

# Executive functioning in children with unilateral cerebral palsy: protocol for a cross-sectional study

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## ABSTRACT

**Introduction:** Early brain injury, as found in children with unilateral cerebral palsy (CP), may cause deficits in higher-order cognitive tasks known as executive functions (EF). EF has been conceptualised as comprised of four distinct yet inter-related components: (1) attentional control, (2) cognitive flexibility, (3) goal setting and (4) information processing. The aim of this study was to examine EF in children with unilateral CP and compare their performance with a typically developing reference group (TDC). The potential laterality effects of unilateral CP on EF will be explored, as will the relationship between the cognitive measures of EF, behavioural manifestations of EF, psychological functioning and clinical features of unilateral CP.

**Methods and analysis:** This cross-sectional study aims to recruit a total of 42 children with unilateral CP (21 right unilateral CP and 21 left unilateral CP) and 21 TDC aged between 8 and 16 years. Clinical severity will be described for gross motor function and manual ability. Outcomes for cognitive EF measures will include subtests from the Wechsler Intelligence Scale for Children—Fourth Edition, Delis-Kaplan Executive Function System, Rey Complex Figure Test and the Test of Everyday Attention for Children. Behavioural manifestations of EF will be assessed using the Behaviour Rating Inventory of Executive Function, Parent and Teacher versions. Psychological functioning will be examined using the Strengths and Difficulties Questionnaire. Between-groups differences will be examined in a series of one-way analyses of covariance and followed up using linear comparisons. An overall composite of cognitive EF measures will be created. Bivariate correlations between the EF composite and psychological measures will be calculated.

**Ethics and dissemination:** This protocol describes a study that, to our knowledge, is the first to examine multiple components of EF using a cohort of children with unilateral CP. Exploration of potential laterality effects of EF among children with a congenital, unilateral brain injury is also novel. Possible relationships between EF and psychological functioning will also be investigated. Ethics have been obtained through the University of Queensland School of Psychology Ethics Committee and the Queensland Children's Health Services Human Research Ethics Committee. Results will be disseminated in peer reviewed publications and presentations at national and

international conferences. This study is registered with the Australian New Zealand Clinical Trials Registry (ACTRN12611000263998).

## INTRODUCTION

Cerebral palsy (CP) is the leading cause of childhood physical disability in Australia with an incidence of 1 in 500 live-births.<sup>1</sup> Unilateral CP, with a presumed brain lesion occurring congenitally prior to 28 days corrected age, is the most common type of CP among children born full term and the second most common type of CP in children born preterm, with an incidence of 1 in 1300 live-births.<sup>1 2</sup> CP has been defined as 'a group of disorders of the development of movement and posture...that is attributed to non-progressive disturbances that occurred in the developing fetal or infant brain...often accompanied by disturbances of ...cognition'.<sup>3</sup>

Caring for people with CP is costly on the healthcare system as well as families. In 2007, the overall financial expenditure for persons with CP in Australia was AU\$1.47 billion.<sup>4</sup> A population-based register study of children with CP in Australia identified that 45% of children with CP experience cognitive difficulties.<sup>5</sup> In later life, CP is related to reduced educational and employment opportunities.<sup>6</sup> In comparison with research on motor and movement impairments in CP, there is a lack of literature examining cognitive and psychological difficulties faced by children with CP.<sup>7</sup> This is concerning given that these factors are essential to the well-being and overall development of children with CP.<sup>8</sup>

Another diagnostic marker for CP is the damage to the developing fetal or infant brain. A key systematic review by Krageloh-Mann and Horber<sup>9</sup> analysed the MRI brain scan findings for children with CP and found that in children with unilateral CP, periventricular white matter damage was the most common brain

injury, occurring in 36%, followed by cortical deep grey matter lesions in 31%, brain malformations (eg, schizencephaly) in 16% and miscellaneous lesions in 7%. Given that children with unilateral CP have sustained a brain injury, and the fact that research has illustrated a link between brain injuries and reduced cognitive and psychological functioning,<sup>10</sup> an examination of the neuropsychological and psychological functioning in this population is warranted.

### Executive function

'Executive function' (EF) is an umbrella term that encompasses the skills necessary for novel, goal-directed and complex activity.<sup>11–15</sup> Everyday functioning relies on executive skills and deficits in EF may manifest as disorganisation and poor planning, inability to focus and attend to tasks, carelessness in responding to tasks, reduced self-control and taking longer to complete tasks.<sup>16–17</sup> Findings from functional neuroimaging studies, predominantly in the adult brain-injured populations, have indicated that EF is principally mediated by the frontal lobes, particularly the prefrontal cortex.<sup>18–19</sup> The frontal lobes demonstrate rich efferent and afferent connections with nearly all other posterior and subcortical cerebral regions.<sup>20–21</sup> It is thought that the frontal lobes integrate and coordinate information and, in essence, work as the 'control master' of the brain.<sup>22</sup> As a consequence, the frontal lobes are important for EF but it is the integrity of the entire brain that is pivotal for successful executive skills.<sup>16–23</sup> In children and adolescents, the frontal lobes are the last brain region to reach maturity, typically by the end of the second decade of life.<sup>24</sup> The refinement of intricate white matter tracts from these underlying brain regions to the frontal lobes and ongoing myelination are also important aspects of prefrontal maturation and, in turn, the progression of executive skills.<sup>24</sup>

There is some evidence suggesting lateralisation of the verbal and spatial aspects of executive functioning among adults. For example, utilising positron-emission tomography, asymmetrical organisation of visual and verbal working memory skills, a component of EF, was shown among a cohort of female adults aged 18–30 years. A predominantly left lateralisation occurred during a verbal memory task, whereas right lateralisation was shown during a spatial working memory task.<sup>25</sup> However, within the paediatric literature, there is a paucity of research exploring the possible laterality of EFs among children, and findings from adult cohorts cannot be extrapolated to children, given their ongoing development. Moreover, among unilateral CP, a congenital brain injury has occurred, rather than one acquired later during development. This may also change the picture of potential lateralisation, given the possibility of functional reorganisation in the developing brain.<sup>26</sup>

Specifically, there is some evidence of functional relateralisation of the lower level cognitive functions in children, particularly related to the visuospatial and

language skills, following early brain injury.<sup>27</sup> Research by Lidzba *et al*<sup>27</sup> has highlighted that children with both left and right unilateral CP show preserved language functions at the cost, however, of poorer visuospatial skills. It appears that visuospatial deficits in children with early left hemispheric lesions are a consequence of lesion-induced right hemispheric language reorganisation. This phenomenon is known as the cognitive-crowding hypothesis.<sup>26</sup>

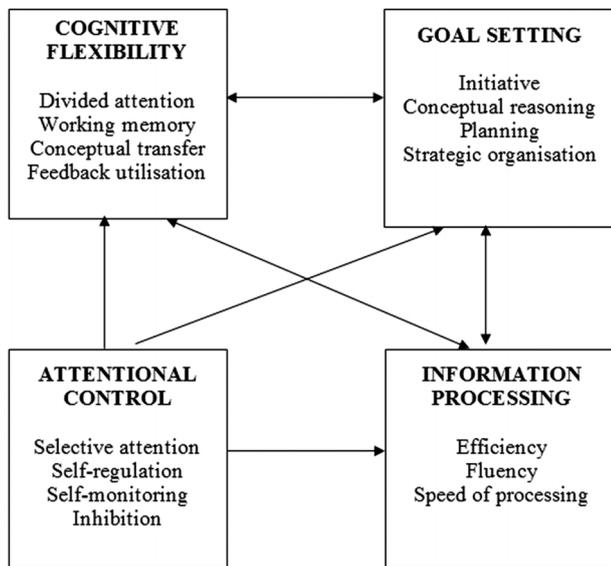
### Development of EFs

As executive skills show a prolonged development through childhood and adolescence, it is important to understand the normal development of these skills in order to identify deviations from the projected maturational patterns. An analogous relationship between the maturing frontal lobes and the unfolding of executive skills is seen.<sup>11</sup> This parallel relationship typically emerges along a hierarchical developmental trajectory often in growth 'spurts' rather than developing in a uniform fashion.<sup>28–29</sup> Major neurophysiological growth spurts occur from birth to 2 years, 7 to 9 years and again in adolescence from 16 to 19 years.<sup>11–30–31</sup> These time frames involve peak periods of synaptogenesis and increased myelination with corresponding improvements in specific EF domains.<sup>28</sup>

A conceptual framework of EF in typically developing children, proposed by Anderson,<sup>32</sup> operationalises EF as an overall control system that comprises four distinct, yet inter-related, executive components: (1) attentional control, the earliest EF domain to emerge, involves the ability to maintain and focus attention for extended periods of time and the capability to selectively focus one's attention towards target stimuli; (2) cognitive flexibility, the ability to correct and learn from errors, flexibly shift from one response set to another and generate multiple and alternative strategies to problems; (3) goal setting, the ability to generate novel goals and initiatives, plan actions and strategies and complete tasks in an organised and proficient manner and (4) information processing, the ability to fluently and efficiently complete tasks and the overall processing speed and speed of output (figure 1). This model of EF is unique in the paediatric neuropsychological literature as it incorporates a developmental context, highlights that each executive component operates in an integrative manner and considers each component as having a separate developmental trajectory.

### EFs in CP

Despite the importance of executive skills for the successful achievement of academic, behavioural, social and adaptive day-to-day functions, there is a paucity of research examining EF in children and adolescents with unilateral CP. Research among the other paediatric populations, such as in childhood stroke<sup>33</sup> and focal frontal lobe lesions,<sup>34</sup> has shown that EF is particularly susceptible to early brain insult during the prenatal and



**Figure 1** Model of executive function in children proposed by Anderson.<sup>32</sup>

perinatal periods. Recent research has also established that executive difficulties are present following early brain insult to any region of the brain—it does not need to be a frontal lesion for executive deficits to be seen.<sup>35–37</sup> Brain injury sustained early in development (ie, before age 3) has been shown to result in global executive deficits across several executive components.<sup>35–38</sup>

Recent research has noted EF deficits among children and adolescents with CP.<sup>39–42</sup> In Bottcher *et al's*<sup>39</sup> study, children (9–13 years) with either unilateral CP (n=14) or diplegia (n=18) were found to have attentional deficits, as measured by subtests from the Test of Everyday Attention for Children (TEA-Ch), EF deficits, as measured by the Contingency Naming Test, and deficits in behavioural manifestations of EF in everyday life as measured by the Behaviour Rating Inventory of Executive Function (BRIEF). It was found that both the unilateral and diplegia CP groups scored significantly below the age-based norms on all measures and there was a non-significant trend for children with diplegia to perform poorer than those with unilateral CP.<sup>39</sup> In a similar study, a smaller cohort of children (8–17 years), again with either unilateral CP (n=8) or diplegia (n=9), were rated as showing clinically significant impairments on measures of attention, impulsivity and vigilance from the Conners' Continuous Performance Test, with children with diplegia showing significantly higher impairments than those with unilateral CP.<sup>42</sup>

The relationship between arithmetic difficulties and EF in children with CP has also been investigated.<sup>40–41</sup> In one study, first graders (mean age of 7 years) with CP were split into two groups—those attending a mainstream school (n=16) and those attending a special school (n=41).<sup>41</sup> A control group of 16 first graders without CP, again with a mean age of 7 years, who were attending a mainstream school, were also included.

Within the CP mainstream school group, 12 children had unilateral CP, 3 had diplegia and one had ataxia, while the CP special school group comprised 10 children with unilateral CP, 29 with diplegia and 2 with ataxia.<sup>41</sup> Executive skills, specifically verbal and visuo-spatial working memory, were assessed using a Digits Forwards and Backwards task and the Knox Blocks test.

Interestingly, the CP mainstream group had the lowest score on the Digits Forwards tasks, followed by the CP special school group and then the controls. Although the CP mainstream group performed more poorly than the CP special school group and the CP special school group lower than the controls, neither of these differences reached clinical significance. On the Digits Backwards task, the CP special school group performed significantly worse than the CP mainstream group and the CP mainstream group performed significantly poorer than the control group. Finally, on the Knox Blocks task, the CP special school group performed significantly worse than the CP mainstream group; however, there was no difference in performance between the CP mainstream and control groups.<sup>41</sup> Structural equation modelling revealed that tasks assessing working memory skills (ie, Digits Forwards/Backwards and Knox Blocks) mediated arithmetic ability in both CP groups, such that poorer working memory abilities predicted a lower arithmetic ability.<sup>41</sup> A follow-on study by the same authors confirmed that EFs, particularly working memory skills, are lower in children with CP (CP types included hemiplegia, diplegia and ataxia) compared with their typically developing peers and that these predict poorer arithmetic ability.<sup>40</sup>

EFs have also been examined in a study of 21 school-age children (mean age of 8 years) who had been born preterm with a periventricular haemorrhagic infarction.<sup>43</sup> Of these children, 13 had unilateral CP, three had diplegia, one had minor neurologic dysfunction and four were neurologically normal. The BRIEF was used as the outcome measure for executive skills with the results showing executive impairments in 18% (parent's report) and 29% (teacher's report) of the sample.<sup>43</sup> Other research has used the Wisconsin Card Sorting Test (WCST) to examine executive skills among 37 children with unilateral CP and 15 children with diplegia (mean age 11 years) and 50 matched typically developing peers.<sup>44</sup> Results found that children with CP, compared with controls, made more non-preservative errors, completed fewer categories, required more trials to complete the first category and gave fewer conceptual responses.

This current literature is limited as all existing studies examine mixed groups of CP and/or investigate only one discrete component of EF; thus, the heterogeneous nature of CP and the multidimensional nature of EF are not accounted for and the results may be misleading. Furthermore, the majority of studies lack a typically developing reference group and also do not include both cognitive and behavioural measures of EF. Furthermore, the relationship between cognitive EF and

the behavioural manifestations of EF and psychological functioning has also not been previously explored. This study aims to remedy these gaps in the literature.

### EFs following early brain injury

Research among children who had sustained an early brain injury has also uncovered EF deficits.<sup>35 45</sup> Using a cross-sectional, retrospective group design, Anderson *et al*<sup>35</sup> examined EF among 164 children (aged 10–16 years) who had sustained a brain injury at varying developmental time points: congenital, perinatal, infancy, preschool, middle childhood and late childhood. Children with diverse focal pathologies and diagnoses were included across all study groups, such as stroke, penetrating head injury and contusions, tumours, cysts and abscesses.<sup>35</sup> The study utilised Anderson's<sup>32</sup> conceptual model of EF to assess these skills in children across four components—attentional control, cognitive flexibility, goal setting and processing speed. Subtests from the Delis-Kaplan Executive Function System (D-KEFS), TEA-Ch and the Rey Complex Figure were used to assess the four executive domains. Behavioural manifestations of EF in everyday life were also examined using BRIEF.

Results showed that compared with normative expectations, children who sustained a brain injury before the age of 3 years experienced the most severe and global EF deficits across all domains.<sup>35</sup> Regardless of the location (ie, frontal versus non-frontal regions), the presence of brain pathology was found to lead to executive dysfunction. These findings lend further support for the early vulnerability hypothesis of brain insult sustained early in development, as children with earlier lesions were most at risk for global EF impairments. The study findings also support the notion that injury to any part of the brain may disrupt neural circuits involved in EF and that there appears to be a lack of functional specificity in the immature brain.<sup>35</sup> Although children with unilateral CP, by definition, have sustained damage to the developing fetal or infant brain, there is a paucity of research specifically on EF and unilateral CP.

### Aims and hypotheses

The broad aim of this prospective cross-sectional study of children with unilateral CP (21 right-sided and 21 left-sided unilateral CP) is to examine their performance on the four domains of EF and to compare this with a group of typically developing age-matched and gender-matched children. The primary aim of the current study is to determine the pattern of EF in children and adolescents with unilateral CP with the following hypotheses and research questions:

1. It is hypothesised that children with unilateral CP will demonstrate poorer performance on tasks assessing the following EF components:
  - A. Attentional control;
  - B. Cognitive flexibility;
  - C. Goal setting;

- D. Information processing;
  - E. In everyday life.
2. It is hypothesised that children with higher levels of EF (ie, better executive skills) would show fewer difficulties across the following domains:
    - A. Behavioural manifestations of executive dysfunction in everyday life, as measured by the BRIEF;<sup>46</sup>
    - B. Emotional functioning, as measured by the Strengths and Difficulties Questionnaire (SDQ);<sup>47 48</sup>
    - C. Behavioural functioning, as measured by the SDQ;<sup>47 48</sup>
    - D. Social functioning, as measured by the SDQ.<sup>47 48</sup>
  3. Finally, the profile EF across the EF components (ie, attentional control, cognitive flexibility, goal setting, information processing and in everyday life) will be explored for children with left unilateral CP versus right unilateral CP in order to ascertain the potential laterality effects of EF following a congenital brain injury.

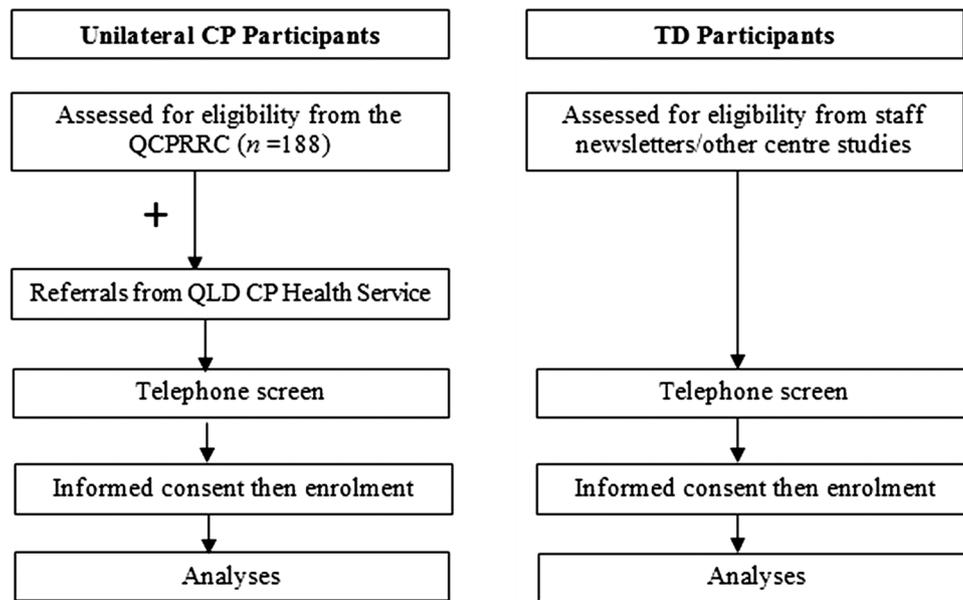
## METHODS AND ANALYSES

### Ethics

Ethics approvals have been gained through the University of Queensland School of Psychology Ethics Committee (10-PSYCH-DCP-32-JM) and the Queensland Children's Health Services Human Research Ethics Committee (HREC/10/QRCH/31). There is no known safety risks associated with any aspect of the study. All parents or legal guardians will give written informed consent and children aged  $\geq 12$  will provide assent, and will be able to withdraw from the study at any time without penalty or any effect on the child's care. Data collected in this study will be stored in a coded reidentifiable form with ID number. Each child will have one appointment during which all assessment measures will be completed. If desired by parents, all children will receive a brief neuropsychological report outlining their results on EF measures and general strategies to assist any identified cognitive weaknesses.

### Recruitment

Children will be identified from the research database of the Queensland Cerebral Palsy & Rehabilitation Research Centre and from the Queensland Cerebral Palsy Health Service at the Royal Children's Hospital, Brisbane, Australia. The treating clinician will then ask the parents of these children, identified as potentially suitable participants, if they would like further information about the study. After expressing interest and providing consent to be contacted, informed consent will proceed with the researchers. Participants will be assessed for eligibility using a brief parent telephone-screening interview based on the study criteria (see below). A provisional psychologist will conduct all telephone-screening interviews. If the participant meets the study-selection criteria, they will be invited to take part in the study and will be emailed/posted a study



**Figure 2** Study flow chart.

information sheet/consent form. An appointment will then be given enabling them to take part in the study.

Typically, developing children (age-matched and gender-matched) will be recruited as a reference sample. Siblings and friends of children with unilateral CP will be invited to take part in the study, as well as recruitment through staff newsletters and from other studies within the centre. A provisional psychologist will again conduct a brief telephone-screen interview again to ensure that they meet the study selection criteria (see below) (figure 2).

### Selection criteria

#### Inclusion criteria

Children will be invited to participate in the study if they have a confirmed unilateral CP diagnosis that was diagnosed within 28 days postnatally, are aged 8–16 years at study entry, have English as their first language, are able to communicate through a spoken language and live within Queensland.

#### Exclusion criteria

Children will be excluded from the study if they have an uncontrolled seizure disorder or if CP was acquired postnatally.

#### Typically developing reference sample

Children are eligible to participate in the reference sample if they are aged between 8 and 16 years, have English as their first language and do not have a history of developmental, neurological, physical or psychiatric conditions.

#### Sample size

A power analysis was conducted using *g* power and it revealed that at least 21 children per group needed to be recruited in order to have sufficient power (0.80) to

detect a large effect size (0.80) utilising an analysis of variance with three comparison groups.<sup>49</sup> Large effect sizes have been found in previous research comparing the performance of children with CP on tests of attention and EF, such as the TEA-Ch.<sup>39</sup>

### Classification measures

#### Family background questionnaire

Parents will complete an adapted version of the family background questionnaire that gathers basic demographic and background information pertaining to both the parent and the child.<sup>50</sup> This includes the presence or absence of seizures and, if present, whether they are controlled by medication.

#### Gross motor function classification system (GMFCS)

This measure will enable the classification of the unilateral CP participants' gross motor functioning (eg, the ability to sit, stand, walk and climb stairs) over a five-level classification system.<sup>51</sup> Research has found strong construct validity between the gross motor function classification system (GMFCS) and the Gross Motor Function Measure ( $r=0.91$ ), a criterion-referenced measure that evaluates change in the gross motor function in children with CP.<sup>52</sup> High test–retest reliability ( $r=0.79$ ),<sup>53</sup> inter-rater reliability between professionals ( $\kappa=0.74$ )<sup>54</sup> and an intrarater reliability between professionals and parents ( $r=0.94$ )<sup>55</sup> have also been documented.

#### Manual ability classification system

This measure will be used to classify the manual ability of children with unilateral CP to use their hands when handling objects in daily activities over a five-level classification system.<sup>56</sup> Research has shown good construct validity between the manual ability classification system and the GMFCS ( $r=0.79$ ), as well as a high inter-rater

reliability between therapists ( $r=0.97$ ) and an intra-rater reliability between parents and therapists ( $r=0.96$ ).<sup>56</sup>

**Strengths and difficulties questionnaire-extended version**

Parents will complete the SDQ, a 33-item questionnaire measuring parents' perceptions of prosocial and difficult behaviours in their child.<sup>47 48</sup> The SDQ is able to discriminate well between community and clinic samples and has good construct validity in associations with the Achenbach Child Behaviour Checklist (CBCL;  $r=0.87$  and  $r=0.81$ ).<sup>57 58</sup> The SDQ total difficulties score has a high internal consistency ( $\alpha=0.73$ ) and a high test-retest reliability ( $r=0.85$ ).<sup>59</sup> The SDQ total scale scores (ie, Emotional Symptoms, Conduct Problems, Inattention/Hyperactivity, Peer Problems and Prosocial Behaviour) and the overall total difficulties score will be used as outcome measures for children's emotional, behavioural and social functioning.

**BRIEF-parent form and BRIEF-teacher form**

Parents and schoolteachers will complete the BRIEF—an 86 item behavioural measure of EF in the child's everyday life.<sup>46</sup> BRIEF yields two index scores: the behavioural regulation index (BRI; including initiate, working memory, plan/organise, organisation of materials and monitor) and the metacognition index (MCI; including inhibit, shift and emotional control). The BRI and MCI combined form a global executive composite (GEC) score. Both the indexes and the composite score can be converted into T scores with higher T scores, indicating a greater level of executive dysfunction, and a T score of 65 and above, indicative of an abnormal elevation.<sup>46</sup>

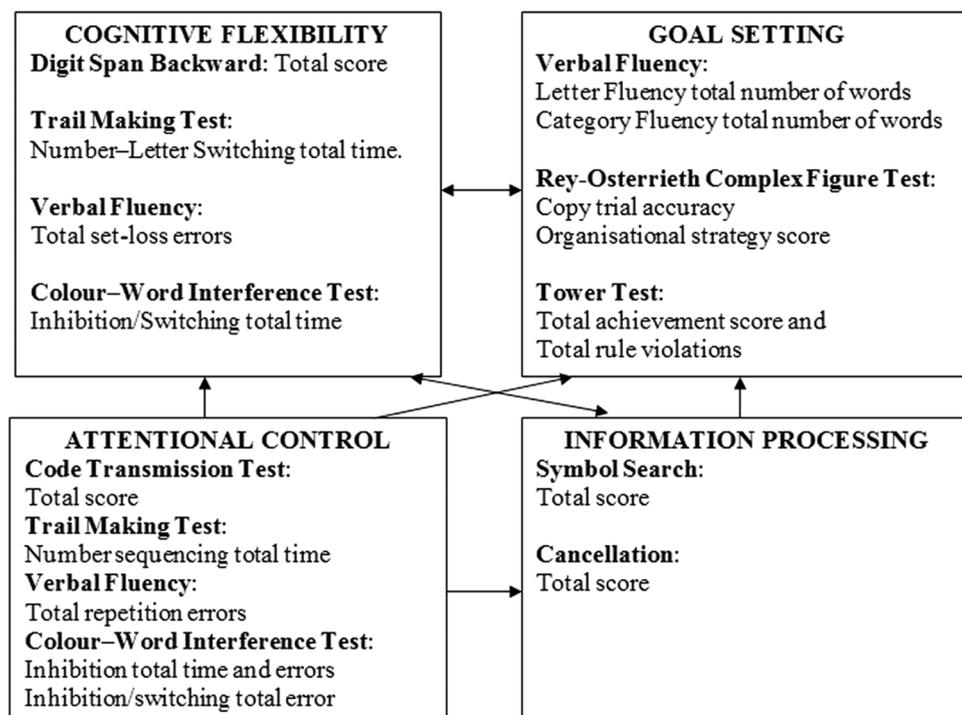
BRIEF has good convergent and divergent validities with the CBCL and the Behaviour Assessment System for Children.<sup>60</sup> High internal consistency, with Cronbach's  $\alpha$  coefficients ranging from 0.80 to 0.98 for both the parent and teacher forms, has also been shown.<sup>46 61</sup> Moderate intrarater reliability between parents and teachers has been found ( $r=0.32$ ), as have high test-retest reliability statistics for the parent form on the BRI ( $r=0.84$ ), MCI ( $r=0.88$ ), and the GEC ( $r=0.86$ ), and for the teacher form on the BRI ( $r=0.92$ ), MCI ( $r=0.90$ ) and the GEC ( $r=0.91$ ).<sup>46 61</sup>

**Outcome measures of EF**

Anderson's<sup>32</sup> conceptual model of EF will be used to operationalise EF. Ten neuropsychological measures were selected to evaluate the four components (ie, attentional control, cognitive flexibility, goal setting and information processing) of this model. The model of EF and list of the neuropsychological measures are reported in figure 3.

**Digit span backward from the Wechsler Intelligence Scale for Children-Fourth Edition**

Digit Span Backwards (range 0–16) is a verbal working memory task that requires children to temporarily store and manipulate information.<sup>62</sup> The child has to repeat a number string that increases from 2 to 8 digits in the reverse order. Higher scores indicate a greater level of the cognitive flexibility. Good internal consistency has been documented for Digit Span Backward ( $\alpha=0.80$ ) and it has high test-retest reliability ( $r=0.74$ ).<sup>63</sup>



**Figure 3** Model of executive function with corresponding neuropsychological assessments.

### Trail making test from the D-KEFS

The Number Sequencing subtest and the NumberLetter Switching subtest from the Trail Making Test will be used as measures of attentional control and cognitive flexibility, respectively.<sup>64</sup> These pencil and paper tasks require children to connect numbers in numerical order from 1 to 16 (Number Sequencing) or to switch back and forth between connecting numbers in numerical order and letters in alphabetical order (NumberLetter Switching). The outcome is the time taken to complete the test. Higher scores indicated greater difficulty with attentional control (for Number Sequencing) or cognitive flexibility (for NumberLetter Switching). High test–retest reliability for Number Sequencing ( $r=0.77$ ) and moderate test–retest reliability for NumberLetter Switching ( $r=0.20-0.55$ ) were reported.<sup>65</sup>

### Verbal fluency from the D-KEFS

Letter Fluency and Category Fluency subtests from Verbal Fluency will be used as measures of attentional control, cognitive flexibility and goal setting. In Letter Fluency, children are told that they have 60 s to name as many words as they can think of that begin with a specified letter (F, then A, then S), following specified rules (eg, not using names of people). In Category Fluency, children are informed that they again have 60 s but that this time they have to name as many different animals and then boy's names as they can think of.<sup>64</sup>

The total number of words generated for Letter Fluency and Category Fluency will be used as an outcome measure for goal setting; the total number of repetition errors across both Letter Fluency and Category Fluency will be used as a measure of attentional control; and the total number of set-loss errors (ie, saying a word that does not belong in the specific category) across Letter Fluency and Category Fluency will be used as a measure of cognitive flexibility. Higher scores for the total number of words generated and fewer numbers of repetition and set-loss errors indicate greater levels of goal setting, attentional control and cognitive flexibility, respectively. Moderate-to-high levels of internal consistency for Letter Fluency and Category Fluency in children and adolescents is documented ( $\alpha=0.53-0.80$ ).<sup>65</sup> The test–retest reliability for people aged 8–19 years is high for Letter Fluency ( $r=0.67$ ) and Category Fluency ( $r=0.70$ ).<sup>65</sup>

### Colour-Word Interference test from the D-KEFS

Inhibition and Inhibition/Switching subtests from the Colour-Word Interference Test will be used as measures of attentional control and cognitive flexibility.<sup>64</sup> For Inhibition, children have to name the ink colour that colour the words (ie, 'red', 'green') are printed in. The total time taken in seconds to complete the task and the total number of errors will be used as outcome measures for cognitive flexibility, with higher scores indicating a greater difficulty with cognitive flexibility. For Inhibition/Switching, children have to switch between reading the

word and saying the colour of the ink in which the colour word is printed. The total time in seconds to complete the task will be used as a measure of cognitive flexibility while the total number of errors will be used as a measure of attentional control. For people aged 8–19 years, an excellent level of test–retest reliability has been shown ( $r=0.90$ ).<sup>65</sup> Divergent validity between Inhibition and a measure of verbal memory, the California Verbal Learning Test–Second Edition (CVLT-II;  $r=0.90$ ) has been documented ( $r=0.27$ ).<sup>65</sup>

### Tower test from the D-KEFS

Tower Test will be used as a measure of goal setting. Across nine items, children move five disks across three pegs to build a target tower shown in a picture, within a specified time limit following specified rules (eg, use the fewest number of moves possible).<sup>64</sup> The total achievement score, which is based on the number of moves needed to make the tower, and the total number of rule violations will be used as outcome measures of goal setting. A higher total achievement score and a lower number of rule violations score indicates a higher goal setting ability. Moderate-to-high levels of internal consistency has been found for the Tower Test for people aged 8–19 years ( $\alpha=0.43-0.84$ ).<sup>65</sup> Adequate test–retest reliability has also been shown for people aged 8–19 years ( $r=0.51$ ).<sup>65</sup> Evidence for divergent validity has been demonstrated by a low correlation ( $r=0.19$ ) between the Tower Test total achievement score and the CVLT-II.<sup>65</sup>

### Rey-Osterrieth complex figure test

The Rey Figure will be used as a measure of goal setting. Children are instructed to copy a complex geometric figure.<sup>66-68</sup> The examiner records the order that the child drew the figure, which will allow for the child's strategic decision-making and organisation to be rated on a scale from 1 (*unrecognisable or substitution*) to 7 (*excellent organisation*), as per Anderson *et al.*<sup>69</sup> Osterrieth's<sup>67</sup> accuracy scoring procedure (score range: 0–36, with higher scores indicating greater spatial organisation;  $M=32$ ,  $SD=4.2$ ) and the organisational strategy score will be used as measures of goal setting. Higher scores on both measures indicate a greater goal setting ability.

The Rey Figure accuracy score has good convergent and divergent validities with significant correlations with related tests such as the Hooper Visual Organization and no significant correlations with language measures such as the Benton Sentence Repetition Test.<sup>70</sup> A moderate level of convergent validity between the organisational strategy score and other measures of EF has also been documented.<sup>69</sup> A high test–retest reliability has been shown for the accuracy scores on the immediate recall trial ( $r=0.76$ ) and the delayed recall trial ( $r=0.89$ ), as well as for the organisational strategy score ( $r=0.79-0.94$ ).<sup>69 70</sup> Using Osterrieth's<sup>67</sup> scoring procedure, an excellent level of inter-rater reliability for the copy trial ( $r=0.96$ ) has been documented.<sup>71</sup> Similarly,

the organisational strategy score has shown a high level of inter-rater reliability ( $r=0.85-0.92$ ).<sup>69</sup>

#### Code Transmission Test from the TEA-Ch<sup>72</sup>

The Code Transmission Test will be used as a measure of attentional control. This auditory sustained attention task requires children to listen to a tape recording that recites 360 consecutive numbers (40 targets) that are heard at regular intervals. The child had to identify when they hear two number fives in a row (eg, '5-5') and then say out loud the number that came before the two number fives. The total number of correctly identified targets will be used as the outcome measure, with a higher number indicating great attentional control (range=0-40). A high level of test-retest reliability has been documented for the Code Transmission Test ( $r=0.78$ ).<sup>72</sup> Overall, the TEA-Ch has been shown to be a valid assessment instrument, based on its factor structure, correlation with other measures and utility in clinical populations.<sup>72</sup>

#### Symbol search from the WISC-IV (Wechsler, 2004)

Symbol Search will be used as a measure of information processing. Children are required to visually scan and search a group of symbols and indicate, by placing a line through the word 'yes' or 'no', whether or not a target symbol is in the search group. Children are instructed to work as quickly as they can and are given a 2 min time limit. The total score is generated by subtracting the total number of incorrectly identified symbols from the total number of correctly identified symbols. A higher score (range=0-60) indicates a greater level of information processing. Raw scores can also be converted into scaled scores ( $M=10$ ,  $SD=3$ ). Good internal consistency has been shown for Symbol Search ( $\alpha=0.79$ ), as has a high level of test-retest reliability ( $r=0.80$ ).<sup>63</sup>

#### Cancellation from the WISC-IV<sup>62</sup>

Cancellation will also be used as a measure of information processing. In this task, children have to visually scan both a random and structured arrangement of pictures and mark the animals. They are instructed to work as quickly as possible and are given 45 s for each of the picture arrangement. The total score will be calculated by subtracting the number of incorrectly identified pictures from the number of correctly identified pictures, with higher scores indicating a higher level of information processing (range=0-136). Good internal consistency has been demonstrated for both Cancellation random ( $\alpha=0.70$ ) and Cancellation structured ( $\alpha=0.75$ ).<sup>63</sup> Similarly, a high level of test-retest reliability has been shown for Cancellation random ( $r=0.72$ ) and Cancellation structured ( $r=0.76$ ).<sup>63</sup> The WISC-IV's overall validity has been demonstrated, based on the test's content, response processes, internal structure and relationships to other variables.<sup>63</sup>

#### Statistical considerations

To test study the hypotheses and research questions 1 (a)-(e) and 3, a series of one-way analyses of covariance

will be conducted for each of the neuropsychological assessment measures, controlling for age and presence/absence of seizure disorders. If significant between-groups differences are found, each will be followed up using two a priori linear contrasts: the first comparing the control group with all the unilateral CP participants and the second comparing the left are right unilateral CP participants. Standardising all measures, reversing selected items so that higher scores equalled better performance, and then aggregating all measures will create an overall composite of the cognitive EF measures. A series of multiple regressions will be used to test hypotheses 2 (a)-(d).

## CONCLUSION

This study protocol highlights a prospective cross-sectional study of children with unilateral CP purposely sampled for age and gender from an equal group of children with right-sided and left-sided brain lesion to examine their EFs and compare them with a group of typically developing children. To our knowledge, this protocol outlines the first study to examine multiple components of EF among a cohort of children solely with unilateral CP and explores the possible laterality effects of EF among children with a congenital brain injury. In addition, this study examines the relationship between cognitive EF measures, behavioural manifestations of EF in everyday life and psychological functioning. The results of this study are scheduled to be published in peer reviewed publications and will be presented at national and international conferences.

**Contributors** HLB was the chief investigator and, together with KW, OL and RNB, designed and established this research study as part of her clinical doctorate study. HLB, KW and RNB were responsible for ethics applications and reporting. All authors were responsible for the selection of measures. HLB was responsible for recruitment and data collection and HLB and KW were responsible for data analysis. All authors have read and approved the final manuscript.

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**Data sharing statement** Further details of the study protocol can be requested from the corresponding author.

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