Cognitive Abilities in Adolescents with Cerebral Palsy

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List of Abbreviations

ACPR	Australian Cerebral Palsy Register
CHC	Cattell-Horn-Carroll Theory of Cognitive Abilities
CP	Cerebral palsy
fMRI	Functional Magnetic Resonance Imaging
FSIQ	Full Scale Intelligence Quotient
g	General Ability Factor
Gc	Crystallized Intelligence
Gf	Fluid Intelligence
GMFCS	Gross Motor Function Classification System
IQ	Intelligence Quotient
LBW	Low Birth Weight
MACS	Manual Ability Classification System
MFIQ	Motor Free IQ
MRI	Magnetic Resonance Imaging
PRI	Perceptual Reasoning Index
RCI	Reliable Change Index
SCPE	Surveillance of Cerebral Palsy in Europe
SGA	Small for Gestational Age
VCI	Verbal Comprehension Index
VCPR	Victorian Cerebral Palsy Register
WISC-V	Wechsler Intelligence Scale for Children – Fifth edition
WMI	Working Memory Index
WPPSI-III	Wechsler Preschool and Primary Scale of Intelligence - Third edition

Abstract

Cognitive impairment is common in children with Cerebral Palsy (CP), and some deficits may only become apparent during adolescence in the context of increasing demands of age-appropriate functioning. However, cognitive assessment is challenging in the context of motor impairment which can be a significant barrier to participation on cognitive tests. Chapter 1 provides an introduction to CP followed by a literature review of cognitive abilities in CP while considering the impact of early brain injury to brain development, intelligence theories, and the impact of motor impairment on cognitive tests in Chapter 2. Subsequently, findings from three related research studies are presented which aim to characterise cognitive abilities in a cohort of adolescents with mild-to-moderate CP. In Chapter 3, standardised administration of the Wechsler Intelligence Scale for Children (Fifth edition; WISC-V) – the most commonly used intelligence test – was shown to underestimate the Intelligence Quotient (IQ) of adolescents with CP compared to short-forms of the WISC-V which minimised motor demands. Chapter 4 characterised a motor-free cognitive profile. Results showed that motor-free IQ scores fell significantly below the normative data and rates of borderline and impaired cognitive abilities were higher in the CP group. A strength in verbal abilities and relatively weaker non-verbal and working memory skills were also noted. Severity of motor impairment and small for gestational age predicted cognitive abilities across domains while seizure history was related to lower verbal abilities. Chapter 5 describes a 10-year follow-up of a cohort of adolescents who had a previous cognitive assessment during early childhood. Results showed a significant decline in Non-verbal IQ scores over time, as well as a marginally significant decline in FSIQ, in the context of stable verbal abilities. At the individual level, reliable change scores indicated 39 to 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 while decline in Nonverbal IQ was associated with higher initial IQ. General discussion in Chapter 6 highlights the importance of ongoing monitoring of cognitive abilities in adolescents with CP using methods which accommodate their motor impairment, particularly for those with more severe motor impairment, born small for gestational age, a history of seizures, and higher initial IQ scores.

List of Manuscripts

Chapter 3

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Chapter 4

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Chapter 5

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Statement of Authorship

Except where reference is made in the text of the thesis, this thesis contains no material published elsewhere or extracted in whole or in part from a thesis accepted for the award of any other degree or diploma. No other person's work has been used without due acknowledgement in the main text of the thesis. This thesis has not been submitted for the award of any degree or diploma in any other tertiary institution.

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Monika Coceski: Conceptualisation, Method, Investigation, Formal analysis, Writing – Original Draft, Writing – Review & Editing, Visualization. **Darren R. Hocking:** Supervision, Writing – Review & Editing **Hisham M. Abu-Rayya:** Formal analysis, Supervision, Writing – Review & Editing **Sarah Sherwell:** Conceptualization, Methodology **Susan M. Reid:** Conceptualization **Dinah S. Reddihough:** Conceptualization **Jacquie Wrennall:** Supervision **Robyn Stargatt:** Conceptualization, Methodology, Supervision, Writing – Review & Editing.

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Chapter 1: Introduction to Cerebral Palsy

Cerebral Palsy (CP) is the most common physical disability in childhood, affecting approximately two in 1000 live births in developed countries (Australian Cerebral Palsy Register, 2018; Surveillance of Cerebral Palsy in Europe, 2000). CP has been well recognised as a physically disabling neurodevelopmental disorder by health professionals and the general public since it was first described in the latter half of the 19th century (Little, 1862). However, as a neurodevelopmental disorder diagnosed based on clinical signs and symptoms rather than underlying aetiology, the term 'CP' has proved challenging to define, with multiple interpretations available in the literature (Bax, 1964; MacKeith & Polani, 1959; Mutch et al., 1992).

Historically, definitions have focused on the motor disorder of CP, conceptualised by Bax (1964) as "a disorder of movement and posture due to a defect or lesion of the immature brain" (p. 295). However, such a simple definition was rendered unsatisfactory due to the heterogeneity of disorders covered by CP, numerous aetiologies, and emerging understanding of development in infants with early brain damage. As a result, CP was defined by Mutch et al. (1992) as "an umbrella term covering a group of non-progressive, but often changing motor impairment syndromes secondary to lesions or anomalies of the brain arising in the early stages of its development" (p. 549). Although CP is a permanent disorder, it is not unchanging, as various manifestations of atypical brain development appear more prominent in different people and at different stages of life (Rosenbaum et al., 2007). In addition, an increased understanding that developmental motor impairment is often associated with a range of other disabilities required this definition to be expanded to capture the heterogeneity of CP.

The most recent consensus definition emphasises the motor disorder characteristic of CP, although recognises that other cognitive and behavioural disorders can accompany it.

> CP describes a group of permanent disorders of the development of movement and posture, causing activity limitation, that are attributed to non-progressive disturbances that occur in the developing fetal or infant brain. The motor disorders of CP are often accompanied by disturbances of sensation, perception, cognition, communication, and behaviour, epilepsy, and secondary musculoskeletal problems (Rosenbaum et al., 2007, p. 9).

Recognition of the impairments commonly associated with CP have emphasised a move toward the multidimensional approach and management of CP.

CP registers collect population-based data on children with CP across several centres worldwide (e.g. the Surveillance of CP in Europe (SCPE), the Australian Cerebral Palsy Register (ACPR)). They aim to monitor the prevalence of CP over time and identify interventions, causal pathways, and preventative strategies (Australian Cerebral Palsy Register, 2018). CP registers draw on various references when considering the definition of CP, and this has proven problematic in terms of agreement on inclusion criteria and comparing data across centres. The ACPR adopted the approach recommended by the SCPE, which outlined five key inclusion criteria common to the definitions published by Bax (1964), Rosenbaum et al. (2007) and Mutch et al. (1992). The criteria state that CP:

> 1) is an umbrella term for a group of disorders; 2) is a condition that is permanent but not unchanging; 3) involves a disorder of movement and/or posture and of motor function; 4) is due to a non-progressive interference, lesion, or abnormality, and 5) the interference, lesion, or abnormality originates in the immature brain (Smithers-Sheedy et al., 2014).

The various definitions of CP cover a wide range of clinical presentations and degree of activity limitation. Therefore, it is necessary to further categorise individuals with CP to describe the nature and severity of the disorder, and to provide information regarding their current and future care needs.

Classifications

While CP is characterised by an abnormal pattern of movement and posture, there are three major classifications that can be applied to describe the nature and severity of the motor impairment.

Motor Type

The motor impairments of CP are classified into four main subtypes: spastic, dyskinetic, ataxic, and hypotonic. Spastic CP is the most common subtype, accounting for approximately 85% of CP cases in Australia (Australian Cerebral Palsy Register, 2018). It is characterised by increased muscle tone and pathological reflexes including hyperreflexia or pyramidal signs (Cans et al., 2007). Hypertonic muscles display a velocity-dependent resistance to stretch (McIntyre et al., 2011). Spasticity can result in contracture or dislocation of the joints, pathological posturing of the limbs (e.g. legs in 'scissored' position), and pain.

Dyskinetic CP accounts for approximately 6% of all CP cases in Australia (Australian Cerebral Palsy Register, 2018) and is characterised by involuntary, recurring, and occasionally stereotyped movements and fluctuating muscle tone (Cans et al., 2007). Dyskinetic CP can be either dystonic or athetoic. Dystonic CP is hypokinetic with abnormal twisting postures or repetitive movements and hypertonia. In contrast, athetoid CP is hypotonic and hyperkinetic, with writhing movements that can co-occur with chorea.

Ataxic CP, accounting for approximately 4% of cases, involves a loss of coordination and problems with balance and depth perception whereby movements are

poorly organised in terms of force, rhythm and accuracy. Ataxic gait and tremor are typical features (Australian Cerebral Palsy Register, 2018; Cans et al., 2007).

The ACPR recognises hypotonic CP as the fourth and least common subtype of CP, characterised by diminished muscle tone in the trunk and limbs without other signs of motor impairment (McIntyre et al., 2011). However, Badawi et al. (1998) and Cans et al. (2007) argue that hypotonia is commonly associated with intellectual disability and preterm birth, and it would be over-inclusive to diagnose hypotonia in isolation as CP.

A proportion of individuals with CP present with 'mixed' motor symptoms. In these cases, the motor type should be classified according to the dominant clinical feature, with any additional abnormalities in movement or tone listed as secondary types—that is, a predominantly spastic motor pattern with dyskinesia (Delacy & Reid, 2016).

Topography

Spastic CP can be further subdivided based on the anatomical distribution of motor impairment. Traditionally, the terms quadriplegia, diplegia, and hemiplegia have been used extensively in research and clinical practice to describe the topographical distribution of the motor disorder. Quadriplegia refers to spasticity in all four limbs whereby the effect on the upper limbs is usually equal to, or more than, that on the lower limbs. Trunk and orofacial involvement is also expected. The sparing of one limb, although rare, is known as triplegia. Diplegia is characterised by spasticity in both legs with greater involvement of the lower limbs than the upper limbs. Hemiplegia is the involvement of the arm and leg on one side of the body, typically with greater involvement of the arm than the leg. The term monoplegia describes the involvement of only one limb.

However, the use of these classifications was criticised due to inconsistent application and low inter-rater reliability. In particular, the distinction between diplegia and quadriplegia was unclear and there was wide variation in the use of these terms

(Surveillance of Cerebral Palsy in Europe, 2000). For example, the prevalence of spastic diplegia as a proportion of all CP cases recorded across European CP registers ranged from 13 to 55%, almost a fourfold difference. Similarly, analysis of epidemiological studies revealed the prevalence of diplegia ranged from 19-62% of spastic CP cases across studies. The authors argued that such a large difference was unlikely to be a real one, and was explained by coding differences (Colver & Sethumadhavan, 2003). The main criteria used to distinguish diplegia from quadriplegia was a subjective rating of the level of involvement of the arms, ranging from 'none' to 'less than that of the legs'. The validity of this comparison was questioned due to the structural and functional differences between the upper and lower limbs, and additionally, the classification failed to consider the involvement of the trunk and oro-pharynx. Moreover, Gorter et al. (2004) reported very small differences and large overlap in functional ability of children with CP when they were grouped by topographical distribution (diplegia, triplegia, quadriplegia). They suggested that the use of these terms did not provide additional prognostic information and was not useful for intervention planning.

Alternatively, the SCPE proposed a simpler approach to the classification of spastic CP that differentiates between unilateral CP (which covers the terms hemiplegia and monoplegia) versus bilateral CP (which subsumes the terms diplegia, triplegia, quadriplegia; Cans et al., 2007). The authors acknowledge that this distinction may still be blurred by some motor involvement on the contralateral side in unilateral CP, or asymmetrical involvement in bilateral CP. Nonetheless, classification based on this distinction has shown good reliability and has been recommended for use in conjunction with standardised ratings of functional impairment in the arms and legs (Krägeloh-Mann & Cans, 2009).

Severity

The functional impairment resultant of the involvement of the upper and lower limbs can be separately classified using objective functional scales to complement the diagnosis of CP and its subtypes (Rosenbaum et al., 2007). The Gross Motor Function Classification System (GMFCS) is a five-level ordinal grading system (where Level V is the most severe motor impairment) based on self-initiated movement with particular emphasis on sitting and walking. Distinctions between levels of gross motor function are based on functional limitations, the need for assistive technology including mobility devices and wheeled mobility, and the quality of movement (Palisano et al., 1997). The GMFCS has been shown to be stable between less than 2 and 12 years of age and interrater reliability was high (Wood & Rosenbaum, 2000). It is widely used in research and clinical settings to describe the level of motor impairment in children with CP.

Similarly, the severity of fine motor impairment can be classified using The Manual Ability Classification System (MACS). MACS provides a functional description of the child's ability to use their hands to handle objects in age-appropriate daily activities, such as eating, dressing, playing, and drawing or writing (Eliasson et al., 2006). MACS describes five levels based on the child's overall ability and does not classify the function of each hand separately. Although the MACS has received some criticism for not taking into account possible asymmetry in hand functions (Cans et al., 2007), it has been recommended for use as a common data element in clinical research studies due to its strong psychometric properties (Schiariti et al., 2018).

Both the GMFCS and MACS aim to characterise the child's usual performance in the home, school and community settings rather than to describe their best capacity or potential for improvement. The GMFCS and/or MACS coupled with the CP subtype provide clinicians with an overall impression of a child's motor functioning. In addition,

other classification instruments have been developed to characterise the impact of CP on other aspects of the child's functioning and ability to participate in daily activities including the Viking Speech Scale (Pennington et al., 2013), the Communication Function Classification System (Hidecker et al., 2011), the Eating and Drinking Ability Classification System (Sellers et al., 2014), and Visual Function Classification System (Baranello et al., 2020). These classification systems provide complementary information, however level of function in one system seldom predicts classification in another system (Hidecker et al., 2012). Nevertheless, in combination these classification systems can provide a comprehensive picture of the child's functional profile.

Causal Pathways and Risk Factors

CP is generally understood to be a result of brain injury in the early developmental period; however, for many cases the causal pathway is not well understood. According to the Australian Cerebral Palsy Register (2018), the vast majority of CP cases are pre- or perinatally acquired, and only 5.9% of cases are due to brain injury during the post-neonatal period before two years of age. Classically, CP was considered the result of birth asphyxia—that is, hypoxic-ischaemic events during delivery. However, this notion was challenged by population-based studies and growing literature that indicated only a small minority of CP cases were associated with sentinel events during birth (Blair & Stanley, 1988; Strijbis et al., 2006). Rather, CP is recognised as the likely result of multiple interacting factors rather than of a single cause (Nelson & Chang, 2008).

A decline in the overall prevalence of CP has been observed over the past 20-30 years (Hollung et al., 2018; Reid et al., 2016). For example, the prevalence of CP in Norway declined from 2.62 per 1000 live births in 1999 to 1.89 in 2010 (Hollung et al., 2018). In addition, the authors found a decrease in the proportion of children with severe motor impairment, communication impairments, epilepsy and intellectual disability. The

authors contend these findings may be explained by improvements in obstetric and neonatal care during the study period which included fewer pregnancies with pre-eclampsia, multiple pregnancies, children born preterm, and perinatal deaths (Hollung et al., 2018).

Despite the decline in CP prevalence overall, two risk factors that remain commonly associated with CP include gestational age and birthweight. Data from the Australian Cerebral Palsy Register (2018) indicates that 43% of children with CP were born preterm (<37 weeks gestation) compared to the Australian population where only 6-8% of live births were born preterm. Moreover, prevalence of CP increases nonlinearly as gestational age at birth decreases (Hirvonen et al., 2014). A systematic review of population-based studies by Oskoui et al. (2013) indicated the prevalence was highest at 112 per 1000 live births among children born extremely preterm (<28 weeks gestation).

Similarly, combined data from the Australian states and territories indicated 43% of children with CP were born with low birth weight (LBW; <2500g) compared to 6.2% of live births in the Australian population (Australian Cerebral Palsy Register, 2018). A European population-based study has shown a decline in the prevalence of CP in children born with LBW between 1980 and 2003, which likely to be related to improvements in peri- and neonatal care over this period (Sellier et al., 2016). Nonetheless, children born with LBW remain at increased risk of CP compared to children born with normal birthweight (Oskoui et al., 2013; Sellier et al., 2016). CP prevalence is highest in children weighing <1500g at 59 per 1000 live births and lowest in those weighing >2500g (1.33 per 1000 live births; Oskoui et al., 2013).

Although infants born preterm or LBW are at increased risk of CP, it should be acknowledged that this accounts for less than half of all CP cases. In most cases, multiple

risk factors act in combination to culminate in CP. Other prenatal risk factors include congenital malformations, intrauterine growth restriction, placental abnormalities, maternal disease during pregnancy (e.g. cardiorespiratory disease, seizures), hypertension and pre-eclampsia, infection, bleeding during the second and third trimester, and multiple pregnancy. Intrapartum risk factors include birth asphyxia, meconium aspiration, instrumental deliveries, and breech delivery (McIntyre et al., 2013). Post-neonatal factors include seizures, respiratory distress, hypoglycaemia, infection, cerebrovascular accidents and head injury (McIntyre et al., 2013). Brain abnormalities associated with CP can arise at different times during brain development. The same cause may give rise to different patterns of impairment depending on the timing of the interference with brain development.

Neuroimaging

Neuroimaging studies show abnormal findings in more than 80% of children with CP (Himmelmann et al., 2017; Korzeniewski et al., 2007; Reid et al., 2014). Diagnostic guidelines recommend magnetic resonance imaging (MRI) to help establish aetiology and prognosis in children with CP (Ashwal et al., 2004). MRI can aid understanding of the pathogenesis of the disorder, timing of the insult, as well as provide insight to the structure-function relationship of the brain. Himmelmann et al. (2017) developed a classification system for MRI findings of children with CP which recognises five main pathogenic patterns: maldevelopments, predominant white matter injury, predominant grey matter injury, miscellaneous, and normal.

Brain maldevelopments occur as a result of disordered cortical formation, i.e. disordered proliferation, migration, or organisation of neuronal precursor cells and neurons during cortical neurogenesis in the first or second trimester of pregnancy (Marret et al., 2013). Maldevelopments were identified in 9% of CP cases overall, but occurred

more often in children born preterm (Krägeloh-Mann & Horber, 2007). CP subtypes frequently associated with maldevelopments are severe forms of bilateral and unilateral spastic CP. While reports of MRI findings in ataxic CP tend to be rare, two studies indicate 18% of cases are associated with maldevelopments on MRI (Krägeloh-Mann & Horber, 2007; Reid et al., 2014).

Predominant white matter injury is by far the most common pathogenic pattern identified on neuroimaging, reported in 56% of CP cases. It is especially common in children with CP born preterm, accounting for 90% of MRI findings in this group (Krägeloh-Mann & Horber, 2007). Predominant white matter injury encompasses periventricular leukomalacia and sequelae of intraventricular haemorrhage or periventricular haemorrhage. Predominant white matter injury typically occurs in the third trimester or peri/neonatally and tends to be associated with milder forms of spastic CP (Krägeloh-Mann & Cans, 2009).

Predominant grey matter injury includes basal ganglia and thalamus lesions, cortico-subcortical lesions or arterial infarctions which typically occur late in the third trimester of pregnancy. As such, this pattern is more commonly identified in children born at term and tends to be associated with severe bilateral spastic CP or dyskinetic CP (Himmelmann & Uvebrant, 2011; Krägeloh-Mann & Horber, 2007).

The ‘miscellaneous’ classification as defined by Himmelmann et al. (2017) encompasses MRI findings including cerebellar atrophy, cerebral atrophy, delayed myelination, calcifications, ventriculomegaly, brainstem lesions, and sequelae of haemorrhage not covered under predominant white matter injury. A review of population-based MRI studies indicated miscellaneous abnormalities were detected in 4-23% of cases and reported for children with all CP subtypes.

Previous studies indicate that 11-33% of people with CP can present with normal MRI findings (Benini et al., 2013; Krägeloh-Mann & Horber, 2007; Reid et al., 2014). Normal MRI findings were more likely in children with mild motor impairment functioning at GMFCS Level I or II (Himmelmann & Uvebrant, 2011) or ataxic CP (Krägeloh-Mann & Horber, 2007; Reid et al., 2014). There was no association between normal MRI findings and term or preterm birth (Reid et al., 2014). It has been suggested that in the case of normal-appearing findings on MRI, more refined imaging techniques that evaluate the integrity of white matter tracts like diffusion tensor imaging may detect alterations in corticospinal and sensory pathways (Benini et al., 2013). Furthermore, a genetic cause is suspected in those who have normal imaging in the absence of risk factors or hypoxic ischaemic events (Benini et al., 2013; Fahey et al., 2017).

Genetics

Recent evidence suggests that 10-30% of CP cases have a genetic aetiology (MacLennan et al., 2015). While a detailed review of the genetic basis of CP is beyond the scope of this thesis, a brief overview is provided in this section. To date, research has indicated that there is no single 'CP gene' but rather that CP is more likely to involve complex genetic pathways similar to other neurodevelopmental disorders including autism spectrum disorder. Four main types of genetic variation contribute to CP pathogenesis including individual gene mutations, epigenetic adaption of function, copy number variations, and mitochondrial DNA (Fahey et al., 2017). The effects of the genetic mutation can vary depending on the type, environmental factors, and interaction with other genes. In some cases, a single genetic mutation may be sufficient to cause CP while, in other cases, CP is the cumulative result of multiple, less severe genetic mutations and/or the presence of environmental factors (e.g. ischemia). Given evidence

indicates a substantial proportion of CP cases are due to genetic mutations, this may provide targets for treatment and prevention.

There has been some discussion in the literature as to whether the clinical diagnosis of CP should be retained if an underlying genetic cause is identified. Some of the genetic variants identified in CP have been previously linked to rare, genetic neurodevelopmental disorders (McMichael et al., 2015) and therefore raises the possibility that the clinical diagnosis of CP should be reclassified in light of a genetic diagnosis. However, a clinical consensus statement from MacLennan et al. (2019) indicated the diagnosis of CP should not be changed despite the identification of a genetic or nongenetic cause. As a disorder that has many known and unknown causal pathways, the authors argue that identification of genetic aetiology provides a more nuanced understanding of the disease pathology and thus provides targets for treatment, prevention, and genetic counselling.

Associated impairments

In addition to the disorder of movement and posture which is the core feature of CP, the Rosenbaum et al. (2007) definition recognises that people with CP often have associated impairments of 'sensation, perception, cognition, communication, behaviour, epilepsy, and secondary musculoskeletal problems' (p. 9). According to a meta-analysis of 30 population-based studies by Novak et al. (2012), moderate to high quality evidence indicated:

> Among children with CP, 3 in 4 were in pain; 1 in 2 had an intellectual disability, 1 in 3 could not walk; 1 in 3 had hip displacement, 1 in 4 could not talk, 1 in 4 had epilepsy; 1 in 4 had behaviour disorder; 1 in 4 had bladder control problems; 1 in

> 5 had sleep disorder; 1 in 5 dribbled; 1 in 10 were blind; 1 in 15 were tube fed; and 1 in 25 were deaf (p. 20).

Rates of associated impairments were strongly related to severity of motor impairment—that is, children with severe motor impairment were more likely to have co-existing impairments. However, as discussed later in Chapter 2, the correspondence between CP severity and functional outcomes is not a one-to-one relationship as there is substantial heterogeneity among children with the same level of motor impairment (Stadskleiv et al., 2018). In contrast, pain was common at all levels of motor impairment while behavioural problems were more likely to occur in children with mild motor impairment.

In addition, the motor disorder of CP often impacts communication. An Australian Cerebral Palsy Register study found that 61% of cases on the register were considered to have speech impairment at 5 years of age, and 24% were non-verbal (Delacy & Reid, 2016). In contrast, an Icelandic population-based study of 152 preschool-aged children with CP found 84% communicated verbally while 16% were nonverbal (Sigurdardottir & Vik, 2011). However, among the 128 children who communicated verbally, 15% were able to produce only one-word utterances while the remainder were able to communicate in sentences (Sigurdardottir & Vik, 2011). Non-verbal children were more likely to have bilateral CP, dyskinetic CP, severe motor impairment and other associated disturbances including epilepsy, intellectual disability, and visual impairment (Sigurdardottir & Vik, 2011).

Visual perception is often impaired in children with CP either due to the initial brain insult or involvement of peripheral visual structures. A systematic review of 15 studies published between 1990 and 2011 found that almost half of children with CP had visual impairment (Ego et al., 2015). While the degree of visual impairment was found to

be influenced by severity of the brain injury, the findings were inconsistent across studies regarding the impact of CP subtype, intellectual disability, seizure history, or neuro-ophthalmological deficits (Ego et al., 2015).

Summary

Although CP is primarily considered a disorder of movement and posture, this chapter has outlined the inherent complexity and heterogeneity of the condition. As an umbrella term that subsumes a variety of aetiologies, motor types, levels of severity, and associated conditions, it can be difficult to predict the functional implications of a CP diagnosis. Similar to the wide range of motor impairment seen in CP, cognitive abilities can vary widely as a primary consequence of the initial brain injury or secondary effect of developmental delay as a result of functional limitations. The following chapter will examine the impact of early brain injury on cognitive abilities and longitudinal cognitive outcomes in children and adolescents with CP, and discuss limitations of commonly used assessments of intellectual ability in this population.

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Chapter 2: Cognitive Abilities in Cerebral Palsy

This chapter provides a brief review of the impact of early injury on brain development and outlines theories of cognitive abilities prior to a review of the literature on the cognitive abilities and long-term cognitive outcomes in children and adolescents with CP. In addition, the inherent limitations of cognitive assessment in children with varying degrees of motor impairment and CP are explored and potential solutions are considered.

Impact of Early Injury to the Developing Brain

The development of the human brain is a complex and protracted process that begins early in gestation and continues well into early adulthood (Stiles & Jernigan, 2010). The prenatal period is primarily concerned with the gross morphological development of the brain and central nervous system including neural networks that eventually subserve the development of high-level cognitive functions. The postnatal period is characterised by rapid dendritic growth and synaptogenesis in the first 2-3 years, followed by a gradual decrease in connectivity through selective "pruning" over the next two decades. Pruning provides an opportunity for brain development to be influenced by experience and environmental factors (Stiles & Jernigan, 2010). The initial overproduction of synapses gives rise to the developing brain's capacity for adaptation and may provide scope for good recovery following brain insult in the prenatal or postnatal period.

Early plasticity is based on the 'Kennard Principle' which posits the developing brain shows an advantage in recovery from early insult, and is associated with better outcome than similar brain injuries later in life (Kennard, 1942; Kolb et al., 2011; Kolb & Gibb, 2007). Proponents of early plasticity have argued that at birth, the two cerebral hemispheres are 'equipotential' for cognitive functions and they become increasingly specialised over the course of childhood and adolescence to resemble the adult model of

hemispheric specialisation (Lennenberg, 1967). Prior support for plasticity was drawn from a classic study by Basser (1962) who investigated the impact of hemispherectomy on speech function in 35 children with intractable epilepsy. Basser (1962) found that speech was unaffected in the majority of cases regardless of the side of lesion and subsequent hemispherectomy. Given evidence that speech was developed and maintained in the intact hemisphere, it was concluded that the left and right hemispheres were equipotential. More recently, neuroimaging evidence has shown that despite the preference of language functions for the left hemisphere in most typically developing people, language functions can be transferred to the right hemisphere if there is left-hemispheric damage during the pre- or perinatal period (Staudt, 2010). However, the theory of early plasticity fails to account for instances of poor recovery and outcome following early brain injury.

An alternative school of thought, which has come to be known as 'early vulnerability' is based on the 'Hebb Principle'. Hebb (1949) proposed that early brain injury was more detrimental than injury later in life based on a study of children with frontal lobe damage during infancy. The theory of early vulnerability is based on the assumption that development of cognitive functions is dependent on the integrity of certain brain structures, and if these structures are damaged, the brain is never able to adequately compensate. This view argues for the 'innate specialisation' of cortical structures for particular cognitive functions, and therefore damage to these structures is likely to lead to permanent impairment of the particular cognitive ability. According to this theory, early damage to the pre-specified language area of the brain is predicted to result in specific language impairment (V. Anderson et al., 2005; Hebb, 1949).

Support for early vulnerability has been provided by Anderson and colleagues in their studies of children with acquired brain injuries (V. Anderson et al., 2000, 2004, 2005, 2010; Crowe et al., 2012). Early brain injury was associated with significantly

reduced performance across cognitive domains including language, visuospatial skills, memory, attention, executive function, and processing speed. However, the degree of recovery/vulnerability following early brain injury is influenced by a number of factors including age at injury, severity, environment and learning experience (V. Anderson et al., 2011). In particular, younger age and injury severity were associated with poorer outcomes on cognitive measures. Children who sustained moderate to severe brain injury before the age of 2 years showed global and severe deficits while children aged 7 or above at the time of injury performed comparably to population norms (V. Anderson et al., 2010). Further support for early vulnerability is provided by longitudinal studies investigating post-brain injury outcomes across the lifespan. In their study of over 700 children and adults with unilateral focal brain injury aged 0 to 84 years, Duval et al. (2008) showed that older age at injury was associated with better cognitive outcomes on Intelligence Quotient (IQ) tests. Indeed, younger age at injury was associated with IQ decline over time while older age at injury was associated with IQ recovery over time. Taken together, the early vulnerability theory indicates early injury increases risk of long-term cognitive impairment but does not necessarily provide an explanation for good recovery post-early injury.

Neither early plasticity nor early vulnerability can account entirely for the range of cognitive outcomes observed post early brain injury. If early plasticity and early vulnerability represent two ends of a continuum (V. Anderson et al., 2011), the theory of ‘cognitive crowding’ may represent somewhat of a middle ground. Cognitive crowding refers to the developing brain’s capacity for functional reorganisation following early injury; however, recognises a hierarchy in cognitive development whereby verbal skills are prioritised. The cognitive crowding hypothesis was initially posited to explain the relative sparing of language functions - and unexpected perceptual deficits - after early

damage to the left hemisphere (Teuber, 1974). The cognitive crowding hypothesis assumes "impairment in nonverbal cognitive function is due to competition for terminal space in the brain induced by displacement or reorganisation of speech in the right hemisphere" (Satz et al., 1994, p. 256). In the Satz et al. (1994) study of people with epilepsy with left-hemisphere dysfunction that occurred before or after 12 months of age (early vs late onset), the crowding effect was observed only in patients with a lesion that occurred during the pre- or perinatal period. Age at onset had no impact on verbal skills but the early-onset group were impaired on all nonverbal cognitive tasks. Hence, the authors concluded the crowding effect occurred in the context of left hemisphere damage and was restricted to nonverbal skills (Satz et al., 1994). Conversely, in the case of early injury to the right hemisphere, Muter et al. (1997) posit the selective lowering of nonverbal skills is a direct consequence of damage to the cortical areas predisposed for development of visuospatial functions. As such, basic visuospatial processing may be subsumed by the left hemisphere, but more complex nonverbal abilities are compromised.

In the context of CP, several cross-sectional (Carlsson et al., 1994; Ito et al., 1997; Sigurdardottir et al., 2008) and longitudinal studies (Gonzalez-Monge et al., 2009; Muter et al., 1997) provide support for the cognitive crowding hypothesis with evidence of relative sparing of verbal skills at the expense of nonverbal functions in children with CP. These findings were corroborated by a functional MRI (fMRI) study of language organisation in young adults with unilateral spastic CP affecting the right side (i.e. lesion to the left hemisphere of the brain) compared to controls (Lidzba, Staudt, Wilke, & Krägeloh-Mann, 2006). They found that verbal and nonverbal functions shared the same right-hemispheric network in individuals with left-sided lesions, with evidence that the right middle frontal gyrus and right premotor cortex were activated in both verbal and nonverbal conditions. In contrast, these areas showed left lateralisation for verbal tasks

and right lateralisation for non-verbal tasks in controls (Lidzba, Staudt, Wilke, Grodd, et al., 2006). Moreover, participants with CP and lesion-induced right-hemispheric organisation of language functions showed deficits on visuospatial tasks compared to controls matched according to age, sex, and verbal IQ (Lidzba, Staudt, Wilke, & Krägeloh-Mann, 2006).

Not only is the development of cognitive abilities in the context of CP affected by the initial brain insult, but it is also complicated by the cumulative demands of ongoing development throughout childhood and adolescence. Drawing on the paediatric acquired brain injury literature, a severe brain injury causes widespread and persistent cognitive deficits as well as a reduced rate of skill acquisition over time compared to typically developing controls (Ewing-Cobbs et al., 1997). Slower development of cognitive abilities leads to the "cumulative" impact of the initial brain injury which is not immediately observable but may "emerge" during later developmental stages and transitional periods (i.e. adolescence) in the context of increasing demands of independence and age-appropriate functioning (V. Anderson & Catroppa, 2005). Thus, the trajectory of cognitive development in children with CP is likely to diverge over time when compared to typically developing children, and therefore emphasises the need for ongoing assessment and monitoring throughout development.

Theories of Cognitive Abilities

In the absence of a consensus definition of intelligence, an often-cited definition describes intelligence as:

> a very general mental capability that, among other things, involves the ability to reason, plan, solve problems, think abstractly, comprehend complex ideas, learn quickly and learn from experience. It is not merely book learning, a narrow academic skill, or test-taking smarts. Rather, it reflects a broader and deeper

> capability for comprehending our surroundings – 'catching on,' 'making sense' of things, or 'figuring out' what to do (Gottfredson, 1997, p. 13).

This definition alludes to the ongoing debate in the field of intelligence research: is intelligence best described as a general ability factor (g) or a host of broad abilities? Reflective models of intelligence consider g to represent a general cognitive ability which is the underlying cause of all other cognitive abilities (Carroll, 1993; Spearman, 1904). Conversely, formative models of intelligence acknowledge the existence of g as a statistical construct but question its existence as a psychological construct (Conway & Kovacs, 2015; Horn & Blankson, 2012). Rather, g is considered an emergent property that is a necessary statistical consequence when testing a profile of correlated cognitive abilities that are functionally linked and rarely performed in isolation (Kovacs & Conway, 2019).

Spearman (1904) initially proposed the reflective view of g as a latent variable underlying individual differences in task performance. His theory was based on the "positive manifold" - the finding of positive correlations among different tests of cognitive abilities. That is, if a person performs well on one cognitive test, they tend to perform well on others. Spearman coined this common variance the general intelligence factor or "g". In contrast, Thurstone (1938) offered an opposing view of intelligence based on his factor analysis of 56 cognitive tests. According to this view, intelligence is conceptualised as a set of primary abilities (verbal, spatial, reasoning, number, fluency) from which g can be obtained as a second order factor.

Building on the work of Spearman (1904) and Thurstone (1938), the Fluid-Crystallised theory of cognitive abilities was proposed by Cattell (1963), who posited a shift toward the formative view of intelligence and away from the reflective view of g as a single cognitive factor. Cattell had a dichotomous view of g whereby fluid intelligence

(Gf) represented an innate ability to respond to novel situations, while crystallized intelligence (Gc) consisted of acquired knowledge that reflected acculturation. He noted that Thurstone's verbal, reasoning, and number primary abilities loaded onto Gc while spatial ability loaded on Gf. Shortly thereafter, the Gf-Gc model was expanded by Cattell's student, Horn (1965) to include four additional broad abilities: visual processing, short term memory, long-term storage and retrieval, and processing speed. By the 1990s, factors representing auditory processing ability, quickness in reaction time and decision making, quantitative reasoning and broad reading-writing ability were added to complete Horn's 10-factor model (Horn & Blankson, 2012). Although the name Gf-Gc theory was retained, the 10 broad abilities were considered equals and not part of a hierarchy.

In contrast to this non-hierarchical model, Carroll (1993) conceptualised cognitive ability as a hierarchical construct comprised of three distinct strata, referred to as the Three-Stratum Model, based on his factor analytic study of over 460 datasets. Carroll placed g at the apex of the model (stratum III) as the broadest and most general cognitive ability consistent with Spearman's reflective view of g. Carroll (1993) also identified eight stratum II broad abilities, defined as "basic constitutional and longstanding characteristics of individuals that can govern or influence a great variety of behaviours in a given domain" (p. 634), which mapped closely onto the Gf-Gc model. The broad abilities subsumed over 70 stratum I narrow cognitive abilities which "represent greater specialisations of abilities, often in quite specific ways that reflect the effects of experience or learning, or the adoption of particular strategies of performance" (Carroll, 1993, p. 634).

To resolve some of the differences between the Gf-Gc model and the Three-Stratum Model, McGrew (1997) integrated these models to form the Cattell-Horn-Carroll (CHC) theory of cognitive abilities. CHC remains the prevailing theory of human

intelligence which has been used to interpret the most commonly used Wechsler scales (Flanagan, 2000), and has been particularly influential in the development of other intelligence tests over the past two decades (McGill & Dombrowski, 2019). The CHC model has undergone revision and restructuring over the years and the current version includes 16 broad abilities which subsume over 80 narrow abilities (Schneider & McGrew, 2012). Although the CHC theory initially acknowledged g as a general intelligence factor, the more recent conceptualisations have omitted g from the theoretical framework (Schneider & McGrew, 2012).

Exploratory factor analytic studies of the WISC-V indicate the most variance is associated with the general intelligence factor and limited unique variance is associated with the first-order factors (i.e. broad abilities) (Canivez et al., 2016, 2020a). In line with these findings, it was suggested that interpretation should focus on the Full Scale Intelligence Quotient (FSIQ) because it accounts for the most common variance. However, there seems to be a disconnect between this view and clinical practice whereby interpretation of test results tends to occur at the level of broad abilities (Wechsler, 2014).

Proponents of the formative view of g conceptualise IQ as a profile of different abilities rather than a reflection of general intelligence (Conway & Kovacs, 2015; Horn & Blankson, 2012; McGrew, 2005). Arguments against the interpretation of a general intelligence factor include evidence indicating the strength of the positive manifold is not universal. For example, the positive manifold is stronger at lower levels of IQ and weaker at higher levels (Molenaar et al., 2017), which suggests that intelligence is more general at the lower end of the continuum and more specific at the higher end. Other evidence has shown that when both g and Gf are included in factor analytic studies, they are statistically indistinguishable and therefore g is considered redundant (Caemmerer et al., 2018; Kan et al., 2011).

Furthermore, the broad abilities have been shown to predict specific academic outcomes over and above the effect of g (Caemmerer et al., 2018; Corimer et al., 2016; Floyd et al., 2007; Taub et al., 2008). Caemmerer et al. (2018) examined the effects of CHC broad abilities on specific academic achievement areas using the WISC-V and Wechsler Individual Assessment Test (Third edition; WIAT-III) co-norming sample. The results showed the effect of g on achievement was strong but indirect through broad abilities and the effect of g was redundant when fluid reasoning was considered. In contrast, the effects of the broad abilities were domain specific because they influenced achievement in some but not all academic skills: verbal comprehension was related to all reading and most writing skills; fluid reasoning was related to essay writing and achievement in mathematics, and processing speed was related to fluency in reading, mathematics, and calculation skills. An exception was working memory which exerted effects on most reading, mathematics, and all writing skills assessed by the WIAT-III and was particularly important in younger children (Caemmerer et al., 2018).

Assessment of Cognitive Abilities

The Wechsler Intelligence Scale for Children - Fifth edition (WISC-V) is the latest version of the most widely used intelligence test for children and adolescents internationally (Oakland et al., 2016). The WISC-V has a hierarchical structure similar to the CHC model: it is comprised of 16 subtests (see Table 1) that assess narrow abilities which are then combined to provide primary index scores that represent broad cognitive abilities, as well as a composite score that represents a general ability factor (FSIQ). The FSIQ is comprised of seven subtests and an additional three subtests are required to calculate the index scores. Consistent with the CHC model, the WISC-V is ambiguous about FSIQ and emphasises interpretation of broad abilities. Although the WISC-V interpretative guidelines recognise the FSIQ score as the most reliable score and

traditionally the first score considered in profile analysis, they state it is best interpreted by considering the contribution of the primary index scores. Indeed, analysis of the primary index scores is "recommended as the principal level of clinical interpretation," particularly in cases with considerable variability across cognitive domains (Wechsler, 2014, p. 157). The WISC-V has strong psychometric properties. The reliability of the FSIQ composite is excellent ($r = 0.96$) and reliability coefficients for the primary index scores range from .88 to .93.

The WISC-V has undergone major revisions since the previous edition and now includes five primary index scores (Wechsler, 2016). While the Verbal Comprehension, Working Memory, and Processing Speed indexes have been retained, the former Perceptual Reasoning Index of the WISC-IV has been split into the Visual Spatial Index and Fluid Reasoning Index to improve alignment with CHC theory. However, independent exploratory and confirmatory factor analytic studies have failed to provide support for the five-factor structure of the WISC-V (Canivez et al., 2016, 2020b). The Fluid Reasoning Index did not load as a separate factor, and results supported a four-factor model with a dominant general factor resembling the structure of the WISC-IV.

The WISC in its various editions is the most widely used intelligence test across 64 countries over the past three decades (Oakland et al., 2016). WISC assessments are used by school psychologists to estimate students' academic aptitude, inform school placement decisions, and diagnose intellectual disability or learning disorders (Oakland et al., 2016). In other clinical settings, the WISC has been shown to be sensitive to epilepsy-related cognitive impairments (MacAllister et al., 2019), distinguish between children with neurodevelopmental disorders (Alloway et al., 2016), identify cognitive impairments in children with autism spectrum disorder (Audras-Torrent et al., 2020), and track recovery and inform rehabilitation programs post paediatric traumatic brain injury

(Bardoni et al., 2013). Relatedly, the WISC is frequently used to assess cognitive abilities in children and adolescents with CP, but as discussed in detail later in this chapter, standardised administration is burdened by fine motor demands that disadvantage children with motor impairment (Yin Foo et al., 2013).

General Cognitive Ability in CP

The impact of the cognitive impairment which often accompanies CP has been gaining recognition since its inclusion in the updated definition of CP (Rosenbaum et al., 2007). The consensus statement by Rosenbaum et al. (2007) recognises that disturbances in cognition can be either global or specific and occur as a result of the primary disturbance which causes CP or as a secondary consequence of activity limitation which restricts learning experiences. Understanding the trajectory of cognitive development in the context of CP is important for parents and clinicians alike in order to make predictions and guide expectations about future functioning and outcomes. IQ has been identified as an important determinant of functioning throughout the life course of people with CP and has implications for participation in education (Jenks et al., 2007), self-care, domestic and community tasks (Van Gorp et al., 2018), employment (Magill-Evans et al., 2008) and independent living (Reddihough et al., 2013). People with CP and IQ in the normal range tend to achieve the expected level of performance in these domains, albeit later compared to typically developing peers of the same chronological age. However, achievement of lifestyle goals is less favourable for adolescents with CP who have intellectual disability, with little improvement in functioning seen over time (Van Gorp et al., 2018).

Table 1

Descriptions of the 10 primary and 6 secondary subtests of the Wechsler Intelligence Scale for Children – Fifth edition

Primary Index	Subtest	Description
Verbal Comprehension	Similarities*	The child is read two words that represent common objects or concepts and is required to describe how they are similar.
	Vocabulary*	The child is required to either name a depicted object, or define a word that is read aloud to them.
	Information	The child answers questions about a broad range of general knowledge topics.
	Comprehension	The child answers questions based on their understanding of general principles and social situations.
Visual Spatial	Block Design*	Working within a specified time limit, the child views a model or picture and uses two-coloured blocks to recreate the design.
	Visual Puzzles	Working within a specified time limit, the child views a completed puzzle and selects three response options that, when combined, reconstruct the puzzle.
Fluid Reasoning	Matrix Reasoning*	The child views an incomplete matrix or series and selects the response option that completes the matrix or series.
	Figure Weights*	Within a specified time limit, the child to views a scale with a missing weight and selects the response option that keeps the scale balanced.
	Picture Concepts	The child views two or three rows of pictures and selects one picture from each row to form a group with a common characteristic.
	Arithmetic	The child mentally solves arithmetic problems within a specified time limit.
Working Memory	Digit Span*	An auditory working memory task in which a sequence of numbers is read aloud by the examiner and the child is required to recall the numbers in the same order, reverse order, or ascending order.
	Picture Span	A visual working memory task in which the child views a stimulus page with one or more pictures of objects for a specified time, and then selects the pictures (in sequential order) from options on a response on a response page.
	Letter-number sequencing	The child is read a series of letter and numbers and required to repeat them back with the numbers in numerical order and with letters in alphabetical order
Processing Speed	Coding*	The child works within a two-minute time limit and uses a key to copy symbols that correspond with simple geometric shapes or numbers.
	Symbol Search	The child scans search groups and indicates whether target symbols are present, while working within a two-minute time limit.
	Cancellation	The child scans two arrangements of objects (one random, one structured) and marks target objects while working within a specified time limit

*subtest contributes to Full Scale Intelligence Quotient

Intellectual disability is defined as "deficits in general mental abilities and impairment in everyday adaptive functioning, in comparison to an individual's age-, gender-, and socio-culturally matched peers" with onset during the developmental period (American Psychiatric Association, 2013, p. 37). Diagnosis is based on "clinical assessment and standardized testing of intellectual and adaptive functions that is individually administered and psychometrically valid, comprehensive, culturally appropriate [and] psychometrically sound" (American Psychiatric Association, 2013, p. 37). When applying an IQ test, intellectual disability is commonly diagnosed when the FSIQ score falls two standard deviations (SD) or more below the mean for age-matched typically developing peers. In the context of the Wechsler scales that have a mean of 100 and SD of 15, this involves FSIQ scores less than 70. Moreover, caution needs to be taken when interpreting the FSIQ score in the face of an uneven cognitive profile which, as discussed later in this chapter, is common in CP.

According to the Australian Cerebral Palsy Register (2018) and similar estimates from the US (Van Naarden Braun et al., 2015), UK (Surman et al., 2006) and Europe (Surveillance of Cerebral Palsy in Europe, 2000), intellectual disability occurs in approximately half of children with CP by 5 years of age. As discussed in greater detail later in this chapter, higher rates of cognitive impairment have been generally associated with spastic quadriplegia (Himmelmann et al., 2006; Reid et al., 2018; Sigurdardottir et al., 2008) or hypotonic CP (Delacy & Reid, 2016), bilateral motor impairment (Sigurdardottir et al., 2008; Smits et al., 2011), and greater severity of motor impairment (Sigurdardottir et al., 2008; Smits et al., 2011; Stadskleiv et al., 2018).

Profile of Broad Cognitive Abilities in CP

In contrast to the high rates of intellectual disability, a substantial proportion of children with CP have IQ scores within the normal range, particularly those with mild

motor impairment (GMFCS Level I) and unilateral subtypes (Sigurdardottir et al., 2008). Nonetheless, median IQ scores remain significantly lower in children with CP than the general population (Sigurdardottir et al., 2008). In addition, FSIQ scores in the normal range may mask selective deficits or areas of relative strength. Studies have revealed the presence of an uneven cognitive profile in children with CP characterised by stronger verbal than non-verbal skills (Carlsson et al., 1994; Ito et al., 1997; Pagliano et al., 2007; Sigurdardottir et al., 2008). For example, in a sample of 4- to-6 year old children with CP, Sigurdardottir et al. (2008) found median verbal IQ to be within normal limits, while median performance IQ was within the *Borderline* range. These authors identified verbal skills as a relative strength across CP subtypes and levels of motor impairment, while deficits in non-verbal skills appeared to increase with increasing CP severity.

Nonverbal skills have also been found to be impaired in both left- and right-unilateral CP groups compared to controls, despite FSIQ scores within the normal range (Carlsson et al., 1994). While there was no significant difference in nonverbal skills between unilateral groups, there was, however, a significant difference in verbal abilities: those with left-unilateral CP (and radiologically confirmed right-hemisphere lesions) outperformed those with right-unilateral CP (and radiologically confirmed left-hemisphere lesions) on verbal tasks. These findings were interpreted in the context of the cognitive crowding effect. Impaired nonverbal function in unilateral CP, irrespective of the side, highlighted the prioritization of language function at the expense of nonverbal skills in atypical brain development (Carlsson et al., 1994). An alternative explanation for lower nonverbal functions pertains to the impact of motor impairment on nonverbal tasks which have a higher motor component (e.g. the Performance IQ of the WISC). Thus the extent to which the crowding effect or motor demands contribute to the uneven profile of verbal versus non-verbal skills is yet to be determined.

It remains unclear the nature of the relation between verbal and non-verbal skills compared to working memory and processing speed in CP because earlier versions of the WISC did not provide the 4-5 factor structure of recent versions. The earlier versions of the WISC (i.e. WISC-Revised, WISC-III) included only two indices: Verbal IQ comprised of verbal comprehension and auditory working memory tasks and Performance IQ comprised of visual-spatial, perceptual reasoning, and processing speed tasks. In a more recent study of executive functions in children and adolescents with unilateral CP (GMFCS Level I-II), Bodimeade et al. (2013) found poorer working memory performance on the Digit Span subtest of the WISC-IV compared to typically developing age-matched controls. The authors found that working memory scores in the CP group were significantly below controls, but nonetheless remained within the normal range. Within the CP group, there was no significant difference between children with left- or right-unilateral CP in the working memory domain.

Similarly, in a study that included children and adolescents with CP and varying levels of motor impairment (GMFCS Level I-V), Stadskleiv et al. (2018) showed that children with severe motor impairment who were capable of answering working memory tasks performed as well as those with less severe motor impairment. In fact, working memory performance was comparable in unilateral and bilateral CP, as well as across all levels of motor impairment measured by the GMFCS (Stadskleiv et al., 2018). These findings highlight that tests with minimal motor demands do not place children with CP at a disadvantage, increase access to cognitive tests and allow them to demonstrate their true cognitive abilities. While this may be achieved in the working memory domain, motor-free assessment of other broad abilities such as processing speed is an inherently more complicated issue.

Slower processing speed in children with CP would not be surprising given the initial brain injury. Predominant white matter injury is the most common pathogenic pattern identified on neuroimaging in children with CP (Krägeloh-Mann & Horber, 2007), while white matter volume is positively correlated with processing speed (Magistro et al., 2015). Accordingly, white matter injury was associated with lower processing speed scores on the WISC in adolescents who were born very preterm (Soria-Pastor et al., 2008). However, as discussed later in this chapter, assessment of processing speed is complicated in the context of CP due to the motor component of processing speed tasks on the WISC. In the study by Bodimeade et al. (2013), children and adolescents with unilateral CP (GMFCS Level I-II) performed worse than typically developing controls on both Symbol Search and Cancellation subtests of the WISC-IV. Although these subtests were selected due to their reduced motor requirements compared to the Coding subtest, they nonetheless require manipulation of a pencil and rapid responses. As such, it is impracticable to determine if lower scores on these subtests are due to slow processing or slow movement time, which can both be impacted by the CP type and severity.

Impact of Clinical Factors on Cognitive Abilities in CP

CP is a heterogenous condition that encompasses a wide range of motor types, topographies, and levels of activity limitation. In turn, the range of cognitive abilities in children with CP varies widely and is influenced by a range of features associated with the disorder. The evidence for an association between CP type and cognitive abilities remains somewhat unclear in the literature. A longitudinal study showed children with dyskinetic CP achieved the lowest scores on Ravens Coloured Progressive Matrices compared to children with spastic CP, and showed the least improvement in scores between the ages of 5 to 9 years (Smits et al., 2011). Similarly, an Icelandic showed that

children with dyskinesia had the lowest median FSIQ score, but intellectual impairment was more common in spastic quadriplegic CP (64%) compared to dyskinetic CP (57%; Sigurdardottir et al., 2008). In contrast, other studies have shown no significant association between CP type and intellectual impairment (Reid et al., 2018; Türkoğlu et al., 2017). Reid et al. (2018) showed that children with dyskinetic CP were 5 times were likely to have severe motor impairment and 8 times more likely to have bilateral motor involvement compared to children with spastic CP. However, after accounting for motor topography, there was no strong evidence to suggest poorer cognitive outcomes in children with dyskinetic CP compared to children with spastic CP. Moreover, other evidence suggests that at similar levels of motor impairment, people with dyskinetic CP demonstrate higher IQ than those with spastic CP (Laporta-Hoyos et al., 2019). Therefore the degree of motor involvement may be more clinically relevant than CP subtype when predicting cognitive outcomes.

Based on the recommendation of the Surveillance of Cerebral Palsy in Europe (2000), it may be more meaningful to differentiate cognitive outcomes according to unilateral versus bilateral CP. Bilateral motor impairment is consistently associated with less favourable cognitive outcomes than unilateral motor impairment (Sigurdardottir et al., 2008; Smits et al., 2011). In fact, unilateral CP has been associated with the lowest rates of intellectual impairment (Himmelmann et al., 2006; Stadskleiv et al., 2018) and highest median FSIQ scores (Sigurdardottir et al., 2008).

Another factor that has been consistently associated with cognitive outcomes is severity of motor impairment as measured by the GMFCS – that is, more severe motor impairment is related to greater cognitive impairment (Himmelmann et al., 2006; Sigurdardottir et al., 2008; Smits et al., 2011; Stadskleiv et al., 2018; Türkoğlu et al., 2017). In fact, based on data from the Australian CP Register, the rate of intellectual

impairment increases with each GMFCS level from 26% at Level I to 85% at Level V (Delacy & Reid, 2016). The relation between motor impairment and cognitive development is complex in so far as the motor impairment is not only related to the initial brain injury but has ongoing implications for the child's ability to engage with, and learn from, their environment. Previous MRI studies have shown that findings of brain maldevelopment, basal ganglia lesions, and cortical/subcortical lesions were associated with severe gross motor impairment as well as severe intellectual disability (Himmelmann & Uvebrant, 2011). Moreover, motor impairment which restricts the child's mobility or communication also restricts cognitive development due to limited ability to explore their environment, engage in play-based learning, and participate in social interactions and classroom activities (Bottcher, 2010). However, it is important to note that there is substantial heterogeneity among adolescents with the same level of motor impairment. Stadskleiv et al., (2018) reported that one third of CP participants with severe motor impairment in their sample had normal cognitive ability, highlighting the importance of motor-free cognitive assessments that would enable children with all levels of motor impairment to demonstrate their true cognitive abilities.

Impact of Associated Conditions on Cognitive Abilities in CP

In addition to the influence of CP-related features on cognitive abilities, a range of associated factors have been identified as contributing to individual differences in IQ including seizure history, gestational age at birth, and birth weight. Previous studies have consistently provided evidence for an association between a history of seizures and reduced FSIQ scores in children with CP at preschool-age (Sigurdardottir et al., 2008), during early childhood (Muter et al., 1997), and into adolescence (Cheng et al., 2013; Gonzalez-Monge et al., 2009). In a study of children and young adults with CP between the ages of 5 and 20 years, Cheng et al. (2013) observed that seizure history had a

deleterious effect on IQ scores over time. They reported that adolescents over the age of 15 years with CP and seizure history were more likely to have IQ < 80 compared to children under the age of 15 with CP and seizure history. These findings suggest seizures may impede the emergence of more complex cognitive abilities during adolescence. Similarly, a study by Gonzalez-Monge et al. (2009) of 32 adolescents aged 4 years 6 months at baseline found a history of seizures was related to significant decline in nonverbal IQ at 7-year follow-up as well as a slower progression of verbal IQ. The authors interpreted these findings in terms of the crowding effect whereby nonverbal skills are more affected by early brain injury than verbal skills.

Previous studies examining the association between gestational age at birth and cognitive outcomes in CP have been largely inconsistent. Hemming et al. (2008) examined a UK CP register of over 4000 cases and found that gestational age was not associated with cognitive impairment in unilateral spastic CP, except for in extremely pre-term cases born before 28 weeks gestation. However, infants with bilateral CP born at term were more likely to have severe intellectual impairment (IQ < 50) than those with bilateral CP born preterm (Hemming et al., 2008). The authors suggested the higher rates of severe intellectual impairment with increasing gestational age in bilateral CP may reflect a reduced ability for the brain to reorganise when there is more diffuse injury at later gestational periods.

In contrast, another study by Pagliano et al. (2007) showed that although children with bilateral CP born preterm and term were similar with regard to overall cognitive ability and verbal skills, preterm birth was associated with poorer nonverbal skills at 6 years of age. The authors suggest this was a direct result of prematurity – the truncated intrauterine period may be insufficient to allow reorganisation of visual pathways after the initial brain injury (Pagliano et al., 2007).

The relationship between birth weight and cognition in CP has also shown mixed evidence in the literature. Cheng et al. (2013) reported birth weight was not associated with cognitive abilities in their sample of children and young adults with CP. In contrast, Hemming et al. (2008) demonstrated that birth weight was a significant predictor of severe cognitive impairment in CP cases on the UK register. These inconsistent findings are likely explained by the way birth weight was measured across these studies. For example, Cheng et al. (2013) examined absolute birth weight for associations with cognitive abilities, while Hemming et al. (2008) calculated standardised birth weight for gestational age. Infants born small for gestational age (SGA; birth weight $<10^{th}$ percentile of expected weight for gestational age) were at increased risk of severe intellectual impairment relative to those born with appropriate weight for gestational age (Hemming et al., 2008). It seems restricted growth may be a more important predictor of cognitive outcomes in CP than low absolute birth weight. This is in line with recent studies in children without CP that identified SGA as a significant predictor of poorer cognitive outcomes in infancy into adulthood independent of preterm birth, birth weight, socioeconomic status, and parent-child relationship (Eves et al., 2020; Sacchi et al., 2020).

In summary, many children with CP have other conditions including seizures, preterm birth, and low birth weight. While there is consistent evidence for the negative impact of seizures on cognitive outcomes, the impact of preterm birth and low birth weight on cognition in the context of CP is yet to be clarified.

Longitudinal Studies of Cognitive Development in CP

Although this disability is a non-progressive disorder, the extent of cognitive and functional deficits in CP may not be identified for many years, and only become apparent when the child is expected to act independently in their environment. Understanding the

developmental trajectory through adolescence is important to guide expectations and support young people with CP in their current functioning and transition to adulthood. Despite this, there are limited longitudinal studies investigating cognitive development in children with CP, and even fewer that examine development into adolescence.

Longitudinal studies of cognitive development during early childhood in the context of CP indicate an early divergence in the development of verbal and nonverbal skills. The uneven cognitive profile is identifiable as early as 3-years of age and characterised by stronger verbal than nonverbal abilities (Fedrizzi et al., 1993; Muter et al., 1997). Verbal skills were within the normal range between the ages of 3 to 6 years in children with unilateral and bilateral CP while a consistent decrement in nonverbal skills was observed over time (Fedrizzi et al., 1993; Ito et al., 1997; Muter et al., 1997). Moreover, the gap between verbal and nonverbal skills was shown to widen due to a slower rate of skill acquisition in the nonverbal domain (Ito et al., 1997; Muter et al., 1997). Factors affecting the development of nonverbal skills included severity of motor impairment (Fedrizzi et al., 1993; Smits et al., 2011), hand-eye coordination (Fedrizzi et al., 1993), seizures (Muter et al., 1997), and school type (Ito et al., 1997).

A limitation of these longitudinal studies of preschool- and primary-school aged children with CP is the restricted age range of participants and short follow-up periods, which may not capture the full extent of cognitive deficits. For example, a study of children with CP born preterm by Pleacher et al. (2004) showed that IQ at age 3 years was not predictive of IQ at age 8 years, suggesting that substantial changes occur during this time. A significant period of cognitive development occurs between the ages of 7 and 9 in typically developing children, which corresponds with a growth spurt in the frontal lobes, and then another spurt between the ages of 11 and 13 years (P. Anderson, 2002).

Longitudinal studies that span these development periods are required to capture the trajectory of cognitive abilities in adolescents with CP.

The few studies that have tracked cognitive change or stability throughout adolescence were limited by substantial heterogeneity in their samples. In a study of 15 children with early unilateral brain injury by Levine et al. (2005), cognitive abilities were assessed once before the age of 7 and at follow-up between 1.5-15 years later. Although there was a significant decline in both verbal and nonverbal IQ scores over time, it was difficult to draw conclusions about any developmental period given the wide age range of participants. A similar decline in IQ was observed by Dahlgren Sandberg et al. (2006) in a study of six children with CP and severe speech and motor impairments. Although the authors were primarily concerned with reading and spelling abilities, they noted an average decline of 23 FSIQ points on Raven's Progressive Matrices between the ages of 6 and 12 years. However, at the individual level, the IQ of two children remained stable over time while the remaining four showed a decrease in IQ ranging between 15 and 45 points (Dahlgren Sandberg, 2006). Despite the same level of motor impairment, their cognitive development was highly disparate and reasons for this were unknown. Similarly, Gonzalez-Monge et al. (2009) conducted a 7-year follow-up study of 32 adolescents with unilateral CP and found that despite a selective decline in nonverbal IQ, verbal skills and FSIQ remained stable over time. However, while group mean difference scores might indicate temporal stability of FSIQ, individual change scores ranged from -27 to +34 IQ points (Gonzalez-Monge et al., 2009). The reliance on group means to describe the developmental trajectory of cognitive abilities in children and adolescents with CP may be misleading because they are unlikely to capture the heterogeneity in the range of cognitive scores.

In summary, longitudinal studies of cognitive development in the context of CP are limited by two main factors: a) short follow-up periods in studies of preschool- or primary school-aged children that do not capture the full extent of later cognitive deficits which may emerge during later childhood or adolescence; and b) the reliance on group mean differences to characterise change in IQ over time, which may be misleading as to the representative range of cognitive abilities in children and adolescents with CP.

Moreover, it is important to note that decline in IQ scores over time does not necessarily represent a regression in cognitive abilities, but rather a reduced acquisition rate of new skills. Levine et al. (2005) explained that in order to maintain the same IQ score over time, the child must continuously rise to task demands which increase with age. Hence, the disparity between children with CP and typically developing children may be expected to widen during adolescence when cognitive tasks demand increasingly more complex abstract reasoning skills as well as sufficient motor abilities.

Impact of Motor Responses on Cognitive Tests

As alluded to earlier in this chapter, a major limitation in the literature is that performance on cognitive tests have been rarely interpreted in the context of the motor impairment characteristic of CP. A diagnosis of intellectual disability, by definition, requires evidence of "deficits in intellectual functions…confirmed by both clinical assessment and individualised, standardised intelligence testing" (American Psychiatric Association, 2013, p. 33). However, standardised assessment in children with CP is challenging because most general intelligence tests require good fine motor skills, rapid responses, and expressive language ability (Kurmanaviciute & Stadskleiv, 2017). The motor impairment inherent to CP is a barrier to participation on cognitive tests and even mild fine motor impairment can negatively impact test performance. Previous studies indicate one-third of children with CP do not have the fine motor skills required to

respond to, or obtain, a formal measure of intelligence (Sherwell et al., 2014; Sigurdardottir et al., 2008; Stadskleiv et al., 2015). In cases where a child does not have the motor skills required to respond to an intelligence test, intellectual disability is commonly determined based on clinical judgement (Andersen et al., 2008; Himmelmann et al., 2006) or severity of motor impairment (Hutton et al., 2002). However, Sigurdardottir et al. (2008) showed that 20% of preschoolers with CP who were unable to respond to the Wechsler Primary and Preschool Scale of Intelligence (WPPSI) achieved a developmental quotient in the normal range (i.e. >85) on tests with fewer motor demands (e.g. Reynell-Zinkin Developmental Scale). This highlights that assessments conducted using standardised procedures will not necessarily be an accurate reflection of the child's cognitive abilities due to the reliance on motor responses, and emphasise the crucial need to develop motor-free methods of assessing intelligence in CP.

Cognitive assessments in CP are not straightforward even for those who have sufficient motor skills to perform the tasks. Losch and Dammann (2004) performed factor analyses to quantify the impact of motor responses on performance on standardised cognitive tests in a group of 6-year-old children who were born very low birth weight. Their sample was comprised of children with no or minimal motor impairment, "clumsy" children, children with CP, and children with ADHD or behavioural problems. The cognitive tests required either verbal or fine motor responses including pointing, the manipulation of a pencil to provide handwritten responses, or arranging pictures in a given order. Results showed that 16% of the variance in test performance was explained by motor skills, even in children without obvious motor impairment. This study highlights important clinical implications for assessing children with CP—that is, children with severe motor impairment are likely to show clear difficulties while responding to subtests that require fine motor demands and results should be interpreted

with caution. Unfortunately, it is often the case that clinicians assume that children with mild motor impairment do not require adjustments to standardised assessments of intelligence, which appears not to be consistent with these findings (Losch & Dammann, 2004).

The WISC-V was developed primarily with typically developing children, and although some special group studies were conducted during standardisation (e.g. children with specific learning disorders), children with motor impairment were not included. As such, the WISC-V does not provide standardised procedures for assessing children with motor impairment. There are two aspects of cognitive assessment using the WISC-V that are particularly challenging for children and adolescents with CP—that is, fine motor control required to manipulate stimuli and speeded responses on timed tasks. For example, the Coding subtest of the WISC-V is a timed, pencil-to-paper task in which the child is required to use a key and copy symbols that correspond with numbers. In addition, the Block Design subtest requires the manipulation of small, three-dimensional blocks using one or both hands to reconstruct a pictured puzzle. The inclusion of these subtests in the calculation of FSIQ has the potential to invalidate results and underestimate cognitive abilities. In a study of preschool children, Sherwell et al. (2014) showed that the inclusion of Coding and Block Design underestimated FSIQ by 2-7 standard score points on the Wechsler Preschool and Primary Scale of Intelligence (Third edition; WPPSI-III) compared to a short-form of the WPPSI-III based on motor-free subtests. The difference was significant even for those with the mildest level of motor impairment (MACS Level I) and the gap between FSIQ and the short-form increased with more severe motor impairment. This issue is further complicated by the WISC-V substitution guidelines which restrict clinicians' options when assessing children with motor impairment. According to the WISC-V substitution guidelines, only one subtest

may be substituted when calculating FSIQ, and the substituted subtest must be from the same domain (Wechsler, 2016). Therefore, only one of Block Design or Coding can be substituted, and the other subtest must be used in calculating FSIQ, which has the potential to underestimate cognitive abilities in children with motor impairment.

Methods for Assessing IQ Which Minimise Motor Demands

To date, although clinicians commonly make adaptations to standardised administration procedures to accommodate individuals with CP (e.g. use of large print), there are some modifications (e.g. extra time) that may affect the construct being measured and thus limit comparison to the normative data (American Psychological Association et al., 2014). In recent years, there has been an increase in the use of touchscreen technology in neuropsychological assessment. The WISC-V is now available via Q-interactive, a digital platform that delivers assessment tasks on an iPad tablet. Equivalence of the pencil-to-paper and digital formats of the WISC-V was reported by Daniel et al. (2014) and Raiford et al. (2016), and this allowed the norms, reliability, and validity information gathered for the WISC-V pencil-to-paper version to be applied to the digital format. Nonetheless, neither of these studies examined the equivalence of the primary indices or FSIQ scores, which is problematic given interpretation usually occurs at this level. Notwithstanding this limitation, the Q-interactive administration of the WISC-V replaces the handwritten responses required by the pencil-to-paper format of Coding with an onscreen touch/point response. In their study of preschool children with CP, Sherwell et al. (2014) showed that while 82% of their cohort were able to provide a simple point response, only 66% were able to complete the pencil-to-paper Coding subtest and enable calculation of a Full Scale IQ score. It remains to be determined the extent to which children with CP are capable of responding with a touch response to an

iPad as well as the current versions of the WISC-V, and in older children and adolescents with CP.

By eliminating the need for handwritten responses, the use of touchscreen technology may improve the accessibility of the WISC-V for children with CP. However, the administration of Block Design has remained unchanged on Q-interactive and continues to be problematic when assessing IQ in children with CP. In the study by Sherwell et al. (2014), 10% of preschool children with CP were unable to respond to Block Design, and those who did showed poorer performance when compared to other nonverbal tasks. According to the WISC-V substitution guidelines, the Visual Puzzles subtest may be substituted for Block Design when calculating FSIQ (Wechsler, 2016). Visual Puzzles requires the child to reconstruct a puzzle by choosing three of six multiple choice options (Wechsler, 2016). It has excellent internal consistency (r = .87; Wechsler, 2016) and factor analysis has shown that Block Design and Visual Puzzles both saliently load on the Visual Spatial domain (Canivez et al., 2016). Therefore, Q-interactive may be used in conjunction with the substitution guidelines to reduce the fine motor requirements of the WISC-V, but there have been no studies to date examining the utility of this method in children and adolescents with CP.

Although the motor demands of the WISC-V can be minimised using Q-interactive, the issue of speeded responses remains a significant barrier when assessing cognition in CP. Processing speed as a construct is comprised of four factors including visualisation speed, perceptual speed, decision time, and movement time (O'Connor & Burns, 2003). Although we may expect slower cognitive processing speed in CP due to the initial brain injury, particularly in the context of predominant white matter injury (Soria-Pastor et al., 2008), its measurement by the WISC-V is confounded by movement time. In a study examining bilateral hand skill using a computerised Peg Moving Task,

Dellatolas et al. (2005) showed that movement time in children with unilateral and bilateral CP was significantly slower than age-matched controls. Interestingly, in the unilateral group, both the affected and the unaffected hand were significantly slower than controls. Furthermore, in an electroencephalographic study of reaction time that required only a simple push-button response, Hakkarainen et al. (2012) showed that movement time in children with CP remained slower than typically developing controls; however, event-related potential analysis revealed that stimulus evaluation time and motor planning were not significantly different between groups. The authors concluded that the processes involved in stimulus evaluation were intact, but movement time remained slow in the CP group. Together, these findings suggest that despite the reduced motor requirements of Coding on Q-interactive, performance will likely remain more reflective of the child's motor speed than purely information processing speed.

An approach to overcoming the limitations of fine motor demands and measures of processing speed in intelligence tests has been proposed by Piovesana et al. (2019). These authors developed a formula for a short-form of the WISC-V to estimate IQ based on six motor-free subtests. The Motor-free IQ is a composite score that combines Similarities, Vocabulary, Visual Puzzles, Figure Weights, Digit Span, and Letter-Number Sequencing. Descriptive statistics and look-up tables were generated using the normative and psychometric data provided in the WISC-V manual. The Processing Speed Index, which has the weakest loading on the general intelligence factor in both normative (Canivez et al., 2016) and clinical samples (Canivez et al., 2020a) has been omitted from the battery due to the motor requirements of the subtests in this domain. Nonetheless, the Motor-free IQ has been shown to have excellent internal consistency ($r = 0.97$) comparable to that of the WISC-V FSIQ (Piovesana et al., 2019), and provides a method

of reducing the impact of motor difficulties on testing without compromising standardised administration procedures and comparison to normative data.

More recently, a similar nonmotor version of the WISC-V was developed by the test publishers in response to the unique needs of clinicians during the COVID-19 pandemic and made available to Pearson subscribers via their online platform. The 'WISC-V International Nonmotor Full Scale Score' was developed to be used in place of the FSIQ composite when blocks and the Coding response booklet cannot be used during telehealth assessments (Pearson Clinical, 2020). Although the Nonmotor score is derived from a different combination of subtests compared to the Motor-free IQ by Piovesana et al. (2019), it provides another useful approach to overcome the limitations of fine motor responses and substitution when estimating IQ in children and adolescents with CP. However, it is yet to be determined whether and to what extent the Nonmotor and Motor-free methods are useful in overcoming the impact of motor impairment on cognitive assessment in a clinical sample of children and adolescents with CP.

Chapter Outline for Empirical Papers

The overarching aim of this thesis was to examine cognitive abilities in adolescents with CP and explore clinical factors which may be related to cognitive impairment. In order to achieve this objective, this thesis contains a series of published papers and a manuscript submitted for publication framed by introductory chapters and a general discussion chapter. Given the motor impairment inherent to CP is a barrier to participation on cognitive tests, it was necessary first to determine the most appropriate method for assessing cognitive abilities in adolescents with CP. To this end, Chapter 3 is a published empirical paper that examines the utility of cognitive assessment methods which minimise motor demands compared to standardised administration of the WISC-V in a cohort of adolescents with CP. In Chapter 4, the utility of a "motor-free" method of

administration of the WISC-V was evaluated to characterise a cognitive profile for adolescents with CP and explore the association between IQ scores and clinical factors and conditions associated with CP. In Chapter 5, individual differences in cognitive outcomes were explored in a 10-year follow-up study that focused on change or stability in cognitive function (rather than relying only on group mean differences) from preschool age to adolescence in a cohort of children with CP. Since the empirical papers are presented in formatted journal style, there will be unavoidable repetition in some sections. Finally, in Chapter 6, the general discussion outlines the main findings across the empirical papers and provides an overview interpretation in the context of previous research, theoretical perspectives of early brain injury and plasticity, limitations and clinical implications, and suggested directions for future research.

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Chapter 3: Assessing IQ in Adolescents with Cerebral Palsy Using the WISC-V

Preamble to empirical paper 1

This study compared motor-free methods for assessing cognitive ability in adolescents with Cerebral Palsy (CP) to the traditional method using the Wechsler Intelligence Scale for Children – Fifth edition (WISC-V). Given previous evidence that subtests of the Wechsler scales that demand quick motor responses place children with CP at an inherent disadvantage (Sherwell et al., 2014), this chapter explored several methods for minimising motor demands while measuring Full Scale Intelligence Quotient (FSIQ) including the use of touchscreen technology and short-forms of the WISC-V based on motor-free subtests. Two short-forms of the WISC-V which minimised motor demands were compared in the following published study: the Nonmotor method (Pearson Clinical, 2020) and the Motor-free method (Piovesana et al., 2019). Although similar, these short-forms were comprised of different combinations of WISC-V subtests, and thus both were included in the present study to determine the most appropriate methods for assessing cognitive abilities in adolescents with CP.

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Assessing IQ in Adolescents With Mild to Moderate Cerebral Palsy Using the WISC-V

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Abstract

Objective: To examine the influence of subtests that require fine motor responses on measures of intellectual ability, and compare three approaches to minimizing motor demands while assessing cognitive abilities in adolescents with Cerebral Palsy (CP) to the traditional method of the Wechsler Intelligence Scale for Children – Fifth edition (WISC-V). *Method:* Seventy adolescents with CP (*M* = 14 years 6 months, *SD* = 10 months) who were able to provide either a verbal or point response were assessed using the WISC-V administered via Q-interactive. The pencil-to-paper version of Coding was also administered. Performance on Block Design and pencil-to-paper Coding was compared to Visual Puzzles and Coding on Q-interactive, respectively. Full Scale IQ (FSIQ) scores derived according to the Traditional method of the WISC-V were compared to alternative estimates of FSIQ derived according to the Q-interactive, Nonmotor, and Motor-free methods, which minimized motor demands. *Results:* An additional 7–12% of participants were able to respond to Visual puzzles and Coding on Q-interactive compared to Block Design and pencil-to-paper Coding, respectively, and performance was marginally but significantly better. For 54 adolescents (Gross Motor Function Classification System (GMFCS) Level I-III) who were able to obtain FSIQ scores, the Traditional method underestimated FSIQ by 3-6 points compared to the alternative methods and the difference was most pronounced for those with more severe CP as measured by the GMFCS. *Conclusion:* Adolescents with CP are at an inherent disadvantage when cognitive ability is assessed using the Traditional method of the WISC-V. Findings suggest clinicians should employ the Nonmotor or Motor-free methods when assessing IQ in adolescents with CP.

Keywords: Adolescents, cerebral palsy, cognitive assessment, motor-free, WISC-V

Assessing IQ in Adolescents With Cerebral Palsy Using the WISC-V

Cerebral Palsy (CP) affects approximately two in 1000 live births in developed countries (Australian Cerebral Palsy Register, 2018; Surveillance of Cerebral Palsy in Europe, 2000). CP has proved challenging to define as it encompasses a range of types and topographies of motor impairment and levels of functioning (Bax, 1964; MacKeith & Polani, 1959; Mutch et al., 1992). The most recent consensus definition emphasises the disordered movement and posture which characterises CP, and also highlights the non-motor features that can accompany it, including disturbances of cognition (Rosenbaum et al., 2007). It is generally accepted that intellectual impairment is common in CP, however reports of prevalence can vary widely, ranging from 17% to 60% (Ashwal et al., 2004; Blair, 2010). Cognitive assessments are often required to assess intellectual abilities in children with CP and subsequently determine eligibility for school placement, government support, and additional resources. However, accurately characterising cognitive abilities in children with CP depends on the suitability and quality of neuropsychological assessments that accommodate their motor coordination problems (Stadskleiv, 2020).

Cognitive assessment in children with CP is challenging because their motor impairment is a barrier to participation and can impact verbal responses, speeded responses and manipulation of stimuli on cognitive tests. Previous studies indicate one-third of children with CP do not have the motor skills required to respond to, or obtain, a formal measure of intelligence (Sherwell et al., 2014; Sigurdardottir et al., 2008; Stadskleiv et al., 2015). Moreover, children with more severe motor impairment who are unable to provide appropriate motor or verbal responses are often deemed ‘unassessable’ and intellectual ability may be estimated on the basis of clinical judgement or severity of gross motor impairment (Stadskleiv, 2020; Yin Foo et al., 2013).

The Wechsler Intelligence Scale for Children - Fifth edition (WISC-V) is the latest version of the most widely used intelligence test for children and adolescents (Flanagan & Alfonso, 2017; Wechsler, 2016). Although some special group studies were conducted during standardization, the WISC-V was developed primarily with typically developing children and procedures for assessing children with motor impairment are not provided. Two subtests which contribute to the Full Scale Intelligence Quotient (FSIQ) are particularly challenging for children with CP: Coding and Block Design. The Coding subtest is a timed, pencil-to-paper task which requires handwritten responses, while the Block Design subtest requires the manipulation of small, three-dimensional blocks using one or both hands to reconstruct a pattern. To date, clinicians may have made adaptions to standardized administration procedures to accommodate individuals with CP (e.g. use of large print); however some modifications (e.g. extra time) may affect the construct being measured and thus limit comparison to the normative data (American Educational Research Association et al., 2014).

In recent years, there has been an increase in the use of touch-screen technology in neuropsychological assessment. The WISC-V is now available via Q-interactive, a digital platform that delivers assessment tasks on an iPad tablet. Q-interactive administration may increase the accessibility of the WISC-V for children with CP as it replaces the handwritten responses required by the pencil-to-paper format of Coding with an onscreen touch/point response. In their study of preschool children with CP, Sherwell et al. (2014) showed that while 82% of their cohort were able to provide a point response, only 66% were able to complete the pencil-to-paper Coding subtest and enable calculation of a FSIQ score. However, the administration of Block Design remains unchanged on Q-interactive and continues to be problematic when assessing IQ in children with CP. In the same study by Sherwell et al. (2014), 10% of preschool children with CP were unable to

respond to Block Design, and those who did performed poorly compared to other non-verbal tasks. According to the WISC-V substitution guidelines, one subtest may be substituted when calculating FSIQ, and the substituted subtest must be from the same domain (Wechsler, 2016). As such, the Visual Puzzles subtest may be substituted for Block Design when calculating FSIQ. Visual Puzzles requires the child to reconstruct a puzzle by choosing three of six multiple choice options. It has excellent internal consistency (r = .87; Wechsler, 2016) and factor analysis has shown that Block Design and Visual Puzzles both saliently load on the Visual Spatial domain (Canivez et al., 2016). Therefore, Q-interactive can be used in conjunction with the substitution guidelines to reduce the fine motor requirements of the WISC-V.

Although the motor demands of the WISC-V can be minimized using Q-interactive, the issue of speeded responses remains, which may in fact be the greatest barrier when assessing cognition in CP. Processing speed as a construct is comprised of four factors including visualization speed, perceptual speed, decision time, and movement time (O'Connor & Burns, 2003). Although we may expect slower processing speed in CP due to the initial brain injury, particularly in the context of predominant white matter injury (Soria-Pastor et al., 2008), its measurement by the WISC-V is confounded by movement time. Studies of bilateral hand skill have shown that movement time in the affected and unaffected side is significantly slower in children with unilateral CP compared to typically developing controls (Dellatolas et al., 2005; Steenbergen & Meulenbroek, 2006). Similarly, an electroencephalographic study of reaction time to a computerized task that required a simple motor response (i.e. push button to indicate yes or no) showed that the movement time of children with CP was slower than typically developing controls (Hakkarainen et al., 2012). However, event-related potential analysis revealed that stimulus evaluation time and motor planning were not significantly different

between groups. The authors concluded that the processes involved in stimulus evaluation were intact, but movement time was slow. Together, these findings suggest that despite the reduced motor requirements of Coding on Q-interactive, performance will likely remain more reflective of the child's movement time rather than their cognitive processing speed.

An approach to overcoming the limitations of fine motor demands and measures of processing speed in intelligence tests was proposed by Piovesana et al. (2019) and adopted by the current study. These authors developed the "Motor-free method", a short-form of the WISC-V which estimates FSIQ based on six motor-free subtests. The Motor-free IQ has been shown to have excellent internal consistency ($r = 0.97$) comparable to that of the WISC-V FSIQ (Piovesana et al., 2019), and provides a method of reducing the impact of motor difficulties on testing without compromising standardized administration procedures and comparison to normative data. To our knowledge, this motor-free method has not yet been applied to a clinical sample of children with CP.

More recently, a similar, "Nonmotor method" of the WISC-V was developed by the test publishers in response to the unique needs of clinicians during the COVID-19 pandemic and made available to Pearson subscribers via their online platform. The 'WISC-V International Nonmotor Full Scale Score' was developed to be used in place of the FSIQ composite when blocks and the Coding response booklet cannot be used during telehealth assessments (Pearson Clinical, 2020b). Although similar, the Nonmotor and Motor-free methods are comprised of different combinations of WISC-V subtests and thus both approaches were included in the present study in an effort overcome the limitations of fine motor responses and substitution when estimating IQ in children with CP.

The first aim of the present study was to investigate the ability of adolescents with CP aged 12 to 16 and varying levels of motor impairment to complete subtests of the WISC-V that require fine motor responses (pencil-to-paper Coding and Block Design) compared to similar subtests with reduced fine motor demands (Coding on Q-interactive and Visual Puzzles, respectively). It was hypothesized that more children with CP would be able to complete Coding on Q-interactive and Visual Puzzles, compared to pencil-to-paper Coding and Block Design, and they would achieve higher scores on the subtests with reduced motor demands. The second aim was to compare FSIQ scores derived using the "Traditional method" to alternative estimates of FSIQ which minimize motor demands, and investigate how scores vary across levels of CP severity. It was hypothesized that FSIQ scores derived using methods which minimize motor demands (Q-interactive, Nonmotor, Motor-free) would be higher than scores derived using the Traditional method, and benefit would be greater for children with more severe motor impairment.

Method

Participants

Participants were recruited from the Victorian Cerebral Palsy Register (VCPR) in Australia. In order to be included on the VCPR, a case must fulfil the criteria contained in the following definitional elements for CP. The VCPR defined CP as:

> an umbrella term for a group of disorders; a condition that is permanent but not unchanging; involves a disorder of movement and/or posture and of motor function; is due to a non-progressive interference, lesion, or abnormality; and the interference, lesion, or abnormality originates in the immature brain (Smithers-Sheedy et al., 2014, p. 324).

The process of participant recrtuitment, screening, and assessment is illustrated in Figure 1. There were 481 families on the VCPR with children born between January 2003 and December 2006 inclusive, living in Victoria and contactable by VCPR staff. Using an opt-out method, 420 families provided consent to be contacted by study personnel, 177 responded to the letter of invitation or a follow-up phone call and 75 were enrolled in the study. There were no significant differences between responders who participated and responders who declined participation in terms of age, $t(174) = .282$, $p = .778$, or gender distribution, $\chi^2_{(1)} = 1.60$, $p = .253$. However, there was a significant association between participation and CP severity as measured by the Gross Motor Function Classification System (GMFCS; Palisano et al., 1997). Responders classified as GMCFS Level IV or V were less likely to participate in the study, $\chi^2_{(4)} = 19.17$, $p = .001$. One adolescent declined to participate in the cognitive assessment and another was excluded due to a co-existent diagnosis of Sturge Weber Syndrome. Thus, 73 participants were included in the screening phase of this study.

Classifications

Clinical information related to diagnosis and associated impairments was obtained from the VCPR and parental report. CP subtypes were categorised as spastic, dyskinetic, or ataxic. Topographical distribution of motor impairment was classified as unilateral or bilateral (Krägeloh-Mann & Cans, 2009). Motor function was measured using the GMFCS, a five-level ordinal grading system (where Level V is the most severe motor impairment) based on self-initiated movement with particular emphasis on sitting and walking (Palisano et al., 1997). Fine motor ability was classified using the Manual Ability Classification System (MACS), a five-level ordinal grading system which provides a functional description of the child's ability to use their hands to handle objects in age-appropriate daily activities including eating, dressing, playing, and drawing or writing

(Eliasson et al., 2006). Additional information was obtained regarding gestational age at birth, birth weight and history of seizures. None of the participants had hearing impairment. Two of the adolescents had a visual impairment that hindered completion of some of the measures used in this study.

The study was approved by the La Trobe University Human Ethics Committee and The Royal Children's Hospital Human Research Ethics Committee.

Screening

Participants' ability to partake in neuropsychological assessment was determined using basic screening measures of pointing ability and expressive language, as described below.

Pointing Ability

The participants' ability to point to complex stimuli typically encountered in standardized cognitive assessment via an iPad was determined using the Peabody Picture Vocabulary Test, Fourth edition (PPVT-4; Dunn & Dunn, 2007). The first 10 items of the PPVT-4 were administered twice, and 'adequate point response' was defined as at least 8 out of 10 concordant responses between the first and second administration. These items were selected as they were considered to have low cognitive load, thereby increasing the likelihood that variations in responding were more probably due to physical limitations rather than cognitive ability. Sixty-nine of 73 participants (94.5%) demonstrated an adequate point response. Characteristics of the participants who were unable to provide an adequate point response are detailed in Table 1.

Expressive Language Ability

The first 10 items of the Picture Naming subtest from the Wechsler Preschool and Primary Scale of Intelligence, Fourth edition (WPPSI-IV; Wechsler, 2012) were

administered to determine each child's ability to provide a coherent verbal response. These items were selected because they were considered to have low cognitive load, thereby increasing the likelihood that unintelligible responses were due to difficulty with speech production rather than difficulty with language skills. 'Adequate verbal response' was defined as eight out of 10 coherent verbal responses. Sixty-eight of the 73 participants (93.2%) could be easily understood during the screen of verbal ability. Characteristics of participants unable to provide an adequate verbal response are outlined in Table 2.

Overall, 66 participants had both adequate point and adequate verbal response. Three participants had adequate point response only, and one had adequate verbal response only. Three participants had neither adequate verbal nor point responses, and were therefore excluded from any further assessment. The final sample comprised 70 adolescents with CP aged between 12 and 16 years (M = 14 years 6 months, SD = 10 months). Table 3 provides demographic and clinical characteristics of the sample.

Cognitive testing

General Intellectual Ability

The WISC-V (Wechsler, 2016) was used to assess general intellectual ability in children with adequate verbal and/or adequate point response. The WISC-V was administered using Q-interactive, a digital system available on iPad which replaces traditional pencil-to-paper administration. The examinee's iPad displays visual stimuli and captures touch/point responses. Equivalence of the pencil-to-paper and digital formats of the WISC-V was reported by Daniel et al. (2014) and Raiford et al. (2016), and this allowed the norms, reliability, and validity information gathered for the WISC-V pencil-to-paper version to be applied to the digital format. An exception was Block Design, with format effect (d = .20) larger than the cutoff established by the authors;

however the authors deemed this acceptable given minimal interaction of the examinee with the tablet during this subtest, and a previous study showed an effect size of only .02 (Daniel et al., 2014). The 10 primary subtests of the WISC-V and secondary subtest Letter-number sequencing were administered. All subtest scores are reported as scaled scores with a mean of 10 (*SD* = 3). FSIQ scores are reported as standard scores with a mean of 100 (*SD* = 15).

Processing Speed Tasks

The processing speed tasks for the Q-interactive format required substantive changes by the publisher to eliminate the writing requirements of the traditional pencil-to-paper format (Raiford et al., 2016). In this study, both versions of processing speed tasks were administered to participants with an adequate point response to investigate the impact of reduced fine motor requirements on a measure of processing speed. For both versions of Coding, the child is required to work within a two-minute time limit and use a key to match a symbol with its corresponding number. The pencil-to-paper version required a handwritten response, i.e. the child was required to draw the symbol that corresponds with each number. In contrast, during Coding on Q-interactive, the numbers appear on the screen one at a time, and the child is required to select the corresponding symbol from five multiple choice options using a touch/point response. Order of administration of the pencil-to-paper and digital versions of the processing speed tasks was counterbalanced.

An equivalence study by Raiford et al. (2016) found high correlations for raw and scaled scores between formats, and no meaningful effect of format on performance for Coding or Symbol Search in typically developing children. However, in February 2020 in an email to Q-interactive customers, Pearson reported an inconsistency with the rate at which digital Coding items advance from one to the next following a response (Pearson

Clinical, 2020a). This inconsistency in presentation rate inflated Coding subtest scores on fifth generation 2017 iPad models, which were used to collect data in the current study. Coding scaled scores were adjusted for each participant based on the results of an equating study conducted by Pearson for the purposes of this paper prior to all analyses (Personal Communication, Pearson). No other WISC-V subtests were affected by inconsistencies in presentation rate.

Procedure

Assessments took place between April 2018 and September 2019. The current study was part of a larger, longitudinal study investigating cognitive and behavioural outcomes in adolescents with CP. The adolescents were assessed with a neuropsychological test battery by a Clinical Neuropsychology Registrar. Assessments were conducted over one or two sessions at the Murdoch Children's Research Institute, La Trobe University Psychology Clinic or during a home visit.

Subtests of the WISC-V were administered using Q-interactive. In addition, the pencil-to-paper version of Coding was administered as described above. Fifty-four participants were able to complete all 10 primary subtests plus Letter-Number Sequencing and the pencil-to-paper tasks. For three participants who demonstrated adequate point response only, subtests requiring a verbal response (Similarities, Vocabulary, Digit Span, Letter-number sequencing) were omitted. Conversely, only verbal subtests were administered to the one adolescent who demonstrated adequate verbal response only. Twelve participants with adequate point and adequate verbal response completed some but not all of the subtests: two had behavioural difficulties; five assessments were limited by time constraints/low tolerance for assessment; and five participants did not complete pencil-to-paper tasks because they were unable to hold/manipulate the pencil.

Calculating FSIQ

Several procedures were used to calculate FSIQ, as outlined in Table 4. First, FSIQ was calculated according to the "Traditional method" which included the seven primary subtests Similarities, Vocabulary, Block Design, Matrix Reasoning, Figure Weights, Digit span and the pencil-to-paper version of Coding. Second, FSIQ was derived according to the "Q-interactive method" which included Coding on Q-interactive instead of the pencil-to-paper version, and Visual Puzzles was substituted for Block Design. Third, the WISC-V International Nonmotor Full Scale Score (the "Nonmotor method") was calculated, which included Visual Puzzles in place of Block design and Coding is omitted (Pearson Clinical, 2020b). Fourth, the Piovesana et al. (2019) "Motor-free method" was used to estimate FSIQ using their short-form of the WISC-V comprised of six subtests which require either a verbal or simple point response and no processing speed tasks.

Statistical analysis

All statistical analyses were conducted using SPSS version 26. Visual inspection of stem-and-leaf plots and Q-Q plots for each variable indicated a normal distribution. To investigate participants' ability to complete subtests that require fine motor responses, Fischer's exact tests were used to compare the number of participants able to respond to subtests that require fine motor responses compared to similar subtests with reduced motor demands (Pencil-to-paper Coding vs Q-interactive Coding; Block Design vs Visual Puzzles). Dependent samples *t*-tests were conducted to compare participants' scaled scores on these tasks.

To examine the influence of fine motor responses on measures of intelligence and determine the most appropriate way to measure FSIQ in adolescents with CP using the WISC-V, frequency analysis was conducted to determine the number of participants able

to obtain a FSIQ score according to the method of derivation. A repeated measures analysis of variance (ANOVA) was conducted with FSIQ scores derived from each method (FSIQ-Traditional method, FSIQ-Q-interactive method, FSIQ-Nonmotor method, FSIQ-Motor-free method) as the within-subjects factor, and CP severity (GMFCS Level I, Level II-III) as the between-subjects factor. A combined Level II-III group was formed to preserve cell sizes. Mauchly's test indicated that the assumption of sphericity was not met for the repeated measures ANOVA, therefore Greenhouse-Geisser corrected tests were reported for these analyses (Abdi, 2010). Post-hoc comparisons were conducted as a follow-up to a significant interaction using a series of one-way repeated measures ANOVAs to examine how FSIQ scores varied according to method of derivation within levels of CP severity.

Results

Sample Characteristics

Sample characteristics are presented in Table 3. Of the total 70 participants, 49 (70.0%) were male and 21 (30.0%) were female. On examining the motor types of CP, 87.1% of participants had spastic CP. Forty-four participants (58.6%) had bilateral CP. Severity of gross motor functional impairment was classified as Level I (24.3%), Level II (35.7%), or Level III (18.6%) for the majority of the sample. In terms of fine motor impairment, 17 participants (24.3%) were classified as Level I, 25 (35.7%) were Level II, 13 (18.6%) were Level III, and two participants (2.9%) were Level IV. The majority of participants were born at term (61.4%) and recorded birth weight in the normal range (67.1%). Less than a third reported a history of seizures.

Coding

Coding was administered to the 69 participants who demonstrated adequate pointing ability during the screening phase. Fifty-seven (83%) of these 69 participants were able to complete both the pen-to-paper and Q-interactive versions of Coding. There was an additional subset of eight participants (12%) who were able to complete Coding on Q-interactive, but not the pencil-to-paper version. The majority of this subset of participants had bilateral CP (88%), GMFCS $\geq$ II (88%), and MACS $\geq$ III (86%). Two participants were unable to complete either version of Coding. These two participants had bilateral CP, GMFCS Level III - IV, and MACS level IV – V. An additional two participants with adequate point ability did not attempt either version of Coding due to behavioural difficulties.

A Fisher's exact test demonstrated a significant association between mode of administration (pencil-to-paper or Q-interactive) and participants' ability to complete the Coding subtest, $\chi^2_{(1)} = 20.17$, $p = .001$.

For the 57 participants (GMFCS Level I-III) who were able to complete both pencil-to-paper and Q-interactive versions of Coding, a paired-samples t-test was conducted to investigate performance on Coding according to mode of administration. On average, participants performed better on the Q-interactive version ($M = 5.72$, $SD = 3.47$) compared to the pen-to-paper version ($M = 5.02$, $SD = 3.29$) of Coding, $t_{(56)} = 6.79$, $p = .002$, but the effect size of this difference was relatively small ($d = .21$).

Block Design and Visual Puzzles

Due to the standardized order of administration (Block Design is the first subtest administered, Visual Puzzles is the eighth subtest administered), there was a difference in the number of participants who attempted each task. Sixty-three (91%) of 69 participants

who had adequate pointing ability were able to respond to Block Design. In contrast, five participants who demonstrated adequate pointing ability during the screening phase were unable to manipulate the blocks due to poor fine motor control during the sample items and thus the subtest was discontinued. On the other hand, 63 (98%) of 64 participants who attempted the Visual Puzzles subtest were able to respond to the task. For four of the participants who completed Block Design but not Visual Puzzles, the secondary subtests of the WISC-V were not administered due to time constraints/low tolerance for assessment. One participant refused to attempt either task.

In total, 58 participants (GMFCS Level I-III) were able to complete both Block Design and Visual Puzzles. There were five additional participants who were able to complete Visual Puzzles but not Block Design. These five participants had bilateral CP, GMFCS $\geq$ II, and MACS $\geq$ III. A Fischer's exact test did not reveal an association between the participants' ability to obtain a measure of their visuospatial ability according to the task administered (Block Design or Visual Puzzles), $\chi^2_{(1)} = 9.82$, $p = .094$.

For the 58 participants (GMFCS Level I-III) who completed both Block Design and Visual Puzzles, a paired sample *t*-test showed that participants performed better on Visual Puzzles ($M = 6.53$, $SD = 3.28$) compared to Block Design ($M = 5.86$, $SD = 3.13$), $t_{(57)} = 2.88$, $p = .006$, but effect size of the difference was relatively small ($d = .21$).

FSIQ

The total number of participants who were able to obtain a FSIQ score according to the method of derivation is provided in Table 5. The number of participants with CP GMFCS Level I and Level II who were able to obtain a FSIQ remained relatively stable across the methods. Three participants with GMFCS Level II who were able to obtain FSIQ using the Traditional method did not complete the Visual Puzzles subtest due to

time constraints, and therefore were unable to obtain FSIQ scores using any of the alternative methods. Compared to the Traditional method, the number of CP GMFCS Level III participants who obtained a FSIQ increased by four when motor demands were reduced using the Q-interactive, Nonmotor, and Motor-free methods. The only participant with CP GMFCS Level IV was able to obtain a FSIQ score using the Nonmotor and Motor-free methods. The sole participant with CP GMFCS Level V obtained a FSIQ using the Nonmotor method only.

A total of 54 participants were able to obtain a FSIQ score using all four methods of derivation. This included 25 participants with GMFCS Level I, 20 participants with GMFCS Level II and 9 participants with GMFCS Level III. There were no significant differences in FSIQ scores between participants with GMFCS Level II and Level III, and they were therefore combined to form a Level II-III group in order to preserve cell size.

There was a significant main effect for method, $F_{(2.29, 119.17)} = 64.01$, $p < .001$, $\eta_p^2 = .55$, which reflected an overall difference between IQ scores derived from the different methods. Bonferroni adjusted pair-wise comparisons indicated that FSIQ scores in all non-Traditional methods were higher than the FSIQ score derived from the Traditional method, and scores derived specifically from the Nonmotor and Motor-Free methods were the highest (see Table 6). There was also a significant interaction between FSIQ-method and CP severity, $F_{(2.29, 119.17)} = 7.50$, $p < .001$, $\eta_p^2 = .13$.

For participants identified as GMFCS Level I, a post-hoc follow-up showed a statistically significant difference between FSIQ scores among this subgroup, $F_{(2.63, 63.11)} = 12.84$, $p < .001$, $\eta_p^2 = .35$. Bonferroni adjusted pair-wise comparisons indicated that FSIQ scores derived using the Nonmotor and Motor-Free methods were the highest (see Table

6). Compared to the Traditional method, there was a mean difference of 3.00 – 3.36 points when FSIQ was derived using the Nonmotor or Motor-free methods, respectively.

Similarly, a post-hoc comparison showed a statistically significant main effect among participants identified as GMFCS Level II-III, $F_{(1.91, 53.40)} = 64.06$, $p < .001$, $\eta_p^2 = .70$. Bonferroni adjusted pair-wise comparisons indicated that FSIQ scores in all non-Traditional Methods were higher than the FSIQ score derived from the Traditional method, and scores derived specifically from the Nonmotor- and Motor-Free methods were the highest (see Table 6). Compared to the Traditional method, there was a mean difference of 6.00 – 6.21 points when FSIQ was derived using the Motor-free and Nonmotor methods, respectively. There were no other significant differences between FSIQ scores.

Discussion

The present study compared motor-free methods of assessing cognitive ability in adolescents with CP to the traditional method using the WISC-V. Clinical experience and previous research suggests cognitive assessment is challenging in CP because intelligence tests require good fine motor skills and rapid responses (Kurmanaviciute & Stadskleiv, 2017). To overcome the challenges of IQ testing in CP, the current study employed touchscreen administration of the WISC-V to replace handwritten responses as well as altenative methods for deriving a FSIQ score based on motor-free subtests. The current findings indicate adolescents with CP have more difficulty on subtests of the WISC-V that require quick fine motor responses compared to similar subtests with reduced motor demands. As such, adolescents with CP are at an inherent disadvantage when cognitive ability is assessed according to the Traditional method of the WISC-V, and benefit significantly from alternative estimates that minimize motor requirements. Taken

together, these findings suggest that clinicians should employ the Nonmotor or Motor-free methods for neuropsychological assessments of cognitive ability in adolescents with CP.

The hypothesis that adolescents with CP would perform better on WISC-V subtests with reduced motor demands was supported in the current study. It is important to note that employing subtests with reduced motor demands improves the accessability of the WISC-V for adolescents with CP. An additional 7–12% of participants were able to respond to Visual Puzzles and Coding on Q-interactive compared to Block Design and pencil-to-paper Coding, respectively. Even for those participants with CP who were able to complete the WISC-V subtests in the traditional manner, performance was marginally but significantly better on tasks with reduced motor demands. When considering how subtests that require fine motor responses influence estimates of intelligence, the difference in FSIQ scores appears to be driven by the inclusion (or omission) of Coding. Although the difference at the subtest level was small, it appeared to have a substantial influence on estimates of FSIQ and thus may impact decisions based on the outcome of the cognitive assessment.

Consistent with our hypothesis, the short-forms of the WISC-V which minimized motor demands produced higher FSIQ scores than the Traditional method, and the benefit was greater for those with more severe motor impairment (i.e. GMFCS Level II-III). The Traditional method underestimated FSIQ by about 3 points for participants functioning at GMFCS Level I, and up to 6 points for those functioning at GMFCS Level II-III. While the classification system relates to gross motor function, findings suggest participants were also having trouble with fine motor skills. It would appear that participants with more severe motor impairment were disadvantaged to a greater degree by the fine motor component of pencil-to-paper Coding that required them to grip and manipulate a pencil.

Like many other functional skills, the development of the fine motor control required for handwriting is delayed in CP even for those with mild motor impairment and good hand function (Öhrvall et al., 2010). A 13-year longitudinal study of hand function in children with CP showed that while speed improved over time on measures of grip and object manipulation, it nonetheless remained four times slower than typically developing controls (Eliasson, Forssberg, et al., 2006). Therefore, strategies like adjusting the requirements of a task or providing alternative measures are essential to accommodate adolescents with CP and minimize disadvantage due to poor fine motor control on intelligence tests.

While results indicated some benefit from Coding on Q-interactive compared to the pencil-to-paper version at the subtest level, FSIQ derived using the Q-interactive method remained significantly below scores derived using the Nonmotor and Motor-free methods. These findings support the contention that processing speed tasks in estimates of intelligence place adolescents with CP at an inherent disadvantage due to the nature of their motor disorder. Although the motor demands of Coding on Q-interactive were reduced compared to the pencil-to-paper version, it still requires point responses that are accurate and in quick succession, which becomes fundamentally more difficult with more severe motor impairment.

The current findings may be interpreted in the context of Fitts' Law (Fitts, 1954), which describes the trade-off between speed and accuracy such that smaller targets lead to slower movements when accuracy is important. A previous study investigating speed and accuracy on a simple pointing task on an iPad showed that adolescents with CP were slower than the healthy controls at all levels of task difficulty (Bertucco & Sanger, 2014). Within the CP group, movement time slowed as the targets became smaller. Similarly, Fernani et al. (2017) identified severity of motor impairment as a significant predictor of

performance on a computerized task such that higher GMFCS level was associated with slower movement time when the target was smaller and demanded better accuracy. Given the small size of the targets on the Q-interactive version of Coding, and their close proximity to other response options, it is likely that the adolescents with CP were constrained to reduce the speed of their movements in order to achieve accurate responses. Future iterations should consider adapting the format of Coding on Q-interactive to increase target size, or alternatively, use a non-motor measure of processing speed.

Clinical Implications

Given the WISC-V is the most widely used intelligence test (Flanagan & Alfonso, 2017), evidence that the Traditional method is underestimating IQ in adolescents with CP is concerning. Accurate characterisation of intellectual abilities is crucial given cognitive assessments are often required in CP to determine eligibility for funding, support and entry into educational programs and settings. Based on the current findings, the Nonmotor and Motor-free methods appear to be the most appropriate way to measure cognitive ability in adolescents with CP using the WISC-V. An issue with the Nonmotor method is that Pearson did not provide information regarding the reliability or validity of this composite score. In contrast, the Motor-free method has been shown to have strong psychometric properties comparable to that of the WISC-V (Piovesana et al., 2019). A particular benefit of the Motor-free method is that it requires the administration of only six WISC-V subtests, which can then be combined to produce a Motor-free IQ score and three domain-level scores (Verbal Comprehension, Perceptual Reasoning, and Working Memory; Piovesana et al., 2019). In doing so, the Motor-free method reduces assessment time, which can be a limiting factor when assessing children with CP due to behavioural difficulties or low tolerance, and provides a more nuanced cognitive profile.

Although the Motor-free method appears to be the most appropriate method currently available for assessing IQ in adolescents with mild to moderate CP using the WISC-V, it is not without limitation. The Processing Speed Index has been omitted due to the motor requirements of subtests in this domain but, in doing so, an element of IQ – *cognitive* processing speed – has been overlooked which may be particularly relevant in CP due to the nature of the initial brain injury. Predominant white matter injury is the most common pathogenic pattern identified on neuroimaging in CP cases (Krägeloh-Mann & Horber, 2007); and white matter injury has been associated with lower processing speed scores on the WISC in adolescents who were born very preterm (Soria-Pastor et al., 2008). While some studies using inspection time tasks with minimal motor demands indicate slower cognitive processing speed in individuals with CP compared to typically developing controls (Kaufman et al., 2014; Shank et al., 2010), others indicate no significant difference between the two groups (Hakkarainen et al., 2012). Future research may consider developing an inspection time task to be used as a substitute for the processing speed tasks of the WISC-V.

On the other hand, although processing speed is typically included as a 'broad ability' in models of intelligence (Carroll, 1993; McGrew, 2005); exploratory and confirmatory factor analyses have shown that the Processing Speed Index has the weakest loading on the general intelligence factor of the WISC-V in both normative (Canivez et al., 2016) and clinical samples (Canivez et al., 2020; Stephenson et al., 2021). Moreover, other standardised intelligence tests (e.g. Kaufman Assessment Battery for Children - Second edition (KABC-II), Stanford-Binet Intelligence Scales - Fifth edition) do not assess processing speed at all. In fact, the inclusion of time bonuses on the KABC-II resulted in a model that provided a poorer fit and thus weaker explanations for the cognitive abilities measured by the test (Reynolds et al., 2007). There is evidence to

suggest that the effect of processing speed on intelligence is mediated almost entirely by working memory such that improvements in processing speed lead to increased working memory capacity, which in turn, is related to improvements in fluid intelligence (Fry & Hale, 1996). In fact, working memory was shown to be highly related to general intelligence, and the impact of processing speed on intelligence was neglible after accounting for the short-term storage component of working memory (Colom et al., 2004, 2008).

Limitations

Given the aim of this study was to determine how the reliance on fine motor responses impacted performance on intelligence tests, it may have seemed more intuitive to classify severity of motor impairment according to MACS rather than GMFCS. Unfortunately, missing MACS data for 18% of the sample precluded this analysis. Interestingly, however, Fernani et al. (2017) found that MACS level was not related to movement time on a computerized task. In fact, GMFCS was the only factor that significantly influenced movement time. The authors posited that MACS was not sensitive since their participants were classified as MACS Level I-III and therefore were all sufficiently able to manipulate the mouse to complete the task (Fernani et al., 2017). We may have expected a similar result since the composition of our sample was comparable, however, given the differences between mouse-pointing and pointing onscreen, this would warrant further research. Moreover, previous studies have shown a significant association between GMFCS level and cognitive ability. Not only is GMFCS level related to the type of initial brain injury in CP (Himmelmann & Uvebrant, 2011), but children with more severe impairment and restricted mobility receive less sensory input from their environment, which in turn limits cognitive development (Bottcher, 2010).

Another limitation of the current study was the under-representation of adolescents with more severe motor impairment. Four adolescents with CP GMFCS Level IV-V participated in the screening phase, and unfortunately only two went on to the cognitive assessment. Neither of these two participants were able to complete Block Design or either version of Coding due to their level of motor impairment which precluded their inclusion in the subtest level analysis and comparison of FSIQ scores. However, it is encouraging and important to note that we were able to obtain FSIQ scores for these two participants using the Nonmotor or Motor-free methods, when previously they may have been considered 'unassessable'. Despite the terms "Nonmotor" and "Motor-free", the six subtests included in these methods require either a verbal or point response which demand a certain level of motor ability. Indeed, many adolescents with CP and severe motor impairment are unable to point or produce a verbal response, highlighting that the Nonmotor and Motor-free methods are more likely to be an appropriate solution for those with mild to moderate motor impairment. Future research should consider validating the WISC-V for use with alternative response modes to accommodate participants with more severe motor impairment. Kurmanaviciute and Stadskleiv (2017) found that alternative response modes including gaze pointing and partner-assisted scanning did not influence how typically developing children scored on tests of verbal comprehension and visuospatial reasoning. While alternative response modes may be more time consuming, they provide the opportunity for children with motor impairment to demonstrate their true cognitive abilities (Kurmanaviciute & Stadskleiv, 2017).

A further limitation is that the iPad presentation of the Coding subtest was susceptible to technical difficulites. Due to the presentation rate inconsistency and inflation of scores on Q-interactive, Coding scaled scores were adjusted for each

participant based on the results of an equating study (Personal Communication, Pearson). The adjustment was based on their observation that, regardless of Coding format, 65-70% of examinees with clinical conditions typically obtain Coding scores that are equal to or below the mean scaled score of the eight primary subtests that are not Processing Speed tasks. Based on their analysis, Coding scores obtained on Q-interactive were generally higher than the mean of the eight subtests due to the presentation rate inconsistency, and the differences across formats became larger as Coding ability level increased. This statistical adjustment reduced the mean score for Coding on Q-interactive by 1.3 scaled score points and thus influenced the magnitude of difference observed across modes of administration. Given findings that Coding on Q-interactive was more accessible to adolescents with motor impairment than the pen-to-paper version, future research should revisit a digital version of Coding and explore the impact of mode of administration on IQ test scores in CP.

Conclusion

Cognitive assessment using the WISC-V is challenging in children with CP because the standardised procedures do not account for their motor coordination difficulties. Subtests that require quick fine motor responses, Coding and Block Design, place adolescents with CP at an inherent disadvantage. The current findings have shown that the Traditional method significantly underestimates FSIQ by three to six points depending on GMFCS level compared to non-Traditional methods with reduced motor demands. Furthermore, these findings suggest that the Nonmotor or Motor-free methods, which combine six motor-free subtests of the WISC-V to estimate IQ, are the most appropriate methods to assess intelligence in adolescents with mild to moderate CP. Accurate assessment of intelligence in CP is particularly important in the context of intellectual disability, where the outcome of a cognitive assessment can influence

decision-making regarding support, school placement and interventions, and potentially have lifelong consequences. Future research may consider adapting the WISC-V for use with alternative response modes to further improve access for children with CP and severe motor impairment.

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Table 1

Characteristics of participants with cerebral palsy unable to provide adequate point response

ID	Motor Type	Topographical pattern	GMFCS level	MACS level
1	Dyskinetic	Bilateral	III	IV
5	Dyskinetic	Bilateral	V	V
24	Spastic	Bilateral	IV	IV
34	Spastic	Bilateral	III	-[a]

Note. GMFCS = Gross Motor Function Classification System; MACS = Manual Ability Classification System

[a]MACS data was not available for this participant

Table 2

Characteristics of participants with cerebral palsy unable to provide an adequate verbal response

ID	Motor Type	Topographical pattern	GMFCS level	MACS level
1	Dyskinetic	Bilateral	III	IV
5	Dyskinetic	Bilateral	V	V
10	Dyskinetic	Bilateral	III	III
24	Spastic	Bilateral	IV	IV
65	Spastic	Bilateral	II	-[a]

Note. ID = Participant identification number; GMFCS = Gross Motor Function Classification System; MACS = Manual Ability Classification System

[a]MACS data was not available for this participant

Table 3

Participant characteristics

	Participants	
	n	%
Gender		
Male	49	70.0
Female	21	30.0
Motor type		
Spastic	61	87.1
Dyskinetic	8	11.4
Ataxic	1	1.4
Topography		
Unilateral	29	41.4
Bilateral	41	58.6
Gross motor function (GMFCS)		
Level I	27	38.6
Level II	24	34.3
Level III	17	24.3
Level IV	1	1.4
Level V	1	1.4
Fine motor ability (MACS)		
Level I	17	24.3
Level II	25	35.7
Level III	13	18.6
Level IV	2	2.9
Unknown	13	18.6
Gestational age at birth		
<28 weeks	6	8.6
28 - <32 weeks	8	11.4
32 - <37 weeks	12	17.1
>37 weeks	43	61.4
Unknown	1	1.4
Birth weight		
<1000g	5	7.1

1000 – 1499g	7	10.0
1500 – 2499g	8	11.4
>2500g	48	68.6
Unknown	2	2.9
History of seizures		
Yes	20	27.1
No	50	72.8

Note. GMFCS = Gross Motor Function Classification System; MACS = Manual Ability Classification System

Table 4

Calculating alternative Full Scale Intelligence Quotient scores

Traditional method	Q-interactive method	Non-motor method[a]	Motor-free method[b]
Similarities	Similarities	Similarities	Similarities
Vocabulary	Vocabulary	Vocabulary	Vocabulary
Block Design	Visual Puzzles	Visual Puzzles	Visual Puzzles
Matrix Reasoning	Matrix Reasoning	Matrix Reasoning	Figure Weights
Figure Weights	Figure Weights	Figure Weights	Digit span
Digit span	Digit span	Digit span	Letter-Number Sequencing
Coding (pencil-to-paper)	Coding (Q-interactive)		

[a] Pearson Clinical. (2020b). *WISC-V International Nonmotor Full Scale Score*. https://qglobal.pearsonclinical.com/qg/viewRestrictedPdfS.seam

[b] Piovesana, A. M., Harrison, J. L., & Ducat, J. J. (2019). The Development of a Motor-Free Short-Form of the Wechsler Intelligence Scale for Children–Fifth Edition. *Assessment*, *26*(8), 1564–1572. https://doi.org/10.1177/1073191117748741

Table 5

Total number of participants with cerebral palsy able to obtain a Full Scale Intelligence Quotient according to method of derivation and severity of motor impairment

Severity (GMFCS)	*n*	Traditional method	Q-interactive method	Non-motor method[a]	Motor-free method[b]
Level I	27	25	26	26	26
Level II	24	23	20	20	20
Level III	17	9	13	13	13
Level IV	1	0	0	1	1
Level V	1	0	0	1	0
Total	70	57	59	61	60

Note. GMFCS = Gross Motor Function Classification System.

[a] Pearson Clinical. (2020b). *WISC-V International Nonmotor Full Scale Score*. https://qglobal.pearsonclinical.com/qg/viewRestrictedPdfS.seam

[b] Piovesana, A. M., Harrison, J. L., & Ducat, J. J. (2019). The Development of a Motor-Free Short-Form of the Wechsler Intelligence Scale for Children–Fifth Edition. *Assessment*, *26*(8), 1564–1572. https://doi.org/10.1177/1073191117748741

Table 6

Means and standard deviations of Full Scale Intelligence Quotient scores according to method of derivation and severity of motor impairment in adolescents with cerebral palsy who were able to complete all subtests of the Wechsler Intelligence Scale for Children (5th edition).

Severity (GMFCS)	*n*	Traditional method M*(SD)*	Q-interactive method M*(SD)*	Non-motor method[a] M*(SD)*	Motor-free method[b] M*(SD)*
Level I	25	90.64 *(14.54)*	91.88 *(14.62)*	93.64 [c,d] *(13.78)*	94.00 [c,d] *(14.01)*
Level II-III combined	29	73.62 *(15.16)*	75.24 [c] *(15.88)*	79.83 [c,e] *(15.25)*	79.62 [c,e] *(16.64)*
Total	54	81.50 *(17.50)*	82.94 [c] *(17.33)*	86.22 [c,e] *(16.04)*	86.28 [c,e] *(16.96)*

Note. GMFCS = Gross Motor Function Classification System

[a] Pearson Clinical. (2020b). *WISC-V International Nonmotor Full Scale Score*. https://qglobal.pearsonclinical.com/qg/viewRestrictedPdfS.seam

[b] Piovesana, A. M., Harrison, J. L., & Ducat, J. J. (2019). The Development of a Motor-Free Short-Form of the Wechsler Intelligence Scale for Children–Fifth Edition. *Assessment*, *26*(8), 1564–1572. https://doi.org/10.1177/1073191117748741

[c] Scores are significantly different to the Traditional method at $p < .001$

[d] Scores are significantly different to the Q-interactive method at $p < .05$

[e] Scores are significantly different to the Q-interactive method at $p < .001$

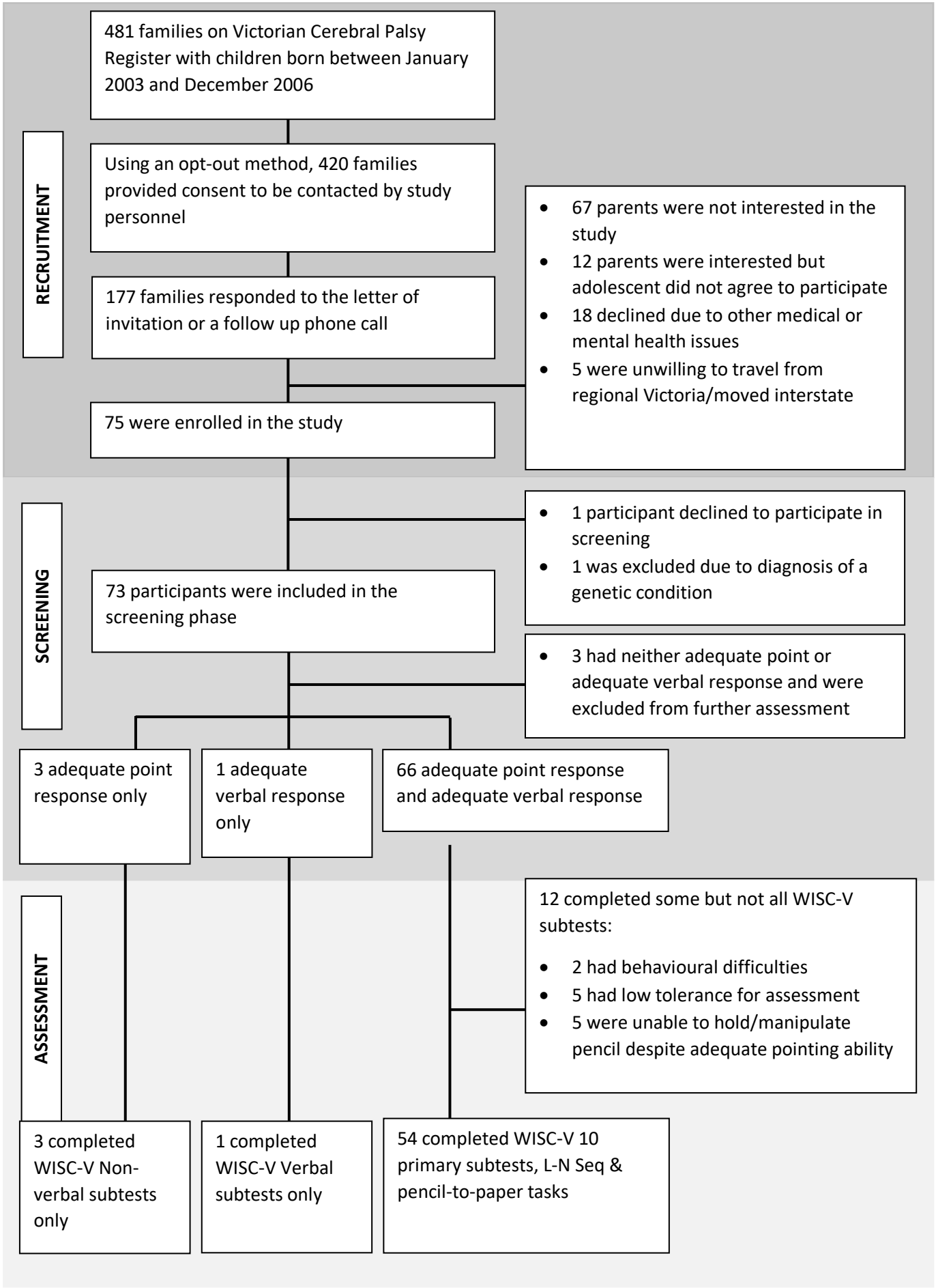
RECRUITMENT
481 families on Victorian Cerebral Palsy Register with children born between January 2003 and December 2006
Using an opt-out method, 420 families provided consent to be contacted by study personnel
177 families responded to the letter of invitation or a follow up phone call
75 were enrolled in the study
67 parents were not interested in the study
12 parents were interested but adolescent did not agree to participate
18 declined due to other medical or mental health issues
5 were unwilling to travel from regional Victoria/moved interstate
SCREENING
73 participants were included in the screening phase
1 participant declined to participate in screening
1 was excluded due to diagnosis of a genetic condition
3 had neither adequate point or adequate verbal response and were excluded from further assessment
3 adequate point response only
1 adequate verbal response only
66 adequate point response and adequate verbal response
ASSESSMENT
12 completed some but not all WISC-V subtests:
2 had behavioural difficulties
5 had low tolerance for assessment
5 were unable to hold/manipulate pencil despite adequate pointing ability
3 completed WISC-V Non-verbal subtests only
1 completed WISC-V Verbal subtests only
54 completed WISC-V 10 primary subtests, L-N Seq & pencil-to-paper tasks

Figure 1

Flow diagram of participant recruitment, screening and assessment

Note: L-N Seq = Letter Number Sequencing; WISC-V = Wechsler Intelligence Scale for Children – Fifth edition

Chapter 4: WISC-V Motor-Free Cognitive Profile in Adolescents with Mild to Moderate Cerebral Palsy

Preamble to empirical paper 2

The preceding chapter determined that cognitive assessment using the Wechsler Intelligence Scale for Children – Fifth edition (WISC-V) in the traditional manner disadvantaged adolescents with CP and significantly underestimated Full Scale Intelligence Quotient (FSIQ) compared to the Nonmotor and Motor-free methods which minimised motor demands. Given findings that FSIQ scores derived using the Nonmotor and Motor-free methods were not significantly different, the Motor-free method was recommended for use with adolescents with CP based on its strong psychometric properties and capacity to produce three primary index scores (Piovesana et al., 2019). Subsequently, the following published paper characterised the motor-free cognitive profile of adolescents with mild to moderate CP using the WISC-V and explored clinical factors and associated conditions related to cognitive impairment. Findings provide new insights regarding cognitive strengths and weaknesses as well as risk factors for cognitive impairment in adolescents with CP.

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WISC-V Motor-Free Cognitive Profile and Predictive Factors in Adolescents with Cerebral Palsy

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Abstract

Background: The most commonly used intelligence tests – the Wechsler Scales – do not provide standardised procedures for assessing children with motor impairment, and as a result, may underestimate the intelligence quotient (IQ) of young people with CP.

Aims: To characterise a motor-free cognitive profile of adolescents with CP using the Wechsler Intelligence Scale for Children – Fifth edition (WISC-V) and explore the influence of clinical factors on cognitive abilities.

Methods and procedures: The WISC-V was used to assess cognitive abilities in 70 adolescents (M = 14 years 6 months, SD = 10 months). Sixty-six adolescents (Gross Motor Function Classification System (GMFCS) Level I, n = 26 ; II, n = 23; III, n = 15; IV, n = 1; V, n = 1) obtained either a Motor-free IQ or index score using the motor-free method.

Outcomes and results: MFIQ and index scores fell below the normative data and rates of borderline and impaired cognitive abilities were significantly higher in the CP group. Scores showed an uneven cognitive profile with a relative strength in verbal abilities. Severity of motor impairment and small for gestational age (SGA) were associated with lower IQ scores. A history of seizures was related to lower verbal abilities.

Conclusions and implications: Cognitive abilities of adolescents with CP are significantly below expectation compared to normative data. Severity of motor impairment, SGA, and seizures need to be recognised by health professionals as risk factors for cognitive impairment. A substantial proportion of adolescents showed borderline cognitive abilities, constituting a group with CP which are relatively neglected in the literature.

Keywords: adolescents, cognitive assessment, cerebral palsy, motor-free, WISC-V

What this paper adds

This study is the first to use motor-free administration of the Wechsler Intelligence Scale for Children – Fifth edition (WISC-V) to characterise the cognitive profile of a cohort of adolescents with Cerebral Palsy (CP). Standardised administration of the WISC-V assumes good fine motor ability, and consequently, places children and adolescents with CP at a disadvantage. While the motor-free administration is not without limitation, the present study provides precedent for its use in the context of CP or other developmental disorders characterised by motor impairment.

The current findings highlighted an association between cognitive impairment across domains and clinical factors including severity of motor impairment, small for gestational age (SGA) and seizures, which need to be recognised as risk factors by health professionals. While the relation between severe motor impairment and cognitive impairment is well-established, the present study confirms this association regardless of the assessment method. Similarly, seizures have previously been associated with a decrement in overall IQ; however, the present study suggests a significant association between seizures and lower verbal IQ specifically. In contrast, few studies have investigated the impact of SGA on cognition in CP. The present findings suggest the early effects of SGA on growth and cognitive development are associated with lower cognitive performance in adolescence.

While intellectual disability is generally understood to be common in CP, the current study highlighted a substantial proportion of adolescents with borderline cognitive abilities, i.e., their scores fell below the 'normal' range but above the cut-off for intellectual disability. This group often goes unrecognised and is at-risk for negative academic, social, and behavioural outcomes.

Introduction

Cerebral Palsy (CP) is a permanent disorder of movement and posture attributed to non-progressive disturbance of the developing foetal or infant brain (Rosenbaum et al., 2007). As an umbrella term, CP encompasses a wide range of types, topographies and degrees of motor impairment. Similarly, cognitive abilities vary widely in persons with CP and can range from severe intellectual disability to age-appropriate cognitive functioning. Accurate assessment of intellectual abilities is challenging in CP because the standardised administration procedures of the most commonly used intelligence tests – the Wechsler scales – fail to account for the core feature of motor impairment in CP. Indeed, assessments using such measures have been shown to significantly underestimate the Intelligence Quotient (IQ) of children and adolescents with CP (Sherwell et al., 2014; Yin Foo et al., 2013). This highlights the need for a 'motor-free' method of assessing cognitive abilities using the Wechsler Intelligence Scale for Children (Fifth edition; WISC-V) to accurately characterise the cognitive profile of children and adolescents with CP.

A recurring limitation in the literature is that the cognitive abilities of many children with CP are not assessed because they do not have the motor skills required to respond to intelligence tests. Previous studies suggest one-third of children with CP are unable to respond to tests that require quick, fine motor responses (Sherwell et al., 2014; Sigurdardottir et al., 2008). However, a Norwegian CP register study suggests this figure is much higher; only 29% of cases on the register had their intellectual abilities assessed using a cognitive test (Andersen et al., 2008). In cases where a child does not have the motor skills required to respond to an intelligence test, intellectual disability is commonly assumed based on clinical judgement (Andersen et al., 2008; Himmelmann et al., 2006) or severity of motor impairment (Hutton et al., 2002). However, Sigurdardottir et al. (2008) showed that 20% of preschoolers with CP who were unable to respond to the Wechsler Primary and Preschool Scale of

Intelligence (WPPSI) achieved a developmental quotient in the normal range (i.e. >85) on tests with fewer motor demands (e.g. Reynell-Zinkin Developmental Scale). This further highlights that assessments conducted using standardised procedures of the WPPSI or WISC, even when attempted, will not necessarily be an accurate reflection of the child's cognitive abilities due to the reliance on motor responses.

Cognitive assessments in CP are not straightforward even for those who can perform the tasks; mild motor impairment can impact test performance and 'normal range' IQ scores may mask subtle deficits or areas of relative strength. Gonzalez-Monge et al. (2009) showed that while Full Scale (FSIQ) and verbal intelligence remained fairly stable over 7 years in a group of children with CP, there was a significant decline in non-verbal intelligence over time. Similar findings of an uneven cognitive profile characterised by stronger verbal than non-verbal skills have often been reported in the literature (Ito et al., 1997; Muter et al., 1997; Sigurdardottir et al., 2008). However, the relative weakness in non-verbal skills seen in these studies may be driven in part by the use of Block Design and Coding – non-verbal subtests of the WPPSI and WISC that require quick, fine motor responses. These subtests have been shown to disadvantage children with CP and underestimate cognitive abilities when included in measures of IQ (Sherwell et al., 2014). An approach to overcoming this issue has been suggested by Piovesana et al. (2019) who developed a motor-free short-form of the WISC-V, which allows the derivation of a Motor Free IQ (MFIQ) and three index scores based on six motor-free subtests. Coceski et al. (2020) showed the motor-free method was the most appropriate way to measure IQ in adolescents with CP using the WISC-V, and therefore, characterising a cognitive profile among adolescents with CP using this method is warranted.

In addition to the issue of accuracy of conventional tests of IQ, a range of clinical factors have been identified which may lead to differences in IQ in CP. These factors can be broadly categorised as CP-related factors or associated factors. CP-related factors include

motor type (spastic, dyskinetic, ataxic, hypotonic), topography of motor impairment (unilateral, bilateral), and severity of motor impairment classified according to the Gross Motor Classification System (GMFCS; Palisano et al., 1997). Higher rates of cognitive impairment have been associated with spastic quadriplegic (Himmelmann et al., 2006; Reid et al., 2018; Sigurdardottir et al., 2008) or hypotonic CP (Delacy & Reid, 2016), bilateral motor impairment (Sigurdardottir et al., 2008; Smits et al., 2011), and greater severity of motor impairment (Sigurdardottir et al., 2008; Smits et al., 2011; Stadskleiv et al., 2018). These findings tend to be explained by the inference that bilateral CP and severe motor impairment are associated with widespread brain injury and therefore limited intellectual development.

In addition to motor impairment, other factors that have been linked to poorer cognitive function in CP include seizure history, gestational age at birth, and birth weight. There is strong evidence that links seizure history with poorer cognitive outcomes in CP (Cheng et al., 2013; Muter et al., 1997; Sigurdardottir et al., 2008; Stadskleiv et al., 2018). In a study of children and young adults with CP between the ages of 5 and 20 years, Cheng et al. (2013) observed that seizure history had a deleterious effect on cognition over time. Adolescents over the age of 15 years with CP and seizure history were more likely to have IQ < 80 than children under the age of 15 with CP and seizure history.

The relationship between gestational age at birth and cognitive outcomes in CP remains unclear, with inconsistent findings in the literature. Hemming et al. (2008) examined a UK CP register of over 4000 cases and found that gestational age was not associated with cognitive impairment in unilateral spastic CP, except for in extremely pre-term cases born before 28 weeks gestation. However, infants born at term with bilateral CP were more likely to have severe intellectual impairment (IQ < 50) than those born preterm (Hemming et al., 2008). The authors suggested the higher rates of severe intellectual impairment with

increasing gestational age in bilateral CP may reflect a reduced ability for the brain to reorganise when there is more diffuse injury at later gestation.

In contrast, another study of children with bilateral spastic CP indicated that those born pre-term and term were similar in terms of overall cognitive ability and verbal skills; however, preterm birth was associated with poorer non-verbal skills at 6 years of age (Pagliano et al., 2007). More specifically, visuomotor skills were significantly lower in the preterm group than visuoperceptual (i.e. non-motor) skills. The authors suggest this was a direct result of prematurity – the truncated intrauterine period may be insufficient to allow reorganisation of visual pathways after the initial brain injury. Moreover, eye movements which impact perception were more often impaired in preterm children, in line with Himmelmann et al., (2006) who showed that earlier gestational age at birth is associated with more severe motor deficits.

Similarly, the relationship between birth weight and cognition in CP has shown mixed evidence in the literature. Cheng et al. (2013) reported birth weight was not associated with cognitive abilities in their sample of children and young adults with CP. In contrast, birth weight was identified as a significant predictor of severe cognitive impairment by Hemming et al. (2008). These inconsistent findings are likely explained by the way birth weight was measured across these studies. Cheng et al. examined absolute birth weight for associations with cognitive abilities, while Hemming et al. calculated standardised birth weight for gestational age. Infants born small for gestational age (SGA; birth weight $<10^{th}$ percentile of expected weight for gestational age) were at increased risk of severe intellectual impairment relative to those born with appropriate weight for gestational age (AGA; Hemming et al., 2008).

The present study aimed to characterise the cognitive profile of a cohort of adolescents with CP using motor-free administration of the WISC-V. Given previous

findings of higher rates of cognitive impairment and uneven cognitive profiles in CP, it was hypothesised that adolescents with CP would perform below expectations across domains compared to normative data and demonstrate a relative weakness in the non-verbal domain. A secondary aim of the current study was to explore the association between cognitive abilities in CP, assessed with motor-free administration of the WISC-V, and clinical factors including CP type, severity of motor impairment, topography of motor impairment, seizure history, gestational age, birth weight and SGA. Although previous research provided some evidence that these factors are related to cognitive abilities in CP, it was deemed important to examine their relatedness using the motor-free cognitive profile to establish stability of effects regardless of the assessment method. It was expected that bilateral motor impairment, severe motor impairment, seizure history, and SGA would be associated with poorer cognitive abilities. No specific prediction was made for gestational age or birth weight due to mixed evidence in the literature.

Materials and methods

Participants

Participants were recruited through the Victorian Cerebral Palsy Register (VCPR). VCPR staff identified and contacted 481 parents/carers of children who were born between January 2003 and December 2006 inclusive and were living in Victoria. Using an opt-out method, 420 families provided consent to be contacted by study personnel, 177 responded to the subsequent letter of invitation or follow-up phone call, and 75 adolescents were enrolled in the study. There were no significant differences between children of responders who agreed and those who declined to participate in terms of age, $t(174) = .282$, $p = .778$, or gender distribution, $\chi^2_{(1)} = 1.60$, $p = .253$. However, there was a significant association between participation and CP severity as measured by the GMFCS. Responders with children

functioning at GMCFS Level IV or V were less likely to enrol their child in the study, $X^2_{(4)} = 19.17$, $p = .001$.

Screening

Participants' ability to respond to standardised neuropsychological assessment tasks was determined using basic screening measures of pointing ability and expressive language. Participants' ability to point to stimuli typically encountered in standardised cognitive assessment via an iPad was determined using the Peabody Picture Vocabulary Test, Fourth edition (PPVT-4; Dunn & Dunn, 2007). The first 10 items of the PPVT-4 were administered twice, and 'adequate point response' was defined as at least 8 out of 10 concordant responses. Participants' ability to provide a coherent verbal response was determined using the first 10 items of the Picture Naming subtest from the WPPSI-IV (Wechsler, 2012). 'Adequate verbal response' was defined as eight out of 10 coherent verbal responses. Three participants had neither adequate point nor adequate verbal response and thus were excluded from further assessment. Three participants demonstrated adequate point response only, and one participant demonstrated adequate verbal response only. One participant declined to participate in screening procedures, and one was excluded retrospectively due to a comorbid diagnosis of Sturge Weber syndrome. The final sample was comprised of 70 adolescents with CP aged between 12 and 16 years (M = 14 years 6 months, SD = 10 months).

Classifications

Clinical information related to diagnosis and associated impairments was obtained from the VCPR and parental report. CP subtypes were categorised as spastic, dyskinetic, or ataxic. The topography of motor impairment was classified as unilateral or bilateral (Krägeloh-Mann & Cans, 2009). Gross motor function was classified using the GMFCS (Palisano et al., 1997) and fine motor ability was classified according to the Manual Ability Classification System (MACS; Eliasson et al., 2006). Additional information was obtained

regarding history of seizures, gestational age at birth, and birth weight. The VCPR defines epilepsy as "two or more afebrile seizures before age 5 years, excluding neonatal seizures, irrespective of seizure control" (Delacy & Reid, 2016, p. 51). To determine SGA, expected birth weight for gestational age was obtained from Kiserud et al. (2017). None of the participants had hearing impairment. Two of the adolescents had a visual impairment that may have hindered completion of some of the measures used in this study.

Research Ethics

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).

Cognitive testing

The Wechsler Intelligence Scale for Children – Fifth edition (WISC-V) was used to assess general intellectual ability in children with adequate verbal and/or adequate point response. The 10 primary subtests of the WISC-V and secondary subtest Letter-number sequencing were administered using Q-interactive on iPad. Motor-free IQ scores and indices were derived using the motor-free method described by Piovesana et al. (2019), which involves calculation of the Verbal Comprehension Index (VCI), Perceptual Reasoning Index (PRI), Working Memory Index (WMI), and Motor-Free IQ (MFIQ). Subtest raw scores were converted to scaled scores with a mean of 10 (SD = 3). MFIQ and index scores have a mean of 100 (SD = 15).

Procedure

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. The current study was part of a larger, longitudinal study investigating cognitive and

behavioural outcomes in adolescents with CP. The adolescents were assessed with a neuropsychological test battery by a Clinical Neuropsychology Registrar.

Not all of the adolescents completed all subtests of the WISC-V. Sixty participants completed all six subtests required to calculate the MFIQ and index scores. One participant with adequate verbal response only was administered Similarities, Vocabulary, Digit Span, Letter-number Sequencing. Conversely, three participants with adequate point response only were administered Visual Puzzles and Figure Weights. Assessment was discontinued prematurely for six participants due to behavioural difficulties, low tolerance for assessment, or because they did not return for a second assessment session.

Analyses

All statistical analyses were conducted using SPSS version 26. Visual inspection of stem-and-leaf plots and Q-Q plots for each variable indicated normal distribution. Observed mean values for each of the MFIQ and index scores were compared with the normative data using one-sample t-tests. For participants who obtained a MFIQ score, a repeated-measures analysis of variance (ANOVA) with index scores (VCI, PRI, WMI) as the within-subjects factor was conducted to examine differences in performance across domains. Chi-square tests were conducted to compare the distribution of borderline and impaired MFIQ and index scores in the CP group relative to the expected distribution based on the normative sample. Normal function was defined as within 1.0 SD of the test mean or better, i.e. standard scores > 85. Borderline cognitive ability was defined scores between one and two standard deviations below the test mean, i.e. standard scores between 70 – 84 inclusive. Impaired cognition was defined as two or more standard deviations below the mean, i.e. standard scores < 70. Expected rates of IQ scores in the normative sample are 14% borderline and 2% impaired. Regression analyses were conducted to identify the predictive ability of clinical factors (topography of motor impairment, severity of motor impairment, seizure history,

gestational age at birth, and SGA) on MFIQ and index scores. Motor type was not included as a predictor due to an insufficient number of participants in the dyskinetic and ataxic groups. Gestational age at birth and birth weight were highly correlated, therefore gestational age at birth was retained and birth weight was omitted from the analysis given the inclusion of an alternate weight variable, SGA. Assumptions required for multiple linear regression including linearity, multicollinearity, normality, and homoscedasticity were adequately met.

Results

Sample Characteristics

As shown in Table 1, of the total 70 participants, 49 (70.0%) were male and 21 (30.0%) were female. The majority had spastic CP (87.1%) and bilateral distribution of motor impairment (58.6%). Participants were predominantly classified as GMFCS Level I-II (72.9%) and MACS Level I-II (60.0%). The majority was born at term (61.4%) and recorded a birth weight in the normal range (68.6%). Less than a third reported a history of seizures (27.1%).

Cognitive abilities

Means and standard deviations for MFIQ, VCI, PRI and WMI scores according to motor type, motor topography, level of GMFCS and MACS, gestational age at birth, birth weight, SGA, and history of seizures are provided in Table 2.

A series of one-sample t-tests showed that the CP group as a whole scored significantly lower relative to the normative data on the MFIQ, $t_{(59)} = -6.87$, $p < .001$, $d = .89$ VCI, $t_{(65)} = -6.16$, $p < .001$, $d = .76$, PRI, $t_{(62)} = -8.44$, $p < .001$, $d = 1.06$, and WMI, $t_{(59)} = -6.06$, $p < .001$, $d = 0.78$. As demonstrated by Cohen's d, these effect sizes were large.

For the 60 participants who obtained a MFIQ score, repeated-measures ANOVA showed a significant difference in index scores, $F_{(2, 118)} = 12.77$, $p < .001$, $\eta_p^2 = .18$.

Bonferroni-adjusted pairwise comparisons indicated that VCI scores (M = 89.32, SD = 15.65) were significantly higher than PRI (M = 82.23, SD = 17.41), $p < .001$, and WMI (M = 84.77, SD = 19.48), $p = .002$. There was no significant difference between PRI and WMI scores, $p = .10$.

Rates of impairment

Frequency of borderline and impaired MFIQ and index scores in the CP group are shown in Figure 1. A series of Chi-square tests showed the observed distribution of borderline and impaired IQ scores in the CP group was significantly different from the normal distribution for MFIQ, $\chi^2_{(2)} = 217.04$, $p < .001$, VCI, $\chi^2_{(2)} = 110.68$, $p < .001$, PRI, $\chi^2_{(2)} = 239.06$, $p < .001$, and WMI, $\chi^2_{(2)} = 151.67$, $p < .001$. Rates of borderline and impaired cognitive ability in the CP group were higher than the normal distribution for MFIQ and each Index score. Rates of impairment ranged from 20-29% across domains, with the highest rate of impairment seen in the PRI. Rates of borderline cognitive ability ranged from 20-27%, with the highest rate of mild impairment seen in WMI.

Predictors of cognitive abilities

A series of standard multiple regression analyses were conducted to assess the ability of CP-related factors and associated conditions to predict VCI, PRI, and WMI (Table 3). The combination of CP-related factors and associated conditions was significantly related to verbal comprehension ability (VCI), $F_{(5, 58)} = 9.34$, $p < .001$, accounting for 44.60% of the variance in VCI scores. The results showed that severity of motor impairment as measured by GMFCS, SGA and history of seizures were significant predictors in the model. More severe motor impairment as measured by higher GMFCS level, SGA, and presence of seizure history were associated with lower VCI scores. Inspection of the regression standardised coefficients indicate that GMFCS contributed almost 1.5 times more than SGA and about three times more than seizure history.

The combination of CP-related factors and associated conditions was significantly related to perceptual reasoning ability (PRI), $F_{(5, 55)} = 6.20$, $p < .001$, accounting for 36.00% of the variance in PRI scores. Severity of motor impairment (GMFCS level) made the strongest contribution to predicting PRI scores. SGA also significantly contributed to PRI scores. Higher GMFCS level and SGA were associated with lower PRI scores, and inspection of the regression standardised coefficients indicate that GMFCS contributed almost two times more than SGA.

The combination of CP-related factors and associated conditions was significantly related to working memory ability (WMI), $F_{(5, 52)} = 6.05$, $p < .001$, accounting for 36.80% of the variance in WMI scores. Again, results showed that severity of motor impairment as measured by GMFCS and SGA were significant predictors in the model. Higher GMFCS level and SGA were associated with lower WMI scores.

Discussion

The present study aimed to characterise a motor-free cognitive profile of adolescents with CP using the WISC-V. Previous research has shown that standardised administration of the Wechsler scales fails to account for the motor impairment in CP and consequently underestimates the IQ of children with CP (Sherwell et al., 2014; Yin Foo et al., 2013). To overcome this challenge, motor-free administration of the WISC-V as described by Piovesana et al. (2019) was employed in the present study to derive MFIQ and index scores. Adolescents with CP performed significantly below expectations compared to normative data across domains, and rates of borderline and impaired cognitive abilities were significantly higher than expected. Moreover, results showed an uneven cognitive profile characterised by a relative strength in verbal skills. A secondary aim was to examine the association between the motor-free cognitive profile in adolescents with CP and clinical factors which have been previously linked to poorer cognitive abilities. Severity of motor impairment and SGA were

identified as significant predictors of cognitive abilities across domains while a relation between seizure history and verbal skills was also noted.

The hypothesis that adolescents with CP would perform below expectations across cognitive domains compared to normative data was supported in the current study. Results showed that MFIQ scores as well as VCI, PRI and WMI scores were all significantly below expectations and effect sizes were large. Rates of cognitive impairment in our CP sample ranged from 20-29% across domains – that is 10 to 15 times more often than expected compared to normative data. However, the rates of impairment observed in the current study likely represent an underestimate due to the under-representation of adolescents with CP GMFCS Level IV and V in our sample. CP register studies that include the whole CP population suggest rates of cognitive impairment are closer to 50% (Australian Cerebral Palsy Register, 2018; Novak et al., 2012).

A novel finding was that rates of borderline cognitive ability ranged from 20-27% across domains and is 1.5 to twice as common in the CP group compared to normative data (14%). In this study, borderline cognitive ability was defined as scores falling between one and two standard deviations below the test mean, i.e., scores fall below what is considered the 'normal range' but above the cut-off for intellectual disability. Little attention has been paid to this level of cognitive impairment in the literature, particularly in the context of CP, and it often goes unrecognised (Pulina et al., 2019). Previous studies have shown that primary and lower secondary school students with IQ between 70-85 struggle to cope with the academic demands of mainstream education (Karande et al., 2008; Macmillan et al., 1998; Pulina et al., 2019) but often do not qualify for special classroom assistance. Macmillan et al. (1998) showed that the academic performance of second grade students with borderline cognitive ability in arithmetic and spelling closely resembled students with intellectual disability (IQ < 70), while both groups differed significantly from students with IQ above 85. Students with

borderline cognitive ability are at risk of failure in the classroom, school drop-out, anxiety and depression, social problems and behavioural problems including aggression and withdrawal (Karande et al., 2008). In the context of CP, children with borderline cognitive ability need to be identified at an early stage so they can be taught with methods and pace which not only accommodates their motor impairment but is also appropriate for their learning ability.

Consistent with expectations, the current findings demonstrated an uneven cognitive profile in adolescents with CP. Previous studies have described a relative weakness in the non-verbal domain (Gonzalez-Monge et al., 2009; Ito et al., 1997; Sigurdardottir et al., 2008); however, the present findings indicate the profile is more accurately characterised by a relative strength in the verbal domain. VCI scores were significantly higher than PRI and WMI scores, and there was no significant difference between the PRI and WMI. These findings deviate slightly from the previous studies that used earlier versions of the WISC (i.e. WISC-Revised, WISC-III), perhaps due to a change in the test structure. The earlier versions of the WISC included only two indices: Verbal IQ comprised of verbal comprehension and auditory working memory tasks and Performance IQ comprised of visual-spatial, perceptual reasoning, and processing speed tasks. In contrast, the motor-free version of the WISC-V allows the derivation of three indices which provide a clearer delineation between cognitive domains and therefore a more nuanced cognitive profile (Piovesana et al., 2019).

The current findings of a relative strength in verbal intelligence compared to perceptual reasoning and working memory domains can be interpreted in the context of the 'Cognitive Crowding' hypothesis. First described by Teuber (1974) in adults with brain injury and later applied to hemiplegic children by Carlsson et al. (1994), the Cognitive Crowding hypothesis refers to the developing brain's capacity for reorganisation after injury. However, the hypothesis recognises a hierarchy in cognitive development starting with

language. Several cross-sectional studies have suggested that verbal skills tend to be spared after injury to either hemisphere, but the cost of this flexibility is borne by visuospatial functions (Carlsson et al., 1994; Ito et al., 1997; Pagliano et al., 2007). The present findings build on this and suggest that working memory skills may also be impaired in CP as a result of this early neural compromise. However, as a component of executive functions, working memory deficits may not be noticeable until later childhood or adolescence in the context of increasing demands of independent functioning (Anderson, 2002). Deficits in working memory and perceptual reasoning have also been shown in other developmental disorders including specific learning disorder, autism spectrum disorder, and intellectual disability (Alloway & Archibald, 2009). This common profile lends support to findings that working memory and nonverbal IQ share neural substrates (Gray et al., 2003) which show vulnerability to disruption during disordered development and may not be unique to CP.

The hypothesis that bilateral motor impairment, severe motor impairment, seizure history, and SGA would be associated with poorer cognitive abilities was partially supported. Regression analyses identified severity of motor impairment and SGA as significant predictors of cognitive abilities across domains, while seizure history was also associated with verbal abilities. Even with the motor-free administration of the WISC-V, severity of motor impairment was the strongest predictor of cognitive abilities. Higher GMFCS level was related to poorer cognitive outcomes across domains.

The relation between motor impairment and cognitive development is complex in so far as the motor impairment is not only related to the initial brain injury, but has ongoing implications for the child's ability to engage with their environment. Previous MRI studies have shown that findings of brain maldevelopment, basal ganglia lesions, and cortical/subcortical lesions were associated with severe gross motor impairment as well as severe intellectual disability (Himmelmann & Uvebrant, 2011). Moreover, motor impairment

which restricts the child's mobility or communication also restricts cognitive development due to limited ability to explore their environment, engage in play-based learning, participate in social interactions and classroom activities (Bottcher, 2010). However, it is important to note that although severity of motor impairment is a predictor of cognitive abilities in CP at the group level, Stadskleiv et al., (2018) reported that one third of participants with severe motor impairment in their sample had normal cognitive ability. Although children with severe motor impairment should therefore be identified by medical and allied health professionals as 'at-risk' of intellectual disability, motor-free cognitive assessment is needed to provide them with the opportunity to demonstrate their true cognitive abilities.

Our findings revealed that SGA was also associated with poorer cognitive abilities across domains. There is limited research investigating the impact of SGA on cognition in CP. The current findings build on previous evidence by Hemming et al. (2008) who reported that infants born SGA were at increased risk of severe intellectual impairment relative to those born AGA. However, this is in contrast to other studies which have shown no significant association between SGA and cognitive abilities in infants born at term (O'Keefe et al., 2003), preterm (Graz et al., 2015), or with very low birth weight (VLBW; Latal-Hajnal et al., 2003). This discrepancy between previous and current results may be explained by findings by Latal-Hajnal et al., (2003) who showed that insufficient postnatal growth, rather than SGA, determined later neurodevelopmental outcome. In their study, VLBW infants born SGA who showed "catch-up growth" by 2 years of age had cognitive outcomes comparable to those born AGA. In contrast, SGA infants who remained below the 10th percentile at 2 years of age showed impairments. Moreover, lack of catch-up growth has been associated with poor cognitive outcomes in adulthood (Lundgren et al., 2003). In the context of CP specifically, SGA and severe motor impairment (GMFCS Level III-V) is associated with restricted growth in height, weight, and head circumference such that the gap between infants

born SGA and AGA continues to widen to at least 5 years of age (Strand et al., 2016). Reduced head growth in particular has been correlated with reduced brain tissue volume and impairment on cognitive assessment in early childhood (Cheong et al., 2008). The current findings suggest the early effects of SGA on growth and neurodevelopment persist into adolescence and highlights the need to identify this at-risk group for early intervention.

The findings of the current study also showed an association between seizure history and verbal abilities in adolescents with CP. Previous studies have reported that the presence of seizures were associated with a decrement in overall IQ in children with CP at preschool-age (Sigurdardottir et al., 2008), during early childhood (Muter et al., 1997), and adolescence (Cheng et al., 2013). However, an association between seizures and verbal IQ is less commonly described. Gonzalez-Monge et al. (2009) showed that while the presence of seizures did not predict verbal IQ at 7-years of age, seizures slowed the progression of verbal abilities over a 7-year follow-up period. Our sample is comparable in age to that of Gonzalez-Monge et al. at follow up, perhaps suggesting the effect of seizures on verbal skills is only detectable on assessment during later childhood or adolescence. This would be consistent with Cheng et al. (2013) who argued that seizures had a deleterious effect on cognition over time. Given the broad categorisation of seizure history, we were unfortunately unable to disentangle the impact of seizure type, frequency, or antiepileptic medication on cognition. Although CP is a non-progressive disorder, the consequences for cognition evolve over time, highlighting the need for ongoing assessment and monitoring throughout adolescence particularly for those with seizure history.

While the motor-free administration has been shown to be the most appropriate way to assess IQ in adolescents with CP using the WISC-V, it is not without limitations. First, although complex fine motor responses are avoided using the motor-free method, the test still requires either a verbal or point response to answer questions which is impossible for some

people with CP. Unfortunately, two participants functioning at GMFCS Level IV-V were unable to provide a verbal or point response during the screening phase of this study and were subsequently excluded from further assessment. Adapting the WISC-V for use with alternative response modes like gaze pointing would provide the opportunity for children with severe speech and motor impairment to demonstrate their true cognitive abilities (Kurmanaviciute & Stadskleiv, 2017).

Second, the MFIQ does not include a measure of processing speed. The processing speed tasks of the WISC-V require quick, fine motor responses (i.e. gripping and manipulating a pencil to provide handwritten responses) and have been shown to place adolescents with CP at an inherent disadvantage due to the nature of their motor disorder (Coceski et al., 2020). Given evidence that movement time is slower in children with CP at all levels of severity compared to typically developing controls (Fernani et al., 2017), nonmotor versions of processing speed tasks are required to accurately capture true information processing speed rather than motor speed/movement time in the context of CP and other motor disorders. Future research may consider employing the WISC-V Integrated to circumvent the issues of verbal and point responses, as well as the omission of processing speed tasks. The WISC-V Integrated includes alternate presentation modes and response formats for some subtests which can be used in conjunction with the traditional WISC-V subtests to calculate nonmotor composite scores (i.e. Multiple Choice Verbal Comprehension Index).

The sample size may have resulted in a lack of power to detect group differences. In addition, we were unable to investigate an association between CP type and cognitive abilities due to a small number of participants with dyskinetic and ataxic CP in our sample. The composition of our sample is comparable to previous studies that have used multiple regression in CP (Stadskleiv et al., 2018) and is representative of the distribution of cases by

CP type on the Australian CP Register (2018). Eighty-five per cent of cases on the register have spastic CP while rates of dyskinetic and ataxic CP are 7% and 4%, respectively, with the remainder classified as hypotonic, mixed, or unknown (Australian CP Register, 2018). While increasing the number of participants with dyskinetic or ataxic CP in studies to investigate group differences would be ideal, they are difficult to recruit due to a smaller pool from which to draw and the sample would not be representative of the wider population.

In addition to the clinical factors identified in the current study, future research may consider the impact of social and environmental factors on development of IQ in adolescents with CP. A previous longitudinal study of term-born infants without CP showed that socioeconomic status and the parent-child relationship impact IQ in adulthood, and the effect was comparable to being born SGA (Eves et al., 2020). Social participation has been shown to affect cognitive development in CP (Bottcher, 2010), and these factors may be particularly relevant since they can affect access to education and intervention programs.

Conclusion

Notwithstanding the motor-free administration of the WISC-V used in this study, our findings have shown that the MFIQ and index scores of adolescents with CP fell significantly below expectations compared to normative data. The motor-free cognitive profile was characterised by a relative strength in verbal intelligence and comparatively weaker performance in perceptual reasoning and working memory domains. Clinical factors including severity of motor impairment and SGA were linked to poorer cognitive performance across domains while a history of seizures was related to lower verbal abilities. These factors should be recognised by health professionals and prompt ongoing assessment and monitoring of cognitive abilities throughout childhood and adolescence. While severe cognitive impairment is generally understood to be common in CP, the findings highlighted a substantial proportion of adolescents with CP with borderline cognitive ability – a level of

impairment which has been given very little attention in CP. Young people with CP and borderline cognitive ability may struggle to cope with mainstream education and need to be identified early and taught with methods and pace which accommodate their motor impairment and learning ability in order to avoid negative outcomes including anxiety, social and behavioural problems and school-drop out.

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Declaration of interest

No potential conflict of interest was reported by the authors.

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Table 1

Sample characteristics

	Participants	
	n	%
Sex		
Male	49	70.0
Female	21	30.0
Motor type		
Spastic	61	87.1
Dyskinetic	8	11.4
Ataxic	1	1.4
Topography		
Unilateral	29	41.4
Bilateral	41	58.6
Gross motor function (GMFCS)		
Level I	27	38.6
Level II	24	34.3
Level III	17	24.3
Level IV	1	1.4
Level V	1	1.4
Fine motor ability (MACS)		
Level I	17	24.3
Level II	25	35.7
Level III	13	18.6
Level IV	2	2.9
Unknown	13	18.6
Gestational age at birth		
<28 weeks	6	8.6
28 - <32 weeks	8	11.4
32 - <37 weeks	12	17.1
>37 weeks	43	61.4
Unknown	1	1.4
Birth weight		
<1000g	5	7.1
1000 – 1499g	7	10.0
1500 – 2499g	8	11.4
>2500g	48	68.6
Unknown	2	2.9
Small for gestational age		
Yes	11	15.7
No	57	81.4
Unknown	2	2.9
History of seizures		
Yes	20	27.1
No	50	72.8

Note: GMFCS = Gross Motor Functional Classification System; MACS = Manual Ability Classification System; Small for gestational age was defined as birth weight <10th percentile of expected weight for gestational age.

Table 2

Mean Motor-free IQ and Index Scores in Adolescents with Cerebral Palsy According to Motor Type, Topography, Gross and Fine Motor Function, Gestational Age, Birth Weight, and Seizure History

		MFIQ		VCI		PRI		WMI
	n	M*(SD)*	*n*	M*(SD)*	*n*	M*(SD)*	*n*	M*(SD)*
Total	60	82.78 *(19.42)*	66	87.55 *(16.42)*	63	81.10 *(17.78)*	60	84.77 *(19.48)*
Motor type								
Spastic	53	84.57 *(16.22)*	58	89.47 *(15.89)*	55	82.56 *(18.00)*	53	86.21 *(19.21)*
Dyskinetic	6	70.67 *(17.34)*	7	75.71 *(13.78)*	7	71.00 *(13.94)*	6	74.17 *(21.21)*
Ataxic	1	61.00	1	59.00	1	71.00	1	72.00
Topography								
Unilateral	27	84.85 *(15.19)*	29	89.34 *(12.54)*	27	85.04 *(13.68)*	27	85.56 *(17.78)*
Bilateral	33	81.09 *(22.38)*	37	86.14 *(18.97)*	36	78.14 *(20.00)*	33	84.12 *(21.03)*
Gross motor function (GMFCS)								
Level I	26	92.23 *(16.43)*	26	97.12 *(12.60)*	26	90.15 *(15.18)*	26	93.04 *(18.22)*
Level II	20	78.80 *(15.15)*	23	85.30 *(12.46)*	21	78.05 *(16.62)*	20	80.70 *(13.99)*
Level III	13	72.54 *(22.46)*	15	77.07 *(19.16)*	14	73.14 *(15.89)*	13	77.15 *(22.93)*
Level IV	1	50.00	1	73.00	1	51.00	1	50.00
Level V	-	-	1	62.00	1	51.00	-	-
Fine motor ability (MACS)								
Level I	16	90.19 *(19.89)*	17	96.06 *(14.16)*	16	86.63 *(19.42)*	16	92.31 *(19.44)*
Level II	23	87.13 *(15.30)*	24	91.50 *(12.52)*	23	86.30 *(14.35)*	23	88.57 *(17.05)*
Level III	10	66.10 *(17.62)*	12	74.50 *(14.27)*	11	67.09 *(14.38)*	10	69.10 *(18.97)*
Level IV	1	43.00	2	56.50 *(7.78)*	2	54.00 *(4.24)*	1	50.00

Gestational age at birth								
Term (>37 weeks)	35	84.06 *(19.62)*	39	87.03 *(16.75)*	37	82.65 *(18.27)*	35	86.31 *(19.36)*
Pre-term (<37 weeks)	24	81.00 *(19.80)*	26	88.38 *(16.54)*	25	78.40 *(17.33)*	24	82.92 *(20.21)*
Birth weight								
>2500g	39	84.67 *(19.33)*	43	87.30 *(16.56)*	41	84.02 *(17.75)*	39	86.49 *(19.40)*
< 2499g	19	77.37 *(18.81)*	21	86.76 *(16.10)*	20	72.95 *(14.75)*	19	80.63 *(19.87)*
Small for gestational age								
No	47	85.19 *(18.41)*	53	88.60 *(15.78)*	50	82.48 *(17.46)*	47	87.45 *(19.16)*
Yes	11	69.82 *(18.84)*	11	80.00 *(17.54)*	11	70.91 *(14.95)*	11	72.27 *(17.89)*
History of seizures								
No	44	86.48 *(18.74)*	48	90.62 *(15.95)*	45	84.22 *(18.00)*	44	88.41 *(18.92)*
Yes	16	72.63 *(18.05)*	18	79.33 *(15.15)*	18	73.28 *(14.95)*	16	74.75 *(17.93)*

Note: GMFCS = Gross Motor Classification System; MACS = Manual Ability Classification System; MFIQ = Motor-free Intelligence Quotient; PRI = Perceptual Reasoning Index; VCI = Verbal Comprehension Index; WMI = Working Memory Index; Gestational age at birth was unknown for one participant; Birth weight was unknown for two participants and as a result weight for gestational age could not be calculated. MACS data was not available for 11 participants.

Table 3

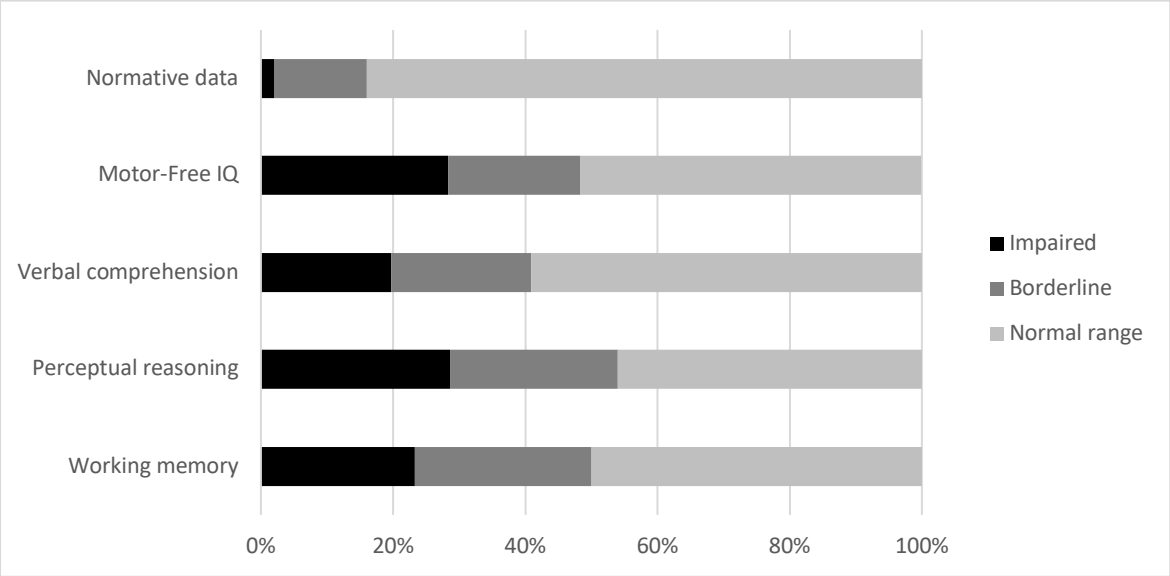
Multiple Linear Regression Model of Variance in Verbal Comprehension Index, Perceptual Reasoning Index, and Working Memory Index

	B	SE B	Standardised ß	*t*	*p*
Verbal Comprehension Index					
GMFCS Level	-11.39	2.19	-.63	-5.19	<.001
SGA	-14.98	4.49	-.35	-3.34	.001
Seizures	-7.59	3.72	-.21	-2.04	.046
Gestation	5.17	3.53	.16	1.47	.148
Topography	5.33	4.03	.16	1.32	.192
Perceptual Reasoning Index					
GMFCS Level	-9.58	2.57	-.50	-3.73	<.001
SGA	-15.05	5.24	-.33	-2.87	.006
Seizures	-7.37	4.28	-.19	-1.72	.091
Gestation	0.11	4.16	.01	-0.02	.979
Topography	2.37	4.64	.07	0.51	.612
Working Memory Index					
GMFCS Level	-11.63	3.31	-.50	-3.51	.001
SGA	-19.32	5.85	-.39	-3.30	.002
Seizures	-9.25	5.15	-.21	-1.80	.078
Gestation	1.24	4.77	.03	0.26	.795
Topography	8.42	5.44	.21	1.55	.128

Note: GMFCS = Gross Motor Classification System, SGA = Small for gestational age. Coded GMFCS = 0 (Level I); 1(Level II); 2 (Level III); Level IV and V were excluded as each had only one value; SGA = 0 (No); 1 (Yes); Seizure history = 0 (No); 1 (Yes); Gestation = 0 (Term); 1(Pre-term); Topography = 0 (Unilateral); 1 (Bilateral).

Figure 1

Rates of Borderline and Impaired Cognitive Abilities as Measured by Motor-Free IQ and Index Scores in the Cerebral Palsy Group Compared to Normative Data



Chapter 5: Motor-Free IQ in Adolescents with Mild to Moderate Cerebral Palsy: A 10-Year Follow-Up Study

Preamble to empirical paper 3

The previous chapter showed that cognitive abilities in adolescents with mild to moderate CP fell significantly below the normative data. Moreover, the uneven cognitive profile showcased stronger verbal than nonverbal skills even with the use of the Motor-free method for assessing IQ. However, the development of these cognitive skills from childhood to adolescence remains unclear given the cross-sectional nature of the study in the previous chapter. In fact, the literature devoted to understanding the trajectory of cognitive development in the context of CP is sparse despite the pervasive, lifelong impacts of the condition. To address this gap in the literature, this chapter, presented in the form of a submitted manuscript to the journal *European Journal of Paediatric Neurology*, examined cognitive change and stability over a 10-year period from preschool age to adolescence. In addition to group-level analyses, this chapter is the first to utilise reliable change statistics to illustrate the change in cognitive abilities over time. This chapter provides new insight into the heterogeneity of cognitive outcomes seen in CP.

Cognitive ability in children with mild to moderate cerebral palsy: A 10-year follow-up study

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Abstract

Aim: To examine cognitive change and stability over a 10-year period from preschool age to adolescence at both the group- and individual-levels in a cohort of children with CP.

Method: Cognitive abilities of 28 children (21 males, 7 females) were assessed using motor-free short-forms of the Wechsler scales at Time 1 (M = 4 years 6 months, *SD* = 7 months) and Time 2 (*M* = 14 years 6 months, *SD* = 9 months). Average time between assessments was 10 years and 4 days (*SD* = 6 months). 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 over time at the individual level.

Results: At the group level, nonverbal IQ scores declined significantly. Decline in FSIQ did not reach significance, and verbal IQ remained stable. At the individual level, reliable change scores indicated nine to 11 children showed a clinically significant decline 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.

Interpretation: Intra-individual changes in IQ indicate considerable heterogeneity in the development of cognitive abilities in a group of adolescents with CP, which may not be accurately captured with a single mean difference score. The findings highlight the importance of ongoing monitoring and individual assessment throughout development.

Keywords: assessment, cerebral palsy, cognitive ability, follow-up, longitudinal

Highlights

- Nonverbal IQ decline noted at 10-year follow up in adolescents with cerebral palsy

- Verbal IQ remains stable over 10-year period in children with cerebral palsy
- Intra-individual decline in Full Scale IQ related to epilepsy
- Intra-individual decline in nonverbal IQ related to higher IQ at preschool age

Cerebral Palsy (CP) develops in about 2 per 1000 livebirths in developed countries (Australian Cerebral Palsy Register, 2018; Surveillance of Cerebral Palsy in Europe, 2000). The term CP is one of clinical utility, encompassing a wide range movement disorders, levels of motor impairment and functional limitation. Many children with CP have associated conditions including disturbances of cognition, behaviour, and epilepsy (Rosenbaum et al., 2007). According to population CP registers (Australian Cerebral Palsy Register, 2018; Surveillance of Cerebral Palsy in Europe, 2000), intellectual disability occurs in approximately half of children with CP by 5 years of age. However, previous studies suggest one-third of children with CP lack the fine motor skills required to respond to cognitive tests (Sherwell et al., 2014), and intellectual disability is commonly assumed based on clinical judgement (Andersen et al., 2008) or severity of motor impairment (Hutton et al., 2002). Although CP is non-progressive, the manifestations of the disability 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 (Anderson, 2002). Despite this, there are few longitudinal studies that investigate the developmental trajectory of cognitive abilities from childhood to adolescence in young people with CP (Gonzalez-Monge et al., 2009).

Intelligence Quotient (IQ) is an important determinant of functioning throughout the life course and has implications for participation in education (Jenks et al., 2007), self-care, domestic and community tasks (Van Gorp et al., 2018), employment (Magill-Evans et al., 2008) and independent living (Reddihough et al., 2013). People with CP and IQ in the normal range are more likely to achieve the expected level of performance in these domains, albeit later compared to typically developing peers of the same chronological age. However, achievement of lifestyle goals is less favourable for adolescents with CP and cognitive impairment, with little improvement in functioning seen over time (Van Gorp et

al., 2018). Therefore, tracking cognitive change or stability through adolescence is important to guide expectations, counsel families, and set realistic treatment goals to support young people with CP during this transitional phase.

Two previous studies have assessed cognitive change or stability from childhood through adolescence in individuals with CP. Dahlgren Sandberg (2006) was primarily concerned with reading and spelling abilities in children with severe speech and motor impairments that required aided communication but also noted a significant decline in Full Scale IQ (FSIQ) over time. On the other hand, Gonzalez-Monge et al. (2009) showed stability in FSIQ and verbal abilities, but a selective decline in nonverbal skills. The use of nonverbal subtests that rely on motor coordination to manipulate stimuli may lead to inaccurate estimation of IQ scores in this domain (Sherwell et al., 2014) and contribute to the substantial heterogeneity observed in the performance of children of the same age and severity of motor impairment (Dahlgren Sandberg, 2006; Gonzalez-Monge et al., 2009). With such heterogeneity, an inherent limitation of previous studies has been the reliance on group differences to characterize 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 (Gonzalez-Monge et al., 2009). Reliance on group mean differences may be misleading and is unlikely to capture the considerable range of cognitive abilities in children with CP.

An alternative, perhaps complementary, approach is to characterize cognitive change at the individual level using the Reliable Change Index (RCI) [see Jacobson & Truax (1991) for a full description]. 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 in children with autism (Waizbard-Bartov et al., 2020), attention and memory post acquired brain injury (Catroppa et al.,

2015), and executive functioning in children with CP (Piovesana et al., 2015). Although most research in CP has been reported at the group level with the intention of generalizing findings to the population, it can be difficult to apply group findings to an individual in a clinical setting. Understanding individual differences is important for prognosis and treatment planning in a heterogeneous condition like CP. To address the limitations of previous longitudinal studies that relied on group-level analyses, the aim of this 10-year follow-up study was to examine change and stability in cognitive abilities from preschool age to adolescence at both the group and individual levels in a cohort of children with CP.

Method

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).

Participants

Children with CP were recruited through the Victorian Cerebral Palsy Register (VCPR) and participated at two data collection timepoints. To be included on the VCPR, a case had to fulfil the definitional criteria for CP (Smithers-Sheedy et al., 2014). Between 2008 and 2009, 80 children with CP aged 4–6 years participated in a study by Sherwell et al. (2014) that examined cognitive abilities in preschool children with CP (Time 1). In 2018, VCPR staff 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 non-participation: 12 parents were not interested in returning for Time 2, three parents were interested but the adolescent did not agree to participate, four declined due to other medical/mental health issues, and one was unwilling to travel from regional Victoria. After enrolment, one adolescent declined to participate in the cognitive assessment and another

was excluded retrospectively due to missing cognitive data at Time 1. Written informed consent was obtained from the parent/caregiver.

Clinical information was obtained from the VCPR and parental report. CP motor types were categorized as predominantly spastic or dyskinetic and topography as unilateral or bilateral (Krägeloh-Mann & Cans, 2009). Gross and fine motor function were respectively described using the Gross Motor Function Classification System (GMFCS; Palisano et al., 1997) and the Manual Ability Classification System (MACS; Eliasson et al., 2006). The VCPR defined epilepsy as "two or more afebrile seizures before age 5 years, excluding neonatal seizures, irrespective of seizure control" (Delacy & Reid, 2016, p. 51). Preterm birth was defined as <37 weeks gestation. Low birth weight was defined as <2499g.

Measures

The core subtests of the Wechsler Preschool and Primary Scale of Intelligence – Third edition (WPPSI-III; Wechsler, 2002) were used to assess general intellectual ability at Time 1. To minimise the impact of fine motor responses, a prorated Performance IQ was derived using Matrix Reasoning and Picture Concepts subtests (Sherwell et al., 2014). An estimated FSIQ score was calculated according to the Tellegen & Briggs (1967) procedure using five motor-free subtests (Information, Vocabulary, Word Reasoning, Picture Concepts, and Matrix Reasoning).

The Wechsler Intelligence Scale for Children – Fifth edition (WISC-V;Wechsler, 2016) was administered at Time 2 using Q-interactive on iPad. Motor-free IQ (MFIQ), Verbal Comprehension Index (VCI), and Perceptual Reasoning Index (PRI) scores were derived according to the method described by Piovesana et al. (2019). The MFIQ and indices have strong psychometric properties that are comparable to the WISC-V FSIQ and

index scores (Piovesana et al., 2019). A factorial invariance study of the Wechsler scales (WPPSI, WISC, and Wechsler Adult Intelligence Scale (WAIS)) 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 (Niileksela & Reynolds, 2019). All IQ and index scores have a mean of 100 (*SD* = 15).

Procedure

The procedure for Time 1 assessments has been described in detail by Sherwell et al. (2014). 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). Participants' ability to respond to standardized neuropsychological assessment tasks was determined using basic screening measures of pointing ability and expressive language (Coceski et al., 2021). Not all the adolescents completed all subtests of the WISC-V required to calculate a MFIQ. One participant did not attempt Visual Puzzles and two participants did not attempt Letter-Number Sequencing subtest due to low tolerance for assessment.

Statistical analysis

Statistical analyses were conducted using SPSS version 26. 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 .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 using the following formula:

$$RCI = \frac{X_2 - X_1}{SD\sqrt{2 - r_1 - r_2}}$$

where X_1 is IQ at Time 1, X_2 is IQ at Time 2, SD is the standard deviation of IQ scores, r_1 is the reliability coefficient of the WPPSI-III FSIQ composite or index and r_2 is the reliability coefficient of the WISC-V MFIQ composite or index. Reliability coefficients were obtained from the test manual for the WPPSI-III and were Piovesana et al. (2019) for the WISC-V motor-free composites. RCI scores above or below ±1.96 represent a statistically and clinically significant improvement or deterioration at the level $\alpha = .05$.

A series of point-biserial correlations was conducted to explore relations between RCIs for FSIQ and nonverbal IQ and dichotomized 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.

Results

Participant characteristics

The final sample comprised 28 adolescents with CP 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, $t(77) = -.71$, $p = .48$, or gender distribution, $\chi^2_{(1)} = .47$, $p = .49$. However, there was a significant association between participation at Time 2 and severity of motor impairment as measured by the GMFCS (Palisano et al., 1997), $\chi^2_{(4)} = 11.88$, $p = .018$. Parents of

children functioning at GMFCS Level IV-V were less likely to enrol their children in the study at Time 2.

Mean age at Time 1 was 4 years 6 months (*SD* = 7 months). 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 CP and were predominantly classified as functioning at GMFCS Levels I-II and MACS Level II; almost half had a history of seizures (Table 1). FSIQ, verbal- and nonverbal IQ scores at Time 1 were available for 23, 24, and 26 participants, respectively.

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, $t_{(22)} = 2.00$, $p = .059$ and effect size was moderately small, $d = 0.32$. Mean nonverbal IQ scores declined significantly between Time 1 and Time 2, $t_{(25)} = 2.12$, $p = .044$, and the effect size was again moderately small, $d = .31$. In contrast, there was no significant change in verbal IQ scores between Time 1 and Time 2, $t_{(23)} = 0.11$, $p = .917$.

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, remained stable for 11, and improved significantly for three participants.

A minimum change of 11 standard score points represented a clinically significant change in verbal IQ. Verbal IQ scores remained stable for 17 participants, the majority of

the sample. In contrast, four participants demonstrated a significant decline in verbal IQ whereas three 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. Thirteen participants showed stable nonverbal IQ and only two showed a significant improvement.

Correlations

Reliable change in FSIQ was significantly correlated with epilepsy, r_{pb} = -.51, 95% BCa CI [-.783, -.097], p = .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 = -.65, 95% BCa CI [-.805, -.361], p = .001, as well as nonverbal IQ score at Time 1, r = -.65, 95% BCa CI [-.812, -.371], p = .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.

Discussion

To date, this study is 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 CP. 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 to 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 CP.

Our findings highlighted a significant decline in nonverbal IQ over time in this group of children with CP. This is consistent with a previous longitudinal study where selective decline in nonverbal IQ scores over a 7-year period in young people with CP was reported (Gonzalez-Monge et al., 2009). 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 emphasize 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 standardized sample. As such, the current results suggest children with CP 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 (Dykens et al., 2006); however, in CP it appears that this plateau is more discernible for nonverbal abilities. According to the 'cognitive crowding' hypothesis (Teuber, 1974), cognitive development following early brain injury occurs in a hierarchy whereby verbal abilities are prioritized 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 begin to demand abstract reasoning skills (Levine et al., 2005). As such, nonverbal impairments in children with CP are more clearly identifiable at an age at which tests become more demanding on nonverbal reasoning skills.

Similarly, our findings indicated children with CP and a history of seizures showed slower-than-expected development of FSIQ over time. This is consistent with a previous

studies that suggest seizures have a deleterious effect on cognition (Cheng et al., 2013; Muter et al., 1997). Indeed, an interplay between age, seizures, and cognitive abilities has been raised—an adolescent with CP and seizures is more likely to have low IQ than a younger child with CP and seizures (Cheng et al., 2013). 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 (Paldino et al., 2017; Widjaja et al., 2013) and in turn, is associated with IQ decline (Paldino et al., 2017). Seizures, therefore, seem to contribute to a reduced rate of cognitive skill acquisition in children with CP 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 should be acknowledged. Firstly, our study was underpowered and non-ambulant children were underrepresented. Given an average decline of 23 IQ points by the age of 12 has previously been reported in children with severe motor impairment (Dahlgren Sandberg, 2006), our findings may underestimate the magnitude of change from childhood to adolescence in CP due to participation bias. We attempted to mitigate this limitation by conducting reliable change statistics which provide useful change-pattern data in small samples while taking into account test reliability and clinical significance.

A second limitation was the broad categorization of epilepsy, and therefore, we were unable to disentangle the impact of epilepsy-related variables including age of seizure onset, frequency, or number of antiepileptic medications which have been previously related to cognitive outcomes in children and adolescents with epilepsy (MacAllister et al., 2019). This may account for variation in cognitive development in adolescents with CP 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 CP 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 results, a cognitive assessment conducted at preschool age is unlikely to accurately inform the needs of a child with CP 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 a risk factor for later decline. We suggest that clinicians use this information to prepare parents and guide expectations for future functioning. Finally, the study emphasizes the importance of seizure control in children with CP and the need for close monitoring throughout development.

Conclusion

The current study is the first to investigate cognitive change or stability over a 10-year period in children with mild to moderate CP using motor-free methods for assessing IQ as well as group- and individual-level analyses to characterize change over time. Intra-individual changes indicate considerable heterogeneity in the development of cognitive abilities in a group of adolescents with CP, which may not be accurately captured with a single mean difference score. 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. Cognitive development is a dynamic and competitive process that is influenced by many factors including the brain, the

environment, and behaviour. Future research may consider environmental and social factors for their potential contribution to cognitive development in the context of CP.

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Declaration of interest

No potential conflict of interest was reported by the authors.

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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
Gestational age at birth	
Term	22
Preterm	6
Birth weight	
Normal	22
Low birth weight	6

Note. GMFCS = Gross Motor Function Classification System; MACS = Manual Ability Classification System

Table 2

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

	n	*M*	*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

Note. IQ = Intelligence quotient

Table 3

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

	Time 1			Time 2		
Participant	FSIQ	Verbal	Non-verbal	FSIQ	Verbal	Non-verbal
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*

* indicates a significant decline in scores between Time 1 and Time 2

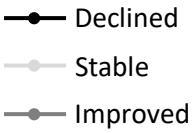
+ indicates a significant increase in scores between Time 1 and Time 2

- unable to calculate score

FSIQ = Full Scale Intelligence Quotient

Figure 1.

Legend



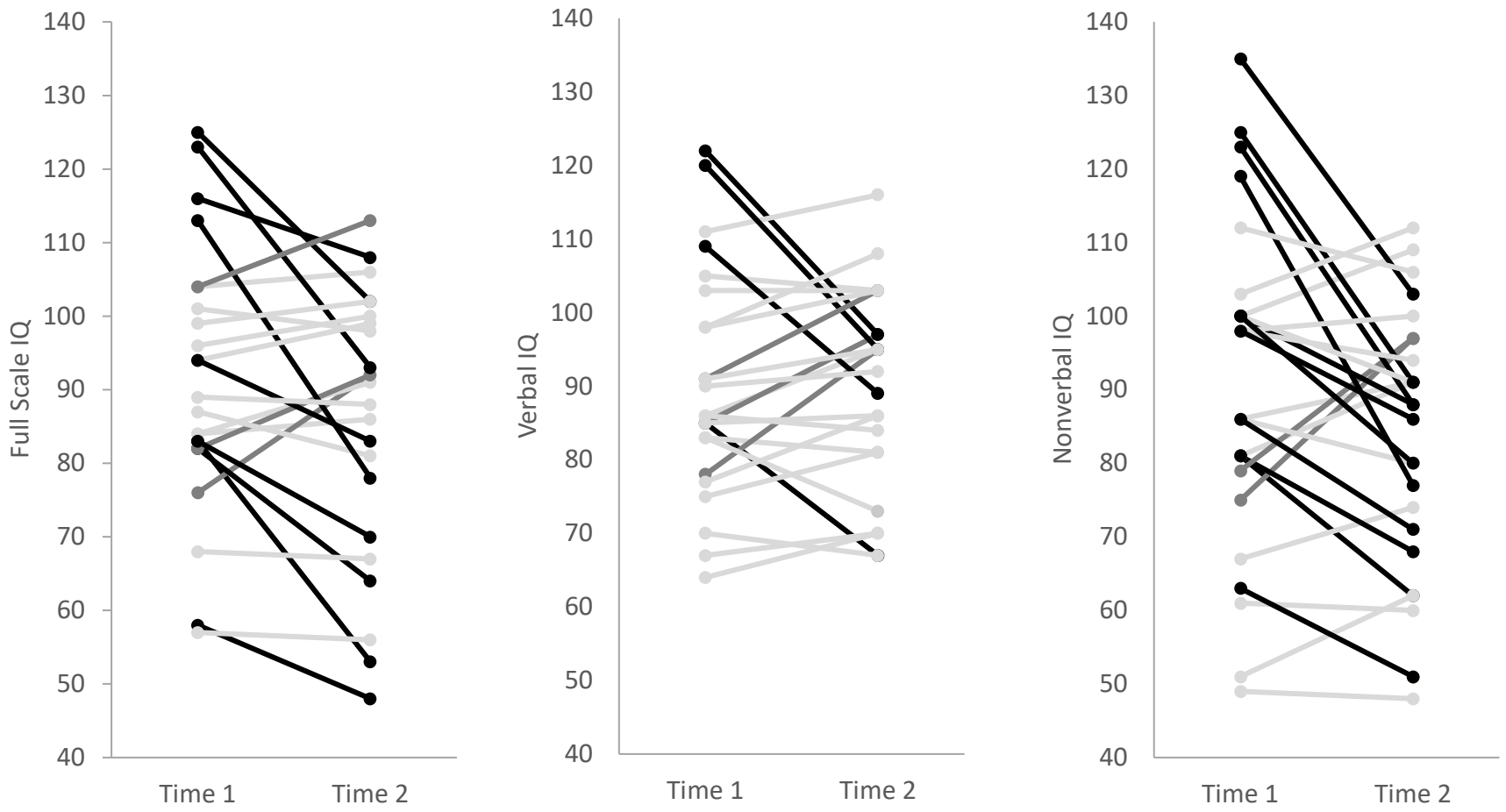


Figure 1

Reliable change in Full Scale IQ, Verbal IQ, and Nonverbal IQ scores between preschool age (Time 1) and adolescence (Time 2) in children with cerebral palsy

Chapter 6: General Discussion

The overarching aim of this thesis was to examine cognitive abilities in adolescents with Cerebral Palsy (CP) and explore clinical factors related to cognitive impairment and change or stability over time. This chapter integrates the findings, highlights their implications regarding theories of recovery following early brain injury, discusses clinical implications for assessment of cognitive abilities in adolescents with CP, and outlines limitations and future directions.

The findings of this thesis make several important contributions to the literature. Firstly, cognitive assessment using the Wechsler Intelligence Scale for Children – Fifth edition (WISC-V) in the traditional manner significantly underestimates the intelligence quotient (IQ) of adolescents with CP. More severe motor impairment was associated with greater disadvantage on the WISC-V. Interestingly, IQ was also underestimated in the group with the mildest motor impairment who, at first blush, may seem to have sufficient motor ability to respond to the tasks. This is particularly concerning given evidence the WISC is the most commonly used intelligence test for children and adolescents internationally (Oakland et al., 2016), and is frequently used to assess cognitive abilities in children and adolescents with CP.

An uneven profile of cognitive abilities in CP characterised by stronger verbal than nonverbal skills was confirmed, even after the impact of motor responses was considered. Although an uneven cognitive profile has been previously reported in the literature, studies were limited by the use of nonverbal subtests or indexes with a motor component which disadvantaged children with CP and contributed to poor performance in this domain. The use of the motor-free method minimised the motor demands of the WISC-V and characterised a motor-free cognitive profile in adolescents with CP. To date, this is the first application of the Piovesana et al. (2019) motor-free method in a clinical

sample and provides precedent for its use in other conditions characterised by motor impairment.

Another contribution to the literature was the use of individual-level analysis to examine cognitive change or stability over a 10-year period from preschool age to adolescence in a cohort of young people with CP. Intra-individual changes highlight considerable heterogeneity in the development of cognitive abilities which is unlikely to be captured accurately at the group level. Rather, the findings suggest three distinct trajectories of cognitive development: clinically significant decline, relative stability over time, and rarely, clinically significant improvement. Importantly, the findings illustrate how cognitive abilities in children with CP evolve over time and selective deficits may not be observable until a later age.

General Overview of Empirical Studies

Chapter 3, which compared methods that reduced motor demands to standardised administration of the WISC-V for assessing cognitive abilities in adolescents with CP offered two main contributions to the literature. First, adolescents with CP were found to have more difficulty on subtests of the WISC-V that require quick fine motor responses compared to similar subtests with reduced motor demands. Second, adolescents with CP are at an inherent disadvantage when cognitive abilities are assessed using the WISC-V in the traditional manner and benefit significantly from alternative methods for estimating IQ which minimise motor demands. Although the Nonmotor (Pearson Clinical, 2020) and Motor-free (Piovesana et al., 2019) methods were comparable in terms estimating IQ with minimal motor demands, the Motor-free method was recommended as the preferred choice given the strong psychometric properties and capacity to derive three primary index scores.

Chapter 4 subsequently employed motor-free administration of the WISC-V to characterise a motor-free cognitive profile of a cohort of adolescents with CP and explore clinical factors related to cognitive impairment. Findings highlighted a global decrement in the cognitive abilities of adolescents with CP. Notwithstanding the motor-free administration of the WISC-V, within-profile analysis confirmed an uneven cognitive profile characterised by a relative strength in verbal skills alongside a relative weakness in perceptual reasoning and working memory. Severity of motor impairment, low birth weight for gestational age, and seizures were identified as significant predictors of cognitive impairment in adolescents with CP.

Chapter 5 examined cognitive change or stability from preschool age to adolescence in children with mild to moderate CP using a longitudinal study design. At the group level, the results showed a significant decline in nonverbal IQ scores over a 10-year period with a marginally significant decline in FSIQ in the face of stable verbal abilities. At the individual level, reliable change scores indicated substantial heterogeneity in cognitive development. Clinically significant decline in FSIQ and Nonverbal IQ was associated with a history of seizures and higher initial IQ scores, respectively. The implications of these findings are that selective deficits can "emerge" during adolescence in CP, highlighting the importance of repeated cognitive assessment throughout development.

Theoretical Implications

While it is generally agreed the brain has some capacity to restore functions following early insult, the findings of this thesis suggest there are limitations to the brain's ability to compensate for early damage. The theoretical models of recovery following early injury to the developing brain have attempted to explain the range of functional outcomes following early brain insult. On one hand, early plasticity posits the

developing brain shows an advantage in recovery from early insult, and is associated with better outcome than similar brain injuries later in life (Kennard, 1942; Kolb et al., 2011; Kolb & Gibb, 2007). The pattern of global cognitive deficits identified in Chapter 4 suggest the development of cognitive abilities in adolescents with CP is not compatible with the early plasticity model. Rather, the findings of this thesis indicate cognitive development in the context of CP is perhaps more consistent with early vulnerability and cognitive crowding theories which suggest the severity of early brain injury has a significant impact on cognitive outcomes later in life.

Early vulnerability is based on the assumption that development of cognitive functions is dependent on the integrity of certain brain structures, and if these structures are damaged, the brain is never able to adequately compensate (V. Anderson et al., 2005; Hebb, 1949). Consistent with this perspective, the pattern of diffuse cognitive deficits identified in Chapter 4 suggests the acquisition of cognitive skills in adolescents with CP lags behind their typically developing peers as a consequence of early brain injury. Moreover, severity of motor impairment was identified in Chapter 4 as the strongest predictor of cognitive abilities across domains – more severe motor impairment as measured by the Gross Motor Function Classification System (GMFCS) was associated with greater cognitive impairment. If, in the absence of neuroimaging data, severity of motor impairment is used as a proxy for the severity of the initial brain insult, then these findings align well with the assumption of the early vulnerability model that more severe injury at a younger age results in global and persistent deficits.

Despite the global decrements in cognitive abilities in adolescents with CP compared to the normative average, within-profile analysis in Chapter 4 identified an uneven cognitive profile characterised by a relative strength in verbal skills alongside weaker perceptual reasoning and working memory abilities, lending support to the

cognitive crowding hypothesis. According to this theory, cognitive functions disrupted by early brain insult are subsumed by adjacent healthy tissue; however, more severe brain injury means less intact tissue to support the reorganisation of cognitive functions. To put it simply, multiple functions need to be supported by less brain. As a consequence of this competition for neural space, the cognitive crowding hypothesis postulates verbal skills are prioritised over nonverbal skills (Satz et al., 1994; Teuber, 1974). Although previous studies have reported an uneven cognitive profile in children and adolescents with CP (Carlsson et al., 1994; Gonzalez-Monge et al., 2009; Muter et al., 1997), it remained unclear if the nonverbal deficits were due to the crowding effect or due to the use of nonverbal tests with a motor component which, as shown in Chapter 3, disadvantage children with CP. The motor-free administration of the WISC-V in Chapter 4 addressed this limitation of previous studies. The persistence of an uneven cognitive profile despite the use of the motor-free method adds credence to the cognitive crowding hypothesis in two ways: first, it supports the contention of preferential development of verbal skills at the expense of nonverbal functions and second, it suggests that difference in verbal and nonverbal skills seen in previous studies cannot be attributed entirely to the impact of motor components of nonverbal subtests.

Moreover, the general pattern of cognitive change and stability from preschool age to adolescence observed in Chapter 5 can also be interpreted in view of the cognitive crowding hypothesis. Although Chapter 4 showed that verbal skills in adolescents with CP were lower than the normative average, the relative stability of verbal skills over time shown in Chapter 5 suggests a consistent rate of verbal skill acquisition. Based on the cognitive crowding effect, this relative sparing of verbal skills occurs at the expense of nonverbal abilities which, as shown in Chapter 5, declined significantly over time. Although basic visuospatial processing generally remains intact, more complex nonverbal

skills are compromised due to the competition for terminal space in the brain following early insult (Muter et al., 1997; Satz et al., 1994). As such, the extent of nonverbal impairment becomes more clearly identifiable at an age at which tests become more demanding on nonverbal reasoning skills, manifesting as a decline in nonverbal IQ during adolescence as seen in Chapter 5.

The impact of cognitive crowding on working memory ability has not been previously discussed in the literature. The findings from Chapter 4 showed that working memory ability of adolescents with CP fell significantly below the normative data. Moreover, verbal IQ is significantly stronger than both working memory and nonverbal abilities (which were not significantly different to one another), suggesting they may share common neural underpinnings that might be disrupted by the cognitive crowding effect. Functional MRI in typically developing adults has shown that working memory and nonverbal IQ share neural substrates in the lateral prefrontal and parietal regions (Gray et al., 2003). It is important to note that working memory was assessed by Gray et al. (2003) using both verbal and nonverbal working memory tasks while only verbal working memory tasks from the WISC-V were used in the current study. Overlap of nonverbal IQ and working memory ability has also been described by Alloway et al. (2016) in the context of neurodevelopmental disorders. While both verbal- and nonverbal working memory scores were lower compared to typically developing controls, severity of impairment in nonverbal working memory and nonverbal IQ predicted group membership for children with specific learning disorder, autism spectrum disorder, and intellectual disability (Alloway et al., 2016). This common profile suggests that the neural substrates which underpin nonverbal IQ and working memory might be vulnerable to cognitive crowding or disruption during development and may not be unique to CP.

Alternatively, the profile of weaker nonverbal and working memory skills compared to verbal abilities in adolescents with CP may be interpreted in the context of two distinct developmental trajectories. While the discrepancy between verbal and nonverbal skills may be explained by the cognitive crowding effect, the weaker working memory skills may be related to the protracted development of working memory as a component of executive functions (P. Anderson, 2002; Gathercole et al., 2004). In typically developing children, working memory development is an ongoing process from infancy through adolescence (Gathercole et al., 2004) with 'spurts' of rapid development between the ages of 7 to 9, and then again between 11 and 13 when working memory skills approach maturity (P. Anderson, 2002). As such, the deficits in working memory observed in Chapter 4 may be due to slower-than-expected acquisition of working memory skills in adolescents with CP.

Clinical Implications

Given the Weschler scales are the most widely used intelligence tests (Oakland et al., 2016), evidence that administration of the WISC-V in the traditional manner is underestimating IQ in adolescents with mild to moderate CP is important for several reasons. Firstly, accurate characterisation of intellectual abilities is crucial given cognitive assessments are often required in CP to determine eligibility for funding, support and entry into educational programs and settings. This is particularly relevant in the context of intellectual disability where diagnosis (or misdiagnosis) can have significant implications related to school placement and cognitive development (Ito et al., 1997). Secondly, building on previous findings by Losch and Dammann (2004), the findings in Chapter 3 highlight that accommodations during cognitive assessment are not only required for those with severe motor impairment who have clear, visible difficulties responding, but also for those with mild motor impairment who may appear to have sufficient motor

ability to respond to subtests. Finally, it was concluded in Chapter 3 that the Motor-free short-form (Piovesana et al., 2019) of the WISC-V is the most appropriate method for assessing cognitive abilities in adolescents with mild to moderate CP. A particular benefit of the Motor-free method is that it requires the administration of only six WISC-V subtests to produce a Motor-free IQ score and three primary index scores (Piovesana et al., 2019). As such, the Motor-free method reduces assessment time by almost half which is likely to be particularly beneficial when assessing children with low tolerance for assessment due to behavioural difficulties or fatigue. Thus, the findings from this thesis provide clear recommendations for clinicians in so far as the Motor-free method should be used in the assessment of IQ in CP.

The findings from Chapter 4 and 5 identified several factors that should be recognised by health professionals as risk factors for cognitive impairment in children and adolescents with CP. Severity of motor impairment and small birth weight for gestational age (SGA) were identified as significant predictors of cognitive abilities across domains, while seizure history was also associated with lower verbal abilities. Although previous studies have provided consistent evidence of link between severity of motor impairment and cognitive impairment, Chapter 4 highlights that motor severity remains an important predictor even after accounting for motor demands during assessment. In contrast, few studies have considered the impact of SGA on cognitive abilities in the context of CP. A novel finding in Chapter 4 identifies SGA as a significant predictor of cognitive impairment across domains and highlights the need for clinicians to recognise children with CP born SGA as at risk of later cognitive impairment and prompt ongoing monitoring of cognitive function throughout development.

While there is consistent evidence linking a history of seizures with a decrement in overall IQ in children with CP at preschool-age (Sigurdardottir et al., 2008), middle

childhood (Muter et al., 1997), and into adolescence (Cheng et al., 2013), an association between seizures and verbal IQ specifically, as seen in Chapter 4, is less commonly described in the context of CP. However, the negative association between seizures and verbal IQ has been previously reported in children with epilepsy (without CP). In a study of 136 children aged 8-18 years with either Temporal Lobe Epilepsy (TLE), Benign Epilepsy with Centrotemporal Spikes (BECTS), Juvenile Myoclonic Epilepsy, or absence epilepsy, results showed that verbal IQ, vocabulary, verbal reasoning, and category fluency were affected across all groups with the exception of children with BECTS (Jackson et al., 2019). Similarly, a significant decline in verbal IQ was observed over a two-year period between the ages of 9 and 11 in children with epilepsy while performance IQ remained stable over time (van Iterson et al., 2013). Previous research by Gonzalez-Monge et al. (2009) which suggested that seizures slowed the progression of verbal abilities over time in children with CP, taken together with findings from Chapter 5 that identified seizures as a risk factor for IQ decline, adds support for the view of an interplay between age, seizures, and cognitive abilities (Cheng et al., 2013). Seizures appear to contribute to a slower-than-expected rate of skill acquisition over time in children with CP highlighting the importance of strategies to control seizures in order to minimise cognitive consequences.

Chapter 5 also identified that children with CP and higher IQ scores at preschool age were more vulnerable to decline in nonverbal IQ over time. These findings suggest an ‘advance-plateau’ developmental trajectory of nonverbal abilities which is seemingly characterised by age-appropriate development to the age of 5 followed by slower progress during mid-childhood or adolescence compared to peers. Clinically, these findings suggest that a cognitive assessment at preschool age is unlikely to accurately inform the needs of a child during secondary school. Average or above average IQ at preschool age

should not be taken as a sign that the child will continue to be free of later cognitive impairments. Indeed, the findings from this thesis suggest that these adolescents with CP are at greater risk of decline, highlighting the need for ongoing assessment and monitoring of cognitive abilities throughout development to track changes over time.

Limitations and Future Directions

This thesis has three broad limitations that must be acknowledged. First, due to the small sample size, particularly the longitudinal cohort in Chapter 5, the study was underpowered to detect medium effect sizes. This limitation was partially offset by the use of reliable change analyses, which provides useful change-pattern data in small samples while taking into account test reliability and clinical significance. In addition, adolescents with severe motor impairment (GMFCS Level IV-V) were under-represented in the sample. Based on our experience with recruitment, a common limitation of studies in CP is that parents of children with severe motor impairment are less likely to participate, as previously mentioned in Chapter 4. Nevertheless, four adolescents with CP GMFCS Level IV-V participated in the screening phase, and two participants proceeded to the cognitive assessment. It is encouraging and important to note that we were able to obtain FSIQ scores for these two participants using the Nonmotor or Motor-free methods, when previously their cognitive assessment may have been based on limited subtests or subjective clinical judgement.

Despite the terms “Nonmotor” and “Motor-free”, the six subtests included in these methods require either a verbal or point response which demand a certain level of motor ability. Indeed, many adolescents with CP and severe motor impairment are unable to point or produce a verbal response, highlighting that the Nonmotor and Motor-free methods are more likely to be an appropriate solution for those with mild to moderate motor impairment. To this end, the use of additional classification measures such as the

Communication Function Classification System (Hidecker et al., 2011) or the Visual Function Classification System (Baranello et al., 2020) may have provided a better description of the sample and allowed for the control of variables other than motor impairment which may impact performance on cognitive assessments. Future research should consider validating the WISC-V for use with alternative response modes to improve access for participants with more severe motor impairment. Kurmanaviciute and Stadskleiv (2017) found that alternative response modes including gaze pointing and partner-assisted scanning did not influence how typically developing children scored on tests of verbal comprehension and visuospatial reasoning. While alternative response modes may be more time consuming, they provide the opportunity for children with motor impairment to demonstrate their true cognitive abilities (Kurmanaviciute & Stadskleiv, 2017).

Second, the Motor-free administration of the WISC-V may be criticised for potentially overestimating IQ as a result of omitting the Processing Speed Index due to the motor requirements of subtests in this domain. In doing so, an element of IQ – *cognitive* processing speed – has been overlooked which may be particularly relevant in CP due to the nature of the initial brain injury. Predominant white matter injury is the most common pathogenic pattern identified on neuroimaging in CP cases (Krägeloh-Mann & Horber, 2007), and white matter injury has been associated with lower processing speed scores on the WISC in adolescents who were born very preterm (Soria-Pastor et al., 2008). Although processing speed is typically included as a 'broad ability' in models of intelligence (Carroll, 1993; McGrew, 2005), exploratory and confirmatory factor analyses have shown that the Processing Speed Index has the weakest loading on the general intelligence factor of the WISC-V in both normative (Canivez et al., 2016; Wechsler, 2016) and clinical samples (Canivez et al., 2020; Stephenson et al., 2021). In

addition, there is evidence to suggest that the effect of processing speed on general intelligence is mediated almost entirely by working memory (Fry & Hale, 1996). Similarly, factor analytic studies have shown that working memory is highly related to general intelligence, and the impact of processing speed on intelligence remains negligible after accounting for the short-term storage component of working memory (Colom et al., 2004, 2008). Nonetheless, it remains a possibility that the motor-free method overestimated IQ as a function of omitting the Processing Speed index. Future research may consider utilising inspection time tasks (which manipulate stimulus presentation time and measure response accuracy rather than reaction time) as a measure of processing speed with minimal motor requirements (Hakkarainen et al., 2012).

Finally, Chapter 5 would be strengthened by the inclusion of an adaptive functioning measure to examine how motor-free IQ scores relate to daily functioning. Considering evidence of the pervasive effects of CP on lifestyle outcomes (Van Gorp et al., 2018), it would be important to understand how change or stability in motor-free IQ over time influences achievement of age-related milestones in personal, domestic and community activities of daily living. In addition, repeated assessments at regular intervals would increase understanding of the developmental trajectory of cognitive and adaptive functioning in young people with CP and potentially identify sensitive periods or triggers for cognitive change which could then be used as targets for intervention.

Conclusion

This thesis examined cognitive abilities in adolescents with CP and explored clinical factors related to cognitive impairment and change or stability over time. A significant shortcoming of previous studies was highlighted by findings that traditional cognitive assessment using the WISC-V placed adolescents with CP at an inherent disadvantage and underestimated FSIQ scores. The motor-free cognitive profile of

adolescents with CP was uneven, marked by relatively stronger verbal than nonverbal abilities which may be interpreted in the context of early vulnerability and cognitive crowding theoretical frameworks. The findings of this thesis highlighted that long-term cognitive outcomes are heterogenous in CP, in so far as cognitive abilities evolve from preschool-age to adolescence and selective deficits may not be observable until a later age in the context of increasing expectations of independence. Taken together, the implications of this thesis emphasise the need for cognitive assessments that cater for all levels of motor impairment in order to improve the understanding of cognitive development in young people with CP over time.

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