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ORIGINAL ARTICLE

Academic skills in children with cerebral palsy and specific learning disorders

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Abstract

Aim: To investigate the prevalence and clinical manifestations of reading, writing, and mathematics disorders in children with cerebral palsy (CP). We explored how the clinical profile of these children differed from those with specific learning disorders (SLDs), taking into account several factors, particularly IQ scores, neuropsychological aspects, and the presence of a visual impairment.

Method: A prospective cross-sectional study was conducted in 42 children with CP (mean age 9 years 8 months; SD = 2 years 2 months) and 60 children with SLDs (mean age 10 years; SD = 1 year 7 months). Clinical characteristics, neuromotor and cognitive profiles, neuropsychological aspects (speech performance, academic skills, visual attention, phonological awareness, working memory), and signs of visual impairment (visual acuity, contrast sensitivity, visual field, oculomotor functions) were assessed. A machine learning approach consisting of a random forest algorithm, where the outcome was the diagnosis and the covariates were the clinical variables collected in the sample, was used for the analyses.

Results: About 59% of the children with CP had reading, writing, or mathematics disorders. Children with CP with learning disorders had a low performance IQ, normal phonological awareness, and working memory difficulties, whereas children with SLDs had normal performance IQ, impaired phonological awareness, and mild working memory difficulties. There were no differences in verbal IQ between the two groups.

Interpretation: Learning disorders are frequently associated with CP, with different clinical characteristics, compared with SLDs. Assessment of academic skills is mandatory in these children, even if the IQ is normal. At school age, specific interventions to promote academic skills in children with CP could be a major rehabilitative goal.

Cerebral palsy (CP) is a lifelong neurodevelopmental disorder that has a profound impact on all aspects of daily life.[1] CP is usually associated with sensory, perceptive, cognitive, communicative, and behavioural impairments due to the brain lesions themselves, which cause other symptoms related to adaptive functions in addition to motor deficits.[2] The coexistence of these concomitant impairments is highly variable, depending on the extent, topography, nature or severity, and timing of the brain damage.[2,3]

Intellectual disorders are frequently associated with CP and are present in about 40.0% to 50.0% of children,[4] with significant differences related to neuromotor involvement, which is less prevalent in children with unilateral forms (11.0–19.0%), but particularly debilitating in those with bilateral spastic forms (22.0–33.0% in bilateral [diplegic] CP to 90.0–100.0% in bilateral [quadriplegic] CP). Children with CP have a high prevalence of visual impairments[5,6] (16.0–90.0%), mainly represented by cerebral visual impairment

Abbreviations: CVI, cerebral visual impairment; PIQ, performance IQ; SLD, specific learning disorder; VIQ, verbal IQ; WISC, Wechsler Intelligence Scale for Children.

(CVI), defined as 'a verifiable visual dysfunction which cannot be attributed to disorders of the anterior visual pathways or any potentially co-occurring ocular impairment'.[7] Hearing loss affects approximately 40% of children with CP, with a tendency to be bilateral and with a severity that correlates with the degree of motor and neurological disability.[8] Approximately 60.0% of children with CP experience communication difficulties, which may include dysarthria, impaired language skills, or a combination of impaired speech and language skills.[9] In addition, behavioural difficulties, such as poor peer relationships, emotional symptoms, and hyperactivity are seen in approximately 30% of school-age children with CP and are not associated with socio-demographic variables and physical and cognitive characteristics.[10]

Children and adolescents with CP are also at risk for executive function difficulties, which are associated with damage to white matter tracts in the prefrontal and posterior regions of the brain.[11] Executive function includes higher-order skills such as attentional control, working memory, cognitive flexibility, goal setting, and information processing, which are necessary for goal-directed behaviour to complete activities.[12] Despite the growing interest in the assessment of executive function in children and adolescents with CP, there are no clear data on the prevalence of executive dysfunction in CP because of the diversity of tests used,[13] the type of CP considered (mainly spastic CP), the neurofunctional involvement[14,15] (unilateral vs bilateral), the time of injury[16] (prenatal vs perinatal or postnatal), and the executive function component examined, that is, attention,[17] cognitive flexibility,[18] inhibition and shifting,[19] and working memory.[19–22] The attention, inhibition, and shifting components of executive function have been described as being frequently impaired in children with CP,[15,17,23,24] especially in children with bilateral lesions.[15] Moreover, the prevalence of attention-deficit/hyperactivity disorder, a neurodevelopmental disorder in which executive dysfunction is a core symptom, has been estimated to be present in 20.0% to 30.0% of children with CP, particularly in those with low IQ scores and severe motor or language deficits.[25] Many studies also examined the association between certain executive function skills and academic achievement.[19–22]

Learning disorders, that is, persistent difficulties in learning key academic skills such as reading, written expression, and spelling, or mathematics, have also been frequently reported in children with CP, with a prevalence ranging from 30.0% to 70.0%.[26–29] According to the DSM-5,[30] learning disorders may be present in a specific form (specific learning disorders [SLDs], 5.0–15.0% of the general population) or in an unspecified form when other medical conditions such as sensory, intellectual, or neurological disorders are present, as in children with CP. The DSM-5 lists SLDs specifiers for: 'impairment in reading', also used in its alternative term dyslexia, expressed by difficulties in word reading accuracy, reading speed or fluency, and reading comprehension; 'impairment in written expression', expressed by difficulties in spelling accuracy, grammar, and punctuation accuracy in written expression, clarity, or organization of written expression; and 'impairment in mathematics', also used in its alternative term dyscalculia, expressed by difficulties in number sense, memorization of arithmetic facts, accurate or fluent calculation, and accurate mathematical reasoning.

What this paper adds

- Reading, writing, and mathematics disorders in cerebral palsy have specific clinical characteristics.
- Their underlying mechanisms differ from those described in specific learning disorders.
- Working memory impairment can be considered a hallmark of learning disorders in children with cerebral palsy.

When associated with CP, learning disorders cannot be described as specific because they may be justified by the broader neurocognitive and sensory deficits caused by prenatal, perinatal, and postnatal structural brain injury or connectivity disorders that affect these children.[31,32]

Learning disorders have received less research attention in children with CP, probably because of their complex profile, which results in their inability to sustain typical reading, writing, and mathematics performance. Children with CP show a delay in emergent literacy skills, even before formal reading and writing education begins.[33] Difficulties in literacy skills may vary according to the type of CP and tend to be more preserved in ambulant and verbal children.[32] Children with CP are more likely to experience difficulties with mathematics,[19,22,34–39] in particular poorer numeracy skills,[35] difficulties in subitizing,[37] counting, developing a mental number line,[38] and arithmetics.[39] Finally, handwriting, an important component of writing performance, is also often impaired in children with CP and upper-limb involvement[40] because of the impaired finger proprioception and bimanual coordination that children with CP typically experience.[41]

Several co-occurring variables have been described in association with learning disorders in CP. These include IQ,[36] auditory perception,[11] phonological awareness,[21] which is the ability to reflect on and manipulate the sound structure of spoken words,[33] and working memory,[19–22] a component of executive function that has a key role in understanding situations that evolve over time, making sense of linguistic information, mentally manipulating elements, and linking previous ideas to form new concepts.[42,43] Translated into an educational setting, working memory is important for mentally retaining letter–sound associations during reading tasks and for monitoring incoming information during mathematical skills.[44,45] In children with CP, phonological awareness, speech expression, and non-verbal intelligence have been specifically described as strong predictors of literacy, while working memory has been seen as a strong

predictor of mathematics skills.[19–22,34] In particular, children with CP and learning disorders have impaired phonological awareness, especially when coexisting with a speech impairment because of difficulties in retrieving whole-word phonology.[33]

It is also important to consider the role that vision can have in the development of academic skills because a considerable portion of school learning activities (70% of the school day[46]) involve tasks based on visual functions that may be impaired in children with CP. The main signs and symptoms associated with a visual impairment (mainly represented by CVI in children with CP) are:[47] eye problems; oculomotor dysfunction (impaired fixation, smooth pursuit, and saccades); basic visual function deficits (such as visual acuity deficit, altered contrast sensitivity, and visual field limitation); and cognitive and visual disorders (visual–spatial, visual-perceptual skills, and visual orientation dysfunctions). All of these visual signs and symptoms are usually reported in children with CP[5,6] and could further impair learning. These complex clinical problems partly justify the possibility that children with CP may have difficulties with academic skills.

As far as SLDs are concerned, the main problem highlighted in the literature is phonological deficit. According to these theories, reading difficulties result from a cognitive deficit in the representation and processing of speech sounds.[48] Visual or visual and attention deficits have been proposed as an alternative cause of dyslexia,[49] but may affect only a subset of individuals with dyslexia, which can present in different forms.[50] A combination of multiple deficits (including working memory, visuospatial skills, and visual attention) seems to underlie mathematical skills.[44,45]

To broaden new perspectives on learning disorders in CP, this study aimed to: (1) investigate the prevalence and clinical manifestations of learning disorders (reading, writing, and mathematics disorders) in a group of children with CP with normal verbal IQ (VIQ) and reduced motor involvement (Gross Motor Function Classification System [GMFCS] levels I–III, Manual Ability Classification System [MACS] levels I and II); and (2) explore how the clinical profile of a group of children with CP and learning disorders differs from that of children with CP without learning disorders and children with SLDs, considering several factors, particularly IQ scores, neuropsychological aspects, and the presence of visual impairment, through the use of machine learning methods, to better understand and visualize the relationships among variables.

METHOD

Participants

In this prospective cross-sectional study, carried out from February 2016 to April 2020, among 145 children with CP referred by the Centre for Diagnosis and Treatment of Children with Neurovisual Problems and Multidisabilities at the Unit of Child Neurology and Psychiatry of ASST Spedali Civili, University of Brescia, Italy, a group of 44 children with CP were invited to participate according to the following inclusion criteria: (1) classified in levels I to III on the GMFCS;[51] (2) classified in levels I or II on the MACS;[52] (3) IQ within the normal range or borderline intellectual functioning (VIQ > 85 or full-scale IQ ≥ 70) consistent with the Wechsler Intelligence Scale for Children, Third Edition (WISC-III) testing;[53] (4) ability to speak Italian and good speech intelligibility (level I on the Viking Speech Scale);[54] (5) normal or near-normal visual acuity (no less than 4/10 with binocular viewing); (6) no hearing impairment as assessed by audiological examination; (7) absence of other neurodevelopmental disorders according to the DSM-5 classification;[30] and (8) regular attendance of at least the second year of primary school in a mainstream educational setting.

Two families did not give informed consent to participate in the study. Finally, a group of 42 children (20 females, 22 males; mean age 9 years 8 months; SD = 2 years 2 months, age range: 7–16 years) participated in the study. A learning support teacher assisted 23 children (54.8%) during school hours, although children followed the regular classroom learning programme; 27 children (64.3%) needed help to get to school.

A group of 60 children (29 males, 31 females; mean age 10 years; SD = 1 year 7 months; age range: 8–16 years) with SLDs (the group with SLDs) was also recruited for comparison at the Unit of Child Neurology and Psychiatry of ASST Spedali Civili, University of Brescia. All individuals in the group with SLDs were consecutively invited from the wider sample admitted to the Unit for suspected SLDs and underwent specific neuropsychological assessment for diagnosis, detailed in the 'Procedures' section of this article.

All the children meeting the following criteria were diagnosed with SLDs:[55] (1) presence of subjective and objective learning difficulties for at least 6 months despite the provision of extra help or targeted intervention; (2) completed and regularly attended at least the second year of primary school in a mainstream educational setting; (3) academic underachievement was not due to intellectual disability (VIQ > 85 and full-scale IQ ≥ 70), other mental or neurological disorders, visual or hearing problems, or poor or inappropriate academic instruction; (4) ability to speak Italian; (5) performance below the fifth centile or − 2 SD in reading, writing, or mathematics tasks. Specifically, the adopted diagnostic criteria for reading disorders required performance on at least two tasks to be below −2 SD or the fifth centile in speed or accuracy for a positive diagnosis.[56] The diagnostic criteria for writing disorders required performance on at least three tasks to be below −2 SD or the fifth centile.[56] Mathematics disorders were then diagnosed if the total score was below −2 SD or the fifth centile in at least four subtests in the battery. Children with one or more associated coded neurodevelopmental disorders were excluded.

All families of the selected participants gave written informed consent to participate in the study; the children were referred to the Centre for the Diagnosis and Treatment of Children with Neurovisual Problems and

Multidisabilities at the Unit of Child Neurology and Psychiatry of ASST Spedali Civili, University of Brescia, to assess the visual aspects.

All participants with CP and with SLDs attended nursery school in Italy as infants, were educated in mainstream schools, and had parents who were proficient in Italian. No socioeconomic issues were reported based on available national registry information or were revealed during history taking. All the children completed the evaluation and no missing data were reported.

Procedures

The study was approved by the ethics committee of Brescia (Protocol number 1324). All study procedures were performed in accordance with relevant guidelines and regulations. Written informed consent was obtained from 42 parents or legal guardians of the 44 children with CP and from all the parents or legal guardians of the children with SLDs.

All children underwent neurological, neuromotor, cognitive, neuropsychological, and neurovisual evaluation. Neuromotor skills were assessed only in children with CP using the GMFCS,[51] while fine motor skills were measured using the Italian version of the MACS.[52] The WISC-III was used to assess cognitive skills.[53] It was preferred to the fourth edition to be consistent with those children who had already been tested to assess IQ level using the WISC-III (see Appendix S1 for a comparison between the WISC-III and WISC-IV scores in this study). Speech intelligibility was evaluated using the Viking Speech Scale.[54]

The neuropsychological evaluation included: (1) assessment of visual attention skills using the Bells cancellation task,[57] with results on the sustained attention component; (2) evaluation of phonological awareness using the phonological processing task from the NEPSY II;[58] and (3) evaluation of working memory using the listening recall task,[59] in its Italian version. In this task, participants listened to sets of grammatical utterances, decided whether each statement was true or false while repeating the last word of each utterance, and recalled these words at the end of each set. The number of words recalled represented the listening span of each participant. All results were expressed as z-scores.

Learning skills were assessed in both children with SLDs and CP according to the Italian guidelines for the evaluation of SLDs.[55,56] Participants' abilities were tested in terms of speed and accuracy in reading and calculation, and accuracy in writing. Reading ability was assessed using MT text reading tests,[60–62] and the single unrelated words and single unrelated non-words subtests.[63] Both the number of errors (accuracy) and the time taken to complete the task (speed) were recorded. Writing difficulties were expressed by orthographic coding, assessed in terms of the number of errors present in the written text,[64] untimed single unrelated words,[63] untimed single unrelated non-words under the dictation subtests,[63] and spontaneous text writing.[64] The evaluation of mathematic disorders was carried out using standardized dyscalculia batteries.[62,65] The handwriting component was not included in the neuropsychological assessment battery because of the difficulties that children with CP may have with a skill that requires particularly specific and accurate fine motor skills.

The neurovisual assessment was conducted according to Galli et al.[5] and Fazzi et al.[6] It included the evaluation of refractive errors (assessed in cycloplegia), oculomotor functions (fixation, smooth pursuit, and saccades), and basic visual functions (visual acuity, contrast sensitivity, and visual field). If refractive errors were present, children were asked to wear spectacles with maximum refractive correction for the evaluation. Visual fixation, smooth pursuit, saccades, visual field, and contrast sensitivity were defined as normal or impaired according to Galli et al.[5] Visual acuity was evaluated under maximum refractive correction using letter optotypes and expressed in tenths.

For both CP groups (i.e. with and without learning disorders), neuropsychological evaluations consisting of phonological awareness, working memory, visual attention, and learning skills assessments were conducted at the end of the evaluation protocol. For children with SLDs, neurovisual evaluation was conducted at the end of the evaluation protocol and after enrolment (see Figure S1 for the details). A multidisciplinary team consisting of an ophthalmologist, an orthoptist, a paediatric neurologist, two paediatric neuropsychologists, and a rehabilitation therapist conducted the evaluations. An expert paediatric neurologist, assisted by a paediatric therapist, performed the neurological, neuromotor, and neurovisual examination, preceded by a preliminary ophthalmological and orthoptic evaluation. These clinicians were blinded to the group to which the patients with CP they evaluated belonged (whether they were children with CP with learning disorders or children with CP without learning disorders, defined according to the neuropsychological evaluation) to limit possible bias. Two paediatric neuropsychologists conducted the cognitive, linguistic, and neuropsychological evaluation, including the assessment of learning skills, in all three groups.

The most important clinical variables according to previous literature, clinical experience, and the presence of significant differences between the three groups, were then used for the machine learning analysis.

Statistical analysis

A total of 102 children were included in the study (41.2% with CP and 58.8% with SLDs). To define the sample size, the maximum uncertainty scenario was chosen (i.e. CP and SLDs occurred in 50% of the sample). Consequently, the precision of the study, measured as the half-width of the confidence interval (CI) of the proportion, was equal to 0.1, with a CI ranging between 0.4 and 0.6.

Descriptive statistics were calculated, including the mean, SD, 95% CI, median, first (Q1) and third (Q3) quartiles, and

range (min–max) for the quantitative variables, while frequencies (absolute and relative percentage) were calculated for the categorical variables. When data were stratified according to diagnosis, a Wilcoxon rank-sum test (or Kruskal–Wallis test when comparing more than two categories) was calculated for the quantitative variables, and a Fisher's exact test for the qualitative variables. When tests were applied to more than two groups, pairwise comparisons were calculated (using Holm–Bonferroni correction to adjust the *p*-value).

Using classification models, it is possible to identify which specific clinical variables had a major impact on predicting the diagnosis (children with CP without learning disorders, children with CP with learning disorders, or children with SLDs). Specifically, a machine learning approach was used, namely the random forest algorithm, where the outcome is the diagnosis and the covariates are the clinical variables collected in the sample (i.e. VIQ, performance IQ [PIQ], phonological awareness, working memory, visual fixation, smooth pursuit, saccades, and visual acuity); missing values were imputed using the missForest algorithm.[66] Random forest is an ensemble method often used for classification problems; it combines thousands of classification trees to provide accurate predictions, thus overcoming the instability problem (i.e. small changes in the data can cause large changes in the results) of a single tree. It estimates a classification model using the diagnosis as the outcome and all other variables (qualitative and quantitative) as the covariates. The benefit of this method is that it can model non-linear relationships between the outcome (which is categorical) and the covariates; it can also address collinearity issues.

In this study, the random forest algorithm was not used in a predictive sense, but rather to understand the relationships between covariates and outcomes. Therefore, additional information was extracted. This included (1) the relative variable importance measure and (2) the three-dimensional partial dependence plots.[67–70]

In addition, a single classification tree was developed because it allowed the identification of homogeneous clusters of patients with respect to the outcome (diagnosis) of the model. Further details of the algorithms used can be found in Appendix S2.

All analyses were computed with R v4.2.1 (R Foundation for Statistical Computing, Vienna, Austria), using the following libraries: arsenal, MASS, ggplot2, missForest, randomForest, pdp, rpart, and rpart.plot.

RESULTS

Prevalence and clinical manifestations of learning disorders in a group of children with CP

Twenty-five of the 42 children with CP (59.5%) had at least one learning disorder (group of children with CP with learning disorders: 14 males and 11 females; mean age: 9 years 11 months; age range: 8–15 years). Reading and writing disorders were found in 19 (45.2%) and 17 (40.4%) children respectively. Fifteen children (35.7%) had a mathematics disorder. Multiple learning disorders were found in 16 children (38.1%), of which 10 showed a disorder involving mathematics, reading, and writing skills, four showed reading and writing disorders, one showed reading and mathematics disorders, and one showed writing and mathematics disorders.

Seventeen children did not present with learning disorders (group with CP without learning disorders: 6 males and 11 females; mean age: 9 years 11 months; age range: 7–16 years).

No statistically significant differences were found based on sex, age, preterm birth, type of CP (bilateral vs unilateral involvement), GMFCS and MACS levels, IQ levels, and CVI signs and symptoms, between children with CP with learning disorders and children with CP without learning disorders (Table 1).

In children with CP with learning disorders, reading disorders were present in 76.0%, writing disorders in 68.0%, and mathematics disorders in 60.0%. In children with SLDs, 83.3% presented a reading disorder, 80.0% a writing disorder, and 70.0% a mathematics disorder. Additionally, children in the SLDs group presented either two or three of the assessed learning disorders at the same time. No statistically significant differences were observed with respect to the distribution of reading ($p = 0.544$), writing ($p = 0.268$), or mathematics ($p = 0.450$) disorders between children with SLDs and children with CP with learning disorders.

Clinical profile of the three groups

Complete details of the clinical profiles of the three groups (children with CP with learning disorders, children with CP without learning disorders, children with SLDs) are shown in Table 2. The evaluation of neuromotor skills using the GMFCS and MACS was conducted only for children with CP as described in Table 1. Children with CP with learning disorders showed the lowest PIQ values, and these were significantly different from the values in children with SLDs. Refractive errors, strabismus, visual acuity deficit, fixation, and smooth pursuit impairments were all significantly more frequent in children with CP compared to children with SLDs. No differences were found between children with CP without learning disorders and children with CP with learning disorders. Visual attention skills were similar among the three groups. Children with CP with learning disorders and children with CP without learning disorders both showed preserved phonological awareness, which was lower in the group with SLDs by comparison. Children with CP with learning disorders showed the greatest difficulties in the working memory task, and scores were significantly lower than in children with CP without learning disorders.

A random forest classifier was estimated using the diagnosis (children with CP with learning disorders, children with CP without learning disorders, and children with SLDs)

TABLE 1 Descriptive statistics on the stratified clinical features of children with CP with and without learning disorders.

Clinical feature	Children with CP without learning disorders ($n = 17$)	Children with CP with learning disorders ($n = 25$)	p
Neuromotor classification (Hagberg)			1.00[a]
Bilateral	8 (47.1%)	12 (48.0%)	
Unilateral	9 (52.9%)	13 (52.0%)	
Sex			0.222[a]
Male	6 (35.3%)	14 (56.0%)	
Female	11 (64.7%)	11 (44.0%)	
Age			0.53[b]
Mean (95% CI)	9.9 (8.6–11.3)	9.9 (9.1–10.8)	
Median (Q1–Q3)	9.0 (8.0–11.0)	9.0 (9.0–10.0)	
Range	7.0–16.0	8.0–15.0	
Preterm birth			1.000[a]
No	10 (58.8%)	14 (56.0%)	
Yes	7 (41.2%)	11 (44.0%)	
GMFCS levels			0.497[a]
I	7 (41.2%)	15 (60.0%)	
II	8 (47.1%)	7 (28.0%)	
III	2 (11.7%)	3 (12.0%)	

Abbreviations: CI, confidence interval; CP, cerebral palsy; GMFCS, Gross Motor Function Classification System.

[a]Fisher's exact test.

[b]Wilcoxon rank-sum test. Q1, first quartile; Q3, third quartile.

as the outcome and VIQ, PIQ, phonological awareness, working memory, visual fixation, smooth pursuit, saccades, and visual acuity as the covariates. Contrast sensitivity and visual field were not included in the model because of the smaller number of participants with these disorders. Using the relative variable importance measure extracted using the random forest algorithm, phonological awareness, PIQ, and working memory were strong determinants in classifying patients with the three diagnoses. The algorithm assigns to these three covariates a relative variable importance measure greater than 60.0% (threshold chosen according to the literature[67]). Figure 1 shows the ranking from the most significant (phonological awareness with relative variable importance measure = 100%) to the least significant (saccades with variable importance measure = 12.7%) variable. Three-dimensional partial dependence plots were extracted from the random forest algorithm (Figure 2) to visualize the probability of being classified with one of the three diagnoses according to the most significant covariates (phonological awareness, PIQ, working memory). Figure 2a, which shows the probability of being classified as with CP without learning disorders with varying phonological awareness, PIQ, and working memory, and 2b (the probability of being classified as with CP with learning disorders) showed a similar trend. Figure 2c (the probability of being classified as with SLD) showed a reverse trend. Specifically, Figure 2a shows that children with high scores for phonological awareness and working memory but with low or average scores for PIQ had a high probability (0.4) of being classified as with CP without learning disorders. Figure 2b shows that children with high scores for phonological awareness but with low scores for working memory and PIQ had a high probability (0.6) of being classified as with CP with learning disorders. Finally, Figure 2c shows that children with high scores in PIQ and mild working memory difficulties but with low scores for phonological awareness had a high probability (0.8) of being classified as with SLDs.

The classification tree (Figure 3) was created using the diagnosis (CP with learning disorders, CP without learning disorders, and SLDs) as the outcome, with VIQ, PIQ, phonological awareness, working memory, visual fixation, smooth pursuit, saccades, and visual acuity as the covariates. The algorithm identifies the clinical variables (and corresponding thresholds) that cluster children into homogeneous groups according to the outcome. The classification tree confirmed the importance of phonological awareness, PIQ, and working memory for partitioning the data, which are found in the first, second, and third levels of the tree (see the top of the tree structure). Low phonological awareness scores were specific for the group with SLDs (blue rectangles; nodes at the base of the tree). High phonological awareness scores along with low PIQ scores were typically present in both groups with CP (violet and pink nodes). Moreover, high phonological awareness with low PIQ and low working memory scores specifically identified the group with CP with learning disorders (pink nodes). Both groups with CP were on the left side of the tree (violet and pink nodes), while the group with SLDs was on the right side (blue nodes). This separation helped to confirm that, among the selected variables, children with CP associated

TABLE 2 Descriptive statistics on the possible clinical determinants of learning disorders, stratified according to the three groups: children with CP without learning disorders, children with CP with learning disorders, and children with SLDs.

Clinical feature	Children with CP without learning disorders ($n = 17$)	Children with CP with learning disorders ($n = 25$)	Children with SLDs ($n = 60$)	p
Full IQ[a]				0.056[b]
Mean (95% CI)	100.0 (92.0–108.0)	92.0 (86.0–98.1)	98.8 (96.1–101.5)	
Median (Q1–Q3)	98.0 (93.0–113.0)	89.0 (82.0–100.0)	101.0 (90.5–106.0)	
Range	70.0–130.0	70.0–126.0	80.0–121.0	
Verbal IQ[a]				0.288[b]
Mean (95% CI)	106.2 (99.1–113.3)	99.4 (92.6–106.2)	103.9 (100.5–107.2)	
Median (Q1–Q3)	107.0 (93.0–114.0)	98.0 (90.0–112.0)	104.0 (96.0–112.0)	
Range	85.0–140.0	86.0–128.0	82.0–140.0	
Performance IQ[a]				**0.002**[b]
Mean (95% CI)	97.7 (87.0–108.4)	90.2 (83.2–97.1)	105.2 (101.4–109.0)	CP without learning disorders vs CP with learning disorders: 0.332[c]
Median (Q1–Q3)	96.0 (89.0–104.0)	87.0 (77.0–106.0)	102.5 (95.0–116.0)	CP without learning disorders vs SLDs: 0.332[c]
Range	45.0–129.0	66.0–127.0	71.0–135.0	CP with learning disorders vs SLDs: **0.001**[c]
Refractive error				**<0.001**[d]
No	2 (11.8%)	2 (8.0%)	50 (83.3%)	CP without learning disorders vs CP with learning disorders: 1.000[c]
Yes	15 (88.2%)	23 (92.0%)	10 (16.7%)	CP without learning disorders vs SLDs: **<0.001**[c] CP with learning disorders vs SLDs: **<0.001**[c]
Strabismus				**<0.001**[d]
No	10 (58.8%)	12 (48.0%)	58 (96.7%)	CP without learning disorders vs CP with learning disorders: 0.543[c]
Yes	7 (41.2%)	13 (52.0%)	2 (3.3%)	CP without learning disorders vs SLDs: **<0.001**[c] CP with learning disorders vs SLDs: **<0.001**[c]
Visual field deficit				**0.017**[c]
No	17 (100.0%)	22 (88.0%)	60 (100.0%)	CP without learning disorders vs CP with learning disorders: 0.519[c]
Yes	0 (0%)	3 (12.0%)	0 (0%)	CP without learning disorders vs SLDs: 1.000[c] CP with learning disorders vs SLDs: 0.070[c]

TABLE 2 (Continued)

Clinical feature	Children with CP without learning disorders ($n = 17$)	Children with CP with learning disorders ($n = 25$)	Children with SLDs ($n = 60$)	p
Visual acuity				**<0.001**[b]
Mean (95% CI)	9.3 (8.7–9.9)	8.7 (8.0–9.5)	9.8 (9.4–10.1)	CP without learning disorders vs CP with learning disorders: 0.331[c]
Median (Q1–Q3)	10.0 (9.0–10.0)	10.0 (8.0–10.0)	10.0 (10.0–10.0)	CP without learning disorders vs SLDs: **0.030**[c]
Range	6.0–10.0	4.0–10.0	9.0–10.0	CP with learning disorders vs SLDs: **<0.001**[c]
Visual fixation				**<0.001**[d]
Normal Impaired	12 (70.6%) 5 (29.4%)	16 (64.0%) 9 (36.0%)	57 (95.0%) 3 (5.0%)	CP without learning disorders vs CP with learning disorders: 0.747[c] CP without learning disorders vs SLDs: **0.022**[c] CP with learning disorders vs SLDs: **0.002**[c]
Smooth pursuit				**<0.001**[d]
Normal Impaired	10 (58.8%) 7 (41.2%)	13 (52.0%) 12 (48.0%)	54 (90.0%) 6 (10.0%)	CP without learning disorders vs CP with learning disorders: 0.758[c] CP without learning disorders vs SLDs: **0.012**[c] CP with learning disorders vs SLDs: **<0.001**[c]
Saccades				**0.003**[d]
Normal Impaired	10 (58.8%) 7 (41.2%)	10 (40.0%) 15 (60.0%)	47 (78.3%) 13 (21.7%)	CP without learning disorders vs CP with learning disorders: 0.346[c] CP without learning disorders vs SLDs: 0.249[c] CP with learning disorders vs SLDs: **0.003**[c]
Visual attention[e]				0.216[b]
Mean (95% CI)	−0.8 (−1.5 to 0)	−1.3 (−1.9 to −0.7)	−0.9 (−1.2 to −0.6)	
Median (Q1–Q3)	−0.8 (−1.6 to 0)	−1.2 (−2.1 to −0.5)	−0.8 (−1.3 to −0.4)	
Range	−4.6 to 1.9	−3.8 to 1.7	−4.1 to 2.2	
Working memory[e]				**0.030**[b]
Mean (95% CI)	−0.1 (−0.9 to 0.8)	−1.3 (−1.8 to −0.8)	−0.9 (−1.2 to −0.5)	CP without learning disorders vs CP with learning disorders: **0.039**[c]
Median (Q1–Q3)	0.4 (−1.3 to 0.8)	−1.2 (−2.1 to −0.5)	−0.7 (−1.9 to 0.1)	CP without learning disorders vs SLDs: 0.122[c]
Range	−3.6 to 2.6	−3.7 to 2.1	−3.5 to 1.4	CP with learning disorders vs SLDs: 0.153[c]
Phonological awareness[e]				**<0.001**[b]
Mean (95% CI)	0.7 (0.2–1.3)	0.1 (−0.5 to 0.6)	−1.4 (−1.8 to −0.9)	CP without learning disorders vs CP with learning disorders: 0.104[c]
Median (Q1–Q3)	1.1 (0.4–1.4)	−0.2 (−1.0 to 1.0)	−1.3 (−2.4 to −0.1)	CP without learning disorders vs SLDs: **<0.001**[c]
Range	−1.8 to 1.9	3.5–2.6	−5.9 to 1.5	CP with learning disorders vs SLDs: **0.002**[c]

Abbreviations: CI, confidence interval; CP, cerebral palsy; SLD, specific learning disorder.

[a]Expressed as standard scores (mean = 100; SD = 15).

[b]Kruskal–Wallis test.

[c]Pairwise comparison (Holm–Bonferroni-adjusted p-value).

[d]Fisher's exact test.

[e]Expressed as z-scores (mean = 0, SD = 1). $p < 0.05$ are shown in bold. Q1, first quartile; Q3, third quartile.

with learning disorders presented a different clinical profile compared to children with SLDs.

DISCUSSION

To date, data on the association between CP and learning disorders in the literature have been scarce and inconsistent.[26–29] In this study, we demonstrated that 59.5% of children with CP, with normal VIQ, classified in GMFCS levels I to III and MACS levels I and II, presented with learning disorders, with manifestations of reading, writing, and mathematics difficulties similar to those of children with SLDs.

The prevalence we observed lies within the 30% to 70% range of previous reports,[26–29] which probably reflects the heterogeneity of the study samples and the different methodologies used to assess the presence of learning disorders in individuals with CP. For example, Schenker et al.[27] and Gillies et al.[28] collected data retrospectively from national databases but provided no further information about the definition of learning disorders nor did they report IQ levels. In a study by Frampton et al.,[26] children with unilateral CP were tested prospectively on their reading, writing, and mathematics skills, while considering their intellectual level; many children performed too poorly in the attainment tests to be able to determine whether or not they had learning disorders. In the current study, the detailed assessment protocol, following the national learning disorder diagnosis guidelines,[55,56] permitted an accurate evaluation of the presence and clinical characteristics of academic competencies. It also provided a more reliable comparison with the neurocognitive profile of children with CP and children with SLDs as they had a similar age, and showed similar levels of visual attention, full-scale IQ, VIQ, and speech performance.

The random forest analysis clearly demonstrated that working memory, PIQ, and phonological awareness were the most significant variables to discriminate between children in the three groups (children with CP with learning disorders, children with CP without learning disorders, children with SLDs).

Children with CP with learning disorders showed significant working memory difficulties and low PIQ scores, as well as normal or mildly impaired phonological awareness. Children with CP without learning disorders differed from the group with learning disorders only in higher working memory scores. On the other hand, the group with SLDs showed normal or only mildly impaired working memory, above-normal PIQ scores, and low phonological awareness scores.

Working memory scores had a prominent role in helping to differentiate the presence of learning disorders in CP as their scores were significantly lower in children with CP with learning disorders than in children with CP without learning disorders. Working memory is the core executive function that permits us to work with information no longer perceptually present.[12] It is important in supporting the development of other cognitive skills; it is critical for making sense of the world information that continuously reaches our senses, to mentally reorder items, consider alternatives, and derive general principles from related information.[12] It is particularly relevant for academic performance because it is necessary to make sense of written or spoken language, to perform mathematical calculations in the head, for numerical cognition,[44,45,71] and with early numeracy.[35] Working memory is frequently described as being impaired in children with CP, especially in the bilateral forms,[72] along with other executive function components that affect mathematics skills, such as shifting and inhibition, which are predictors of future mathematics skills.[19,22]

While low working memory scores indicated the presence of learning disorders in children with CP, low PIQ scores were significant in differentiating children with CP (lower scores) from children with SLDs (higher scores). Low PIQ scores are frequently described in children with bilateral forms of CP,[73–76] whose motor impairment affects speedy responses and manipulation of stimuli, which can limit performance in specific subtests. In particular, the block design subtest, which determines PIQ, is difficult for children with CP, leading to an IQ underestimation of three to six points.[77] Lower PIQ scores can be a marker of visual impairment, particularly CVI, in children with CP, which is related to the specific vulnerability of white matter tracts, as occurring in periventricular leukomalacia.[76] In the current study, CVI signs and symptoms, such as visual acuity deficits and oculomotor dysfunctions, were significantly more frequent in children with CP than in children with SLDs (Table 2) and may have indirectly contributed to the lower performances in terms of PIQ and learning disorders. Associations have previously been reported between reading performance and refractive errors and visual acuity,[78] contrast demand, sustained accommodative and vergence ability,[46] oculomotor findings,[79] and visual-perceptual skills.[80] Vision also has an impact on mathematics skills, as demonstrated by the reported links between mathematical components and basic visual perception,[81] visual movement perception,[82] visual working memory,[45,83] visual attention,[84] and visuospatial mental rotation.[85] However, the signs and symptoms of CVI were less useful for distinguishing the presence of learning disorders when inserted in the random forest algorithm, despite the prominent role of PIQ.

It is possible that these CVI signs and symptoms may have helped to strengthen the role of PIQ as a strong determinant for classifying patients as with CP or SLDs; however, further studies that specifically analyse the role of visual functions in each learning disorder are needed.

Low phonological awareness scores are crucial for distinguishing children with SLDs (lower scores) from children with CP with and without learning disorders (higher scores). While phonological processing is one of the main causes of literacy disorders in children with SLDs,[48,86,87] irrespective of the spoken and coded language, in our study this association was less evident in children with CP.

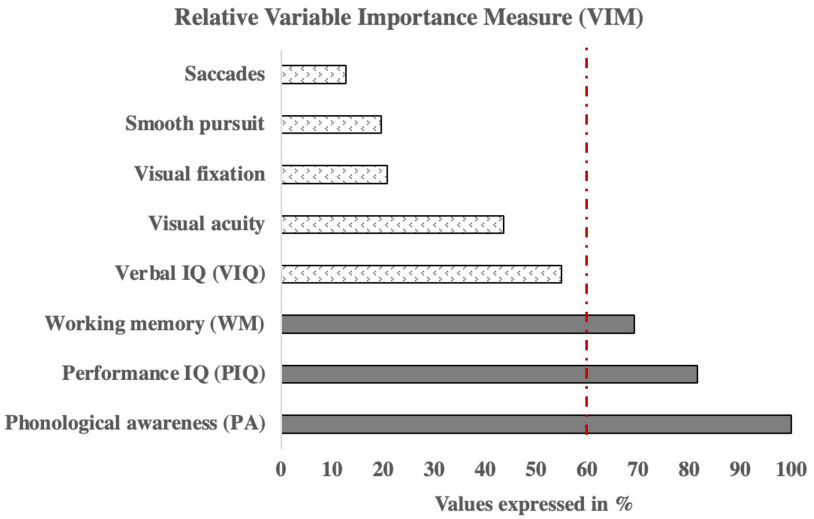

FIGURE 1 Relative variable importance measure (VIM) identified which variables had a strong impact on outcome, that is, the diagnosis (children with cerebral palsy [CP] without learning disorders, children with CP with learning disorders, children with specific learning disorders). It is extracted from a random forest model where the diagnosis was the outcome and verbal IQ (VIQ), performance IQ (PIQ), phonological awareness, working memory, visual fixation, smooth pursuit, saccades, and visual acuity were the covariates. Relative VIM normalizes the most significant values to simplify the interpretation of the results; it is defined as the percentage improvement with respect to the most important predictor. It produces a ranking from the most significant (phonological awareness with relative VIM = 100.0%) to the least significant (saccades with relative VIM = 12.7%) variable. The grey bars represent those variables with a high value relative to the VIM (>60.0%), that is, the three strong diagnostic predictors. The cut-off used for variable selection (60.0%, identified by the red dashed line) was chosen according to recommendations in the literature.

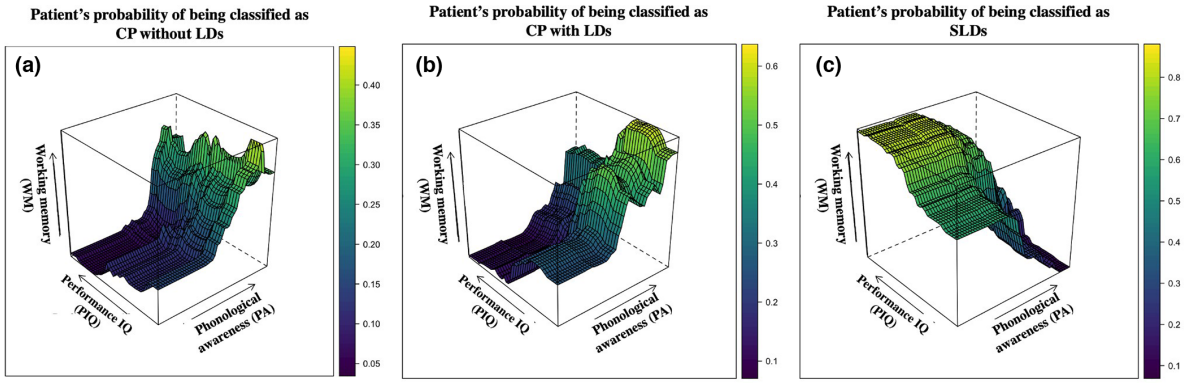

FIGURE 2 Three-dimensional partial dependence plots showing how model classifications (children with cerebral palsy [CP] without learning disorders (LDs), children with CP with learning disorders, and children with specific learning disorders [SLDs]) changed according to the values of the most important covariates selected by the random forest algorithm and reported in the three-dimensional space (phonological awareness in *x*, performance IQ [PIQ] in *y*, and working memory in *z*). The colour fades from blue to yellow as the probability of being classified with one of the three diagnoses increases (the legends with the magnitude of the probability are displayed on the right of each plot). The arrows on the *x*, *y*, and *z*-axes show the directions in which the scores of the three variables selected by the relative VIM increased. (a) Probability of being classified as with CP without learning disorders, reaching a maximum of 0.4 as shown by the yellow peak when children obtained high scores for phonological awareness and working memory but low or median scores for PIQ. (b) Probability of being classified as with CP with learning disorders, reaching a maximum of 0.6 as shown by the yellow peak when children obtained high scores for phonological awareness, lower scores for working memory and low scores for PIQ. (c) Probability of being classified as SLD, reaching a maximum of 0.8 as shown by the yellow peak when children obtained high scores in PIQ and working memory but low scores for phonological awareness.

Associations have previously been reported between phonological awareness and literacy skills in CP in the presence of speech difficulties. In a study by Peeters et al.[21] investigating possible precursors (phonological awareness, phonological short-term memory, speech perception, speech production, and non-verbal reasoning) to early reading development in neurotypical development and in CP, phonological awareness best predicted early reading skills in the former while speech production was the most important predictor in CP. In studies recruiting adolescents with CP and typical communication skills, phonological awareness was significantly, although not highly, correlated with literacy skills.[88] More recently, Critten et al.[89] found an association between different components of phonological processing (among them phonological awareness) and reading skills in children with CP. However, as declared by the same authors, most of the non-reading participants had a slow speech that further affected both the scores in

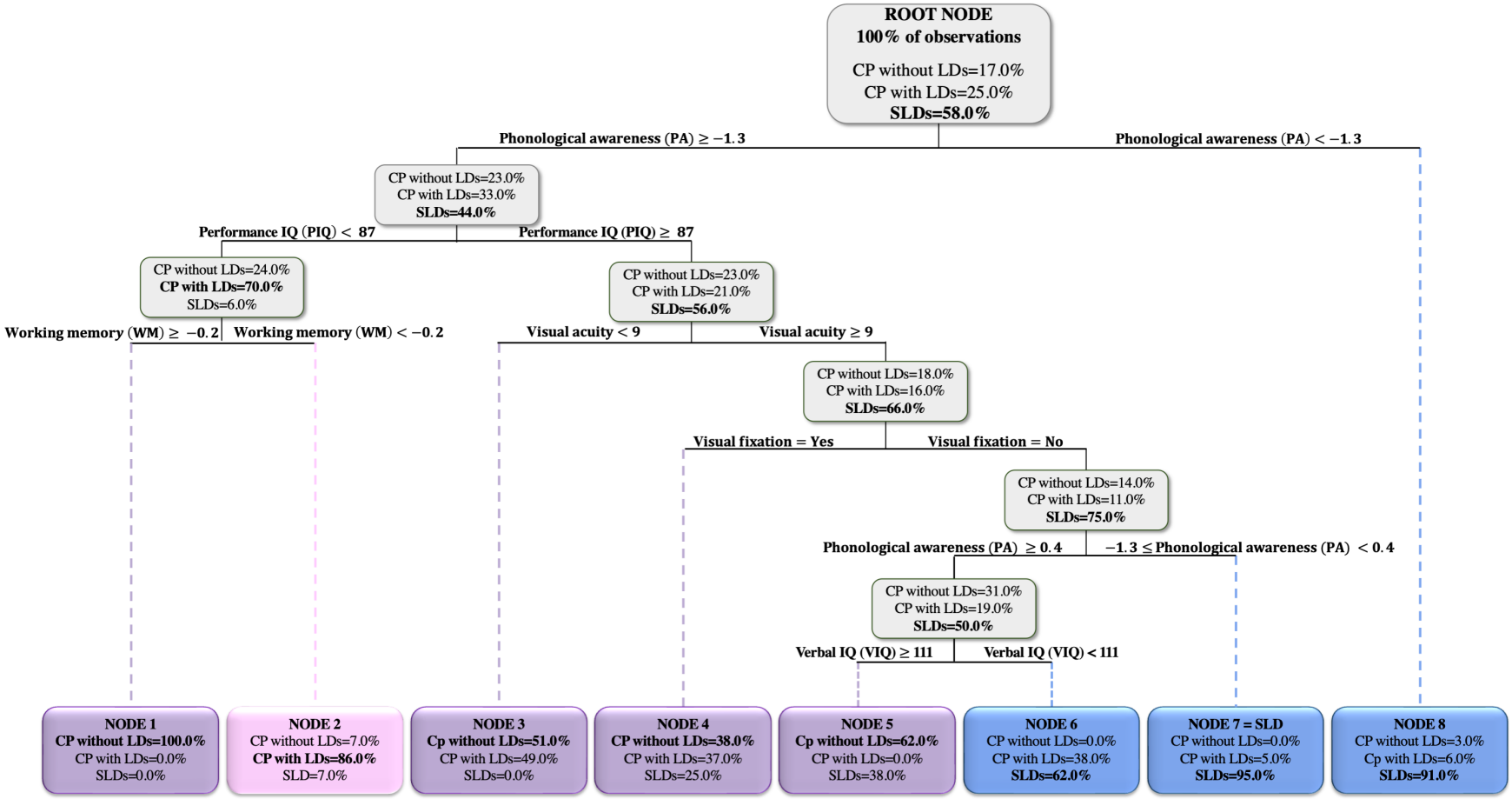

FIGURE 3 Classification tree displaying the participants grouped into homogenous clusters according to their diagnosis. At the top of the dendrogram (ROOT NODE) are all the individuals collected for the study. At each step, the algorithm divides participants into two subgroups using a splitting criterion that selects the best covariate and corresponding cut-off values. Above each split (horizontal segment), the selected variable and corresponding rules of thumb are given. The grey rectangles represent the participant clusters created by the splits and contain the corresponding percentage for each diagnosis (the most frequent is shown in bold). Pruning was used to stop tree growth. Consequently, the optimal size of the model was reached automatically. At the bottom of the dendrogram, there are eight final nodes (coloured rectangles), which are classified according to the most representative diagnosis (in bold) in the node. Purple, pink, and blue nodes signify children with cerebral palsy (CP) without learning disorders, children with CP with learning disorders, and children with specific learning disorders (SLDs) respectively. The coloured dashed lines lead from the farthest nodes (grey rectangles) to the final nodes of the tree (i.e. the homogenous group of children with regard to diagnosis). The lines are coloured violet, pink, or blue when they reach nodes classified as children with CP without learning disorders, children with CP with learning disorders, and children with SLDs respectively. The most important variables were selected at the first, second, and third level of the tree (i.e. the top of the tree structure). Abbreviations: PIQ, performance IQ; VIQ, verbal IQ.

phonological awareness and reading tasks, which were time-responsive. In the current study, only children with preserved language and speech functions were included (level I on the Viking Speech Scale[54]) to avoid the influence of speech disorders on reading performances and to enable a reliable comparison with children with SLDs. We hypothesize that, in controlling speech production skills, phonological awareness ceased to be strongly related to learning skills in children with CP.

This study has several limitations. Stratifying the study sample according to the presence of at least one learning disorder can lead to problems concerning the control of each single clinical variable with respect to reading, writing, or mathematics skills. The potential comorbidity or co-occurrence of different learning disorders was remarkably high in the current study, reaching 68.0% in children with CP with learning disorders and 85.0% in children with SLDs. A small body of research sought to explain the cognitive mechanisms underlying the co-occurrence of SLDs,[90–92] reporting a core deficit underpinning mathematics, reading, and writing skills.[90] Moreover, in the DSM-5[30] there is one overarching category of SLDs, with 'specifiers' to characterize learning difficulties in three major academic domains, that is, reading, writing, and mathematics as further proof of a unique neurodevelopmental disorder in which similar underpinning mechanisms characterize specific phenotypes. Our results approach this debate and may help point to new associations not previously reported, at least with respect to learning disorders in CP.

The limited number of recruited participants underpowered the generalization of the results. This is particularly true for children with CP because of difficulties in finding children with CP with relatively good IQ scores, only mildly impaired gross and bimanual motor skills, and normal or near-normal acuity, as previously explained by Gillies et al.[28] Moreover, the recruitment of children with no other associated neurodevelopmental disorders—a very common situation in both CP and SLDs—adds a certain degree of selection bias, thus limiting the overall generalizability of our results across the heterogeneous clinical profile of CP and SLDs. This strict recruitment served to ensure a reliable assessment of different neurocognitive functions and to facilitate a more reliable comparison between children in the groups with SLDs and CP. The age range of the participants was quite broad, although mean ages were not statistically different across the three groups. The mechanisms behind the academic abilities in the first years of primary school differed from those in secondary school and the wide age range recruited may have prevented the clear distinction of the role of each single learning skill component.

As for the procedure, not all possible explanatory factors related to learning disorders in CP were inserted in the algorithm, that is, the different executive function components according to the model by Diamond,[12] and the overall CVI signs and symptoms assessed, especially in their basic and ocular components in this study. The possible explanatory factors, inserted in the model, included previous literature data, clinical experience, and major differences between the three groups. This choice may have limited the scope for identifying further interesting associations.

An old version of the IQ test was used to minimize differences between children who were tested with the previous version of the WISC-III test. To test the influence of the WISC-III scores in the model, we computed Spearman's rank correlation coefficients between WISC-III and WISC-IV scores for a limited number of participants, retested with this newest version in the years after their study participation. The results showed that the WISC-III and WISC-IV IQs were highly correlated; consequently, we considered the use of the WISC-III or WISC-IV as substantially indifferent and thus not influencing the results of the analysis (Appendix S1).

In conclusion, data from the current study show that learning disorders are very frequent even in children with CP, classified in GMFCS levels I to III and MACS levels I and II, and with normal VIQ. It is also possible to hypothesize that the underlying factors sustaining the manifestation of learning disorders differ in children with CP compared to children with SLDs, with working memory having the most significant role in differentiating children with CP who developed a learning disorder from those who did not. Low PIQ scores helped to differentiate children with CP from children without CP; low phonological awareness scores had a prominent role in helping to identify children with SLDs.

School-age children and adolescents with CP require an overall neuropsychological profile assessment, in particular a detailed screening of learning disorders, to gain insight beyond what is surmised from IQ measurements alone. Early detection of learning disorders in children with CP would facilitate the implementation of specific learning skill-oriented programmes, which are one of the main rehabilitation tools at school age.

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DATA AVAILABILITY STATEMENT

The datasets used and analyzed during the current study are available from the corresponding author on reasonable request.

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SUPPORTING INFORMATION

The following additional material may be found online:

Appendix S1: Comparison between WISC-III and WISC-IV.
Appendix S2: Statistical analysis.
Figure S1: Study flow chart.

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