Title: An Investigation of the Use of Computerized Attention/Executive Functioning Measures with School Age Children with Neurofibromatosis Type 1

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Introduction: Children with neurofibromatosis type 1 (NF1) are at an increased risk of developing difficulties with executive function. About one third to one half of children with NF1 meet diagnostic criteria for Attention Deficit/Hyperactivity Disorder (ADHD; North, Hyman, & Barton, 2002), with even more children demonstrating sub-clinical executive difficulties (Casnar & Klein-Tasman, 2016; Plasschaert et al., 2016). The NIH Toolbox and the Cogstate computerized assessment battery are relatively new computerized measures of attention and executive functioning; the performance of children with NF1 on these measures has not been described. Computerized measures of executive function tend to be easy to administer and offer continually updated normative data. There is a need to identify measures that are sensitive to the attention and executive functioning challenges of children with NF1, especially for use in clinical trials research (Walsh et al., 2016).

Method: School age (9-13 years; Mage=11.02) children with NF1 (N = 37; 20 male) were seen in the context of a larger study. Participants were administered several National Institute of Health (NIH) Toolbox attention and executive function measures, including the Dimensional Change Card Sort Task (DCCS; Zelazo et al., 2013), List Sort Working Memory (LSWM; Tulsky et al., 2015), and Flanker (Zelazo et al., 2013). All of the NIH Toolbox measures yield standard scores (SS; M=100, SD=15). The frequency of difficulties on each measure was examined (mild difficulty: SS<85; severe difficulty: SS<70). Participants were also administered the Cogstate (Cogstate Ltd. 2002) Identification and One Back Tasks. The primary outcome measure on the Identification task is log transformed reaction time, and the outcome on the One Back is Arcsine transformed accuracy. The Cogstate outcome scores were converted to z-scores (M=0, SD=1) for analyses. The frequency of difficulties on each measure was examined (mild difficulty: Z>1; severe difficulty: Z>2).

Results: Initial analyses were conducted examining the frequency of at least mild difficulties across measures. 21.6% of children demonstrated mild difficulty on the DCCS, 16.2% on the LSWM, 47.2% on the Flanker, 51.3% on Identification, and 35.9% on One Back. Cochran’s Q test determined that there was a significant difference in the proportion of identified difficulties across tasks, \( \chi^2(4)=19.69, p = .001 \). Follow-up McNemar’s tests demonstrated significant differences between the DCCS and Flanker (\( p=.022 \)), DCCS and Identification (\( p=.012 \)), LSWM and Flanker (\( p=.006 \)), and LSWM and Identification (\( p=.002 \)). Next, the frequency of severe difficulties across measures was examined. 2.7% of children had severe difficulty on the DCCS, 5.4% on the LSWM, 8.3% on the Flanker, 28.2% on Identification, and 15.4% on One Back. Cochran’s Q indicated a significant difference in identification of severe difficulties across tasks, \( \chi^2(4)=18.59, p = .001 \). Follow-up McNemar’s tests demonstrated significant differences between the DCCS and Identification (\( p=.002 \)), LSWM and Identification (\( p=.012 \)), and Flanker and Identification (\( p=.039 \)) tasks.

Discussion: We examined whether computerized measures of executive abilities differentially identify difficulties in children with NF1. The findings of the present study suggest that the NIH Flanker and Cogstate Identification tasks identify significantly more children with executive difficulties than the DCCS, LSWM, and One Back tasks. Furthermore, the Identification task identifies significantly more severe executive difficulties than all NIH Toolbox executive tasks. The NIH Flanker and Cogstate Identification tasks may be particularly useful tools for detection of these difficulties in clinical trials and developmental research with children with NF1. Further implications including potential limitations will be discussed.

References:

- Cogstate Ltd. (2002). *Cogstate* (Version 2.1.0). Melbourne, Australia


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