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Executive function in school-aged children with cerebral palsy: Relationship with speech and language

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ABSTRACT

Background and Aims: Although children with cerebral palsy (CP) are at an increased risk for developing speech, language, and executive function (EF) impairments, little is known regarding the relationship among these risk factors. The current study examined how different profiles of speech and language impairment might be associated with impairments in EF skills in school-aged children with CP.

Methods and Procedures: Forty-seven school-aged children with CP were included. Each child contributed between one and four data points for a total of 87 data points. Children were classified into speech and language profile groups at each data point. EF skills were examined using the Behavior Rating Inventory of Executive Function questionnaire. Outcomes and Results. Compared to a mean of 50 from a normative population of children, mean scores on all measures of EF were significantly elevated for all groups ($p < .05$). The proportion of children with CP with elevated EF scores was significantly higher for all groups compared to the expected proportion in a normal population of children ($p < .05$).

Conclusions and Implications: Children with CP who do not have impairments in speech or language may be at risk for EF difficulties which may negatively affect social communication, academic performance, and functional independence.

1. Introduction

Cerebral palsy (CP) is a complex and heterogeneous disorder that can have a profound impact on all aspects of life (Kennes et al., 2002; Liptak et al., 2001). Due to brain insult experienced early in life, children with CP are at an increased risk for co-occurring problems, including impairments with sensation, perception, cognition, communication, and behavior (Rosenbaum et al., 2007).

CP is associated with a range of underlying neuropathologies including basal ganglia damage, focal infarct, cortical/subcortical damage, and malformations (Bax, Tydeman, & Flodmark, 2006). The predominant type of injury to the brain associated with CP results from damage to white matter (otherwise referred to as periventricular leukomalacia (PVL)), found in 42.5% of children with CP (Bax et al., 2006). Functional neuroimaging studies have shown that the prefrontal cortex and white matter tracts connecting the prefrontal and posterior regions of the brain (Christ, White, Brunstrom, & Abrams, 2003) are especially important for the development of executive functioning (EF) skills (Krasenegor, Lyon, & Coldman-Rakic, 1997).

Broadly defined, EF skills are a set of higher order cognitive abilities that are responsible for guiding and managing an individual's functions and behaviors (Welsh, Pennington, & Groisser, 1991). Owing to underlying neuropathologies mentioned above, children

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with CP are at risk for difficulties with EF skills (Odding, Roebroeck, & Stam, 2006). Only recently have researchers begun to examine EF skills in children with CP. Studies have found that compared to typically developing peers, adolescents and school-aged children with CP show deficits in inhibitory control, working memory, visual and auditory attention, flexibility, planning, and information processing (Anderson et al., 2010; Bodimeade, Whittingham, Lloyd, & Boyd, 2013; Christ et al., 2003; Edgin et al., 2008; Pirila, van der Meere, Rantanen, Jokiluoma, & Eriksson, 2011; Schatz, Craft, White, Park, & Figiel, 2001; White & Christ, 2005). These studies have primarily focused on examining EF skills with regard to a specific type of CP (mainly spastic) or in relation to the type of anatomical involvement (i.e., left unilateral vs. right unilateral (Bodimeade et al., 2013); bilateral vs. unilateral (Pirila et al., 2011)), or time of injury (congenital vs. peri-natal vs infancy vs. different points during childhood (Anderson et al., 2010)). Several studies have also examined the association between certain EF skills and skills from other domains, such as mathematical ability (Jenks, De Moor, & Van Lieshout, 2009; Jenks, van Lieshout, & de Moor, 2012).

Although recent work has contributed to our understanding of EF abilities in children with CP, the relationship between EF skills and speech and language problems - another common area of impairment for children with CP - is unknown (Cheney & Palmer, 1997; Krigger, 2006; Rosenbaum et al., 2007). Approximately 60% of children with CP experience some type of communication difficultly (Bax et al., 2006). These impairments may include dysarthria, a motor speech disorder affecting about half of all persons with CP (Nordberg, Miniscalco, & Lohmander, 2014), impaired language abilities, also affecting about half of all persons with CP, or a combination of impaired speech and language skills (Himmelmann, Lindh, & Hidecker, 2013; Hustad, Gorton, & Lee, 2010; Sigurdardottir & Vik, 2011). In 2010, Hustad and colleagues proposed and validated a four-group classification model based on the speech and language abilities of children with CP (Hustad et al., 2010). Children with CP who had no clinical evidence of a speech or language impairment were classified as NSMI (no speech motor involvement); those children with a clinical diagnosis of dysarthria and typical receptive language skills were classified as SMI-LCT (speech motor involvement-language comprehension typical); those with a clinical diagnosis of dysarthria and impaired receptive language skills were classified as SMI-LCI; and those children who were able to produce 5 or fewer spoken words were classified as ANAR (anarthric or the inability to produce functional speech). This model has provided a useful framework for studying different patterns of developmental change in speech and language abilities and as a tool for predicting longer term outcomes (Hustad et al., 2017).

Past research has highlighted the important role that language plays in the development of cognitive skills (Barkley, 1997; Cohen et al., 2000; Liss et al., 2001; Russell, 1997) and research from other populations of individuals with neurodevelopmental disabilities has shown that children with language impairment often present with co-occurring deficits in EF skills (Henry, Messer, & Nash, 2012; Wittke, Spaulding, & Schechtman, 2013). Less is known about the relationship between speech development and EF skills; however, due to the cognitive underpinnings of spoken language one might expect an important linkage (Barkley, 1997).

The current study utilized the four-group classification system described above (Hustad et al., 2010) in order to examine EF skills in groups of children with similar speech and language abilities, with a particular focus on the three groups of children who were able to produce speech (NSMI, SMI-LCT, and SMI-LCI). A better understanding of the relationship between speech, language, and EF abilities would help to further define the profiles of associated impairments in children with CP. Furthermore, due to the negative impacts that speech, language and EF deficits may have on functional independence in school and in the community, a more comprehensive understanding of this relationship is crucial to the development of better interventions tailored to meet needs of individual children.

The current study addressed the following questions:

1) Are there differences in mean scores on standardized measures of EF among three profile groups of children with CP (NSMI, SMI-LCT, and SMI-LCI)?
2) Are mean scores on standardized measures of EF within each of three profile groups of children with CP different from an expected typically developing mean score of 50?
3) Are there differences in the proportion of clinically elevated scores on standardized measures of EF between the three groups of children with CP?
4) Is the proportion of children with clinically elevated scores on standardized measures of EF within each of the three profile groups of children with CP greater than the expected proportion in typically developing children?

2. Methods

2.1. Participants

Participants were selected from a larger cohort of children (n = 139) participating in a longitudinal study on communication development in children with CP. Between 2005 and 2012, children were recruited though local and regional neurology and physiatry clinics in the Upper Midwestern region of the US. Children were between 2;0 and 5;0 years old upon initial enrollment. Children were seen twice a year, roughly 6 months apart, until their 8th birthday, after which children were seen yearly. Criteria for inclusion into the larger study required that children have a medical diagnosis of CP and have hearing within normal limits as documented by either formal audiological evaluation or distortion product otoacoustic emission screening. Inclusion criteria for the current study were: 1) have at least one completed data collection session between the ages of 5;0 and 15;0; and 2) have at least one valid Behavior Rating Inventory of Executive Function (BRIEF; Gioia, Isquith, Guy, & Kenworthy, 2000) completed by a parent. A total of 47 children with CP (27 males) met criteria and were included in the current study. Table 1 presents demographic characteristics of these children. All children were born between 2001 and 2009. Because the larger study was longitudinal, each child

Table 1
Demographic characteristics of children with CP.

	NSMI n = 13	SMI-LCT n = 14	SMI-LCI n = 20
Male: Female ratio	10:3	7:7	10:10
Type of CP			
Spastic			
Diplegia	4	3	5
Hemiplegia (left)	5	1	3
Hemiplegia (right)	3	1	8
Triplegia	0	1	0
Quadriplegia	0	3	0
Dyskinetic	0	1	0
Ataxic	0	3	1
Mixed	0	0	0
Unknown	1	1	3
GMFCS			
I	11	7	4
II	2	3	11
III	0	2	2
IV	0	2	1
V	0	0	2

NSMI: no speech motor involvement; SMI-LCT: speech motor involvement – language comprehension typical; SMI-LCI: speech motor involvement – language comprehension impaired.

contributed between one and four data points for a total of 87 data points across the 47 children included in this study. Seventeen children contributed one data point; 21 children contributed 2 data points; 8 children contributed 3 data points; and one child contributed 4 data points. Time between data points ranged from 6 months to 25 months.

2.2. Materials and procedures

As part of the larger longitudinal study, a standard research protocol was administered to participants at each visit (see Hustad et al., 2010). For the present study, EF abilities, as measured by the BRIEF questionnaire, were of primary interest. Additionally, measures of speech production, spontaneous spoken communication, and language comprehension were used for the classification of children into speech-language profile groups.

2.2.1. Executive function

The BRIEF (Gioia et al., 2000) is a parent questionnaire used to examine EF skills beginning at 5;0 years of age. We began having parents complete this measure as part of our longitudinal study in 2014. Data for the current study was collected between October 2014 and December 2016. At this time, the youngest child in the study was 5;5 years old; however, most participants were between 8 and 9 years old. The BRIEF consists of 86 items assessing eight domains of EF: Inhibit, Shift, Emotional Control, Initiate, Working Memory, Plan/Organize, Organization of Materials, and Monitor. Caregivers are asked to rate their child's behavior on a 3-point Likert-type scale (1 = behavior is never a problem; 2 = behavior is sometimes a problem; 3 = behavior is often a problem). Two index scores (Behavioral Regulation Index (BRI) and Metacognition Index (MI)) and one composite score (Global Executive Composite (GEC)) are derived from parent responses to questions.

Both index scores (BRI and MI) as well as the composite score (GEC) from the BRIEF were used as measures of EF in the current study. According to the technical manual, the BRI "represents the child's ability to shift cognitive set and modulate emotions and behaviors via appropriate inhibitory control" (Gioia et al., 2000, p. 20). The BRI is comprised of the Inhibit, Shift, and Emotional Control subscales. The MI reflects the child's "ability to cognitively self-manage tasks and reflects the child's ability to monitor his or her performance" (Gioia et al., 2000, p. 20) and is comprised of the Initiate, Working Memory, Plan/Organize, Organization of Materials, and Monitor subscales. Raw scores from each of the index measures of EF (BRI, MI, GEC) were used to generate normative T scores based on the age and gender of the individual and the raw score distribution in the standardization sample. The child's T scores for these measures have a mean of 50 and standard deviation of 10 in a normative population of children (Gioia et al., 2000). T scores at or above 65 (50 + 1.5 SD) are considered abnormally elevated and clinically significant. The questionnaire was completed at the time of participation or by mail just prior to participation. Per the directions on the questionnaire, caregivers were asked to answer all questions regarding their child's behavior over the past 6 months.

2.2.2. Classification of speech-language profile groups

Children were classified into speech-language profile groups following Hustad et al. (2010). All classifications were made by the first author, who was involved in the data collection sessions for all participants. Classifications were made for each visit either immediately following the data collection session or by listening to collected audio and video samples of elicited and spontaneous spoken communication. Classifications were made based on clinical judgement of the presence or absence of speech motor impairment and the presence or absence of language impairment.

Speech motor impairment was determined through clinical observation of the presence or absence of common features of dysarthria, including facial asymmetry, drooling, hypernasality, short breath groups, breathy, harsh, or wet vocal quality, imprecise articulation, and consonant or vowel substitutions, distortions, or omissions that were not age-appropriate. Perceptual judgements were made during two tasks: a) a delayed imitation task in which the child produced a standard set of sentences ranging from 2 to 7 words in length following an adult model (Hodge & Daniels, 2007), and b) a spontaneous speech sample either between the child and parent or the child and clinician.

The presence or absence of receptive language impairment was based on clinical impressions of language ability and considering performance on the Peabody Picture Vocabulary Test – 4 (PPVT-4; Dunn & Dunn, 2007) or the Test of Auditory Comprehension of Language –3 (TACL-3; Carrow-Woolfolk, 1999). Both measures are norm-referenced assessments of receptive language (vocabulary for PPVT-4; and a broader range of vocabulary, grammar, and syntax for TACL-3). In each test, children are presented with an array of pictures and asked to select the picture that best matches the stimulus item spoken by the examiner. In the current study, the PPVT-4 was administered to participants who were unable to tolerate the longer TACL-3 or were outside the upper age limit of the TACL-3. Although the PPVT-4 provides a narrower examination of language comprehension, it has been widely used a basic tool for language comprehension measurement (Burchinal, Peisner-Feinberg, Pianta, & Howes, 2002; Spere, Schmidt, Theall-Honey, & Martin-Chang, 2004).

Both the PPVT-4 and TACL-3 yield raw scores which can be converted to age-normed standard scores with a mean of 100 and a standard deviation (SD) of 15. For classification purposes, children with standard scores lower than 1 SD below the mean (i.e., 84 or lower) were classified as language impaired and children with standard scores equal to or higher than 1 SD below the mean (i.e., 85 or higher) were classified as having typical language skills.

In the current study, 13 children were classified in the NSMI profile group, 14 children in the SMI-LCT group, and 20 children were classified as SMI-LCI. Children who contributed more than one data point in the current study did not change profile group membership. There were four instances where a valid measure of receptive language could not be obtained for a data collection session due to child factors such as fatigue, inattentiveness, or behavior. For these four cases, profile group membership was made based on clinical impressions and test scores from previous data points.

2.3. Theory/calculation

Mean T scores on the two index measures (BRI and MI) and one composite measure (GEC) were estimated for each clinical profile group in a linear mixed model using all data points from all participants (Laird & Ware, 1982), model predictors were indicator (dummy) variables for the two SMI groups (and NSMI serving as the reference group). In pre-planned analyses, a child-level random effect was used to account for the correlation between repeated measures on the same participant – a method that is valid even for varying numbers of observations per participant, allowing us to use all available measures on each participant in each group. Using the fitted model, differences in mean T scores (contrasts) between each pair of the three groups were also estimated, and tested using two-sided hypothesis tests. 95% confidence intervals were constructed around the estimated means and contrasts. Two degree-of-freedom tests of equal means across the three groups were performed to test whether the three groups had the same mean T score, versus the alternative that at least one group differs from at least one other group. Finally, in the context of the fitted model, the estimated T score means for each clinical profile group were compared to a normative value of 50, the average in a normal population of children (Gioia et al., 2000), using one-tailed tests for an alternative mean greater than 50 versus a null hypothesis equal to 50.

In order to extract the estimated proportion of children with CP with T scores over 65 (indicating abnormally elevated and clinically significant EF abilities) in each clinical profile group, we classified T scores into < 65 and > 65 and modelled the binary response of being > 65. In these models, each participant's response is his or her proportion of the number of instances (or data points) of T scores over 65, divided by the total number of scores for that participant. These proportions were modelled using logistic regression for binomial data, with predictors being indicator (dummy) variables for the two SMI groups (and NSMI serving as the reference group). We allowed for overdispersion in the binomial model, accounting for extra variance due to correlated repeated binary measurements from each participant making up the proportion (McCullagh & Nelder, 1989). Using the fitted model, we constructed 95% confidence intervals for the proportion of participants with T scores on any of the three BRIEF measures over 65. Two degree-of-freedom tests of equal proportions across the three groups were performed to examine whether the clinical profile group proportions differed in any one group. Finally, estimated group percentage > 65 was compared to a reference of 6.68%, the proportion of children in a normal population with mean 50 and SD 15 who would have a T score above 65, using one-tailed tests for an alternative proportion greater than 6.68%.

3. Results

Fig. 1 plots the distribution of T scores by clinical profile group for each BRIEF measure of EF. Descriptive results suggest considerable variability and overlap on each measure of EF for all profile groups. Means and standard deviations (SD) of each BRIEF score by group are also given in Table 2.

3.1. Between group differences in mean scores on EF measures

Table 2 shows mixed model-based mean T scores on each of the three measures of EF for each profile group, along with 95%

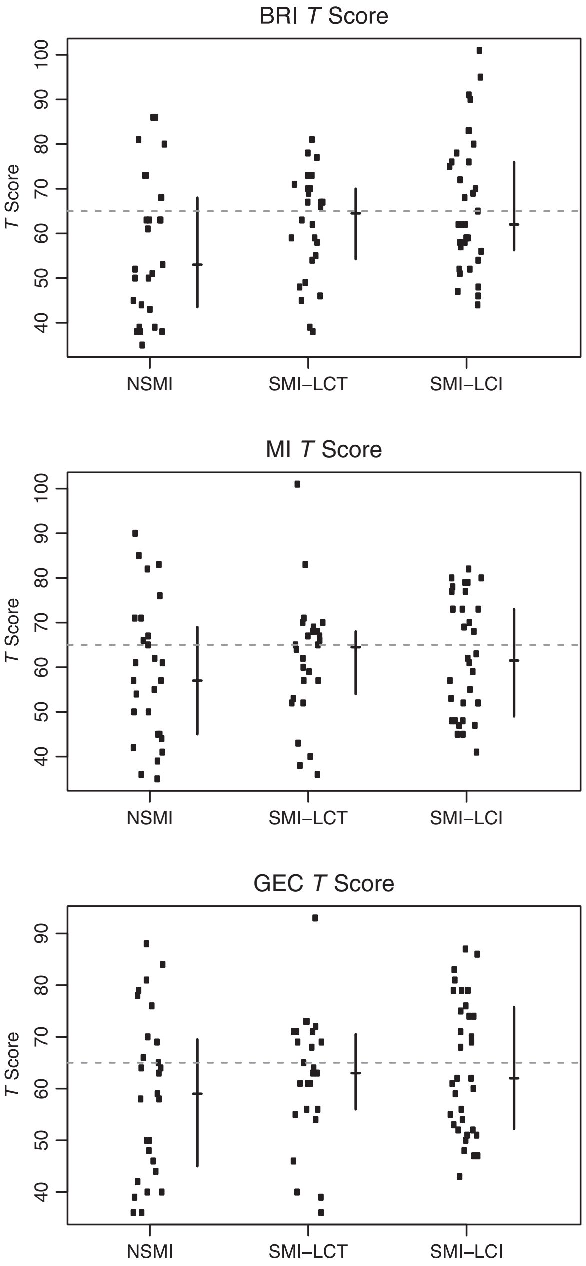

Fig. 1. Descriptive plot of *T* scores for each measure of EF by speech profile group. Top = BRI (Behavior Regulation Index); middle = MI (Metacognition Index); Bottom = GEC (Global Executive Composite). NSMI = no speech motor impairment; SMI-LCT = speech motor impairment and typical language abilities; SMI-LCI = speech motor impairment and impaired language skills. Notes: Points are observed *T* values, jittered horizontally to avoid overlap. Bars run from 1st to 3rd quartile with hashmark indicating the median. Dotted line is threshold of 65 for an elevated *T* score. Plot contains 87 points on 47 children.

confidence intervals derived from the model. Inferential chi-square statistics, presented in a footnote to Table 2, showed no significant difference among clinical profile group means on the BRI, MI, or GEC *T* scores. The pairwise contrasts in mean *T* scores between the three groups yielded only one significant difference, although the general pattern was the SMI-LCI had the highest scores, followed by SMI-LCT, and then by NSMI.

We also note as a side comment that repeated measures over time on these three measures were strongly correlated, the intraclass correlations for measures on the same child estimated as 0.81, 0.67, and 0.73 for BRI, MI, and GEC *T* scores respectively.

Table 2

Estimated mean *T* scores (compared to an expected typically developing mean score of 50), group contrasts, and confidence intervals on the Behavioral Regulation Index (BRI), Metacognition Index (MI), and Global Executive Composite (GEC) by speech-language profile group from linear mixed models.

	Group	Mean	LCL	UCL	*Z*-value[a]	*P*-value[b]
BRI	SMI-LCT vs. NSMI[c]	4.8	−5.8	15.5	0.89	0.37
	SMI-LCI vs. NSMI	10.4	0.5	20.3	2.06	0.04*
	SMI-LCI vs. SMI-LCT	5.6	−4.2	15.3	1.12	0.26
	NSMI	57.3 (15.9)[d]	49.6	65.0	1.86	0.031*
	SMI-LCT	62.1 (12.0)	54.7	69.6	3.19	< 0.001*
	SMI-LCI	67.7 (14.9)	61.4	73.9	5.53	< 0.001*
MI	SMI-LCT vs. NSMI	4.7	−5.5	14.8	0.90	0.37
	SMI-LCI vs. NSMI	3.5	−5.9	13.0	0.74	0.46
	SMI-LCI vs. SMI-LCT	−1.1	−10.4	8.2	−0.24	0.81
	NSMI	58.7 (15.7)	51.5	66.0	2.36	0.009*
	SMI-LCT	63.4 (13.9)	56.3	70.5	3.70	< 0.001*
	SMI-LCI	62.3 (13.2)	56.3	68.3	4.01	< 0.001*
GEC	SMI-LCT vs. NSMI	4.4	−5.6	14.3	0.86	0.39
	SMI-LCI vs. NSMI	6.6	−2.6	15.8	1.40	0.16
	SMI-LCI vs. SMI-LCT	2.2	−6.9	11.3	0.48	0.63
	NSMI	58.4 (15.8)	51.3	65.6	2.32	0.01*
	SMI-LCT	62.8 (12.4)	55.9	69.8	3.62	< 0.001*
	SMI-LCI	65.0 (13.1)	59.2	70.9	5.03	< 0.001*

BRI: Test of Equal Means among 3 groups: Chisq = 4.423, Df = 2, $P = 0.11$.
MI: Test of Equal Means among 3 groups: Chisq = 0.937, Df = 2, $P = 0.626$.
GEC: Test of Equal Means among 3 groups: Chisq = 2.052, Df = 2, $P = 0.358$.

[a] *Z*-value for means is for testing against the null (normative) value of 50. *Z*-value for contrasts is for testing against the usual null value of 0.

[b] *P*-value for means is for one-sided alternative that mean is greater than 50. *P*-value for contrasts is for two-sided alternative that contrast differs from 0.

[c] NSMI: no speech motor involvement; SMI-LCT: speech motor involvement – language comprehension typical; SMI-LCI: speech motor involvement – language comprehension impaired.

[d] Mean's are from fitted mixed effects models; SD's are the crude values computed from all observations.

* P-values of 0.05 or less were considered significant.

3.2. Differences in mean scores on EF measures relative to expected population means

Table 2 shows model-estimated mean *T* scores on each of the three measures of EF for each profile group as compared to an expected typically developing mean score of 50. When compared with the mean of 50 from a normative population of children, mean *T* scores on the BRI, MI, and GEC were all significantly higher than 50 for all clinical profile groups in one-sided tests (*P*-values given in Table 2).

3.3. Between group differences in proportion of elevated scores on EF measures

On each measure of EF, *T* scores at or above 65 indicate clinically significant EF dysfunction. Table 3 shows binomial model-based estimates of the percentage of participants in each clinical profile group with elevated *T* scores (greater than 65) for each BRIEF measure of EF, along with 95% confidence intervals for those percentages. Chi-square test results given in the footnote to Table 3 show no significant differences between groups on the BRI, MI, or GEC *T* scores.

3.4. Differences in proportion of elevated scores on EF measures relative to expected population proportions

In a population of typically developing children, 6.68% are estimated to have an abnormally elevated and clinically significant *T* score (at or above 65). Inferential statistics, including *Z*- and *P*-values, presented in Table 3, show that the percentage of children with elevated BRI *T* scores was significantly higher than 6.68% for all clinical profile groups, and similar results were obtained for MI and GEC, with these proportions varying from 30% to 50%, and the lowest lower confidence limit being 14%, all considerably higher than 6.68%.

4. Discussion

Results from the current study revealed several important findings. First, children with CP present with EF difficulties. Our results showed that children with CP in all profile groups had average BRIEF scores that were higher than the typically developing population mean of 50. Similarly, the proportion of children with CP with EF difficulties in each profile group was much higher than would be expected in a typically developing population of children. These findings are in line with past research showing that children with CP are at a greater risk for having EF difficulties due to the nature of the brain insult (Odding et al., 2006); however, the

Table 3
Estimated proportion of clinically elevated T scores (i.e., $T > 65$) on the Behavioral Regulation Index (BRI), Metacognition Index (MI), and Global Executive Composite (GEC) by speech-language profile group from binomial logistic models with overdispersion.

	Group	Proportion (%)	LCL	UCL	Z-value[a]	P-value[b]
BRI	NSMI[c]	29.6	13.5	53.3	3.49	< 0.001*
	SMI-LCT	50.0	28.4	71.6	5.58	< 0.001*
	SMI-LCI	47.1	28.3	66.7	6.08	< 0.001*
MI	NSMI	37.0	18.5	60.3	4.35	< 0.001*
	SMI-LCT	50.0	28.2	71.8	5.53	< 0.001*
	SMI-LCI	44.1	25.7	64.3	5.72	< 0.001*
GEC	NSMI	37.0	18.8	59.8	4.44	< 0.001*
	SMI-LCT	42.3	22.5	64.9	4.93	< 0.001*
	SMI-LCI	47.1	28.5	66.5	6.16	< 0.001*

BRI: Test of Equal % among 3 groups: Chisq = 2.782, Df = 2, $P = 0.384$.
MI: Test of Equal % among 3 groups: Chisq = 0.913, Df = 2, $P = 0.734$.
GEC: Test of Equal % among 3 groups: Chisq = 0.621, Df = 2, $P = 0.803$.

[a] Z-value for testing against the null value of 6.68%, the percent over 65 in a normative population with mean 50 and SD = 10.

[b] P-value for one-sided alternative that percent over 65 is greater than 6.68%.

[c] NSMI: no speech motor involvement; SMI-LCT: speech motor involvement – language comprehension typical; SMI-LCI: speech motor involvement – language comprehension impaired.

* P-values of 0.05 or less were considered significant.

proportions of children with EF difficulties were much larger than expected. When there is an injury to the brain, such as PVL, it is unlikely that the damage will be contained to one area. For example, interruption of motor tracts can lead to motor deficits, lesions in the periventricular white matter in anterior and parietal regions of the brain can result in impaired attention and executive functioning (Schatz et al., 2001; White & Christ, 2005; Pavlova, Sokolov, Birbaumer, & Krägeloh-Mann, 2008). An injury to the brain can comprise the entire cerebral system and in turn, cause problems in multiple areas including vision, sensation, behavior, communication and cognition (Bottcher, 2010). It may be the case then, that in CP, EF impairment is a comorbidity or a co-occurring difficulty that may be present in children and is less influenced by or related to other impairments, as in the case for other neurodevelopmental disorders.

Results of this study also suggest that the association between EF skills and speech-language profile group membership for children with CP may not be as straightforward as has been found in other populations (Henry et al., 2012; Wittke et al., 2013). In our study, there was no difference among the three profile groups of children with CP in mean global EF scores or in proportion of global EF scores that were clinically elevated. It is possible, however, that the speech deficits of children with CP with typical language skills (i.e., those in the NSMI and SMI-LCT profile groups) may contribute to the presentation of EF difficulties. Children in the SMI-LCT group had a clinical diagnosis of dysarthria, a speech motor disorder that often leads to reduced intelligibility. Although children in the NSMI group had no clinical evidence of a speech motor disorder, past research from our group has shown that these children have reductions in speech intelligibility relative to typically developing peers (Hustad, Schueler, Schultz, & DuHadway, 2012). Reductions in speech intelligibility can lead to communication difficulties and may manifest as social and/or behavioral challenges (Whittingham, Fahey, Rawicki, & Boyd, 2010). Children with social or behavioral challenges may exhibit certain behaviors, such as becoming tearful easily, acting wilder or sillier than others, or not noticing that their behavior causes a negative reaction. These behaviors could be interpreted as having problems with emotional control, behavior inhibition, or behavior monitoring – all of which fall under the construct of EF. It is also important to note that in the current study, all measures of EF - Behavioral Regulation (BRI), Metacognition (MI), and the Global Executive Composite (GEC) – had similar means and proportions of children with elevated scores. Thus, our findings suggest that children with CP experience EF deficits that spread across various subdomains of EF and that these deficits are not directly associated with a child's speech or language abilities. More research involving a larger sample size is needed; however, and future studies should explore this relationship at a deeper level by examining specific aspects of speech, receptive language, and EF abilities.

Taken together, results of the current study suggest that careful considerations need to be made when designing interventions for children with CP. Intervention for children with CP and impaired speech and language skills should consider all areas of impairment related to language and cognition. The finding that even children with CP who present with typical speech and/or language skills also struggle with EF skills holds great significance when considering whether these children are receiving services to address difficulties. It may be that their strong speech and language skills mask EF difficulties and these children may be overlooked for intervention. This becomes especially important to consider as children enter school-age years. More demands are placed for independent learning and functioning, and children with CP may start to struggle academically and socially due to their EF difficulties. This susceptibility for EF deficits should be recognized and considered for all children with CP – even those without co-occurring impairments in speech and language.

4.1. Conclusions and limitations

This study provides new information concerning the relationship between speech, language and EF abilities in school-aged children with CP. Compared to a normal population of children, children with CP present with EF deficits, regardless of their speech and language abilities. In addition, the proportion of children with CP with clinically significant elevated EF scores is significantly higher than the expected proportion in a normal population of children. Both of these findings hold true for all children regardless of speech-language profile group membership.

There were several limitations of the current study. First, the sample size was small and was not fully representative of the larger population of children with CP. In particular, our sample did not include children in the anarthric speech-language profile group. Children with anarthria are often more severely affected in terms of gross motor impairment and cognitive involvement. Because the BRIEF includes items that require a certain level of independent functioning, many items do not apply for children with more complex needs.

Another related limitation to the current study was the use of BRIEF, a caregiver report measure, to access EF abilities in children with CP. There are several issues with using a caregiver report measure that are important to discuss. First, although the BRIEF is a standard clinical tool for measuring EF skills in children, it is an indirect measure and ratings are obtained through the lens of parents. There are other ways to characterize EF skills in children by utilizing more direct assessment tools. Future studies should consider including both direct and indirect measurements of EF in order to fully characterize EF skills. Second, although the use of caregiver rating systems to access different areas of child functioning is frequently used and well-documented in the literature (Achenbach, 1991; Conners, 1989; Reynolds & Kamphaus, 1992), it is possible that parent ratings of EF problems are influenced by the severity of a child's disability. Depending on the severity of their child's disability, it may be that caregivers are over-reporting or under-reporting behaviors as problematic. For example, a child with CP and co-occurring speech and language impairments likely receives speech and language therapy and may have extra support during their school day. With extra support, the number of opportunities to independently demonstrate certain behaviors such as handing in homework on time or checking work for mistakes, may be reduced or interpreted by caregivers as being completed independently and therefore, not problematic. It is also possible that caregiver expectations are influenced by the severity of their child's disability. Parents of children with CP who are developing typically in terms of speech and language skills may have higher expectations of independent EF and therefore report more behaviors as problematic. Future studies should examine both direct and indirect assessments of EF in order to understand the nature of EF deficits in school-aged children with CP.

Finally, the current study did not examine EF deficits in relation to gross motor functioning or manual abilities. Future studies should explore this association by examining subdomains of functioning.

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