: General Coordination.

Table 1:

Sample size and demographic information for the SPARK sample used in this analysis.

 $\mathscr{V}_{\text{Each row}}$ is independent, i.e., the percentages do not need to sum to 100.

Table 2:

Factor analysis to assess the factor structure based on maximum likelihood method. For each question, shown value indicates the factor to which the question most likely belongs. Order of these factors is based on the percent variance explained by each factor and is therefore different from the factors shown in the other tables. For example, Factor 1 is similar to the Fine Motor (FM) subdomain. Factor 2 is similar to the Control During Movement 1 (CDM1) subdomain. Factor 3 is similar to the Control During Movement 2 (CDM2) subdomain. Factor 4 is similar to the General Coordination 1 (GC1) subdomain. Factor 5 is similar to the General Coordination 2 (GC2) subdomain. Note that Q13 could belong in any of the Factors 1, 4, or 5.

Table 3:

Correlations across DCD-Q subdomains/items, measure-based impairment categories (SCI and RB severity), and parent-reported delay categories (Cognitive, Language, and Functional delays).

SCI: Social Communication Impairment, RB = Repetitive Behaviors, CDM: Control During Movement, FM: Fine Motor, GC: General Coordination. All correlations are statistically significant with a p-value of 0.0023 (after Bonferroni correction) or lower. Correlation values ≥0.2 or ≤−0.2 are highlighted to show important relations.

Table 4:

Wald chi-square statistic to predict the impairment categories based on SCI and RB severity as well as Cognitive, Language, and Functional delay using age, sex, and DCD-Q total/subdomains/item scores within ordinal logistic regression analyses. Predictor significance is based on FDR-corrected p-values.

Note that **indicates $p < 0.007$, *indicates p<0.05). SCI: Social Communication Impairment, RB = Repetitive Behaviors, CDM: Control During Movement, FM: Fine Motor, GC: General Coordination, FDR: False Discovery Rate.

Table 5:

Subgroup effect sizes across DCD-Q subdomains/items, measures-based impairment categories (SCI and RB severity), and parent-reported delay categories (Cognitive, Language, and Functional delays).

SCI: Social Communication Impairment, RB = Repetitive Behaviors, CDM: Control During Movement, FM: Fine Motor, GC: General Coordination. Effect sizes ≥0.8 or ≤−0.8 (large) are highlighted in green and 0.5 to 0.8 or −0.5 to −0.8 (moderate) are highlighted in gold to show how motor performance (DCD-Q) distinguishes the extreme categories based on other impairments/delays.