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Dissociations among linguistic, cognitive, and auditory-motor neuroanatomical domains in children who stutter

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ABSTRACT

The onset of developmental stuttering typically occurs between 2 to 4 years of age, coinciding with a period of rapid development in speech, language, motor and cognitive domains. Previous studies have reported generally poorer performance and uneven, or "dissociated" development across speech and language domains in children who stutter (CWS) relative to children who do not stutter (CWNS) (Anderson, Pellowski, & Conture, 2005). The aim of this study was to replicate and expand previous findings by examining whether CWS exhibit dissociated development across speech-language, cognitive, and motor domains that are also reflected in measures of neuroanatomical development. Participants were 66CWS (23 females) and 53CWNS (26 females) ranging from 3 to 10 years. Standardized speech, language, cognitive, and motor skills measures, and fractional anisotropy (FA) values derived from diffusion tensor imaging from speech relevant "dorsal auditory" left perisylvian areas (Hickok & Poeppel, 2007) were analyzed using a correlation-based statistical procedure (Coulter, Anderson, & Conture, 2009) that quantified dissociations across domains. Overall, CWS scored consistently lower on speech, language, cognitive and motor measures, and exhibited dissociated development involving these same measures and white matter neuroanatomical indices relative to CWNS. Boys who stutter exhibited a greater number of dissociations compared to girls who stutter. Results suggest a subgroup of CWS may have incongruent development across multiple domains, and the resolution of this imbalance may be a factor in recovery from stuttering.
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1. Introduction

Stuttering is conventionally considered to be a disorder associated with deficient speech motor control, the final step in linguistic and speech processing preceding overt speech production (Caruso, Gracco, & Abbs, 1987; Forster & Webster, 2001; Ludlow & Loucks, 2004; Max, Guenther, Gracco, Ghosh, & Wallace, 2004). While stuttering does not seem to be directly related to apparent higher order cognitive or linguistic difficulties, there are numerous theories, models (see Bloodstein, 2002, 2006; Karniol, 1995; Ratner, 1995) and investigations that point to a relationship between language processing and stuttering (Smith, Goffman, Sasisekaran, & Weber-Fox, 2012; Smith, Sadagopan, Walsh, & Weber-Fox, 2010; Spencer, Packman, Onslow, & Ferguson, 2005; but see Nippold (2012) for opposing view). For example, stuttering is more likely to

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occur on initial sounds and words (Brown, 1945; Wingate, 1982), consonants (Taylor, 1966), unfamiliar words (Hubbard & Prins, 1994), and during longer utterances (Robb, Sargent, & O'Beirne, 2009).

The loci of stuttering do not appear to be consistent across development. With age, there is a shift from stuttering more on function words to content words (Au-Yeung, Gomez, & Howell, 2003; however, see Buhr and Zebrowski, 2009; Howell, Au-Yeung, & Sackin, 1999; Rommel, 2001). Research suggests a link between linguistic development, cognitive capacity, and stuttering frequency; children who stutter (CWS) exhibit higher rates of disfluencies with increasing utterance length (Sawyer, Chon, & Ambrose, 2008; Zackheim & Conture, 2003). The interaction between factors such as length of utterance, and grammatical and syntactic complexity may be salient particularly during early speech development in CWS (Logan & Conture, 1995; Ratner & Sih, 1987; Watson, Byrd, & Carlo, 2011; Weiss & Zebrowski, 1992; Yaruss, 1999).

Some studies have hinted at weaker language abilities in CWS relative to children who do not stutter (CWNS). At similar developmental stages, CWS, particularly those with persistent stuttering, tend to have lower scores – although still within the norm – on standardized expressive and receptive assessments compared to CWNS and children who recovered from stuttering (Anderson & Conture, 2000; Coulter et al., 2009; Yairi, Ambrose, Paden, & Throneburg, 1996). However, other studies have reported average or above-average language skills in CWS. Watkins, Yairi and Ambrose (1999) reported expressive language skills that were above developmental expectations for CWS with earlier stuttering onset relative to those with later stuttering onset. Additionally, children who recovered from stuttering performed better than children with persistent stuttering on all language measures.

Language deficits that may exist in CWS may be more conspicuous in some domains compared to others, with some studies reporting poorer expressive than receptive language skills in CWS (Byrd & Cooper, 1989; Ntourou, Conture, & Lipsey, 2011; Ryan, 1992), a pattern that is opposite to the developmental trend reported in CWNS (Ryan, 1992). These language deficits may also extend to lexical skills (see Hall, 2004; for review). Similar to adults who stutter, CWS display poorer lexical ability, including reduced lexical diversity, relative to CWNS (Anderson & Conture, 2000; Newman & Bernstein Ratner, 2007; Silverman & Bernstein Ratner, 2002; Wagovich & Bernstein Ratner, 2007). Children who stutter showed slower speech reaction times during picture naming tasks relative to CWNS even when semantically primed (Pellowski & Conture, 2005). Relative to CWNS, CWS also show greater syntactic priming effects (that is, greater difference between primed and non-primed conditions) (Anderson & Conture, 2004) and greater frequency of stuttering with increasing syntactic complexity (Kadi-Hanifi & Howell, 1992). Taken together, these findings suggest a speech motor system in CWS that may be more vulnerable to breakdown with increased language processing demands.

1.1. Studies examining dissociations among speech, motor, and linguistic domains in children who stutter

One explanation for stuttering in early development is related to the notion of dissociations across multiple linguistic domains (Bates, Appelbaum, Salcedo, Saygin, & Pizzamiglio, 2003). Dissociations may appear as uneven abilities in disparate tasks. In CWS, dissociations or imbalances in the speech language system may prompt greater allocation of resources in reconciling these mismatches, and subsequently, result in fewer resources available for fluent speech (Anderson et al., 2005; Coulter et al., 2009). Evidence for these imbalances has been reported by a number of studies (Anderson et al., 2005; Coulter et al., 2009).

In a study of 20CWS between 3 and 5 years and their matched controls, Anderson and Conture (2000) reported greater incongruity between receptive/expressive skills and receptive vocabulary in CWS compared to CWNS, although no correlation was found between the magnitude of dissociation and stuttering frequency. In a subsequent study of similarly aged CWS ($n = 45$) and their matched peers ($n = 45$), Anderson et al. (2005) found greater likelihood of dissociations across several speech language domains (e.g., vocabulary, oral communication, comprehension) in CWS relative to CWNS. A replication of this study by Coulter et al. (2009) with 85CWS and their matched controls ($n = 85$) reported similar findings; CWS were more likely to exhibit dissociations involving receptive language and sound development skills relative to CWNS. It is essential to note that in both studies, some CWS did not present dissociations, while a small number of CWNS were found to exhibit dissociations, prompting the speculation that there may be subtypes in stuttering, and that incongruities in the speech language domain may not be the primary component in the manifestation of the disorder.

In their study, Anderson et al. (2005) found children with dissociations had lower receptive language skills compared to those without dissociations. However, Anderson et al. did not find any differences in the stuttering duration (time since stuttering onset) between CWS with and without dissociations, which in part may be related to the narrow range of ages (3 to 5 years) in their sample, a period when the trajectory of stuttering persistence versus recovery may not yet have been established. Accordingly, a longitudinal study that tracks the pattern of dissociation across a larger age range encompassing older age groups may uncover subtypes that are associated with persistence versus recovery from stuttering.

The idea that both language and motor components might be affected in stuttering (Peters & Starkweather, 1990; Van Riper, 1982), has prompted a number of studies to extend their evaluations beyond incongruities in the linguistic domain in CWS. A recent study by Hollister, Alpermann, & Zebrowski (2012) reported dissociations within the motor domains, specifically in speech motor articulation (diadochokinesis [DDK]) and speech rate for CWS ($n = 45$) between 4 and 7 years compared to CWNS ($n = 29$). In addition, CWS with dissociations between expressive language and speech-motor abilities (as measured by DDK rate) exhibited more stuttering-like disfluencies (SLD). In another study examining sentence repetition performance in CWS between 4 and 6 years, MacPherson and Smith (2013) reported greater variability in lip aperture movement (a reflection of upper lip, lower lip, and jaw movement coordination as a function of time) for longer sentences

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