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# Relations Between Fine Motor Skill and Parental Report of Attention in Young Children with Neurofibromatosis Type 1

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RELATIONS BETWEEN FINE MOTOR SKILL AND PARENTAL REPORT OF  
ATTENTION IN YOUNG CHILDREN WITH NEUROFIBROMATOSIS TYPE 1

by

Christy Casnar

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ABSTRACT  
RELATIONS BETWEEN FINE MOTOR SKILL AND PARENTAL REPORT OF  
ATTENTION IN YOUNG CHILDREN WITH NEUROFIBROMATOSIS TYPE 1

by

Christy Casnar

The University of Wisconsin-Milwaukee, 2013  
Under the Supervision of Professor Bonita P. Klein-Tasman

Neurofibromatosis type 1 (NF1) is one of the most common genetic disorders presenting in approximately 1 in 3,000 live births. NF1 is a highly variable condition with a large number of complications. A common complication is neuropsychological problems, including developmental delays and learning difficulties that affect as many as 60% of patients. Research has suggested the children with NF1 often have poorer fine motor skills and are at greater risk for attention difficulties than the general population. Furthermore, recent research is beginning to demonstrate a relationship between fine motor skills and attention in older children; however, very little research has examined this relationship in young children. Thirty-eight children with NF1 and 23 typically developing children between the ages of 4 and 6 are enrolled in the study. Varying levels of fine motor functioning were examined (simple, mid-level, and complex fine motor tasks). For children with NF1, significant difficulties were demonstrated on lab-based, mid-level and complex fine motor tasks, even after controlling for nonverbal reasoning abilities. These findings suggest that children with NF1 do not differ significantly from TD children on lab-based, simple fine motor tasks. Additionally, these findings were corroborated by parental report of difficulties in adaptive fine-motor functioning. The current study also examined relations between fine motor ability and parental report of

attention difficulties. No significant correlations were found between complex fine motor ability and attention difficulties. This study provides much needed descriptive data on the early emergence of fine motor difficulties in young children with NF1. The results can help guide further research into potential early intervention programs that may be able to improve overall motor and possibly attention function in children with NF1.

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## **Introduction**

Neurofibromatosis type 1 (NF1) is one of the most common genetic disorders presenting in approximately 1 in 3,000 live births (North et al., 1998). NF1 is a highly variable condition with a large number of complications. A common complication is neuropsychological problems, including developmental delays, learning difficulties and attention problems that affect as many as 60% of patients. Children with NF1 are also at much higher risk for receiving interventions for learning, behavior, speech and motor problems in school (Krab et al., 2008). Research has suggested the children with NF1 are a greater risk for attention difficulties than the general population; it had been reported that as many as one-third to one-half of children with NF1 meet diagnostic criteria for ADHD (Hachon et al., 2011). Additionally, children with NF1 are reported to 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). Considering the prevalence of both attention and fine motor difficulties in older children with NF1, it is important to further investigate and characterize these difficulties in young children. Furthermore, recent research is beginning to demonstrate a relationship between fine motor skills and attention in older children; however, very little research has examined this relationship in young children. By exploring the relationship between fine motor abilities and attention in young children with NF1, we may learn more about the exact nature of that relationship.

This introduction will examine the current literature on the fine motor abilities and attention difficulties often reported in children with NF1. First, I will provide general background information about Neurofibromatosis type 1. I will briefly describe medical



features and the common cognitive and behavioral characteristics of individuals with NF1. Second, I will discuss current research describing attention difficulties often found in individuals with NF1. Third, I will review the current literature on the fine motor abilities of children with NF1. Fourth, current research examining the relationship between fine motor abilities and attention will be explored. Finally, I will outline the research goals, hypotheses, and procedures of the proposed investigation.

### **Neurofibromatosis Type 1**

Neurofibromatosis type 1 is an autosomal dominant disorder with an incidence of approximately 1 in 3,500 live births (Huson & Hughes, 1994; North et al., 1997). NF1 is caused by a mutation of a gene on the long arm of chromosome 17, which is responsible for encoding neurofibromin. As mentioned above, NF1 has an autosomal dominant pattern of inheritance, although, half of all cases result from a spontaneous mutation. Diagnosis requires the presence of two or more of the following criteria: (1) 6 or more café au lait spots, (2) axillary or inguinal freckling, (3) 2 or more cutaneous neurofibromas, (4) 1 plexiform neurofibroma, (5) 2 or more iris Lisch nodules, (6) an optic glioma, (7) a characteristic body lesion, or (8) first degree relative with NF1 (NIH Consensus Development Conference, 1988). The most common manifestations to first appear in childhood are café au lait spots and axillary freckling. A study of children with NF1 by Cnossen et al. (1998) reported that as many as 96.7% of children diagnosed with NF1 displayed six or more café au lait spots by 3 years of age. Freckling was found in 85.3% of children with NF1 by the 4 years of age (Cnossen et. al, 1998). Recent advances in genetic testing have made it possible to confirm diagnosis in approximately 95% of individuals with NF1 (Tonsgard, 2006).

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 at higher risk for cognitive problems, as well as learning and attention difficulties. A recent review of the literature by Hachon, Iannuzzi and Chaix (2011) reports that although NF1 children's cognitive abilities fall in the average range, the IQ curve is shifted to the left (with a mean around 90) when compared to unaffected children. Additionally, approximately 50% of children with NF1 have a learning disability, with variability in the nature of the learning disability (Brewer et al., 1997). Current literature suggests that up to half of children with NF1 have visuospatial ability difficulties, with performance falling one standard deviation or more below population norms even when controlling for IQ and attention difficulties (Hyman et al., 2005; Levine et al., 2006; Schrimsher et al., 2003). Fine motor coordination impairment and low motor speed have also been reported in 20-30% of children with NF1 (Hyman et al., 2005; Levine et al., 2006). Finally, Hachon et al. (2011) report that 30-50% of children with NF1 meet the diagnostic criteria for ADHD.

### **Attention in Children with NF1**

ADHD is the most commonly diagnosed psychological disorder in childhood and children with NF1 are about 3 times more likely to be diagnosed with ADHD than their unaffected siblings (Diamond, 2005; Hyman et al., 2005). These high rates of attention difficulties in the NF1 population make it one of the hallmark cognitive/behavioral impairment of the disorder, although the exact relationship between NF1 and ADHD remains unclear. Several studies have examined the behavioral difficulties related to ADHD in older children with NF1 (Hyman et al., 2005; Koth et al., 2000; Kayl & Moore,

2000). Data obtained from parents and teachers suggest that difficulties with concentration, inattention, impulsivity and hyperactivity are more common in children with NF1 than their unaffected siblings (Dilts et al., 1996; Koth et al., 2000; Hoffman et al., 1994; Mazzocco et al., 1995). Some researchers also report that these symptoms may hinder psychosocial and academic success more generally (Kayl & Moore, 2000). More specifically, recent research suggests that children with attention difficulties (e.g., ADHD) show significantly lower social functioning, based on parent and teacher report, than children without attention difficulties (Greene et al., 1996; Hinshaw, 2002; McConaughy, Volpe, Antshel, Gordon & Eiraldi, 2011). This finding was substantiated by Barton and North (2004) in a sample of 79 children with NF1 between the ages of 8 and 16. These authors found that while, overall, children with NF1 had significantly poorer social skills than their unaffected siblings, the presence of ADHD was the major risk factor for poor social functioning (Barton & North, 2004).

There is limited research examining specific aspects of attention affected in NF1. A recent research review by Templer, Titus and Gutmann (2012) examined the current literature on attention problems and executive function in NF1, concluding that inhibition is the most frequently demonstrated deficit in children with NF1 with and without ADHD. In particular, Hyman et al. (2005) found deficits in sustained attention, even when controlling for intellectual functioning, in children with NF1. Hyman et al. (2005) reported that 54% of children with NF1 scored at least one standard deviation below their unaffected siblings and 63% scored at least one standard deviation below the normative mean on a lab based measure of sustained attention. With regards to working memory, Huijbregts et al. (2010) revealed that children with NF1 tend to make more mistakes as

demands on working memory increases. Additionally, Huijbregts et al. (2010) found, in a cross-sectional developmental study, that impairments in working memory do not diminish with age and are observed in many adults with NF1. Cognitive flexibility has also been shown to be impaired in children with NF1 when compared to age-matched controls. Rowbotham et al. (2009) found, in a sample of adolescents, that as the demand for cognitive flexibility increased on tasks, children with NF1 demonstrated greater impairment than controls.

A recent study by Sangster, Shores, Watt and North (2011) included assessment of the attention abilities of young children with NF1, while examining their broader cognitive and behavioral functioning. The performance of 26 children with NF1, ages 4 through 6, were compared to 21 control children, comprised of siblings and typically developing children from the community, on measures of sustained attention (K-CPT) and parental report of attention difficulties (CADS). The NF1 group demonstrated poorer performance than controls on omission errors, reaction time errors, and variability. However, group differences were no longer significant after controlling for intellectual functioning. Additionally, parental report of attention problems revealed no differences between groups when controlling for maternal education and intellectual functioning. However, the authors note that, qualitatively, children with NF1 were more likely to be classified as having potential attention difficulties than their peers as measured by the K-CPT (58.8% versus 15%) and were more likely to be rated as inattentive by their parents than controls (Sangster et al., 2011).

Generally, there is fairly compelling evidence supporting the presence of attention difficulties in older children and adults with NF1 (Kayl & Moore, 2000; Koth et al.,

2000; Mautner et al., 2002; North et al., 2002; Huijbregts et al., 2010; Templer et al., 2012) and mixed support for attention difficulties in young children with NF1 (Sangster et al., 2011; Legius et al., 1994). Studies examining attention abilities in young children with NF1 suffer from several limitations. Sample sizes are often small and may make it difficult to detect significant differences between groups. In the Sangster et al (2011) study, the comparison group differed significantly on IQ and maternal level of education. While the authors were able to control for those variables statistically, areas of subtle difficulty may have been masked. Additionally, given the limited number of studies reporting attention abilities in young children with NF1, it is difficult to determine if results would generalize to a larger sample. Therefore, research that specifically examines attention difficulties in young children with NF1 is much needed.

### **Fine Motor Abilities in Children with NF1**

Previous research on fine motor abilities in NF1 has focused on examining difficulties mainly in school-aged children. Descheemaeker et al. (2005) found significant impairment on complex psychomotor tasks in a sample of children with NF1 between the ages of 7 and 11. Additionally, Gilboa et al. (2010) analyzed the process and product of handwriting among 8 to 16 year old children with NF1. Results from this study showed significant differences between the NF1 group and a typically developing control group. Johnson et al. (2010) recently examined motor ability in 26 NF1 children between the ages of 4 and 15, and found significantly lower scores than the normative sample for the Total Motor Composite within the Bruininks-Oseretsky Test Version 2 (BOT-2). Additionally, large effect sizes were found for fine motor precision, upper limb coordination, bilateral coordination, balance, run speed/agility and strength. Several other

studies examining the cognitive and behavioral profile of school-aged children with NF1 have demonstrated fine motor difficulties (Billingsley et al., 2003; Hofman et al. 1994; Hyman et al., 2005; Levine et al., 2006).

Understanding early motor difficulties in young children with NF1 may help predict later difficulties in fine motor, visual-motor integration and fine motor coordination. A recent study published by Sangster, Shores, Watt and North (2011) included examination of fine motor abilities in 26 children with NF1 and 21 typically developing preschoolers using the Beery Visual Motor Integration Test (VMI), as part of a larger study of the cognitive profile of preschool-aged children with NF1. Contrary to the findings in the literature of school-aged children, Sangster et al. (2011) did not find any significant differences between groups on the VMI once intelligence and maternal level of education were controlled for statistically. However, as mentioned above, these results should be interpreted with caution since the groups were not well matched on important variables, such as age, which may have obscured group differences. In fact, considering that the contrast group was approximately 6 months younger than the NF1 group, the findings provide suggestive evidence that young children with NF1 are demonstrating difficulties with fine motor and are performing similarly to children who are 6 months younger. Therefore, these findings highlight the importance of examining fine motor difficulties more specifically, with a well-matched control group, in young children with NF1.

Only one additional study has specifically focused on motor abilities in young children with NF1. Lorenzo, Barton, Acosta & North (2011) analyzed motor functioning using the Bayley Scales of Infant Development, Second Edition (BSID-II) in 39 toddlers

(aged 21-30 months) with NF1 compared to 42 control children matched by age and mother's years of education. The motor scale on the BSID-II examines both gross and fine motor abilities, such as crawling, walking, scribbling and manipulation of small objects. Their study found that toddlers with NF1 had significantly poorer motor skills compared to control children. Difficulties included delays in fine motor and gross motor development. Lorenzo et al. state that "monitoring children's fine and gross motor milestones is imperative...and may be 'red flags' that warrant clinical attention and further investigation." One major limitation of this study is that because the BSID-II does not produce separate scores for fine motor and gross motor development, it is not possible to distinguish between fine and gross motor impairment in young children with NF1.

Overall, there is very little research focused specifically on the fine motor characteristics of young children with NF1. There is compelling evidence that fine motor difficulties, especially for more complex fine motor tasks, are present for many school-aged children with NF1 (Billingsley et al., 2003; Hofman et al. 1994; Hyman et al., 2005; Levine et al., 2006). However, the vast majority of these studies utilizes small sample sizes and only examines fine motor abilities using one or two measures, usually, within the larger context of a general cognitive and neuropsychological evaluation. Future research that explores both simple and complex fine motor skills would help identify more precisely where the difficulties with fine motor abilities lie. The Johnson et al. (2010) study provides a peek into early fine motor abilities in young children with NF1; however, a more thorough look at these abilities in a cross-sectional design with a control group is needed.

## **Fine Motor Abilities and Attention**

Recent studies have begun to examine the relationship between fine motor abilities and attention in childhood by studying the fine motor abilities of children with ADHD. Motor problems, including delays in achieving motor milestones and problems with motor planning, execution and coordination, are frequent coexisting problems for many children with ADHD (Kalff et al., 2002; Piek & Dyck, 2004). As many as 30% to 50% of children with ADHD suffer from co-occurring motor difficulties (Fliers et al., 2008). Several studies have sought to explore the relationship between fine motor and attention using correlational study designs (Tseng et al., 2004; Whitmont & Clark, 1996; Marcotte & Stern, 1997; Piek et al., 1999). A study by Whitmont and Clark (1996) reported significantly lower fine motor scores for 9-year-old children with ADHD than controls using the Fine-Motor Composite of the Bruininks-Oseretsky Test of Motor Proficiency (BOTMP). Marcotte and Stern (1997) reported similar findings of impairment on a measure of graphomotor abilities in a group of 80 children with ADHD ages 8 through 13. Tseng, Henderson, Chow and Yao (2004) examined fine motor abilities (using the BOTMP), teacher and parent report of attention difficulties, and sustained attention and impulse control (using Gordon Diagnostic System) in 84 children with and without ADHD. Groups were matched for age, sex and handedness and included children between the ages of 6 and 11 years. Results revealed significant group differences on measures of attention and visuomotor control (Tseng et al., 2004). Fine motor abilities were significantly correlated with impulse control, sustained attention and impulsivity. Regression analysis indicated that sustained attention and impulse control were the best predictors of fine motor skills and accounted for 45.7% of the variance



(Tseng et al., 2004). Fine motor skills have also been associated with severity of ADHD symptoms (Whitmont & Clark, 1996; Doyle et al., 1995; Piek et al., 1999).

It has been suggested that this relationship between fine motor and attention can be interpreted in two ways. First, it is possible that poor inhibitory control and attention interferes with fine motor abilities. This would be consistent with Barkley's (1997) proposed model that suggests poor inhibitory control is a core deficit in ADHD that affects other associated abilities, such as fine motor skill. Therefore, deficits in fine motor abilities that are often found in ADHD would be a direct result of deficits in inhibitory control. A few studies have substantiated this theory by finding no significant difference in simple fine motor abilities in children with and without ADHD, when using tasks such as fingertip tapping or grooved pegboard where inhibitory control demands are low, but reporting significant differences on more complex fine motor tasks that require better attention and self-regulation (Grodzinsky & Diamond, 1992; Leung & Connolly, 1998; Mariani & Barkley, 1997).

Alternatively, a second interpretation of the relationship between fine motor difficulties and attention problems would suggest that ADHD and fine motor deficits are co-occurring problems that may share a common neural connectivity, possibly through the cerebellum, or may be under the influence of a shared dopamine system that would hinder both abilities if impaired. A study by Flapper, Houwen and Schoemaker (2006) recently supported this theory by examining the effects of stimulant medication on fine motor abilities of children with ADHD and Developmental Coordination Disorder (DCD) compared to an age- and sex-matched control group. Substantial improvements in fine motor abilities were seen in children with ADHD-DCD on stimulant medication.

However, the authors report that some fine motor problems still remained after inattention and hyperactivity symptoms had been ameliorated by medication. Therefore, Flapper et al. (2006) propose that attention and fine motor difficulties are comorbid conditions that may share a common neural underpinning, but are two distinct problems (Flapper et al., 2006).

Most studies examining the relationship between fine motor abilities and attention abilities are conducted with school-aged children. Only one study specifically examines both simple and complex fine motor functioning in young children with ADHD. Kalff et al. (2003) compared the performance of 5-6 year old children at risk for ADHD on a low-level fine motor control task (Tracking task), high-level fine motor control task (Pursuit task), and movement speed task (Purdue Pegboard). Children at risk for ADHD (and confirmed at a 1.5 year follow-up) were compared to typically developing control children, children with 'borderline ADHD', and children with other pathological disorders. Results show that 5-6 year old children who were later diagnosed with ADHD were less accurate and more variable on both tasks examining fine motor control. There were no differences between groups on the movement speed task. Kalff et al. (2003) suggested that "young children at risk for ADHD have a more pronounced deficit in movement patterns when task demands are high, demonstrating a specific deficit in higher-order controlled processes" (pg 1054).

Few other studies have systematically examined the level of complexity needed to complete different fine motor tasks; however, there are many benefits from examining fine motor abilities in this way. As proposed by Barkley (1997), breaking down fine motor tasks into different classifications depending on their level of executive control can

help disentangle where the primary deficit in fine motor difficulties lie. Two other studies have alluded to the importance of studying fine motor difficulties at varying complexity levels, including studies by Kroes et al. (2002) and Egeland, Ueland, and Johansen (2012). As Kroes et al. (2002) highlighted in their study of quantitative and qualitative aspects of fine motor performance in preschooler with ADHD, qualitative aspects of motor skills (e.g. quality of fine motor control and coordination) were predictive of later development of ADHD, whereas quantitative performance (e.g. simple motor speed) was not. Additionally, a recent article by Egeland et al. (2012) found a difference in performance on complex, but not simple, fine motor tasks in the different ADHD subtypes.

In sum, there is some compelling evidence suggesting a relationship between fine motor abilities and attention, however this research is still in its infancy. The theoretical model underlying the co-occurrence of attention and fine motor skills deficits is in need of further research. Most studies examining this relationship are correlational in nature and use differing experimental methods to conceptualize and measure fine motor and attention. While the design of the proposed study will not allow for the direct analysis necessary to compare the theoretical models presented above, the proposed study will better characterized the specific components of fine motor skills affected in NF1 and their relation to attention abilities in young children with and without NF1.

### **Summary**

Previous research has focused on behavioral and cognitive impairments of school-aged children with NF1 (Descheemaeker et al., 2005; Huijbregts et al., 2010; Hyman et al., 2005). Currently, there is very little research analyzing the fine motor abilities of

young children with NF1. Additionally, there is very little research examining the relationship between fine motor and attention difficulties in young children more generally. By better understanding the characteristics of the early fine motor abilities and parental report of early attention difficulties in young children with NF1, we may be able to better understand the relationship between fine motor abilities and attention. The proposed study aims to explore those abilities and relationships and provide much-needed descriptive data. Additionally, given that fine motor difficulties are often noticed at an earlier age than attention difficulties, if relations between fine motor abilities and attention are observed, this research would provide an important step toward a contention that early intervention programs developed to improve fine motor abilities may also lower the risks for children to develop attention difficulties over time.

### **Hypotheses**

#### *Hypothesis 1*

*Group differences in complex fine motor abilities but not simple fine motor abilities are expected. A larger proportion of children with NF1 will display difficulties in complex fine motor abilities but not simple fine motor abilities on lab-based measures than typically developing children.*

#### *Hypothesis 2*

*Group differences in parental report of fine motor abilities are expected. A larger proportion of children with NF1 will display deficits in fine motor abilities as measured from parental report compared to typically developing children.*

### *Hypothesis 3*

*A positive relationship will exist between lab-based measures and parental report of fine motor difficulties.*

### *Hypothesis 4*

*A positive relationship will exist between lab-based measures of complex fine motor difficulties and parental report of inattention and hyperactivity.*

## **Methods**

### **Participants**

Participants include 38 children diagnosed with NF1 and 23 typically developing children between the ages of 4 and 6 years. The children with NF1 diagnoses were based on the NIH Consensus Conference criteria (NIH, 1988) by a physician specializing in such diagnoses. The typically developing group consists of children recruited from the community and siblings of children in the NF1 group. 59% of the sample was male (36 males, 25 females). 48 of the participants were Caucasian (79%), 4 were African American (7%), 5 were Hispanic (8%), 2 were Asian (3%), and 2 were of mixed ethnicity (3%). Children who did not speak English were excluded from the study.

### **Procedures**

Participants with NF1 and their siblings were recruited through the Neurofibromatosis Clinics at the Children's Hospital of Wisconsin, Medical College of Wisconsin and University of Chicago Hospitals. Additional typically developing children were recruited using flyers that were posted and handed out at preschools, daycare centers and public libraries in the community.

Participants completed the assessment either at the Child Neurodevelopment Research Lab (CNRL) at the University of Wisconsin-Milwaukee or at the Pediatric Neuropsychology Clinic (PNC) at the University of Chicago Hospitals. Each child was administered age appropriate norm-referenced neuropsychological assessments by a trained member of the study team. Parents were interviewed about adaptive behavior using the Scales of Independent Behavior, Revised (excluding Maladaptive Behavior section) during their child's assessment in an adjacent room.

### **Measures**

All measures chosen have been developed for use with children from ages 4 to 6 and are widely used in preschool and school aged assessment and research. All neuropsychological measures are norm-referenced and have demonstrated strong psychometric properties, including good reliability and validity. These measures have been used with both typically developing children and children with a variety of developmental disorders. These measures were selected to pick up on subtle impairment that is commonly found in young children with NF-1.

*Differential Ability Scales-Second Edition: Early Years Form (DAS-II; Elliot, 1990).* The DAS-II is a commonly used, comprehensive measure of cognitive abilities for children ages 2-6 to 17-11. The DAS-II is empirically derived and demonstrates excellent internal consistency, test re-test reliability and correlates highly with other commonly used measures of cognitive abilities. The DAS-II provides good normative data collected on a large representative national sample and contains excellent floor and ceiling levels, making it appropriate for children with neurodevelopmental disorders. This measure yields an overall composite score (GCA) that is equivalent to a full-scale

IQ score. The GCA includes into two cluster scores, including Verbal Abilities and Nonverbal Abilities. In this study, participants will complete the core subtest for the Early Years Form (including Verbal Comprehension, Naming Vocabulary, Picture Similarities, Matrices, and Copying) to yield a GCA, which will be used to assess intellectual functioning generally. Additionally, the DAS-II Copying subtest score will be examined to assess fine motor abilities. The Copying subtest examines visual-perceptual matching and fine motor coordination in copying line drawings. Subtest scores will be reported in t-scores (mean of 50, standard deviation of 10). Given that the Copying subtest score (which is the main measure used for examining complex motor ability in this study) is embedded in the GCA composite score, the DAS-II Nonverbal Abilities (NV) composite score will be used to assess and control for the role nonverbal reasoning ability on all levels of fine motor ability.

*NEPSY – Second Edition (NEPSY-II; Korkman, Kirk, & Kemp, 2007).* The NEPSY-II is a widely used measure that assesses children's performance in areas of six theoretically derived domains, including Attention and Executive Functioning, Language, Memory and Learning, Sensorimotor, Social Perception and Visuospatial function. Administration of selected subtests takes approximately 5-10 minutes and is designed for children 3-16 years old. This measure yields scaled scores (mean of 10, standard deviation of 3) and percentile ranges for each subtest. For this study, performance on the Sensorimotor subtests (Fingertip Tapping, Imitating Hand Positions, and Visual Motor Precision) will be examined.

Fingertip Tapping is a timed subtest comprised of two components in which the child is asked to copy a series of finger motions demonstrated by the examiner as quickly

as possible. The first part (Repetitions) is designed to assess finger dexterity and motor speed. The second part (Sequences) is designed to assess rapid motor programming. This subtest was normed and only administered to 5 and 6 year old children; therefore, participants who were 4 years old did not complete this subtest. Imitating Hand Positions is a subtest designed to assess the ability to imitate hand and finger positions demonstrated by the examiner. Difficulty on this subtest could indicate difficulty with fine-motor coordination and sensorimotor differentiation. Visuomotor Precision is a timed subtest designed to assess graphomotor speed and accuracy by drawing lines inside of tracks as quickly as possible. Both precision and speed are measured. This subtest examines manual fine-motor coordination and manual motor speed. Imitating Hand Positions and Visuomotor Precision was normed for children as young as 3, therefore, all participants will complete both subtests.

*Summary of Fine Motor Measures.* As discussed in the introduction, there are many benefits from examining differing levels of complexity of fine motor tasks. For the purpose of this study, the Fingertip Tapping – Repetitions subtest will be classified as a simple fine motor task. Fingertip Tapping – Sequences, Imitating Hand Positions, and Visuomotor Precision will be classified as mid-level fine motor tasks. Finally, Copying will be classified as a complex fine motor task. These classifications are based off the similar studies within the current fine motor ability literature. Table 1 details the specific fine motor ability scores that will be examined in the NEPSY-2 and DAS-II subtests, as well as, their classified level of complexity.

*Scales of Independent Behavior – Revised (SIB-R; Bruininks, Woodcock, Weatherman, & Hill, 1996).* The SIB-R is a comprehensive, normative referenced



measure of adaptive behavior for children and adults. The SIB-R demonstrates excellent reliability and validity. The SIB-R will be administered in an interview format with the child's parent. Administration takes approximately 40 minutes to assess 14 adaptive behavior domains. This measure yields an overall composite of Broad Independence. Clusters include composite scores in Motor Skills, Social Interaction and Communication Skills, Personal Living Skills, and Community Living Skills. The Motor Skills domain is made up of two subscales including Fine Motor and Gross Motor. For the purpose of this study only the Fine Motor domain score will be analyzed. This will be done using raw scores (range 1 to 57) and skill level categories ("age appropriate and above", "limited to age-appropriate", and "limited"), as no standard scores are yielded for this domain.

*The Conners' Parent Rating Scales—Revised Short Form (Conners; Conners, 2001).* The Conners' is a widely used measure of attention difficulties in children and is often used to assist in the evaluation of ADHD in children. The Conners' was developed for use with children between the ages of 3 and 17 and is completed by parents. It was standardized using over 2,000 children and adolescents and has demonstrated good reliability and validity. The measure includes four indexes, including the Hyperactivity, Cognitive Problems/Inattention, Opposition and ADHD index. This study will analyze the results of all four scales, which are reported in t-scores (mean of 50, standard deviation of 10).

## **Results**

In this section, the data from laboratory-based fine motor abilities and parental report of fine motor ability and attention difficulties are provided. First, descriptive statistics examining group differences in general cognitive functioning will be provided.

Next, performance on lab-based fine motor tasks, examined at three levels of complexity (simple, mid-level, complex), will be provided for both children with NF1 and TD children. Analysis examining the role of nonverbal skill in fine motor ability will also be provided. Next, parental report of fine motor abilities measured using the SIB-R will be reviewed and then compared to performance on lab-based measures of fine motor ability. Finally, relations between fine motor ability and parental report of attention difficulties will be examined.

The data were analyzed using IBM SPSS for Windows, version 20. Findings are interpreted with respect to both statistical significance and effect size. For all analysis, a “difficulty” was operationalized as a score one or more standard deviations below the normative mean. A p-value of .05 was used, but given the small sample size, p-values between .05 and .1 were considered trends and are also reported to decrease the chances of dismissing significant findings because of low power. Interpretations of Cohen’s *d* are as follows: negligible effect = 0 – .14; small effect = .15 – .39; medium effect = .40 – .74; large effect = .75 and above. Spearman’s rho was used when correlational analyses were conducted and interpretations of correlation effect size (Cohen, 1988) are as follows: small = .1 – .3; medium = .3 – .5; large = .5 – 1. The stability of the correlations must be interpreted with caution given the small sample size. Table 2 summarizes the demographic data for each group.

### **General Cognitive Abilities**

The GCA composite score from the DAS-II was used to examine general cognitive functioning. For children with NF1, the mean GCA score fell in the average range ( $M = 94.45$ ,  $SD = 14.33$ ). The mean GCA score for TD children also fell in the

average range ( $M = 109.61$ ,  $SD = 9.02$ ). An independent-samples t-test was conducted to compare the GCA scores for children with NF1 and TD children. There was a significant difference between groups ( $t(59) = -4.54$ ,  $p < .001$ , two-tailed). The Nonverbal Abilities (NV) composite score from the DAS-II was the primary measure used to examine nonverbal reasoning skills. For children with NF1, the mean NV score fell in the average range ( $M = 96.55$ ,  $SD = 13.90$ ), as did the TD children's NV score ( $M = 104.43$ ,  $SD = 11.57$ ). An independent-samples t-test was conducted to compare the NV scores for children with NF1 and TD children. There was a significant difference between groups ( $t(59) = .47$ ,  $p = .026$ , two-tailed). Given that the GCA is a composite that includes the Copying subtest score, the NV score will be used as a covariate in the analysis of variables that may account for differences in fine motor ability scores.

### **Fine Motor Functioning**

**Simple Fine Motor Abilities.** The Fingertip Tapping – Repetitions (FTT) subtest from the NEPSY-II was the primary measure used to examine simple fine motor abilities in young children with and without NF1. The Fingertip Tapping - Repetitions subtest measures finger dexterity and motor speed. Not all children were administered FTT Repetitions because the subtest is only standardized for children 5 and older, therefore, 25 children with NF1 and 16 TD children completed this measure.

For the children with NF1 ( $n = 25$ ), the mean FTT Repetitions score fell in the average range ( $M = 10.04$ ,  $SD = 2.70$ ). One-sample t-tests were conducted to compare the children with NF1's mean scores to the mean from normative data and the rates of difficulties were also examined using frequency analysis. Performance on FTT repetitions was not significantly lower than would be expected based on the normative

data [ $t(24) = 0.07, p = .942$ ]. Three children (12%) had FTT Repetition scores one or more standard deviations below the mean. For 16 TD children, the mean FTT Repetitions score also fell in the average range ( $M = 11.25, SD = 2.32$ ). One-sample t-tests were conducted to compare the TD children's mean scores to the mean from normative data and the rates of difficulties were also examined using chi-square. Performance on FTT repetitions was significantly higher than would be expected based on the normative data [ $t(15) = 2.15, p = .048$ ]. None of the TD children (0%) had FTT Repetition scores one or more standard deviations below the mean.

An independent-samples t-test was conducted to compare the simple fine motor scores for children with NF1 and typically developing children. There was no significant difference in FTT Repetition scores for children with NF1 and TD children ( $t(39) = -1.47, p = .148$ , two-tailed). A medium effect size was observed ( $d = 0.48$ , mean difference = -1.21, 95% CI: -2.86 to .449). A chi-square test indicates there was no significant difference in the proportion of children with NF1 falling in the delayed range (12%) as compared with the value of 0% TD children [ $\chi^2(1, n = 41) = .207, p = .150$ ].

**Mid-Level Fine Motor Abilities.** The Fingertip Tapping – Sequencing (FTT), Imitating Hand Positions (IHP), and the Visual Motor Precision (VMP) subtests from the NEPSY-II were the primary measures used to examine mid-level fine motor abilities in young children with and without NF1. The Fingertip Tapping – Sequencing subtest measures rapid motor programming. The Imitating Hand Positions subtest measures fine-motor coordination and sensorimotor differentiation and the Visual Motor Precision subtest measures fine-motor coordination and manual motor speed. Not all children were administered all three of these measures because FTT Sequencing is only standardized

for children 5 and older, therefore, only 25 children with NF1 and 16 TD children completed this measure. All children were administered Imitating Hand Positions and Visual Motor Precision.

For the children with NF1 ( $n = 25$ ), mean FTT Sequencing score fell in the average range ( $M = 8.32$ ,  $SD = 3.44$ ). When examining all children with NF1 in the sample ( $N = 38$ ), the IHP scores fell in the low average range ( $M = 7.11$ ,  $SD = 2.02$ ), and the VMP scores fell in the low average range ( $M = 7.79$ ,  $SD = 2.83$ ). One-sample t-tests were conducted to compare the children with NF1's mean scores to the mean from normative data and the rates of difficulties were also examined using frequency analysis. Performance on FTT Sequencing was significantly lower than would be expected based on the normative data [ $t(24) = -2.43$ ,  $p = .023$ ]. Three children (12%) had FTT Sequencing scores one or more standard deviations below the mean. Performance on IHP was also significantly lower than would be expected based on the normative data [ $t(37) = -8.81$ ,  $p < .001$ ]. Fifteen children (39.5%) had IHP scores one or more standard deviations below the mean. Additionally, performance on VMP was significantly lower than would be expected based on the normative data [ $t(37) = -4.79$ ,  $p < .001$ ]. Thirteen children (34.2%) had VMP scores one or more standard deviations below the mean

FTT Sequences was available for 16 TD children. The mean FTT Sequencing score fell in the average range ( $M = 9.75$ ,  $SD = 2.93$ ). When examining all TD children in the sample ( $N = 23$ ), the IHP scores fell in the average range ( $M = 8.87$ ,  $SD = 1.93$ ), and the VMP scores fell in the average range ( $M = 9.96$ ,  $SD = 4.06$ ). One-sample t-tests were conducted to compare the TD children's mean scores to the mean from normative data and the rates of difficulties were also examined using frequency analysis. Performance on

FTT Sequencing was not significantly lower than would be expected based on the normative data [ $t(16) = -.34, p = .738$ ]. One child (6.2%) had a FTT Sequencing score one or more standard deviations below the mean. Performance on IHP was significantly lower than would be expected based on the normative data [ $t(22) = -2.79, p = .010$ ]. Three TD children (13%) IHP scores one or more standard deviations below the mean. Additionally, performance on VMP was not significantly lower than would be expected based on the normative data [ $t(22) = -0.51, p = .960$ ]. Four children (17.4%) VMP scores one or more standard deviations below the mean

Independent-samples t-tests were conducted to compare the mid-level fine motor scores for children with NF1 and typically developing children. There was no significant group difference in FTT Sequencing ( $t(39) = -1.37, p = .178$ , two-tailed). A medium effect size was observed ( $d = 0.45$ , mean difference =  $-1.430$ , 95% *CI*:  $-3.47$  to  $.612$ ). There was a significant group difference in IHP scores ( $t(59) = -3.352, p = .001$ , two-tailed). A large effect size was observed ( $d = 0.9$ , mean difference =  $-1.76$ , 95% *CI*:  $-2.818$  to  $-.711$ ). Additionally, there was a significant group difference in IHP scores ( $t(59) = -2.45, p = .017$ , two-tailed). A medium effect size was observed ( $d = 0.66$ , mean difference =  $-2.167$ , 95% *CI*:  $-3.937$  to  $-.397$ ). A chi-square test indicates there was a significant difference in the proportion of children with NF1 falling in the delayed range on IHP (39.5%) as compared with the value of 13% TD children ( $\chi^2(1, n= 61) = .481, p = .028$ ). However, no significant differences were found on FTT Sequencing (NF = 12%, TD = 6.2%;  $\chi^2(1, n= 41) = .366, p = .545$ ) and VMP (NF = 34.2%, TD = 17.4%;  $\chi^2(1, n= 61) = .201, p = .156$ ).

**Complex Fine Motor Abilities.** The Copying subtest from the Differential Ability Scales – Second Edition (DAS-II) was the primary measure used to examine the complex fine motor abilities in young children with and without NF1. The Copying subtest measures visual-perceptual matching and fine motor coordination. For children with NF1, the mean Copying score fell in the low average range ( $M = 41.84$ ,  $SD = 8.525$ ). One-sample t-tests were conducted to compare the children with NF1's mean scores to the mean from normative data and the rates of difficulties were also examined using frequency analysis. Performance on Copying was significantly lower than the normative data [ $t(37) = -5.89$ ,  $p < .001$ ]. Thirteen children (34.2%) had Copying scores one or more standard deviations below the mean. For TD children, the mean Copying score fell in the average range ( $M = 52.74$ ,  $SD = 8.00$ ). Performance on Copying was not significantly lower than the normative data [ $t(22) = 1.64$ ,  $p = .115$ ]. One TD child (4.3%) had a Copying score one or more standard deviations below the mean.

An independent-samples t-test was conducted to compare the complex fine motor scores for children with NF1 and typically developing children. There was a significant group difference in Copying scores ( $t(59) = -4.95$ ,  $p < .001$ , two-tailed). A very large effect size was observed ( $d = 1.33$ , mean difference =  $-10.89$ , 95%  $CI$ :  $-15.30$  to  $-6.49$ ). A chi-square test indicates there was a significant difference in the proportion of children with NF1 falling in the delayed range (34.2%) as compared with the value of 4.3% TD children [ $\chi^2(1, n = 61) = .722$ ,  $p = .007$ ]. Table 3 summarizes the findings for fine motor ability.

Given that the groups differed in nonverbal abilities, a repeated measures multivariate analysis of covariance was performed to statistically control for group

differences in nonverbal reasoning (NV). Preliminary assumption testing was conducted to check for normality, linearity, univariate and multivariate outliers, homogeneity of variance-covariance matrices and multicollinearity, with no serious violations noted. There was a statistically significant omnibus group difference, taking into account nonverbal reasoning abilities on the combined dependent variables,  $F(4, 55) = 5.228$ ,  $p = .001$ ; Wilks' Lambda = .725; partial eta squared = .275. When the results for the dependent variables were considered separately, both Copying,  $F(1, 59) = 17.492$ ,  $p < .001$ , partial eta squared = .232 and IHP,  $F(1, 59) = 7.238$ ,  $p = .009$ , partial eta squared = .111 showed group differences even after NV abilities were taken into account. This finding indicates that group differences in complex fine-motor functioning persist even after controlling for nonverbal reasoning skills.

### **Parent Measure of Fine Motor Functioning**

The Fine Motor (FM) subscale of the Scales of Independent Behavior – Revised (SIB-R) was used to measure parental report of fine motor difficulties in young children with and without NF1. The Fine Motor subscales measures everyday adaptive fine motor functioning. For children with NF1, the raw FM score fell in the “age-appropriate” range ( $raw = 34.03$ ,  $SD = 5.856$ ). Frequency analysis was used to examine group differences in the proportion of children falling in the developmentally delayed category provided by the SIB-R normative data. Twelve children (31.6%) had FM scores falling in the developmentally delayed range. For TD children, the raw FM score fell in the “age-appropriate” range ( $raw = 37.22$ ,  $SD = 5.469$ ). Frequency analysis was used to examine group differences in the proportion of children falling in the developmentally delayed



category provided by the SIB-R normative data. Three TD children (13%) had FM scores falling in the developmentally delayed range.

An independent-samples t-test was conducted to compare the parental report of fine motor abilities (raw scores) for children with NF1 and typically developing children. There was a significant difference in FM scores for children with NF1 and TD children ( $t(59) = -2.114, p = .039$ , two-tailed). A medium effect size was observed ( $d = .57$ , mean difference = -3.191, 95% *CI*: -6.212 to -.170). A chi-square test indicates there was no significant difference in the proportion of children with NF1 falling in the developmentally delayed range (31.6%) as compared with the value of 13% TD children [ $\chi^2(1, n = 61) = .265, p = .103$ ].

### **Relations between Lab Based Fine Motor and Parental Report**

Relations between fine motor abilities (as measured by FTT Repetitions, FTT Sequences, IHP, VMP, and Copy) and parental report of fine motor abilities (as measured by FM subscale from the SIB-R) were investigated using Pearson product-moment correlation coefficients. Preliminary analyses were performed to ensure no violation of the assumptions of normality, linearity and homoscedasticity. For children with NF1, there was a large, positive correlation between Copying and FM ( $r = .443, n = 38, p = .005$ , two tailed), with higher performance on Copying associated with higher scores reported by parents on the FM subscale of the SIB-R. There were no significant correlations found for between parental report of fine motor difficulties and lab-based measures of fine motor ability for TD children.

### Parental Report of Attention Difficulties

The Hyperactivity, Cognitive Problems/Inattention, Opposition and ADHD Index of the Conners' Parent Rating Scales – Revised (Conners) served as the primary measures of attention difficulties in young children with and without NF1. Table 4 provides descriptive statistics for scores yielded from the Conners. One-sample t-tests were conducted to compare the children with NF1's mean scores to the mean from normative data. Parental report of attention difficulties was significantly lower than would be expected based on the normative data in the Cognitive Problems/Inattention [ $t(37) = 4.71, p < .001$ ], Hyperactive [ $t(37) = 3.18, p = .003$ ], and ADHD Index [ $t(37) = 5.61, p < .001$ ] scales. Parental report of attention difficulties was not significantly different from the normative data for TD children.

An independent-samples t-test was conducted to compare parental report of attention difficulties for children with NF1 and typically developing children. There was no significant difference in Opposition index scores for children with NF1 and TD children [ $t(59) = .752, p = .455$ , two-tailed]. However, there was a significant difference between groups on the Cognitive Problems/Inattention [ $t(59) = 2.99, p = .004$ , two-tailed], Hyperactive [ $t(59) = 2.58, p = .012$ , two-tailed], and ADHD Index [ $t(59) = 3.77, p < .001$ , two-tailed] scales. A large effect size was observed for Cognitive Problems/Inattention ( $d = 0.8$ , mean difference = 9.399, 95% CI: 3.113 to 15.68), a medium effect size for Hyperactive ( $d = 0.7$ , mean difference = 8.348, 95% CI: 1.897 to 14.799), and a large effect size for the ADHD Index ( $d = 1.01$ , mean difference = 9.974, 95% CI: 4.681 to 15.267) scales.

### **Relations between Fine Motor Functioning and Attention Difficulties**

Relations between fine motor abilities (as measured by FTT Repetitions, FTT Sequences, IHP, VMP, and Copy) and parental report of attention difficulties (as measured by the Conners) were investigated using Pearson product-moment correlation coefficients. Preliminary analyses were performed to ensure no violation of the assumptions of normality, linearity and homoscedasticity. There was a large, positive correlation between Opposition and FTT Repetitions ( $r = .611, n = 25, p = .001$ ), with higher performance on Repetitions associated with more oppositional behaviors reported by parents on the Opposition index scale on the Conners.

### **Discussion**

The goal of the current study was to provide more information about the early emergence of fine motor difficulties and its relationship with attention difficulties in young children with NF1 compared to typically developing children. Fine motor abilities were examined at varying complexity level, from simple to complex. Results of the current study indicate that children with NF1 do exhibit significant impairment on mid-level and complex fine motor tasks when compared to typically developing children. Difficulties in fine motor abilities persisted even after nonverbal reasoning was taken into account. This study also examined the relationship between fine motor performance and parental report of attention difficulties. However, no significant relationship between fine motor abilities and parental report of attention difficulties were found. In the following section, I summarize the findings from the analyses and discuss how these results relate to the proposed hypotheses. I describe some limitations of the current study, as well as, provide general conclusions and directions for future research.

## **Hypotheses Revisited**

*Group differences in complex and mid-level fine motor abilities but not simple fine motor abilities are expected. A larger proportion of children with NF1 will display deficits in complex and mid-level fine motor abilities but not simple motor abilities on lab-based measures than typically developing children.*

Significant group differences in complex and mid-level fine motor tasks were demonstrated in the current study. Children with NF1 had more difficulty than typically developing children on two mid-level fine motor tasks that measured fine motor coordination, sensorimotor differentiation, and manual motor control. Additionally, children with NF1 also showed more impairment than typically developing children on a complex fine motor task that examined fine motor coordination, control, and visual-perceptual matching. These findings suggest that fine motor difficulties can be demonstrated in young children with NF1, as suggested by Lorenzo et al.'s (2011) study that found significant fine motor impairment in toddlers with NF1. Additionally, these findings lend support to other studies that demonstrate impairment on complex fine motor tasks in school-aged children with NF1 (Billingsley et al., 2003; Descheemaeker et al., 2005; Gilboa et al., 2010; Hofman et al. 1994; Hyman et al., 2005; Johnson et al., 2010; Levine et al., 2006). However, the findings demonstrated in the current study are contrary to the results reported by Sangster et al.'s (2011) study of fine motor abilities in a sample of preschool children with and without NF1, in which no significant difference in Beery VMI scores were found after statistically controlling for general cognitive ability and maternal level of education. One possibility of explaining this discrepancy is that the Sangster et al. (2011) study did not utilize a well-matched control group, which may have

hindered the ability to demonstrate significant differences between groups. Future studies replicating the findings of the current study are needed to clarify the discrepancy seen in the fine motor abilities in young children with NF1. Additionally, these findings help support a growing body of literature emphasizing the importance of examining fine motor abilities at different complexity levels.

*Group differences in parental report of fine motor abilities are expected. A larger proportion of children with NF1 will display deficits in fine motor abilities as measured from parental report compared to typically developing children.*

A significant group difference in parent-reported fine motor abilities was demonstrated in the current study. This finding suggests that children with NF1 are showing more difficulties with everyday, fine motor tasks compared to typically developing children. It also indicated that the difficulties with fine motor activities are pervasive enough to impair performance on adaptive fine motor tasks, not just lab-based measures.

*A positive relationship will exist between lab-based measures and parental report of fine motor difficulties.*

A large positive correlation between parental report of fine motor abilities and a complex fine motor was seen for children with NF1, but not for typically developing children. Results suggest that better performance on lab-based complex fine motor tasks was associated with parental report of better everyday, fine motor functioning. Parent-reported fine motor functioning was not significantly associated to other lab-based measures of fine motor ability. This is an important finding that may indicate that the complex lab-based fine motor tasks may be the best measure of everyday, adaptive fine

motor functioning. Specifically, these findings suggest that parents are reporting more difficulties on fine motor tasks that require higher-order controlled of fine motor tasks (e.g. tying shoelaces, writing first and last name), rather than difficulties on lower-order controlled fine motor tasks (e.g. picking up objects with two fingers). Additionally, these results could suggest that everyday, adaptive fine motor tasks are typically more complex in nature and tend to require higher-ordered control processes.

*A positive relationship will exist between lab-based measures of complex fine motor difficulties and parental report of inattention and hyperactivity.*

No significant correlation was found between complex fine motor difficulties and parental report of inattention and hyperactivity. This finding is contrary to previous literature that suggests a relationship between attention and hyperactivity and fine motor difficulties (Fliers et al., 2008; Kalff et al., 2002; Marcotte & Stern, 1997; Piek et al., 1999; Piek & Dyck, 2004; Tseng et al., 2004; Whitmont & Clark, 1996). However, the lack of a significant correlation in this study may be more suggestive of a different profile of ADHD symptoms for children with NF1. Some recent research suggests that children with NF1 present with a different profile of ADHD symptoms than typical children with ADHD (North et al., 1995; Hofman et al., 1994). The findings from these studies report that children with NF1 have more symptoms related to difficulties with inattention and are more likely to be diagnosed with ADHD – Inattentive Type, when compared to other children with ADHD (Ferner et al., 1996; North et al., 1995; Hofman et al., 1994). This may explain why the theoretical model presented by Barkley (1997) does not fit the results of the current study. As discussed in the introduction, Barkley suggested that difficulties in fine motor abilities are a result of a deficit in inhibitory control. If children

with NF1 show the greatest difficulty with sustained attention rather than inhibitory control, then it seem likely that their impairment on complex fine motor tasks would not be as evident as it is for children with ADHD, more generally.

Finally, a lack of significant findings in the current study may be due to the age of the children examined. The majority of the previous studies that demonstrated a relationship between attention and fine motor abilities were conducted in school-aged children with ADHD. Only one study has examined the relationship in young children with ADHD (Kalff et al., 2003); therefore, more research is needed in order to make a definitive conclusion on the nature of the relationship between fine motor skill and attention in young children.

### **Limitations and Future Directions**

The current study represents the first investigation that examined differing levels of fine motor abilities and their relations to attention difficulties in young children with NF1 compared to same-aged typically developing children. However, the current study is not without several limitations. First, the current study utilized a relatively small sample size. Further studies using a larger number of participants, including younger and older children, would be helpful for further understanding the development of fine motor skills and attention difficulties in children with NF1, more generally. Second, the current study employed a cross-sectional design to examine fine motor abilities in 4-6 year old children, which limits the ability to interpret the results from a developmental viewpoint. Future studies using a longitudinal design would be better able to describe the course and development of fine motor difficulties and its direct relationship to attention difficulties. Finally, the current study only examined one measure of simple fine motor ability and

only one measure of complex fine motor ability. Future studies would benefit from using more simple and complex fine motor tasks in order to better generalize the results.

### **Conclusions**

The present study outlines the fine motor difficulties seen in young children with NF1 compared to typically developing children. For children with NF1, significant difficulties were demonstrated on lab-based, mid-level and complex fine motor tasks, even after controlling for nonverbal reasoning abilities. These findings suggesting that children with NF1 do not differ significantly from TD children on lab-based, simple fine motor tasks. Additionally, these findings were corroborated by parental report of difficulties in adaptive fine-motor functioning. The current study also examined the relation between fine motor ability and parental report of attention difficulties. While no significant correlations were found between complex fine motor ability and attention difficulties, results may be related to the possibility of attention difficulties for children with NF1 that is qualitatively different from children with ADHD alone. Future research using larger sample sizes and longitudinal designs is needed to further explore the relationship between fine motor abilities and attention.



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Table 1.

*NEPSY-2 and DAS-II Subtest Score Descriptions*

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

Table 2.

<i>Demographic Data</i>		
	NF1 (n=38)	TD (n=23)
Gender		
Male	21 (55%)	15 (65%)
Female	17 (45%)	8 (35%)
Age in Months		
Mean	63.08 months	64.70 months
Range	48-82 months	48-82 months
Ethnicity		
Caucasian	28 (74%)	20 (88%)
African American	3 (8%)	1 (4%)
Hispanic	5 (13%)	0 (0%)
Asian	1 (2.5%)	1 (4%)
Mixed	1 (2.5%)	1 (4%)

Table 3.

*Group differences on selected NEPSY-II and DAS-II motor-related subtests*

Subtest	NF1			TD			Sig
	<i>N</i>	<i>Mean</i>	<i>(SD)</i>	<i>N</i>	<i>Mean</i>	<i>(SD)</i>	
Fingertip Tapping							
Repetitions	25	10.04	2.70	16	11.25 +	2.32	
Sequences	25	8.32	3.44 +	16	9.75	2.93	
Imitating Hand Positions	38	7.11	2.02 ++	23	8.87 ++	1.93	**
Visuomotor Completion	38	7.79	2.83 ++	23	9.96	4.06	*
Copying	38	41.84	8.52 ++	23	52.74	7.99	**

Significantly different from normative data in one-sample t-test +  $p < .05$ ; ++  $p < .001$

Significant group differences \*  $p < .05$ ; \*\*  $p < .001$

Table 4.

*Group differences on the Conners' Parent Rating Scale*

Scale	NF1			TD			Sig
	<i>N</i>	<i>Mean</i>	<i>(SD)</i>	<i>N</i>	<i>Mean</i>	<i>(SD)</i>	
Opposition	38	54.21	14.02	23	51.65	10.65	
Inattention	38	59.92	12.96 ++	23	50.52	9.83	*
Hyperactivity	38	57.00	13.57 ++	23	48.65	9.46	*
ADHD Index	38	59.97	10.95 ++	23	50.00	8.18	**

Significantly different from normative data in one-sample t-test +  $p < .01$ ; ++  $p < .001$

Significant group differences \*  $p < .01$ ; \*\*  $p < .001$