Research in Developmental Disabilities 35 (2014) 203–214

Contents lists available at ScienceDirect

Research in Developmental Disabilities

Gait profiles as indicators of domain-specific impairments in executive control across neurodevelopmental disorders

Darren R. Hocking [a,b,*], Jasmine C. Menant [c], Hannah E. Kirk [a], Stephen Lord [c], Melanie A. Porter [d]

[a] School of Psychology and Psychiatry, Faculty of Medicine, Melbourne, Australia
[b] Olga Tennison Autism Research Centre, School of Psychological Science, La Trobe University, Bundoora, Australia
[c] Neuroscience Research Australia, University of New South Wales, Sydney, Australia
[d] Department of Psychology, Macquarie University, Sydney, Australia

ARTICLE INFO

Article history:
Received 23 August 2013
Received in revised form 2 October 2013
Accepted 2 October 2013
Available online 28 October 2013

Keywords:
Williams syndrome
Down syndrome
Gait
Dual-tasking
Gait variability
Executive function
Executive control
Verbal working memory
Attention set-shifting

ABSTRACT

In neurodevelopmental disorders, unique profiles of executive control and attention appear to co-occur with poor motor coordination. However, less is known about how syndrome-specific cognitive profiles interact with motor control and impact behavioural outcomes in neurodevelopmental disorders such as Williams syndrome (WS) and Down syndrome (DS). Here we aimed to examine the extent to which specific components of executive function interact with gait control when performing cognitive dual-tasks (verbal fluency, digit span) in WS and DS. Spatiotemporal gait characteristics and intra-individual variability of gait were assessed in individuals with WS who were matched on spatial ability to individuals with DS, and chronologically age (CA) matched controls. During the concurrent verbal fluency task, the WS group had greater dual-task costs on spatiotemporal gait parameters and variability than CA controls. Conversely, individuals with DS had selective gait interference during the concurrent digit span task when compared to CA controls, but only under increased demands on cognitive control where there was greater variability in step timing in DS. The interrelationships between cognitive-motor interference and behavioural measures of executive functioning appeared to differentiate between WS and DS, and emphasise the importance of task modality in unpacking the executive control profile in these neurodevelopmental disorders. These findings support the notion that associated cerebellar-cortico abnormalities may produce quite distinct profiles of executive control across cognitive and motor domains that impact on behavioural outcomes in neurodevelopmental disorders.

1. Introduction

Neurodevelopmental disorders of different genetic origin, such as Williams syndrome or Down syndrome, are characterised by contrasting profiles of cognitive and behavioural difficulties. Williams syndrome (WS) is a relatively rare genetically based disorder with prevalence estimates between 1 in 7500 and 1 in 20,000 births (Stromme, Bjornstad, & Ramstad, 2002), and is caused by a de novo microdeletion of 25–28 genes on the long arm of chromosome 7, specifically 7q11.23 (Ewart et al., 1993). Individuals with WS generally show relative strengths in verbal abilities alongside core

* Corresponding author at: Olga Tennison Autism Research Centre, School of Psychological Science, La Trobe University, Bundoora, Australia.
E-mail address: d.hocking@latrobe.edu.au (D.R. Hocking).

0891-4222/$ – see front matter © 2013 Elsevier Ltd. All rights reserved.
http://dx.doi.org/10.1016/j.ridd.2013.10.005

weaknesses in visual short-term memory (STM), visuospatial construction and visuomotor control (Hocking, Bradshaw, & Rinehart, 2008; Jarrold, Baddeley, & Hewes, 1999; Karmiloff-Smith, 2012). Down syndrome (DS) is one of the leading causes of intellectual disability affecting 1 in every 700–1000 live births (Kittler, Krinsky-McHale, & Devenny, 2008), and is caused by additional material on chromosome 21. In addition to an increased risk of age-related cognitive decline and dementia (Rowe, Lavender & Turk, 2006), the DS profile is characterised by weaknesses in verbal abilities (expressive language) and verbal STM, alongside relative strengths in visuospatial construction when compared to their developmental level (Vicari, 2006). In the context of these roughly opposing cognitive profiles, core motor coordination problems have been reported separately in WS and DS, especially in relation to poor balance (WS: Barozzi et al., 2013; DS: see Enkelaar, Smulders, van Schrojenstein Lantman-de Valk, Geurts, & Weerdesteyn, 2012, for a review) and gait abnormalities (WS: Hocking, McGinley, Moss, Bradshaw, & Rinehart, 2010; Hocking et al., 2011, 2013; DS: Smith & Ulrich, 2008; Smith, Ashton-Miller, & Ulrich, 2010). However, the extent to which the unique cognitive profiles interact with motor control across the lifespan in these neurodevelopmental disorders is hitherto unknown. As such, studying the dynamic interplay between cognitive and motor functioning may provide important insights into the extent to which cross-domain deficits impact behavioural outcomes across neurodevelopmental disorders.

Recent studies in both genetic syndromes have provided evidence to point to a fractionation of executive functions and attentional control skills (Borella, Carretti, & Lanfranchi, 2013; Carney, Brown, & Henry, 2013; Lanfranchi, Jerman, Dal Pont, Alberti, & Vianello, 2010; Rhodes, Riby, Park, Fraser, & Campbell, 2010; Rhodes, Riby, Fraser, & Campbell, 2011). Executive function is a broad term incorporating an extensive set of higher-order operations that organise and regulate goal-directed behaviour by modulating more posterior brain systems via the attention network (Miyake, 2000). It is well established that executive control can be fractionated into separable, but not completely independent, components processes, namely the ability to shift between different mental sets or tasks (“Shifting”), updating and monitoring of working memory representations (“Updating”), and selectively attending to stimuli and inhibiting prepotent responses (“Inhibition”) (Miyake et al., 2000; Miyake & Friedman, 2012). With regard to WS, set-shifting, STM and working memory impairments have been observed on tasks requiring manipulation of visuospatial (Menghini, Addona, Costanzo, & Vicari, 2010; Rhodes et al., 2010), but not verbal, material (but see Jarrold, Baddeley, & Phillips, 2007). In the domain of inhibition in WS, Atkinson et al. (2003) found a relative difficulty on inhibitory tasks requiring spatial or motor responses (antiscaccade and detour box reaching tasks), but with a relative preservation of performance on Stroop like naming tasks (day and night task). In contrast, there is evidence for greater impairment in shifting and verbal aspects of memory in individuals with DS when compared to mental age (MA) matched controls (Carney et al., 2013; Lanfranchi et al., 2010). Similarly, in the verbal domain, individuals with DS make more intrusion errors during verbal working memory tasks (Kittler, Krinsky-McHale, & Devenny, 2006), show poorer response inhibition when using verbal-based material (Borella et al., 2013; Porter, Coltheart, & Langdon, 2007), and exhibit poorer control of concurrent cognitive tasks (Brunamonti et al., 2011; Kittler et al., 2008). However, both neurodevelopmental disorders show impairments in executive working memory tasks which require a high level of cognitive control (Costanzo et al., 2013; Lanfranchi, Carretti, Spanò, & Cornoldi, 2009; Lanfranchi, Jerman & Vianello, 2009; Rhodes et al., 2010). Together, these findings suggest that the atypical development of separable domains of executive control and their interactions may result in a complex pattern of cognitive performance across neurodevelopmental disorders.

The cross-domain effects of impairments in executive control may also be implicated in the motor control problems in these neurodevelopmental disorders. Abnormalities of gait, impairments in stair and surface stepping, and poor visuomotor integration are common characteristics of individuals with WS (Cowie, Braddick, & Atkinson, 2012; Hocking et al., 2011; Withers, 1996). The evidence for associations between variable stride length and non-verbal abilities such as perceptual organisation and processing speed (Hocking, Rinehart, McGinley, & Bradshaw, 2009), and increased variability of stride time during attentional cueing of stride length (Hocking et al., 2010) point to the view that higher-order cognitive functions may contribute to the motor control problems in WS (Hocking et al., 2008). Significantly, some commonalities in gait have been reported between individuals with WS and DS, with slower walking speed, shorter stride length and a wider base of support (Hocking et al., 2009, 2010; Rigoldi, Galli, and Albertini, 2011), which may be compensatory strategies to maintain postural stability. Similarly, both neurodevelopmental disorders show evidence of poor anticipatory gait adjustments during obstacle crossing (Hocking et al., 2011; Vimercati, Galli, Rigoldi, & Albertini, 2013). Despite these cross-syndrome commonalities, at a finer-tuned level, individuals with DS show marked temporal variability of gait during obstacle crossing (Smith & Ulrich, 2008; Vimercati et al., 2013), whereas individuals with WS show a lack of integration of visual and/or spatial information when approaching an obstacle early in the gait cycle suggestive of poor planning abilities (Hocking et al., 2011). Taken together, these findings suggest cross-syndrome similarities in anticipatory control during gait adaptation, but also imply syndrome-specific profiles that may stem from a higher demand on the reduced executive control resources within the visuomotor domain.

Cross-syndrome commonalities and differences can be explored using dual-task paradigms, which examine the performance on a primary cognitive or motor task while concurrently performing a secondary task. The differential effect of interference from a secondary task can be interpreted within an influential model of working memory (Baddeley, 1986, 2007) which comprises a limited-capacity central executive for the control of attention, two separable components for the temporary storage of verbal (‘phonological loop’), and visuospatial (‘visuospatial sketchpad’) material, and an episodic buffer, which provides a passive store for combining information from the two storage systems. One component of the central executive is the ability to coordinate the performance of multiple tasks (Kane & Engle, 2003; Alloway, Gathercole, & Pickering, 2006). In studies on DS individuals, when compared to younger MA-matched controls, impairments in dual-task