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Ten-year follow-up study found that motor-free intelligence quotient declined in children with mild-to-moderate cerebral palsy

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Abstract

Aim: This 10-year follow-up study examined cognitive change in a cohort of children with cerebral palsy from preschool to adolescence at the group and individual levels.

Methods: The Wechsler Preschool and Primary Scale of Intelligence was administered to 80 children with cerebral palsy (mean = 4 years 6 months, standard deviation = 7 months) at baseline (Time 1). At 10-year follow-up (Time 2), 28 adolescents (mean = 14 years 6 months, standard deviation = 9 months) returned for assessment with the Wechsler Intelligence Scale for Children. Motor-free intelligence quotient (IQ) scores were calculated and paired-samples t-tests and the Reliable Change Index (RCI) were used to investigate change in IQ over time.

Results: At the group level, nonverbal IQ scores declined significantly. At the individual level, RCI indicated nine and 11 children showed a clinically significant decline in Full Scale IQ (FSIQ) and nonverbal IQ scores, respectively. Decline in FSIQ was related to a history of seizures whereas decline in nonverbal IQ was associated with higher initial IQ.

Conclusion: Cognitive abilities in children with cerebral palsy evolve over time and selective deficits may not be observable until a later age, highlighting the importance of repeated cognitive assessment throughout childhood and adolescence.

KEYWORDS
assessment, cerebral palsy, cognitive ability, follow-up study, longitudinal

1 | INTRODUCTION

Cerebral palsy describes the disordered development of movement and posture causing activity limitation as a result of nonprogressive disturbances in the developing brain, occurring in about 2 per 1000 live births.[1,2] The known or presumed timing of insults may be antenatal, perinatal or post-neonatal. Many children with cerebral palsy have associated conditions, including disturbances of cognition, behaviour and epilepsy.[3] Although cerebral palsy is, by definition, related to static brain anomalies, the manifestations of the disability

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may change over time. The extent of cognitive deficits may not be identified for many years, and only become apparent when the child is required to act independently in their environment.[4] Despite this, there have been few longitudinal studies that have investigated the developmental trajectory of cognitive abilities from childhood to adolescence in young people with cerebral palsy.[5]

According to population cerebral palsy registers,[1,2] intellectual disability is reported in approximately half of children with cerebral palsy by 5 years of age. However, assessing intelligence quotient (IQ) in the context of cerebral palsy is challenging. Previous studies suggest one-third of children with cerebral palsy lack the fine motor skills required to respond to cognitive tests.[6] For those who are able to participate in cognitive testing, the use of nonverbal subtests that rely on motor coordination and speed has been shown to significantly underestimate IQ compared to performance on motor-free assessments.[6,7] IQ is an important determinant of functioning throughout the life course and has implications for participation in education,[8] self-care and independent living,[9] and employment.[10] People with cerebral palsy and IQ in the normal range are more likely to achieve the expected level of performance in these domains. However, outcomes are less favourable for adolescents with cerebral palsy and cognitive impairment, with little improvement in functioning over time.[9] Tracking cognitive change or stability through adolescence is important to guide expectations, counsel families and set realistic treatment goals to support young people with cerebral palsy during this transitional phase.

Two previous studies assessed cognitive change or stability from childhood through adolescence in individuals with cerebral palsy. Both Dahlgren Sandberg[11] and Gonzalez-Monge et al.[5] noted a significant decline in nonverbal IQ over time, with evidence of stable Full Scale IQ (FSIQ) and verbal skills[5]. However, the use of nonverbal subtests that rely on motor coordination may have led to inaccurate estimation of IQ scores in this domain[6] and contributed to the substantial heterogeneity observed in the performance of children of the same age and severity of motor impairment.[5,11] With such heterogeneity, an inherent limitation of previous studies has been the reliance on group differences to characterise change in IQ scores over time. For example, while group mean difference scores indicated temporal stability of FSIQ, individual change scores ranged from −27 to +34 IQ points.[5] Reliance on group mean differences may be misleading and is unlikely to capture the considerable range of cognitive abilities in children with cerebral palsy.

An alternative, perhaps complementary, approach is to characterise cognitive change at the individual level using the Reliable Change Index (RCI). Jacobson & Truax[12] have described this approach in detail. The RCI indicates the amount of change required on a cognitive test to be considered clinically significant and has previously been used to track individual differences on measures of symptom severity[13] and cognitive abilities[14] in children with neurodevelopmental disorders. Although most research in cerebral palsy has been reported at the group level with the intention of generalising findings to the population, it can be difficult to apply group findings to an individual in a clinical setting. The current study aimed to examine change or stability in IQ from early childhood to adolescence in

> **Key notes**
> - This study used a motor-free method to assess change in Intelligence Quotient (IQ) across childhood into adolescence in cerebral palsy.
> - Group-level analyses of motor-free IQ scores showed a decline in nonverbal intelligence over the 10 years follow-up period.
> - The presence of epilepsy and higher IQ scores at preschool age were associated with intra-individual decline in motor-free IQ scores over time.

cerebral palsy. In a 10-year longitudinal follow-up study, changes in cognitive abilities were examined at both the group and individual levels in a cohort of children with cerebral palsy.

2 | METHOD

2.1 | Participants

Children with cerebral palsy were recruited through the Victorian Cerebral Palsy Register and participated at two data collection time points. To be included in the Register, a case had to fulfil the definitional criteria for cerebral palsy.[15] Between 2008 and 2009, 80 children with cerebral palsy aged 4–6 years participated in a study by Sherwell et al[6] that examined cognitive abilities in preschool children with cerebral palsy (Time 1). In 2018, staff from the Register contacted the parents/carers of these 80 children, 67 families provided consent to be contacted by study personnel, 50 responded to a subsequent letter of invitation or follow-up phone call, and 30 were enrolled in the current study (Time 2). Reasons for nonparticipation included lack of interest in returning for the second time point, declining due to other health issues and unwillingness to travel from regional Victoria. After enrolment, one adolescent declined to participate in the cognitive assessments and another was excluded retrospectively due to missing cognitive data at Time 1. Written informed consent was obtained from the parent or caregiver.

Clinical information was obtained from the Victorian Cerebral Palsy Register and parental report. Cerebral palsy motor types were categorised as predominantly spastic or dyskinetic and topography as unilateral or bilateral.[16] Gross and fine motor function were, respectively, described using the Gross Motor Function Classification System (GMFCS)[17] and the Manual Ability Classification System[18]. The Victorian Cerebral Palsy Register defined epilepsy as a history during childhood of '*two or more afebrile seizures, excluding neonatal seizures, irrespective of seizure control*'.[19] Preterm birth was defined as <37 weeks of gestation. Low birth weight was defined as <2499 grams. Aetiology was grouped as developmental, perinatal insults in children born preterm, perinatal insults in term-born children born and post-neonatal insults before 2 years in a typically developing child.

Ethics approval was obtained from the Human Ethics Committee at La Trobe University, Melbourne, Australia (HEC17-094) and The Royal Children's Hospital Human Research Ethics Committee, Melbourne, Australia (37343A).

2.2 | Measures

The Wechsler Preschool and Primary Scale of Intelligence—Third Edition (WPPSI-III)[20] was used to assess general intellectual ability at Time 1. Verbal IQ was calculated according to standardised procedures. To minimise the impact of fine motor responses, a prorated Performance IQ was derived using Matrix Reasoning and Picture Concepts subtests.[6] A prorated FSIQ score was calculated using five motor-free subtests including Information, Vocabulary, Word Reasoning, Picture Concepts and Matrix Reasoning.[6]

The Wechsler Intelligence Scale for Children—Fifth Edition (WISC-V)[21] was administered at Time 2 using Q-interactive on iPad (Pearson Education). Motor-free IQ, Verbal Comprehension Index and Perceptual Reasoning Index scores were derived according to the method described by Piovesana et al.[22] The Motor-free IQ and indices have strong psychometric properties that are comparable to the WISC-V FSIQ and index scores[22]. All IQ and index scores have a mean of 100 (standard deviation, $SD = 15$). A factorial invariance study of the Wechsler scales and their revisions showed that the constructs measured by these scales are generally the same and consistent across versions despite differences in item-level questions and subtests.[23]

2.3 | Procedure

The procedure for Time 1 assessments has been described in detail previously.[6] Briefly, the children were assessed with a neuropsychological battery including the WPPSI-III administered by a Psychologist (SS). Time 2 assessments took place between April 2018 and September 2019 at the Murdoch Children's Research Institute, La Trobe University Psychology Clinic or during a home visit. All adolescents were assessed with a neuropsychological test battery by a Clinical Neuropsychology Registrar (MC). The participants' ability to respond to standardised neuropsychological assessment tasks was determined using basic screening measures of pointing ability and expressive language.[7] Three of the adolescents did not complete all subtests of the WISC-V required to calculate a MFIQ and were excluded from the analysis.

2.4 | Statistical analysis

Statistical analyses were conducted using SPSS version 26 (IBM Corp,). Visual inspection of stem-and-leaf plots and Q-Q plots for each variable indicated normality within acceptable ranges. For a dependent samples t-test, a priori power analysis indicated a sample of 34 participants would be required to detect a medium effect size

with 80% power when employing the traditional 0.05 criterion of statistical significance. A series of paired-samples t-tests were conducted to investigate change in IQ scores at the group level between Time 1 and Time 2.

The Reliable Change Index (RCI) was used to investigate change in IQ scores over time at the individual level. RCI was calculated on the basis of Jacobson and Truax's[12] formula:

$$RCI = \frac{X_2 - X_1}{SD1\sqrt{2(1 - r_{xx})}}$$

where X_1 is IQ at Time 1, X_2 is IQ at Time 2, $SD1$ is the standard deviation of IQ scores at Time 1, r_{xx} is the IQ test reliability. Since the WPPSI-III was administered at Time 1 and WISC-V at Time 2, r_{xx} was calculated as the average reliability of the two composite scores as they reasonably represent parallel forms of IQ assessment. Reliability coefficients were obtained from the test manual for the WPPSI-III FSIQ ($r = 0.96$) Verbal IQ ($r = 0.95$) and Performance IQ ($r = 0.91$) and from Piovesana et al[22] for the WISC-V Motor-free IQ ($r = 0.97$), Verbal Comprehension Index ($r = 0.92$) and Perceptual Reasoning Index ($r = 0.94$). RCI scores above or below ±1.96 represent a statistically and clinically significant improvement or deterioration at the level $\alpha = 0.05$.

A series of point-biserial correlations was conducted to explore relations between RCIs for FSIQ and nonverbal IQ and dichotomised clinical variables—topography of motor impairment, severity of motor impairment (GMFCS Level I, Level II-III), history of seizures (epilepsy, no epilepsy), gestational age at birth (term, preterm) and birth weight (normal, low birth weight). Pearson correlations were conducted to explore relations between IQ scores at Time 1 and RCIs for FSIQ and nonverbal IQ.

3 | RESULTS

3.1 | Participant characteristics

The final sample comprised 28 adolescents with cerebral palsy aged between 13 and 16 years. Sample characteristics are presented in Table 1. There were no significant differences between children who participated at Time 2 and those who were lost to follow-up in terms of age ($t[77] = -0.71$, $p = 0.48$) or gender distribution, (chi-square $= 0.47$, $p = 0.49$). However, there was a significant association between participation at Time 2 and severity of motor impairment as measured by the GMFCS (chi-square $= 11.88$, $p = 0.018$). Parents of children functioning at GMFCS Level IV-V were less likely to enrol their children in the study at Time 2.

The mean age at Time 1 was 4 years 6 months ($SD = 7$ months). The mean age at Time 2 was 14 years 6 months ($SD = 9$ months). Average time between assessments was 10 years and 4 days ($SD = 6$ months). Most children had unilateral spastic cerebral palsy and were predominantly classified as functioning at GMFCS Levels I-II; almost half had a history of seizures (Table 1). FSIQ, verbal- and

TABLE 1 Sample characteristics

	Participants N
Sex	
Male	21
Female	7
Motor type	
Spastic	25
Dyskinetic	3
Motor topography	
Unilateral	18
Bilateral	10
Gross motor function (GMFCS)	
Level I	11
Level II	11
Level III	4
Level IV	1
Level V	1
Manual ability (MACS)	
Level I	6
Level II	14
Level III	7
Level IV	1
History of seizures	
Yes	12
No	16
Causal group	
Developmental	4
Perinatal insult in preterm	6
Perinatal insult in term-born	16
Post-neonatally acquired	1
Unknown	1
Gestational age at birth	
Term	22
Preterm	6
Birth weight	
Normal	22
Low birth weight	6

Abbreviations: GMFCS, Gross Motor Function Classification System; MACS, Manual Ability Classification System.

nonverbal IQ scores at Time 1 were available for 23, 24 and 26 participants, respectively.

3.2 | Changes in IQ scores at the group level

Mean IQ scores are presented in Table 2. Mean FSIQ scores declined between Time 1 and Time 2; however, this change did not reach statistical significance ($t_{(22)} = 2.00$, $p = 0.059$) and effect size

TABLE 2 Mean Full Scale IQ, Verbal IQ and Nonverbal IQ scores at Time 1 and Time 2

	n	Mean	SD	Min.	Max.
Time 1					
Full Scale IQ	23	91.39	18.15	57	125
Verbal IQ	24	90.08	15.60	64	122
Nonverbal IQ	26	90.65	22.23	49	135
Time 2					
Full Scale IQ	23	85.65	18.38	48	113
Verbal IQ	24	89.83	13.60	62	116
Nonverbal IQ	26	83.73	17.57	48	112

Abbreviation: IQ = Intelligence quotient.

was moderately small ($d = 0.32$). Mean nonverbal IQ scores declined significantly between Time 1 and Time 2 ($t_{(25)} = 2.12$, $p = 0.044$) and the effect size was again moderately small ($d = 0.31$). In contrast, there was no significant change in verbal IQ scores between Time 1 and Time 2 ($t_{(23)} = 0.11$, $p = 0.917$).

3.3 | Reliable change in IQ scores at the individual level

Individual change in IQ scores between Time 1 and Time 2 is illustrated in Figure 1. Participants who demonstrated a reliable decline or improvement in IQ scores are indicated in Table 3. Based on the RCI, change of 8 or more standard score points represented a clinically significant change in FSIQ. FSIQ scores declined significantly between Time 1 and Time 2 for nine participants (39%), remained stable for 11 (48%) and improved significantly for three participants (13%).

A minimum change of 11 standard score points represented a clinically significant change in verbal IQ. Verbal IQ scores remained stable for 17 participants (71%), the majority of the sample. In contrast, four participants (17%) demonstrated a significant decline in verbal IQ whereas three (12%) showed a significant improvement.
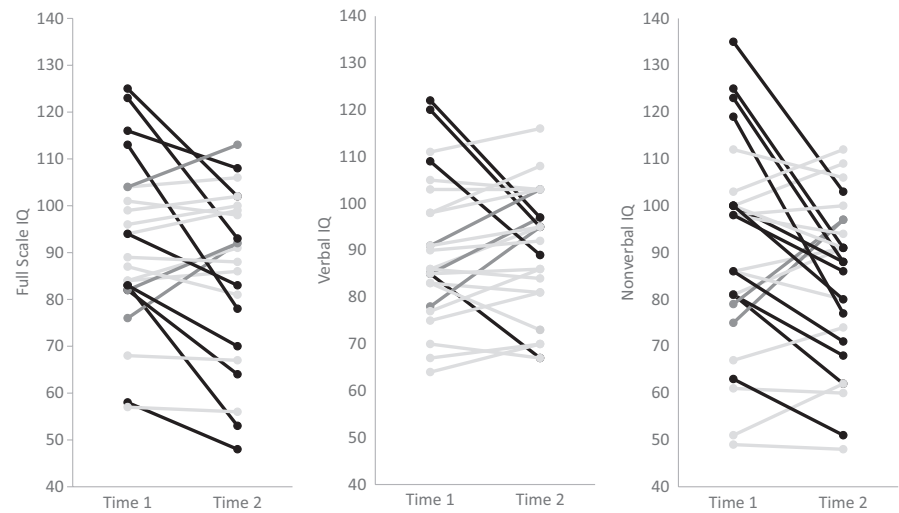
A change of at least 12 standard score points represented a clinically significant change in nonverbal IQ. Nonverbal IQ decreased significantly in 11 participants (42%). Thirteen participants (50%) showed stable nonverbal IQ and only two (8%) showed a significant improvement.

3.4 | Correlations

Reliable change in FSIQ was significantly correlated with epilepsy, $r_{pb} = -0.51$, 95% Bias-corrected and accelerated bootstrap (BCa) Confidence Interval (CI; −0.783, −0.097), $p = 0.014$. Epilepsy accounted for 26% of the variance in reliable change in FSIQ.

Reliable change in nonverbal IQ was negatively correlated with FSIQ score at Time 1, $r = -0.65$, 95% BCa CI (−0.805, −0.361),

$p = 0.001$, as well as nonverbal IQ score at Time 1, $r = -0.65$, 95% BCa CI ($-0.812, -0.371$), $p = 0.007$. That is, higher FSIQ or nonverbal IQ scores at Time 1 were associated with greater decline as indicated by the RCI. FSIQ or nonverbal IQ scores at Time 1 accounted for 42% of the variance in reliable change of nonverbal IQ at Time 2. There were no other significant correlations.

4 | DISCUSSION

The current study was the longest known follow-up study to investigate change or stability in cognitive abilities from preschool age to adolescence in children with mild-to-moderate cerebral palsy. At the group level, results showed a significant decline in nonverbal IQ scores over the 10-year period, which drove a marginally significant decline in FSIQ, in the face of stable verbal abilities. At the individual level, reliable change scores indicated 39% and 42% of children showed a clinically significant decrease in FSIQ and nonverbal IQ scores, respectively. Decline in FSIQ was related to a history of seizures whereas decline in nonverbal IQ was associated with higher initial IQ. These findings suggest a slower-than-normal rate of cognitive development through adolescence in many young people with cerebral palsy.

Our findings highlighted a significant decline in nonverbal IQ over time in this group of children with cerebral palsy. This is consistent with a previous longitudinal study, which reported a selective decline in nonverbal IQ scores over a 7-year period in young people with cerebral palsy.[5] Moreover, the RCI in the current study indicated that children with higher initial IQ scores were more vulnerable to decline over time. It is important to emphasise that the decline in IQ scores does not necessarily represent a regression in cognitive abilities but rather a reduced rate of skill acquisition compared to the standardised sample. As such, the current results suggest children with cerebral palsy demonstrate age-appropriate development of nonverbal skills until the age of 5 years, followed by slower progress during mid-childhood or adolescence compared with age peers. This advance-plateau

trajectory of development has previously been described in the context of Down syndrome[24]; however, in cerebral palsy it appears that this plateau is more discernible for nonverbal abilities. According to the cognitive crowding hypothesis[25], cognitive development following early brain injury occurs in a hierarchy whereby verbal abilities are prioritised over nonverbal. Although elementary visuospatial functions tend to be preserved, more complex nonverbal abilities are compromised. To maintain the same IQ score over time, the child must continuously rise to the increasing demands of age-related functioning. Through mid-childhood and beyond, IQ tests begin to demand abstract reasoning skills. As such, nonverbal impairments in children with cerebral palsy are more clearly identifiable at an age at which nonverbal reasoning tests become more demanding.

Similarly, our findings indicated children with cerebral palsy and a history of seizures showed slower-than-expected development of FSIQ over time. This is consistent with previous studies that suggest seizures have a deleterious effect on cognition.[26,27] Indeed, there is an interplay between age, seizures and cognitive abilities—that is, an adolescent with cerebral palsy and seizures is more likely to have lower IQ than a younger child with cerebral palsy and seizures.[26] This contention is supported by MRI studies, which have shown that increasing epilepsy duration is associated with reduced efficiency of brain networks that support higher order cognitive functions[28,29] and in turn, is associated with IQ decline.[28] Therefore, seizures appear to contribute to a reduced rate of cognitive skill acquisition in children with cerebral palsy, which manifests as a decline in FSIQ scores when the child is expected to respond to increasingly complex demands.

Some limitations of the current study are acknowledged. Firstly, our study was underpowered and nonambulant children were underrepresented. Only 28 out of the 80 children with cerebral palsy enrolled in the first study phase as pre-schoolers were included in the assessment of their motor-free IQ as adolescents. The dropout rate was clearly substantial, however, was not surprising considering the long gap between the two assessments and other scheduling reasons. We attempted to mitigate this limitation by conducting

TABLE 3 Full Scale IQ, Verbal IQ and Nonverbal IQ Scores for Each Participant at Time 1 and Time 2

Participant	Time 1			Time 2		
	FSIQ	Verbal	Nonverbal	FSIQ	Verbal	Nonverbal
1	83	85	81	53*	67*	62*
2	104	105	100	106	103	109
3	-	-	67	85	89	74
4	84	83	86	86	81	91
5	76	78	75	92+	95+	97+
6	82	83	81	64*	73	68*
7	84	86	81	91	95	91
8	82	85	79	92+	97+	97+
9	87	86	86	81	84	80
10	123	122	125	93*	97*	91*
11	58	67	49	48*	70	48
12	116	111	123	108*	116	88*
13	-	-	100	81	78	88*
14	125	120	135	102*	95*	103*
15	101	103	98	98	103	94
16	83	77	86	70*	86	71*
17	57	64	51	56	70	62
18	68	75	61	67	81	60
19	113	109	119	78*	89*	77*
20	94	91	98	99	103+	100
21	94	90	100	83*	92	91
22	72	70	81	-	67	-
23	104	98	112	113+	103	106
24	96	91	103	100	95	112
25	99	98	100	102	108	80*
26	-	-	-	-	62	51
27	-	-	63	50	73	51*
28	89	85	98	88	86	86*

Abbreviation: FSIQ, Full Scale Intelligence Quotient.
*indicates a significant decline in scores between Time 1 and Time 2.
+indicates a significant increase in scores between Time 1 and Time 2.
-Unable to calculate score.

reliable change statistics, which provide useful change pattern data in small samples while taking into account test reliability and clinical significance. Nevertheless, given previous reports of an average decline of 23 IQ points by the age of 12 in children with severe motor impairment,[11] our findings may underestimate the magnitude of change from childhood to adolescence in cerebral palsy due to selection bias.

A second limitation was the broad categorisation of epilepsy; and therefore, we were unable to disentangle the impact of epilepsy-related variables including seizure type, frequency or antiepileptic medication on cognition over time. This may account for variation in cognitive development in adolescents with cerebral palsy and seizures. Although epilepsy was associated with decline in FSIQ, not all children with seizures showed a decline, and in fact, one showed a clinically significant improvement.

Despite these limitations, the current study highlights a range of implications for clinical practice. First, cognitive abilities in children with cerebral palsy evolve over time and selective deficits may not be observable until a later age, highlighting the importance of repeated cognitive assessment throughout childhood and adolescence. Based on our findings, a cognitive assessment conducted at preschool age is unlikely to accurately inform the needs of a child with cerebral palsy during secondary school. Second, average or above average IQ scores at preschool age should not be taken as a sign that the child will continue to be free of later cognitive impairments. In fact, our results suggest the contrary—high initial IQ scores were associated with later decline suggesting children may grow into a deficit. We suggest that clinicians use this information to prepare parents and guide expectations for future functioning. Finally, the study emphasises the impact of seizures

on cognition in children with cerebral palsy and the need for close monitoring throughout development.

5 | CONCLUSION

The current study is the first to investigate cognitive change or stability over a 10-year period in children with mild-to-moderate cerebral palsy using motor-free methods for assessing IQ as well as group- and individual-level analyses to characterise change over time. While some participants showed cognitive stability over time, many showed clinically significant decline in FSIQ and nonverbal IQ scores, and few showed clinically significant improvement. The findings highlight the importance of ongoing monitoring and individual assessment of cognitive skills throughout development to guide expectations and need for support. Future research may consider environmental and social factors for their potential contribution to cognitive development over time across childhood and adolescence in cerebral palsy.

CONFLICT OF INTEREST
The authors have no conflicts of interest to declare.

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