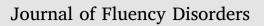
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Atypical non-verbal sensorimotor synchronization in adults who stutter may be modulated by auditory feedback



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A R T I C L E I N F O

Keywords: Stuttering sensorimotor control timing synchronization

ABSTRACT

Purpose: To investigate if non-verbal sensorimotor synchronization abilities in adult individuals who stutter (IWS) differ from non-stuttering controls (NS) under various performance conditions (tempo, auditory feedback, use of hands [single/both] and rhythm).

Methods: Participants were 11 IWS (5 males, 6 females, *Mean age* = 25.8, SD = 8.7) and 11 ageand gender-matched controls (*Mean age* = 24.4, SD = 8.4). During the experiment, participants were asked to prepare three melodies and subsequently perform them with a metronome at different rates and auditory feedback modalities (non-altered and suppressed). For each task/ condition we tracked timing asynchrony related to the steady metronome beat.

Results and conclusions: : Overall, IWS displayed significantly higher timing asynchrony. Of all conditions, auditory-feedback distinguished IWS from NS most strongly, a subgroup of IWS significantly benefitting from the absence of auditory feedback. In addition, IWS showed a non-significant trend of higher negative mean asynchrony (NMA) and were more affected by the slower rate and increased rhythmic complexity and occasionally suggested poorer beat perception. These results suggest aberrant timing of sensorimotor network interaction associated with the origin of developmental stuttering.

1. Introduction

Developmental stuttering is a neurobiological disorder affecting (speech) fluency in approximately 5% of all pre-school age children and 1% of the teenage and adult population (Yairi & Ambrose, 2013). Although the exact etiology and interplay of factors in stuttering remain unclear, precise timing of movements and successful integration of sensorimotor processes appear to be necessary for effortless, fluent speech production (Civier, Tasko, & Guenther, 2010; Zelaznik, Smith, & Franz, 1994). In IWS, differences in timing and integration of sensorimotor processes have been found during both episodes of stuttering and perceptually fluent speech (Caruso, Abbs, & Gracco, 1988; Zimmerman, 1980b). There is evidence that during stutter-free speech, IWS could be identified on the basis of aberrant speech timing patterns, such as atypical rate and less accurate rhythms (Wendahl & Cole, 1961). Based on these observations, it has been proposed that impaired *timing* mechanisms may be a central component to the disorder (Andrews et al., 1983; Harrington, 1988; Kent, 1984; Van Riper, 1973). More specifically, some have argued that the core dysfunction in stuttering may be related to a defective 'medial system', comprised of the basal ganglia (BG) and supplementary motor area (SMA) (Goldberg, 1985), to produce 'internal' (i.e., in the absence of any external signal) timing cues for the initiation of the next motor segment (Alm, 2004), an hypothesis recently echoed by Etchell et al. (2014). Consequently, IWS may rely on secondary systems that utilize external

Received 4 November 2016; Received in revised form 13 April 2017; Accepted 19 May 2017 Available online 27 May 2017 0094-730X/ © 2017 Elsevier Inc. All rights reserved.

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http://dx.doi.org/10.1016/j.jfludis.2017.05.004

timing cues to sequence (speech) movements. Support for this viewpoint, amongst other things, comes from the fact that IWS are able to reduce their stuttering markedly when speaking synchronized to a metronome or another person – also known as 'choral reading' (e.g., Andrews, Howie, Dozsa, & Guitar, 1982). Further evidence in favor of this hypothesis has emerged from both behavioral and neuroimaging studies of speech production. For example, more temporal variability in IWS has been shown with respect to respiratory control (e.g., Zocchi et al., 1990), voice onset time (Zimmerman, 1980a), voice initiation time (Cross & Luper, 1979), vowel duration (Hand & Luper, 1980) and articulatory kinematics (e.g., Max & Gracco, 2005); these are consistent with findings of abnormal functioning of the basal-ganglia thalamo-cortical (BGTC) network in adults (Braun et al., 1997; Chang, Horwitz, Ostuni, Reynolds, & Ludlow, 2011; Watkins, Smith, Davis, & Howell, 2008) and children who stutter (Chang & Zhu, 2013), which may disappear when speaking under fluency-inducing conditions (Toyomura et al., 2011Toyomura, Fujii, & Kuriki, 2011) or after successful therapy (Giraud et al., 2008). Interestingly, areas as the BG and SMA – known to be involved, amongst other things, in the selection, initiation, timing and automatization of speech movement patterns – appear to play a role as well in rhythm and beat processing (Grahn & Brett, 2007), in line with recent findings that suggested atypical rhythm perception in children who stutter (Wieland, McAuley, Dilley, & Chang, 2015).

By contrast – but certainly not mutually exclusive with the aforementioned claims – others have posited auditory-motor coupling, sensorimotor integration and forward modeling as fundamental underlying problems of stuttering (Max, 2004; Neilson & Neilson, 1987). Consistent with this line of reasoning is the fact that IWS often immediately reduce stuttering when their own auditory feedback is altered (e.g., speaking under delayed/frequency-altered auditory feedback, masking noise or simultaneous with a second speaker; for a review, see Lincoln, Packman, & Onslow, 2006). Support for aberrant sensorimotor processes and learning in stuttering has also included findings of reduced compensation to somatosensory (Caruso et al., 1987Caruso, Gracco, & Abbs, 1987) and auditory feedback manipulations (Cai et al., 2012) as well as abnormalities in learning verbal sequences (Smits-Bandstra & De Nil, 2009), implicit speech sequence learning (Smits-Bandstra & Gracco, 2015) and overall limited retention in learning speech motor sequences (Namasivayam & van Lieshout, 2008; Smits-Bandstra & Gracco, 2015). Neuroimaging studies are generally consistent with an underlying sensorimotor abnormality by demonstrating over-activation of (primarily right-hemisphere) motor areas, increased cerebellar activity and under-activation of auditory areas (Brown, Ingham, Ingham, Laird, & Fox, 2005; Chang, Kenney, Loucks, & Ludlow, 2009; De Nil, Kroll, & Houle, 2001; Fox et al., 1996; Watkins et al., 2008), abnormal grey and white matter integrity in speech motor regions (Beal, Gracco, Brettschneider, Kroll, & De Nil, 2013; Beal, Gracco, Lafaille, & De Nil, 2007; Chang et al., 2011; Chang, Zhu, Choo, & Angstadt, 2015), atypical symmetry of the planum temporale (Foundas, Bollich, Corey, Hurley, & Heilman, 2001) and reduced functional connectivity in premotor cortical areas (Chang & Zhu, 2013; Lu et al., 2010).

One of the challenges associated with many of the aforementioned studies measuring speech motor control in IWS is dissociating whether the findings may reflect either a primary (or secondary) abnormality, or a conscious or unconscious attempt not to stutter (i.e., compensatory reaction). This problem is largely absent when one investigates performance in non-speech tasks, such as manual reaction times, finger-tapping, non-verbal dual-tasks or synchronization-continuation tasks, as IWS have generally not experienced concrete stuttering during such tasks and thus are unlikely to display any corrective or compensatory behavior.

Based on the premise that common mechanisms may be responsible for rhythmic movements across both speech and non-speech fine motor tasks (Franz, Zelaznik, & Smith, 1992), differences between IWS and controls in non-verbal timing and rhythmic tasks have been reported by some (e.g., Borden, 1983; Cooper & Allen, 1977; Falk, Müller, & Dalla Bella, 2015; Howell, Au-Yeung, & Rustin, 1997; Zelaznik, Smith, Franz, & Ho, 1997) but were not replicated by others (Hulstijn, Summers, van Lieshout, & Peters, 1992; Max & Yudman, 2003; Till, Goldsmith, & Reich, 1981). These inconclusive findings may be explained by differences in task and methodology; usually more demanding tasks elicited more pronounced differences [e.g., Hulstijn et al., 1992; Zelaznik et al., 1997] and neuroimaging may reveal differences, which may be obscured using behavioral measures.

One factor that has been overlooked in previous studies that investigated non-verbal sensorimotor behavior in IWS is the presence or manipulation of auditory feedback of self-initiated actions. Usually, tapping tasks do not elicit any complex and meaningful auditory feedback comparable to speech, raising the possibility that group differences may be modulated by this factor. Specifically, it could be that tapping or rhythmic manual movements may be less accurate and/or more variable in IWS when the complexity and meaningfulness of self-initiated auditory feedback increases. This may be a plausible possibility as both children and adults who stutter have shown aberrant auditory processing as a function of increased stimulus complexity (Corbera, Corral, Escera, & Idiazábal, 2005; Gonçalves, Andrade, & Matas, 2015). Additionally, to our knowledge, no studies to date have examined whether rhythmic complexity may be a factor that potentially affects non-verbal timing more in IWS than in controls.

1.1. Current study

Using a non-verbal synchronization task, the current experiment attempts to extend results of previous studies by investigating short-term sensorimotor learning. Here we are interested whether distinct modalities of auditory feedback (non-altered and suppressed) along with different rhythmic and manual complexity and various rates will have consequences for the timing of sequential finger movements. Participants learned to play 3 different melodies on a digital piano. The three tunes varied in complexity, with the first using only equal note values synchronized to the metronome (resembling a typical finger-tapping task), the second making alternate use of half and quarter notes and the third using both hands. This allowed us to identify also which type of complexity may result in a relative higher amount of timing asynchrony. Based on previous studies (e.g., Falk et al., 2015; Zelaznik et al., 1997), we expected that IWS would perform less synchronized to the beat than NS, especially for the more complex melodies. More specifically, if timing accuracy in stuttering is primarily modulated by rhythmic features, we expect that IWS will experience relatively more difficulty with melody 2. If specific coordination patterns (of the two hands) pose greater challenges to IWS (as shown by Zelaznik

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