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Cognitive flexibility in preschool children with and without stuttering disorders

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ABSTRACT

Purpose: Multifactorial explanations of developmental stuttering suggest that difficulties in self-regulation and weak attentional flexibility contribute to persisting stuttering. We tested this prediction by examining whether preschool-age children who stutter (CWS) shift their attention less flexibly than children who do not stutter (CWNS) during a modified version of the Dimension Card Change Sort (DCCS), a reliable measure of attention switching for young children.
Methods: Sixteen CWS (12 males) and 30 children CWNS (11 males) participated in the study. Groups were matched on age (CWS: M = 49.63, SD = 10.34, *range* = 38–80 months; CWNS: M = 50.63, SD = 9.82, *range* = 37–74 months), cognitive ability, and language skills. All children completed a computer-based variation of the DCCS, in which they matched on-screen bivalent stimuli to response buttons based on rules that switched mid-task.
Results: Results showed increased slowing for CWS compared to controls during the postswitch phase, as well as contrasting patterns of speed-accuracy tradeoff for CWS and CWNS as they moved from the preswitch to postswitch phase of the task.
Conclusions: Group differences in performance suggest that early stuttering may be associated with difficulty shifting attention efficiently and greater concern about errors.

Findings are consistent with a growing literature indicating links between weak attentional control and persisting developmental stuttering.

1. Introduction

Cognitive flexibility refers to a critical set of skills that enable us to shift attention, focus on relevant cues in a changing environment, and adapt to new situations. Along with inhibitory control (the ability to resist impulsive or automatic responses) and working memory (ability to temporarily store and manipulate information in the face of ongoing activity), cognitive flexibility is considered a core executive function (EF; Diamond, 2013) and shows significant growth during the preschool years (Davidson, Amso, Anderson, & Diamond, 2006). Executive function is an umbrella term referring to a range of cognitive skills (inhibitory control, working memory, and attention switching) that are essential to cognitive and psychological development, and that predict academic achievement, employment productivity, and overall life success. When cognitive flexibility is impaired, one consequence is perseveration, which occurs when a previously appropriate response is repeated in a context for which it is no longer appropriate (Sandson

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& Albert, 1984). Although there are many theoretical explanations for perseverative behavior, as discussed below, behavioral and neurophysiological findings suggest that flexible switching is supported by an underlying ability to sustain attention to relevant task information and exclude the irrelevant (Benitez et al., 2017; Moriguchi & Hiraki, 2009). This ability to deploy attention strategically, and engage with certain stimuli but disengage and shift attention away from others, is denoted by the term attentional control (e.g., Kane, Conway, Miura, & Colflesh, 2007).

It is well accepted that the development of stuttering involves complex interactions among cognitive, linguistic, emotional, and motor factors (Smith & Kelly, 1997; Smith et al., 2010; Smith & Weber, 2016); however, unique contributions of individual factors, particularly those related to attentional control, are not well understood. Attentional control plays a critical role in children's ability to self-regulate and manage emotional reactions to stress (Blair & Ursache, 2011). Within the context of stuttering, it has been proposed that children's risk for chronic stuttering may increase if they are unable to shift attention, and "move on" from experiences of disfluency (Karrass et al., 2006; Walden et al., 2012). Based on the dual diathesis-stressor (DD-S) model (Walden et al., 2012), a child's ability to regulate emotions and attention is one of several key factors that contribute to chronic stuttering. Results of parent-rated questionnaires and several behavioral studies indicate that relative to fluent peers, CWS have more difficulty controlling and regulating attention (Anderson et al., 2003; Arnold et al., 2011; Eggers, De Nil, & van den Bergh, 2012; Kefalianos et al., 2014). If attentional control is compromised in CWS, perseverative patterns of behavior are likely to be observed. Based on this reasoning, the present study examined whether young CWS show deficits in controlling attention and acting flexibly to resolve cognitive conflict on a rule-switch task. Exploring cognitive flexibility in this clinical population can clarify how aspects of attention switching interact with vulnerable capacities in other areas of development (e.g., motor speech incoordination) and influence the development and maintenance of stuttering.

1.1. Measuring cognitive flexibility

The ability to shift attention based on changing situational demands has traditionally been measured in adults using the Wisconsin Card Sorting Test (Berg, 1948; Grant & Berg, 1948) or similar paradigms. In this task, participants sort cards according to changing rules (e.g., color, number, shape), which are deduced from the experimenter's feedback. The Dimension Card Change Sort (DCCS; Frye et al., 1995) is a widely-used adaptation of this task for young children. In this simpler task, preschoolers are presented with bivalent stimuli that vary in color and shape. Children are required to sort a single set of stimuli by one dimension (e.g., color), then switch to the other (e.g., shape). Studies utilizing this paradigm consistently report a robust effect of age. A recent meta-analysis of 69 studies using the DCCS (Doebel & Zelazo, 2015) showed an estimated 41% pass rate at age 3 years, 5 months, compared to 69% and 88% pass rates at ages 4 and 5, respectively. Age-related changes in DCCS performance are assumed to reflect underlying development of the prefrontal cortex (PFC), which shows a protracted course of maturation (Fuster, 2002) and is part of a larger cognitive control network that supports flexible rule use (Ezekiel et al., 2013). In line with this view, results of near-infrared spectroscopy (NIRS; Moriguchi & Hiraki, 2009) showed increased inferior prefrontal activity during the DCCS compared to a control task that did not involve switching and in children who successfully switched compared to those that perseverated. Similarly, event-related changes in young children's frontal EEG coherence were positively associated with their performance on tasks requiring attentional control (visual search) and cognitive flexibility (DCCS) (Whedon et al., 2016).

1.2. Theoretical accounts for perseveration

Several theoretical frameworks for perseverative behavior have been proposed but no single account is universally accepted. According to inhibition or attentional inertia accounts (Kirkham, Cruess, & Diamond, 2003; Koch, Gade, Schuch, & Philipp, 2010), flexible switching requires a mechanism that can reduce activation of current task goals to enable switching to a new task. Children perseverate when they are unable to inhibit the tendency to view stimuli from a particular perspective (e.g., color or shape). Other researchers (Morton & Munakata, 2002) attribute task switching performance to the strength of activated representations in working memory (WM). When currently and previously relevant information conflict, flexible behavior can only occur when active representations are stronger than latent ones. Thus, perseveration is sometimes viewed as a weakness in WM.

It is also possible that no single mechanism fully accounts for switching performance. Diamond, for example, asserts that cognitive flexibility builds upon both WM and inhibitory control, and for this reason, emerges later than both prerequisite EFs (Diamond, 2016). To successfully shift attention to a new stimulus feature, children must activate a new goal in WM and inhibit the one used previously. It has further been demonstrated that EFs in children are not as distinct or separable as they are in adults and that children show a lack of EF differentiation throughout the preschool period (Nelson, James, Chevalier, Clark, & Espy, 2016; Wiebe et al., 2011). A possibility raised by these authors and others (Garon, Bryson, & Smith, 2008) is that basic elements of attention, particularly the ability to sustain attention, selectively attend, and adjust attentional focus, serve as critical building blocks for individual EFs during early development.

This perspective is supported by recent literature related to the DCCS. Results of a recent meta-analysis (Doebel & Zelazo, 2015) indicated that in addition to age, switching success was reliably predicted by certain task manipulations, such as reducing the number of preswitch trials or requiring labeling of the relevant stimulus dimension. Fewer preswitch trials results in weaker representation of the preswitch rule, making it easier to disengage attention and switch to the new sorting rule. Similarly, labeling the relevant stimulus dimension facilitates switching by drawing attention to the relevant dimension and related response. Although findings of Doebel and Zelazo did not converge to support any specific developmental mechanism or theory for flexible switching, results suggested that performance generally improved when experimental conditions increased attention to current task goals. Thus, although the DCCS is classically viewed as a measure of set shifting and cognitive flexibility (Zelazo et al., 2013), children's performance appears to reflect a more basic, underlying ability to sustain and control attention in the context of changing task demands (Benitez, Vales, Hanania, & Smith, 2017; Moriguchi & Hiraki, 2009).

Collectively, these views suggest that perseverative behavior can be interpreted as evidence of weakness in inhibitory control, WM, or more fundamental aspects of attention. This point is important because it helps place the present study within the larger stuttering literature and clarifies potential links to previous studies examining attention, as well as individual EF components, in individuals who stutter.

1.3. Stuttering as a form of perseveration

The nature of stuttering, particularly its most overt speech symptoms such as sound repetitions, prolongations, and blocks, intuitively suggests an association between stuttering and perseveration. This notion was considered in several early studies, with some findings indicating that adults who stutter (AWS) perseverate more than nonstutterers on a variety of psychomotor tasks. Relative to controls, AWS had more difficulty on tasks involving switching between closely related procedures, such as alternating between capital vs. lowercase letters in a copying task, performing addition vs. multiplication calculations, and providing antonyms and synonyms for given words (Eisenson & Pastel, 1936; Eisenson & Winslow, 1938; Wingate, 1966). King (1961) showed that perseverative tendencies were a pervasive characteristic of AWS and affected their performance across tasks measuring psychomotor flexibility as well as sensory processing (e.g., shifting perspectives on the Necker cube task).

It is difficult to draw conclusions from this early literature for several reasons. First, research examining perseveration in people who stutter has not always yielded similar findings. Although some studies report greater perseveration in people who stutter compared to nonstuttering controls (Eisenson & Pastel, 1936; Eisenson & Winslow, 1938; King, 1961; Wingate, 1966), other studies show no group differences on similar tasks (Kapos & Standlee, 1958; Martin, 1962). There is also a significant lack of consistency in how perseveration is operationally defined and measured. Although perseveration is most often assessed using psychomotor tasks (e.g., Eisenson & Pastel, 1936; Eisenson & Winslow,1938), some researchers defined perseveration as an aspect of temperament (King, 1961) or as the duration of aftereffects of sensory stimulation (e.g., Eisenson & Winslow, 1938; King, 1961).

Overall, previous research suggests compelling links between stuttering and perseveration; however, collective findings remain inconclusive due to an unclear definition of perseveration and inconsistent means of measuring this phenomenon. In the present study, we addressed these limitations by approaching perseverative behavior from a theoretical perspective that is consistent with current developmental cognitive psychology literature (Benitez et al., 2017; Carroll et al., 2016; Diamond, 2013; Morton & Munakata, 2002; Zelazo et al., 2003). We also used a reliable and widely-studied task for measuring perseverative behavior in young children (Zelazo, 2006), as described above.

1.4. Stuttering and attention

Recent research in the area of stuttering has not focused on cognitive flexibility, specifically, but has examined closely related aspects of attention. Successful switching on rule-switch tasks is highly dependent on the ability to sustain and control attention in changing contexts (Benitez et al., 2017; Moriguchi & Hiraki, 2009). Thus, if individuals who stutter demonstrate weaknesses in attentional control, we might also expect these speakers to demonstrate poorer cognitive flexibility on classic switching tasks.

Data from parent-rated questionnaires indicate that parents of CWS perceive their children as having more difficulty regulating attention and adapting to novelty than fluent peers (Anderson, Pellowski, Conture, & Kelly, 2003; Eggers, De Nil, & Van den Bergh, 2010; Felsenfeld et al. 2010; Karrass et al., 2006; Kefalianos, Onslow, Ukoumunne, Block, & Reilly, 2014). For example, parents indicated that CWS were less flexible in adapting to unexpected changes in routine, or shifting from one activity to another. Results of more direct behavioral research are less consistent than parent reports overall (Ofoe, Anderson, & Ntourou, 2015); however, available research provides a fair amount of evidence suggesting an association between stuttering and weak attentional control. Relative to fluent children, CWS had more difficulty orienting attention efficiently on a child-adapted version of the Attention Network Test (Eggers et al., 2012) and demonstrated longer RT on trials that required controlled attention toward cues that predicted the location of targets (Heitmann et al., 2004). In comparison to fluent peers, CWS also showed weaker sustained and selective attention on verbal and nonverbal auditory attention tasks (Anderson & Wagovich, 2016) and had more difficulty controlling attention in the presence of distracting background stimuli (Schwenk et al., 2007) and conversations (Arnold, Conture, Key, & Walden, 2011).

Several neurophysiological studies using event-related potentials (ERP) report similar findings of weakness in attentional control associated with stuttering, sometimes in the absence of behavioral differences between stuttering and fluent participants. For example, Kaganovich and colleagues examined amplitude of the P3 component (believed to index allocation of attentional resources and WM updating) in CWS and controls using an auditory oddball paradigm involving standard and deviant pure tones (Kaganovich et al., 2010). Averaged P1 and N1 components in each group were similar, indicating comparable detection and early encoding of auditory stimuli. Group differences were evident in the processing of deviant tones, which elicited a significant P3 component in the CWNS, but not in CWS. Based on these findings, the authors concluded that preschoolers who stutter have difficulty allocating attention and that these neurophysiological differences do not appear to be a consequence of disfluency (i.e., these characteristics are already evident in young children who have not stuttered very long). Chou (2014) similarly found that school-aged CWS (8–12 years) controlled attention less efficiently than controls during an auditory Go/No-Go task. This conclusion was based on significant between-group differences in the peak latency of 1RON, a component associated with attentional reorientation following distraction. In other words, CWS and CWNS attended to and processed distractions similarly, however, CWS had more difficulty recovering from these distractions.

Overall, converging evidence from neurophysiological studies, behavioral tasks, and parent perceptions suggests an association between stuttering and weaknesses in attention control. Tasks used to measure attention, however, vary widely and do not consistently indicate group differences in performance (Blood et al., 2007; Johnson et al., 2012). The DCCS has been used in more than 150 studies over the past two decades, resulting in a rich literature that details developmental changes in children's performance, cognitive mechanisms underlying switching success, and atypical patterns in clinical populations. This task is therefore ideal for exploring and understanding emerging attention switching abilities in preschoolers with early stuttering.

1.5. Aims

The present study focused on preschool children (3–6 years old) based on consistent data showing that typically developing children begin to demonstrate flexible behavior during these years (Doebel & Zelazo, 2015; Frye, Zelazo, Palfai, Frye, & Zelazo, 1995; Zelazo, 2006; Zelazo et al., 2013) and because stuttering typically emerges at this time (Bloodstein, 2006; Bloodstein & Bernstein Ratner, 2008; Yairi, 2004). We used a computer-based rule-switching task modeled after the DCCS to examine the relationship between cognitive flexibility and stuttering. Our primary hypothesis was that relative to fluent peers, stuttering preschoolers would have more difficulty focusing on relevant task goals and show more evidence of perseveration, particularly during the postswitch phase, which involves greater flexibility of attention. We expected perseverative tendencies to be manifested as a greater reduction in accuracy and lengthening of reaction times (RT) during the postswitch phase of the sorting task. Although DCCS performance in children is generally measured in terms of accuracy (pass rates), we included RT to capture cognitive biases that may be present even when responses were correct, as they would likely be for older children (Diamond & Kirkham, 2005a). The predominance of categorical analyses in DCCS literature is considered a shortcoming by some researchers (Yerys and Munakata, 2006), who emphasize the need for continuous data that can potentially reveal subtle attentional processes underlying performance. We further expected all children to show general improvements in performance (respond more quickly and accurately) over the course of the experiment (regardless of phase) based on typical learning patterns.

2. Methods

2.1. Participants

Participants included 46 children between the ages of 3 years, 0 months and 6 years, 6 months, who were recruited from local preschools and speech/language clinics throughout the New York metropolitan area. All children were monolingual English speakers and had no known neurological or psychological disorder, hearing loss, or significant medical history, based on responses provided by parents on an informal written developmental screening questionnaire. Additional inclusion criteria included a multiple composite score within at least the average range (85 or higher) on a test of general cognitive ability (*Kaufman Assessment Battery for Children,* K-ABC; Kaufman & Kaufman, 1983), and core language score within at least the average range (85 +) on the *Clinical Evaluation of Language Fundamentals − Preschool, Second Edition (*CELF-Preschool 2; Wiig et al. 2004). Sixteen of the children (12 males) had confirmed stuttering disorders; the remaining 30 children (11 males) served as fluent controls. Stuttering diagnosis was determined for the children who stutter (CWS) based on diagnostic information provided by a speech-language pathologist or parent, and rating of at least mild stuttering (score ≥ 11) on the Stuttering Severity Instrument − 4th Edition (SSI; Riley, 2009) at the time of the study. Stuttering severity for the CWS ranged from mild to severe based on SSI results. Fluency measurements were derived from recorded speech samples (approximately 250-500 syllables) obtained during an unstructured play interaction between the child and examiner (20–30 min). Testing, scoring procedures, and disfluency counts were completed by the first author, who is an ASHA-certified and state-licensed speech language pathologist; assistance was provided by trained graduate students. Informed parental consent and child assent were obtained before beginning experimental procedures. Children were compensated with small, age-appropriate rewards when tasks were completed. An attempt was made to recruit equal numbers of male and female children; however, boys represented a larger proportion of the stuttering group (3:1) than the control group (0.57:1) ($\chi^2(1, 46) = 6.13$, $p = 0.01$). The disproportionate representation of males in the stuttering group is consistent with prevalence data reported for fluency disorders (Bloodstein & Bernstein Ratner, 2008); however, the differences in gender ratio between groups was accounted for in statistical analyses, as described below. Gender differences in switching performance are also not typically found on the DCCS (Müller, Dick, Gela, Overton, & Zelazo, 2006; Zelazo, 2006; Zelazo, Müller, Frye, & Marcovitch, 2003). The two groups were closely matched in age (CWS: $M = 49.63$, $SD = 10.34$, *range* = 38–80 months; CWNS: $M = 50.63$, $SD = 9.82$, *range* = 37–74 months) as well as in cognitive and linguistic abilities, as assessed by the K-ABC and CELF-Preschool 2, respectively. Demographic details and results of standardized testing for each participant group are summarized in Table 1.

2.2. Stimuli and procedures

Stimuli for the computer-based adaptation of the DCCS task consisted of three target pictures (blue hexagon, yellow parallelogram, pink curved rectangle) and six test pictures that each shared a single dimension with two of the target pictures (e.g., yellow hexagon, pink parallelogram, blue curved rectangle). Although most standard versions of the DCCS use familiar objects (e.g., rabbit, boat) and primary colors (red, blue) as exemplars for shape and color dimensions, respectively (Doebel & Zelazo, 2015; Frye et al., 1995; Zelazo, 2006; Zelazo et al., 2013), our adaptation used figures that were difficult to label and that were shaded in light

Table 1
Participant characteristics. Mean (SD).

	CWNS (n = 30)	CWS (n = 16)	F	p
Chronological age (months)	50.63 (9.82)	49.63 (10.34)	0.10	
K-ABC (MPC)	104.76 (10.47)	99.86 (7.74)	2.42	0.128
CELF-P2 (CLS)	105.71 (9.51)	107.45 (16.37)	0.17	0.68

Notes: CWNS = Children Who Do Not Stutter; CWS = Children Who Stutter; K-ABC = Kaufman Assessment Battery for Children; MPC = Multiple Performance Composite; CELF-P2 = Clinical Evaluation of Language Fundamentals – Preschool, 2nd Edition; CLS = Core Language Score. K-ABC data available for 29 CWNS and 14 CWS; CELF-P2 data for 28 CWNS and 11 CWS.

nonprimary colors. We selected these stimuli and dimensions in order to minimize the likelihood of children utilizing verbal strategies or relying on verbal WM, as these mechanisms were not the focus of the present study and may be an area of weakness for CWS (e.g., see Bajaj, 2007 for review). Each of the three target pictures was affixed to a 3-inch round response button; buttons were organized in an array that remained constant for the duration of the experiment. On each trial, a single test picture appeared in the center of the computer screen. Children were instructed to match the stimulus on the screen to one of the response buttons based on the sorting rule (color or shape) specified. The relevant sorting dimension remained consistent for 12 trials (preswitch phase), followed by presentation of a new sorting rule and second set of 12 trials (postswitch phase). The order of rules (color/shape) was counterbalanced and presentation order of individual stimuli within each phase was randomized. See Fig. 1 for a sample trial and Fig. 2 for a diagram of the general task structure.

Each phase (pre- and postswitch) began with instructions presented auditorily by the examiner based on written scripts onscreen. The matching rule was explained using age-appropriate language and repetition, consistent with standard versions of the DCCS (Frye et al., 1995; Zelazo, 2006): *"This is the shape (color) game. In the shape (color) game, you look at the picture on the screen and find the matching shape (color) on the big buttons. I will show you how to do it and then it will be your turn."*

Correct responses were demonstrated by the examiner for three trials, with each response preceded by an explanation that highlighted the relevant dimension: "*This shape (color) is the same as… this shape (color)."* Demonstration items were followed by three practice trials, each cued with the relevant rule (e.g., *"This shape [color] is the same as…")* and followed by immediate onscreen feedback. Following practice, children proceeded to 12 trial items that comprised the preswitch phase. Participants were then exposed to the new sorting rule with instructions that emphasized the conflict between the old and new rules (this feature is included in most standard versions of the DCCS and significantly influences switching performance, as discussed by Doebel & Zelazo, 2015): "*Okay, now we're going to switch and play a new game. This game is the color (shape) game. We are not going to play the shape (color) game anymore. We are going to play the color (shape) game and the color (shape) game is different. In the color (shape) game…* (remaining instructions were identical to those provided for the preswitch rule but emphasized the second dimension)." As in the preswitch phase, participants viewed three demonstration items, responded to three practice items with feedback, and completed a set of 12 test trials independently. All participants were tested individually. Screening procedures and experimental tasks were completed in a single session lasting approximately two hours.

2.3. Equipment

Experimental tasks were presented on a laptop computer with a 13-inch screen. Response buttons with target pictures were joined to a Quizworks USB switch interface connected to the laptop and positioned two inches away from the edge of the table directly in front of each participant. Stimulus presentation was controlled via E-Prime 2.0 software, which recorded accuracy and RT data for all manual responses.

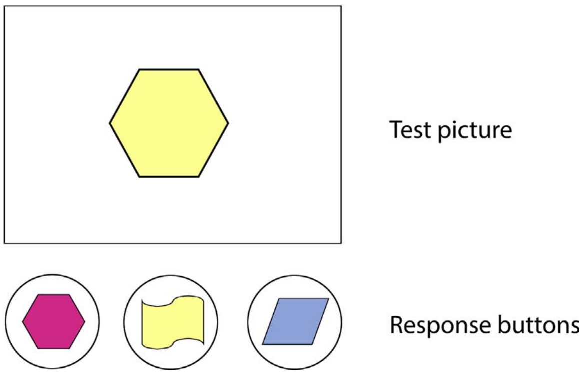

Fig. 1. Sample trial of sorting task.

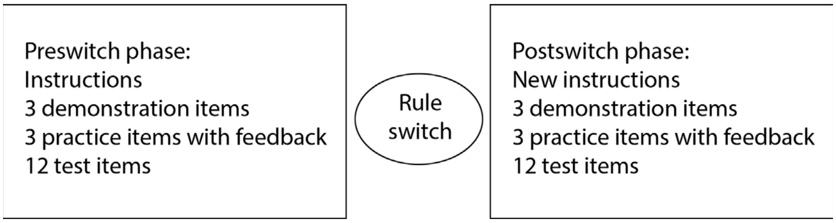

Fig. 2. General task structure.

2.4. Data processing and analysis

Dependent variables included accuracy of each trial (0 or 1) and RT for correct responses. Although methods for combining accuracy and RT in a single two-vector score to capture speed/accuracy tradeoff effects have been described (Zelazo et al., 2013), this scoring system is recommended primarily for older children who maintain high levels of accuracy ($> 80\%$) and are completing mixed blocks including multiple rule switches. We analyzed accuracy and RT individually because our task only involved a single switch and participants were young (under 6). Raw RT data were screened via a two-step process, which included removal of extreme outliers (< 200 ms or $> 10,000$ ms) and trimming of within-subject RT distributions to values within four standard deviations above or below the mean value for each subject. A total of 18 RT values (0.8%) were excluded based on these criteria, and these same values were flagged using the Median Absolute Deviation criteria advanced by Leys and colleagues (Leys, Ley, Klein, Bernard, & Licata, 2013). Excluded values consisted of 6 extreme outliers (0.3%) and 12 RT values that were more than 4 *SD* above subject means (0.5%). Accuracy results for trials with outlier RT values were also removed. Additional sensitivity analyses including all outliers and using varying outlier criteria (i.e., 2 or 3 *SD/MAD*) yielded essentially identical results, except that key findings were statistically stronger when using more liberal outlier criteria. Because RT values were not normally distributed (*skew* = 5.39, *kurtosis* = 52.86), we followed recommendations of Whelan (2008) and log transformed these data (using natural logarithm) before entering them in analyses (*skew* = 0.54, *kurtosis* = 3.83).

Generalized multilevel linear models were utilized to model accuracy and reaction time, as these models provide maximum flexibility and robustness when analyzing multi-level experimental data and can account for both within- and between-subject factors and unbalanced designs (Gelman & Hill, 2006; Hoffman & Rovine, 2007). Accuracy data were binary and therefore analyzed using multilevel logistic regression models; RT was log transformed and analyzed using multilevel regression models. These models were estimated using the Lme4 package in R (Bates et al., 2014). Inferential tests of fixed coefficients utilized Satterthwaite approximation and were conducted using the lmerTest package (Kuznetsova et al., 2016). Plots were created using the sjPlot package in R (Lüdecke, 2017).

For each analysis, a series of increasingly complex models was estimated. Predictors were entered in an a priori selected order such that random effects representing between-subject differences on mean performance variables and within-subject changes over time were entered first. Subsequent models added variables to test key hypotheses (task phase, speaker type, and trial number), followed by corresponding interactions in increasing complexity (two-, then three-way). Non-significant variables and interactions were removed at each stage unless statistically required to test higher-order interactional hypotheses. Successive models were compared using the log-likelihood ratio test, as well as Akaike Information Criterion (*AIC*) and Bayesian Information Criterion (*BIC*) fit statistics.

3. Results

3.1. Accuracy

Switching accuracy was examined by comparing the accuracy of postswitch responses relative to preswitch responses for the two speaker types (CWS vs. CWNS). Analyses also considered whether changes in accuracy occurred over the course of the experiment (i.e., trial number effect, regardless of phase). Descriptive data by task phase and speaker type are provided in Table 2. A series of successively complex models was estimated to examine effects of speaker type, task phase, and trial number on task performance (see Table 3). Examination of coefficients for the final model (Table 4) indicated that task phase had an overall effect on accuracy, such

Table 2
Accuracy (proportion of correct responses) and reaction time by task phase and speaker type. Mean (SD).

	Accuracy		*Reaction Time (ms)*	
	Preswitch	*Postswitch*	*Preswitch*	*Postswitch*
CWNS	0.895	0.787	1794.93 (1084.39)	2041.71 (1014.0)
CWS	0.818	0.764	2228.31 (1355.30)	2420.80 (1470.77)

Notes: CWNS = Children Who Do Not Stutter; CWS = Children Who Stutter.

Table 3
Model fit of sequential multilevel logistic regression model of accuracy data.

	df	*AIC*	*BIC*	*-2LL*	X^2	*p*
M0: Random Intercept Only	2	890.67	900.66	-443.34		
M1: Added Fixed Slope (Trial Number)	3	878.75	893.73	-436.37	13.92	0.0002
M2: Added Random Slope (Trial Number)	5	872.09	897.06	-431.05	10.66	0.005
M3: Added Phase	6	868.98	898.94	-428.49	5.11	0.024
M4: Added Phase * Trial Number	7	868.07	903.02	-427.03	2.92	0.088
M5: Added Speaker	8	869.01	908.96	-426.51	1.05	0.305
M6: Added Speaker * Trial Number	9	870.77	915.71	-426.39	0.24	0.625
M7: Added Speaker * Phase	10	866.82	916.75	-423.41	5.95	0.015

Note: Logistic models were fit using Maximum Likelihood Estimation and included a random intercept for each subject.

Table 4
Results of final model of accuracy (M7).

Fixed Effects	Unstandardized Coefficient	*OR*	*SE*	*z*	*p*
Intercept	2.81		0.54	5.25	< 0.0001
Phase	−2.61	0.07	0.81	−3.23	0.001
Trial	0.003	1.003	0.05	0.06	0.949
Speaker	−0.22	0.81	0.69	−0.32	0.747
Phase * Trial	0.09	1.09	0.06	1.52	0.129
Trial * Speaker	−0.09	.91	0.06	−1.50	0.135
Phase * Speaker	1.81	6.12	0.75	2.42	0.016

Note: Model was fit using Maximum Likelihood Estimation (Laplace Approximation) and included the random effect of an individual-level intercept Model was based on 1089 observations across 46 subjects.

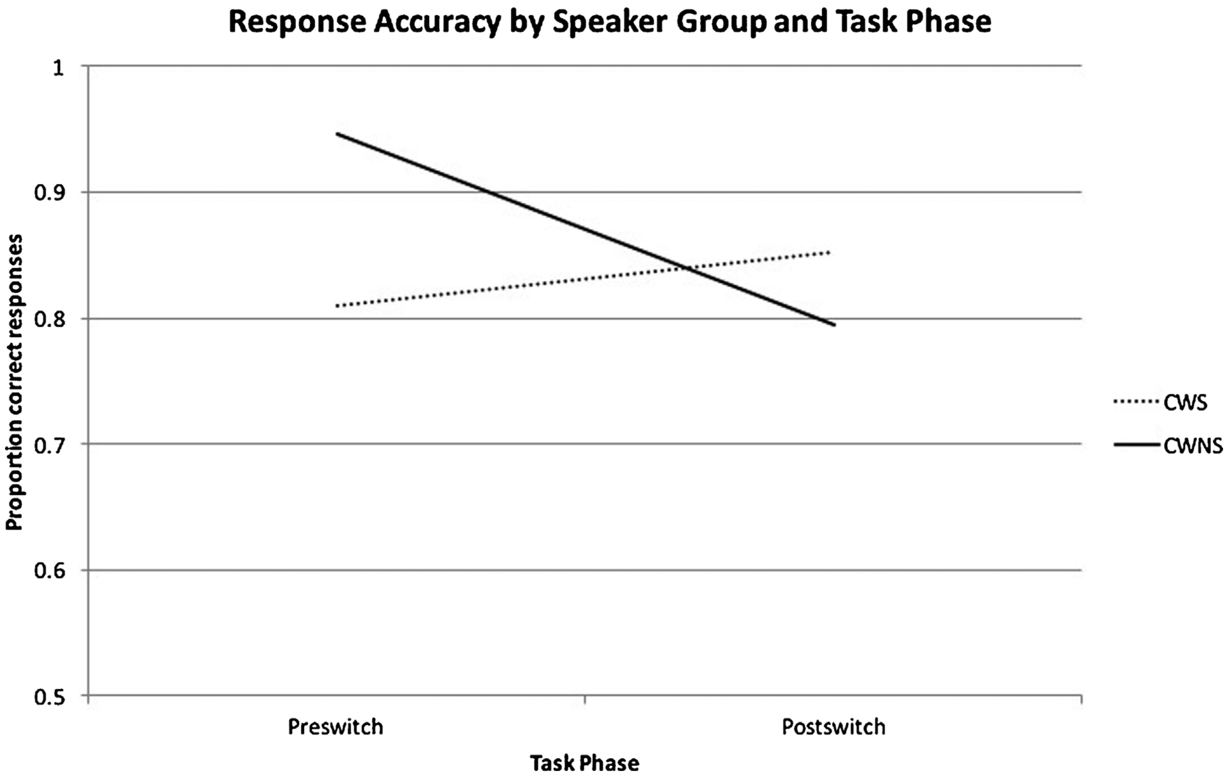

Fig. 3. Accuracy results by task phase (pre- and postswitch) and speaker group (CWS = Children Who Stutter and CWNS = Children Who Do Not Stutter).

that the odds of responding correctly was 7% lower during the postswitch compared to preswitch phase ($\beta = -2.61$, $SE = .81$, $OR = .07$, $z = -3.23$, $p = .001$). There was no main effect for speaker type ($\beta = -0.22$, $SE = .69$, $OR = .81$, $z = -0.32$, $p = 0.75$) or trial number ($\beta = 0.003$, $SE = 0.05$, $OR = 1.003$, $z = 0.06$, $p = 0.95$);

Table 5
Model fit of sequential multilevel regression models of reaction time data.

	df	*AIC*	*BIC*	*-2LL*	X^2	*p*
M0: Random Intercept Only	3	1043.26	1057.66	−518.63		
M1: Added Fixed Slope (Trial Number)	4	1045.25	1064.45	−518.62	0.01	0.9219
M2: Added Random Slope (Trial Number)	6	951.99	980.78	−469.99	97.26	< 0.0001
M3: Added Phase	7	829.94	863.53	−407.97	124.05	< 0.0001
M4: Added Phase * Trial Number	8	829.4	867.79	−406.7	2.54	0.1109
M5: Added Speaker	9	828.95	872.14	−405.47	2.45	0.1175
M6: Added Speaker * Trial Number	10	826.23	874.22	−403.12	4.72	0.0299
M7: Added Speaker * Phase	11	815.28	868.07	−396.64	12.95	0.0003

Note: Logistic models were fit using Maximum Likelihood Estimation and included a random intercept for each subject.

however, speaker type significantly interacted with task phase ($\beta = 1.81$, $SE = 0.75$, $OR = 6.12$, $z = 2.42$, $p = 0.016$). Specifically, CWS did not show a large decrement in their performance accuracy on the postswitch compared to preswitch phase. Relative to CWS, children in the control group showed a greater decline in accuracy on the second task phase. See Fig. 3 for a graph of interaction effects between task phase and speaker type on performance accuracy. This interaction maintained significance in a model that included gender and age as control variables, although older age showed a small but significant association with increased accuracy overall ($\beta = .06$, $SE = 0.02$, $OR = 1.06$, $z = 2.61$, $p = 0.009$). These accuracy findings were not consistent with our hypothesis, which predicted a greater reduction in accuracy for CWS relative to controls.

3.2. Reaction time

Descriptive statistics for RT by task phase and speaker type are provided in Table 2. As with accuracy data, a series of successively complex models was used to examine effects of task phase, trial number, and speaker type on task performance (see Table 5). Coefficients of the final model (Table 6) indicated significant main effects for each of the three factors. Based on these coefficients, RT for the postswitch phase was slower compared to preswitch RT ($\beta = 0.27$, $SE = .10$, $t = 2.60$, $p = .0096$); RT tended to become faster for each successive trial over the course of the experiment ($\beta = -0.03$, $SE = 0.01$, $t = -3.95$, $p < 0.0001$); and CWS responded more slowly than controls overall ($\beta = 0.48$, $SE = .14$, $t = 3.35$, $p = .0017$). Both task-related factors (phase and trial) also interacted with speaker type. Specifically, CWS showed greater increases in speed over the course of the experiment relative to CWNS ($\beta = -0.04$, $SE = 0.01$, $t = -3.88$, $p < 0.0002$; Fig. 4). There was also a significant interaction between speaker type and task phase ($\beta = 0.35$, $SE = 0.10$, $t = 3.60$, $p = .0003$). Thus, relative to fluent controls, stuttering children showed greater RT slowing during the postswitch phase of the task (Fig. 5). As with results for accuracy, these interactions remained significant when controlling for age and gender, although age was significantly correlated with slightly faster overall RT ($\beta = -0.01$, $SE = 0.004$, $t = 2.63$, $p = .01$). RT results supported our primary hypothesis, in which we predicted greater postswitch slowing for CWS relative to controls.

Given unexpected and contrasting patterns between accuracy and RT, we considered the possibility that CWS and CWNS applied different strategies during the postswitch phase, resulting in contrasting speed-accuracy tradeoffs. To examine this hypothesis, we calculated mean RT and percentage correct for each participant and tested their relationship using non-parametric Spearman's rank correlation coefficients. Results showed that longer mean RT tended to be associated with a lower percentage of accurate responses during preswitch (Rs (46) $= -0.33, p = 0.02$) and postswitch phases (Rs (46) $= -0.36, p = 0.01$), indicating that participants who performed more poorly took a longer time on this task. However, this relationship between RT and accuracy varied based on speaker group and task phase. For CWNS, mean RT was only weakly correlated with accuracy (percent correct) on preswitch trials (Rs (30) $= -0.20, p = .29$), but became more strongly associated postswitch (Rs (30) $= -0.42, p = 0.02$). In contrast, correlation results for CWS indicated a strong relationship between mean RT and accuracy preswitch (Rs (16) $= -0.60, p = 0.01$), but weaker relationship postswitch (Rs (16) $= -0.35$, $p = 0.18$). Thus, across both groups, participants with slower RTs tended to be less accurate overall; however, CWNS demonstrated this association more clearly postswitch; whereas CWS showed the opposite pattern (strong

Table 6
Results of final model of RT (M7).

Fixed Effects	Unstandardized Coefficient	*SE*	*t*	*p*
Intercept	7.56	0.09	86.40	< 0.0001
Phase	0.27	0.10	2.60	0.0096
Trial	-0.03	0.01	−3.95	0.0001
Speaker	0.48	0.14	3.35	0.0017
Phase * Trial	0.01	0.01	1.71	0.0876
Trial * Speaker	-0.04	0.01	−3.88	0.0002
Phase * Speaker	0.35	0.10	3.60	0.0003

Note: Model was fit using Restricted Maximum Likelihood Estimation (Satterthwaite Approximation) and included the random effect of an individual-level intercept. Model was based on 897 observations across 46 subjects.

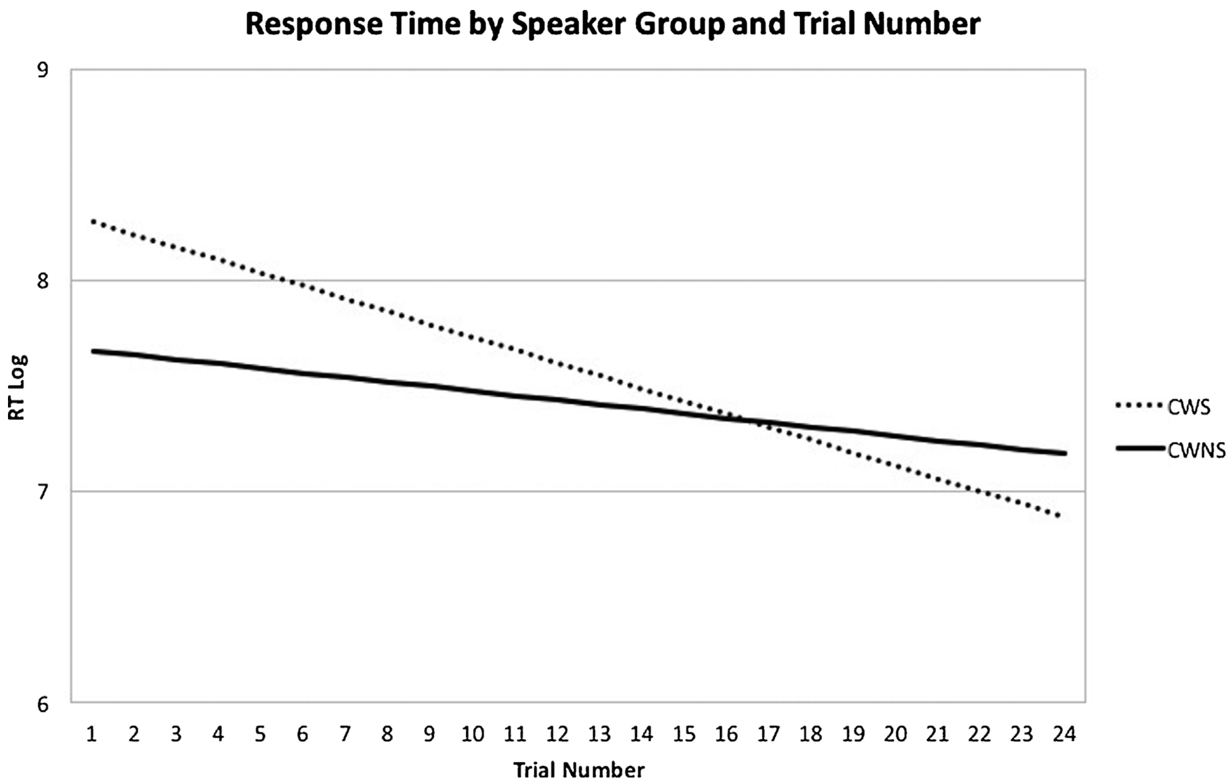

Fig. 4. Reaction time by trial number and speaker group (CWS = Children Who Stutter and CWNS = Children Who Do Not Stutter).

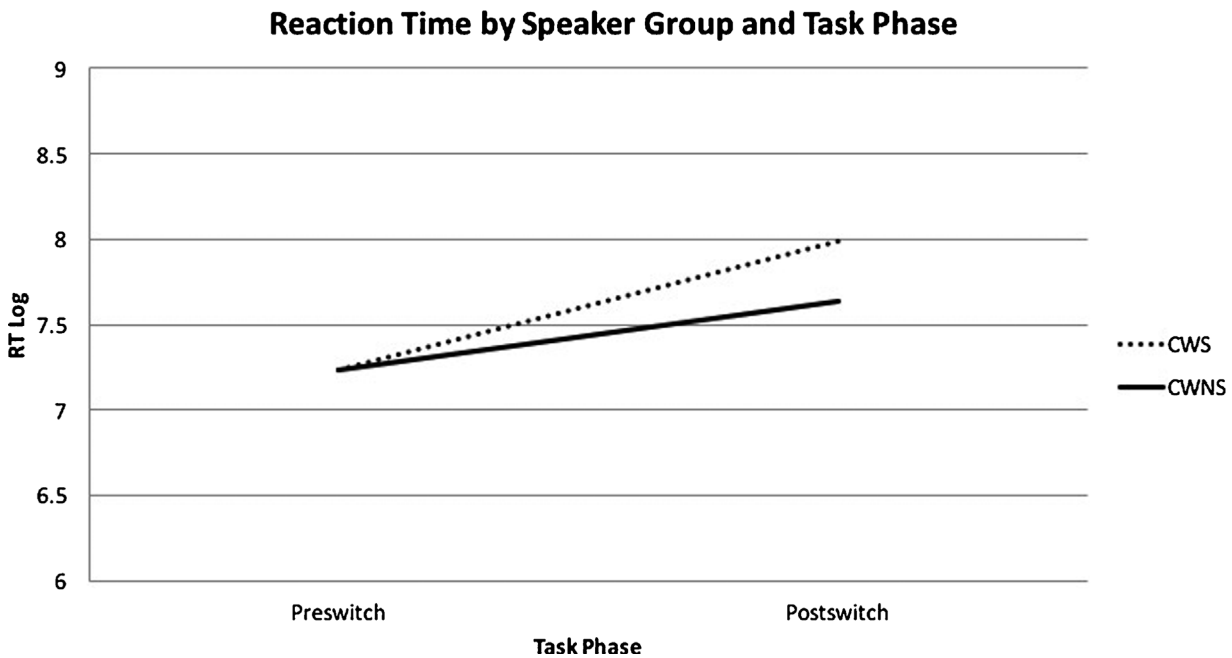

Fig. 5. Reaction time by task phase (pre- and postswitch) and speaker group (CWS = Children Who Stutter and CWNS = Children Who Do Not Stutter).

association preswitch but weak relationship postswitch). This result suggests a possible tradeoff between speed and accuracy for CWS postswitch, in which greater accuracy was maintained by sacrificing the speed of responses.

4. Discussion

The key finding emerging from the study was that stuttering was associated with more difficulty controlling attention in response to changing task goals. Compared to fluent peers, CWS had more difficulty reconfiguring their cognitive system to the new sorting rule, as manifested by greater postswitch slowing. Consistent with our primary hypothesis, RT results suggested that CWS have weaknesses in cognitive flexibility and are less efficient in disengaging and switching attention. This finding supports a multifactorial etiology for stuttering and identifies attentional differences that may contribute to developmental stuttering, as discussed in more detail below. Our hypothesis related to accuracy (greater postswitch reduction in accuracy for CWS) was not supported and these results contrasted with those of RT. This unexpected finding led us to consider potential differences in speed-accuracy tradeoff between groups. These analyses suggested that CWS approached demands of the postswitch phase differently and tended to maintain high levels of accuracy at the expense of RT. Although shifts in speed-accuracy tradeoffs are expected as children move beyond the preschool stage (Davidson et al., 2006), we did not anticipate between-group differences in response strategies. The more cautious strategy of CWS suggests greater concern about errors and to our knowledge, is a novel behavioral finding in the stuttering literature. Interesting results related to accuracy-speed tradeoffs were recently reported for CWS (Fortunato-Tavares, Howell, Schwartz, & De Andrade, 2017); however, these tradeoffs represented contrasting approaches used by CWS on different tasks (comprehension of relative clauses vs. reflexive assignment) rather than between-group differences on the same task.

Overall, children showed expected patterns of performance on the switch task. Introduction of a new sorting rule elicited changes in performance that reflected perseveration. These changes were manifested as significantly reduced accuracy and corresponding increases in RT during the postswitch phase. RT data also reflected practice effects over the course of the experiment, with faster responses on successive trials, regardless of phase. A third finding was that increased age was associated with higher accuracy and speed. This relationship is consistent with the vast literature on the DCCS paradigm, which shows reliable, robust effects of age on switching performance, particularly during the preschool years (Doebel & Zelazo, 2015; Zelazo et al., 2013). These three general effects across both groups indicate that the experimental task worked as expected.

4.1. Attention, perseveration, and stuttering

Compared to controls, CWS had more difficulty flexibly shifting attention in response to changing task demands. They showed a stronger bias toward previously relevant information, as manifested by significant group differences in RT (analyzed for correct responses only) during the postswitch phase. The tendency of the cognitive system to remain focused on what it was focused on previously is often referred to as “attentional inertia” (Diamond & Kirkham, 2005b) and can be reflected in perseverative sorting (incorrect responses) or, as found in the present study, in longer RT for correct responses after the sorting criterion changes.

Our findings most clearly align with a growing literature indicating attentional weakness in individuals who stutter based on performance differences on behavioral tasks (Anderson & Wagovich, 2016; Arnold et al., 2011; Chou, 2014; Eggers et al., 2012; Heitmann, Asbjørnsen, & Helland, 2004; Kaganovich et al., 2010; Schwenk, Conture, & Walden, 2007). Collectively, these studies suggest that stuttering is associated with difficulties focusing attention, regulating attention, resisting distractions, and recovering from distraction. The present study extends these findings by demonstrating that CWS also have difficulty shifting attention from one learned task goal to another. The fact that our sample consisted of preschoolers indicates that atypical patterns of attention switching are already apparent during the developmental period when these skills first emerge (Zelazo et al., 2003). This result extends findings of earlier studies reporting perseverative tendencies in adults who stutter and suggests that differences in attention shifting may play a role in early stuttering development. Greater susceptibility to attentional inertia in CWS compared to controls is consistent with parental observations that CWS tend to be more hypervigilant and have more difficulty adapting to novelty, shifting attention from one activity to another, and suppressing inappropriate responses than their fluent peers (Anderson et al., 2003; Eggers et al., 2010). As explained by multifactorial models of stuttering (e.g., Walden et al., 2012), these differences in attentional flexibility could contribute to persisting stuttering by making it difficult for children to move past disfluencies and react with resilience. Instead, some children may respond to disfluencies by increasing attention inward and trying to control articulatory movements through maladaptive tensing and fragmentation of motor speech sequences. This view of stuttering development is reflected in Bloodstein’s perspective of stuttering as an “anticipatory struggle” (Bloodstein, 1972, 1975, 1984), as well as the more recent vicious circle hypothesis (Vasić & Wijnen, 2005), which attributes stuttering to hypervigilant (and counterproductive) monitoring of discontinuities. Attention control in young CWS may be an important characteristic that not only predicts higher order cognitive abilities but also shapes aspects of self-regulation that can directly influence how these children manage and react to disfluencies.

4.2. Cognitive strategies

Although greater RT slowing relative to controls indicates that CWS had more difficulty disengaging from previously relevant task rules, they tended to maintain accuracy during the postswitch phase. The significant interaction between speaker type and task phase showed that although overall odds of an incorrect response were higher postswitch; this tendency was less true for CWS compared to controls. These different patterns of change for each group over the course of the task were unexpected and suggest that the two groups applied contrasting strategies to manage new task demands after the rule switch. Fluent children showed a greater likelihood of error but were less affected in response speed. In contrast, CWS maintained accuracy at the expense of speed.

This interpretation of the data is supported by results of within-group correlations between RT and accuracy. These findings indicated a general association between slow responses and reduced accuracy; however, this relationship was less true for CWS than for CWNS during the postswitch phase. Within CWS, the strength of this relationship also decreased considerably from preswitch to postswitch. This finding suggests that long RTs for CWS, specifically during the postswitch phase, may reflect use of a strategy in which accuracy was emphasized rather than speed. This pattern is more typical of older children, as mentioned above (Davidson et al., 2006; Zelazo et al., 2013) and suggests that relative to controls, CWS may be more concerned about making errors and show a greater tendency toward perfectionism. Other researchers have reported increased perfectionistic tendencies in individuals who stutter (Brocklehurst, 2008; Brocklehurst, Drake, & Corley, 2015); however, these data were based on surveys administered to adults. To our knowledge, no previous studies have demonstrated behavioral evidence of perfectionism in people who stutter.

Several behavioral studies have found the opposite pattern for CWS (fast RT with frequent errors) and suggested that stuttering is associated with impulsivity and poor response control (Blood, Blood, Maloney, Weaver, & Shaffer, 2007; Chou, 2014; Eggers, De Nil, & Van den Bergh, 2013). It is difficult to directly compare these results, given the range of behavioral tasks used (Continuous Performance Task, classic visual Go/No-Go, auditory Go/No-Go with task-irrelevant stimulus changes); however, time constraints built into certain tasks could explain why outcomes might differ from those reported in the present study. In each of these studies, target stimuli were presented for a limited amount of time and responses outside of that timeframe were considered incorrect. This type of task structure could pressure participants with more deliberate response styles to respond before they are ready and produce more errors as a result. In contrast, DCCS stimuli remained visible until the child made a selection; this design (consistent with standard protocol) would therefore allow longer RT to become apparent. The inherent conflict between dimensions of the bivalent stimuli and the relevant task rule could also have elicited a more cautious approach than tasks that do not present this sort of interference.

Although careful, deliberate response styles may support performance across many cognitive tasks, this mode of processing can be disadvantageous in other contexts. In particular, the tendency to attend excessively to articulatory processes during spontaneous speech production appears to be maladaptive (Eichorn, Marton, Schwartz, Melara, & Pirutinsky, 2016) and has been implicated in certain theoretical explanations for stuttering (Bloodstein, 1984; Vasić & Wijnen, 2005).

4.3. Speed and automatization

Reaction time data also showed group differences in learning patterns. Overall, CWS performed more slowly than fluent controls. Slower RT is a consistent finding for individuals who stutter and has been demonstrated in manual (e.g., finger tapping) as well as verbal tasks (Namasivayam & van Lieshout, 2008a; Smits-Bandstra, 2010). In the present study, RT for the two speaker groups differed initially but was comparable at the end of the task (i.e., end of postswitch phase). Thus, although CWS tended to respond more slowly at the start of the experiment, they benefited from the repetition provided by the consistent trial structure and, relative to CWNS, showed greater improvement in response efficiency over time. The control group, in contrast, showed a steadier RT pattern throughout. This result suggests that CWS require more time to show practice effects and develop automaticity for learned responses. Similar findings have been reported for AWS who took longer than controls to acquire finger tapping and nonsense syllable sequences (Smits-Bandstra et al., 2006) and showed slower stabilization and coordination of speech movement patterns (Namasivayam & van Lieshout, 2008b). These results support perspectives attributing stuttering to a less efficient underlying learning system (e.g., Smits-Bandstra & Gracco, 2013). The present study did not examine motor learning; however, slower practice effects in CWS suggest that learning differences associated with stuttering are evident at a young age and may not be limited to the motor domain.

An alternative explanation for RT changes observed in the present study may be that CWS simply had more room for improvement, whereas fluent children achieved the fastest possible response speed for children in this age group. A more challenging task (e.g., involving multiple switches) may help clarify whether group differences in improvement reflect true differences in learning patterns or are related to ceiling effects resulting from the simple nature of the task.

Methodologically, our findings related to group differences in postswitch RT, practice effects and speed/accuracy tradeoff point to the importance of continuous data in interpreting DCCS performance. Although studies using this paradigm generally analyze performance categorically (pass/fail scores) (Benitez et al., 2017; Blackwell, Chatham, Wiseheart, & Munakata, 2014; Carlson, 2005; Frye et al., 1995; Zelazo, 2006), continuous data can highlight subtle processes underlying switching performance (Yerys & Munakata, 2006).

4.4. Limitations

Several limitations in the present study warrant consideration. First, our task was relatively simple. Although it is not clear that children were performing at ceiling, many children achieved high levels of accuracy and the control group, in particular, may have reached the fastest possible RT for children their age. The addition of more demanding conditions could have provided stronger support for our interpretation of the findings, specifically those related to group differences in cognitive strategies. Additionally, the size of our sample was too small to appropriately assess the impact of individual differences in stuttering severity on switching performance. This potential relationship is an important consideration for future research on this topic. Finally, although slower responses postswitch clearly reflected the influence of preswitch rules, inaccurate responses did not unambiguously indicate a bias toward previously relevant task goals. The two foils for each trial included one stimulus consistent with the preswitch sorting rule as well as one that was not consistent with either preswitch or postswitch rules. It is possible that some incorrect responses reflected general inattention; however, error types were not differentiated. This limitation is shared by many studies using the DCCS, particularly those that only include two response options (see Carroll et al., 2016 for further discussion).

4.5. Future research

Cognitive flexibility and temperament are assumed to overlap theoretically and practically (see Eisenberg & Zhou, 2016 for recent review). Merging insights from the two perspectives could help clarify whether temperamental characteristics associated with stuttering are mirrored in performance on cognitive tasks, and how variations in the ability to modulate attention interact with early disfluencies. Additionally, although our findings provide evidence for perseverative tendencies in young CWS, it is unclear how many of these children will continue to stutter and whether difficulties in attentional control can predict stuttering persistence (vs. recovery). Future research could compare attention shifting in CWS who persisted and those who recovered. Such findings could clarify whether behavioral measures of attention may be diagnostically useful.

4.6. Clinical implications

Based on multifactorial models of stuttering, chronic stuttering may be caused, at least in part, by difficulties in shifting attention away from speech disfluencies. Recent research suggests that a child's ability to regulate attention and emotions is malleable (Swingler et al., 2015); thus, clinical intervention for CWS may benefit from novel approaches that encourage attentional flexibility (e.g., see Flook, Goldberg, Pinger, & Davidson, 2015), specifically away from stimuli perceived as negative. The tendency of CWS to maintain high levels of accuracy at the expense of slowed RT postswitch also suggests an association between stuttering and perfectionistic tendencies. Corroborating evidence from studies in adults (e.g., Brocklehurst et al., 2015) and clinical reports (Boucand et al., 2014) highlights the need for stuttering interventions that address young children's concern about errors and ability to tolerate imperfection. Suggestions for implementing this kind of approach are discussed by Boucand et al. (2014). Jones et al. (2014) also describe considerations for selecting treatment approaches based on children's attentional and temperament profiles. As an example, treatment approaches that focus on modifying disfluencies or training fluency targets to high levels of accuracy likely increase attention to disfluencies and may be counterproductive for children who are strongly reactive and have difficulty shifting their attentional focus.

5. Conclusion

In sum, results of the present study demonstrate evidence of poorer cognitive flexibility and slower rates of learning in CWS relative to fluent peers. Processes underlying perseverative tendencies and weak flexibility in CWS are not entirely clear but appear to reflect difficulty controlling attention. This finding supports multifactorial explanations of stuttering which attribute chronic stuttering to a combination of motor, linguistic, cognitive, emotional, and environmental factors. Although further research is needed to clarify the role of attention in stuttering development and its relationship to aspects of temperament, our results suggest that attentional profiles should be considered when designing and implementing interventions for young children with early stuttering. Our finding that CWS and CWNS relied on different cognitive strategies to manage task demands further indicates the relevance of error awareness, error monitoring, and tolerance of imperfection in future research and clinical approaches related to childhood stuttering.

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