

Relations between fine motor skills and parental report of attention in young children with neurofibromatosis - type 1

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Background

Neurofibromatosis type 1 (NF1) is one of the most common genetic disorders presenting in approximately 1 in 3,000 live births. NF-1 is a highly variable condition with a large number of medical complications. While physical medical features are indeed problematic for some with NF1, the most common complaints from parents of children with NF1 are not medical in nature, but rather neuropsychological and behavioral.

Children with NF1 are much more likely than the general population to receive interventions for learning, behavior, speech and motor problems in school (Krab et al., 2008). Research has suggested that school-age children with NF1 have poorer performance in fine motor functions than their siblings who are unaffected (Hyman et al., 2006; Chapman et al., 1996; Hofman et al., 1994). Additionally, children with NF1 are a greater risk for attention difficulties; as many as one-third to one-half of school-age children with NF1 meet diagnostic criteria for ADHD (Hachon et al., 2011).

Considering the prevalence of both fine motor and attention difficulties in older children with NF1, it is important to further investigate and characterize these difficulties in young children. Additionally, there is very little research examining the relations between fine motor and attention difficulties in young children more generally.

Methods

Participants

Thirty-eight children with NF1 recruited through Midwestern Neurofibromatosis clinics, and 23 unaffected children (siblings and community members) ages 4 through 6 years were enrolled in the study. There was no significant difference between groups in age ($p = .511$), sex ($p = .619$), ethnicity ($p = .423$), or SES ($p = .149$).

Measures

- o *The Differential Ability Scale (DAS-II)*: The General Conceptual Ability (GCA) score was used to assess intellectual functioning and the Nonverbal Abilities (NV) score was used to assess nonverbal reasoning skills. The Copying subtest was used to assess fine motor abilities.
- o *NEPSY – Second Edition (NEPSY-II)*: The Fingertip Tapping (Repetitions and Sequences – FTT), Imitating Hand Positions (IHP), and Visuomotor Precision (VMP) subtests was used to examine fine motor abilities.
- o *Scales of Independent Behavior – Revised (SIB-R)*: The Motor Skills domain was used to examine parent-reported motor functioning.
- o *Conners' Parent Report Rating Scales – Revised: Short Form (Conners)*: The Hyperactivity, Cognitive Problems/Inattention, Opposition, and ADHD Index scores were used to assess attention.

Fine motor tasks were grouped based on their perceived level of complexity and level of required cognitive control, as described as below:

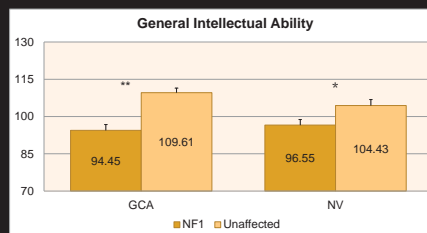
Subtest	Score	Complexity	Score Type	Measures
Fingertip Tapping	Repetitions Combined Score	Simple	Scaled	Fine-motor control and dexterity
	Sequences Combined Score	Mid-level	Scaled	Fine-motor programming
Imitating Hand Positions	Total Score	Mid-level	Scaled	Fine-motor coordination, sensorimotor differentiation
	Combined Score	Mid-level	Scaled	Fine-motor coordination, manual motor speed
Copying	Subtest Score	Complex	T-score	Fine-motor coordination, motor control

Results

General Intellectual Ability

General intellectual functioning ($p < .001$) and Nonverbal Abilities ($p = .026$) were significantly different between groups.

- o There was no significant age or gender effects on GCA or NV.



Given that the GCA is a composite that includes the Copying subtest score the NV score was used as a covariate in the analysis of variables that may account for differences in fine motor ability scores.

Fine Motor Skills

Group differences and descriptive statistics on selected DAS-II and NEPSY-II motor subtests are presented below:

Subtest	NF1			Unaffected			Group Differences	
	N	Mean	SD	N	Mean	SD	Sig	d
Fingertip Tapping								
Repetitions	25	10.04	2.70	16	11.25	2.32 +		0.48
Sequences	25	8.32	3.44 +	16	9.75	2.93		0.45
Imitating Hand Positions	38	7.11	2.02 ++	23	8.87	1.93 ++	***	0.90
Visuomotor Completion	38	7.79	2.83 ++	23	9.96	4.06	**	0.66
Copying	38	41.84	8.52 ++	23	52.74	7.99	***	1.33
Fine Motor Scale SIB-R	38	Raw = 34.03	5.86	23	Raw = 37.22	5.47	*	0.57

Significantly different from normative data in one-sample t-test + $p < .01$; ++ $p < .001$
 Significant group differences * $p < .05$; ** $p < .01$; *** $p < .001$

Results (cont)

Fine Motor Skills (cont)

As the groups differed in nonverbal abilities, a MANCOVA was performed to examine group differences in motor skills while statistically controlling for group differences in NV.

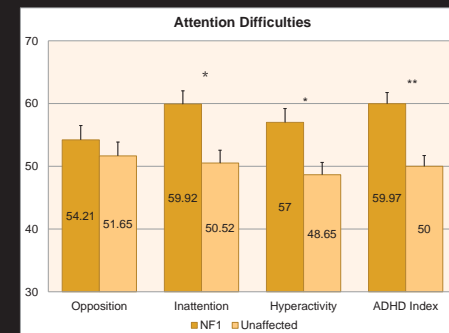
- o Copying and IHP showed group differences even after nonverbal reasoning abilities were taken into account.

Parent Measure of Fine Motor Functioning

An independent t-test indicates there was a significant difference in FM scores for children with NF1 and unaffected children ($p = .039$) on the SIB-R Fine Motor Scale.

Parental Report of Attention Difficulties

Descriptive statistics and group differences on the Conners' Parent Rating Scale are presented below:



Relations between Fine Motor Abilities & Attention

Fine motor difficulties were not significantly related to attention problems.

Discussion

The goal of the current study was to provide more information about the early emergence of fine motor difficulties in young children with NF1 and relations with attention difficulties. Fine motor abilities were examined at varying levels of complexity. Results of the current study indicate that:

- o Children with NF1 do exhibit significant difficulties on mid-level and complex fine motor tasks compared to unaffected children.
- o While intellectual functioning level played a role in fine motor abilities, group differences in fine motor abilities persisted even after nonverbal reasoning was taken into account.
- o Fine motor difficulties were also seen based on parental report of fine motor functioning in everyday life. Hence, the difficulties with fine motor activities are pervasive enough to impair performance on adaptive everyday fine motor tasks, not just lab-based measures.
- o No significant relations between fine motor abilities and parental report of attention difficulties were found, such that the fine motor difficulties observed are not merely due to difficulties with attention or impulsivity.

These results suggest that many children with NF-1 may benefit from early interventions that target fine motor skills development. Additional research is needed to further explore fine motor difficulties as a risk factor for later difficulties in learning and attention.