Prediction of Functional Academic Outcomes by Fine Motor Skills in Individuals With Sickle Cell Disease

Lakia Kearson, Christina Dandar, Catherine Hoyt, Jennifer Longoria, Victoria Okhomina, Darcy Raches, Brian Potter, Guolian Kang, Jane Hankins, Clifford Takemoto, Andrew Heitzer

Importance: Individuals with sickle cell disease (SCD) are at heightened risk of poor neurocognitive and academic outcomes. The relationship between fine motor skills and academic outcomes is not well understood.

Objective: To compare the fine motor skills of individuals with SCD with normative expectations, test whether demographic and medical factors are associated with fine motor performance, and determine the impact of fine motor performance on academic performance.

Design: Cross-sectional.

Setting: St. Jude Children's Research Hospital.

Participants: Individuals with SCD ($N = 376$; ages 8–24 yr).

Outcomes and Measures: Fine motor outcomes included visual–motor integration, manual dexterity, and graphomotor speed. Academic outcomes included math fluency and word reading. Demographic and medical variables were obtained via medical records and interviews.

Results: Compared with normative expectations, the performance of individuals with SCD on all fine motor measures was lower than expected. Male sex, lower socioeconomic status, and lower oxygen saturation was associated with slower graphomotor speed. Lower socioeconomic status and older age were associated with lower visual–motor integration scores. Performance on all fine motor measures was positively associated with math fluency and word reading.

Conclusions and Relevance: Individuals with SCD exhibited poorer than expected fine motor skills across multiple motor domains, and these deficits were associated with poorer academic outcomes. Early referral to intervention services for fine motor skills may facilitate improved academic outcomes for individuals with SCD.

Plain-Language Summary: This study had three objectives: (1) Compare the fine motor skills of people with sickle cell disease (SCD) with normative expectations, (2) test whether demographic and medical factors are associated with fine motor performance, and (3) determine the impact of fine motor performance on academic performance. We found that SCD is a risk factor for lower than expected fine motor performance across multiple fine motor domains and that these deficits also affect functional academic skills.

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Sickle cell disease (SCD) is an inherited blood disorder affecting approximately 100,000 people in the United States (Knight et al., 2021; Marchese et al., 2022). SCD consists of a group of hemoglobinopathies and is characterized by a change in form and flexibility of red blood cells that can lead to irregular blood flow and supply of oxygen to tissue (Burkhardt et al., 2017; Marchese et al., 2022). Thus, cerebrovascular complications, including silent cerebral infarctions and overt ischemic or hemorrhagic stroke, are more common among individuals with SCD than in the general population (Aggeli et al., 2021; Marchese et al., 2022). However, the risk of neurological complications is often moderated by the inherited hemoglobin gene

variant (Quinn, 2016), and those with more severe forms of the disease, such as the HbSS and HbSβ^0genotypes, tend to have more frequent and severe neurological complications than those who have milder forms of SCD, such as the HbSC and HbSβ^+ genotypes (Quinn, 2016). Nonetheless, varying degrees of symptoms (e.g., hemolytic anemia and vaso-occlusive events) can be observed across all SCD genotypes (Steinberg, 2008). Individuals with SCD and a history of a stroke or silent infarct display greater neurocognitive deficits than those without this history; however, even in the absence of these complications, individuals with SCD display greater neurocognitive deficits than their peers, likely because of chronic insufficiencies in oxygen or glucose in the brain (Aggeli et al., 2021).

Fine motor performance is a particular skill that has been shown to be at risk in this population. In general, individuals with SCD show significantly weaker fine motor skills compared with healthy control participants (Burkhardt et al., 2017; Gentry et al., 1997; Hijmans et al., 2011; Newby et al., 2018; Schatz & Roberts, 2007). However, fine motor performance in daily life involves a range of associated and interdependent skills, including manual dexterity, graphomotor speed, and visual–motor integration. *Manual dexterity* refers to efficient manipulation of objects (Strooband et al., 2020), and *graphomotor speed* refers to rapid hand motor actions executed manually, typically during writing or drawing (Knaier et al., 2022). *Visual–motor integration* encompasses how well an individual can skillfully coordinate visual input with motor movements (Carsone et al., 2021). The research that has examined these individual skills in the context of SCD has been limited. Of these studies, a majority have investigated fine motor skills using a global composite (Schatz & Roberts, 2007) or included a single measure of fine motor skills (e.g., only visual–motor integration; Hijmans et al., 2011). Only one available study simultaneously investigated the numerous components of fine motor performance (Newby et al., 2018). This study found that individuals with SCD exhibited significantly weaker visual–motor integration skills than healthy control participants, but average manual dexterity (Newby et al., 2018). Nonetheless, because of the limited research, whether these skills are equally vulnerable to the disease-related impacts of SCD is unclear.

Within the population with SCD, older age (Kral et al., 2006; Schatz & Roberts, 2007), lower level of parent education (Drazen et al., 2016), and lower socioeconomic status (Drazen et al., 2016; Tarazi et al., 2007) have been associated with poorer fine motor skills. Fine motor deficits have also been observed in individuals with SCD with (Brown et al., 2000; Gold et al., 2008; Schatz et al., 2001) and without (Berkelhammer et al., 2007; Daly et al., 2008; Grueneich et al., 2004; Kral et al., 2006) a history of cerebrovascular incidents and regardless of SCD genotype (Grueneich et al., 2004; Johnston et al., 2022; Tarazi et al., 2007). However, the studies that have investigated the effects of demographic or medical factors on fine motor skills among individuals with SCD have been limited by their assessment of fine motor performance and included measures that assess only one or two fine motor components.

In the general population, age-typical fine motor skills facilitate cognitive and academic development (Brons et al., 2021; Cools et al., 2009; Escolano-Pérez et al., 2020) because fine motor skills are required for an abundance of academic tasks, including writing and typing (Gaul & Issartel, 2016). Those who develop early fine motor skills at an age-appropriate rate have been shown to exhibit greater academic success in the areas of mathematics and early literacy (Gaul & Issartel, 2016; Strooband et al., 2020; Suggate et al., 2016). Individuals with SCD demonstrate a host of academic difficulties and poor educational attainment (Epping et al., 2013; Schatz, 2004; Schatz et al., 2001). However, no studies have examined the impact of fine motor skills on academic performance in this population. Understanding the impact of fine motor skills on academic outcomes among children with SCD may facilitate earlier referral to intervention.

In the current study, our first objective was to compare the fine motor skills of a large sample of individuals with SCD with normative expectations. The second objective was to determine whether demographic components and medical factors (e.g., sickle cell genotype, age, sex) were associated with fine motor performance among individuals with SCD. The third objective was to determine whether fine motor performance was associated with academic performance among individuals with SCD. We hypothesized that children with SCD would perform below normative expectations, and poor fine motor performance would be associated with age, socioeconomic status, and genotype. We also predicted that fine motor performance would be positively associated with math fluency and word reading.

Method

Participants

Participants who were enrolled in the Sickle Cell Clinical Research Intervention Program (SCCRIP) were included in our analysis. The organizational structure and design of SCCRIP has previously been described (Hankins et al., 2018). In brief, SCCRIP is a longitudinal lifetime cohort study that collects clinical, psychosocial, neurocognitive, and health-related outcomes of children, adolescents, and young adults with SCD (Hankins et al., 2018). Neurocognitive evaluations occur across four developmental stages: school age (ages 8–9 yr), early adolescence (ages 12–13 yr), late adolescence (ages 16–17 yr), and young adulthood (ages 19–24 yr; Hankins et al., 2018). Approval was provided by the St. Jude Internal Review Board, and written informed consent was provided by all participants before their participation in the SCCRIP study.

If a participant received multiple assessments, we used the most recent in our analysis.

Demographic and Medical Variables

Demographic and medical variables were abstracted from the SCCRIP database. According to National Heart, Lung, and Blood Institute guidelines, all individuals with HbSS or HbSβ^0 genotypes received hydroxyurea and those with HbSC or HBSβ^+ genotypes received hydroxyurea on the basis of the frequency of disease-related complications (Luchtman-Jones et al., 2016). We ascertained age of hydroxyurea initiation and duration of treatment. Hematologic indices included hemoglobin concentration, fetal hemoglobin, white blood cell count, and platelet count. These indices were either obtained on the same day as neurocognitive testing or were the average of lab values recorded within 3 mo before testing. Daytime hemoglobin oxygen saturation was acquired on the same day as neurocognitive testing. The Social Vulnerability Index was calculated for each participant on the basis of zip code; it measures socioeconomic disadvantage related to poverty, education level, and housing (Cutter et al., 2003; Flanagan et al., 2018). Scores are presented as a percentile (0–100), and higher scores indicate greater risk. The Barratt Simplified Measure of Social Status (BSMSS) was used to measure household socioeconomic status on the basis of parental education and occupation status. Total composite scores range from 8 to 66, with lower scores indicating lower socioeconomic status (Barratt, 2006).

Neurocognitive Measures

Neuropsychological assessments were administered or supervised by licensed psychologists. Assessment measures were selected to examine cognitive, academic, and fine motor outcomes quantitatively. The Coding subtest from the Wechsler Intelligence Scale for Children, Fourth or Fifth Editions (WISC–IV or WISC–V, respectively; Wechsler, 2003, 2014b) or the Wechsler Adult Intelligence Scale, Fourth Edition (WAIS–IV; Wechsler, 2014a), was used to measure graphomotor speed, dependent on participant age. For this subtest, participants use an index to copy symbols with a pencil as quickly as possible within a specified time limit. The Beery–Buktenica Developmental Test of Visual–Motor Integration, Sixth Edition (Beery VMI), was used to assess visual–motor integration. This assessment requires participants to copy geometric forms with a pencil. The forms are arranged in developmental sequence from less to more complex (Beery, 2010). The Grooved Pegboard Test was used as a measure of manual dexterity. This assessment involves quickly placing keyhole-shaped pegs into 25 holes one at a time in a prescribed order (Klove, 1963). Each peg must be rotated to match the hole before it can be inserted. Participants first complete the task with their dominant hand as quickly as possible (i.e., timed measure) and then do so with their nondominant hand (Klove, 1963).

The Wechsler Abbreviated Scale of Intelligence, Second Edition (WASI–II; Wechsler, 2011) was used to measure general cognitive ability (four-subtest IQ). Hand dominance was based on parent report. Academic achievement measures included the Math Fluency and Letter–Word Identification subtests from the Woodcock–Johnson Tests of Achievement, Third or Fourth Edition. The Math Fluency subtest measures speed of computation or the ability to solve simple addition, subtraction, and multiplication problems within a specified time limit (McGrew, 2014). The Letter–Word Identification subtest measures an individual's ability to identify letters and read words as they become increasingly difficult (McGrew, 2014). Tests were selected according to participant age at the time of the evaluation. For tests with multiple editions, the most up-to-date version of each test was administered to each participant. The correlation between the WISC–IV and WISC–V Coding subtests was acceptable. For participants with multiple neuropsychological assessments, only the most recent scores were included.

Analyses

Participants' demographics, clinical characteristics, and SCD disease-modifying therapy were reported using means and standard deviations or frequencies and percentages. Differences between SCD genotypes were tested using χ^2 or Fisher's exact tests for categorical characteristics and two-sample t tests or Wilcoxon rank-sum tests for continuous characteristics.

To address our first objective, t tests and Wilcoxon signed-rank tests were used to compare fine motor skills in our sample with normative expectations. Cohen's d was also calculated to assess differences between our sample and normative expectations. Linear regressions determined associations between fine motor performance and demographic and medical factors to address our second objective. Factors associated with fine motor performance at $p < .10$ in univariate models were used as potential predictors in multivariable stepwise selection models that assessed the relationship between patient factors and fine motor function independent of the effects of other factors. SCD genotype was included in each final model on the basis of previous literature (Heitzer et al., 2021). Each full model (i.e., fine motor skill score = all potential covariates) was checked for multicollinearity, and variables with a variance inflation factor greater than 2 with SCD genotype included in the model were excluded as potential covariates. The final predictors in the multivariate models were based on the model's Akaike information criterion (AIC).

Spearman correlations and linear regressions were used to determine whether fine motor performance was associated with academic performance.

Multivariable linear regression models created using AIC-based stepwise variable selection were used to assess whether significant relationships between fine motor performance and academic performance remained after controlling for demographic and medical factors related to academic performance. To further assess the association of fine motor performance with academic performance independent of global cognition, an abbreviated IQ score (WASI–2 four-subtest IQ) was also included in multivariable models with selected demographic and medical factors.

False discovery rate–adjusted *p* values (*p*FDRs) were provided to indicate significance after accounting for analyses with multiple testing. Unless otherwise stated, *p* values were two-sided and considered significant when $p < .05$ or $p\text{FDR} < .05$ (for multiple comparisons). All analyses were performed in SAS (Version 9.4).

Results

Demographic and Clinical Characteristics

Demographic and clinical characteristics by SCD group (HbSC/HbSβ$^+$ and HbSS/HbSβ^0) are displayed in Table 1. A total of 376 individuals (ages 8–24 yr) were administered a neurocognitive evaluation with fine motor subtests. Approximately 41% were diagnosed as having the HbSC or HbSβ$^+$ genotype, and 59% were diagnosed as having the HbSS or HbSβ^0 genotype. On average, individuals were age 13.35 yr (*SD* = 4.19) at evaluation and right hand dominant (72.87%), and roughly half the sample was male (50.53%). Mean SVI and BSMSS scores were 64.95 (*SD* = 26.16) and 30.23 (*SD* = 13.20), respectively, indicating that the total sample, on average, had higher social vulnerability. As expected, differences were found in medical treatment and lab values, based on SCD genotype. Consistent with other sickle cell cohorts, those with HbSS and HbSβ^0 genotypes were more

Table 1. Demographic and Clinical Characteristics by SCD Genotype

Characteristic	*n*	Total Sample (*N* = 376)	HbSS or HbSβ^0 (*n* = 222)	HbSC, HbSβ$^+$, or Other (*n* = 154)	*p*FDR
		n (%)			
Sex	376				.40
Female		186 (49.47)	105 (47.30)	81 (52.60)	
Male		190 (50.53)	117 (52.70)	73 (47.40)	
Race	376				.25
African American		373 (99.20)	221 (99.55)	152 (98.70)	
Other		1 (0.27)	1 (0.45)	0 (0.00)	
White		2 (0.53)	0 (0.00)	2 (1.30)	
Hand dominance	322				.07
Right		274 (72.87)	163 (73.42)	111 (72.08)	
Left		48 (12.77)	34 (15.32)	14 (9.09)	
Current HU use	376	208 (55.32)	172 (77.48)	36 (23.38)	**<.001**
		M (*SD*)			
Age at evaluation, yr	376	13.35 (4.19)	13.32 (4.35)	13.40 (3.97)	.71
Age started HU, yr	250	7.73 (5.01)	6.78 (4.37)	11.50 (5.65)	**<.001**
Duration of HU, yr	376	3.78 (4.52)	5.74 (4.67)	0.96 (2.25)	**<.001**
SVI[a]	375	64.95 (26.16)	65.90 (26.50)	63.59 (25.68)	.43
BSMSS[b]	302	30.23 (13.20)	29.99 (12.68)	30.57 (13.93)	.72
Hemoglobin, g/l	365	10.08 (1.77)	9.07 (1.24)	11.53 (1.37)	**<.001**
Fetal hemoglobin, %	320	12.47 (10.06)	16.81 (9.42)	4.83 (5.61)	**<.001**
White blood count, $\times 10^9$/l	365	8.72 (3.56)	9.36 (3.61)	7.79 (3.28)	**<.001**
Platelet count, $\times 10^9$/l	365	342.68 (174.15)	395.90 (188.93)	266.39 (113.49)	**<.001**
Oxygen saturation, %	211	99.48 (1.04)	99.36 (1.18)	99.63 (0.82)	.25

Note. Values in bold are statistically significant at a *p*FDR < .05. BSMSS = Barratt Simplified Measure of Social Status; *p*FDR = false discovery rate–adjusted *p* value; HU = hydroxyurea; SCD = sickle cell disease; SVI = social vulnerability index.

[a]Classifies individuals based on social vulnerabilities at the neighborhood level (e.g., poverty, education, housing data); a higher percentile score (0–100) indicates higher social vulnerability.

[b]Classification system that codes occupations based on skill, power, and social position in society. Lower scores indicate lower socioeconomic status, ranging from 8 to 66.

likely to be treated (77.48% treated) with hydroxyurea than those with HbSC and HbSβ$^+$ genotypes (23.38% treated; *p*FDR < .001). The HbSS–HbSβ^0 subgroup had lower total Hb values (*p*FDR < .0001) than the HbSC–HbSβ$^+$ subgroup.

Comparison With Normative Expectations

A comparison of fine motor performance among individuals with SCD with normative expectations is displayed in Table 2. When compared with normative values, individuals with SCD scored significantly lower on all fine motor measures, including graphomotor speed, visual–motor integration, and bilateral manual dexterity (all *p*FDRs < .001).

Associations Between Demographic and Medical Factors and Fine Motor Skills

Associations between fine motor skills and demographic and medical factors among individuals with SCD are displayed in Table 3. In multivariable analyses adjusted for relevant covariates and forcing SCD genotype into the model, male sex was associated with lower performance on graphomotor speed ($p < .001$) and dominant hand manual dexterity ($p = .003$). Higher household socioeconomic status was associated with better performance on measures of graphomotor speed ($p = .006$) and visual–motor integration ($p = .003$). There was an inverse relationship between age and performance on a measure of visual–motor integration ($p < .001$). Daytime oxygen saturation was positively associated with graphomotor speed performance ($p = .042$). Across all analyses, fine motor performance was not dependent on SCD genotype (all *p*s > .05).

Associations Between Fine Motor and Academic Performance

Multivariable analyses examining the associations between fine motor and academic performance are displayed in Table 4. When accounting for demographic and medical covariates, graphomotor speed, visual–motor integration, and dominant and nondominant hand manual dexterity were positively associated with performance on a measure of math fluency (all *p*s < .001). Graphomotor speed, visual–motor integration, and nondominant hand manual dexterity were positively associated with performance on a measure of basic word-reading skills (all *p*s < .05).

To account for the potential influence of global cognition on the relationship between fine motor skills and academic performance, multivariable analyses were repeated after forcing an abbreviated IQ measure (WASI–II four-subtest IQ) into the models. The models with abbreviated IQ included are displayed in Table A.1. in the Supplemental Material (available online with this article at https://research.aota.org/ajot). After controlling for global cognition and other relevant covariates, graphomotor speed and dominant and nondominant manual dexterity continued to be positively associated with a measure of math fluency ($p < .05$). Only visual–motor integration remained a significant predictor of basic word-reading skills ($p = .006$) when the four-subtest IQ was included in the model, with higher visual–motor integration scores associated with better word-reading skills.

Discussion

Age-typical fine motor skills facilitate cognitive and academic development and are necessary to carry out activities of daily living. Children and adolescents with SCD are at increased risk of neurocognitive and fine motor deficits due to disease-related complications. In comparison with normative expectations, the performance of individuals with SCD in this study was significantly lower across all areas of fine motor functioning assessed. Likewise, demographic (e.g., age, sex, socioeconomic status) and medical (e.g., oxygen saturation) factors were associated with fine motor outcomes, and fine motor skills were predictive of functional academic outcomes. These results highlight the need for careful monitoring of fine motor skills among children with SCD, with referral to occupational therapy as appropriate. Given the relevance of

Table 2. Fine Motor Skills Among Individuals With SCD Compared With Normative Expectations

Variable	*M* (*SD*)		Cohen's *d*	*p*FDR
	Individuals With SCD	Normative Expectations		
Graphomotor speed[a]	7.93 (2.82)	10 (3)	−0.71	**<.001**
Visual–motor integration[b]	84.71 (13.05)	100 (15)	−1.09	**<.001**
Manual dexterity[c]				
Dominant hand	−1.51 (1.94)	0 (1)	−0.98	**<.001**
Nondominant hand	−1.30 (2.06)	0 (1)	−0.80	**<.001**

Note. Values in bold are statistically significant at *p*FDR < .05. *p*FDR = false discovery rate–adjusted *p* value; SCD = sickle cell disease.
[a]Scaled score from Coding subtest of the Wechsler Intelligence Scale for Children, Fourth or Fifth Edition, or the Wechsler Adult Intelligence Scale, Fourth Edition.
[b]Standard score from Beery–Buktenica Developmental Test of Visual–Motor Integration, Sixth Edition.
[c]*Z* score from Grooved Pegboard Test.

Table 3. Effects of Demographic and Medical Factors on Fine Motor Skill Performance

Effect	Graphomotor Speed[a]				Visual–Motor Integration[b]				Manual Dexterity							
									Dominant Hand[c]				Nondominant Hand[d]			
	β	Std β	*SE*	*p*	β	Std β	*SE*	*p*	β	Std β	*SE*	*p*	β	Std β	*SE*	*p*
Genotype	−0.12	−0.02	0.38	.758	−1.44	−0.06	1.48	.331	0.08	0.02	0.21	.715	0.15	0.04	0.22	.501
Sex	−1.24	−0.24	0.38	**.001**	—	—	—	—	−0.60	−0.15	0.20	**.003**	—	—	—	—
BSMSS score	0.04	0.20	0.02	**.006**	0.17	0.18	0.06	**.003**	—	—	—	—	—	—	—	—
Daytime oxygen saturation	0.43	0.15	0.21	**.042**	—	—	—	—	—	—	—	—	—	—	—	—
Age at evaluation	—	—	—	—	−0.71	−0.24	0.17	**<.001**	—	—	—	—	—	—	—	—

Note. Values in bold are demographic and medical factors statistically significant at $p < .05$. Dashes indicate that values were not reported because the factor did not remain in the model. SCD genotype was included in each final model based on previous literature. BSMSS = Barratt Simplified Measure of Social Status.

[a]Scaled score from Coding subtest of the Wechsler Intelligence Scale for Children, Fourth or Fifth Edition, or the Wechsler Adult Intelligence Scale, Fourth Edition. Scaled scores have $M = 10$ ($SD = 3$). Final model: Coding ~ genotype + sex + BSMSS + oxygen saturation.

[b]Standard score from the Beery–Buktenica Developmental Test of Visual–Motor Integration, Sixth Edition; Standard scores have M = 100 ($SD = 15$). Final model: VMI ~ genotype + BSMSS + age at evaluation.

[c]Z score from the Grooved Pegboard Test. Z scores have $M = 0$ ($SD = 1$). Final model: dominant hand score ~ genotype + sex.

[d]Z score from the Grooved Pegboard Test. Z scores have $M = 0$ ($SD = 1$). Final model: nondominant hand score ~ genotype.

fine motor skills to scholastic achievement, early intervention and access to supportive services to develop age-appropriate fine motor skills may facilitate improved academic outcomes in this population.

Youth with SCD demonstrated deficits in visual–motor integration and manual dexterity, along with an even greater deficit in graphomotor speed performance compared with normative expectations. These findings are consistent with literature that has identified poorer fine motor outcomes among children with SCD, including global fine motor skills (Schatz & Roberts, 2007) and visual–motor integration (Brown et al., 2000; Burkhardt et al., 2017; Gentry et al., 1997; Gold et al., 2008; Kral et al., 2006; Schatz et al., 2001). Our results extend these findings to suggest that children with SCD demonstrate deficits across numerous domains of fine motor development, rather than just one. Furthermore, we found that fine motor skills were sensitive to the effects of socioeconomic status and male sex. Consistent with expectations, lower socioeconomic status was associated with slower graphomotor speed and weaker visual–motor integration skills, which aligns with prior literature on the impact of environmental enrichment on functional outcomes (Drazen et al., 2016; Tarazi et al., 2007). These findings suggest that there are groups of individuals with SCD who have a greater likelihood of requiring intervention services, specifically individuals with higher social vulnerability and more barriers to accessing health care. Social vulnerability status may help to identify individuals at increased risk for fine motor difficulties and may inform clinical decision making among health care providers to facilitate referral for evaluation or intervention.

Male sex was also associated with weaker dominant hand manual dexterity and slower graphomotor speed, compared with female sex. Prior literature investigating the impact of sex on fine motor outcomes in other populations has been mixed, although some studies have reported that males exhibit weaker visual–motor integration in comparison with females (Coallier et al., 2014; Memišević & Hadzic, 2013). These sex differences have been attributed to both biological (e.g., muscle development and approach to visual–motor tasks; Giammarco et al., 2016) and environmental (e.g., gender normative play; Dinkel & Snyder, 2020) factors, with females having an early advantage in fine motor skills (Escolano-Pérez et al., 2021; Reikerås et al., 2017). Our study extends these findings to show that, specifically in the population with SCD, male sex is associated with weaker manual dexterity and slower graphomotor speed.

Similar to previous studies, we found a negative association between age and fine motor performance, with older youth displaying weaker visual–motor integration (Kral et al., 2006; Schatz & Roberts, 2007). This pattern may be reflective of the cumulative and progressive nature of SCD in neurocognitive deterioration. Lower oxygen saturation was associated with slower graphomotor speed, likely indicative of disease

Table 4. Effect of Fine Motor Skills on Academic Performance

Effect	Math Fluency[e]				Letter-Word Identification[f]			
	β	Std β	*SE*	*p*	β	Std β	*SE*	*p*
Graphomotor speed[a]	3.35	0.58	0.37	**<.001**	1.82	0.33	0.40	**<.001**
Visual motor integration[b]	0.30	0.22	0.11	**.009**	0.44	0.44	0.07	**<.001**
Manual dexterity, dominant hand[c]	2.78	0.36	0.58	**<.001**	1.23	0.14	0.64	.055
Manual dexterity, nondominant hand[d]	2.15	0.28	0.62	**.001**	1.43	0.19	0.57	**.013**

Note. Values in bold are associations between fine motor and academic performance statistically significant at $p < .05$. BSMSS = Barratt Simplified Measure of Social Status.

[a]Scaled score from Coding subtest from the Wechsler Intelligence Scale for Children, Fourth or Fifth Edition, or the Wechsler Adult Intelligence Scale, Fourth Edition. Scaled scores have $M = 10$ ($SD = 3$). Final models: Math Fluency ~ Coding + genotype + age at evaluation + Social Vulnerability Index, Letter–Word Identification ~ Coding + genotype + age at evaluation + BSMSS + social vulnerability index.

[b]Standard score from Beery–Buktenica Developmental Test of Visual–Motor Integration, Sixth Edition. Standard scores have $M = 100$ ($SD = 15$). Final models: Math Fluency ~ visual–motor integration + genotype + age at evaluation + Social Vulnerability Index, Letter–Word Identification ~ visual–motor integration + genotype + age hydroxyurea initiated + BSMSS.

[c]*Z* score from Grooved Pegboard Test. *Z*-scores have $M = 0$ ($SD = 1$). Final models: Math Fluency ~ dominant hand score + genotype + age at evaluation + Social Vulnerability Index, Letter–Word Identification ~ dominant hand score + genotype + age at evaluation + BSMSS.

[d]*Z* score from Grooved Pegboard Test. *Z* scores have $M = 0$ ($SD = 1$). Final models: Math Fluency ~ nondominant hand score + genotype + age at evaluation + Social Vulnerability Index, Letter–Word Identification ~ nondominant hand score + genotype + age at evaluation + BSMSS.

[e]Standard score from Math Fluency subtest of the Woodcock–Johnson Test of Achievement, Third or Fourth Edition.

[f]Standard score from Letter–Word Identification subtest of the Woodcock–Johnson Test of Achievement, Third or Fourth Edition.

severity and chronic oxygen deficiencies. The graphomotor speed task used in the current study required a high cognitive processing demand, a cognitive skill that is particularly vulnerable to the individual disease-related impacts of SCD (Prussien et al., 2019; Stotesbury et al., 2018). Thus, the deficits observed in the current study likely represent the combined effects of graphomotor and cognitive processing speed.

Regarding academics, fine motor skills robustly predicted functional academic outcomes. Manual dexterity, visual–motor integration, and graphomotor speed were associated with math fluency and word-reading performance. However, after controlling for general intelligence, fine motor skills were more consistently associated with math skills rather than with word reading. Functional math skills were assessed with a timed math fluency task, which has a higher fine motor demand compared with a typical untimed calculation task. As such, the observed relationship between fine motor performance and math fluency is likely related in part to graphomotor demand. At baseline, children with SCD typically demonstrate below-grade-level academic performance (Brown et al., 2000; Schatz, 2004; Schatz et al., 2001; Wang et al., 2001), and the present results suggest that fine motor difficulties may represent an additional barrier to scholastic achievement (Knight et al., 2021; Newby et al., 2018).

Limitations and Future Directions

Several limitations of the current study should be considered. We did not include a demographically matched or sibling control group; as such, it is uncertain how demographic factors may have contributed to the observed differences between our SCD population and normative expectations. Detailed information regarding prior occupational therapy services was not available. Future research should include additional intervention data to further elucidate the relationship between fine motor skills and academic outcomes, as well as the effect of demographic and medical factors on fine motor outcomes. Fine motor outcomes were measured quantitatively, and future research should also examine how functional fine motor skills may affect adaptive functioning outcomes in populations with SCD using additional assessment measures. Future studies should examine the longitudinal trajectory of fine motor outcomes in adults with SCD. Additional neurocognitive domains that may have affected performance, such as processing speed, were not fully evaluated or accounted for in our analyses. Thus, it is difficult to isolate the impact of SCD on fine motor performance. Deficits in fine motor performance are apparent, but additional information regarding related neurocognitive performances could likely provide further context for tailoring occupational therapy services. Additionally, not all individuals in our study received neuroimaging. Therefore, the potential impact of cerebrovascular events (e.g., silent cerebral infarcts) on fine motor functioning is unknown.

Implications for Occupational Therapy Practice

The findings of this study have the following implications for occupational therapy practice:

- Diagnosis of SCD places an individual at risk for fine motor and functional difficulties that could benefit from occupational therapy services.

Individuals with SCD who are of lower socioeconomic status, male sex, or older age or who have lower oxygen saturation have even greater risk of fine motor difficulties, and these variables may help inform clinical decision making for referral to evaluation or intervention.
- Children with SCD should be consistently screened in academic settings for deficits affecting occupational performance.
- Measures such as the Beery VMI can be used to carefully screen individuals with SCD. Precision drawing apps administered on a tablet, such as the Standardized Tracing Evaluation and Grapheme Assessment, may also allow for increased access to these types of assessments (Philip et al., 2023) to determine whether occupational therapy services are needed.

Conclusion

In this study, children with SCD exhibited lower-than-expected fine motor performance across multiple motor domains, and these deficits were associated with poorer academic outcomes. Given that the attainment of adequate fine motor skills is essential to any child's development, both adaptively and academically, adequate screening, referral, and implementation of early intervention support for the population with SCD becomes especially important.

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Lakia Kearson, PsyD, is Neuropsychology Fellow, Department of Psychology and Biobehavioral Sciences, St. Jude Children's Research Hospital, Memphis, TN.

Christina Dandar, MA, is Neuropsychology Intern, Department of Psychology and Biobehavioral Sciences, St. Jude Children's Research Hospital, Memphis, TN.

Catherine Hoyt, PhD, OTD, FAOTA, OTR/L, is Assistant Professor of Occupational Therapy, Neurology, and Pediatrics, Department of Pediatrics and Medicine, School of Medicine, Washington University in St. Louis, St. Louis, MO.

Jennifer Longoria, PhD, is Neuropsychologist, Department of Psychology and Biobehavioral Sciences, St. Jude Children's Research Hospital, Memphis, TN.

Victoria Okhomina, MPH, MS, is Biostatistician, Department of Biostatistics, St. Jude Children's Research Hospital, Memphis, TN.

Darcy Raches, PhD, is Neuropsychologist, Department of Psychology and Biobehavioral Sciences, St. Jude Children's Research Hospital, Memphis, TN.

Brian Potter, PhD, is Neuropsychologist, Department of Psychology and Biobehavioral Sciences, St. Jude Children's Research Hospital, Memphis, TN.

Guolian Kang, PhD, is Biostatistician, Department of Biostatistics, St. Jude Children's Research Hospital, Memphis, TN.

Jane Hankins, MD, is Director of Global Hematology Program, Department of Hematology, St. Jude Children's Research Hospital, Memphis, TN.

Clifford Takemoto, MD, is Director of Clinical Hematology, Department of Hematology, St. Jude Children's Research Hospital, Memphis, TN.

Andrew Heitzer, PhD, is Neuropsychologist, Department of Psychology and Biobehavioral Sciences, St. Jude Children's Research Hospital, Memphis, TN; andrew.heitzer@stjude.org