Research Article

The Role of Effortful Control in Stuttering Severity in Children: Replication Study

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Background: In 2014, Kraft et al. assessed the temperament, home environment, and significant life events of 69 North American children who stutter to examine the combined and compounded effects of these individualized factors on mediating overt stuttering severity. The temperament domain of effortful control was singularly found to be significantly predictive of stuttering severity.

Purpose: Because of the clinical significance of the initial study's findings, a replication study with a different, larger cohort of children who stutter was warranted to validate the reported outcomes.

Method: The current study assesses 98 children who stutter, ages 2;4 to 12;6 (years; months, $M = 6;7$), recruited from Perth, Australia.

Results: The results support the previous findings of Kraft, Ambrose, and Chon (2014), with effortful control remaining the sole significant contributor to variability in stuttering severity, as rated by both parents and clinicians.

Conclusion: These cumulative and consistent outcomes support the need to develop targeted intervention strategies that specifically strengthen aspects of effortful control as a means to support positive therapeutic change in children who stutter.

The etiology of developmental stuttering is multifactorial and yet to be fully understood (Bloodstein & Bernstein Ratner, 2008). Accordingly, the factors contributing to the variability of stuttering severity between, and within, children who stutter (CWS) appears similarly complex (Yaruss, 1997). Kraft, Ambrose, and Chon (2014) postulated that a child's individual temperament profile may influence the severity of their stutter as he or she interacts with, and responds to, major life events and their daily environment. Their initial study assessed 69 North American CWS, 2;4 to 5;9 (years; months, $M = 3;7$), and evaluated the impact that these factors might have on the generalized overt severity of stuttered speech (Kraft et al., 2014). The interaction of these variables was hypothesized to have an additive effect and would be reflected in parent- and clinician-rated stuttering severity scores for these children. The results of the study, however, were surprising. A single domain of temperament, known as *effortful control* (EC), was found to be significantly predictive of stuttering severity (Kraft et al.,

2014) with no other measure showing additive or predictive significance. Given the implications of such findings for the clinical management of stuttering, a follow-up study was pursued to see if these results would be replicated in a new larger cohort of children diagnosed with developmental stuttering from Perth, Australia.

Stuttering and Severity

Developmental stuttering is a disorder that disrupts speech fluency (Bloodstein & Bernstein Ratner, 2008). Typically, CWS encounter an onset of symptoms anywhere between 2 and 6 years of age (Yairi & Ambrose, 2013) and exhibit an overt speech profile that includes repetitions, sound distortions, and/or blocking (Van Riper, 1982; Wingate, 1964). As we learn more about the onset and developmental trajectory of stuttering, traditional incidence estimates of approximately 5% are now being revised to rates in excess of 8% (Dworzynski, Remington, Rijdsdijk, Howell, & Plomin, 2007; Reilly et al., 2009; Yairi & Ambrose, 2013). Indeed, a longitudinal Australian study reports a cumulative incidence rate of 11% by 4 years of age (Reilly et al., 2013). Should this be applied to the current estimated number of 4-year-old children residing in the United States (3,987,656; US Census Bureau, 2015), approximately 438,642 will have stuttered during some stage in their development, with approximately one fifth (approximately 88,000) likely to persist into adulthood. Within this hypothetical population, the relative severity of each child's stutter will vary

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considerably, ranging from a mild disfluency with minimal negative impact on the child's ability to speak to a severe disfluency that significantly impedes verbal communication. Indeed, for those children whose stutter persists, the severity of their stutter can impact much more than their speech (O'Brian, Jones, Packman, Menzies, & Onslow, 2011).

The severity of an individual's stutter has been found to influence peer acceptance (e.g., an individual with a mild stutter is described more positively than an individual whose stutter is more severe, after just one brief observation; Gabel, 2006). The overt, or observable, characteristics of an individual's stutter are just one component included in a global evaluation of stuttering severity, with covert stuttering (e.g., avoidance, emotional response) an important variable to consider (Bloodstein & Bernstein Ratner, 2008; Guitar, 2014). Stigmatized individuals are seen to possess characteristics that are undesirable and/or different from social norms. People who stutter are susceptible to stigmatization, through negative stereotypes and discrimination, which can have a substantial adverse effect (Craig, Blumgart, & Tran, 2009; Link & Phelan, 2006). Bullying, victimization, and negative social experiences associated with childhood stuttering have all been well documented in the literature (Betz, Blood, & Blood, 2008; Blood & Blood, 2004, 2007; Blood et al., 2011; Davis, Howell, & Cooke, 2002; Hartford & Leahy, 2007; Langevin, Packman, & Onslow, 2009; Mooney & Smith, 1995; Woods & Williams, 1976). There is also evidence that stuttering affects academic performance (Williams, Melrose, & Woods, 1969), with individuals suffering the difficult consequences of bullying and poor self-esteem, thus confounding their educational experience (Hugh-Jones & Smith, 1999). Such individuals become even less able to communicate effectively (Crichton-Smith, 2002) and have increased difficulties concentrating on learning while also worrying about, and attempting to control, stuttering behaviors (Daniels, 2007). As such, studies have identified an inverse relationship between stuttering severity and level of educational attainment and self-perceived communicative competence (O'Brian et al., 2011). We surmise, the more severe the child's stutter is, the more likely they will be perceived as "different" with an increased liability for experiencing stigmatization by those in their daily living environment.

Links between stuttering severity and anxiety, self-esteem, and other factors that affect quality of life (QoL) outcomes have been explored with some equivocal results (Ezrati-Vinacour & Levin, 2004; Mulcahy, Hennessey, Beilby, & Byrnes, 2008). Recent studies have found that factoring in the role of temperament as a potential mediator highlights the relationship temperament has on perceived stuttering impact. Koedoot, Bouwmans, Franken and Stolk (2011), for example, found that there are stronger relationships between severity and QoL when outcomes were analyzed in the context of individual coping styles. Overall, the study suggested that, the more coping strategies an individual could flexibly apply to various stressful circumstances, the greater their QoL scores were measured to be.

From a clinical perspective, the severity of a child's stutter can be a significant factor in prompting parents to

seek services or connect with and participate in recommended treatment programs. Consistent monitoring of global severity ratings over time—by both parents and clinicians—also assists clinicians to gauge each child's responsiveness to intervention and facilitates the planning of appropriate treatment strategies for ongoing management of the disorder (Guitar, 2014; Yaruss, 1997).

Temperament

Contemporary literature describes temperament like many other heritable traits: determined genetically and evident early in development but maturing over time and influenced by our experiences (Bates, Freeland, & Lounsbury, 1979; Henderson & Wachs, 2007; Putnam, Ellis, & Rothbart, 2001; Rothbart & Bates, 2006). The domains of temperament define individual differences in motor, attentional, and emotional reactivity to stimuli and self-regulation processes that serve to inhibit reactivity (Henderson & Wachs, 2007; Rothbart & Derryberry, 1981). Studies using terms such as *emotional regulation/reactivity*, *inhibitory control*, *behavioral regulation*, *positive/negative affectivity*, *attentional/executive control*, and/or *perceptual sensitivity* are typically referring to one or more components of an individual's overall temperamental profile (Henderson & Wachs, 2007; Rothbart & Bates, 2006).

According to Rothbart and Bates (2006), there are three major domains of temperament: (a) surgency/extraversion, (b) negative affectivity, and (c) EC. *Surgency/extraversion* refers to reactivity factors, such as sociability, enjoyment of high-intensity pleasures, impulsivity, and activity levels. Negative affectivity is also reactive and includes fear, discomfort, anger/frustration, and sadness. Both reactive factors of temperament begin to emerge in the early months of development (Rothbart, Ahadi, & Evans, 2000; Rothbart, Ahadi, Hershey, & Fisher, 2001; Rothbart, Sheese, & Posner, 2007). The final factor, EC, is self-regulating through volitional and nonvolitional aspects and is thought to emerge at approximately 2 to 3 years of age (Rothbart et al., 2007; Rueda, Posner, & Rothbart, 2005). EC is defined as "the ability to inhibit a dominant response in order to activate a subdominant response" (Rothbart et al., 2007, p. 3). It is a critical aspect of temperament that is linked with the development of consciousness (Kochanska, Murray, & Harlan, 2000). As it matures, children become more able to consciously direct their attention, inhibit their behavior to follow set instructions, and cope with stimuli, enabling them to plan more appropriate responses and regulate reactive emotions (Rueda & Rothbart, 2009; Simonds, Kieras, Rueda, & Rothbart, 2007).

Although temperament is constitutionally based, it can only be inferred through observing behaviors that reflect an individual's cognitive and emotional traits (Goldsmith, 1996; Rothbart et al., 2000). Consequently, research into temperament predominantly relies on data collected through clinical observation and/or comprehensive questionnaires (Goldsmith, Buss, & Lemery, 1997; Rothbart et al., 2001). The most common method of data collection is in the form

of behavior questionnaires that reflect broad profiles of temperament, completed by parents or primary caregivers (Bates et al., 1979; Carey, 1970; Carey & McDevitt, 1978; Goldsmith, 1996; Henderson & Wachs, 2007).

Temperament and Stuttering

There has been a notable increase in the number of studies investigating behavior, personality, and temperament within the realm of developmental stuttering in recent years (Alm, 2014; Ambrose, Yairi, Loucks, Seery, & Throneburg, 2015; Anderson, Pellowski, Conture, & Kelly, 2003; Bleek et al., 2012; Bush, 2006; Choi, Conture, Walden, Jones, & Kim, 2016; Choi, Conture, Walden, Lambert, & Tumanova, 2013; Clark, Conture, Walden, & Lambert, 2015; Eggers, De Nil, & Vanden Bergh, 2010, 2012, 2013; Embrechts et al., 2000; Fowle & Cooper, 1978; Glasner, 1949; Haitani & Kumano, 2016; Hollister, 2015; Iverach et al., 2016; Johnson, Walden, Conture, & Karrass, 2010; Karrass et al., 2006; Kefalianos, Onslow, Block, Menzies, & Reilly, 2012; Kefalianos, Onslow, Ukoumunne, Block, & Reilly, 2014, 2017; Ntouriou, Conture, & Walden, 2013; Schwenk, Conture, & Walden, 2007; K. A. Smith et al., 2017; Tumanova, Zebrowski, Throneburg, & Kulak Kayikci, 2011; Walden et al., 2012). The vast majority of these studies have employed a case-control design to identify whether the temperament of CWS differs significantly from that of their fluent peers. Although there are differing findings in this area of research, a broad perspective on this body of work suggests that CWS (compared with fluent peers) have increased difficulty in the regulation and/or adaptation of their behavioral and emotional responses to everyday scenarios within a variety of contexts (Alm, 2014; Kefalianos et al., 2012), which leads to increased emotional reactivity to stressful stimuli (Alm, 2014; Eggers et al., 2010, 2012, 2013).

Temperament and Stuttering Severity

The literature is sparse with regard to investigations of stuttering severity and temperament. It is very difficult to find studies aimed at understanding individual variability especially within sample populations and profiles of children who all stutter. Thus, we must draw our inferences from studies that compare CWS with their typically developing peers.

Schwenk et al. (2007) assessed the attentional control and adaptation to background stimuli of preschool CWS and compared it with that of their fluent peers. Within the group of preschool CWS, they found moderately significant correlations with both distractibility and attentional control, against the child's rate of observed disfluencies (stutter like, nonstutter like, or total). The effect sizes associated with these correlations were reported to be considerable. Another study used a "disappointing gift" to assess the emotional regulation of preschool children who do and do not stutter (Johnson et al., 2010). This study found that CWS exhibited a significant increase in nonstuttering-like

disfluencies following the receipt of a *desired* gift, compared with baseline or the receipt of a disappointing gift. An increase in stutter-like disfluencies was also noted following the receipt of a desired gift, but the differences from baseline were not found to be significant (Johnson et al., 2010).

Arnold, Conture, Key, and Walden's (2011) study examined the use of four emotional regulation strategies (i.e., distraction, self-stimulation, cognitive restructuring, and instrumental coping) following emotional stimulation (positive, negative, and neutral), on the rate of disfluency for CWS compared with their fluent peers. Their results highlighted a noticeable reduction in duration and frequency of regulatory strategies that coincided with an increase in severity for CWS (Arnold et al., 2011). This study, although able to offer insight into the relationship between regulatory strategies and speech fluency for CWS, did not include measures of general emotional reactivity as a mediating variable (e.g., greater emotional reactivity may inhibit/stimulate use of regulatory strategies; Arnold et al., 2011) and does not suggest, in agreement with the current authors, that temperament is causal of stuttering but supports an interplay between speech fluency and emotional regulation. A study testing the dual diathesis-stressor framework outlined by Walden et al. (2012) addressed the potential interplay of regulatory strategies, emotional reactivity, and speech fluency. Researchers collected measures of parent-reported emotional reactivity and, then, introduced emotional stress conditions (positive, negative, and neutral) via overheard conversations, after which they recorded rates of stuttered disfluencies through a narrative task. Within the CWS only, children who exhibited greater negative emotion while speaking stuttered more frequently. That is, unless the negative emotion was also followed by self-regulatory behaviors, then no increase in severity was noted (Walden et al., 2012). To investigate this further, a study by Choi et al. (2016) indicated that positive emotional reactivity in CWS was positively associated with severity of stuttering, regardless of the emotional stressors introduced. Negative reactivity in CWS, on the other hand, was only positively associated with increased stuttering under positive emotional stress conditions (Choi et al., 2016). The authors of both studies concluded that changes in emotional stimuli affected the capacity of preschool CWS to remain fluent, with the child's emotional reactivity predicting the conditions under which they would be most affected. Ntouriou et al. (2013) conducted a similar study but employed more active emotion-eliciting tasks (as opposed to the passive audio- or video-watching paradigm employed in the previous two studies). Narratives were recorded after each time CWS and controls were asked to complete two tasks, one neutral and one designed to elicit frustration. Within the CWS cohort, the emotion regulation behaviors of "distraction" and "self-speech" resulted in opposing influences on the percentage of stuttered disfluencies observed in the subsequent narrative task. In this instance, the use of self-speech was positively correlated with stuttering frequency, whereas the use of distraction was negatively correlated (Ntouriou et al., 2013). These results indicate that some self-regulatory strategies

may be more effective than others in supporting fluent speech in the context of emotional stressors.

In contrast, other studies have been unable to find any significant correlation between stuttering severity and the temperament (Eggers et al., 2010; Tumanova et al., 2011). The assessments used by Eggers et al. (2010) were similar to Kraft et al. (2014) with the use of the Children's Behavior Questionnaire (CBQ)–Short Form (Dutch version) to collect temperament profiles, but only clinical ratings of stuttering severity were collected. Eggers et al. (2010) noted that limitations of their study might have influenced their findings, as they did not have data to explore differential correlations of the various elements contributing to stuttering severity. Tumanova et al. (2011) specifically assessed the relationship between CBQ outcomes with the average duration of sound prolongations from a single speech sample recorded in a clinical setting. Despite their null outcomes, both authors acknowledged that temperament is likely an important factor within biopsychosocial models of stuttering, with researchers yet to fully understand how this is so.

Choi et al. (2013) investigated differences between CWS and their nonstuttering peers in behavioral inhibition, a characteristic of temperament that can be measured through observation and latency (to the sixth spontaneous comment) in a conversation with an unfamiliar examiner. Although they found no significant difference in measures of behavioral inhibition between preschool children who do (CWS) and do not stutter (CWNS), they did observe differences within the CWS group (Choi et al., 2013). Again, contrary to the findings of Kraft et al. (2014) and others, CWS who were more behaviorally inhibited produced more stutter-like disfluencies than CWS who demonstrated less behavioral inhibition.

Given that temperament, and more specifically EC, matures over time, adult studies that address any potential relationships with stuttering severity are relevant and of interest. A recent experimental study comparing the attentional function of adults who stutter (AWS; $n = 17$) and adults who do not stutter (AWNS; $n = 18$) found that attentional function did not significantly differ between the groups (Haitani & Kumano, 2016). However, Haitani and Kumano (2016) did find that, within the AWS group, individuals who had faster reaction times and gave more incorrect responses stuttered more severely. These findings support the notion that, even into adulthood, AWS who exhibit greater reactivity to stressors within their environment are less able to produce fluent speech.

Although it is evident that EC regulates one's emotional response, its involvement in attention and cognition is also of interest. Attention and cognition have historically served as the foundation for many clinical intervention strategies (Nye et al., 2013). Laboratory measures of orientation and executive attention have been correlated with ratings of EC (Rothbart, Ellis, Rueda, & Posner, 2003), and a recent review of EC and other self-regulation–related constructs has found that EC best reflects trait-level mapping of cognitive control and, in particular, executive attention (Nigg, 2017). By establishing the presence of significant and measurable contributing

factors to stuttering severity, researchers and clinicians would have an opportunity to guide the development of more effective and efficient individualized treatment programs for CWS.

Home Environment and Life Events

Although temperament is biologically based and described as being predominantly stable, the development of an individual's temperament can be mediated by their life experiences (Henderson & Wachs, 2007; Plomin & Asbury, 2005). Consequently, it is prudent to assess temperament within the context of the child's external environment and significant life experiences.

While temperament is the unique filter through which we experience all life's challenges, it is also, in part, a product of our daily environment. Each child's home environment can vary significantly across living conditions, culture, and experiences—contributing to differences in environmental stress levels, from one family to the next. Chaotic home environments (e.g., homes with high noise levels, disorganization, confusion, and less routine) have been shown to mediate the relationship between socioeconomic status and different aspects of social and emotional development, including self-regulation (Evans, Gonnella, Marcynyszyn, Gentile, & Salpekar, 2005). For example, a child living in a chaotic home is less likely to receive support to develop foundational skills of self-efficacy and self-regulation, increasing their vulnerability to poorer cognitive skills, aggression, impulsivity, and internalization/externalization of emotional issues (Coldwell, Pike, & Dunn, 2006; Dumas et al., 2005; Shamama-tus-Sabah & Gilani, 2011). The extent to which home environments and genetics influence the development of a child's temperamental profile was the focus of a recent large-scale study (Lemery-Chalfant, Kao, Swann, & Goldsmith, 2013). Through the assessment of 807 twin pairs, Lemery-Chalfant et al. (2013) found that home environments were less chaotic for children with high EC, whereas children with high surgency/extraversion experienced greater levels of chaos in their home environments. Both of these correlations were found to be genetically mediated. As such, this study found that home environments both facilitated and suppressed the heritability of different aspects of a child's temperament, that is, surgency/extraversion was more heritable and EC was less heritable in a chaotic home (Lemery-Chalfant et al., 2013). As developmental stuttering is also a heritable trait (Kraft & Yairi, 2012), the interplay between gene expression, temperament, and home environment is particularly relevant for researchers examining the variables associated with the severity of stuttering in children.

Some clinicians working with CWS have heard parents and caregivers draw connections between significant life events (e.g., changing day care, birth of sibling, new school, divorce) and their child's ability to achieve and maintain speech fluency (Langevin et al., 2009). Significant life events—both positive and negative—can be stressful, as they are capable of upsetting an individual's psychological and physiological equilibrium (Beautrais, Fergusson, & Shannon, 1982; Garnezy & Masten, 1990; Heisel, Ream,

Raitz, Rappaport, & Coddington, 1973). Behavioral and neurobiological researchers report that encountering moderate levels of stress promotes the development of resilience as they cope with, and overcome, stress-inducing events (Masten, 2001; Russo, Murrough, Han, Charney, & Nestler, 2012). Yet, there are significant detrimental effects on child behavior, coping mechanisms, and social-emotional development if a child is exposed to repeated events that are highly stressful (Donaldson, Prinstein, Danovsky, & Spirito, 2000; Russo et al., 2012). The number of stressful events experienced during childhood highly influences the risk of the child developing psychological disturbances (Höök, Hägglöf, & Thernlund, 1995). This effect, again, appears to be mediated by a child's temperament, as a large-scale longitudinal study conducted in Sweden found children with an easy temperament exhibit greater resilience against high levels of adversity (Agnafors et al., 2016).

In spite of the strong interconnectedness between temperament, significant life events, and daily home environments, no other studies have addressed the influence of temperament on childhood stuttering severity within the same holistic context since Kraft et al. (2014).

The Original Study

The initial study by Kraft et al. (2014) predicted that an additive effect of the variables—home environment, major life events, and child temperament—would produce a significant relationship with global severity ratings of CWS. Discovering that EC, alone, significantly predicted stuttering severity in CWS was an unexpected result, with considerable implications for the clinical management of CWS (Kraft et al., 2014).

The negative correlation discovered between stuttering severity and EC scores showed low EC to be significantly correlated with higher stuttering severity ratings. As mentioned earlier, low EC is related to poorer regulation of executive attention. Certain individuals have an inability to switch attention while actively engaged. For these individuals, “deficit response modulation is not a consequence of their motivational state, but a fundamental deficiency that impedes self-regulation across a wide range of situations” (Newman & Wallace, 1993, p. 707). Following this theory, children with greater EC are simply more able to voluntarily focus, shift attention, and inhibit or activate their behaviors. One possibility is that these children are able to reduce the severity of their stutter by naturally responding to aberrant speech with shifts in their attention from the stutter itself to the employment of compensatory mechanisms. EC in CWS would likely be demonstrated with environmental stressors as well, as children who have high EC are better able to cope with changes in their environment. This potentially reduces the demands on their cognitive system, freeing up resources for online processing of speech, which may result in greater fluency. Both reactivity and self-regulation are important factors in the development of self-monitoring and awareness in young CWS. Children who are more readily overwhelmed by their attentional,

emotional, and behavioral responses are hypothetically less able to manage stuttered speech or reduce its severity. Rothbart, Ahadi, and Hershey (1994) have proposed that aspects of EC support flexible responsiveness to stimuli that may result in similar behaviors. For example, a child with low reactivity and poor self-regulation may also produce more stuttering behaviors without suppression or flexibility, thus increasing the overt stuttering severity observed. Overall, EC appears to reflect a person's reliance upon emotional self-regulation for the management of their personal and environmental stress (Eisenberg & Spinrad, 2004), with children who exhibit poor EC experiencing greater levels of personal distress (Valiente et al., 2004).

CWS who have not developed these necessary mechanisms will be more frequently taxed beyond their capacities by a variety of stimulus changes, including environmental, life events, and home environments, resulting in more frequent breakdowns of their speech. This may explain why neither home environments nor major life events contributed independently to stuttering severity in the original study by Kraft et al. (2014). For some children with low EC, their reactivity is sensitive enough to respond to typically unremarkable home environments and/or life events. Most instruments only measure major life events, but these children may respond to relatively minor life events (e.g., losing a specific toy), which can be difficult to measure and will likely vary from one child to the next. Furthermore, Kraft et al. (2014) reflected that EC likely mediates situational severity by the quantity and intensity of “in the moment” demands placed on the child. Thus, EC alone—rather than the interaction of temperament, home environment, and life events—significantly predicted stuttering severity in their study.

Goal/Hypotheses

The primary aim of this study was to reexamine the interaction of stuttering severity in children with variables of temperament, home environment, and the presence of significant life events.

This replication study hypothesized that the statistical relationships found in the original study would remain consistent, despite the larger population recruited from a different geographical location. Investigators predicted ratings of stuttering severity in CWS that were negatively correlated with the temperamental domain of EC. Despite the results of the original study, participants' home environment profiles and the presence of significant life events were also explored. The literature largely remains supportive for interaction or additive effects, and as a replication, fidelity with the original study's data analysis was necessary.

Method

Children/Participants

Ninety-eight CWS (30 female and 68 male, ages 2;4 to 12;6, mean age of 6;7) and their parents were recruited through the Curtin Stuttering Treatment Clinic, Curtin University, Perth, Western Australia.

The measures of interest were obtained during a single visit, with the child participants and their parent(s) present. Data collection continued over a period of 4 months through the Curtin Stuttering Treatment Clinic. All children who satisfied the inclusion criteria during this time were included in the current study.

The Australian participant inclusion criteria was kept consistent with the previous study, which were as follows: (a) The child is regarded as exhibiting stuttering both by parent(s) and by a clinically certified speech-language pathologist experienced and specialized in the assessment and treatment of childhood stuttering; (b) the child has a stuttering severity rated as 1 or greater on an 8-point scale (0 = *none*, 7 = *profound*) by clinicians at the time of evaluation; (c) the child is considered within normal limits across other speech and language domains, determined upon intake to the clinic; (d) the child was initially seen by clinician within 12 months poststuttering onset; and (e) the child has no history of hearing loss or neurological impairment. All participants in this study were unpaid volunteers.

Data Collection

All methods of data collection were, wherever possible, kept consistent with the previous study. Some adjustments, however, were necessary in order to accommodate the larger number of participants recruited and the time available to collect the required data. The hearing screening and standardized language assessments that were part of the U.S. study were not included as they were initially administered as part of a larger longitudinal study and would not provide relevant data to address the hypotheses for this study. In its place, the treating clinician and/or clinical supervisor overseeing the child's treatment program provided the clinician report severity rating. The participants' comprehensive most recent clinical records were also referenced to further substantiate the clinician report severity ratings that were provided at the same time to ensure that they were representative. In the previous study, parent report child stuttering severity ratings were obtained separately from both mothers and fathers, when possible, for each child. However, upon analysis, the ratings provided by both parents were very highly correlated. Accordingly, the investigators therefore decided to obtain a severity rating from just one parent or caregiver per child.

During data collection, parents of CWS were asked to complete a short case history with an investigator. The case history included questions about their child's speech and current level of overt stuttering severity. As in the previous study, parent(s) reported their child's stuttering severity globally, as a daily "overall average," based on an 8-point scale (Yairi & Ambrose, 2005), where 0 is *normally fluent* and 7 is *very severe* (see Figure 1). Parents were asked to think "globally" to provide an overall severity rating of their child's stutter, with considerations to frequency, duration of stuttering events, observed tension during stutters, and the involvement of secondary characteristics. The parents recruited through the Curtin Stuttering Treatment Clinic were all experienced in using this stuttering severity rating

system to evaluate and record their child's fluency based on observations in clinic and at home on a daily basis. Half-numbers were accepted.

The following three questionnaires were given to the parent(s) of the child participants, and the majority were completed within the one data collection session. If more time was required, some parents completed the questionnaires at home and returned them within the week. The data for this study were collected in conjunction with a larger international project to uncover the genes associated with developmental stuttering supported by the National Institutes of Health (R03DC015329) and was approved by the Curtin Human Ethics Review Committee and Wayne State University's Institutional Review Board (IRB#091212MP2E).

External Environment Measures: Life Events Checklist/Confusion, Hubbub and Order Scale

Measures of children's external environments were obtained through the Life Events Checklist (LEC; Sarason, Johnson, & Siegel, 1978) and the Confusion, Hubbub and Order Scale (CHAOS; Matheny, Wachs, Ludwig, & Phillips, 1995). The LEC was developed to measure events occurring in the child's life, characterized as either "good" or "bad" for the child (Sarason et al., 1978). Potentially traumatic events are significantly correlated with measures of psychological distress, and the LEC is regarded as a reliable method of measuring exposure to such events (Gray, Litz, Hsu, & Lombardo, 2004). Parents are asked to circle "yes" or "no" to major life events that have occurred within the last 12 months (e.g., moving, divorce, death of family member, birth of sibling), indicating whether the event was perceived to be good or bad for the child in question. Studies pertaining to the reliability and psychometric properties of the LEC have found it to be a temporally reliable assessment, demonstrating strong convergence with established measures of psychopathology that are associated with exposure to traumatic events (Brand & Johnson, 1982; Gray et al., 2004).

The CHAOS is an assessment of everyday home environments, incorporating aspects across daily routine, organization, cleanliness, communication, and noise level (Matheny et al., 1995). To complete this assessment, 15 items were rated by the parent(s) with whom the child lives most of the time. Each item is rated on a 5-point scale, with "1" indicating *not at all like my home* and "5" indicating *very much like my home* (Matheny et al., 1995). The developers of the CHAOS have reported an internal consistency (Cronbach's α) of .79, with test-retest reliability over a 12-month span of .74 (Dumas et al., 2005). This validity of this assessment was also found to be significant across all variables following evaluation against the Purdue Home Simulation Inventory (Matheny et al., 1995).

Child Temperament Measure

The CBQ was designed to provide measurements of child temperament traits (Putnam & Rothbart, 2006). The short form version of the CBQ scale consists of 94 items

Figure 1. Scale used by clinicians and parents to rate stuttering severity in children who stutter.

Stuttering Severity							
0	1	2	3	4	5	6	7
No Stuttering, Normally Fluent	Rarely	Mild		Moderate		Severe	

to be completed by the child's parent or primary caregiver. Each item is rated as follows: 1 = *extremely untrue*, 2 = *quite untrue*, 3 = *slightly untrue*, 4 = *neither true nor untrue*, 5 = *slightly true*, 6 = *quite true*, 7 = *extremely true*, with a "not applicable" option available also. The scale evaluates the child across 18 different behavioral dimensions: activity level, anger/frustration, attentional focusing, approach, discomfort, smiling and laughter, sadness, impulsivity, high-intensity pleasure, inhibitory control, fear, low-intensity pleasure, shyness, EC, perceptual sensitivity, falling reactivity, surgency/extraversion, and negative affectivity (Putnam & Rothbart, 2006). An examination of the CBQ's internal consistency showed that it reached the standard of 0.70 in 11/12 scales, with "sadness" reaching 0.65, which is still considered acceptable (DeVellis, 2017).

Although this questionnaire was designed and norm referenced for children aged 3 to 7 years, its use outside of this age range was deemed appropriate in this context as compiled raw scores (i.e., not standardized against age-related norms) were employed in correlational analysis and linear modeling.

Measurements

Scoring Questionnaires

All scores were calculated by entering questionnaire de-identified responses into computerized templates.

Reliability of Severity Measures

All clinician-reported severity ratings were obtained from certified speech-language pathologists who specialize in fluency disorders. Each clinician was familiar with a proportion of the participants' stuttering profiles and treatment histories. The parents recruited through the Curtin Stuttering Treatment Clinic were all trained and reliably experienced in using a stuttering severity rating system to evaluate and record their child's fluency measures.

Statistical Analysis

All statistical analysis was kept consistent with the previous study to optimize the replication reliability of results.

All measures were tested for normality. If a Shapiro-Wilk test revealed that a distribution violated the assumption of normality, the larger sample size (i.e., > 50) meant that parametric testing might still be appropriate if the calculated z scores of skewness (Z_{Skewness}) and kurtosis (Z_{Kurtosis}) were within appropriate bounds. For samples sized $50 < n < 300$,

an absolute z score of < 3.29 reflects a normal distribution, at an alpha level of .05 (Kim, 2013; Mudholkar, Marchetti, & Lin, 2002). Parent- and clinician-reported severity ratings were compared through descriptive analysis (mean and standard deviation) and a paired t test. Further correlation analysis was applied to test whether the severity ratings, as well as all independent variables, were significantly correlated with each other.

In order to address the hypotheses regarding the multiple contributions and interactions of child temperament, home environment, and life events on children's stuttering severity (parent and clinician ratings), a hierarchical linear regression along with interaction effects was applied. The SPSS 22.0 software program (SPSS Statistics; International Business Machines, 2013) was used for all statistical analysis.

Results

Stuttering Severity Ratings: Parents and Clinicians

The parent- and clinician-reported severity rating distributions were tested for normality. Absolute z scores of skewness and kurtosis were < 3.29 for both parent-reported ($Z_{\text{Skewness}} = 1.07$, $Z_{\text{Kurtosis}} = .13$) and clinician-reported ratings ($Z_{\text{Skewness}} = 2.87$, $Z_{\text{Kurtosis}} = .08$).

Table 1 shows that a paired t test, two tailed, revealed a significant difference between the two severity ratings, $t(97) = -2.89$, $p = .005$. It demonstrated that Australian parent-reported severity ratings were, on average, significantly lower than the clinician-reported severity ratings of the same children. However, a Pearson r correlation analysis of parent- and clinician-reported severity ratings found that they were highly correlated, $r = .759$ ($p < .001$).

The significant difference found between parent- and clinician-reported severity ratings is consistent with the previous study. However, mothers who participated in the U.S. study, on average, reported that their child's severity is significantly higher than did the clinician.

Table 1. Descriptive statistics of severity ratings ($n = 98$).

Rating	<i>M</i>	<i>SD</i>	<i>t</i>	<i>df</i>	<i>p</i>
Parent severity rating	2.95	1.34	-2.89	97	.005*
Clinician severity rating	3.24	1.49			

* $p < .01$.

Relationship Between Children's Stuttering Severity, Home Environment, Life Events, and Temperament

Age

Correlations with age were assessed using nonparametric means, as it violated both Shapiro–Wilk ($p < .001$) and the skewness test of normality ($Z_{\text{Skewness}} = 4.24$). No significant correlations were found between participants' ages and other major variables using Spearman rho.

LEC

The distribution of the LEC was also not normal ($Z_{\text{Skewness}} = 5.29$) and, thus, was assessed using Spearman rho, revealing a significant positive correlation with CHAOS scores ($r_s = .250, p = .013$).

CHAOS

As the distribution of CHAOS scores did not violate Shapiro–Wilk's assumption of normality ($p = .061$), Pearson r correlations were used between this and other normally distributed outcomes. The temperament domains of negative affectivity and EC were both identified as significantly correlated with CHAOS (see Table 2). A positive correlation was shown between CHAOS and negative affectivity ($r = .202, p = .046$), and a negative correlation was found with EC ($r = -.243, p = .016$).

Temperament

All three major domains met Shapiro–Wilk's assumptions of normality ($p = .323, p = .758$, and $p = .070$), so correlational analysis with other normally distributed variables was conducted using Pearson r . As stated earlier, negative affectivity was positively correlated with CHAOS scores ($r = .202, p = .046$), and a correlation between the temperament domain of EC and CHAOS measures also reached significance ($r = -.243, p = .016$), describing a negative relationship. The domain of surgency/extraversion was not significantly correlated with any other major variable in this study. Only EC was significantly correlated with

parent- ($r = -.289, p = .004$) and clinician-reported ($r = -.369, p < .001$) severity ratings (see Table 2).

Parent's Scores and Parent Severity Rating

In applying a hierarchical linear regression model (HLM), the CBQ scores of child temperament were compiled into three primary domains (surgency/extraversion, negative affectivity, and EC).

The hierarchy of models conformed to analysis performed in the original study. As EC would likely be the strongest predictor among other variables for children's stuttering severity, it was placed as the sole variable in Model 1. In the second model, the temperament domains of the Surgency/Extraversion and Negative Affectivity subscales were added. In the third and final model, CHAOS and LEC scores were added (see Table 3).

The HLM results for parent's scores and parent-reported severity, described in Table 4, have indicated that all three models were significantly predictive of the children's severity rating ($F = 8.733, p = .004$ for Model 1; $F = 3.671, p = .015$ for Model 2; and $F = 2.895, p = .018$ for Model 3). However, as shown in Table 3, EC was the only significant contributor to the model and, therefore, is the sole predictor of parent-reported severity ratings.

Further analysis of Model 1 was conducted to assess the relative contribution of EC in variability of stuttering severity. EC was found to account for 8.3% of the variation of parent-reported severity (see Figure 1). Although still highly significant, this contribution is lower than what was reported in the previous study, where EC accounted for 20.1% of the variance in mother-reported severity (see Figure 2).

Nonetheless, the results of the HLM and correlation analysis reveal consistencies with the previous study, demonstrating that stuttering severity is significantly correlated with EC in CWS. All other potential variables reported by parents that could contribute to stuttering severity did not reach significance, and no significant interaction effects were observed among any of the variables.

Table 2. Correlations among major variables.

Variable	Age (months) ^a	CBQ: effortful control	CBQ: surgency/extraversion	CBQ: negative affectivity	Parent severity rating	Clinician severity rating	Total life events (LEC) ^a	CHAOS
Age (months) ^a	—	-.116	.066	.030	-.013	.131	-.013	.012
CBQ: effortful control		—	-.101	.150	-.289**	-.369**	.083	-.243*
CBQ: surgency/extraversion			—	.063	-.066	.058	.173	.029
CBQ: negative affectivity				—	-.160	-.103	.166	.202*
Parent severity rating					—	.759**	.062	.146
Clinician severity rating						—	.140	.090
Total life events (LEC) ^a							—	.250*
CHAOS								—

Note. CBQ = Children's Behavior Questionnaire; LEC = Life Events Checklist; CHAOS = Confusion, Hubbub and Order Scale.

^aSpearman rho correlation used for this variable.

* $p < .05$ (two tailed). ** $p < .01$ (two tailed).

Table 3. Hierarchical linear regression model: predictors of parent-reported severity.

Model	<i>b</i> ^a	SE <i>b</i> ^b	β ^c
Model 1			
Constant	5.691	.937	
CBQ: effortful control	-.523	.177	-.289**
Model 2			
Constant	7.297	1.470	
CBQ: effortful control	-.508	.180	-.281**
CBQ: surgency/extraversion	-.170	.191	-.088
CBQ: negative affectivity	-.220	.193	-.112
Model 3			
Constant	7.037	1.677	
CBQ: effortful control	-.484	.188	-.267*
CBQ: surgency/extraversion	-.200	.191	-.103
CBQ: negative affectivity	-.273	.198	-.140
LEC	.058	.040	.156
CHAOS	.144	.371	.045

Note. CBQ = Children's Behavior Questionnaire; LEC = Life Events Checklist; CHAOS = Confusion, Hubbub and Order Scale.

^aUnstandardized coefficient. ^bStandard error of the unstandardized coefficient. ^cStandardized coefficient.

p* < .05. *p* < .01.

The Australian data validates earlier findings that low scores on measures of EC are related to children with higher parent-rated stuttering severity scores. Conversely, children with low stuttering severity scores show higher scores in the temperament domain of EC, as described in Figure 1. As such, EC remains a significant predictor of parent-reported child stuttering severity ratings.

Parents' Scores and Clinician Severity Rating

Identical procedures were used for HLM and clinician-reported severity ratings. The results of HLM indicated that all three models significantly predicted the children's severity rating (*F* = 15.117, *p* < .001 for Model 1; *F* = 5.058, *p* = .003 for Model 2; and *F* = 3.943, *p* = .003 for Model 3); see Table 5. EC was the sole contributor to Model 2 and

Table 4. Hierarchical linear regression model results for children's scores and parent-reported severity rating significance by model.

Model	<i>F</i>	Significance
1	8.733	.004 ^a **
2	3.671	.015 ^b *
3	2.895	.018 ^c *

Note. *F* = corresponding *F* statistic of overall regression significance by model.

^aModel 1 predictors (constant): Children's Behavior Questionnaire (CBQ): effortful control. ^bModel 2 predictors (constant): CBQ: effortful control, CBQ: surgency/extraversion, CBQ: negative affectivity.

^cModel 3 predictors (constant): CBQ: effortful control, CBQ: surgency/extraversion, CBQ: negative affectivity; Life Events Checklist; Confusion, Hubbub and Order Scale.

p* < .05. *p* < .01.

made the greatest contribution to Model 3 (see Table 6). Another variable, the parent measure of LEC, was also found to be a significant contributor to Model 3, but this outcome is unreliable. LEC was included within the HLM to be consistent with the analysis conducted in the original study; however, it was not normally distributed. Furthermore, LEC did not show any significant correlation with clinician-reported severity when assessed nonparametrically.

The results of this HLM analysis revealed that clinician-reported stuttering severity is also significantly related to EC in CWS. As demonstrated in the scatter plot below (see Figure 3), EC accounts for 13.6% of the variation in clinician-reported severity ratings in this study, which is greater than the 8.7% contribution found in the previous study. This indicates that EC was a significant predictor of clinicians' stuttering severity ratings for CWS.

Discussion

In this requisite replication study, findings were robustly consistent with those reported by Kraft et al. in 2014. The following considerations were made to ensure the procedures had fidelity to the original study where practicable: Hierarchical linear modeling analysis was similarly used and demonstrated that the temperament domain of EC was uniquely predictive of stuttering severity in these children; stuttered speech severity ratings were again reported by both parents and clinicians; and the same standardized parent report quantitative questionnaires were used with results again endorsing the finding that effects were independent of home environment and major life events. These replicated results culminate in important clinical implications for speech-language pathologists working with young CWS and their parents.

The relationship replicated and demonstrated in this study suggests that children who exhibit lower EC are more likely to display more severe stuttering behaviors. The rationale for this negative correlation is supported by the literature, as children with poor EC are less able to regulate their attentional control and less able to shift their attention from stimuli that elicits an emotional response to salient stimuli (e.g., teacher instructions) or original intention (e.g., achieving communication/speaking goal; Bush, 2006). Eggers et al. (2013) describe low EC as lower inhibitory control and greater resistance to modifying a dominant response to particular events. This observed tendency for some CWS to be less able to regulate or adapt their behavioral or emotional responses leads to greater experience of stress in the face of environmental triggers (Karrass et al., 2006; Valiente et al., 2004). For children with higher EC, the effects are typically reversed, with CWS responding to stimuli in a manner that facilitates effective self-monitoring and self-regulation. Perhaps, CWS with high EC are more capable of shifting their attention from a moment of stuttered speech to the use of strategies that facilitate forward-moving speech and, ultimately, their fluent communication goal. CWS with high EC may also find it easier to suppress maladaptive regulatory

Figure 2. Scatter plot for parent-reported severity ratings and effortful control. CBQ = Children's Behavior Questionnaire.

strategies, once they are found to be ineffective (Eggers et al., 2013).

In terms of placing these findings in the broader literature across the life span, the importance of developing strong self-regulation skills in the behavioral aspects of EC has been endorsed in a recent article by Carter, Breen, Yaruss, and Beilby (2017). The authors investigated the relationship between self-efficacy and QoL in AWS and found that the construct of high self-efficacy in a person

Table 5. Hierarchical linear regression model results for children's scores and clinician-reported severity rating significance by model.

Model	F	Significance
1	15.117	.000*
2	5.058	.003*
3	3.943	.003*

Note. F = corresponding F statistic of overall regression significance by model.

* $p < .05$. ** $p < .01$.

Table 6. Hierarchical linear regression model: predictors of clinician-reported severity.

Model	b^a	SE b^b	β^c
Model 1			
Constant	7.150	1.106	
CBQ: effortful control	-.746	.192	-.369*
Model 2			
Constant	7.232	1.610	
CBQ: effortful control	-.725	.197	-.359*
CBQ: surgency/extraversion	.054	.209	.025
CBQ: negative affectivity	-.110	.212	-.051
Model 3			
Constant	7.991	1.827	
CBQ: effortful control	-.772	.205	-.382*
CBQ: surgency/extraversion	.002	.208	.001
CBQ: negative affectivity	-.115	.216	-.053
LEC	.090	.044	.218*
CHAOS	-.311	.404	-.086

Note. CBQ = Children's Behavior Questionnaire; LEC = Life Events Checklist; CHAOS = Confusion, Hubbub and Order Scale.

^aUnstandardized coefficient. ^bStandard error of the unstandardized coefficient. ^cStandardized coefficient.

* $p < .01$.

Figure 3. Scatter plot for clinician-reported severity ratings and effortful control. CBQ = Children's Behavior Questionnaire.

who stutters reflects positively in their improved QoL. Further, the researchers recommended that clinicians should target therapy that specifically increases self-efficacy to “buffer” against the adverse impacts of a stuttering disorder (Carter et al., 2017).

This contemporary theme of targeting temperament regulation skills has recently been addressed in a hallmark position paper postulating “Crystal Ball Gazing” for the future of clinical practice in fluency disorders by esteemed authors Manning and Quesal (2016). In this statement, the authors endorse the findings of Edward Conture and others outlining the unique emotional regulation differences that need to be highlighted empirically so that needs can be identified early to support active change in children’s individual therapy. The results from this study show promise in the identification of predictors underpinning the severity of stuttered speech in children.

It is well known that self-regulatory systems can change very rapidly when the child is young. We must therefore move to employ strategies that help shape EC as it is still developing; a likely pivotal step in shifting the speech

trajectory. Many natural parent-administered early interventions for stuttering address fluency by encouraging behavioral inhibition, that is, advice such as “slow down, take your time, take a moment and try again” are all examples of parent responses addressing the child’s specific high emotional, linguistic, and emphatic communication utterances. In effect, the parents are supporting and encouraging regulatory responses on the part of their child that are more conducive to the enhancement of fluency. Results from this study support extending the repertoire of regulatory parenting and clinical practices that can ultimately decrease the severity and impact of stuttering for very young children.

The “Purdue Stuttering Project” (A. Smith & Weber, 2016) is a diagnostic battery that outlines early stuttering from a multidimensional perspective. The authors state that “heightened emotional reactivity” for CWS warrants an individualized treatment approach that incorporates regulation training and stuttering management concurrently. The authors also make a claim for the fact that “heterogeneity” among CWS is real and evident and an emphasis

on individualized treatments are needed to map onto and support the individual profiles of each child who stutters (A. Smith & Weber, 2016). The consolidated results from this study and the original in 2014 extend the emphasis of these considerations for future discrete clinical management. In fact, they give directional support to using a child's EC profile to guide clinical management and inform treatment strategies. For CWS who demonstrate lower EC, clinicians could preemptively integrate therapy goals that promote the development of self-monitoring and effective self-regulation strategies. To this end, future research investigating the specific self-regulatory behaviors of CWS, across a wide range of EC/temperament profiles, would be highly beneficial. This would enable researchers to see whether specific EC/temperament profiles can predict the type, frequency, or effect of regulatory behaviors observed, as well as the extent that maladaptive strategies are adopted or resistant to extinction/recovery once established. A more detailed investigation of these factors may elucidate findings such as those reported by Choi et al. (2013), where greater behavioral inhibition was associated with increased severity.

Despite the literature supportive of a relationship between home environment and major life events and stuttering severity, these particular variables as measured in both studies (original and replication) were not significantly correlated with stuttering severity in CWS. As previously mentioned by Kraft et al. (2014), this may reflect the tendency for children with low EC to reach their emotional response threshold, even when the event appears to be relatively unremarkable. For these children, even mundane events may be sufficiently stimulating to trigger an emotional reaction that reduces their capacity to effectively regulate other behaviors, including those for speech.

Limitations

The skewed distribution of ages within this cohort (i.e., fewer older children than younger) may have been a factor in altering the contribution EC made to stuttering severity, compared with the original study. Future research in this area may benefit from groups of participants that are evenly distributed across multiple age groups to determine whether age changes the relationship between EC and stuttering severity for CWS. Longitudinal studies that examine these effects within subjects over time would also be of value.

Conclusions

Therapeutic efficacy is enhanced by extending our understanding of the contributions to and underpinnings of severity. The development of treatment approaches from the platform of temperament appears an auspicious philosophy and one that is garnering increased contemporary momentum in the field. Holistic treatment for stuttering in children should include the behavioral components of enhanced self-regulation of EC (i.e., attention, inhibitory

control, and perceptual sensitivity) alongside traditional approaches to augment the effectiveness of each. Historically, clinicians and researchers have addressed limited components of EC and associated aspects of attention regulation in therapy objectives that have targeted self-monitoring skills. Careful consideration should now be made for the specific development of attentional attributes, emotion regulation strategies, vigilance reduction of exercises, and flexible cognitive shifting. Such therapeutic aims would have potentially significant downstream effects on reducing generalized stuttering severity. In essence, coping mechanisms that reduce emotional reactivity and increase inhibitory control capabilities will theoretically build resilience in children and result in fewer speech disruptions. The individual mechanisms driving both volitional and involuntary aspects of EC work in concert with resources for cognitive and motor speech demands. By measuring the individual profiles of each child's temperament in relation to their severity, we could approach stuttering behaviors from a new clinical perspective that embraces an individualized treatment for each child respectful of the multidimensional nature of early stuttering.

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