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Evidence for a rhythm perception deficit in children who stutter

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ABSTRACT

Stuttering is a neurodevelopmental disorder that affects the timing and rhythmic flow of speech production. When speech is synchronized with an external rhythmic pacing signal (e.g., a metronome), even severe stuttering can be markedly alleviated, suggesting that people who stutter may have difficulty generating an internal rhythm to pace their speech. To investigate this possibility, children who stutter and typically-developing children ($n = 17$ per group, aged 6–11 years) were compared in terms of their auditory rhythm discrimination abilities of simple and complex rhythms. Children who stutter showed worse rhythm discrimination than typically-developing children. These findings provide the first evidence of impaired rhythm perception in children who stutter, supporting the conclusion that developmental stuttering may be associated with a deficit in rhythm processing.

1. Introduction

Stuttering is a speech disorder characterized by frequent occurrences of repetitions or prolongations of sounds, syllables, or words that disrupt the rhythmic flow of speech (World-Health-Organization, 2010). Stuttering onset is typically observed between the ages of two and five years when children begin to form simple sentences. Of these children who stutter, up to 80% will recover from stuttering (Andrews et al., 1983; Yairi & Ambrose, 1999). Despite decades of behavioral and imaging research, the exact mechanisms behind speech disruptions in people who stutter remain unclear (e.g., Alm, 2004; Packman, Code, & Onslow, 2007).

One of the hallmarks of skilled motor behavior such as fluent speech production is accurate timing (Zelaznik, Smith, & Franz, 1994). Many models of speech timing have proposed that speech, like other motor activities, is rhythmically structured in time (e.g., Allen, 1973; Cummins, 2009; Cummins & Port, 1998; Dilley, Wallace, & Heffner, 2012; Martin, 1972; Tilsen, 2009). Rhythm can generally be defined as a serial pattern of durations marked by a series of events, and perceptually as the perceived temporal organization of the physical sound pattern (McAuley, 2010). Wendahl and Cole (1961) modified recordings of adults who do and do not stutter to remove disfluencies and then asked participants to evaluate the speech on measures such as rate (i.e., normal tempo) and rhythm. Their results demonstrated that even during fluent productions, adults who stutter had a less typical rate of speech and used less rhythmical speech patterns than adults who do not stutter. DiSimoni (1974) likewise found differences in the timing of productions of speech segments in adults who stutter compared with controls. Kent (1984) suggested that the primary difference between people who stutter and fluent speakers can be found in the capacity to generate temporal structures of action. He suggested that a reduced ability to generate temporal patterns for speech perception and production is a central disturbance in stuttering behavior. Andrews et al. (1983) also suggested that an unreliable mechanism for timing control may exist in adults who stutter.

One well-known phenomenon is that people who stutter become more fluent when synchronizing their speech to an external pacing signal, such as an isochronous metronome (Wingate, 2002; Wohl, 1968). Other conditions such as speaking in unison with another person ("choral speech") (Adams & Ramig, 1980; Ingham & Carroll, 1977), and singing (Glover, Kalinowski, Rastatter, & Stuart, 1996) also have similar fluency inducing effects. These conditions may reduce disfluency in people who stutter because they each provide an external rhythmic timing reference during speech production, something that may not be present during spontaneous speech production. Etchell, Johnson, and Sowman (2014) proposed a theory in which the "core" neurophysiological deficit in stuttering is based on a dysfunction

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within a brain network that supports internal timing, resulting in reliance of a secondary system that utilizes external timing cues to sequence speech movements.

Recent research supports the role of the basal ganglia thalamocortical (BGTC) network in rhythm processing and internal generation of a periodic timing signal (i.e., a beat) (Grahn, 2009; Grahn & Brett, 2007; Grahn & McAuley, 2009), as well as temporal prediction (Schwartze & Kotz, 2013). The BGTC network includes the basal ganglia (putamen), the supplementary motor area (SMA), and pre-motor and auditory regions. Recent findings from functional and structural magnetic resonance imaging (MRI) studies have shown that people who stutter may have deficient connectivity among brain areas that support auditory-motor integration, timing, and rhythm processing (Chang, Horwitz, Ostuni, Reynolds, & Ludlow, 2011; Lu et al., 2010). Moreover, a recent study reported that children who stutter have attenuated functional and structural connectivity in the BGTC network compared to age-matched controls (Chang & Zhu, 2013). One study examined brain activity during induced fluency conditions (i.e., reading in synchrony with a metronome beat and reading in chorus), compared to reading in solo in adults who stutter (Toyomura, Fujii, & Kuriki, 2011). The results showed that under solo reading conditions (where speakers who stutter were markedly disfluent compared to the induced fluency conditions), the basal ganglia (putamen), inferior frontal gyrus, and the other motor cortical regions within the BGTC network had significantly decreased brain activity compared to controls. During the metronome-timed speech condition, motor areas within the BGTC network heightened activity in the group who stuttered and thus the significant group differences observed during solo speech disappeared. In addition, the group who stuttered had bilateral increases in temporal cortex activity during both of the fluency inducing conditions (i.e., metronome-timed and choral speech). In sum, a growing body of work suggests a possible deficit in the BGTC network in people who stutter, and potential deficits in internal generation of rhythm that normally guides the timing of fluent speech. Related to this, it has been posited that impairment in basal ganglia function to produce timing cues may be a major deficit underlying stuttering (Alm, 2004).

We are not aware of any studies examining rhythm *perception* abilities in people who stutter; however, previous work has examined rhythm *production* abilities in this population. Studies comparing adults who stutter with controls on spontaneous or synchronize-continue tapping tasks have shown conflicting results; some finding group differences (e.g., Blackburn, 1931; Brown, Zimmermann, Linville, & Hegmann, 1990; Cooper & Allen, 1977), whereas others found no difference between the groups (e.g., Hulstijn, Summers, van Lieschout, & Peters, 1992; Max & Yudman, 2003; Zelaznik et al., 1994). Two studies conducted with children who stutter found mouth movements had greater timing variability (Howell, Au-Yeung, & Rustin, 1997) and clapping motions had more variable inter-clap-intervals (Olander, Smith, & Zelaznik, 2010) than typically-developing children. Both these studies suggest a fundamental deficit may exist in the ability to internally generate consistent rhythmic motor behaviors in children who stutter compared to typically-developing controls.

Although few studies have directly examined rhythm processing in individuals who stutter, rhythm processing has been examined in other clinical populations with known deficits in the BGTC network, such as in Parkinson's disease (Grahn & Brett, 2009). A number of studies have shown that simple metrical rhythms with a strong beat (i.e., explicitly marked within the sounds) are better discriminated, remembered, and reproduced than complex metrical rhythms with a weak beat (i.e., at least partially induced by the listener) (e.g., Grahn, 2012; Grahn & Brett, 2007; Povel & Essens, 1985). Consistent with the involvement of the BGTC network in rhythm processing and temporal prediction, Grahn and Brett (2009) showed that individuals with Parkinson's disease exhibit poorer rhythm discrimination and a reduced beat-based advantage compared with age-matched controls.

Stuttering is similar to Parkinson's disease in that the initiation and duration of movement execution are affected; specifically movements associated with speech production are affected in stuttering. Moreover, individuals who stutter may show a rhythm discrimination deficit similar to that observed for individuals with Parkinson's disease, given recent evidence of differences in the BGTC network in children (Chang & Zhu, 2013) and adults who stutter (Chang et al., 2011; Lu et al., 2010) compared to controls in areas previously shown to support rhythm processing (Grahn & Rowe, 2009). If so, these findings would support the hypothesis that people who stutter may have a deficit in rhythm processing.

To address this possibility, we compared auditory rhythm discrimination in children who stutter to typically-developing controls using a child-friendly version of a rhythm discrimination paradigm (Gordon, Shivers, Wieland, Kotz, Yoder, & McAuley, 2014). Based on previous research showing attenuated functional connectivity in the BGTC network in children who stutter compared to typically-developing controls (Chang & Zhu, 2013), we hypothesized that children who stutter would show worse rhythm discrimination than their matched controls. If so, this result would provide the first evidence of a rhythm perception deficit in developmental stuttering and support the view that a rhythm processing deficit may underlie developmental stuttering. Moreover, we hypothesized that the predicted group difference in overall rhythm discrimination may be larger for complex rhythms than simple rhythms because discrimination of the former may possibly rely more on internal beat generation than the latter.

2. Methods

2.1. Participants

Participants were 17 children who stutter (stuttering; 9 F, 8 M; age: $M = 8.70$ years, $SD = 1.55$) and 17 typically-developing children (control; 9 F, 8 M; age: $M = 8.79$ years, $SD = 1.53$) ranging from 6.08 to 11.42 years of age (see Table 1). The children were recruited through the Speech Neurophysiology Lab at Michigan State University. All children underwent careful screening to ensure normal speech and language development and typical developmental history except for the presence of stuttering in the stuttering group (see Table 1). Participants were monolingual, native speakers of English, with normal hearing, and without concomitant developmental disorders such as dyslexia, ADHD, learning delay, or other confirmed developmental or psychiatric conditions. The parents also confirmed that no child was taking any medication affecting the central nervous system. The children who stutter and typically-developing controls were matched in chronological age and sex, and did not differ in socioeconomic status (Hollingshead, 1975).

Research procedures were approved by the Michigan State University Institutional Review Board, and both the child and the parent signed informed consents. All participants were given nominal remuneration and small rewards (i.e., stickers) for participation.

2.2. Speech, language, hearing, and cognitive evaluation

Prior to participation in the current study, all participants were given a battery of standardized speech, language, and cognitive tests, audiometric hearing screening, and cognitive evaluations. These tests included the Peabody Picture Vocabulary Test (PPVT-4),