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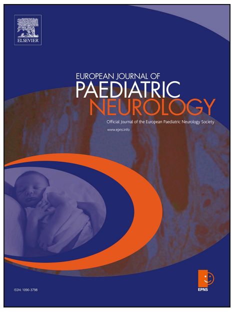

Executive function and general intellectual functioning in dyskinetic cerebral palsy: comparison with spastic cerebral palsy and typically developing controls

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Aim

To comprehensively describe intellectual and executive functioning (EF) in people with dyskinetic cerebral palsy (DCP), by comparing their performance with that of: 1) age- and sex-matched typically developing controls (TDC); and 2) participants with spastic cerebral palsy (SCP) matched for age, sex, term/preterm and gross motor function classification system (GMFCS).

Method

This cross-sectional study was conducted by the University of Barcelona in collaboration with five institutions. Participants were people with DCP (n=52; 24 females, median age 20.5y: 5mo, interquartile range [IQR]= 13.75y: 7mo; GMFCS I-V). As comparison groups, participants with SCP (n=20; 10 females, median age= 20.5y: 5.5mo, IQR= 13.75y 9mo; GMFCS I-V) and TDC (n=52; 24 females, median age= 20y: 4mo, IQR= 12y 7mo) were included. Intelligence and EF were assessed using common tests in all participants.

Results

Both CP groups had lower intelligence than TDC and performed poorer in almost all EF tasks. Intelligence was higher in DCP than SCP (z=-2.51, p=.01). Participants with DCP also performed significantly better in goal-setting tasks (z=2.27, p=.03) and information processing (z=-2.54, p=.01) than those with SCP.

Conclusion

People with DCP present lower general intellectual functioning and poorer EF across multiple domains than typically developing controls. People with DCP have higher general intellectual functioning and better EF than people with SCP when levels of motor severity are similar.

KEYWORDS

Dyskinetic cerebral palsy

Spastic cerebral palsy

Executive function

Intellectual functioning

Goal setting

Executive function and general intellectual functioning in dyskinetic cerebral palsy: comparison with spastic cerebral palsy and typically developing controls

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What this paper adds

This is the first study to compare general intellectual functioning and executive function in a relatively large sample of people with dyskinetic cerebral palsy (DCP) against samples of typically developing controls (TDC) and people with spastic cerebral palsy (SCP) who are similar in terms of age, sex, being term/preterm and gross motor severity. The present study contributes, therefore, to the characterization of cognitive impairments in DCP. Participants with DCP present poorer general intellectual functioning and executive function than TDC in terms of attentional control, cognitive flexibility, goal setting and information processing. People with DCP display stronger general intellectual functioning and executive function than those with SCP. The results suggest that cognitive functions may have been underestimated and masked by gross motor severity in people with DCP. Observations made in SCP cannot be generalized to dyskinetic forms. It is important to properly assess general and specific cognitive functions in people with cerebral palsy (CP) even in the most severe cases.

1. INTRODUCTION

Although cerebral palsy (CP) is primarily a disorder of movement and posture, it often involves disturbances of communication and cognition [1] which may have an impact on quality of life [2]. Dyskinetic cerebral palsy (DCP) is the second largest CP group, with high rates among children born at term and among children with normal birth weight. It is agreed that DCP presents poorer gross motor function than other CP types [3] but there is less scientific evidence of the presence of poorer cognitive abilities in comparison with other CP types, especially when levels of gross motor severity are equivalent between groups [3–10]. Dyskinetic cerebral palsy is characterized by involuntary, uncontrolled, recurring and occasionally stereotyped movements. Primitive reflex patterns predominate, muscle tone is varying, and dyskinesia is differentiated into dystonia and choreoathetosis [11,12].

Some studies have reported that people with DCP present higher rates of cognitive difficulties. Specifically, a study including 474 participants with DCP (16% walked without aids, 24% with aids and 59% were confined to a wheelchair) and 4746 participants with bilateral spastic cerebral palsy (SCP) (36% walked unaided, 42% unable to walk) found learning disability to be more frequent in children with DCP (52%) than in children with bilateral SCP (33%) [3]. Similarly, another study reported that people with DCP have a lower intelligence quotient (IQ) than people with SCP [6]. Verbal IQ was also found to be significantly higher among children with spastic diplegia (n=49) and hemiplegia (n=35), and lower in those with DCP (n=11) [7]. Neither of those studies reported the GMFCS level separately for participants with DCP and other CP types. Only Himmelmann et al.[3] reported gross motor severity by CP type. While 59% of participants with DCP needed wheelchair, only 2% of participants with SCP needed it. Interestingly, Pueyo et al.[4] did not find differences between bilateral DCP, mixed CP, and SCP in nonverbal reasoning using the same measure in all participants. Strauss et al.[5] found that in a sample of people with tetraplegic CP with severe motor dysfunction (defined as tetraplegia with no functional hand use and inability to crawl, creep, scoot, or walk), 95% of those with SCP had an IQ<50 and only 2% had no intellectual disability. Among the participants with DCP, only 40% (n=8) had an IQ<50, and 20% (n=4) had an IQ≥70 or higher. However, the authors reported in the study that "the reliability of the cognitive assessments [was] untested". Indeed, another study [8] found that the percentage of learning disability was higher (100%) in people with spastic tetraplegia (GMFCS IV n=2, V n=21) than in DCP (60%; GMFCS I n=2, II n=4, III n=5, IV n=15, V n=26). Moreover, a recent study suggested that "cognitive quotient" in tetraplegic SCP may be significantly lower than other CP types, including DCP [9]. Finally, Dalvand et al. found that the lowest IQ level was more frequent in spastic tetraplegia than in DCP. Specifically, in this study hypotonic, spastic tetraplegic, and hemiplegic but not dyskinetic participants had the highest odds to assign higher ratings in impaired IQ [10]. Overall, these studies might suggest that when people with DCP are comparable to other CP types in terms of gross motor severity, they might not present poorer cognitive performance. Some aspects of these studies, however, preclude drawing firm conclusions. For example, some studies have small samples of participants with DCP, gross motor severity by CP type is not always reported and groups are not comparable in terms of gestational age and gross motor severity in most studies. Moreover, the different CP classifications and the different terms used between some studies make it difficult to compare results. Table 1 shows further details about the above-mentioned studies.

Interestingly, a systematic review focused on cognition in childhood dystonia has been recently published [9]. Although this work is not specifically focused on secondary dystonia due to CP, it indicates that people with DCP often have mild deficits in memory, impairments of visuospatial functions, information processing speed, and social cognition. This review stresses that available data is very limited and that there is a strong need for case-control studies assessing cognition and using standardized neuropsychological tests, with particular emphasis on attention and executive functioning skills among others. In this review it is further encouraged to assess control groups consisting of patients with other movement disorders [13].

Executive function (EF) is necessary for the successful completion of everyday, novel, goal-directed activities and has been associated with quality of life in DCP [14]. Most studies of EF in CP focus on SCP or unilateral CP, and have shown that people with CP perform significantly worse than typically developing children [15] in all EF domains described by Anderson [16]. Briefly, these domains include attentional control (capacity to selectively attend to specific stimuli, to focus attention for a prolonged period, and impulse control), cognitive flexibility (ability to shift between response sets, learn from mistakes, devise alternative strategies, divide attention, apply working memory and process multiple sources of information simultaneously), goal setting (ability to initiate an activity and devise a plan to complete it) and information processing (fluency, efficiency and speed of output) [16]. Although it has been proposed that DCP may be characterized by an executive dysfunction [4], this hypothesis has still to be tested. The hypothesis is consistent with the lesions of the basal ganglia and thalamic systems which may impair focused attention and executive function [17] and are frequently described in people with DCP [18]. Executive functions depend on the integrity of the entire brain but they are mainly mediated by the frontal lobes and their connections with posterior and subcortical brain regions [15,16]. Specifically, some circuits in the basal ganglia originate in the prefrontal and limbic regions of the cortex which are known to be involved in the executive function [15]. If we focus on the differences between CP types, one study [4] found that EF was the only function in which mean performance was poorer in DCP participants than in participants with SCP; however, the differences were not statistically significant, the sample was small, not all domains of EF were assessed, and other variables that may influence EF such as gestational age and gross motor severity were not controlled. When aiming to identify the association between spasticity/dyskinesia and cognition, influential variables other than CP type, such as gross motor severity and prematurity, should be taken into account, since they may prompt additional cognitive impairments [19].

The International Classification of Functioning, Disability and Health considers intellectual functions to be an essential Core Set for people with CP [20], and intellectual functions play an important role in communication [21], employment [22] and quality of life [14]. Nevertheless, few studies so far have focused on cognitive functions in DCP or have analysed the EF profile in depth, and the results they present are partly conflicting. Most of them have used different measures for assessing cognition in the participants, and the results vary depending on whether the participants with DCP are compared with participants with severe or moderate forms of SCP. No study to date has compared cognitive functioning between a relatively large sample of people with DCP and a group of participants with SCP who are similar in terms of gross motor severity and prematurity. To better understand the cognitive correlates of different CP types, neuropsychological studies able to differentiate between subtypes of the condition are required [23]. The results would help to guide the design of more appropriate interventions and follow-up programs focused on DCP.

Thus, in the present study we aim to map general intellectual functioning and EF in people with DCP by comparing their performance with: 1) typically developing controls (TDC) matched for age and sex, and 2) participants with SCP matched for age, sex, gestational age and gross motor severity. Following on from previous studies, our primary hypothesis was that general intellectual functioning and EF would be poorer in participants with CP compared with TDC. Our second hypothesis was that, with similar gross motor severity in the two CP groups, general intellectual functioning would be higher in participants with DCP than in those with SCP. This hypothesis is in accordance with the studies mentioned above which seem to preliminarily indicate that when people with DCP are comparable to other CP types in terms of gross motor severity, their cognitive performance tends to be as good as that of the other CP types or better. Furthermore, taking into account the brain lesions described in DCP and the results of the only previous study to analyse EF differences between CP types [4], we expected performance on EF to be poorer in participants with DCP.

2. METHOD

2.1. Participants

This study recruited participants with DCP and a comparison group with SCP from the main hospitals in Barcelona that monitor people with CP (the Pediatric Neurology Department and Rehabilitation and Physical Medicine Department at the Hospital Vall d'Hebron and the Neurology Department at Hospital Sant Joan de Déu), other institutions attending people with CP (Health Services and Rehabilitation Services of the ASPACE Cerebral Palsy Association, Centro

Ocupacional Sinia, and Nadís). Some of them were participants in a previous study [4] who were contacted and invited to participate in the current study. Cases were also compared with a group of TDC composed of relatives/friends of the participants with CP and people recruited through advertisements.

The inclusion criteria for participants with clinical diagnosis of CP with predominant dyskinetic features were (1) age older than 6 years and (2) ability to understand instructions, as evaluated by the Spanish Grammar Screening Test (receptive part) [24]. Exclusion criteria were (1) presence of severe visual or auditory disability and (2) lack of an intelligible yes/no response. In case of signs of sensory impairment, only participants in whom the deficit was corrected or/and the sensory impairment did not prevent evaluation were included.

After the recruitment of the participants with DCP, participants who had a clinical diagnosis of SCP and met the inclusion and exclusion criteria described above were recruited and matched by sex (male/female) and age with participants with DCP (Supplemental material 1). Since the influence of age on cognitive performance is stronger in younger people, the age matching criterion was more flexible with older participants: <20 years old +/-2 years; 20<30 years old +/-4 years; ≥30 years old +/-8 years. In order to control for variables other than the CP type, additional inclusion criteria were implemented to ensure that each participant of the SCP group matched a participant with DCP with regards to term vs preterm (≥37 weeks, <37 weeks) and as ambulant vs non-ambulant (GMFCS I-II-III vs IV-V). That is, the prematurity and ambulation level of participants with SCP included was driven by the characteristics of participants with DCP.

Physicians from the institutions mentioned above informed their patients with DCP or SCP who met the inclusion criteria, or their careers, about the possibility of participating in this project. The diagnosis of CP type was based on the neurologist's clinical assessment. All physicians agreed to define DCP as the CP type characterized by abnormal patterns of posture and/or movement accompanied by involuntary, uncontrolled, recurring, and occasionally stereotyped movements. Impaired muscle tone regulation, movement control, and coordination may comprise dystonic and choreoathetotic patterns [11]. Only participants who clearly presented predominant dyskinetic symptoms were included in the DCP group. Participants who presented spastic and dyskinetic symptoms to the same extent were not considered eligible. Participants were further contacted by phone to double-check inclusion/exclusion criteria, to explain the participation procedure, and to take part in the study; the recruitment and data collection period was from 2012 to 2016.

Typically developing people without brain pathology were matched one-to-one by age and sex with participants with DCP. Controls were ineligible if they had been born preterm, were diagnosed with a neurological or psychiatric disorder, or were illicit substance consumers.

All procedures performed in the study complied with the ethical standards of the 1964 Helsinki declaration. Ethical approval was obtained by the University of Barcelona's (CBUB) Institutional Ethics Committee, Institutional Review Board (IRB 00003099, assurance number: FWA00004225; http://www.ub.edu/recerca/comissiobioetica.htm) and the Hospital Universitari Vall d'Hebron. Written informed consent was obtained from all participants or their careers.

2.2. Neuropsychological assessment

Tests were carefully chosen to allow most of the participants to answer in an autonomous way and to permit, when possible, the use of assistive technology for communication. Participants were encouraged to use the response technique best suited to their degree of disability and the communication devices they normally used. See Supplemental material 2 for test details and adaptations used.

The Raven's Coloured Progressive Matrices (RCPM) test was used to measure general intellectual functioning and the four EF [16] domains were assessed as follows:

Attentional control

-Inhibition and sustained attention components were assessed using an adapted version of the Stop Signal Task (SST) of the Cambridge Neuropsychological Test Automated Battery (CANTAB) [25].

-Selective verbal attention was assessed using the digit forward span from either the Wechsler intelligence scale for children (WISC-IV) [26] or the Wechsler adult intelligence scale (WAIS-III) [27].

-Selective visual attention was assessed using the spatial forward span from the Wechsler nonverbal scale of ability (WNV) [28].

Cognitive flexibility

-Feedback utilization was assessed using the 64-item computerized version of the Wisconsin Card Sorting Test (WCST) [29].

-Verbal working memory was assessed using the digit backward span from either WISC-IV [26] or WAIS-III [27].

-Visual working memory was assessed using the backward spatial span from the WNV [28].

Goal setting ability was evaluated by means of the Stockings of Cambridge (SOC) test from the CANTAB [25]. Three scores were used: problems solved in minimum moves, mean moves in items of two moves and mean moves in items of five moves.

Information processing was assessed only in verbal participants, using a lexical verbal fluency task.

2.3. Background measures

Gross motor function was determined based on the Gross Motor Function Classification System (GMFCS) [30] while manual ability was measured by the Manual Ability Classification System (MACS) [31]. The CFCS (Communication Function Classification System) was used to categorize communication [32]. Gestational age and epilepsy status were recorded from parent interviews and medical records.

2.4. Statistical analysis

Statistical analyses were performed using R version 3.3.1 (https://www.r-project.org/). Descriptive statistics were calculated, and a global test based on Kruskal-Wallis and Chi-squared test was used to check differences between groups in matching and descriptive variables. Medians and interquartile ranges (IQR) were used due to the presence of skewness in the empirical distribution of some numerical variables.

Matched pairs were not included in the analyses as blocks because only one measure was recorded for each pair and, therefore, the block effect could not be separated from error. As a result, global tests based on Kruskal-Wallis (H statistic) were performed to analyse differences in cognitive performance between groups. Pairwise contrasts, based on the Mann-Whitney U test, were performed in cases in which the global test yielded a significant result. The Hodges-Lehmann estimator, as well as a 95% bootstrap-percentile confidence interval, was estimated in order to report the differences between groups. The p values were corrected for multiple comparisons (for global and posteriori contrasts) and we used an alpha level of .05 for all statistical tests. As for the reported effect size, probability of superiority was used. Given its relationship with Cohen's d under certain conditions, probabilities greater than .56, .64, and .71 (or lower than .44, .36, and .29, depending on the order in which groups are compared) could be regarded respectively as small, medium, and large effects. Further details about the statistical analysis are available in Supplemental material 3.

3. RESULTS

3.1. Participants

The final sample comprised 52 participants with DCP with age range 6-62 and median age 20.5y: 5mo, IQR= 13.75y: 7mo without visual/auditory abnormalities precluding neuropsychological assessment, able to understand instructions and, at least, able to answer yes/no. Five of these 52 participants had sensorineural hearing loss (two mild, one moderate and two severe hearing loss –

the last three using hearing aids), 17 subjects with corrected refractive errors (myopia, astigmatism and hypermetropia), four subjects with strabismus (two corrected and two non-corrected), two who showed a slight delay in signal transmission in visual evoked potentials but without pathological values and one with decreased visual acuity in one eye. The remaining cases did not report any hearing or visual problem. A comparison group of 52 age- and sex-matched typically developing controls with an age range 7-59 and age median age 20y: 4mo (IQR= 12y: 7mo) were recruited. The final sample for the group with SCP comprised 20 participants (10 females), age range 7-65 and median age= 20.5y: 5.5mo (IQR= 13.75y: 9mo). The recruitment process is described in Figure 1. Due to absence or unintelligible speech and motor severity or comprehension difficulties, some participants were unable to complete some of the cognitive tests used. These missing values are indicated in the footnote of Table 3.

Given the difficulty of finding participants with SCP who met the matching criteria and due to the completion of the research project, a one-to-one matching was not possible. The range of age and GMFCS of participants were similar in both CP groups. For detailed information on the matching, see Supplemental material 1. There were no significant differences in age and sex between the three groups, as regards the rates of term, preterm and very preterm participants (x^2=2.67, p=.26) or GMFCS between CP groups. Demographic and clinical data for the three groups are shown in Table 2.

3.2. General intellectual functioning differences between the three groups

Both CP groups had significantly lower scores than TDC (Table 3, Figure 2), and participants with DCP had significantly higher scores than participants with SCP. The differences were large for DCP and SCP vs TDC and medium for DCP vs SCP.

3.3. Executive function differences between the three groups

Attentional control: Inhibition/sustained attention measured by the SST did not differ significantly between the three groups. Both selective verbal and visual attention were significantly poorer in DCP and SCP than TDC; there were no differences between CP groups.

Cognitive flexibility: Feedback utilization as well as verbal and visual working memory were significantly poorer in DCP and SCP than TDC. There were no significant differences between CP groups in any cognitive flexibility domain.

Goal setting: The mean number of moves in problems that can be solved in two moves did not differ significantly between the three groups. The mean number of moves in problems that can be solved in five moves was significantly lower in TDC and DCP than in SCP. There were no significant differences between DCP and TDC. Both CP groups had significantly worse global scores (problems solved in the minimum number of moves) than TDC. There were no significant differences between CP groups in problems solved in the minimum number of moves.

Information processing: Both CP groups showed significantly poorer verbal fluency than TDC. Verbal fluency was significantly higher in DCP than in SCP.

Interestingly, in the variables in which both CP groups performed worse than TDC but did not differ from each other, mean scores were always higher (though not significantly) in participants with DCP (Table 3, Figure 2).

4. DISCUSSION

The present study contributes to the characterization of cognitive impairments in DCP. To our knowledge, it is the first study to compare general intellectual functioning and performance in all EF domains in a relatively large sample of people with DCP against samples of TDC and people with SCP who are similar in terms of age, sex, gestational age and gross motor severity.

As expected, the comparison between TDC and CP groups showed that people with CP had lower general intellectual functioning and poorer executive function across all the domains described by Anderson [16]. Differences between CP and TDC in all EF domains have previously been reported in unilateral CP [15]. In the present study, performance did not differ significantly between TDC and CP groups for inhibition/sustained attention and for the easiest items of goal setting. The lack of differences between CP and TDC on the easiest items of goal setting shows that CP performance may be similar that of TDC in very basic planning tasks. The lack of differences in inhibition/sustained attention is possibly due to the fact that SST may overestimate the performance of some of the CP participants on “stop trials” (25% of the total score used), as motor slowness precludes inhibition errors.

Our results corroborate those of previous studies which have indicated that learning or intellectual disability is more common in spastic tetraplegia than in DCP [5,8,9]. Although our results seem to be at odds with those of Sigurdardottir et al. [6], when those authors took into account participants with DCP who could not be assessed by the same test as the rest of the sample (and were therefore assessed by different measures) the highest proportion of children with intellectual impairment was found in the group with spastic tetraplegia. In fact, the authors themselves concluded that cognitive skills might be masked by limitations of motor control [6]. Our results, are also at odds with those of other studies [3,4,7]. The differences with regard to two of these studies [4,7] may be due to the small size of the sample of participants with DCP. Himmelmann et al. [3], however, studied a large sample of DCP participants; the differences between our study and theirs might be due to the fact that they did not control for GMFCS and prematurity, and that general intellectual functioning was reported dichotomously using an IQ cut-off point of 50. It has been suggested that the more severe the motor impairments, the higher the percentage of cognitive impairment [3] and so DCP has often been associated with poorer cognitive outcomes. However, our results show that, at similar levels of gross motor severity, dyskinetic forms may present higher general intellectual functioning than spastic forms. Accordingly, some studies suggest that intellectual disability, rather than the degree of motor involvement, is a predictor of verbal comprehension abilities [21,33,34]. Overall, these findings seem to support the hypothesis that people with DCP, even if they often present poorer gross motor functioning than people with SCP, do not necessarily present poorer cognitive functioning. It is worthy of note that similar mismatches have previously been found between motor impairment and communication abilities in DCP [35].

Participants with DCP performed significantly better for goal setting and information processing than participants with SCP. Better performance in goal setting was observed for the most difficult items; that is, both CP groups performed similarly on relatively easy items, but people with DCP performed significantly better when more complex planning is required. In fact, their performance on the most difficult items was close to the performance of TDC, since no significant differences were found. Given that information processing was assessed with a verbal fluency task, the conclusions about the better performance observed in DCP than SCP must be limited to verbal participants. This fact may have biased information processing scoring toward a better performance in both CP groups. Information processing is inherently temporal, so clinical testing of this process is affected by a participant's ability to form a response. In the authors' opinion, comparing performance against people with similar motor difficulties as we have done by comparing DCP and SCP groups is a promising way to control this limitation. Further studies should focus on developing tasks that control this effect at the maximum level possible but, presently, this is a limitation difficult to overcome when researching performance in this kind of tasks in people with CP.

Overall, our results show that people with DCP perform worse than TDC and better than SCP in both general intellectual functioning and EF indicating a general tendency, rather than a specific dysexecutive deficit. The performance of people with DCP tends to be closer to TDC on EF than on general intellectual functioning. At this point it has to be noted that the measure used to estimate general intellectual functioning (Raven's coloured progressive matrices) is a matrix reasoning task. Although the Raven's progressive matrices is regarded as a quintessential measure of fluid intelligence [36], it aims to measure the ability to deduct relationships [37]; other aspects considered in psychometric tests of cognitive abilities such as verbal ability or crystallized intelligence are not considered. Moreover, it has been suggested that the Raven's coloured progressive matrices gives comprehensive information on cognitive performance and it is sensitive to the structural state of the brain in DCP [38]. The executive function results comparing DCP and SCP groups do not confirm DCP as a CP type specifically characterized by an executive dysfunction. Further research including neuropsychological assessment of other cognitive domains is needed to conclusively reject the hypothesis of a dysexecutive deficit in DCP.

The strengths of this study include the recruitment of a large sample of participants with DCP taking into account that this CP type is relatively rare; a wide assessment using reliable and common measures in all participants and the lack of differences in age, sex, GMFCS and prematurity between groups. The limitations include the absence of measurement of dyskinesia and speech production using a quantitative scale, the wide age range of the sample and the lack of a wider description of the functioning and disability of the participants according the International Classification of Functioning, Disability and Health. Moreover, neuro-ophthalmological disorders are among the main symptoms in CP [39] and their interaction with manual ability may have an effect on cognitive performance. To control this effect, the execution time was not considered when scoring performance, but SST and verbal fluency are influenced by execution time. In this regard, it is interesting that there were no significant differences in manual ability between DCP and SCP groups (z= 0.16, p=.88). Additionally, considering neuroimaging as a further matching variable between groups would provide meaningful information and further strength the results of our study. Finally, participants with SCP included in the present study are not representative of the entire SCP population and the sample is small. Caution is therefore required when interpreting the results referring to this group. This is due to the fact that participants with SCP were included *a posteriori*, based on prematurity, ambulation, age and sex characteristics of participants with DCP.

In conclusion, the present study identified difficulties across multiple executive function domains compared with TDC and a better cognitive functioning in people with DCP than SCP. Although performance observed in our sample cannot be generalized to the broad population of DCP (we have included only participants with enough comprehension, an intelligible yes/no response and without severe visual or auditory disabilities), the study of specific cognitive functions is only possible in subsamples of DCP such as these. The results are clinically relevant as they suggest that cognitive functions may have been underestimated and masked by gross motor severity in people with DCP. The present study highlights the importance of properly assessing general and specific cognitive functions in CP, even in the most severe cases. Our study may help to broaden the understanding of the clinical consequences of dyskinesia for cognitive function and, by extension, the interaction between cognitive function, muscle tone, and specific brain damage in early childhood. Once again, the results indicate that observations made in SCP cannot be generalized to dyskinetic forms. A comprehensive understanding of cognitive functioning in each CP type would contribute to improving the accuracy of prognosis and also to the design of educational approaches. Although this study increases our current knowledge about DCP, further studies are needed. Harmonizing nomenclature and cognitive measures between studies is also important, as this would allow researchers to compare results, perform meta-analyses and multicentre studies, as well as to pool data for the sake of achieving larger sample sizes.

Acknowledgments

ACCEPTED MANUSCRIPT

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Table 1. Studies comparing/reporting percentages of general general intellectual functioning in dyskinetic and spastic cerebral palsy

	n	Age range	Motor assessment/ Motor severity by CP type	CP type (n)	IQ assessment	Results
Stadskleiv et al., 2018 [9]	70	5y:1m-17y:7m	GMFCS / - (GMFCS range of the whole sample I-V)	DCP (8) SCP tetraplegia (9) SCP hemiplegia (35) SCP diplegia (18)	Cognitive quotient measured by one of the following options: 1) Cognitive Ability Quotient: *Verbal comprehension* Test for Reception of grammar Vocabulary score (British Picture Vocabulary Scale and/or Receptive Vocabulary) *Perceptual reasoning* WPPSI-III/WISC-IV (substituting Block Design with Picture Completion for children with severe fine motor impairments) 2) Developmental Quotient BSID-III/Receptive Vocabulary	Significantly lower cognitive quotient in the tetraplegic SCP group **DCP** 75% IQ≥85 **SCP tetraplegia** 33% IQ≥85 **SCP hemiplegia** 86% IQ≥85 **SCP diplegia** 78% IQ≥85 **Mixed CP** 76% IQ≥85
Dalvand et al., 2012 [10]	662	3y-14y	GMFCS	DCP (53) SCP tetraplegia (218) SCP hemiplegia (57) SCP diplegia (223) Mixed CP (63) Ataxic CP (18) Hypotonic (30)	WPPSI, WISC-R	**DCP** 26% IQ≥85; 9% IQ 71–84; 19% IQ 50–70; 45% IQ<50 **SCP tetraplegia** 8% IQ≥85; 5% IQ 71–84; 10% IQ 50–70; 77% IQ<50

						SCP hemiplegia 25% IQ≥85; 21% IQ 71–84; 28% IQ 50–70; 26% IQ<50 **SCP diplegia** 24% IQ≥85; 18% IQ 71–84; 22% IQ 50–70; 35% IQ<50 **Mixed CP** 33% IQ≥85; 21% IQ 71–84; 13% IQ 50–70; 33% IQ<50
Sigurdardottir et al., 2011 [7]	111	4y:6y	GMFCS / - (GMFCS range of the whole sample I-V)	DCP (11) SCP tetraplegia (10) SCP hemiplegia (35) SCP diplegia (49) Ataxic CP (6)	n=29 BSID-II n=4 Reynell Zinkin Developmental Scales n=9 Columbia Mental Maturity Scale n=4 The Leiter International Performance Scale	Verbal IQ was lower in DCP than SCP
Himmelmann et al., 2009 [3]	5220	Years of birth 1991-1996	Severe: needing a wheelchair Moderate: ambulation with aids Mild: ambulation without aids / DCP: 16% mild, 24% moderate, 59% severe SCP bilateral: 36% mild, 2% severe	DCP (474) SCP bilateral (4746)	Measure not specified Categorized into IQ above / below 50	**DCP** 52% IQ<50 **SCP bilateral** 33% IQ<50

Sigurdardottir et al., 2008 [6]	118	4y-6y: 6m	GMFCS / - (GMFCS range of the whole sample I-V)	DCP (14) SCP tetraplegia (28) SCP hemiplegia (31) SCP diplegia (45)	50% WPPSI 26% WPPSI in combination with the other developmental scales 12% BSID-II 10% BSID-II in combination with other developmental scales n=4 Reynell Zinkin Scales n=5 Columbia Mental Maturity Scale n=1 Leiter International Performance Scale-Revised in com- bination with TONI-2	Median IQ was lower in DCP than SCP **DCP** 29% IQ/DQ ≥85; 14% IQ/DQ 70 to 84; 21% IQ/DQ 50 to 69; 36% IQ/DQ <50 **SCP tetraplegia** 50% IQ/DQ ≥85; 14% IQ/DQ 70 to 84; 21% IQ/DQ 50 to 69; 14% IQ/DQ <50 **SCP hemiplegia** 61% IQ/DQ ≥85; 19% IQ/DQ 70 to 84; 13% IQ/DQ 50 to 69; 6% IQ/DQ <50 **SCP diplegia** 53% IQ/DQ ≥85; 18% IQ/DQ 70 to 84; 22% IQ/DQ 50 to 69; 7% IQ/DQ <50
Himmelmann et al., 2006 [8]	70	4y-8y	GMFCS / DCP (GMFCS I n=2, II n=4, III n=5, IV n=15, V n=26) SCP (GMFCS IV n=2, V n=21)	DCP (50) SCP tetraplegia (20)	WPPSI-R, WISC-III, Griffith scales or estimated from clinical observation	**DCP** 40% IQ≥70; 18% IQ 50-70; 42% IQ<50 **SCP tetraplegia** 100% IQ<50

Strauss et al., 2005 [5]**⁵ ⁵ ⁵**	446	-	Severe: tetraplegia with no functional hand use and inability to crawl, creep, scoot, or walk / All participants with severe motor dysfunction	DCP (20) SCP (426)	Measure not specified	**DCP** 20% IQ≥70; 40% IQ<50 (profound mental retardation)+ **SCP** 2% IQ≥70; 95% IQ<50 severe mental retardation (75% profound)
Pueyo et al., 2003 [4]	19	16y-38y	- / All bilateral	DCP tetraplegia (2) DCP diplegia (2) DCP triplegia (2) SCP tetraplegia (5) SCP diplegia (6) SCP triplegia (2)	RCPM	No statistically significant differences between DCP and SCP

-, data not provided; + The term used is the same that authors use; CP: cerebral palsy; BSID, Bayley Scales of Infant Development; DCP: dyskinetic cerebral palsy; DQ: developmental quotient; GMFCS: Gross Motor Function Classification System; ICD10: International Statistical Classification of Diseases and Related Health Problems 10th Revision; IQ: intelligence quotient; m, months, RCPM: Raven's coloured progressive matrices; SCP: spastic cerebral palsy; TONI-2: Test of Nonverbal Intelligence, 2nd Edition; WIPPSI: Wechsler Preschool and Primary Scale of Intelligence; WISC: Wechsler Intelligence Scale for Children; y, years.

Table 2. Participants' characteristics

Matching variables	DCP	SCP	TDC	Differences: Statistic (*p*-value) *
Age **Md years: months (IQR years: months)**	20.5: 5 (13.75: 7)	20.5: 5.5 (13.75: 9)	20: 4 (12: 7)	H=0.01 (p=.99)
Sex **n (female/male)**	24/28	10/10	24/28	χ^2=0.02 (p=.99)
Gestational age **n (<32 weeks/32-36weeks/≥37 weeks)**	4/6/42	4/1/15	-	χ^2=2.67 (p=.26)
Gestational age participants born preterm n / Md weeks (IQR weeks)	10 / 34 (6.5)	5 / 28 (4.5)	-	z= 1.48 (p=.12)
Gross motor function (GMFCS) (n)	I (15) II (8) III (6) IV (11) V (12)	I (5) II (3) III (4) IV (5) V (3)	-	z= 0.14 (p=.89)
Other participants' characteristics				
Motor distribution **n (bilateral/unilateral)**	44/8	16/4		
Manual ability (MACS) (n)	I (5) II (10) III (17) IV (10) V (10)	I (1) II (5) III (5) IV (4) V (4)	-	z= 0.16 (p=.88)
Communication (CFCS) (n)	I (17) II (23) III (6) IV (6)	I (6) II (10) III (2) IV (2)	-	z= 0 (p=1)
Epilepsy status[†] **n (no epilepsy/ active/resolved)**	30/16/6	11/4/5	-	χ^2 = 0.93 (p=.63)
Aetiology[a]				χ^2 = 7.47 (.11)
HIE	22	6	-	
Intra-cranial haemorrhage, infarction or hydrocephalus	9	7	-	
Infection	2	2	-	
Kernicterus	2	0	-	
Unclassifiable	17	5	-	
Main communication system[+] **n (Speech/Speech+gestures, communication board and other tools/ gaze, facial expressions and gestures/ communication board/ alphabetic communication board)**	37/5/3/5/2	17/1/2/0/0	-	

†The International League Against Epilepsy criteria were used to determine epilepsy status; *The level of significance was set at a *p*-value < .05 and *p* values were corrected for multiple comparisons (for global and posteriori contrasts); -, not applicable (note that all typically developing controls were born at term); [+]Here there are presented the main communication system used in their daily life but most of participants used other complementary systems depending on the context or nature of the content they want to communicate; [a] Aetiology classification was based on clinical criteria complemented by the information from available neuroimaging, and HIE criteria were based on Himmelmann et al. [40]; CFCS, Communication function classification system; DCP, dyskinetic cerebral palsy; GMFCS, Gross motor function classification system; HIE, hypoxic-ischemic encephalopathy; IQR, interquartile range; MACS, Manual ability classification system; SCP, spastic cerebral palsy; TDC, typically developing controls; -, not applicable; χ^2, Pearson's Chi-squared test; z, transformed score from Mann-Whitney U test.

Table 3. Descriptive statistics and comparisons of cognitive scores between participants with dyskinetic cerebral palsy, typically developing controls and spastic cerebral palsy

	n / Md (IQR)			General contrasts H (*p* value)*	Posteriori contrasts	z (adjusted *p* value)	HL estimator (95% CI)	Effect size (probability of superiority)
	DCP	TDC	SCP					
GENERAL INTELLECTUAL FUNCTIONING								
	52 / 27.5 (11)	52 / 35 (2)	20 / 24.5 (9.75)	55.49 (*p*<0.01)	TDC - DCP	5.96 (*p*<.01)	7 (3;-9)	0.79[L]
					SCP - DCP	-2.51 (*p*=.01)	-5 (-9;-1)	0.29[M]
					SCP - TDC	-6.22 (*p*<.01)	-11 (-15;-9)	0.02[L]
EXECUTIVE FUNCTIONING								
Attentional control								

Inhibition and sustained attention[a]	49 / 283 (17)	52 / 283 (13.25)	18 / 291.5 (18.75)	2.02 (*p*=0.36)	TDC - DCP	-		
					SCP - DCP	-		
					SCP - TDC	-		
Selective verbal attention[a]	47 / 5 (2)	52 / 6 (2)	19 / 5 (2)	24.76 (*p*<0.01)	TDC - DCP	4.61 (*p*<.01)	1 (1;2)	0.68[M]
					SCP - DCP	0.00 (*p*=1)	0 (-1;1)	0.38[S]
					SCP - TDC	-3.36 (*p*<.01)	-1 (-2;-1)	0.16[L]
Selective visual attention[a]	47 / 5 (2)	52 / 6 (1)	19 / 4 (1)	46.21 (*p*<0.01)	TDC - DCP	5.79 (*p*<.00)	2 (1;2)	0.68[M]
					SCP - DCP	-1.64 (*p*=.10)	-1 (-1;0)	0.38[S]
					SCP - TDC	-5.29 (*p*<.00)	-2 (-3;-2)	0.16[L]
Cognitive flexibility								
Feedback utilization †[a]	50 / 9.5 (8)	52 / 7.5 (4)	18 / 10 (16.25)	7.44 (*p*=0.03)	TDC - DCP	-2.21 (*p*=.04)	-2 (-4;0)	0.34[M]
					SCP - DCP	0.88 (*p*=.38)	1 (-2;6)	0.55[S]
					SCP - TDC	2.19 (*p*=.04)	3 (0;9)	0.64[S]
Verbal working memory [a,c]	47 / 4 (2)	52 / 5 (2)	19 / 3 (1)	37.06 (*p*<0.01)	TDC - DCP	4.83 (*p*<.01)	2 (1;2)	0.71[M]
					SCP - DCP	0.00 (*p*=.20)	0 (-1;0)	0.28[M]
					SCP - TDC	-5.24 (*p*<.01)	-2 (-3;-1)	0.03[L]
Visual working memory [a,c]	47 / 5 (2.5)	52 / 6 (0.25)	18 / 4 (1)	50.55 (*p*<0.01)	TDC - DCP	5.85 (*p*<.01)	2 (1;2)	0.75[L]
					SCP - DCP	-1.54 (*p*=.12)	-1 (-1;0)	0.28[L]
					SCP - TDC	-5.96 (*p*<.01)	-2 (-3;-2)	0.02[L]
Goal setting								
Mean moves in tests of two moves†[c]	48 / 2 (0)	52 / 2 (0)	17 / 2 (0)	5.57 (*p*<0.07)	TDC - DCP	-		
					SCP - DCP	-		
					SCP - TDC	-		
Mean moves in tests of five moves†[c]	46 / 6.62 (2.44)	52 / 6.5 (1.75)	17 / 7.75 (1.75)	13.79 (*p*<0.01)	TDC - DCP	-1.80 (*p*=.07)	-0.50 (-1;0)	0.36[S]
					SCP - DCP	2.27 (*p*=.03)	1.12 (0.25;1.75)	0.67[M]
					SCP - TDC	3.68 (<.01)	1.50 (0.75;2.25)	0.77[L]
Problems solved in minimum moves[a]	48 / 8 (3)	52 / 9 (2.25)	18 / 7 (3)	24.69 (*p*<0.01)	TDC - DCP	3.48 (*p*<.01)	2 (1;2)	0.64[S]
					SCP - DCP	-1.91 (*p*=.06)	-1 (-3;0)	0.29[M]
					SCP - TDC	-4.55 (*p*<.01)	-3 (-4;-2)	0.10[L]
Information processing								
Lexical verbal fluency[b]	37 / 30 (23)	52 / 39.5 (14.75)	18 / 12.5 (11.5)	36.21 (*p*<0.01)	TDC - DCP	4.32 (<.01)	13 (7;19)	0.76[L]
					SCP - DCP	-2.54 (*p*=.01)	-11 (-20;-2)	0.27[L]
					SCP - TDC	-5.15 (*p*<.01)	-25 (-31;-18)	0.09[L]

*The level of significance was set at a *p*-value < 0.05 and p values were corrected for multiple comparisons (for global and posteriori contrasts); † Higher scores indicate worse performance; -, not applicable; CI, Bootrap-percentile confidence interval based on 10,000 bootstrap samples; DCP, dyskinetic cerebral palsy; H, Kruskal-Wallis statistic; HL, Hodges-Lehmann estimator; IQR, interquartile range; Md, median; SCP, spastic cerebral palsy; TDC, typically developing controls; z, transformed score from Mann-Whitney U-test. Reasons for missing data (handled with pairwise deletion): [a]absence or inenteligible speech and motor severity; [b] absence or inenteligible speech; [c]No comprehension of test instructions; [L], large effect size; [M], medium effect size; [S], small effect size.

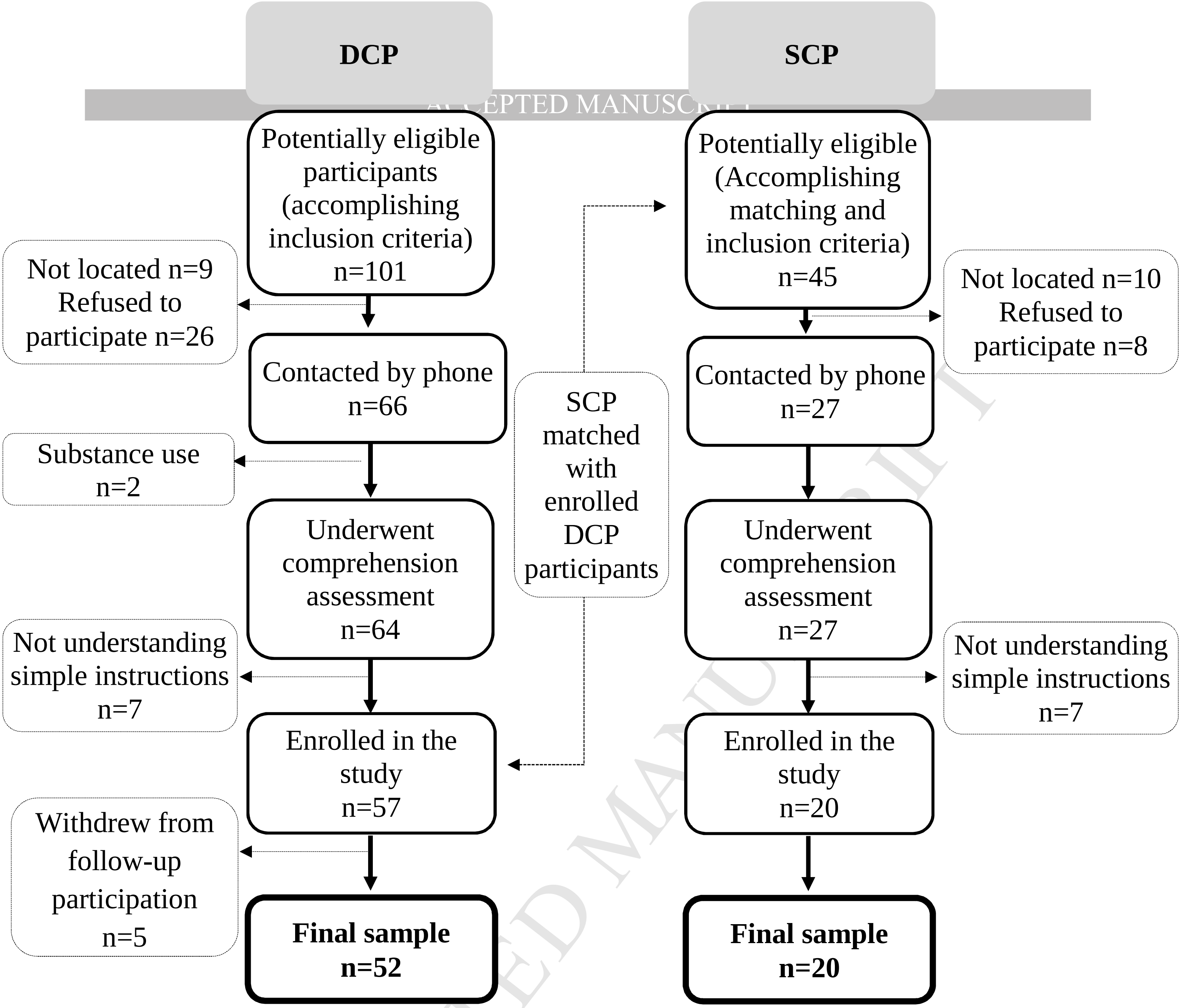

Figure 1. Flowchart showing requitement process for dyskinetic and spastic cerebral palsy participants. DCP, Dyskinetic cerebral palsy; SCP, Spastic cerebral palsy

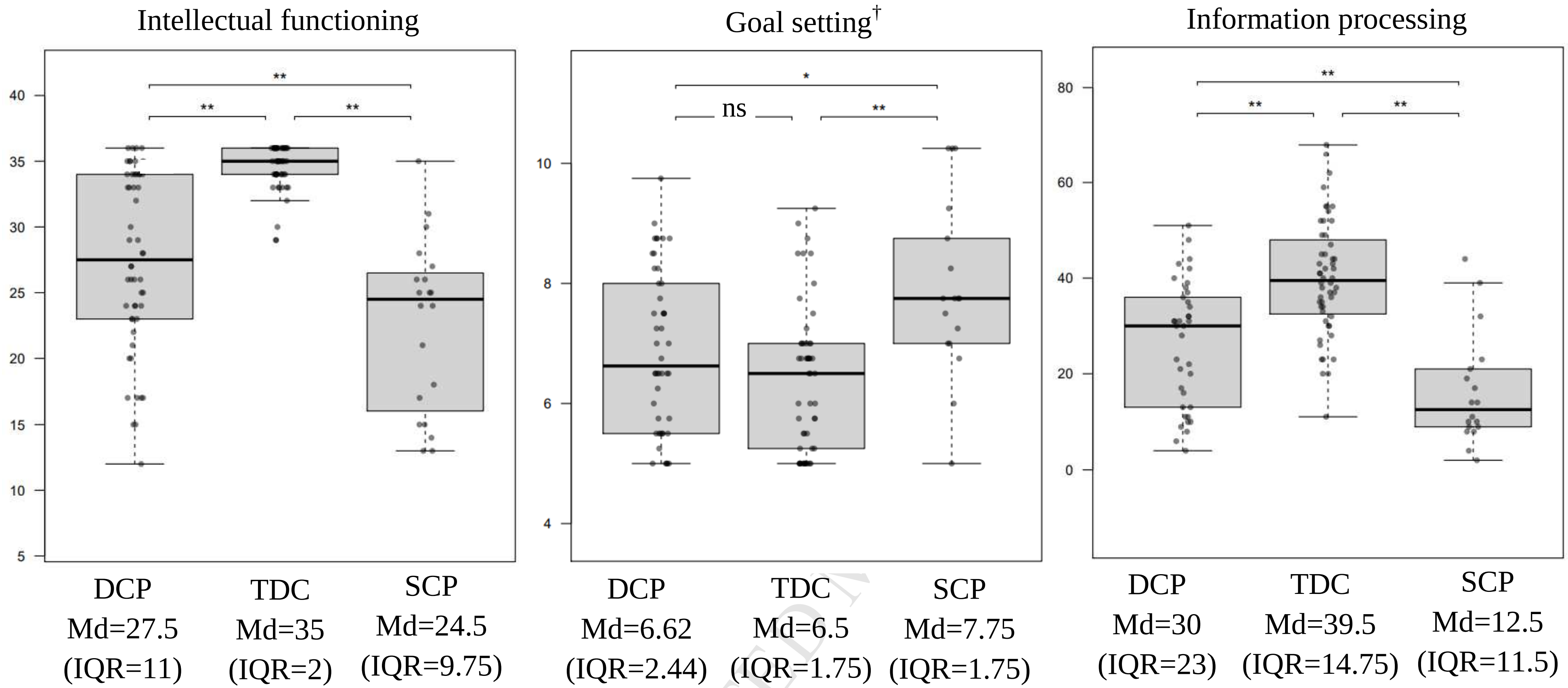

Figure 2. Boxplots showing raw scores (y axis) by groups (x axis) of intellectual functioning, goal setting (mean moves in tests of five moves) and information processing. Only boxplots showing significant differences between participants with dyskinetic and spastic cerebral palsy in table 3 are presented. *p < .05; **p <.01; † Higher scores indicate poorer performance in this task; DCP, Dyskinetic cerebral palsy; IQR, interquartile range; Md, median; SCP, Spastic cerebral palsy; TDC, typically developing controls.

Highlights

- General intellectual functioning is higher in DCP than in SCP.
- People with DCP display stronger executive function than those with SCP.
- People with DCP present poorer executive and general intellectual functioning than controls.

Conflict of interest statement

We wish to confirm that there are no known conflicts of interest associated with this publication and there has been no significant financial support for this work that could have influenced its outcome.

Executive function and intellectual functioning in dyskinetic cerebral palsy: comparison with spastic cerebral palsy and typically developing controls

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