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Understanding macrographia in children with autism spectrum disorders

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ABSTRACT

It has been consistently reported that children with autism spectrum disorders (ASD) show considerable handwriting difficulties, specifically relating to accurate and consistent letter formation, and maintaining appropriate letter size. The aim of this study was to investigate the underlying factors that contribute to these difficulties, specifically relating to motor control. We examined the integrity of fundamental handwriting movements and contributions of neuromotor noise in 26 children with ASD aged 8–13 years (IQ > 75), and 17 typically developing controls. Children wrote a series of four cursive letter *l*'s using a graphics tablet and stylus. Children with ASD had significantly larger stroke height and width, more variable movement trajectory, and higher movement velocities. The absolute level of neuromotor noise in the velocity profiles, as measured by power spectral density analysis, was significantly higher in children with ASD; relatively higher neuromotor noise was found in bands >3 Hz. Our findings suggest that significant instability of fundamental handwriting movements, in combination with atypical biomechanical strategies, contribute to larger and less consistent handwriting in children with ASD.

1. Introduction

Autism spectrum disorders (ASD) refers to a group of neurodevelopmental disorders that share a triad of impairments in communication, social interactions, and restricted and stereotyped patterns of interests and behaviors. In addition to their core diagnostic symptoms, it is now recognized that there are also pervasive motor abnormalities associated with ASD (Fournier, Hass, Naik, Lodha, & Cauraugh, 2010). The need for further investigation of fine motor and handwriting impairments, in particular, is highlighted by the high rate of clinical referrals for assistance in this domain. A study of academic performance in boys with Asperger's disorder (AD) found that 58% of children those aged between 6 and 11 years had received occupational therapy to address fine motor issues such as poor handwriting and scissor manipulation (Church, Alisanski, & Amanullah, 2000). Cartmill, Rodger, and Ziviani (2009) also reported 40% of occupational therapists' caseloads were comprised of children with ASD, and 86% of those cases with twenty or more consecutive referrals were for assistance with handwriting or fine motor skills. Despite the clear clinical and academic importance of evidence-based handwriting therapies for children with ASD, few studies have attempted to characterize the nature of handwriting difficulties in these disorders.

Handwriting is a complex skill, dependent on smooth integration of higher-order linguistic planning working memory, proprioceptive and visuomotor control (Cartmill et al., 2009; Kushki, Chau, & Anagnostou, 2011). A review of the

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handwriting literature in ASD by Kushki et al. (2011) identified consistent deficits in overall legibility and letter formation. Of the small number of studies that have examined handwriting in children and adolescents with ASD, qualitative measures have identified problems with legibility (Church et al., 2000; Fuentes, Mostofsky, & Bastian, 2009) and poor letter formation (Cartmill et al., 2009; Fuentes et al., 2009). Difficulty controlling handwriting size is also commonly reported in children with both high functioning autism (HFA) and AD (Cartmill et al., 2009; Myles et al., 2003). Furthermore, Beversdorf et al. (2001) found that adults with ASD exhibited macrographia (atypically large handwriting). The overarching aim of this study was to therefore move beyond descriptions of handwriting quality and understand the process of producing handwriting movements in children with ASD, specifically focusing on motor control.

All handwriting is comprised of a base unit, a stroke, which is a simple up or down movement; consecutive up and down strokes form a loop, e.g. the cursive letter *l* (Plamondon & Guerfali, 1998). Strokes and loops are chained together to form more complex letters and whole words (Plamondon & Guerfali, 1998). The motor plan for simple handwriting strokes can be or upscaled or downscaled dependent on the context, for example the letter *l* or *e* (Phillips, Ogeil, & Best, 2009). If the internal representation (motor plan) for basic handwriting units is poorly formed or poorly controlled, this has downstream consequences for consistent formation of letters and planning of whole words (Phillips, Ogeil, & Best, 2009; Plamondon & Guerfali, 1998). The approach of using a series of chained loops, such as '*l*'s or '*e*'s to understand the control of handwriting movements has previously been used in children with ADHD (Langmaid, Papadopoulos, Johnson, Phillips, & Rinehart, 2012; Tucha, Mecklinger, Walitza, & Lange, 2006), Huntington's disease (Phillips, Bradshaw, Chiu, & Bradshaw, et al., 1994), Parkinson's disease (Caligiuri, Teulings, Filoteo, Song, & Lohr, 2006), and alcohol dependence (Phillips, Ogeil, & Müller, 2009). Studies assessing the control of simple fine motor movements, such as '*l*'s or small circles, have found that limiting external cues, such as writing guides, is effective in eliciting and assessing the integrity of the internal representation of the motor plan (Langmaid et al., 2012; Martin, Phillips, Iansek, & Bradshaw, 1994).

Following on from findings of inconsistent letter formation in children with ASD (Cartmill et al., 2009; Church et al., 2000; Fuentes et al., 2009), the first specific aim of this study was to therefore assess the integrity of the internal representation of simple handwriting movements in children with ASD, using a cursive *l*'s task. It was hypothesized that children with ASD would show more unstable motor plans for simple handwriting movements (strokes) than typically developing children. Previous studies of children with poor handwriting (van Galen, Portier, Smits-Engelsman, & Schomaker, 1993) and ADHD (Langmaid et al., 2012) have indicated that this would manifest as overall movement size and trajectory being larger and more variable. A recent kinematic evaluation of large circle drawing in children aged 4–8 years with ASD revealed that timing consistency during drawing continuous circles did not differ between ASD and controls, therefore we did not anticipate any greater variability in movement duration (Fleury, Kushki, Tanel, Anagnostou, & Chau, 2013). However, we predicted greater movement velocity (van Galen et al., 1993) and reduced index of ballisticity (i.e. few number of cycles of acceleration and deceleration during a movement; Langmaid et al., 2012).

The second aim of this study was to investigate whether inconsistent letter formation may also relate to inherently greater noise within the neuromotor system (Smits-Engelsman & Van Galen, 1997; van Galen et al., 1993). Previous studies have estimated neuromotor noise using power spectral density analysis, in which the velocity profile of an entire stroke is decomposed into its frequencies using Fast Fourier Analysis (Smits-Engelsman & Van Galen, 1997; van Galen et al., 1993). As the duration of each complete *l* is approximately 1 second, low frequencies (1–2 Hz) relate to the periodicity of the task itself, representing the adductive and abductive finger and wrist movements required to write consecutive letter *l*'s (van Galen, van Doorn, & Schomaker, 1990). Superimposed upon this signal are frequencies associated with visual and proprioceptive feedback corrections, tremor, and neuromotor recruitment. Handwriting movements require ongoing visual feedback to maintain an accurate movement trajectory, resulting in rapid corrections to movement trajectory that can seen within the 3 to 4 Hz range (Miall, Weir, Wolpert, & Stein, 1993; Smits-Engelsman & Van Galen, 1997). Uncontrolled movements such as neuromotor tremor and reflex-based contractions, occur at frequencies greater than 5 Hz (van Galen et al., 1990). In tasks that are highly automated, or well learned, the relative contribution of low frequency (1–4 Hz) decreases, and there is a shift toward frequencies >5 Hz (van Galen et al., 1993). Poor handwriting in typically developing children is associated with greater energy within the 4–7 Hz band of the velocity profile (van Galen et al., 1993). We therefore predicted higher absolute spectra values in the ASD group relative to typically developing controls across all frequencies. Further, we predicted children with ASD would show relatively higher frequencies in bands >4 Hz compared to typically developing children. In light of reports of atypical visuomotor control as a contributing factor to handwriting deficits in ASD (Cartmill et al., 2009), we also predicted higher relative energy within the 3–4 Hz band.

The final aim of this study was to explore whether age, IQ measures, motor skills and the spectral band associated with visual correction of movement (3–4 Hz) were associated with kinematic features of handwriting movements in ASD. Earlier investigations of handwriting quality in ASD found that motor skills as measured by Physical and Neurological Examination for Subtle (Motor) Signs (PANESS; Fuentes et al., 2009), and perceptual reasoning index (Fuentes, Mostofsky, & Bastian, 2010) were significantly predictive of letter formation.

2. Method

2.1. Participants

This study was approved by Monash University and Southern Health Human Research Ethics Committees. Parents of participants provided informed consent prior to the commencement of the study, and written assent was provided by the