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What is quality of life for children with cerebral palsy? A constructivist grounded theory approach with implications for measurement

Elena Clare Swift

ORCID 0000-0002-3080-1971

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Melbourne School of Population and Global Health

The University of Melbourne

Abstract

Cerebral palsy (CP) is the most common physical disability in children in Australia and other high-income countries (Reid et al., 2016; Shaunak & Kelly, 2017). CP has permanent consequences across many areas of life from children's early development throughout the lifespan. In research and health service settings, quality of life (QOL) is recognised as a useful conceptual model for measuring subjective wellbeing across many areas of life, including those that may be impacted by various health conditions. Generic measures aim to capture QOL for all people, while condition-specific measures have been developed to address specific aspects of life that may be affected by specific conditions, such as CP. Four CP-specific measures of QOL have been developed for children, but there are shortcomings in both current conceptual understandings of QOL for children and in these measures, particularly in their inclusion of the views of children themselves. This thesis aimed to develop a constructivist grounded theory of QOL for children with CP including the views of children with CP and of their parents, and to compare this to the content of CP QOL-Child (Waters et al., 2006), an existing condition-specific measure of QOL for children with CP.

In-depth, open-ended interviews were conducted with 18 families of children with varying severity of CP, including 19 parents of children aged five to 12 years. Eight of the children aged nine to 12 years with mild or moderate CP also completed individual interviews. Interview topics ranged over many facets of child and family life, focussing on aspects that were beneficial or a hinderance to a good child QOL. Interviews were analysed using constructivist grounded theory methods proposed by Charmaz (2014). These methods are appropriate to research focusing on understanding lived experiences of participants and led to developing QOL theory that centred child and parent perspectives. Child interviews used a flexible child-friendly approach, including multiple strategies such as visual prompts and drawing materials. The use of multiple strategies contributed to child engagement and added to the richness of the data gathered, both in children's responses to the interviews and in the process of analysis. Analysis of child interviews included examining interactions within the interview, which helped to expose disability-specific assumptions and identities.

Overall, child and parent findings mirrored the same main contributors to child QOL, with some exceptions. Children provided greater insight into the complex impacts of social relationships on their QOL, while parents provided detail of wider family functioning that children may be unaware of, such as managing finances and balancing time between childcare and other responsibilities. For both children with CP and their parents, children's QOL was the result of interactions between person and environmental factors, including issues of accessibility and inclusion. Through relationships with family members, peers, teachers, aides, and other adults in their lives, children negotiated for adaptations to overcome environmental barriers and achieve QOL goals. For parents, supporting child QOL began from the time of diagnosis, and included day-to-day management as well as future considerations for themselves, their children, and other family members. Parents worked towards good QOL for their children through hierarchical tasks, from basic care needs, to participation goals, to future considerations and advocating for systemic change. The interactions between individual personal factors and environmental circumstances reflected in the findings support socio-ecological underpinnings of QOL definitions, and social models of disability. QOL research for children with disabilities would benefit from improved conceptual models of QOL that include disability models and are based on child data for child self-report measurement models, with parent report encompassing family QOL concepts.

Declaration

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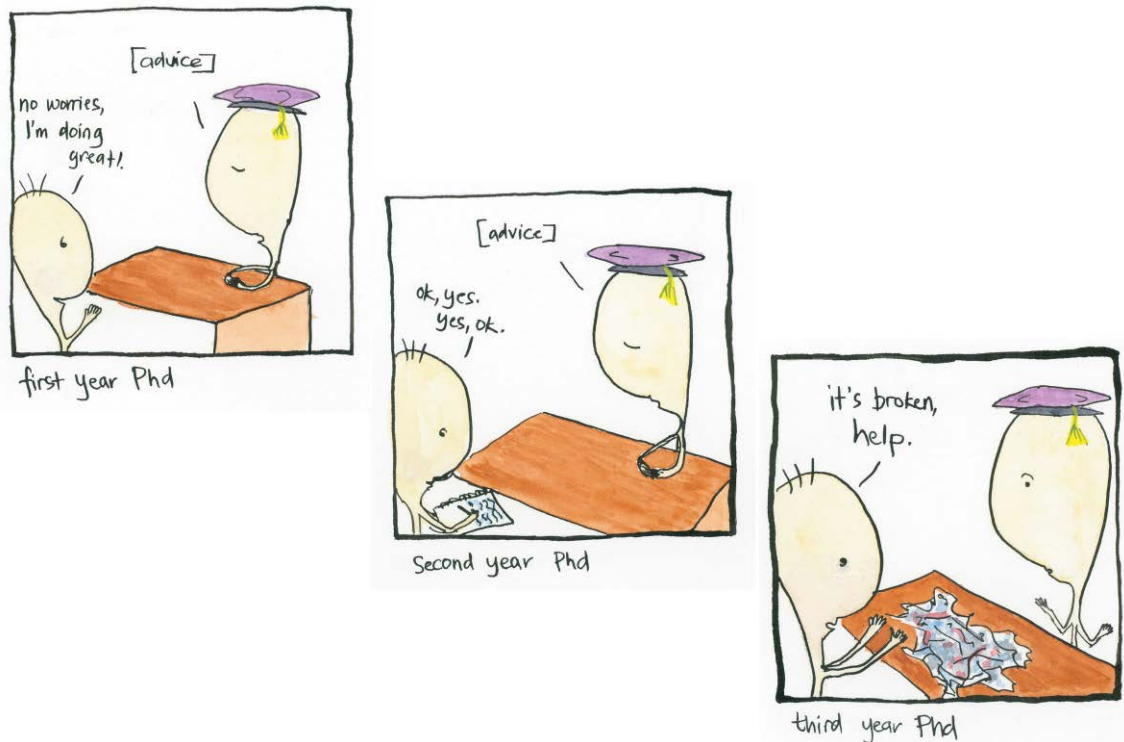
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My eternal gratitude to my supervisors who insisted on reading thousands of pages of incomplete drafts and enabled me to eventually form them into a coherent thesis. The illustration below accurately describes the supervision process.



To all the well-meaning but unwitting friends, acquaintances, and strangers who asked how it was going and opened themselves up to long rants on esoteric research issues: thanks for your patience and for pretending to listen.

I would not have completed without the existential and practical support of my family, friends, co-workers, and PhD 'writing' group. It's now your chance to say, "I told you so!", for all the times you insisted I could do it despite my ongoing scepticism.

This thesis is dedicated to Nannie and Small, with love always.

A brief diagram of the PhD process

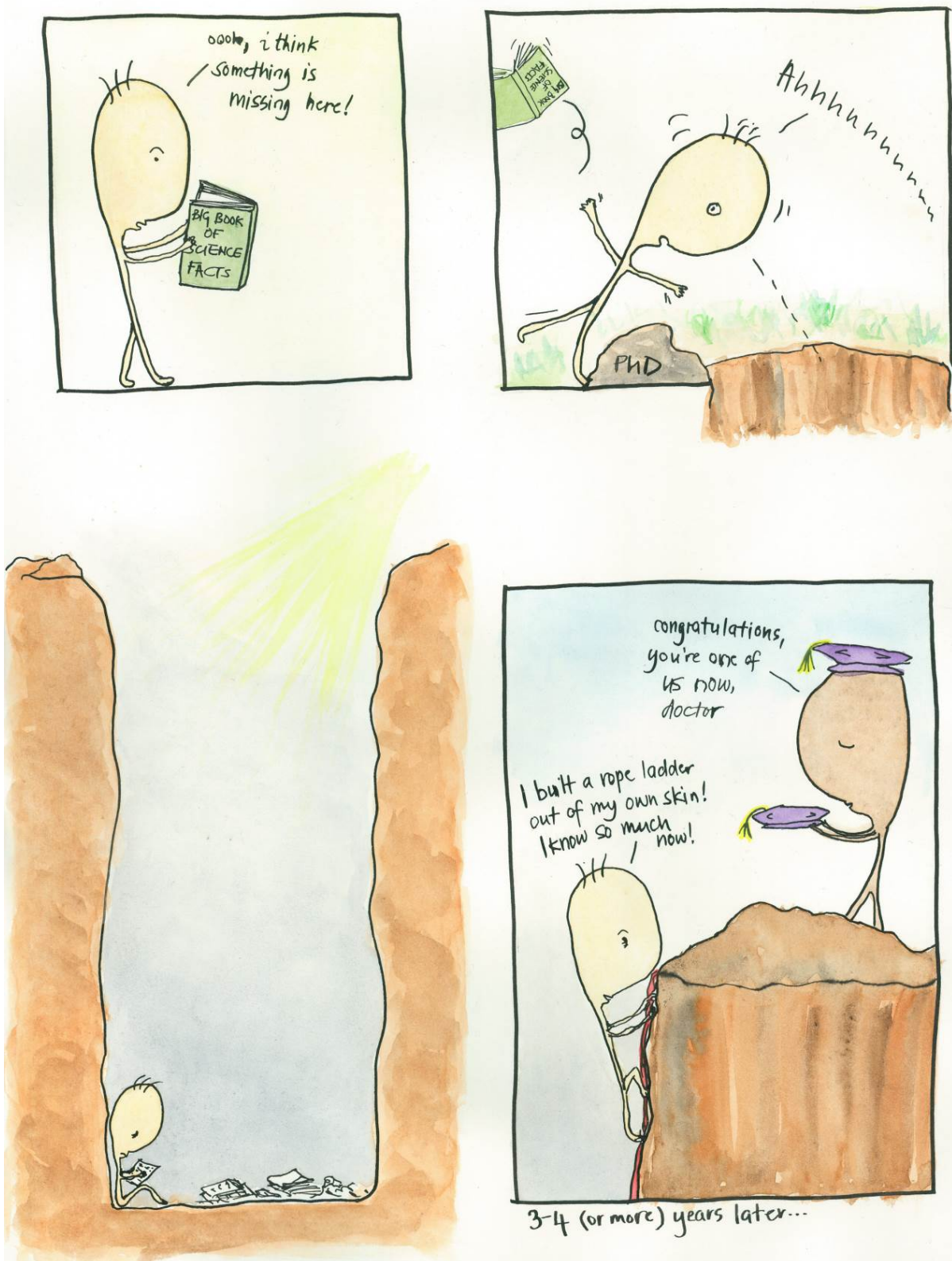


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List of acronyms

AAC	Alternative and augmentative communication
ConGT	Constructivist grounded theory
COSMIN	COnsensus-based Standards for the selection of health Measurement INstruments
CP	Cerebral Palsy
CPQOL-Child	Cerebral Palsy Quality of Life Questionnaire for children
CRC	Convention on the Rights of the Child
CRPD	Convention on the Rights of People with Disabilities
EBM	Evidence-based medicine
GMFCS	Gross Motor Function Classification System
HR-QOL	Health-related quality of life
ICF	International Classification of Functioning, Disability and Health
ICF-CY	International Classification of Functioning, Disability and Health—Child and Youth version
ID	Intellectual disability
ISOQOL	International Society for the Study of Quality of Life
MCID	Minimal clinically important difference
NDIS	National Disability Insurance Scheme
PRO	Patient reported outcome
QOL	Quality of life
SPARCLE	Study of Participation of Children with Cerebral Palsy Living in Europe
VCPR	Victorian Cerebral Palsy Register
WHO	World Health Organisation

Chapter 1: Introduction

Cerebral palsy (CP) is widely recognised as the most common physical disability in childhood, with an approximate prevalence of 1 in every 500 infants (Reid et al., 2016). It is a permanent, lifelong condition which has multiple impacts on many areas of a child's life beyond the motor impairments which are core to the diagnosis. In Australia, most children with CP are diagnosed during infancy or as toddlers (Novak et al., 2017) and throughout their childhood often receive a comprehensive suite of interventions and treatments from a range of health practitioners, including paediatricians, social workers, occupational therapists, and psychologists.

While CP is incurable, most of the interventions offered aim to improve motor and other aspects of function, such as activities of daily living and participation in schooling and recreational pursuits. Ultimately, disability services and interventions aim to ensure each child experiences their greatest possible quality of life (QOL). With the introduction of the National Disability Insurance Scheme (NDIS) and targeted disability funding in Australia, there is increasing emphasis on collecting outcome data for disability service providers (Reddihough et al., 2016). These outcomes frequently include measures of QOL. This raises a need to understand what constitutes QOL for children with CP and how to reliably measure it in the context of clinical interventions.

More than twenty years ago the World Health Organisation (WHO) defined QOL as “the individual's perception of their position in life, in the context of culture and value systems in which they live and in relation to their goals, expectations, standards and concerns” (WHOQoL Group, 1995). Despite its age, this remains the most widely used conceptual definition of QOL, as it encompasses both the subjective and multidimensional characteristics of QOL. Notably, QOL is not a measure of a person's functioning, but their satisfaction with their functioning relevant to their personal and cultural expectations. It encompasses areas of life beyond physical functioning that are likely to be impacted by disability and/or stigma related to disability, such as social inclusion, participation in leisure activities, and social/emotional well-being. Not only does it consider more than physical functioning, but it deliberately situates an individual's wellbeing within their socio-environmental context.

The WHO definition of QOL was carefully considered to be applicable to all people, regardless of health status or cultural context—but was designed for an adult population. How well this definition applies to children, or how it differs, is a matter of debate in research. There are likely developmental changes that impact on what is most significant to QOL as a child ages, which are not adequately explained in this definition. Twenty years ago Eiser (1997) commented on conceptual differences between children's and adults' definitions of QOL. However, in the literature there is still a general lack of a child-specific definition of QOL based on children's own understandings of QOL, which is needed to form a child-specific conceptual model of QOL and the factors influencing it.

Despite these limitations, there are many existing measures of QOL for children, including those applicable to all children (generic measures), and those produced for specific conditions and illnesses (condition-specific measures). Four condition-specific measures of QOL have been developed for children with CP—each with varied content, strengths, and limitations (DISABKIDS-CPM, Baars et al., 2005; CPCHILD, Narayanan et al., 2007; PedsQL, Varni et al., 2006; CP QOL-Child, Waters et al., 2006). While all four measures have been used in published research, there remain limitations both in the measurement of QOL for children with CP and in the conceptualisations of QOL underpinning these measures. The three main areas that require more research in the field of the measurement of QOL for children with CP are conceptual understandings of QOL for children with disabilities, the inclusion of child voice in research and QOL reporting, and how QOL changes over time in individuals.

A review of the use of QOL outcomes in clinical trials found that despite the strengths of existing measures, where such measures have been used in clinical trials for children with CP, they largely show no change in QOL outcomes (Tsoi et al., 2012). This may reflect the homeostatic theory of QOL, which posits that individuals show minimal change over time unless homeostasis is overwhelmed prior to intervention (Cummins, 2000). Therefore, unless children with CP face an overwhelming impact on QOL as a direct result of CP, CP-related interventions may be unlikely to result in changes to measurement of QOL. However, there are also several possible measurement related reasons why trials have generally been unable to demonstrate QOL improvements.

This may reflect the incomplete validation of existing measures—most QOL tools for children with CP have not demonstrated sensitivity to change, and none have information on what is considered a meaningful change score. This continues to present a barrier to use of existing QOL tools despite the increased interest in using QOL outcomes in both research and clinical practice (Carlson et al., 2010). The minimal QOL changes seen in clinical trials could alternately be related to a mismatch between the purpose of the measures and the purpose of the interventions. Interventions for CP usually target functional outcomes, but there is currently limited understanding of the relationship between functional outcomes and QOL for children with CP (McDougall et al., 2014). Without a better understanding of QOL for children with CP, it is hard to know how clinical practice or interventions could make changes to QOL for children, or what areas can be targeted to do so.

An ongoing challenge for the measurement of QOL of children with disabilities is the inclusion of children's perspectives in both development of tools and as reporters. Many existing QOL tools for children with disabilities have not included children's perspectives, or their carers', in the development of measurement content. This is despite definitions of QOL emphasising the subjectivity of QOL (for example, WHOQoL Group, 1995), and guidelines on development of QOL outcome tools stating that content validity relies on the inclusion of patient voice in measure development (for example, Food and Drug Administration, 2009). For children with CP especially, a secondary issue is that children are frequently excluded from self-report as QOL tools are not adapted for very young children, those with intellectual disability, communication impairment, or a combination of these.

Research has repeatedly shown discrepancies between parent report and child report that have not been resolved (for example, E. Davis et al., 2012; Dey et al., 2013; Jardine et al., 2014; Morrow et al., 2012). Parent report may offer additional information on family context which is relevant to child QOL, but this is usually not considered in current measures in which parent report is concerned only with providing individual proxy for children.

The third area of QOL measurement for children with CP that requires further research relates to the conceptual underpinnings of QOL for children with disabilities generally, and CP specifically. Existing measures of QOL for children with CP frequently diverge from multidimensional definitions of QOL by focusing on health status and functioning or measuring illbeing rather than wellbeing (E. Davis et al., 2006). Either case means that children are not able to report good QOL, only an absence of poor QOL. This might also be a reason that trials show limited change even where they have positive outcomes on other measures. More broadly, current theories of QOL that could inform measurement models have been developed for adults and do not consider differences in perspective or experiences of childhood that are qualitatively and developmentally different between children and adults (Priestly, 1998). Although there is a growing body of qualitative literature exploring children's perspectives of wellbeing (e.g. Hill et al., 1996; McWhirter, 2014; Sixsmith et al., 2007) this has not yet been translated into QOL theory.

Beyond literature specific to QOL of children with CP, there are ongoing debates about how QOL and disability are framed in research. The traditional medical model of disability characterises disability as individual, biomechanical impairments resulting from a medical condition, to be addressed in a health context with therapeutic interventions that aim to reduce perceived deficits in functioning (Shakespeare & Watson, 1997). From this perspective, it has been assumed that people with disability will report lower QOL than those without disability and require interventions to experience good QOL. However, many researchers have shown this is not the case (Albrecht & Devlieger, 1999; Dickinson et al., 2007).

The understanding of disability within this thesis is informed instead by social models of disability (Thomas, 2004b), which argue that regardless of individual impairments or differences in abilities, disability is the result of societal barriers that discriminate or exclude people with disabilities from self-determination or active participation in their communities. Rather than focusing on and categorising individual differences, this shifts the focus to rights of people with disability and the environmental context – though prominent disability rights activists and researchers have argued that the social model of disability does not need to exclude consideration of individual impairments

(Shakespeare & Watson, 1997). Therefore, while QOL concepts have often been framed as an outcome of disability interventions framed by the medical model, the WHO definition of QOL closely mirrors the social model of disability in emphasising the relationship between an individual and their environmental context.

Alongside the social model of disability, the thesis is informed by several other relevant conceptual frameworks. The interplay between personal and contextual factors in QOL is well framed by the socioecological model, as adapted for early child development (Irwin et al., 2007), which emphasises the inclusion of family context as well as wider societal consideration. . Family context can be informed by concepts of family QOL, which considers relational aspects of family life and the interdependence of QOL between family members (Boelsma et al., 2017). Individual factors of QOL for children with CP are informed by the International Classification of Functioning, Disability and Health (World Health Organisation, 2001), and child developmental theories and sociology of childhood studies. Lastly, rights-based frameworks for both children and people with disabilities inform the research process for how to include children with disabilities in research about them. Rights-based frameworks are examined through the Convention on the Rights of the Child (United Nations, 1989) and the Convention on the Rights of Persons with Disabilities (United Nations, 2006), both of which challenge researchers to amplify the voice of children with disabilities in research about them. Including rights-based frameworks in QOL research for children with CP strengthens the link to the social model of disability, which is grounded in the concepts of self-determination, inclusion, and participation of people with disabilities.

While there is limited QOL theory developed regarding children, there has been qualitative work focusing on conceptual models of children's QOL. The original work for the condition-specific QOL measure CP QOL-Child included development of a conceptual model based on perspectives of children with CP and their parents (Waters et al., 2005). However, in the time since this work was carried out, there have been significant technological changes in everyday life, including the rise of social media, that may have changed children's experiences of QOL in relation to communication or social interactions. There may also have been changes in therapeutic practice, such as

the increase in family-centred services (G. King et al., 2013), and these may influence children's experiences of QOL and CP.

The need to improve conceptual models of QOL for children with disabilities and to do so in a way consistent with children's rights led to the adoption of a qualitative methodology in this thesis. The qualitative approach is well-suited to explaining phenomena from the unique perspective of participants (Wertz et al., 2011), which parallels the subjective nature of QOL. Meanwhile, the reflexivity and flexibility in data collection allows for incorporating adapted interview methods to encourage engaged participation of children with CP and to examine and discuss influence of methods and interactions between interviewer and interviewee in the analysis of findings.

Specifically, the thesis adopted the constructivist grounded theory method presented by Charmaz (Bryant & Charmaz, 2007; Charmaz, 2014). This method emphasises the co-constructed nature of qualitative research, taking account of an active role played by both researchers and participants in creating data and interpreting participants' experiences in the research findings. The subjectivity and flexibility that Charmaz proposes is therefore ideally suited to exploring concepts of QOL for children with CP.

Better knowledge of children's experiences in relation to QOL and CP can inform a conceptual model of QOL for children with CP that recognises children's rights and values family perspectives.. This can improve existing QOL measurement models that endeavour to assess the need for and the contribution of specific interventions on the child's QOL, which will in turn strengthen clinical practice and improve the lives of children with CP.

1.1 Aims and research questions

The two broad aims of the research in this thesis are: 1) to develop a constructivist grounded theory of QOL for children with CP including the views of children with CP and their parents; 2) use this theory to review the content of the existing CP QOL-Child, for both child report and parent report versions, and discuss implications of any discrepancies.

Within the context of an exploratory conceptual model of QOL for children with CP this thesis will examine the following research questions to achieve these aims:

- a) What do children with CP and their parents consider contributes to the QOL of a child with CP?;
- b) How do child and parent perspectives relate to each other?
- c) How do child and parent perspectives relate to existing quantitative QOL measures for children with CP, namely the CP QOL-Child?

1.2 Thesis overview

This thesis comprises eight main chapters following this introduction. These cover background literature (chapters 2 and 3), methodology (chapters 4 and 5), findings (chapters 6 and 7), and discussion (chapters 8 and 9). Chapter 2 establishes background information on the effects of CP on children and their families, and medical approaches to management of CP and its comorbidities. This chapter also provides a critical review of literature on measuring QOL outcomes for these families, and its place in clinical interventions. Chapter 3 outlines a detailed overview of the relevant theory and conceptual frameworks that guide QOL research for children with CP and situates the thesis within a rights-based approach that incorporates person-environmental interactions of the social models of disability. From the introduction of the socioecological model, frameworks are arranged into those that emphasise environmental factors of children's lives, and those relevant to individual factors of children's QOL. The chapter concludes with an overview of existing QOL theory and research that has examined QOL from the perspective of children with disabilities. Chapter 4 establishes the basis for the use of qualitative methodology for investigating the QOL of children with CP, and outlines the methods used in detail.

The following three chapters summarise results of the research. Chapter 5 reflects on the methods used in the child interviews and Chapters 6 and 7 present findings from the qualitative analysis, from children and parents respectively. Chapter 8 compares and contrasts child and parent perspectives, links these findings to the existing CP QOL-Child, and highlights implications for future revisions of the measure and future work on QOL measurement in children with CP. The overall discussion in Chapter 9

relates the three results chapters to relevant research introduced in earlier chapters, and more broadly to the conceptual frameworks outlined in Chapter 3, while acknowledging limitations of the current work. Lastly, Chapter 9 provides concluding reflections on the current work and its contribution to the wider literature.

Chapter 2: Background

Motor impairments are the hallmark of CP, but these vary at the extremes from global impairments that prevent a child from walking or sitting unaided, to restrictions in fine motor control in one limb only. Necessarily, the corollaries of CP for these children will also vary. Therefore, the first half of this chapter introduces CP in children and some of its consequences and treatments, focusing on outcomes for children and families rather than pathophysiology or aetiology. This section includes how current interventions relate to evidence-based treatment principles and outcome measurement.

This leads to the discussion of the measurement of quality of life (QOL) in the second part of the chapter, from the perspective of both measurement theory and clinical implementation, and a critical review of the existing measures of QOL for children with CP. Finally, what is currently known about QOL for children with CP is reviewed.

2.1 Cerebral palsy in children

CP was first recognised as a disorder in 1861, at that time called ‘cerebral paresis’ (Rosenbaum et al., 2007). Since then the overall definition of CP, as well as how severity and subtypes should be classified, has undergone substantial change. CP results from neurological damage acquired prenatally, at birth, or in infancy (Graham et al., 2016). The severity of this damage and the pattern and severity of resulting impairments varies greatly, however there are commonalities in the process of diagnosis, likely interventions, and family impacts. These issues are discussed below.

2.1.1 Diagnosis of CP

While progress has been made since 1861 in understanding the aetiology of CP, in up to 80% of cases the causes are not fully determined (Novak et al., 2017). Furthermore, identified neural damage does not always correlate with the prognosis or severity of clinical symptoms (Novak et al., 2012). The diagnosis of CP is therefore based on the clinical presentation. In 2006 an international panel of experts defined CP as:

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

Diagnosis of CP is not always a simple process, as the lack of a single marker or cause means many other potential conditions must be excluded. In Australia, a child may be referred to many specialists before a diagnosis is reached, including general practitioners, child health nurses, paediatricians, neurologists and geneticists. Along with a primary diagnosis of CP, a child may receive secondary diagnoses of co-occurring conditions—this is especially likely for those with more severe CP (Novak et al., 2012). Comorbidities include epilepsy, vision or hearing impairment, intellectual disability, and others. These are all likely to contribute to early developmental delays, and additional consequences for the child and their family.

The different pathways to CP may also influence the process of diagnosis. A child born premature or with a traumatic birth requiring resuscitation or other known risk factors of CP is likely to receive close monitoring, resulting in early detection of abnormalities of movement, especially if the symptoms are severe (Graham et al., 2016). By contrast, a child born full term with no apparent difficulties may have seemingly normal development for the first several months of life before showing motor difficulties instigating medical investigation. Similarly, early development may be normal for infants whose CP results from injury or damage occurring in the first few years of life. Generally, altered motor function, or related delays in motor development, can be observed by 12–18 months of age (Rosenbaum, 2003). Children are usually diagnosed with CP between 12 and 24 months of age (Novak et al., 2017).

2.1.2 Population registers and prevalence

Population based registers of people diagnosed with CP have now been established in many countries. In Australia, the first population registers were State-based, with the first founded in Western Australia (Stanley & Watson, 1985), followed by Victoria and

South Australia. Every state and territory now has a register which collectively contribute to a national collaboration, the Australian Cerebral Palsy Register or ACPR (The ACPR Group, 2013). The basis for inclusion in the WA Cerebral Palsy Register is a definitive diagnosis of CP at 5 years of age (Blair et al., 2007). Before this age, a child may be provisionally enrolled on the register. These guidelines have subsequently been adopted by other State-based registers in Australia, as well as the ACPR. CP diagnosis is based on medical record review but referrals to the register may come from any health professional or from families or individuals themselves. Registers in Europe have adopted slightly different guidelines determined by the collaborative group, Surveillance of CP in Europe (Surveillance of Cerebral Palsy in Europe (SCPE), 2000). There, diagnosis is considered definitive at 4 years.

Differences in procedures for diagnosis, including enrolment in state or national registers, have broader implications for clinical guidelines, and to CP epidemiology. Much of the work on population prevalence, frequency of subtypes, and severity of the motor impairments, is based on information collected from population registers (Hurley et al., 2015). These registers now generally show a prevalence of 2 in 1000 live births, which has remained stable over time (Cans et al., 2008). Very recently there has been reports of a slight decrease both in prevalence and severity of the motor disorder across Australia (Galea et al., 2018; Reid et al., 2016).

Beyond epidemiologic research, differences in the timing of diagnosis may impact on the care a family receives or is able to access. A child diagnosed at an early age is likely to receive early intervention services sooner, and have better outcomes (Novak et al., 2017).

2.1.3 Classification of CP

There are three ways in which the motor impairments presented in CP may be classified. CP may be classified according to:

- (i) the bodily location and distribution of the motor impairment, for example, one limb (monoplegia), both legs (diplegia), one side of the body (hemiplegia), or all limbs and trunk (quadriplegia);

- (ii) the type of motor disorder, for example, muscle stiffness (spasticity), abnormal movements such as dystonia and athetosis, and ataxia (tremors), with some children having a mixed movement disorder;
- (iii) the overall severity of motor impairment.

The first two of these, bodily distribution and motor types, are widely used clinically as they give some indication of areas and directions for interventions. However, bodily distribution especially has been criticised for its lack of reference to severity of functional impairment, overlap between categories (e.g. quadriplegia versus diplegia), and lack of uniform application (Gainsborough et al., 2008). That is, an individual child may be classified in two or three different ways even by well-trained clinicians.

A well-recognised method of classifying severity of motor impairment is the Gross Motor Function Classification System (Palisano et al., 1997). The GMFCS was developed based on expert opinion of functional impairments, mainly the ability to walk, or sit unaided. It has a clear checklist for classifying children aged 2–12 years, which can be used by either families or clinicians. Research has shown good reliability and consistency in its application between families and clinicians, and across clinicians (Palisano et al., 2007). The GMFCS has five levels which describe the levels of severity, from level I—able to walk unaided, with impairments only in more advanced gross motor skills, through to level V—uses a wheelchair at all times and has limited gross motor control even with mobility devices.

The GMFCS is now widely reported in samples of children with CP. Prevalence estimates for each level have been reported for many different populations. In Australia, within children with CP the estimated prevalence levels across GMFCS are: level I: 34%, level II: 26%, level III: 12%, level IV: 14%, and level V: 16% (Reid, Carlin, et al., 2011). Although there may be some variation between countries and published prevalence estimates, generally the proportions are higher in the lower levels, such that children at levels I and II comprise the majority. GMFCS level status has been shown to be relatively stable from early childhood through adolescence (Palisano et al., 2007). The usefulness of GMFCS levels is not only restricted to epidemiologic or sample descriptions, but in the prediction of the lifelong consequences for children

with CP, and corresponding advice that can be given to families at the time of diagnosis. With current treatments and interventions, a child classified at age 5 as GMFCS level IV or V will never be able to walk unaided and will require significant carer support to live independently. By contrast, a child classified GMFCS level I can expect to achieve most of the mobility milestones of typically developing children.

2.1.4 Common comorbidities

Common comorbidities include epilepsy, intellectual disability, hearing, visual and communication impairments, and chronic pain (Odding et al., 2006). Frequency of comorbidities varies by subtype of CP and by overall severity. Children at GMFCS I often have no comorbidities, while those at higher GMFCS levels typically have at least one or more, with lifelong consequences for the child's outcomes in terms of immediate participation, medical involvement, need for mobility aids, and independence (Badia et al., 2015).

2.1.4.1 Epilepsy

Epilepsy is experienced in various forms by at least a third of children with CP, particularly those with the most severe motor disorder (Ashwal et al., 2004). The presence and severity of epilepsy generally has consequences for the treatment of CP. Uncontrolled epileptic seizures may cause additional problems through falling, pain in the affected limbs, and reduction in overall QOL (Cross et al., 2013); in severe epilepsy, seizures may occur multiple times a day for hours at a time. In most cases, children with epilepsy are prescribed antiseizure medications to reduce and control seizures, however many medications have undesirable side effects or interfere with other pharmacologic treatments. Where epileptic seizures are very severe and cannot be adequately controlled by medications, clinicians may recommend a ketogenic diet (a specific, highly controlled high fat, low carbohydrate diet) or other strategies such as provision of a vagal nerve stimulator or surgery (Cross et al., 2013). Control of epilepsy may be as great a challenge for parents and children as managing motor symptoms due to the supervision required, dietary manipulation, and administration of one or more medications.

2.1.4.2 Intellectual disability

Intellectual disability (ID) is estimated to be present to some extent in at least a quarter of people with CP, and may be as high as 44% or 1 in 2, depending on subtype (Novak et al., 2012; Odding et al., 2006). In those with very severe CP, i.e. GMFCS level 5, and high levels of communication impairment, intellectual ability is estimated rather than tested, as traditional methods of diagnosing intellectual ability generally cannot be used. In children with fewer motor impairments, traditional assessments of intellectual ability can be used. Overall, intellectual impairment is more likely at higher GMFCS levels (Reid et al., 2016), but importantly does not necessarily correspond with motor impairment. A child may be restricted in their ability to produce speech or move independently, but have no trouble comprehending information or learning in a supported environment. However, presence of intellectual impairment is generally associated with higher levels of other comorbidities, such as communication impairments, visual and hearing impairments, behavioural problems, and epilepsy (Badia et al., 2015; Odding et al., 2006).

2.1.4.3 Vision, hearing, and communication impairments

It can be difficult to estimate hearing and visual impairments in those with very severe motor impairments. Generally, in those with greater motor impairment, there are corresponding higher levels of additional perceptual impairments (Shevell et al., 2009). Hearing loss of any degree has been documented in 13% of children with CP in Victoria, Australia; 7% have at least moderate hearing loss in both ears (Reid, Modak, et al., 2011). Vision impairments of any form are more common, estimated at 28% of children with CP (Ashwal et al., 2004), with registry data showing severe impairment or blindness in 9.5% (Shevell et al., 2009). Hearing and visual impairments necessarily impact on a child's ability to interact with their environment, including communication abilities and use of alternative or augmentative communication aids.

While not technically a comorbidity, communication impairments are experienced by many children with CP, and are observed more frequently and with greater impact in those with higher GMFCS levels. Communication impairments usually take the form of difficulties in speech production or corresponding slowing of speech, related to gross

or fine motor impairments in the mouth and jaw and often accompanied by uncontrolled or excessive drooling (Parkes et al., 2010). Estimates of prevalence of speech production impairment vary according to severity; noticeable differences in speech production may affect 40% of children with CP (Cockerill et al., 2013). Several epidemiologic surveys have estimated approximately 20% of all children with CP are unable to speak or are classified as non-verbal (Andersen et al., 2010); among those at GMFCS levels IV and V, this increases to 60-80% (Cockerill et al., 2013).

Some children with communication impairments rely instead on alternate methods of expressive communication, known as augmentative or alternative communication (AAC). These include pictorial communication books or cards, hand signing, graphic methods, speech producing machines, or a combination of these methods. Recently, greater access to technology such as electronic devices and computer programs designed specifically for communication, adapted laptops and iPads, and smartphone apps, has given children increased options for communication (Light & McNaughton, 2014). For adolescents and young adults especially, social media offers additional opportunities for breaking down communication barriers (Hynan et al., 2014).

In most cases, children with communication impairments are likely to be much more easily understood face-to-face by their immediate families. In many cases, even where children are nonverbal and do not use AAC, parents or primary caregivers distinguish between a child's utterances and movements to understand when they are expressing emotions such as hunger, pain, satisfaction, annoyance, or pleasure.

2.1.4.4 Pain

Chronic pain may be more common than epilepsy in children with CP, but has historically been much more difficult to establish and reported prevalence varies widely (Penner et al., 2013). A recent review found reported pain prevalence of between 14% and 76% for children and young adults with CP (Mckinnon et al., 2018). Pain is generally more commonly reported and is experienced with greater frequency in those with more severe CP (Parkinson et al., 2010). While measures of chronic pain commonly rely on parent or clinician report for those with severe CP, children with limited communication abilities may signal quite clearly when they are in pain (Hadden

& von Baeyer, 2002). The relationship between CP severity and pain severity may be less pronounced for children who can self-report (Parkinson et al., 2010). However, it is likely the chronic pain among all children with CP is underreported and undertreated, and persists lifelong (Kingsnorth et al., 2015).

Chronic pain has multiple causes, but is most often related to musculoskeletal effects of CP and/or gastrointestinal symptoms (Penner et al., 2013). While medication may help reduce pain, it is unlikely to eliminate it, and pain medication may interfere with medications for other conditions and symptoms, such as epilepsy or control of motor symptoms of CP. Unmanaged, or incompletely managed chronic pain, is likely to interfere with many areas of children's lives, including their motor abilities, school functioning, emotional wellbeing, sleep quality, and QOL (Berrin et al., 2006; Vargus-Adams, 2016).

2.1.4.5 Somatosensory alterations

Related to hearing and visual impairments, somatosensory alterations are common in children with CP (Badia et al., 2015). Somatosensory alterations may take the form of specific deficiencies in proprioception, increased sensitivity to light or noise, or tactile sensitivity. Research comparing children with and without CP suggests the alterations in sensitivity to stimuli may originate in underlying neurological damage inherent to CP (Riquelme & Montoya, 2010). In general, somatosensory alterations are likely to increase a child's general level of discomfort in their environment and may restrict their ability to participate in many situations. Furthermore, increased sensitivity to pain may compound already existing pain, already common in children with CP.

2.1.4.6 Fatigue and sleep quality

Fatigue is commonly reported by children with CP at all levels of motor ability (Berrin et al., 2006). Functionally, fatigue reduces what a child with CP can physically achieve. For example, while a child may be able to walk with limited mobility aids, if this causes fatigue after a short distance, they are likely to require a wheelchair throughout their schooling to prevent fatigue interfering with their ability to concentrate and learn.

Fatigue in children with CP may be related to sleep quality and duration. Children with CP may be 7 to 10 times more likely than typically developing children to experience sleep disturbance, with greater prevalence for those at higher GMFCS levels or with other comorbid conditions (Verschuren et al., 2017). Poor sleep is often a direct result of underlying neurologic damage, but may also result from postural discomfort, medications for other symptoms, chronic pain, and in some cases, epilepsy. Unfortunately, poor sleep and the resulting fatigue is likely to feed back into poorer outcomes for all other symptoms and disorders which a child experiences.

2.1.4.7 Behavioural problems

A comorbidity that is often very challenging for families and schools to manage is that of behavioural problems, including emotional and social difficulties, hyperactivity and/or attentional deficits (Badia et al., 2015). Behavioural problems are reported by parents of approximately 40% of children with CP (Brossard-Racine et al., 2012), and may be more likely to be comorbid with intellectual disability (Parkes et al., 2008), and epilepsy (Carlsson et al., 2008). Not only are behaviour problems difficult to manage in a home or school environment, they can interact with a child's ability to adhere to intervention protocols, and indirectly reduce their functional outcomes. Behavioural challenges often include or lead to disruptions in social connections with peers and siblings (Brossard-Racine et al., 2012), thereby negatively affecting their wellbeing, and increasing their chance of bullying and exclusion.

2.1.4.8 Other CP related challenges

Other conditions that are often comorbid with a diagnosis of CP include difficulties with feeding, swallowing, drooling, and endocrinological and urogenital disorders. These disorders may result directly from motor impairments of CP, and can be present in up to half of children with CP (Odding et al., 2006). Children at GMFCS levels IV and V especially may be fed through tubes rather than orally; this is reported in 8% of all children with CP (Shevell et al., 2009). Along with endocrinological disorders, these conditions can result in nutritional difficulties, developmental delays, and weight problems (Badia et al., 2015). In day-to-day life, they also place a high burden on carers during a child's early life. For children with severe feeding difficulties, tube feeding is

usually easier for carers to manage and may circumvent problems such as aspiration with recurrent chest infections, prolonged meal times and poor weight gain. Similarly, in children with urogenital disorders including incontinence, providing guidance for the family in how best to prevent problems such as groin rash and skin breakdown is very important.

Like CP itself, there are guidelines developed by researchers and clinicians for management and treatment of most comorbidities. Depending on the severity of motor impairments, the comorbidities may have greater impact on a child's life than the CP itself. For example, a child at GMFCS level I localised to one arm but with a moderate intellectual disability will likely face greater developmental challenges because of their intellectual disability than motor challenges from their CP.

2.1.5 Impact of CP on children and families

The early onset of both CP symptoms and comorbidities have developmental and functional consequences throughout a child's life. During early childhood, children may be delayed in reaching developmental milestones, especially related to motor activities, such as sitting, crawling and walking (and those more severely affected will never achieve these milestones). These delays, as well as the presence of comorbidities, can result in secondary impairments and negative health outcomes throughout childhood and adulthood. Like many other disabilities, having CP is associated with reduced opportunities for inclusion in schooling and recreational activities, and poorer social outcomes. The financial costs of CP are high for individuals, families and carers, and society in general. Lastly, the impact of caring for a child with disability is likely to be felt by other family members, especially parents. Overall, the consequences of CP are felt in many ways.

2.1.5.1 Secondary impairments

As children age, CP conditions can result in secondary impairments that are not considered features of CP but impact children's lives. For example, motor impairments can lead to hip displacement, as well as other adverse health outcomes related to limited physical activity (Graham et al., 2016). Communication and cognitive impairments also often result in delays in language acquisition, greater challenges in

entering formal education, poorer family relationships, and poorer psychological wellbeing (Parkes et al., 2010). Children with feeding difficulties and gastroenterological symptoms are at greater risk of nutritional deficiencies, even from infancy, if they have difficulty in sucking or swallowing. From childhood through adulthood, people with CP experience significant barriers to achieving recommended levels of physical activity, resulting in poorer health outcomes in multiple areas (Verschuren et al., 2016). Older children and teens with CP also experience much higher rates of psychopathology than their typically developing peers (Rackauskaite et al., 2016). On a broader scale, children with CP are more likely to experience socioeconomic and health inequalities throughout childhood and adulthood—which may be compounded by the higher risk of CP in infants of families of lower socioeconomic status (Sundrum et al., 2005; Woolfenden et al., 2019).

2.1.5.2 Barriers to participation and inclusion

Even in families who do not face financial strain related to a child's disability, their child may face barriers to participation and access to many leisure activities, and in educational opportunities. Despite cultural changes in how people with disabilities are viewed, stigma remains, especially for those with more visible disabilities. Children with disabilities of any kind are more likely than typically developing peers to experience all forms of bullying, and have smaller peer social networks (Carter & Spencer, 2006; Nadeau & Tessier, 2006).

Environmental factors have a direct impact on the participation of children with disabilities in multiple ways (Colver et al., 2012). Mainstream community organisations, such as sporting or recreational clubs, vary in the encouragement they provide for participation of children with disabilities. This ranges from deliberately refusing to make adaptations or alternate ways of participating due to ignorance or lack of resources to do so, to providing specialised staff and adapted environments to support inclusion of children with special needs. Wheelchair users also face physical barriers in accessing recreational opportunities, despite changes in accessibility laws (Stancliffe, 2014). The variable practical difficulties of using public transport in a

wheelchair can reduce opportunities for participation even where inclusive recreation facilities exist (Pyer & Tucker, 2017).

As a precursor to independence and other adult achievements, school attendance and educational attainment necessarily varies greatly among children with CP. Children with mild physical impairments and minimal behavioural or intellectual problems are likely to attend a mainstream school with minimal additional classroom support.

However, even those with mild physical impairments may face challenges in mainstream schooling due to impairments in fine motor control which interfere with writing or limit writing speed. Other children attend specialised schools, or specialised classes within a mainstream school. The choice of enrolling their child in mainstream or special school may be complex for parents, including financial and geographical considerations as well as social and educational attainment goals for their child.

2.1.5.3 Costs of CP

The costs of disability are high for both health care systems and individuals: in 2007 in Australia the total annual cost of CP was approximately \$1.5 billion (*The Economic Impact of Cerebral Palsy in Australia in 2007*, 2008). In Australia, even with a universal public health care system and governmental support for many costs of disability specific services, most families face additional out of pocket costs for supplementary health care, equipment, schooling, and transport, that are not otherwise covered by subsidised services. In families who do not have additional disposable income, children will be more restricted in opportunities for leisure participation, programs or interventions that can increase functional mobility, schooling, and participation. These limitations will also likely extend to other children in the family, and to parental opportunities for leisure and respite from caring duties. Financial costs may be due not only to direct costs of services, but also may result in loss of income if parents are unable to balance employment with caring responsibilities. There are also costs associated with frequent trips to hospital or outpatient services and absenteeism from paid work. In 2007 informal care for people with CP cost an estimated \$129 million (*The Economic Impact of Cerebral Palsy in Australia in 2007*, 2008).

2.1.5.4 Stress on parents and other family members

Meanwhile, parents or primary caregivers generally share a great deal of responsibility for the health care and medical needs of their children. While they may receive assistance with respite and the provision of equipment, transporting a child to and from appointments and hospital stays can be burdensome. The period of time that follows receiving a diagnosis is likely to be traumatic for parents, as they grapple with grief for loss of expected child outcomes, and different expectations for the future. They also need to learn to deal with the often-overwhelming medical needs of their young child (Piggot et al., 2003). Before children are capable of making their own medical decisions, parents are usually responsible for deciding whether children should have invasive surgeries, receive additional interventions, wear assistive devices, and enforce dietary or exercise regimes in the home. They may experience limited support in finding information about their child's condition, or in negotiating what can be a bewildering array of health professionals involved in their child's care. For those with severe CP, parents are faced with the prospect of a child requiring lifelong assistance for basic functions. For those with less severe CP, there often remains a level of uncertainty as to how much independence a child may achieve, or how much assistance they will require as they age.

Within the family, the stress of diagnosis, and subsequent care for a child with a physical disability, impacts on parents' relationships, wellbeing, and finances (Raina et al., 2005; Reichman et al., 2008). These outcomes then have likely flow-on consequences for children (Carona et al., 2013). Many researchers have now shown that caring for a child with a disability results in poorer QOL, poorer physical health, and higher rates of mental illness, than parenting a typically developing child (Dambi et al., 2016). Early research suggested that parents of children with disabilities have higher rates of separation and divorce than parents of typically developing children, however more recent updates show the difference may not be very great (Freedman et al., 2012; Lundeby & Tøssebro, 2008; Risdal & Singer, 2004). Parental stress and mental health can not only increase the burden of caregiving (Carona et al., 2013), it also relates to poorer health outcomes for their children (N. A. Murphy et al., 2006). Some research suggests that siblings of children with a disability are also likely to have

higher probabilities of behavioural or mental health difficulties (Goudie et al., 2013), although there is limited research on outcomes for siblings of children with disabilities.

Overall, while the clinical presentation of CP in children is diverse—varying from mild to severe, with the likelihood of other complications—children with CP experience lifelong barriers to participation, which has a substantial impact on themselves, their parents, and extended community. In Australia, children with CP are usually diagnosed in infancy, live to adulthood, and receive a range of health services throughout childhood and beyond aimed at managing pain and discomfort, increasing mobility, and improving related outcomes. Further details of these services are discussed in the following section.

2.1.6 Health services for children with CP

Although there is no cure for CP, there are many therapeutic interventions which aim to improve various aspects of functioning for children with CP. There are many published articles that report evaluation of intervention trials, as well as many reviews of commonly used therapies. The Cochrane Database of Systematic Reviews includes more than 70 reviews related to interventions for people with CP and related physical conditions, including at least 15 specifically related to children with CP.

2.1.6.1 Treatment goals

As the neurological damage underlying CP cannot be cured with current treatments, many interventions are focused on improving a child's functional abilities in some way that can increase their participation outcomes and decrease their pain and discomfort. Increasingly, interventions are goal-oriented and strive for alternative mobility, inclusion, and participation, rather than achieving 'normal' mobility outcomes (Law & Darrah, 2014). For example, modern therapeutic goals may promote use of a wheelchair for mobility rather than focusing on efforts to walk 'normally'.

Recognition of the importance of factors outside of traditional measures of function relative to normal development has come in part from the World Health Organisation, with the 2001 publication of the International Classification of Functioning, Disability and Health (ICF; World Health Organisation, 2001). The ICF outlines in detail what

should be considered in a person's life to promote a state of perfect health. These factors are separated into two groups—functioning and health, and contextual factors. The recognition of factors outside of an individual's bodily functions and abilities as influencing health has had a great impact in broadening what are considered clinical goals, and what should be measured in considering the impact of medical interventions.

Clinical guidelines for management of CP span several practice disciplines, including paediatrics, surgery, physical and occupational therapy, speech pathology, and others. While treatments children receive vary by symptomatology and severity, most children with CP have consultations with health professionals from a variety of specialties. Each specialty may focus on a specific symptom or set of symptoms of CP. A paediatrician and sometimes also a general practitioner are likely to be the family's contacts for overall advice on their child's medical care and a source of referrals to engage with other specialities.

2.1.6.2 Clinical guidelines

Several clinical and research bodies have published clinical guidelines for the management of CP. These include those for both CP overall (e.g. The Royal Children's Hospital, Melbourne, Australia), and those for specific symptoms or aspects of CP (e.g. spasticity, Thompson et al., 2005). Very recently, the UK National Institute of Health and Care Excellence (NICE), published an extensive overall guideline for what is considered best practice in management of paediatric CP (*Cerebral Palsy in under 25s: Assessment and Management*, 2017), and the American Academy of Cerebral Palsy and Developmental Medicine (AACPDMD) has published a series of care pathways summarising best evidence for clinical care of various aspects of CP management (Fehlings et al., 2018). These clinical guidelines are based on reviews of available peer-reviewed evidence, collated and graded by researchers with significant clinical expertise.

Many more specific interventions are promoted to achieve various functional goals in children with CP, both within conventional and alternative medicine. Within conventional medicine, treatments have varying levels of evidence to support their

use. Even with the increase in evidence-based clinical guidelines, and large-scale reviews of commonly used treatments, there is research suggesting that common practice may not always be best practice (O'Connor et al., 2016; Saleh et al., 2008). Due to the diverse and individualised presentations of CP and accompanying comorbidities, the choice of treatment is often up to a clinician's discretion. Especially in the case of the use of multiple medications, it may be difficult to balance the positive effect of symptom control versus the negative consequences of side effects.

2.1.6.3 Common therapeutic interventions

For almost all children with CP, physiotherapy is the mainstay of treatment for motor impairments (Graham et al., 2016). Common physiotherapy-based interventions may include stretching and strengthening exercises, constraint therapy for children with hemiplegia (restricting the use of non-affected limbs to promote voluntary movements in CP-affected side of the body), or individualised goal-directed learning based on daily activities or participation aims. Aims include development of optimal motor skills, improved function and independence, and increased strength.

Physiotherapy may also be used to teach children and their parents about the use of mobility aids. In recent decades, there has been a gradual change in approach, moving away from promoting achieving 'normal' functioning, to promoting functioning that supports independence, with an increased use of mobility aids (Rosenbaum, 2003).

Common surgical interventions include spinal surgery for scoliosis to improve posture and comfort, muscle lengthening procedures for joint contractures and bony procedures for torsional deformities. Before children reach school age, they may have already experienced several surgical procedures, each requiring a hospital stay and usually a rehabilitation period. Physiotherapy is commonly used for post-surgical rehabilitation, to regain and improve function.

Another frequently-used treatment is botulinum toxin injections, particularly for children with spasticity. Numerous articles have been written on efficacy, dosage, and tolerance for botulinum toxin treatments alone or as adjunct to other therapy for improvement in muscular functioning. Botulinum toxin is also used to reduce drooling (Hay & Penn, 2011) and pain (Heinen et al., 2010). Within the published literature

there is strong evidence to suggest the efficacy of botulinum toxin interventions, as well as clinical guidance for their use (e.g. Heinen et al., 2010; Hoare et al., 2010). However, botulinum toxin injections have limited duration of action, usually with gradually decreasing effects after 3-6 months. Therefore, a child may receive this treatment on a repeat injection schedule of every 6-12 months. Physiotherapy is generally used to promote optimal functional outcomes following botulinum toxin injections, as with surgeries.

Many children with more severe CP receive pharmacologic treatments, prescribed for epilepsy, control of spasticity, spasms and tremors, chronic pain, and gastroenterological problems such as reflux and constipation. In children with very severe CP, prescription of multiple medications may become quite complex, with management of medication side effects, interactions between medications, and timing of dosages requiring additional clinical attention and parental monitoring.

2.1.6.4 Patient and family centred care

Parents usually manage diverse appointments, transporting children to and from them, implementing treatment regimes, advocating for better care, whilst at the same time experiencing their child's and their own distress. Among the bewildering array of treatments and frustration with a system that can be very difficult to manage, parents may choose to seek treatments outside of conventional medicine, especially where these potentially promise greater functional gains. Ultimately, whatever decisions are made by clinicians and families, even a child with very mild CP is likely to experience significant medical and allied health interventions aimed at improving their functional outcomes.

The move away from striving for 'normal' mobility to functional goals positions patient and carer priorities uppermost. Similarly, patient-centred, or family-centred care emphasises the importance of including a patient in decisions on interventions (Dickens et al., 2011). Beginning with discussion of treatment goals, patient-centred care moves away from a traditional model of medicine to a more collaborative approach. In the case of children, this necessarily involves the participation of families. Therefore, in best practice, family-centred care prioritises both collaboration with

families in treatment goals and incorporates existing family roles and strengths in planning how to implement treatment programs (S. King et al., 2004).

2.1.6.5 The Australian context and NDIS

In Australia, children with CP at all levels of functioning typically receive health services through a variety of settings, including hospitals, community rehabilitation centres, non-government agencies, primary practice, and the education system (Meehan et al., 2016). The source of their medical support is generally dictated by their age and may vary from state to state. Up to school age, children with disabilities are eligible to receive many non-hospital health services from early childhood intervention services that are frequently community organisations with multidisciplinary teams that usually include case coordination.

At school age, children transition from early childhood intervention services to more general child disability services, and they receive many interventions through school-supported funding. Whether children are eligible to receive funding and the support services they receive are based on both the severity and complexity of their disabilities, and the school in which they are enrolled. For example, a child in a rural community attending a small school with no other disability-identified children may receive more limited school-based support. In contrast, a child attending a much larger metropolitan school with more children with disabilities, or with previous accessibility improvements for children with physical disabilities may have access to a wider variety of supports within their school. Therefore, a family's choice of whether to send a child to a mainstream or special school may also be dictated by multiple factors—not only by the availability and location of schools, and educational attainment goals—but by the funding and supports that will be available in either situation. Even if families are facing only a choice between mainstream schools, or between public or privately funded mainstream school, they are very likely to be influenced by the accessibility, support and funding available at specific schools, as this has daily and long-term impacts on a child's life.

The transition from primary to secondary school can also be a time at which services for children with disabilities change. Often, as young people move from early

childhood through adolescence, and eventually to adulthood, the range of services that they are able to access decreases—even though their medical or support needs may not change.

Australia has recently enacted substantial changes to disability service funding with the introduction of the National Disability Insurance Scheme (NDIS). The NDIS aims to increase the independence of consumers receiving government disability support, by providing them with individualised funding packages. Each service user is then able to choose what health services they access, rather than having this choice being dictated by systemic factors (Reddihough et al., 2016)—though choice may still be limited by availability of services. The NDIS was first introduced to trial sites in 2013, and from 2016 has been implemented in stages across the country. The ongoing changes to disability health services has resulted in substantial uncertainty for families in accessing funding and services for their children, with poor access to information for families on the scheme (Simpson & Baldwin, 2017), and a complex process to establish funding allocations (Foster et al., 2016). Early reports of experiences of families who have transitioned to the NDIS are mixed (Ranasinghe et al., 2017; Snow & Donnelly, 2017).

With the introduction NDIS, there has been an increased focus on service evaluation across disability related health services (Imms et al., 2015). Not only has the NDIS resulted in funding changes for people with disabilities and their families and carers, the funding structure for most disability service providers is also changing. Consequently, service providers are recognising an increased need for validated outcome measures, both to attract and retain consumers.

2.1.7 Evidence based medicine and patient reported outcomes

The changing landscape of disability services in Australia is leading to an increasing use of service outcomes. However, evidence-based medicine (EBM) itself is not new, nor is the use of validated outcome tools in clinical practice as advocated by EBM. EBM is especially relevant for children with CP, who are likely to receive a diverse range of interventions of varying degrees of evidence. The main principles of EBM in practice include having and using the best evidence for treatment (i.e. high-quality research),

service providers maintaining knowledge of current treatments, and the use of validated outcomes in practice.

2.1.7.1 Use of validated outcome tools in clinical practice

Guidelines for best practice in health services for paediatric disability recommend monitoring patient progress with the use of validated outcome measures and basing treatment protocols on those supported by good quality evidence aligned with patient priorities (Palisano, 2006). Despite this, a recent systematic review of reports of the practices of allied health practitioners in Australia found that very few available evidence-based outcome tools are routinely used in paediatric CP services (O'Connor et al., 2016). Evidence-based outcome tools are those with evidence of acceptable validity and reliability and published information on their purpose, intended use, scoring and interpretation. Tools being regularly used in the review by O'Connor et al. related to classification of impairments and functional assessment, while the use of tools relating to other areas of life (such as QOL) was rarely reported. This review included published literature from 2000 to 2015, including literature on validation of outcome tools. However, some outcome tools specific to childhood CP have only been developed in the last five years, with validation information published more recently, suggesting that practices reported by O'Connor et al may be outdated. Without systematic measurement using validated tools, it is difficult to judge how standard clinical practice is improving the lives of children with CP.

2.1.7.2 Patient reported outcomes (PROs)

Beyond the outcome tools related to functional assessment, the introduction of patient reported outcomes (PROs) to clinical practice is a new area of research that is receiving significant attention. PROs are measures that ask a patient to report directly on various issues of concern to them, in contrast with clinician-based measures, which focus on expert knowledge. Additionally, clinician-based measures usually focus on objective, observable factors; PROs may be more concerned with subjective measures such as feelings about health or various other aspects of life, or patient goals. PROs are particularly relevant for patient- and family-centred practice.

Researchers, policy makers, and others are increasingly advocating for the use of PROs not just in practice, but in clinical trials (Janssens, Thompson Coon, et al., 2015). In 2004 the National Institutes of Health in the USA established a group to develop a series of PROs for use in various chronic illnesses in research and health services (Cella et al., 2010). More recently, the National Health Service in the UK has sought to establish appropriate PROs for evaluation of health services for children and young people with disabilities (C. Morris et al., 2014). Especially in complex, lifelong conditions such as CP, patient perspectives are invaluable to inform directions of practice and research which have impacts observable to the individual. Clinical outcomes may translate to very little change in a person's day-to-day life if they are not relevant to patient goals or day-to-day living environments.

While a strong and sensitive clinical relationship may make such clinical gains or environmental influences obvious, to capture these outcomes systematically, PROs are invaluable (Bevans et al., 2014). There are many different types of PRO measures, varying in content and in how they are applied. Generic instruments are applicable to all people and can be used to make comparisons across population groups, for example, children with CP and typically developing children. By contrast, condition-specific instruments are designed for greater specificity and sensitivity for one health condition but cannot then be used to compare patient groups with different conditions.

To be useful in producing good quality evidence to support practice, PROs must be well-constructed, reliable, and valid for the purpose. There are now numerous publications detailing methodologies for validating PRO instruments, and a similarly large number reviewing the psychometric properties of published PROs. One valuable new tool for exactly this purpose is the COnsensus-based Standards for the Selection of health status Measurement INstruments (COSMIN; Mokkink et al., 2010). Through literature, expert opinion, and a series of publications on its construction, the COSMIN standards were developed with the aim of standardising validation practices, and to assist in evaluating validation studies of PROs. By evaluating the validation of a PRO using the COSMIN standards, clinicians and researchers can determine the likely

usefulness and application of measures and decide how best to measure outcomes for their patients.

Many PROs include content measuring function and overall health status not dissimilar to traditional clinician-based measures. While this is valuable information, the focus of PROs is a patient's own perspective, both during scale development and in responding. This makes them ideally suited to measuring subjective aspects of patient wellbeing such as QOL. QOL measures have emerged as a valuable type of PRO for clinicians and patients to capture an overall picture of the impacts of a disability on a person's life and wellbeing beyond functional status.

2.2 Measuring QOL

One major goal of understanding QOL for individuals, especially in a clinical context, is to maintain or improve it. Appropriate instruments, validated for the purpose, are a necessary component to knowing if this is happening. There are some broad considerations in QOL measurement, including challenges of measuring QOL in children, which are detailed first. Principles of measurement and evaluation of QOL measures need to be understood before evaluating actual measures, so these principles are described next, including how they apply to the development of QOL measures.

2.2.1 Challenges in measuring QOL

As for other PROs, QOL measures may be generic, chronic-generic, or condition-specific. Generic measures are applicable to the general population and may be compared across groups of individuals with or without different health conditions—though many were not developed specifically for use in health contexts. Chronic-generic instruments are designed for individuals with chronic conditions, but may be compared across these conditions, for example, comparing children with CP and children with asthma. Condition-specific measures are targeted towards groups of individuals with a common condition, such as CP, and aim to capture additional information relevant to that condition as well as general QOL. In general, outside of comparing QOL across groups, condition-specific measures are preferred for their

greater specificity. This is especially the case in using measures for evaluating interventions, or for individual clinical practice (Trama & Dieci, 2011).

Given the subjective nature of QOL, it is usually measured through self-report questionnaires. One of the basic challenges of measuring QOL in children, especially in those with disabilities, is in balancing the need for a self-report with what a child is able to report about family circumstances or abstract concepts that may be beyond their ability to convey. The ability of children to self-report can also be dependent on how QOL has been defined and operationalised as a measure. Therefore, defining QOL for condition-specific measurements is discussed below before reviewing challenges in child report versus parent report.

2.2.1.1 Theoretical challenges

In adult literature, the most well-known theory of QOL change is Cummin's homeostatic model which states that subjective QOL in humans is homeostatically maintained within a narrow range such that after significant events or circumstances causing shifts in QOL individuals generally adapt and return to the same previous range (Cummins, 2000). While there is some empirical support for this theory, conflicting research shows it does not fully account for QOL variation within individuals over time. For example, the WHOQOL group in the UK, using the WHOQOL instruments, demonstrated that both chronic pain (Skevington et al., 2001) and untreated mental illness (Skevington & Wright, 2001) reduce overall QOL, and relieving symptoms can lead to increase in overall QOL. The merit in the homeostatic model is that it draws attention to internal personal factors which impact QOL, and which are highly resistant to change over the long term.

2.2.1.2 Defining QOL for measurement

In developing review criteria for QOL instruments, Lohr et al. (Lohr et al., 1996) drew a distinction between conceptual and measurement models of QOL. The conceptual model provides the descriptions of underlying concepts an instrument is intended to target, including their relationships to each other. The measurement model refers to the structure and items of the instrument itself, including how items are scored and the validity of scoring relative to the measurement and conceptual models. More

recently, others have highlighted the paucity of conceptual models in existing paediatric QOL tools, and the limited information provided on how conceptual models have been operationalised (Waters et al., 2009). If the conceptual factors of children's QOL are unknown, or have not been defined for measurement, the resulting measures cannot reasonably claim to be valid.

The challenge for condition-specific measures is to balance the need for specific outcomes relevant to the condition with the use of a QOL definition that maintains the multidimensional concepts of most definitions of QOL, such as that of the WHO (see page 1). One challenge within research on QOL for health outcomes is that historically there has been limited distinction between measures of functioning or health status, health-related QOL (HR-QOL), and a more holistic version of QOL encompassing concepts of wellbeing outside of health and functioning. Furthermore, the definition of QOL used in a measure may differ from definitions of QOL used in conceptual models (Morrow et al., 2010). Whether an instrument truly measures what it purports to relative to its QOL definition or conceptual model is a topic for validation, as explained in measurement principles in the next section. However, the way in which a conceptual model and definition is operationalised for measurement is a necessary part of evaluating existing measures.

Koot (2014) provides an excellent review of some distinctions in definition of QOL concepts in health measurement, noting that how QOL has been operationalised has been almost instrument specific. Nevertheless, most researchers agree on a few key points in defining QOL: that it encompasses multiple areas of life across physical, social and emotional factors, within expected contextual or cultural settings. Within QOL measurement, each area of life is referred to as a domain; a QOL tool may include a physical domain, an emotional domain, an environmental domain, and so on.

Overall QOL differs from measures of function because it encompasses an individual's own experience and feelings about their functioning (E. Davis et al., 2006). Similarly, where health status may be considered a checklist of symptoms or symptom severity, QOL reflects how satisfied a person is with their health, or in what ways their health status impacts their ability to enjoy or achieve in other areas of life. From the

perspective of individuals with chronic illness or disability, they may not have high scores on measures of health status or functioning, but they may still be able to live full and happy lives outside of their condition or have supports to reduce the negative impacts of their condition, thus showing high QOL. This has long been demonstrated by many of those living with disability (Albrecht & Devlieger, 1999).

At the time that QOL was first being considered an important outcome in healthcare, many measures were developed targeting health-related QOL (HR-QOL). Health-related QOL may be considered to be a person's subjective feelings of well-being and satisfaction across domains related specifically to their health, illness, or functioning (E. Davis et al., 2006). HR-QOL is usually now considered to be a subset of overall QOL despite the focus on the respondent's subjective experience of multiple domains of life, as it is restricted to factors specifically related to an illness (Koot, 2014; Morrow et al., 2010). However, confusion exists in the use of HR-QOL versus QOL measures, with many trials reporting outcomes from HR-QOL, or measures of health status and functioning, as a QOL outcome. While HR-QOL may be a valuable outcome for a health service intervention, ultimately it is only a small part of QOL. It is especially common in chronic illness and disability to conflate HR-QOL as QOL in measures. Throughout this thesis, QOL refers to overall QOL including both HR-QOL and other areas of life.

2.2.1.3 Parent report versus child report

Self-report is the preferred method of collecting QOL data, as QOL is based on the subjective evaluation of an individual's own life (Eiser et al., 2000). However, in the case of children, QOL is often measured using parent proxy in place of or in addition to child self-report. Proxy report is usually used when children are too young to understand QOL concepts, or when they have communication or cognitive impairments which limit their ability to complete the questionnaires.

Developing a child report version of QOL relies on two aspects of measurement: the underlying concepts must be understood by children and must be in a form in which items asked can be easily answered by children. Most researchers agree that before the age of 8 or 9 years old children are not well able to understand abstract concepts such as 'wellbeing', or to use and respond accurately to Likert response scales usually

used in PROs (Eiser et al., 2000). A few measures have been developed specifically for younger ages using alternate response methods. For example, the Take 6 is a HRQOL measure for children with chronic illness aged 4-7 years which uses a pictorial Likert response scale based on smiley faces (Chaplin et al., 2008). However, almost all child report versions of QOL measures are restricted to ages 9 and older, including all condition-specific measures for children with CP. In contrast, parent report scales have been used for children as young as 2 years old (for example, the PedsQL in Varni et al., 2007).

Where researchers have been able to capture both child self-report and parent-proxy, there is a trend toward children reporting higher QOL, with notable discrepancies between parent and child report in psychosocial and emotional domains (E. Davis et al., 2012; Dey et al., 2013; Robitail et al., 2007). Several studies have shown higher agreement on physical domains, and lower agreement on subjective/emotional domains (Jardine et al., 2014; Majnemer, Shevell, Law, Poulin, et al., 2008; Morrow et al., 2012), which may explain why it is often cited that there is overall good agreement between child and parent report. Others suggest that there may be greater discrepancy between parent and child report as children age (Cremeens et al., 2006). A recent well-cited review of generic instruments overall concluded that there is poor reliability between parent and child report on QOL instruments (Janssens, Rogers, et al., 2015).

In considering parent report data on child QOL, is it useful to consider other aspects of the reporting parent's life that may impact on the QOL they report for their child. If a parent is distressed by their child's condition, has a current mental health condition, has poor QOL of their own, or is seeking additional support services for their child, they are likely to report poorer QOL for their child—even where the child themselves reports high QOL (E. Davis et al., 2012). Overall, parent report is influenced by the parent's own QOL or psychological distress (Cremeens et al., 2006; E. Davis et al., 2012; White-Koning et al., 2007), possibly as much as by how the child truly feels.

Qualitative research into the discordance between child and parent report data has also highlighted differences in the way children and their parents respond to

questionnaire items (E. Davis et al., 2007). Davis and colleagues used a cognitive interviewing approach where participants worked out aloud how they would answer each item on a QOL scale. This is a commonly used technique in instrument validation, to monitor how items are interpreted, and whether the way they are being answered is very different to how it was intended by the instrument's designers. In Davis et al. (2007), children considered immediate events from their past day or two as indicative of how to answer QOL items. Parents, on the other hand, took a much longer-term view, considering general tendencies in their child and how they felt the item best reflected their situation over a longer period of time. Similarly, other qualitative work with children with chronic illness and their parents has highlighted the additional work parents engage in to increase their children's physical activity, which is invisible to children and highlighted only in parent interviews (Fereday et al., 2009). These studies show that even when there is a high correlation between child and parent report data, it is likely that it is based on different information.

This not only highlights the importance of self-report data, especially where QOL outcomes are used to inform patient priorities for treatment but also suggests that QOL information reported by parents is qualitatively different than children. Recently other researchers reviewing QOL instruments for children have recommended that proxy report should be considered to be a different measurement, and named parent report (Eiser & Morse, 2001; Janssens, Rogers, et al., 2015). That is, children and parents offer different and unique information on child QOL, and both should be taken into account when examining child QOL. That is, parent report instruments should also consider content of issue to parents, because even where this is not strictly child QOL, it is likely to affect a child's life and QOL. It is therefore feasible to consider that a QOL scale for parents to complete on behalf of their child may include significantly different content to a version for the child to report on their own QOL. Condition-specific measures of QOL should provide a self-report form for children wherever it is feasible to do so, adapted to be comprehensible to children of varying ages and level of disability. The next section discusses some considerations in adaptations for young children and those with disabilities which may interfere with their ability to complete a traditional self-report scale.

2.2.1.4 Accessibility for children with disabilities

Questions remain over the appropriateness of QOL measurement for very young children. Children tend to comply with adult instruction and complete a questionnaire even if the items are meaningless to them due to poor comprehension (Eiser et al., 2000), yielding responses that may be inaccurate or meaningless. Contemporary self-report measures of QOL for children have infrequently examined children's comprehension of QOL items, which is an aspect of measurement validity. Children with CP are more likely to experience cognitive difficulties which reduce their comprehension even at older ages (Sakash et al., 2018)—especially for those with intellectual disability. For many children unable to independently complete a measure but able to communicate with a parent or another adult, completing a measure may require only that items are read to them with the child answering verbally, or using an alternate means of communication to indicate a preference. While this may occur naturally with existing child self-report measures, in fact little is reported in the literature about how parent- or clinician-assisted administration may change child responses. This is concerning in the context of the many reports of discrepancies between child-report and parent-report with QOL measures (see page 34).

For other children, alternate response scales, such as visual scales that use either graphic response options or sliding scales instead of words, may make reporting more accessible (for example, with use of smiley faces indicating responses, Chaplin et al., 2008). Similarly, for very young children, some researchers have been successful in simplifying items to language appropriate to a child's level of development, for example by recording children's language in discussing QOL concepts in focus groups (Jirojanakul et al., 2003). This is a necessary process regardless of the abilities or impairments of intended child respondents. However, there are still gaps in research on how this changes the information that children report, or the validity of these scales. For example, visual response scales for young children generally show lower internal consistency than is considered acceptable for health measures (Chaplin et al., 2008), while others have shown that children avoid neutral or unhappy smiley faces in response scales (L. Hall et al., 2016).

Given these conflicting results on reliability of visual scales, more work is needed to ensure that very young children can reliably complete QOL measures. Additionally, if the wording of scale items cannot be easily understood by typically developing children, it can be assumed to be even more daunting or complex for children who have experienced developmental delay, and/or have additional needs for communication support. For these children, it is fundamental to include accessibility and administration support into an instrument's development and use.

2.2.2 Principles of measurement development and evaluation of measures

Beyond the challenges specific to measuring QOL in children with disabilities, there are overarching principles to the development of QOL tools. These principles apply first to the process of producing a new QOL measure, and secondly to evaluating the properties of the measure—primarily by validation of its psychometric properties. The first part of this section discusses the main points to be considered in developing a new QOL tool, including basing the measure on a conceptual model, and ensuring items and domains reflect the perspectives of its anticipated respondents. The second part of this section discusses how to evaluate the measurement properties of developed QOL tools, based on published guidelines (Food and Drug Administration, 2009; Mokkink, Terwee, Patrick, et al., 2010; Reeve et al., 2013).

2.2.2.1 Measure development

Prior to evaluation of a completed instrument, development of an appropriate QOL tool should involve qualitative research with intended respondents to generate content. Historically, literature on PRO development has prioritised psychometric validation rather than principles of qualitative content development (Lasch et al., 2010). However, in 2009 the Food and Drug Administration (FDA) in the US published guidelines on development and evaluation of PROs for use in health research, advocating for measures based on a conceptual framework incorporating qualitative research with the intended respondents (Food and Drug Administration, 2009). Since then, there has been an increase in literature establishing guidelines for this work to meet the FDA's recommendations.

The first purpose of qualitative research in development of a PRO is to identify and clearly define key areas and characteristics of the measurement construct, and to formulate corresponding items that exemplify them (Lasch et al., 2010). For example, initial qualitative research may identify domains of QOL relevant to a specific population. Domain definitions may include direct quotes from participants, which can also form items on the final instrument. Many PRO instruments are developed from qualitative research involving both anticipated instrument respondents, such as patient groups, and expert opinion from researchers or clinicians working with the patient groups. Expert perspectives may identify main areas of interest in a clinical measure, with key concerns in these areas provided by further research with patients. Similarly, reviewing existing literature may provide guidance on issues likely to be of concern to patients. However, as the key component of PROs is the perspective of respondents themselves, they should be included at some stage of initial content development to ensure content validity (Brod et al., 2009; Food and Drug Administration, 2009). This is particularly imperative for those who have historically been excluded from research, such as children and people with disabilities (Mertens et al., 2011).

As well as establishing the necessity of including qualitative research involving PRO respondents, recent guidelines on PRO development have discussed the necessity for rigour and validity in the qualitative methodology itself (Brod et al., 2009; Lasch et al., 2010). There are many types of qualitative research and each includes specific techniques for ensuring methodological rigour. For PRO development, interviews with anticipated respondents, analysed with a grounded theory method, provide both a way to include user perspective, and develop a conceptual model of the contrast for the PRO (Brod et al., 2009; Lasch et al., 2010). The specific techniques of grounded theory methods, in general and in relation to methodological rigour, are discussed in detail in Chapter 4: Methodology, Section 4.1, page 75.

Once initial domains and items reflecting them have been drafted, the second stage of developing content for PROs involves checking the validity of the drafted tool with the anticipated respondents, both for overall conceptual content and the interpretations of individual items. A common method of confirming content validity is “cognitive

interviewing”, in which anticipated respondents are asked to verbalise their thoughts as they complete the items of the PRO instrument (Food and Drug Administration, 2009). By confirming content in this way, researchers can be confident that their interpretations of initial work are similarly interpreted by respondents, and that language used in the items is acceptable to the tool’s intended audience. However, without early open-ended qualitative research, the second stage may not identify gaps in domains of a measure, or that the underlying conceptual model is incomplete. That is, both stages are important to establish the validity of the conceptual model and content of a PRO.

2.2.2.2 Evaluating measurement properties of QOL tools

The final step in development of a PRO measure is to evaluate its measurement properties. Measurement properties, or psychometrics, can be broadly separated into three categories: validity, reliability, and responsiveness (Food and Drug Administration, 2009). Reliability is the extent to which scale scores are free from error, can be reproduced over time, and are rated similarly for similar items. Validity is the extent to which a scale is acceptable to respondents, and measures what it is supposed to relative to the proposed measurement model and hypothesised population differences. Responsiveness—also known as sensitivity to change—is the extent to which a scale can detect changes in the concept it is measuring, and what changes in scale scores mean clinically; it is often considered a component of validity (Reeve et al., 2013). However, while a scale may be valid but unreliable, or reliable but lacking validity, responsiveness relies on both validity and reliability.

There are well-established procedures for establishing validity and reliability of scales through quantitative testing. Guidelines have been published on how to evaluate the quality of psychometric validation of PROs, and whether they met adequate methodological standards. Three of these are especially relevant to QOL tool development. The first is the COSMIN tool (Mokkink, Terwee, Patrick, et al., 2010), mentioned in Section 2.1.7.2 (see page 9), developed for evaluating the measurement properties of health measurement tools, and the quality of studies reporting on these tools. In addition, the International Society for the Study of QOL (ISOQOL) has

developed guidelines for evaluating measures of QOL (Reeve et al., 2013), based on the expert consensus of their members—largely researchers and clinicians working in the field of QOL measure development and use. Lastly, the FDA’s PRO guidelines (mentioned in the previous section, see page 37) were developed by the FDA for evaluating the use of PROs to demonstrate outcomes in clinical trials.

COSMIN includes twelve evaluation criteria: nine of the criteria relate to psychometric properties, one to overall interpretability, and the final two criteria evaluate the quality of the scale’s validation study. While COSMIN is intended to form a basis of choosing a PRO in practice, the checklist focuses almost entirely on measurement properties. The minimum standards reported by ISOQOL (Reeve et al., 2013) cover the same psychometric properties as those included in the COSMIN checklist, but have additional categories aimed at implementing the use of the instruments in practice. These include features of interpretability such as reporting average scores for specific population parameters, and considerations of administration burden. The FDA guidelines include the same considerations as those of COSMIN and ISOQOL, with the addition of greater detail on instrument development and content validity, and the use of the instrument in specific populations, and within clinical trials (Food and Drug Administration, 2009).

All the aspects of measurement evaluation included in these three guidelines are relevant to evaluating existing condition-specific measures of QOL for children with CP. While most aspects of development and measurement properties have been easily established by existing measures, construct validity and sensitivity to change are worth examining in greater detail.

Construct validity is established by showing that a measure is correlated with measures of the same or similar constructs, and uncorrelated with unrelated constructs (Mokkink, Terwee, Patrick, et al., 2010). This can be demonstrated by a QOL measure which produces different scores for populations hypothesised to have different levels of QOL. In condition-specific measure of QOL for children with CP, construct validity is often established through hypothesised differences in QOL scores across GMFCS levels, with higher QOL expected in those with lower overall impairment

(e.g. Varni et al., 2006). However, this conflates functioning with subjective wellbeing. While it might be anticipated that children at higher GMFCS levels would score lower in a physical domain of QOL, research in the broader disability community has repeatedly shown that the presence or severity of disability does not automatically confer low QOL (Albrecht & Devlieger, 1999). If a measure of QOL consistently demonstrates lower QOL in children with more severe impairments, it may more accurately reflect functioning than global wellbeing by presupposing that those with disabilities cannot achieve high QOL. This further reinforces the need for QOL measures to be based on clearly defined conceptual models, with appropriate methods for content development that reflect a multidimensional definition of QOL such as that of the WHO (WHOQoL Group, 1995).

Establishing sensitivity to change has proved challenging in QOL measurement (Drotar, 2004). To establish that a measure is sensitive to change it is necessary to establish first how and where the measured construct will change, which relies on both longitudinal data and knowledge of change on the underlying construct. Many clinical measures establish responsiveness through comparison to changes on an established gold-standard instrument—for example, a measure of a construct known to be correlated with the construct of interest or using a generic measure of the same construct to develop a more sensitive measure for a specific population. For most QOL instruments there are three barriers to establishing responsiveness: limited longitudinal data, no established gold-standard instruments, and theoretical confusion over when and how QOL can be expected to change. Where there is no established gold-standard instrument, responsiveness can be established through comparison to changes in observed clinical outcomes known to impact the measurement construct (Revicki et al., 2008).

For children with CP, there is still limited knowledge on how or when QOL can be expected to change as a result of therapeutic interventions, or how observed clinical changes may relate to QOL changes (Tsoi et al., 2012). In general QOL theory, the influential homeostatic model (Cummins, 2000) argues that QOL resists change in most people, returning to an individually determined setpoint. Sensitivity to change can be considered an extension of construct validity—and if a measure reliably reports

differences between groups expected to have different QOL, when QOL changes, the measure should capture this for all respondents (Revicki et al., 2008). Therefore, where measures have demonstrated reliability and construct validity, a lack of change in QOL scores may reflect homeostatic resistance to change in QOL rather than limitations to measurement sensitivity.

Once sensitivity to change is established, it is essential to establish a measure's minimal clinically important difference (MCID) if it is to be useful in clinical trials and practice to measure the change in an individual's QOL over time. The MCID is the minimum difference in a scale score which translates to a meaningful difference in the outcome in a clinical context—i.e. the smallest difference on the measure that could be felt as a subjective difference by the patient—rather than group differences that may be demonstrated in establishing sensitivity to change (Revicki et al., 2008). The MCID may vary depending on context and the responder and relies on additional information ideally collected through multiple methods. None of the existing condition-specific measures of QOL for children with CP have yet established a MCID. The following section provides an overview of these tools and evaluates their strengths and limitations in development methods and measurement properties.

2.2.3 Condition-specific measures of QOL for children with CP

While comparing the QOL of individuals across health conditions can be important at a population level, this often has little relevance in evaluating interventions and clinical practice. For the purposes of determining patient priorities, and measuring progress in areas targeted by interventions, condition-specific QOL instruments are the most appropriate.

Condition-specific instruments are designed to provide greater detail of the most significant aspects unique to each condition, in addition to evaluating overall QOL. For the paediatric CP population, there are four condition-specific instruments measuring QOL currently available: a CP-specific module for a generic instrument, PedsQL-CP (Varni et al., 2006), a CP-specific module for a chronic-generic instrument, DISABKIDS-CPM (Baars et al., 2005), and two stand-alone CP-specific instruments: CPCHILD (Narayanan et al., 2007), and CP QOL-Child (Waters et al., 2006). All four vary in their

development, domains and items. Additionally, there are published reviews of these and other chronic-generic tools for the paediatric population, examining their content, theoretical basis, psychometric properties, adherence to user rights, and clinical usefulness (Carlon et al., 2010; E. Davis et al., 2006, 2018; Schiariti et al., 2011). Each of these reviews note limitations in the existing tools.

Furthermore, it is now at least ten years since any of these measures were first developed. During this time there have been significant changes in technology which may change the nature of children's participation or therapeutic interventions and aids, including the rise of social media, development of smart phones and tablets, and exponential increase in use of apps for these devices. Impacts of these may not be captured by the current measures but would be relevant to both families and health services.

2.2.3.1 Evaluating existing QOL measures

Accepting the WHO definition of QOL, there are several considerations for evaluating the validity of QOL measures. Measures should have positively-framed items that focus on wellbeing rather than illbeing, domains should include more than just functioning and health, and given the subjective nature of QOL, ideally the measure should include both a self-report and proxy version, with development including input from anticipated respondents (Waters et al., 2009). It is also necessary to note how domains for the measure are derived, and whether they are based on a defined conceptual model. As well as these QOL specific considerations, outcome measures should provide quantitative evidence of psychometric reliability and validity for the intended population, as recommended by guidelines on evaluating PRO measurement properties (Food and Drug Administration, 2009; Mokkink, Terwee, Patrick, et al., 2010; Reeve et al., 2013).

Outside of research settings, brevity and ease of administration are fundamental, as most health practitioners have limited contact time with patients. In research and clinical use, it is essential that a measure to be used as an ongoing evaluation tool is sensitive to change in the outcome being examined, and ideally information on what constitutes a clinically meaningful change will be available. Lastly, cost and availability

of the instrument can present barriers to use in both research and health service contexts if it is distributed commercially.

A summary of these properties for the four condition-specific measures of QOL for children with CP is included in Table 1 below, with further detail provided in the following sections. All four measures have been used in published research and in practice. However, none of the four measures adequately meets all criteria for all children with CP, despite varying strengths and limitations of each measure. Notably, none of the measures have established a MCID, though CPCHILD has provided some indication of change scores that have subsequently been used in trial interventions (A. M. Murphy et al., 2008).

Table 1. Properties of condition-specific QOL measures for children with CP

		PedsQL-CP ¹	DISABKIDS-CPM ²	CPCHILD ³	CP-QOL Child ⁴
Measurement Properties	Clear conceptual basis?	Based on WHO definition of health	Yes	Yes—but not solely QOL	Yes, from qualitative work with families
	Multidimensional definition of QOL?	Core yes, CP module only includes health and functioning	Yes	Primarily function, 5 items on wellbeing, 14 on pain and discomfort	Yes
	Wellbeing focus?	No, all items modules focus only on difficulties	No, all items focus only on difficulties	No, only 2 items have positive response option	Yes
	Respondents included in development?	No	Yes	Caregivers included, but not children	Yes
	Self-report version?	Yes, for ages 5-18 years	Yes, for ages 8-18 years	Yes, but it is unvalidated	Yes, for ages 9-12 years
	Reliability and validity?	Internal consistency reliability, construct validity based on intercorrelations with core scales ¹	Internal consistency reliability, structural validity from factor analysis and item correlations ²	Test-retest reliability, construct validity based on expected differences across GMFCS levels ³	Test-retest and internal consistency reliability, construct validity based on correlations with generic instruments ⁴
Administration properties	Length	Core module 23 items; CP module 35 items	Core module 37 or 12 items; CP module 16 items	76 items	66 items on proxy report, 52 items on self-report
	Time to complete ⁵	5-10 minutes	20 minutes	20-30 minutes	15-25 minutes
	Sensitive to change?	Yes, limited validation	Not demonstrated	Yes, limited validation	Not demonstrated
	Meaningful change defined?	No	No	Estimates based on standard deviation, not formally published	No
		Free for unfunded academic research; funded academic research 990USD; other uses	€60-70 for unfunded academic research; other uses (e.g. clinical) varies by sample size up to €5000		
	Cost?	(e.g. clinical) >2300USD		Free	Free

¹Varni et al., 2006; Varni, Burwinkle, & Seid, 2005; ²Baars et al., 2005; ³Narayanan et al., 2007; ⁴Waters et al., 2006; ⁵Carlson et al., 2010

2.2.3.1.1 PedsQL-CP

The PedsQL has a core generic scale, the PedsQL 4.0 Generic Core Scales, but includes additional modules developed for children with specific conditions, including the PedsQL 3.0 CP Module for children with CP (Varni et al., 2006). The original PedsQL was conceptualised to be a measure of health-related QOL, and the CP module has been conceptualised in the same way. Therefore, while the items match the multidimensional aspect of QOL definitions, they focus on functioning rather than wellbeing. The PedsQL 4.0 Core Scales have 23 items (Varni et al., 2001), with a short form of 15 items (Chan et al., 2005), both of which require payment for use. The parent-proxy items of the CP module have been published in the appendix of its validation publication (Varni et al., 2006). All the modules of the PedsQL are designed to be applicable to children aged 2-18 years, however the self-report versions are limited to those aged 5-18 years.

The construct validity of the PedsQL 4.0 Core Scales was demonstrated by hypothesis testing with differences in scores for typically developing children and children with CP—i.e. children with CP showed lower QOL, and QOL decreased with increasing severity of CP (Varni et al., 2006). Construct validity of the CP module was then established through intercorrelations between subscales of the CP module and the Core Scales. While this is consistent with the aim of the scale, it suggests the items may have a greater focus on functioning than overall QOL, which was shown by other researchers to be unrelated to CP diagnosis (Dickinson et al., 2007). The items of the PedsQL themselves, for both the core and CP scales, focus on ‘difficulties’ in functioning—meaning that children and their parents can at best report an absence of difficulties, rather than positives aspects of their lives. This is inconsistent with accepted definitions of QOL (WHOQoL Group, 1995).

The core scales of the PedsQL have been shown to be sensitive to change (Varni et al., 2002) in a limited clinical sample of children with fractures. Additional work has been published validating responsiveness for ambulatory children with CP (Oeffinger et al., 2008). However, Janssens et al. (2016) criticised the methodology of the validation research when measured against the COSMIN standards required to demonstrate

responsiveness. Therefore, although the initial validation is promising for the PedsQL CP module, additional work is needed to better validate its responsiveness. Furthermore, the validation work does not address PedsQL's heavy focus on functioning rather than QOL.

2.2.3.1.2 DISABKIDS-CPM

The DISABKIDS resulted from a large multi-national expert group in Europe, focused on developing a series of QOL instruments that would be applicable across countries. In order to make comparisons between different population groups, they developed a generic instrument, the KIDSCREEN (a 27 item version, Ravens-Sieberer et al., 2007, a 10 item version, 2010; a 52 item version, Ravens-Sieberer et al., 2008), a chronic generic instrument, the core module of the DISABKIDS (Bullinger et al., 2002), and seven condition-specific modules for different health conditions, including one for CP (Baars et al., 2005), to be used in addition to DISABKIDS core module. Lastly, there is a short version for younger children aged 4 to 7 years, or those over 7 years who did not have reading comprehension for the main questionnaires, that includes a visual response scale (Chaplin et al., 2008). Each instrument was developed following a similar method, with a QOL model derived by expert consensus, followed by a series of focus groups with children from relevant populations to inform item generation (Baars et al., 2005; Herdman et al., 2007; Simeoni et al., 2007).

The psychometric properties of the DISABKIDS Chronic Generic Module (37 items) were demonstrated in a rigorous procedure of pilot and field testing (Petersen et al., 2005). Content was validated through pilot testing while field testing demonstrated satisfactory test-retest and internal consistency reliability, as well as good discriminant validity based on health status and correlations with other generic instruments (Simeoni et al., 2007). DISABKIDS was also tested cross culturally across 7 European countries (Schmidt et al., 2006).

The DISABKIDS CP Module followed a procedure of pilot testing and development consistent with the other DISABKIDS instruments, showing good reliability of items within scale domains (Baars et al., 2005). The authors note that the sample size for the CP group was very small (n=21 in pilot test, n=43 in field test)—a sample of this size

would not meet COSMIN standards to show good reliability and validity (Mokkink, Terwee, Knol, et al., 2010). A more recent study with a larger sample replicated the reliability of the domains and showed acceptable discriminant validity across health status when compared with other scores on other generic QOL instruments (Mueller-Godeffroy et al., 2016). However, in the sample tested by Mueller et al. (2016) just over 50% of the participants achieved the maximum possible score on one of the two domains of the CP Module, known as a ceiling effect. Such a large proportion of the sample scoring so highly suggests that the CP Module does not have the capacity to measure high QOL and may not be applicable to all children with CP.

Furthermore, the DISABKIDS-CPM does not measure well-being; like PedsQL, it focuses on ill-being. The items concentrate primarily on 'difficulties' and impairments, with generally negatively framed items meaning the best a child can report is an absence of difficulty. This not only restricts children's ability to report good QOL but may represent a threat to self-esteem for both children and their caregivers.

There are other limitations to the use of both the core DISABKIDS and the CP module. The original core scale is relatively long, with 37 items, estimated to take 20 minutes to complete (Carlon et al., 2010). The CP module is an additional 16 items. Another bar to its use is the cost involved. While unfunded academic research may use the measure for less than €100, all other uses including standard practice incur significantly higher fees to a maximum of €5000. Lastly, as with the other measures, the lack of information on sensitivity to change, or MCID, limit interpretation of scores in both research and practice contexts.

2.2.3.1.3 CPCHILD

The Caregiver Priorities & Child Health Index of Life with Disabilities, or CPCHILD, is a Canadian instrument developed to consider HRQOL of children with CP in combination with the impact of disability care on the family. The CPCHILD is of limited use to a wide population of children with CP, as it was developed specifically for non-ambulatory children with severe mobility limitations (i.e. GMFCS levels IV and V) and has been validated only for this group (Narayanan et al., 2007). It would be inappropriate to use

this measure for children with CP who have minor impairments, however its content may be more applicable to those with severe CP than other instruments.

Initial validation of the CPCHILD included test-retest reliability and construct validity by hypothesis testing of poorer functioning at higher GMFCS levels (Narayanan et al., 2007). Both were shown to be acceptable. The CPCHILD has since been used in multiple clinical trials for various interventions for children with CP. Although the initial validation did not include evaluation of sensitivity to change, subsequent research by the authors has demonstrated changes as a result of surgical intervention (Narayanan et al., 2011). Other researchers have used an estimate of standard deviation of change as an indicator of the size of meaningful change (A. M. Murphy et al., 2008), though, this has not been formally published by the CPCHILD authors.

Although the CPCHILD overall focuses on functioning and measures illbeing rather than wellbeing, one significant section of the instrument (36 items) asks respondents to rate how important these aspects are to their overall QOL (Narayanan et al., 2007).

Therefore, while a child may show significant difficulties in activities of daily living in other sections of the questionnaire, they are also able to nominate that these difficulties have little impact on their overall QOL. These ratings were used during the testing of the instrument to confirm face validity for caregivers of children with severe CP. This aspect of personal subjectivity is not captured in any of the other CP-specific instruments.

2.2.3.1.4 CP QOL-Child

CP QOL-Child was developed through interviews with children with CP and their parents, using a grounded theory approach allowing themes relevant to participants to emerge (Waters et al., 2005). Themes identified in qualitative analysis were then compared to an accepted definition of QOL and content of comparable scales to develop a conceptual model. The resulting questionnaire includes versions for both child self-report and parent proxy and has been shown to have good measurement properties of reliability and psychometric validity (Waters et al., 2006). Items are positively framed and cover a range of domains beyond functioning and health. Although items use a simple format and clear response scale, item wording is identical

in child report and parent report versions—it may be possible to improve the use of language appropriate for children’s developmental level.

The CP QOL-Child is freely available and does not require training to administer or interpret. However, it is very long, taking approximately 15-25 minutes to complete (Carlson et al., 2010). It was not originally developed specifically for clinical use, and given it measures a range of areas of life nominated by families (i.e. family health, finances, access to services), some domains may not be as useful for clinicians. Lastly, CP QOL-Child has not been evaluated for sensitivity to change, which is a bar to its validity in ongoing evaluation.

A group of researchers in the UK used qualitative interviews to identify concepts of QOL nominated by children with disabilities and their parents, and compared these to domains included in the current CP QOL-Child (Parkinson et al., 2011). The instrument did not address all the concerns nominated by children and parents. Notably, children nominated areas of family connections, relaxation, tiredness, and negative emotions that were not addressed by the existing CP QOL-Child. The authors suggest these differences may relate to the different cultural setting, however an update of the CP QOL-Child including qualitative analysis may help explain how these fit into the QOL concept for children in Australia.

2.2.4 Use of existing condition-specific QOL measures for children with CP

As noted previously (see Section 2.1.7.1), current literature suggests that QOL outcomes are very rarely used in paediatric practice (O’Connor et al., 2016). It is likely that the limited clinical utility of current tools is one barrier to increased use. On the other hand, existing QOL tools for children with CP have been adopted in two other areas of research: as population measures, and as secondary outcomes for clinical trials. Each of these uses presents unique challenges for measurement, and provides implications for what constitutes QOL for children with CP. These are described in more detail below.

2.2.4.1 Population comparisons

It is fundamental to recall that disability does not automatically confer a low QOL—it is the subjective experience of multiple domains of life, not the replication of functional health status. This is evidenced by research at the population level using generic measures to compare QOL of children with CP with typically developing children in the general population. The most widely cited example of this is the SPARCLE studies, a large, multi-centre European project examining the QOL of children with CP using the KIDSCREEN, a generic measure of QOL developed by the same methods and researchers as DISABKIDS (Dickinson et al., 2007). This research was based on a sample of children aged 8-12 years who were able to self-report. The highlighted findings of this paper were that level of impairment did not correspond directly with overall QOL, though there were some specific relationships (such as communication impairments related to poorer family relationships). Furthermore, schooling was the only domain where children from the general population differed from those with CP; children with CP had higher QOL related to schooling. This may relate to greater support offered for children in special schooling compared to children without disabilities in mainstream schools. However, the authors reported the difference as equivocal: sensitivity analysis showed it may have been related to high non-response in one region (Dickinson et al., 2007). Nonetheless, given this research was informed by a generic measure, it is possible that the issues most relevant to QOL for children with CP were not well examined. One basic conclusion to be drawn from this work is that the QOL of children with CP can vary as greatly as among any other population group; the diagnosis of CP alone cannot determine the needs of individuals in improving their QOL.

2.2.4.2 QOL outcomes in clinical trials and clinical practice

There has been a rise in the inclusion of QOL and related measurement in research-based evaluations of interventions, especially in randomised controlled trials. There is now a growing body of literature reporting on short-term QOL outcomes for specific interventions, which inform evidence-based practice. However, for many of these, even where there is good evidence of improvement in primary clinical measures, there has often been no measurable change in overall QOL.

In 2012, Tsoi, Zhang, Wang, Tsang, and Lo published the first systematic review of QOL outcomes in clinical trials for children with CP, based on literature published up to and including 2010. Of the eight trials included in the review, the results were somewhat equivocal: while some demonstrated improvements in specific domains of QOL (especially the physical domain), there was little change in overall QOL in any of those included. The authors noted several limitations in QOL evaluation in intervention trials. Many include outcomes purporting to represent QOL, but without satisfying the conceptual definition such as that of the WHO (WHOQoL Group, 1995). Moreover, even for researchers who chose appropriate QOL outcome measures, none of the CP-specific paediatric measures of QOL have established both sensitivity to change and MCID. Both are necessary for evaluation of QOL outcomes in clinical trials.

Since 2010, there have been at least twenty additional paediatric CP trials including a QOL outcome published (such as Engsberg, Ross, & Collins, 2006; Gates, P.E et al., 2012; Hoving et al., 2009; Van Wely, Balemans, Becher, & Dallmeijer, 2014). These have evaluated interventions and therapies from a variety of disciplines, with diverse functional and activity goals. Similar to those included in the review by Tsoi et al. (2012), they show some evidence of change in specific QOL domains, and more limited evidence that overall QOL can change. However, these changes are inconsistent, and there is little understanding of how to systematically effect changes in QOL from the available interventions.

There are two major challenges to assessing evidence of QOL outcomes in these trials. Firstly, even where appropriate QOL instruments have been used, without validation of their sensitivity to change, it is not possible to know whether lack of change is the result of measurement error (i.e. poor reliability) or if there is no real change in QOL. Secondly, the majority of interventions included in Tsoi et al. (2012) and elsewhere do not explicitly target improvement in QOL, but usually have more immediate functional goals. How intervention aims impact on QOL outcomes, or how functional goals relate to QOL outcomes, is generally not made explicit.

Clearer conceptual understanding of QOL and how it may relate to therapeutic practice is therefore a necessary step to improving QOL for children with CP. This

conceptual basis can then be used for appropriate development or revision of CP specific QOL tools. Once tools are validated with information on sensitivity to change and minimal important clinical difference, they can be implemented and accurately interpreted in research trials and practice for the improvement of children's lives.

2.3 Summary

CP is a varied condition, characterised by motor impairments but often accompanied by wide-ranging comorbidities. Impacts of CP are felt lifelong on health, social, educational, and vocational outcomes, through direct effects of the condition and environmental barriers to participation and inclusion. The costs of care for children with CP are high for society but are particularly felt by families. Parents and other family members are usually responsible for a significant amount of the health care and medical needs of children with CP. From the stress of diagnosis, to the increased care needs for a child with a disability, to experiences of disability stigma and exclusion, parents of children with a disability often experience substantial stress on their physical and mental health, relationships, and finances. These effects on the family have been related to poorer outcomes for their children.

While CP is currently incurable, some comorbid conditions can be successfully treated and there are many interventions which focus on improving functioning and reducing pain and discomfort. Multiple organisations have now published clinical guidelines for diagnosis and treatment of CP and related conditions, based on peer-reviewed evidence of efficacy. In recent years, interventions have moved from focusing on promoting 'normal' movement to promoting increased function and participation, even if this means greater reliance on mobility aids. Changes to treatment goals have occurred in tandem with the rise in patient- and family-centred care, which emphasises the value of collaborating with patients and families in devising treatment goals.

Impacts of CP are diverse, as are existing health services and interventions. The changing landscape of disability service provision in Australia has led to an increased demand for evaluations of those services. There is currently limited use of outcome measures to evaluate health services in practice despite the emphasis of this in

evidence-based medicine. As an outcome measurement, QOL addresses both the holistic focus of modern treatment of CP, and the need to include patient concerns in treatment goals; QOL is defined as a person's subjective appraisal of their wellbeing across multiple domains of life.

There are challenges in developing QOL measures for children with CP. There is limited expert agreement on what constitutes a conceptual model of QOL for children with disabilities. Some existing measures continue to focus on health status or level of function, despite widely accepted definitions of QOL encompassing other domains of life. For young children, or those with communication impairments, measures are often restricted to proxy report by parents, carers, or clinicians—either because no self-report measure has been provided, or because the self-report is not accessible for those using alternate communication methods. However, research has shown proxy report often has poor correlation with the preferred self-report.

There are four measures of QOL specifically for children with CP available. Unfortunately, they have some shortcomings, including limited conceptual grounding and de-emphasis of child voice. Where QOL measures have been used in RCTs for CP interventions, there is largely no difference in QOL as a result of interventions. This may be related to lack of specificity in measurement affecting sensitivity or a lack of understanding of factors of QOL for children with CP. Improved measurement both of QOL in clinical practice and of conceptual models of QOL is needed to further the understanding of how QOL can be improved, or how current interventions already target QOL.

It is likely that QOL for children with CP includes both internal personal factors that are highly resistant to change, including those inherent to CP (e.g. pain) and other non-CP factors such as personality, contextual factors common to all children (e.g. family functioning, psychopathology, health/illness), and the interaction between a child's CP and their environment (e.g. access to services, opportunities for participation). Therefore, as well as considering conceptual models of QOL for children with CP, it is necessary to review theories that situate a child with disability within a wider family and societal context. The following chapter reviews frameworks of disability

considering an individual within context, current conceptual models and theory of QOL, how this applies to disability, and how it applies to children.

Chapter 3: Conceptual frameworks

Definitions of QOL emphasise the internal subjective appraisal of external environmental factors across all areas of life. For children with CP, this means that QOL is determined by internal factors relevant to the child themselves including their CP, as well as their environment, and the interaction of their disability and the environment. As highlighted in Section 2.2 (see page 30), current measurement of QOL for children with CP has been limited in conceptual foundations and inclusion of child voice. This chapter therefore outlines conceptual frameworks that can inform development of QOL theory for children, including the basis for the inclusion of child participants in research, and existing literature on child perspectives of wellbeing.

Children with CP are both individuals with a disability, and children: this is relevant for the wider conceptual frameworks that can inform development of QOL theory. Many theories of disability have been developed from the perspective of adults and therefore do not recognise developmental changes and challenges for children. Meanwhile, disability has largely been missing from theories of child development, which are based on the developmental stages and considerations of typically developing children. However, a common aspect to both fields is the recognition of the interaction of person (child, in this case) and their environmental context. This reflects the central tenets of the socioecological model: consequently, this chapter opens with a brief overview of this model. Subsequent sections are organised around their focus firstly on the environment, and secondly on the individual. This includes examining the frameworks of social models of disability, family QOL, the WHO's International Classification of Functioning, Disability and Health (World Health Organisation, 2001), child developmental theories, and sociological approaches to childhood studies.

Finally, this chapter examines existing theoretical and conceptual models of QOL through the lens of the above frameworks. While existing QOL research provides useful background, this examination reveals that key gaps remain in incorporating child perspectives, family context, and wider disability frameworks for children with CP. Therefore, the conclusion summarises how these frameworks inform the aims of the thesis towards addressing these gaps.

3.1 The socioecological model

There are various ecological models used in public health research that promote research that situates an individual within their ecological system. More recent updates also recognise the importance of social relationships in these interactions and are known as socioecological models. The early childhood development model, proposed by the WHO's Commission on the Social Determinants of Health, is based on a substantial body of earlier research (Irwin et al., 2007). This model presents a non-hierarchical, multi-level framework emphasising the importance of environmental factors in shaping child development. Environmental factors include family, neighbourhood, social communities, and the wider policy and governmental environment. All of these are hypothesised to interact with each other, and all must be considered in promoting child health and wellbeing.

These levels of socioecological models are often replicated in domains of QOL proposed by individuals and families in qualitative research (such as child factors, family relationships, school relationships, and community factors such as access to services). An early model of children's QOL from the Nordic countries which grew from country development indices considered resources and conditions from multiple levels that closely resemble the socioecological model (Lindström & Eriksson, 1993). A few years later the widely accepted WHO definition of QOL was published, which itself considers a person's QOL as an interaction of their own subjective appraisals within their cultural context (WHOQoL Group, 1995).

3.2 The environmental context for children with disability

The first section on environmental context discusses the impact of the social model of disability on the field of disability research. This model redefined the meaning of 'disability' and sparked significant policy changes that affected the lives of people living with disability. While the original model has since been modified, critiqued, and updated, it continues to shape disability research. A newer version of the model, the social relational model of disability, is highlighted for its relevance to this thesis.

This section also explores implications of family and parent function on childhood outcomes. The family context is widely recognised for its impact on child development and may be especially relevant for children with physical or other impairments who have a greater reliance on others for their care. Family structure and function often changes due to the demands of caring for a child with a disability, as discussed in Section 2.1.5.4 in the previous chapter (see page 21). Parent reports on family functioning provide valuable information on the family level of socioecological model, as well as insight on dynamics between the child and their environment that children themselves may not be best positioned to see. The section concludes by exploring what this may mean for QOL.

3.2.1 Social models of disability

The social model of disability was proposed by disability activists in the United Kingdom in the 1970s and argues that, although people may have impairments, their disability is acquired as a result of social infrastructure which does not adjust to different abilities (Bricout et al., 2004). The application of the social model of disability shifts the onus of disability services from effecting change within individuals, such as in the individually focused medical model, to changing contextual factors which result in impaired individuals experiencing poorer outcomes across their lives. This framework informed a new area of social research known as disability studies, that is now the dominant field of social research for people with disabilities. The social model of disability has since informed the underlying principles of both the WHO's International Classification of Functioning, Disability, and Health (World Health Organisation, 2001), and the UN's Convention on the Rights of People with Disabilities (United Nations, 2006).

The original social model of disability remains relevant in the provision of health services for adults and children with disabilities, who still regularly face stigma and barriers to accessing necessary health and other services and to participating fully in community life. However, the social model has been criticised in its negation of direct distress experienced as a result of physical disability (Shakespeare & Watson, 1997). An example of this is chronic pain, which causes significant distress and disability for

individuals regardless of contextual factors. The social model of disability promotes the rights of individuals with a disability but may provide an incomplete understanding of the interaction between an individual and their context.

A more recent update to the social model of disability is Thomas' social relational model of disability (Thomas, 2004a). This model emphasises social relationships in creating and/or bridging disability in context, while recognising the real impacts of impairments and physical or other illness on people's ability to function in their lives, and their identity. Thomas also recognises that the distinction between impairments and disability may be disingenuous, as they exist as a continuum rather than as distinct states.

One drawback to the social model of disability is its failure to consider issues specific to children. Connors and Stalker (2007) discuss child-specific adaptations to the model, and what the social model may mean for children with disability. Their work with children highlighted not only the impact of children's impairments as separate from environmental barriers, but two additional, independent aspects of 'difference' and other people's behaviours. Firstly, Connors and Stalkers' analysis showed that many children with disability lacked language to discuss differences between themselves and typically developing children and formed their identities through comparisons to 'normal'. Secondly, they found that others' behaviours had mitigating or exacerbating impacts on children's experiences of their disability as creating difference and limitations. They concluded that the emphasis of Thomas' social relational understanding of disability may also translate better to children's experiences as much of children's development occurs through social learning and interactions with peers (Connors & Stalker, 2007).

Researchers have repeatedly shown that presence or severity of disability is unrelated to subjective experiences of multi-faceted QOL (e.g. for children with CP: Dickinson et al., 2007). However, due to different environmental contexts highlighted in the social model of disability, models of QOL for people with disabilities may be influenced by specific factors of less relevance to the general population. A model of QOL arising from research in the field of intellectual disability is that proposed by Schalock and

colleagues (Schalock et al., 2016). They propose eight domains of QOL, within three higher order factors (in brackets): emotional, material, and physical wellbeing (wellbeing), self-determination and personal development (independence), interpersonal relations, social inclusion and rights (social participation). This model is based on an ecological framework of disability, such that disability-specific QOL results from an interaction between personal and contextual factors in each of these domains. Specific factors that act as mediators and moderators between context and individual QOL are drawn from published literature, and lead to suggested enhancement strategies (further details of this literature are provided in Schalock et al., 2016). The enhancement strategies are recommended as possible therapeutic goals, with the authors advocating further research to determine whether the suggested strategies are predictive of changes in individual QOL.

However, there are two main limitations to the use of this model of QOL for children with CP. Firstly, the model does not distinguish between conceptual domains for adults and those for children, though the authors imply it is applicable to all people with intellectual disability. In reviewing QOL measurement models for children with CP, Waters et al. (2009) built on earlier work in paediatric oncology (Bradlyn et al., 1996) to argue that child QOL must encompass at least a child's own perception of their social, physical, and emotional well-being, and that any measure using these must be sensitive to changes in such domains across a child's development. Although social, physical and emotional wellbeing may be common domains between young children, adolescents, and adults, researchers have repeatedly shown differences in how children and adults perceive these, and even differences between child and adolescent experiences in these areas of life (both discussed in more detail in Section 2.2.1.3, see page 33). Secondly, the original domains proposed by Schalock et al. (2016) were based on literature review and expert opinion, rather than being raised by people with intellectual disability themselves. While this does not indicate that the domains are not relevant to this population, the subjective nature of QOL means that models should include input from those to whom they apply (Koot, 2014), as with recommended guidelines for PRO development (Food and Drug Administration, 2009).

The commonality for these and other theories of QOL is the consistent proposition that the level of QOL experienced by individuals is the subjective experience of internal personal factors, contextual factors, and their interactions—which parallels recent iterations of social models of disability. Using the Schalock model of QOL, Van Hecke et al. (2018) examined articles reviewing QOL measurement and conceptualisation, and came to the same conclusions: that QOL is the subjective experience of a multidimensional construct including both objective and subjective domains, influenced by a variety of individual and contextual factors.

3.2.2 Family context and family QOL

For all children, the socioecological model emphasises the family context—including parent perspectives and parent support. In fact, the value of support for parents is recognised in several articles of the UN’s Convention on the Rights of the Child (United Nations, 1989). Children with disabilities are already more reliant on their parents and carers than are other children—it may be harder for them to divorce themselves from this context if there are family or parenting stressors.

The examination of individual parent factors is outside the scope of this thesis, which focuses on child outcomes. However, these factors may not be entirely distinguishable from child QOL (see Section 2.2.1.3 in the previous chapter, page 33). A new area exploring these links is the recently developed concept of ‘family QOL’. Family QOL recognises the interplay between the individual QOL of family members, their interactions, and collective family functioning (Boelsma et al., 2017). It may be particularly relevant to children with CP given they usually have higher care needs compared to typically developing children. So far, research on family QOL in the field of childhood disability has focused mostly on the families of children with intellectual disability or autism spectrum disorder—two conditions often involving high levels of behavioural difficulties which have a high impact on family functioning.

One drawback of the family QOL model in disability research is that measures have focused mainly on negative impacts of disability on the family, as in the PedsQL Family Impact Module (Varni et al., 2004). That is, the theory of the family QOL model is that family functioning impacts individual QOL, and vice versa, however the scales and

measurement of it are usually based on the assumption that having a child with a disability is a burden in various ways and exposes other family members to additional difficulties in their lives, including disability-related discrimination; therefore, the whole family is stagnated in achieving their goals. Models that assume that a child's disability is necessarily only a burden on the whole family and on other individuals within that family are not just prejudicial but conflate the notion that wellbeing is only the absence of illbeing. That is, in this model, a family can at best report that a child's disability does not inconvenience them. On the other hand, a family QOL model that emphasises how well a family is able to achieve family goals (Brown et al., 2006) may more accurately reflect the multidimensionality of family life in the presence of a child with a disability, and provide interactional context for an individual child's QOL.

3.3 Individual factors of health and QOL for children with disabilities

Definitions of disability are composed of impairment (individual) and disability (social environment that limits participation, access, stigma). This is clearly delineated in the WHO's International Classification of Functioning, Disability and Health (ICF; World Health Organisation, 2001), which provides a link between environmental concerns of disability for children, and child-specific concerns for children with disability. The first part of this section provides a description of the ICF and its application to QOL. The ICF has been, and still is, an influential document in disability research. It marked a departure from the previous WHO disability classification by recognising the social model of disability. There is also a child and youth specific version that has been adopted by QOL researchers.

Beyond disability specific literature, there are two major approaches to researching children and childhood relevant to individual factors in theorising children's QOL. Crucially, both emphasise the qualitative differences between childhood and adulthood in terms of perspectives, needs, and priorities for wellbeing and health. Theories of developmental psychology emphasise individual learning leading to developmental milestones, against a background of biological change leading to adulthood. On the other hand, childhood studies are informed by a sociological

approach, focusing on experiential knowledge from the perspective of children and child voice as distinct from adults theorising on childhood.

3.3.1 International Classification of Functioning, Disability and Health (ICF)

The International Classification of Functioning, Disability and Health, introduced in 2001, updated the WHO's earlier models of disability, emphasising a distinction between bodily structures (with or without impairments), and functioning. Both the adult (World Health Organisation, 2001) and child and youth (World Health Organisation, 2007) versions of the ICF provide a framework for classifying health and functioning across multiple domains of life for any individual experiencing any health state, including disability. Following the socioecological model, the ICF situates an individual within their particular social context and links individual outcomes to the interaction between an individual person and wider social and structural dynamics. The ICF highlights that relationships between an individual, their context, their physical condition, and their participation in activities are bidirectional. The ICF provides a comprehensive checklist of factors assessing health status, outcomes and determinants. The child and youth version, ICF-CY, goes further in including developmentally specific environmental factors, such as educational settings.

Since its introduction, the ICF has been rapidly adopted, modified, enhanced, and also criticised within disability research. Some disability researchers maintain that despite its update from previous models of disability and functioning, the ICF still preferences the medical model and de-emphasises the importance of context (Heerkens et al., 2017). While the ICF is informed by the socioecological model, the ICF checklist and subsequent research exploring the ICF model have focused primarily on personal factors such as individual impairments.

The proposed links between structure and function in the ICF have received some empirical support in the paediatric CP literature. For example, Wright and colleagues followed up children for 6 months after botulinum toxin injection, a well-known and validated treatment for reducing spasticity (Wright et al., 2008). While there was improvement in direct functional outcomes, activity/participation outcomes were

more strongly related to baseline predictors than to functional changes. This preliminary evidence relevant to the relationships outlined in the ICF model indicates there is a child-context interaction, but shows that these relationships are complex. Similar studies showing promising validation of the ICF model lend further support to the increasing recognition of the importance of measuring outcomes and determinants beyond simple functional status (Majnemer & Mazer, 2004).

The links between the ICF model and QOL have been recognised in a number of ways: as a proposed addition to the model (McDougall et al., 2010), as a framework for evaluating the content of QOL measures (Schariti et al., 2011), and as a method of identifying factors relating to QOL for youth with chronic conditions (McDougall et al., 2014) and children with CP (Chen et al., 2014). The ICF models do not explicitly include QOL as an outcome but the checklist and related modelling can guide thinking on what may impact QOL in paediatric disability, and how to target QOL in practice.

3.3.2 Child developmental theories

Within social sciences, psychological theories of child development first explored the concepts of wellbeing for children, and how to best promote optimal development—recognising that early childhood experiences form the foundations for adult outcomes. There are now multiple well-known developmental theories from psychology research as to how humans develop across the lifespan, especially during childhood. To review each of these in depth is beyond the scope of this thesis, however there are commonalities to these theories that form a context for how researchers interpret childhood.

Developmental theories emphasise change across the lifespan, whether that is from middle to old age, or young children to teenagers. Psychological child developmental theories emphasise the achievement of goals—defined differently in each theory—broadly covering emotional, social, and intellectual spheres. By successfully navigating their developmental goals in the prescribed pattern, individuals pass from childhood to fully functioning adulthood.

While child developmental theories focus on the individual, they do also relate to context around the child: e.g. familial relationships were recognised as highly influential in infancy and child development in early psychological theories, most famously in Bowlby's attachment theory (Bretherton, 1985). More recent theories of child development continue to recognise the role of children's social networks as opportunities for learning and development, such as Bandura's influential social-cognitive learning theory (Bandura, 2001). Contemporary child development research emphasises that a child's development is reliant not only on the child's own intrinsic factors, but external environmental factors as well (Irwin et al., 2007).

Modern child educational theories arose from these frameworks, conceptualising the goal of education to provide an environment for children to acquire skills in both intellectual and social life (Martin et al., 2017). In this context, there are consequences for lifelong development if children are deprived by disability from 'normal' developmental situations and conditions. Therefore, for children with disabilities, the need to achieve developmental goals has led to concepts of primary and secondary deprivation, in which children miss not only primary participation activities, but the secondary opportunities for optimal development (Missiuna & Pollock, 1991).

Lastly, in the developmental model the primary purpose of childhood has been for children to develop into fully functioning adults. The corollary of this perspective is that children are either explicitly or tacitly framed as 'becoming' adults, rather than as whole in themselves (Uprichard, 2008). Historically, this perspective has de-emphasised the validity and value of children's perspectives on their lives. Children have often been excluded from actively contributing to research on the meaning and goals of childhood. With changing social mores over the 21st century, childhood has come to have increased significance, and children are recognised as distinctly different from adults, either in their goals, abilities, or perspectives (Priestly, 1998). Recognition of these differences informs much of sociological research in childhood, explored below.

3.3.3 Sociology of childhood and childhood studies

As cultural views of childhood have shifted, sociological theories of childhood have arisen that recognise children as ‘being’ in their own right, rather than ‘becoming’ adults (Uprichard, 2008). That is, a developmental perspective assumes that children are in the process of becoming adults, and views childhood through an adult lens. By contrast, sociological theories of childhood focus on child experience and recognise that children are able to communicate their own views of childhood (Priestly, 1998). The sociological approach to child research, known as childhood studies, further differentiates between adult views of childhood and childhood concepts, and the experience of childhood for children. For example, child definitions of ‘play’ may be conceptually different to adult views of children’s play (Glenn et al., 2013). Sociological researchers therefore emphasise the integration of children’s views into research about children, rather than preferencing adult perspectives. Childhood studies researchers have repeatedly called for the use of ‘child voice’ in research about children, and suggested that child research that does not provide children the opportunity to express their views is incomplete (Lewis, 2010). This approach echoes Article 12 of the Convention on the Rights of the Child: children’s right to express opinions on matters which concern them (United Nations, 1989). The meaning and implications of a rights-based research framework for children with disabilities is explored in greater detail in the following section (The rights of children with disabilities, page 67).

Within the sociological perspective, children may still be considered passive under adult authority, at risk and in need of protection, or as competent child citizens with personal agency, or somewhere in between (Harden et al., 2000). The position researchers take in regard to children’s competence and need for protection alters both the research agenda and the research methodology. By integrating ideas of children as both at risk and as having specific competencies of value to children’s research, researchers have developed strategies to engage children as participants and enable them to express their views in a research setting (Phelan & Kinsella, 2013). The use of child-specific strategies in qualitative research is described more fully in the

methodological chapter of this thesis (see Section 4.1.5 Qualitative interview strategies for engaging children, page 86).

Changing conceptions of childhood have paralleled changing views of disability in society. Recently, a new theoretical perspective has emerged that draws on the parallels between social models of disability and childhood studies to create what the authors title “disabled children’s childhood studies” (Curran & Runswick-Cole, 2014). Disabled children’s childhood studies not only recognise the distinct differences in meanings of ‘childhood’ for children and adults but call for the inclusion of this perspective in studies for children with disabilities. From this perspective, “it is important to see every child as a child first and with an impairment second” (Connors & Stalker, 2007, p. 23). This is informed by an earlier highly-cited publication by Priestly, who stated even more starkly “to view disabled children as ‘disabled first’ is to deny them their rights as children” (1998, p. 210).

Children with disabilities therefore have the same rights as all children for inclusion, access, education, and recreation in the community. Furthermore, within research they have the same rights to voice their own opinions, regardless of impairment, and to participate in research regarding their childhoods as equally valid cultural experiences without reference to medical models of impairment that classify them as less than ‘normal’. Research must include methods to accommodate children with diverse ways of communicating and accept alternate definitions of child development; to do otherwise reinforces the social structures that disadvantage and exclude people with disabilities.

3.4 The rights of children with disabilities in research

There are two international conventions governing the rights of children with disabilities, both of which provide an imperative for researchers to include perspectives of children and people with disabilities in research about them and not just their caregivers. These two conventions are the 1989 UN Convention on the Rights of the Child (CRC, United Nations, 1989) and the 2006 UN Convention on the Rights of People with Disabilities (CRPD, United Nations, 2006). Australia is a signatory to and has formally ratified both Conventions (the CRC in 1990, and the CRPD in 2008) and,

under international law has an obligation to abide by their principles. Both conventions are useful in defining what is meant by 'childhood' and 'disability', and in guiding ways in which research can serve the best interests of children with disabilities. Each of these is described separately below, followed by a brief discussion on their meaning to research in combination.

The CRC defines children as any person up to the age of eighteen years and requires the provision of rights equally to all children, including those with disabilities, without discrimination. The convention preamble reiterates previous UN rights declarations in stating children's need for protection and particular care above and beyond that of adults (United Nations, 1989). The articles are concerned with providing for the best interests of the child in all situations, to provide for optimal development physically, socially, spiritually, and psychologically.

Two specific articles, Article 12 and 13, have been highly quoted in research concerned with children and children's services: both relate to children's right to participate in research about them. Article 12 requires provision:

to the child who is capable of forming his or her own views the right to express those views freely in all matters affecting the child, the views of the child being given due weight in accordance with the age and maturity of the child. (United Nations, 1989, p. 4)

Article 13 extends the circumstances of children's views by providing children:

the right to freedom of expression; this right shall include freedom to seek, receive and impart information and ideas of all kinds, regardless of frontiers, either orally, in writing or in print, in the form of art, or through any other media of the child's choice. (United Nations, 1989, p. 4)

These two Articles together argue strongly for the inclusion of child participants in all research about them and more broadly, research affecting their lives. In a seminal paper, Lundy (2007) examines the application of Article 12 particularly in education, and argues that this article specifically creates a legal imperative for greater integration of child views into decisions involving children. Lundy critiques many child-

related practices which have either failed to systematically incorporate children's views, or which have ostensibly included 'child voice' but have not met the requirements of Article 12 to give due weight to their opinions. Lundy proposes a model to implement Article 12 that includes four separate concepts: space, voice, influence, and audience. Space and voice combine to satisfy the right for children to express their views, while influence and audience satisfy the right for views to be given due weight in decisions.

Since the adoption of the CRC, child-focused researchers have delineated between 'research on' and 'research with' children to distinguish levels of child participation (McWhirter, 2014). That is, while both may address the same issues in children's lives, 'research on' children de-emphasises child voice and may exclude child participants or use methods that limit children's opportunity to express their views in the research. In contrast, 'research with' children fully recognises children's rights to be involved in research about them and encourages their participation in the research process.

The conditions of Article 13 support the recent development of child-centred approaches in research. The child-centred approach not only recognises children's right to participate, it uses methods designed to encourage the participation of children of different ages and stages of development through alternatives to traditional research methods. For example, using games, toys, or role-playing with very young children to elicit their views on inclusive education (Koller & San Juan, 2015). With the explicit statement in the CRC that these rights apply equally to all children, the right to freedom of expression has particular relevance to children with disabilities in research. To uphold Article 13 in research with children with disabilities, researchers must make provision for the participation of children with communication or other impairments who may be excluded from traditional research approaches. In this way, the CRC directly informs the use of accessible and inclusive research for children with disabilities.

The Convention on the Rights of People with Disabilities (CRPD; United Nations, 2006) was drafted more recently than the CRC and has marked a shift in focus of services and research for people with disabilities. It is the first UN Convention drafted with the

explicit inclusion of the group for whom it is focused (Mittler, 2015) and calls for the inclusion of people with disabilities, including children with disabilities, in all aspects of development of policy and legislation which have material impacts on their lives (United Nations, 2006, Article 4). The CRPD follows decades of activism by people with disabilities and disability advocacy groups, who adopted the phrase “nothing about us without us” to call for increased rights for people with disabilities not only in housing, employment, accessibility, and other areas, but in self-determination (e.g. Charlton, 1998).

The CRPD contrasts with the CRC’s approach to recognising limitations to capacity. The CRC includes provisos to children’s rights to express their views such as “in a manner consistent with evolving capacities” (United Nations, 1989, p. 2, Article 5) and “the child who is capable of forming his or her own views” (United Nations, 1989, p. 4, Article 12). Prior to the adoption of the CRPD, this allowed national legislation to use these clauses to define competencies of children with disabilities, and limit their rights to expression (J. Davis & Watson, 2000). In contrast, the CRPD rejects the notion that capacity or competency negates a person’s right to express their opinion.

Consequently, some researchers have discussed the implications of the wider scope of the CRPD for the rights for children under the CRC, and argued that research methods should attempt to seek views of all children with disabilities, regardless of abilities or competence tests (Sandland, 2017). This reading of the CRPD emphasises the need to provide opportunities for supported participation for children of all abilities, as for adults with disabilities, to ensure fair and equal participation. This is codified in Article 7 of the CRPD:

that children with disabilities have the right to express their views freely on all matters affecting them, their views being given due weight in accordance with their age and maturity, on an equal basis with other children, and to be provided with disability and age-appropriate assistance to realize that right.
(United Nations, 2006, Article 7)

Combining both the CRC and the CRPD challenges researchers who wish to grant children with disabilities their full rights to take additional steps to: a) ensure that

participation in research is fully accessible, regardless of capacity or ability; and b) grant children choice in their manner of participation.

3.5 Perspectives of wellbeing from children with disability

Existing models of QOL for adults can inform child conceptual models of QOL. The homeostatic model (Cummins, 2000) has been highly influential and defines links between individual subjective QOL and contextual conditions. Meanwhile in the field of intellectual disability research, Schalock and colleagues developed a detailed conceptual model of QOL predictors and influencers for individuals with intellectual disability (Schalock et al., 2016). However, there is limited literature on how these models may be adapted to children's developmentally different experiences. Therefore, lastly, this section explores qualitative work directly focused on the experiences of children with disabilities that has proposed measurement models and investigated children's perspectives of wellbeing. These overlap with QOL and offer insights into concepts of QOL for children with CP.

Along with the perspectives of children themselves, understanding of children's contexts, especially in the case of therapeutic goals for disability care, can be informed by research based on the views of experts and parents. A recently developed model of targets for clinical intervention for children with CP is the F-words paradigm (Rosenbaum & Gorter, 2012). There are six 'F-words': function, family, fitness, fun, friendships, and future. The first five of the F-words map onto the ICF framework, recognising the relationships between other areas of a child's life and their physical functioning. The F-words extends the framework by including 'future'—in recognition of the changing nature of child development, and the need for clinical interventions to address future ambitions and expectations as well as current concerns. Although not considered by its creators to reflect QOL, the six 'F-words' may parallel domains of conceptual models of QOL, which usually include similar factors. However, the F-words paradigm may be limited in application to children's perspectives, as it was based on literature review and clinical experience of the authors. Ultimately, the F-words are intended to focus interventions for children with CP on broader considerations than

striving for ‘normal’ functions—another aim of the use of QOL as a health-related outcome.

In research with an explicit focus on QOL as an outcome for services and interventions, Renwick et al. (2003) proposed a conceptual model of QOL for children with developmental disabilities using qualitative methods. Using a grounded theory analysis, this model highlighted changes across age groups for children from 3 years to 12 years, as well as links between parent and family QOL, the broader environmental context, and a child’s own QOL. However, the research was based only on interviews with parents, without children’s involvement. Therefore, it is hard to know if children would have a different perspective on the content of the model.

Based explicitly on the perspectives of young people with CP, a separate qualitative project used constructivist grounded theory to understand positive and negative influences on QOL for adolescents with CP (Shikako-Thomas et al., 2009). The researchers identified a model in which QOL was dependent on three factors: personal (“intrinsic aspects”), environmental (“extrinsic aspects”), and the interaction between personal and environmental factors. The strong links between individual factors and environmental factors supports a socioecological perspective of disability and QOL and align with the WHO definition of QOL. The authors highlighted that one major aspect of good adolescent QOL was a match between personal activity preferences and local availability of opportunities for participation. The model also included ability to manage challenges related to CP, as well as aspects of self-identity and others’ perceptions. These show a slightly different perspective to aspects of QOL than included in other models, which focus on function and social acceptance rather than personal feelings about overcoming challenges or sense of self. These aspects may correspond to the perspectives of children with CP, however the participants in this study were 12 to 16 years old. Increasing independence during teen years may change the focus of QOL factors compared to younger children’s views.

Researchers working with 8-12 year old children in the general population in Ireland used innovative qualitative methods not only to conceptualise children’s ideas about wellbeing, but to understand how they perceived interrelationships between domains

of wellbeing (Gabhainn & Sixsmith, 2006). They asked child participants to take photos that represented wellbeing, and then discussed the photographs and how they related to wellbeing with a second group of children. The authors extended the original research by repeating part of the process with parents and teachers, allowing them to study the differences between adult and child perceptions of wellbeing conceptualisations (Sixsmith et al., 2007). Children's original summary themes included social relationships, activities, food and drink, and animal/pets, whereas parents highlighted school as an important domain but did not nominate pets. This research suggests what may be included in child-based conceptualisations of children's QOL and emphasises how the inclusion of children in the research process can alter conceptualisations of wellbeing, and by extension, QOL for children.

Lastly, a group of researchers in the UK used qualitative methods with children with CP to understand children's conceptions of QOL, and how well they map to two existing instruments for measuring QOL (Parkinson et al., 2011; Young et al., 2007). Both these instruments included children and parents in their development. The first of these was the KIDSCREEN (Ravens-Sieberer et al., 2005; The KIDSCREEN Group Europe, 2006), a generic child health-related QOL measure developed with close parallels to DISABKIDS (see Section 2.2.3.1.2 for further information on DISABKIDS). The second was the CP QOL-Child, a CP-specific measure of QOL for children (discussed in Section 2.2.3.1.4), with content derived from qualitative work with children with CP and their parents (Waters et al., 2005). While the analysis of children's responses largely corroborated the content of both the KIDSCREEN (Young et al., 2007) and CP QOL-Child (Parkinson et al., 2011), neither instrument fully captured aspects of QOL children valued. For example, in both studies, children nominated the importance of relationships with extended family members, though this is not a feature of either instrument. Both instruments did not capture all aspects of children's recreational experiences relevant to QOL, including environmental accommodations and availability, and opportunities for relaxation. Additionally, children's experiences of negative emotions, safety and community environment, and inclusion and fairness may have high impact on their sense of QOL but are not well captured on these two measures. While the authors acknowledge that some of these differences could be due to differences in cultural

context, current knowledge does not adequately explain the differences, or how they fit into a conceptual model of QOL for children with CP. Therefore, a greater breadth of research which includes the voices of children with CP is required to adequately conceptualise QOL for children with CP.

3.6 Summary

Despite research showing that QOL for children with CP is as diverse as QOL in the general population, there are still clear gaps in conceptualising what QOL may mean for children with CP. Both developmental theories of psychology and later sociology of childhood research makes clear that children's needs and experiences of wellbeing and QOL differ from adults. Yet models and concepts commonly applied to children's QOL have been developed for an adult population; it is unclear whether and how QOL may be conceptually different for children. Similarly, despite the acknowledgement of the importance of family and wider societal contextual factors for children's health in general, and for children with disability in particular, this work has not been well applied to existing conceptual models of QOL. UN conventions on the rights of both children and people with disabilities encourage research agendas that are specific and inclusive to the perspectives of children with disabilities. This is especially relevant given the subjectivity of QOL. Therefore, to strengthen conceptual models of QOL for children with disability, research must focus specifically on children's own perspectives.

While there is limited work examining wellbeing from the perspective of children with disabilities, the original work for the CP QOL-Child (Waters et al., 2005) included development of a conceptual model based on qualitative work with families (see Section 2.2.3.1.4, page 49). However, in the years since this work was carried out, there have been significant technological and other changes in everyday life, including the rise of social media, that may well have changed how children's QOL matters in a clinical context, or factors impacting on those areas of QOL related to communication or social interactions. The following chapter outlines the methodology for investigating what these factors may be.

Chapter 4: Methodology

It is necessary to increase our understanding of the QOL construct from the point of view of children with CP and their parents to improve both conceptual understandings of QOL and potentially revise existing QOL measures. This follows from research developing and advocating for a conceptual model to underpin measures of QOL (e.g. E. Davis et al., 2006; Reeve et al., 2013). Therefore, the focus of this qualitative project will be to develop a contemporary conceptual model of QOL from the perspective of children with CP and their parents, who are the intended respondents of the CP QOL-Child. One ultimate aim of this work is to inform a subsequent research project to revise the CP QOL-Child.

Before describing the specific methods of this thesis, this chapter describes the background literature that led to the use of the qualitative methodology. This begins with a basic discussion of how qualitative methods can be used to investigate QOL, and especially how the use of constructivist grounded theory is suited to the research questions of this thesis. More detail is provided on special considerations for conducting qualitative research with marginalised and potentially vulnerable populations, such as children with disabilities. The section concludes by describing new techniques that can be used for improving research practices for child participants. These approaches are presented as alternatives to traditional interviewing.

After the background on qualitative methodology, the second half of the chapter describes the qualitative methods used for this thesis. This includes details of recruitment, interview schedules, and data analysis for interviews with families of children with CP, as well as ethical considerations and the use of pseudonyms.

4.1 Qualitative research principles and methods

Qualitative research methods are well suited to exploring the important areas of life for an individual or group—especially those with a disability—for three main reasons. Firstly, the focus of qualitative research is participants' subjective life experiences—as is the conceptual definition of QOL. Secondly, qualitative research questions

emphasise ‘what’ and ‘how’; they are intended to lead to interrogation and generation of new theory. Lastly, the data collection methods are reflexive, responsive, and can be adapted to suit involvement of those who might otherwise be excluded from research participation, such as children or people with disabilities. From the perspective of rights-based disability principles, this process is not only enlightening for the researcher, but can be empowering for participants as well.

4.1.1 Overview of qualitative research principles

The qualitative research approach broadly questions the meaning of social phenomena (Wertz et al., 2011). Qualitative research focuses on participants’ subjective experiences and their social context, allowing the meaning and interpretation of conceptual relationships to be co-constructed between researchers and participants (Liamputtong, 2013). This is consistent with two of the basic principles of QOL research: firstly, that QOL is a subjective construct, and secondly, that QOL measurement tools must be based on the perspectives of respondents. Qualitative research also offers the opportunity to use the language of participants themselves, through quotes and interview transcripts, and this is recommended for developing specific items for a QOL tool (Brod et al., 2009).

Qualitative methods are reflexive in critiquing researchers’ prior assumptions and ways of theorising, and instead emphasise participants’ own narratives and perspectives in the creation of knowledge, as experts on their own life experiences (Charmaz, 2014). In practice, this means that the qualitative researcher must adapt their research questions to the subject matter raised by participants rather than forcing predefined ideas into the data. By ensuring the research process is responsive and fluid, the qualitative approach can provide rich data about the whole of participants’ subjective experiences. This is particularly relevant when addressing research in QOL, which relies greatly on participants’ subjective judgements of their life experiences. In disability research, the reflexive process requires researchers to interrogate their own assumptions and stigmas related to participants’ differences, and how they are viewed by society.

In an extension of this approach, qualitative researchers have adopted novel methods for collecting data in ways not traditionally included in quantitative research. Novel methods may include drawings, photographs, children's games, stories, or videos produced by participants (Groundwater-Smith et al., 2015). These methods are designed around participants' abilities and knowledge, and encourage participation by those who may be excluded in traditional quantitative research which requires restrictive methods of participation (Liamputtong, 2007).

Qualitative methods may also be effective in empowering participants who otherwise have little opportunity to voice their own experiences, such as children and people with disabilities (Liamputtong, 2013). It is especially important for children with disabilities, whose perspectives have been left out of the development of many theories and frameworks informing public health and QOL research. Including the voices of children in QOL research supports a rights-based perspective of disability research and provides a conceptual understanding of QOL that preserves children's own meanings of QOL. Ultimately, this knowledge can benefit children by providing evidence for how health practitioners can best improve or maintain the QOL of children.

All qualitative approaches focus on participants' subjective experience, are responsive to questions arising in the data, and reflexive in their approach. Therefore, choice of the particular qualitative approach used depends on the researcher's own philosophical position, as well as the research question, context, and participants (Wertz et al., 2011). The following section presents an overview of grounded theory methods, the qualitative approach used in this research.

4.1.2 The grounded theory method

The grounded theory approach was one of the first qualitative methods systematically documented, in the ground-breaking 1967 text "The discovery of grounded theory: Strategies for qualitative research" (Glaser & Strauss, 1967). Since its initial publication the grounded theory method has been interpreted and revised in many different ways, but it remains the most widely used method of qualitative inquiry (Bryant & Charmaz, 2007).

The original grounded theory method espoused by Glaser and Strauss contrasted with prevailing quantitative paradigms by rejecting the hypothesis testing model in favour of 'discovering' theory in the data. Glaser and Strauss argued for social scientists to place greater emphasis on developing usable theories which fit the data they collected, rather than collecting data to fit with pre-conceived theories (Bryant, 2014). In this way, theory is 'grounded' in the data.

The grounded theory method is appropriate to analyse data from many different types of sources, but it has most frequently been used for interviews and focus groups. The salient point of data collection is that it occurs simultaneously with data analysis.

Analysis follows a process of 'constant comparison', between early codes and new data during data collection, and between different sources of data during the analysis. As part of the process of constant comparison, researchers continue to revise both their analysis and data collection by comparing them as they unfold. One of the principles of the original grounded theory method was not only that it should result in usable theory, but that theory must be modifiable as new data changes the codes and categories that form the analysis (Charmaz, 2015).

This classic version of the grounded theory method was widely adopted across the social sciences in the decades following Glaser and Strauss' original publication. However, classic grounded theory methods have been heavily criticised for suggesting that researchers are able to be objective and unbiased by previous knowledge, merely 'discovering' theory as it emerges from the data (Bryant, 2014). An influential publication by Strauss and Corbin (1990) addressed similar concerns that grounded theory methods were not rigorously explained. This later work takes an even more objectivist stance to the discovery of theory, positioning the researcher outside the analysis process objectively bringing theory forward from data through a prescribed set of practices.

In the 1990s a third branch of the ground theory method emerged as an alternative to this objectivist stance. Known as constructivist grounded theory methods, this branch of the grounded theory method has been championed most notably by Kathy Charmaz (for example: Charmaz, 2014). This method maintains many of the original precepts of

classic grounded theory methods but adopts the position that meanings within the data are co-constructed between the researcher and participants. Researchers are therefore encouraged to interrogate their own viewpoints, and to consider how their personal perspective may influence the resulting theory.

The constructivist approach recognises that researchers begin with knowledge of their area of study and may already have hypotheses as to what will emerge in the data. The constructivist process of data collection and analysis is designed such that the researcher acknowledges how initial assumptions may influence the data collection. It is flexible to changing questions based on what emerges in the data (Charmaz, 2014). This approach is appropriate for the revision of existing knowledge, such as in the case of the QOL domains identified in the CP QOL-Child. It maintains the importance of listening closely to participants' lived experiences, and stresses social contexts and personal viewpoints in the interpretation of data. This is especially relevant to both the QOL concept and to research involving people with disabilities. Therefore, the constructivist grounded theory method was used in the current study; this method is described in detail below.

4.1.3 Constructivist grounded theory methods

Constructivist grounded theory method is based on the epistemological position that the researcher and participant co-create knowledge through interpretation of interactions and life experiences (Charmaz, 2014). Charmaz contends it is naïve to assume that researchers begin with no knowledge of their research area, but rather directs researchers to consider how their personal perspective, shaped by what they know, influences the interpretation of data and data analysis. Constructivist grounded theory therefore departs from the original grounded theory method by emphasising the subjectivity and flexibility of the research process rather than assuming that researchers are neutral bystanders 'discovering' theory (Charmaz, 2014).

Methods of data collection used for constructivist grounded theory are similar to those of other qualitative approaches, with open-ended or semi-structured interviews and focus groups among the most common. Charmaz (2015) advocates 'thick description' in data, also including extensive field notes and memos written during the data

collection phase. These notes and memos are then included into the later analysis of interview transcriptions. It is this full understanding of contextual clues and cues that also allows the researchers to more deeply analyse hidden assumptions behind what is being said, and what is not being said. This level of analysis eventually leads to descriptions of phenomena that are generalizable beyond the sample of included participants by comparing findings with existing evidence to corroborate new findings and build interpretive theory.

Most qualitative methods, including grounded theory, examine data by labelling sections based on the content, processes, or underlying meaning being described (Wertz et al., 2011). This process is known as 'coding', and the codes which arise form the basis of qualitative analysis. Since the division of original grounded theory methods into multiple schools, authors from each approach have differentiated their approaches by providing considerably more detail into their own specific form of coding and analysis.

One of the hallmarks of the constructivist grounded theory method is the initial use of line-by-line coding. Hand-in-hand with line-by-line coding, Charmaz (2015) recommends the use of direct transcription of interviews as a way of very closely examining the data and developing new insights. As its name suggests, line-by-line coding involves applying a short code to every line of a transcribed interview. Charmaz recommends that codes use gerunds, or verbs, to remain active and examine how the participant interacts with the subject matter, rather than merely describing the interview content (Charmaz, 2014). The concept of 'lines' is an arbitrary designation, designed to imply examination of individual phrasings and emphasise breaking down assumed meanings. In this way, researchers are likely to identify ideas and nuances, both explicitly and implicitly stated, that may be overlooked in a more general content analysis (Charmaz, 2014). Additionally, coding line-by-line helps to make explicit the connection between developing theory and data and helps prevent researchers' own preconceptions influencing the analysis. Throughout the coding process, researchers write interpretive memos to further describe codes arising in the data, and move forward from description to interpretation and theory.

Following the initial line-by-line coding of some data collected early in the research project, such as the first few interviews, data analysis moves to focused coding. At this stage, the initial coding will have already suggested what may be important analytical directions. With this in mind, the researcher can decide which of the initial codes are most important, or occur most frequently, to carry forward in the analysis (Charmaz, 2015). These focused codes are then used to quickly move through larger quantities of data, such as several additional later interviews. By engaging in constant comparison, between and within interviews and early codes, focused codes can be tested for how well they fit the data, as well as their underlying conceptual properties, and relationships and contexts (Charmaz, 2014). The final stage of coding involves developing focused codes beyond descriptions of the data into conceptual categories. Other forms of qualitative analysis refer to 'themes' within the data, which are broadly equivalent to categories in grounded theory as outlined by Charmaz. As this thesis follows the principles outlined by Charmaz, the following chapters make reference to categories rather than themes.

As Charmaz explains (2014), chosen categories are those that best describe the data, but they also explicate generic, fundamental processes which can be generalised outside of these specific examples. To fully understand categories, focused coding examines the background, context, determinants, and consequences of each category. Complete explanations of categories within the analysis include examinations of relationships between them—again, based on focused coding of the data.

While grounded theorists may begin interviewing participants with specific semi-structured interview prompts, Charmaz (2015) describes how this can change over the course of the study. As coding moves to categories, questions arise over when and how categories do or do not apply, how they relate, and what processes underlie them. These types of questions are tested not only through comparing interviews after they have been conducted, but also by modifying the questions asked in later interviews. This may mean asking entirely new questions, or merely checking and clarifying in more detail information participants provide relevant to those questions. This process necessarily relies on writing detailed memos throughout the interview process, as well as beginning analysis concurrent with data collection.

It is the process of constantly refining interviews and categories that leads to theoretical saturation. Theoretical saturation is achieved when analytical categories are fully described and raised to a conceptual level to form the final grounded theory. Not every analytical category may be fully described or raised to the level of a conceptual category in the theory. The process of interpreting categories to raise them to the abstract level of concepts is one way in which researchers construct meaning from the data—and the interpretations may change from one researcher to another even with the same data (Charmaz, 2014).

The final formation of ‘theory’ is one point which distinguishes grounded theory analysis from other similar types of analysis, such as thematic analysis, and one aspect of grounded theory that is least likely to be followed (Mertens et al., 2011). It is important to note that the term theory as used here may differ from empirical theory as used in positivist research. Charmaz (2014; 2011) describes grounded theory as abstract interpretations of concepts—the extension of analytical categories—and understandings of the relationships between them; there is limited focus on causality. Therefore, theory arising from a constructivist grounded theory analysis is readily applicable to forming a conceptual model of QOL.

The concept of methodological rigour is often raised in response to the use of qualitative methods, including grounded theory. Constructivist grounded theory takes a philosophical stance not only that data are co-constructed by participant and researcher, but that findings are an interpretation of participants’ worlds, rather than an exact depiction of a single ‘truth’ (Charmaz & Belgrave, 2014). This position necessarily requires reflexivity in the analytic process, as researchers must analyse not only participants’ responses, but how their own language and questioning style may have impacted on the interview data. Researchers can improve the validity of their work by describing not only the theoretical analysis of interviews, but details of how interviews were conducted, and the interactions between the researcher and participants (W. A. Hall & Callery, 2001). This not only allows others outside the project to understand the context of the research, but during the interview process it may help to explicate assumptions being made in the interviews as to shared knowledge, adopted roles, or the relevance of discussed topics. Hall and Callery (2001) therefore

recommend notes on interactions form part of memos taken during the interview process; Charmaz (2015) similarly recommends expanding on transcriptions of interviews with detailed descriptive notes of observations at the time of interviews. By engaging in this type of reflexivity, as well as constant comparison between interviews, codes, and theory during the analysis, it is possible to develop a generalisable theory with good 'fit' for the data, i.e. high validity for participants' experiences. The theory is developed and improved through inclusion of existing theory and literature in the final stages of analysis. This stage builds the robustness of the findings, especially in moving from individual participants to generalisable descriptions of phenomena (Daly et al., 2007).

The constructivist grounded theory approach has previously been used for a project similar to this thesis, though focused on a different age group: as mentioned in the previous chapter (see page 72), Shikako-Thomas and colleagues examined factors contributing to QOL from the perspective of adolescents with CP (Shikako-Thomas et al., 2009). The researchers chose this approach for its ability to generate a nuanced, complex theoretical understanding of QOL based on the direct experience of those involved—in this case adolescents with CP. Data were collected through semi-structured interviews that were interviewer led, but flexible to the participants' responses and interests. New ideas and links between ideas in the analysis were then considered in light of what is already known about QOL determinants in other populations. While they could include as participants some adolescents from all GMFCS levels, this more traditional interview format restricted their recruitment to only those capable of engaging in such a dialogue with no or minimal communication assistance from a parent or guardian.

This type of limitation is common in research, including qualitative interviews, with children and those with disabilities. Research with these groups necessarily raises considerations of the meaning and ethics of participation, in terms of accessibility and representation. Additionally, if participants are limited in their ability to participate due to communication impairments or cognitive understanding of the research questions, it invokes a need to develop alternative methods of data collection. Both these points have been explored by qualitative researchers from multiple disciplines

working with children and people with disabilities. Some of this work is addressed in the following section on working with vulnerable populations and informs the methods of the current study as described later.

4.1.4 Qualitative research with potentially vulnerable populations

Qualitative researchers have written specifically on managing research with special population groups referred to as ‘vulnerable populations’ (e.g. Liamputtong, 2007). The concept of ‘vulnerability’ relies on social constructions and is context dependent. Some groups may experience vulnerability in multiple ways. However, generally those considered vulnerable are those who experience diminished autonomy in some way, are marginalised groups within society who are less likely to be included in mainstream research, and are more likely to be at greater risk of poor health outcomes (Liamputtong, 2007). While these factors do not necessarily imply reduced ability to participate in research, they can contribute to increased potential for harm through research participation. However, by recognising ways in which people are potentially vulnerable or subject to exclusion, qualitative research methods can be altered to be more specifically inclusive and provide a voice for people otherwise excluded from traditional quantitative research.

Historically, children and people with disabilities, with other marginalised groups, have experienced research as a practice done to them, rather than with them. This has been a result of coercive practices of obtaining consent, or the assumption that consent is unnecessary for certain groups who did not always receive recognition of basic human rights (Mertens et al., 2011). Some historical research has also involved the privileging of dominant narratives that exclude alternative perspectives of marginalised groups, or do not allow participants an appropriate avenue to fully participate (Liamputtong, 2007).

Modern qualitative research frequently seeks to support the rights of marginalised groups and redress existing inequalities. Yet in most situations there still exists a power differential between the participant and the researcher—who represents some form of authority. Such participants may fear negative consequences of either refusing to participate, or providing data that are contrary to what researchers wish to hear

(Liamputtong, 2007). People who are reliant on provision of social services—such as those with disabilities—may be especially prone to this type of coercion. They may feel pressured to consent if they perceive their services to have a vested interest in the research outcome, or fear withdrawal of essential services should they not cooperate. Moreover, children and adults with intellectual disabilities may be less able to perceive potential negative consequences, or to understand the demands of participation prior to consenting to participate. However, membership of a marginalised group does not automatically confer an inability to understand research demands.

Ideally, procedures for obtaining consent and methods of data collection should be tailored to the participants' abilities, guided by participants themselves, and recognise participants' right either to participate or refuse consent and request their data be withdrawn (Mertens et al., 2011). In this way, marginalised communities retain a more level balance of power with researchers, can make a more meaningful contribution to research, and ultimately achieve more powerful research outcomes for themselves.

Alongside the recognition of rights of potentially vulnerable participants, new approaches to interviewing have been promoted for participants who may be otherwise unable to understand or complete a traditional verbal and written research activity. Such participants may include children, people who are illiterate, have communication or intellectual disabilities, or those whose primary language is not that of the researchers. One alternate approach is intensive open-ended interviews which are flexible to the interviewee interests rather than following a pre-determined guide presented by the researcher (Borer & Fontana, 2012). Flexible, open-ended interviews fit easily within the constructivist grounded theory method, which similarly emphasises the need for flexibility in interviews (Charmaz & Belgrave, 2014).

Other authors have expanded on the value of research flexibility in philosophical considerations of the needs for research participants to have agency over the construction of their worldviews, as part of the recognition of the rights of those in marginalised populations such as disability communities (Mertens et al., 2011). Alongside the recognition of the construction of worldviews is the recognition of the heterogeneity of vulnerable populations: just as within wider communities there may

be differing perspectives on many social issues, or differing experiences with power structures, these exist within minority communities as well. To ensure the full participation and rights-based representation of people with disabilities in research, researchers must be prepared to encourage multiple methods of participation, and divergent views on experiences of power and oppression and the social construction of disability.

Similar points have been recognised in moving from philosophical and methodological considerations to specific methods for engaging in research with children. Advocates for child-centred methods may be making assumptions as to the ability of all children to participate in traditional research—when we should be focusing instead on person-centred methods that recognise children will have diverse interests and abilities to participate (Groundwater-Smith et al., 2015). Not only that, but the use of alternate methods to collect data are likely to impact on the data itself, and how knowledge is co-constructed—which should be critically reflected on in the research process. Specific methods that have been used in research with children, and children with disabilities are described in detail in the following section.

4.1.5 Qualitative interview strategies for engaging children

There is a growing literature showing how young children in general have been engaged in research interviews by the use of a number of innovative research methods designed to elicit more detailed and nuanced responses than traditional interviewing questions (Teachman & Gibson, 2013). Many of these strategies can be applied in face-to-face interview settings, singly or in combination. Some are newer methods from the wider field of participatory action research, adapted or developed specifically for children—these methods move the focus away from researcher-led interviews to a wider range of child-led approaches which may not include interviews at all (Groundwater-Smith et al., 2015).

Innovative methods do overlap, however they generally fall into five categories: alternate verbal strategies, visual tools, technological alternatives, play-based activities, and ethnographic methods. The following sections describe each of these approaches with some of the uses and challenges of each methods. Unfortunately,

despite the promise these methods have in engaging children with disabilities, very little has been published on additional considerations and applications for children with disabilities. Some examples are given where these exist.

Besides focusing on specific techniques, some researchers have successfully used and advocated for combining multiple methods (Darbyshire et al., 2005; Teachman & Gibson, 2013). Therefore, after describing specific strategies, the benefits and challenges of using a flexible combination of strategies are discussed—including why this may be the ideal approach for which to aim.

Though they may be useful in giving children choice, eliciting rich data, and building rapport, innovative methods should be chosen to fit the research questions (Groundwater-Smith et al., 2015). Therefore, this section concludes with considerations in choosing how and when to employ innovative methods with children, which methods to use, and the ethical implications of using them.

4.1.5.1 Alternate verbal strategies

Researchers have maintained the verbal focus of a traditional interview while increasing children's participation with the use of alternate verbal strategies. The simplest strategies involve using conversation, rather than a formal interview (Groundwater-Smith et al., 2015). Conversations may take place while children are engaged in other activities and attempt to mimic a more normal interaction. Although the researcher may still introduce specific topics, conversations allow for greater control by the child, but also rely on active engagement and participation by the child.

Another alternative verbal interview format is using role-plays or drama to create situations or to pose questions to address the research agenda. Through the use of role-plays children may feel freer to express their own opinions or discuss traumatic experiences by distancing themselves into other characters, while providing context to familiar situations (Veale, 2005). However, it may be difficult with both methods to distinguish between creative imagination and children's own experiences or views—particularly with younger children (Groundwater-Smith et al., 2015).

Another verbal technique that researchers have used within a more traditional interview is sentence completion (Teachman & Gibson, 2013). In this activity, participants are provided with the beginning of a sentence and asked to complete it in their own words. For example, Teachman and Gibson (2013) used this technique to elicit children's opinions of therapies they experienced, such as "The best thing about therapy is..." and "The worst thing about therapy is...". While children did not always respond well to this technique, it allows the interviewer to introduce a topic the child might feel is otherwise off-limits and encourages responses from children who may have difficulty formulating their own answers unprompted. Furthermore, this technique requires limited additional input or abilities on the child's part and may be adapted to any child able to participate in an interview.

Cognitive interviewing is a technique often used in measure development with both adults and children, which combines alternate verbal strategies with physical and visual prompts. For example, researchers developing a new measure of wellbeing for younger children investigated how children responded to new items, including pictorial items, by asking them to 'think aloud' as they completed them (Rebok et al., 2001). This demonstrates how both questions and response formats are understood by the intended respondents of a measure and is commonly used in the development of patient reported outcomes. It can prompt discussion of the content but is not used to generate new content. The 'think aloud' method has also been used as a verbal interview to understand discordance between child and proxy responses to a general QOL questionnaire (E. Davis et al., 2007). However, the pictorial response format used by Rebok et al. (2001) to pilot new response formats is very similar to more general visual techniques described below.

4.1.5.2 Visual tools

Visual tools often provide the simplest ways of changing the format of an interview. There are many variations on how to incorporate visual prompts into verbal interviews. The most common include the use of photographs, pictures, and drawing as prompts to elicit narratives and opinions. For example, children are asked to draw a picture of a specific activity related to the research questions, and then to discuss

elements of the drawing and why they chose to draw it, or what feelings it elicits for them (Hill et al., 1996). It has been theorised that by asking participants to engage with visual methods, particularly drawing, they engage with different ways of thinking than with traditional verbal methods; different ways of thinking in turn call to mind additional features and emotions of their responses (Harper, 2002; Sewell, 2011). Some researchers suggest this is especially the case for children whose cognitive understanding is greater than their verbal ability (Horstman et al., 2008).

When used specifically with drawing, this technique is known as 'draw-and-write', due to the common practice of asking participants to make notes on the drawing rather than just explaining it verbally (Sewell, 2011). It has been used primarily with children, exploring a range of topics including health, psychology, education and school experiences. The method is accessible to children who cannot write for themselves by allowing the child to dictate exactly what should be written about their picture (McWhirter, 2014). Researchers generally do not analyse the drawing themselves, focusing instead on children's written or verbal explanations.

Potentially the simplest method of introducing a visual element to interviews is the method of photo elicitation. Photo elicitation involves presenting an interview participant with a photo or image and asking them to respond to it verbally (Harper, 2002). As Harper (2002) discusses in his highly cited paper on photo elicitation, the types of images that can be used are virtually unlimited. Different subjects are likely to evoke different types of responses, but all require participants to engage in both visual and verbal processing; this is hypothesised to lead to richer responses.

Recently, researchers have focused specifically on the use of emoji as visual elicitation tools in research with children (Fane et al., 2018). Emoji are a series of small images, such as facial expressions, widely used in digital media and new technologies to represent various everyday emotions, animals, food and drink, and other objects. With children's increasing use of technology in their everyday lives, emoji offer a new way of including visual images in research, while incorporating communication symbols already familiar to children. The use of emoji already familiar to children supports the inclusion of 'child voice' in the research by elevating them to a position of expert in the

research materials, while providing greater emphasis to meanings children make themselves, as opposed to that framed by the context of photos chosen by researchers (Fane et al., 2018).

An extension to photo elicitation techniques, which use researcher-generated visual material, is 'photovoice', which instead uses participant-generated material. Visual materials in photovoice can be used both as elicitation prompts in verbal interviews, but also form part of the data to be analysed. Data may be in the form of photos or videos, where children are given cameras and encouraged to carry these with them over several days and photograph things which are meaningful to them (Einarsdottir, 2005). This approach was successfully used to understand the meaning of 'wellbeing' for Irish children aged 8-12 years, including a follow-up activity in which a second group of children categorised the photos into wellbeing-related themes (Gabhainn & Sixsmith, 2006). This method helped to expose the relationships between aspects of wellbeing in children's lives, as well as allowing children themselves to be involved in the analysis of the photos.

Studies using photovoice and photo elicitation with children with disabilities were recently reviewed, including recommendations for adaptations (Eisen et al., 2018). The authors concluded not only that these methods can provide valuable insight into the lives and experiences of children with disabilities, but they may be a particularly powerful method for those with communication impairments.

More complex visual methods, such as ecomaps, encourage children to develop maps of relationships between elements of their lives, such as people, places, and activities (Rogers, 2017; Woodgate et al., 2016). This encourages children to show how they engage with different parts of their lives, and which areas are most meaningful for them. These methods are most useful for research questions related to social relationships and social capital.

Visual methods and prompts are useful in discussing sensitive topics with children moving the focus away from directly responding to a researcher's questions, and may simplify otherwise complex issues, or provide additional time to formulate answers. Drawing tasks also allow children to choose topics and direct the interview more than

a traditional question and answer format. Using pictures rather than drawing may be more accessible for children with physical disabilities, while still allowing them to engage with visual methods.

4.1.5.3 Play-based activities

Play-based activities are often used for younger children from pre-school and early primary school age, and often involve researchers engaging in play with children while conversing naturally rather than interviewing (Groundwater-Smith et al., 2015). They are more natural for children of this age, who often engage in non-verbal play, and may find it difficult to sit still and answer questions for a longer period, or to understand abstract verbal constructs. Play-based methods may also encourage researchers to consider broader questions, with data including more descriptions of non-verbal behaviour, such as what children have engaged with, or if there are multiple participants, how and with whom they have interacted.

There are also simpler play-based activities that resemble the use of visual methods. For example, Koller and San Juan (2015) explored children's perspectives of inclusive education using a combination of non-directive play with toys, as well as dolls and pictures depicting both typically developing children and children with visible disabilities. Dolls and pictures were used both as props for play and as prompts for discussion of specific research questions.

These may also overlap with ethnographic methods and ecomapping, in asking children to interact with their environment in ways that are close to their usual lives. While play-based methods are effective in engaging and including younger children in research, these types of activities are likely to be less attractive to older children who engage in different forms of play.

4.1.5.4 Ethnographic interviews and new technologies

Ethnographic interviews provide richer experiential data by encouraging children to physically lead the interviewer through a tour of their lives and their local environment. Children are given far greater agency in these interviews and may entirely direct the content and issues discussed.

The use of photos and videos produced by children, as in photovoice, may be considered an ethnographic method. In a more direct replication of photovoice techniques, a researcher may ask a child to lead them on a tour of their home, or places in their neighbourhood, and point out what matters to them. This has been used successfully for children with physical impairments describing their leisure activities at home (Kanagasabai et al., 2018). Again, while the initial aims may be described (e.g. the children know that the interviewer is interested in their games and pastimes), the interview itself is child-led. The physical environment acts as both a prompt for verbal interactions, and a distraction from what may be intimidating or distressing for children to discuss with a relative stranger.

The use of new technologies such as social media, apps, and wearable recording devices offer new ways for researchers to observe children's everyday worlds, and to engage them in research. Some authors also consider the use of social media for interviews, or online interactions as data gathering methods (Groundwater-Smith et al., 2015). These online methods have been used successfully for older children in research addressing specific online environments and relationships, such as social negotiations in online communities (Boyd, 2007), but may be limiting in more general research. Additionally, the use of online-only contact may be more ethically challenging in sensitive research, where researchers cannot distinguish cues of child distress, nor build researcher-participant relationships that may help children to discuss emotionally-laden topics (A. Morris et al., 2012).

4.1.5.5 Using multiple methods

There may be additional benefits to both children and researchers in combining strategies described above within a single research project. This can be done sequentially in different settings, such as used by Darbyshire et al. (2005), who combined individual interviews using photo elicitation with focus groups involving games. Another option is the flexible 'toolbox' approach adopted by Teachman and Gibson (2013), whereby they offered different strategies within single interviews and proceeded with those to which each child best responded. Similarly, Hill et al. (1996) combined multiple methods within both focus groups and interviews to understand

children's perspectives on wellbeing and emotions. Reporting on multi-method research for children with disabilities, Pickering (2013) emphasises that designing creative research which provides children with disabilities space to participate meaningfully is the best way to increase child 'voice' in disability research and policy.

For children, the use of multiple methods provides additional opportunities to participate in the way which is most meaningful, comfortable, or interesting to them. Necessarily, this will vary depending on the child—not all children will be as interested in using drawing methods, nor will all find prompted sentence finishing exercises useful in discussing difficult topics. Pickering (2013) suggests that having a choice over how they participate directly increases child engagement in research. Additionally, by offering choice over methods and moving toward child-led models of research, multiple methods recognise children's rights to expression in matters which concern them (United Nations, 1989).

For researchers, using multiple methods offers the opportunity to explore the research questions from different perspectives—which may lead to additional insights. For example, Darbyshire and colleagues (2005) found that different data emerged from different ways of asking children to participate in research about their leisure and physical activities. They concluded that had they used just one approach, they would not have uncovered some of their most significant findings.

Just as not all methods are suited to all children, neither are they appropriate for all research questions. Simply adding strategies to the methods without considering their implications will not result in better research. How to choose between strategies and other considerations in using innovative methods are reviewed below.

4.1.5.6 General considerations in choosing alternative interview strategies

As with all qualitative research, the choice of data collection strategies depends on the research questions and how they can best be answered (Groundwater-Smith et al., 2015). For example, questions which focus on place may best be answered with ethnographic data, while ecomaps may be more appropriate if the focus is on relationships and social networks. All methods, if used well, aim to elicit rich data on

children's views and narratives, and can be coded using constructivist grounded theory methods.

Data collection is also subject to available resources, and the ability of participants to actively participate. In the current project, children with diverse physical abilities were being sought as participants. Therefore, ensuring that interviews were inclusive was a key criterion, and the major reason for adopting alternative interview strategies. For this reason, ethnographic methods were not considered, as children with mobility limitations face greater challenges in leading researchers physically through their local environments. Similarly, play-based activities that rely on greater physical involvement were not used in the current study.

However, some of these strategies can be adapted, such as allowing children to lead the interviews verbally by describing the places where they go in their day-to-day life, and which matter most to them. This strategy can be adopted in conjunction with simpler visual methods, such as the use of generic pictures, or drawing tasks for more able children. With any method used, the interviewer must be responsive to the child's abilities and interests to best engage them. Allowing children to lead interviews as much as possible not only suits the use of alternative methods of data collection, but supports the ethical approach to working with vulnerable groups (Darlington & Scott, 2002).

Along with adopting a flexible combination of strategies, Teachman and Gibson (2013) collaborated with parents prior to the child interview to ascertain children's preferences, abilities, and some details to personalise the interview. Teachman and Gibson particularly used collaboration with parents to prepare for use of strategies such as role-play and vignettes, when the characters adopted within these 'stories' could be modelled on the child's own background and activities, such as experience with particular modes of therapy. Similar strategies could be adopted in the use of visual tools, by tailoring which images would be presented to a child based on prior information from the parent as to that child's usual activities. However, parental information used in this way must be carefully considered, in order that child's

perspectives are not falsely limited or replaced by the initial parent report (Teachman & Gibson, 2013).

Throughout the data collection phase, the inductive and reflexive processes of grounded theory methods encourage adjustment not only of the coding strategies and interview questions, but of interview strategies as well (Charmaz, 2014). It is likely that some strategies will be more suited to some children than others, but if some are particularly successful in eliciting rich data, or are universally popular with children and parents, they will be more relied on in later interviews.

One of the drawbacks of adopting such strategies is that this field is new and there is limited critical literature, especially in adapting methods for children with disabilities. Researchers have sometimes assumed that using methods like this provides better data without directly comparing across methods outside of the analysis process. Little is known as to what extent customising approaches to individual children changes the responses they give or limits the topics they will raise. In this thesis, reflecting on and critiquing this process also forms part of the analysis.

4.2 Methods for this thesis

This PhD research used qualitative analysis of in-depth face-to-face interviews with children with CP and their parent or primary caregiver. The aims of the interviews with families were to model the issues and concerns that children with CP and their parents nominate in discussing QOL, including how they may feel about relating these concerns to their current service providers, or how they may relate to the services they receive.

The content of a QOL measure should be guided by the perspective and concerns of its intended target respondents, in this case, children with CP and their parents (E. Davis et al., 2006). The use of qualitative methodology involving children with CP and their families maintains their meaningful contribution in shaping the content of the instrument, and builds conceptual understanding of subjective QOL for children with CP.

Data were analysed using constructivist grounded theory methods, based on the principles described in Section 4.1.3 Constructivist grounded theory methods (page

79). Specific details on how this was applied are provided after describing the recruitment and interviewing process.

4.2.1 Participants

Previous research into QOL with children has suggested that prior to the age of 9 years, children have difficulty completing a self-report questionnaire, and may have very different understandings and priorities of QOL (Eiser et al., 2000). Children aged 4-8 years are able to provide information about their life experiences and treatment preferences relevant to a clinical QOL scale, but these scales require a detailed pictorial format, and are administered in a structured interview. The findings of this thesis are partly intended to inform a model of QOL for comparison with existing QOL scales, and a revision of the current CP QOL-Child would update the content but maintain the current self-report format for both child and parent versions. For younger children (and children with severe disabilities), this format is appropriate only for parent report. Therefore, for children aged 4-8 years, interviews in this research were with parents/primary carers only. Children aged 9-12 years had the opportunity to participate directly, to the extent of their ability, based on parents' recommendations.

Children (and their families) were eligible if they had a current diagnosis of CP and were able to be interviewed in person in Melbourne, or a telephone interview in the case of parental interviews. Purposive sampling was used seeking diversity with respect to overall level of motor function, to ensure the findings were applicable to a wide range of children with CP. That is, recruitment targeted children with varying severity of CP as measured by the GMFCS. Further details of the participants are provided in Section 5.1, page 110, for the child participants, and Section 7.1, page 187 for the parent participants.

4.2.2 Recruitment

Families were recruited through the Victorian Cerebral Palsy Register (VCPR), an independent, voluntary database of individuals with CP founded in 1987 and managed through the Murdoch Children's Research Institute. The VCPR maintains demographic information, birth details, and some clinical details on individuals with CP who have agreed to register, for epidemiologic and other research to reduce risk of CP and to

improve the lives of individuals with CP. All information is securely maintained by the VCPR office, following state privacy legislation, and the VCPR is approved by the Human Research Ethics Committee of the Royal Children's Hospital, Melbourne. The VCPR has ongoing ethics approval to collect basic information on individuals with CP and their families. After being added to the register, families/individuals are contacted to determine if they wish to opt-in to adding additional clinical information, and additionally choose whether or not they wish to be contacted about future opportunities for external research participation. In consultation with the VCPR, a recruitment protocol was established as described below.

The VCPR initially selected a cohort of 80 families based on the recruitment criteria of child's age and GMFCS level, with additional preferences for families located in metropolitan Melbourne. The initial cohort size was chosen in consultation with VCPR staff, based on anticipated response rates and the target sample size of 20-25 families.

Families were first contacted in late October to November of 2016, with interviews taking place from January to June of 2017. The initial recruitment of families was managed by the Victorian Cerebral Palsy Register (VCPR). As the VCPR decided to initially contact families by email, only those with valid email addresses were included—the majority of those on the register have email contact information. Families were eligible if they had a child aged 4-12 years, with the sample stratified to include children from all GMFCS levels across the age range. As all child interviews were to be face-to-face, families with a child aged 9-12 years were included only if they lived in metropolitan Melbourne. Those eligible for parent-only interviews also included a small number located in other areas of Victoria.

The 80 families were then emailed a short study description based on the Plain Language Statement (PLS). They responded directly to the VCPR manager to indicate either interest in participating, or to decline the invitation. Those who did not respond were sent two reminder emails over a two-week period, following the protocol developed for ethical approval. Those declining the research invitation received no further contact from the VCPR.

At the end of the contact period, the VCPR manager forwarded details of families who expressed interest, and for those who did not respond at all. This followed the VCPR protocol agreed to by families on registration. Forwarded details included names for child and primary caregiver, relationship of the primary caregiver to the child, phone number and email for primary caregiver, child's age, GMFCS, probability of intellectual disability where this was available, and the suburb where the family lived. For families who expressed interest, information was provided as to whether a child interview might be possible. Eleven families replied to the initial contact that they would be willing to participate; six of these included a child who could potentially complete an interview. There were also 51 families who did not respond and were eligible to be followed up by researchers directly.

All 11 families who expressed interest were contacted directly by the researcher to further explain the project and schedule an interview. An additional 14 families who did not initially respond were also followed up directly by the researcher. Location of interviews was determined by family preference during these phone calls, however the researcher suggested the participant's home as usually most suitable for child interviews. For parent-only interviews, parents were also offered the choice to complete the interview by phone or skype, though face-to-face was preferred. Formal written consent was obtained at interviews, or by prior arrangement for those conducted by phone/skype. The PLS and consent forms are included in Appendix A for parent and child interviews, and Appendix B for parent-only interviews.

Each family that participated was offered a \$30 gift voucher to recognise their time.

4.2.3 Interview guides

General interview guides for children and parents are provided in Appendix C and D, respectively. These questions were designed to be general in nature, allowing participants to raise specific activities, experiences, or emotions that they or their parent experience in everyday life. Questions were primarily focused on the concept of 'wellness', health and happiness, in keeping with definitions of QOL requiring positive aspects. However, prompts also at times focused specifically on considerations and consequences of the child's impairments, to elicit data specific to living with CP. Some

prompts were included based on positive parent responses during previous CP QOL-Child qualitative research. Additional prompts were also included introducing domains of the current CP QOL-Child or areas of life that have contributed to child QOL in past research.

The interview guides provided initial prompts for interviewees, but interviews were open-ended and flexible, so that not all questions were asked of all participants if the main areas the interview guide addressed were introduced by participants. After initial questions taken from the interview guide, interviews were led by topics introduced by participants themselves, as well as later interviews including specific issues and topics raised by earlier participants. Interviews involved both open-ended questions about children's life experiences and what contributes to their feelings of their quality of life, as well as more focused questions on some of the domains covered by the current CP QOL-Child (such as relationships, schooling, healthcare, access to services), or possible new items (e.g. new technologies such as electronic tablets or apps), where these were not addressed earlier.

Questions for children, especially early in each interview, focused predominantly on positive aspects of their lives, such as their favourite pastimes, friends, or enjoyable aspects of schooling and hobbies. However, when children introduced negative experiences, these were also explored with additional prompts. Once children appeared to be comfortable in the interview discussions, they were also asked if they had other negative or uncomfortable experiences related to having CP. Often during the initial part of each child interview, a higher number of closed questions were used, as these were easier to answer. Additionally, while most parents were present when their children were being interviewed, parents (and other siblings) were requested to allow children to answer questions directly themselves. Where parents assisted children in communicating in any way, questions were still addressed directly to the child, and parent contributions were checked with children where possible.

At the end of each interview, both parents and children were asked if they would feel comfortable discussing the QOL issues raised in the interviews, with their health care or disability service providers. For example, if they felt it would be appropriate or

useful for their health service to ask them to complete the CP QOL-Child or a similar measure. These questions were intended to gauge whether families felt QOL issues were relevant to the services they currently receive.

4.2.4 Interview strategies for engaging child participants

Prior to the child interviews, several strategies were planned to engage child participants. These included 1) seeking parent collaboration prior to the interview; 2) a warm-up activity to begin the interview, and 3) multiple activity props used throughout the interview including drawing materials, and various printed pictures and prompt cards. A combination of these multiple approaches was used in each interview, with planned strategies for each interview varying based on the child's age and the parent's opinions of their abilities. Throughout the data collection phase, these strategies, prompts, and scripts were refined and adapted based on how children in earlier interviews engaged with them or commented on them. Specific activities which prompted more detailed responses and positive engagement were used in preference to the others. This section describes the planned strategies, while the following chapter provides reflections and discussion of the implementation of the strategies and their influence on data production (see page 110).

4.2.4.1 Parent collaboration

In a phone call prior to the child interview, parents were asked to comment on the capabilities and strengths of their child, including their preferences for different types of activities and their ability to read written cards. This information was intended to initially tailor the interview approach to be most receptive to the child. Where parents indicated children were regularly using alternate methods of communication (such as aids of any kind, or signing rather than verbal communication), parents were asked if specific accommodations needed to be made to the interview, or if the parent was required or able to act as interpreter or facilitator to enable the child's full participation. However, in practice parents only volunteered their children to participate where they felt the child was able to complete an interview largely independently. For child participants with an intellectual disability the interview topics and questions were simplified to suit their level of understanding while encouraging

their participation. Parent presence during the child interviews was at the discretion of the parent, allowing them to assist their child to contribute as they felt a need to do so. In all cases, interview methods and questions were flexible and guided by understanding and engagement of participants, as well as by content raised within the interview.

4.2.4.2 Introductions and warm-up activity

Before beginning the interview, children and parents were presented with written information sheets (Plain Language Statement, PLS) and consent forms and the purpose of the research was explained to the child, along with the types of questions that would be asked. The PLS followed University of Melbourne ethics guidance on necessary inclusions, with the child-friendly version using simple language based on similar PLS used in earlier University of Melbourne child research. Children who were able to provide written consent were encouraged to do so; written parental consent was required for all children. Some parents advised that their children would not be able to understand written information and consent forms, and in those cases, children received verbal information only. Following advice from a child reference group in previous research (Moore et al., 2008), children were assured that there are no right or wrong answers, that they didn't have to answer any questions that they didn't wish to answer. Children were encouraged to ask any questions they had about the research, and child verbal assent was required before continuing.

A warm-up game was prepared to begin each child interview, intended as an icebreaker rather than for data collection. This is based on the interviewer card-game warm-up activity described by Teachman and Gibson (2013), which their child participants found engaging. Beginning the interview with a warm-up activity that is not aimed at data collection helps the researcher to gauge the child's interests and abilities and increases the participant's comfort and reduces anxiety. The game is based on two identical sets of cards pre-printed with questions, with one set provided each for the researcher and child participant. The child and researcher then take turns to read and answer the questions on the cards. The child is encouraged to play interviewer first by choosing a card to ask the researcher. By allowing child participants

to assume the role of interviewer, this game additionally de-emphasises the interviewer's authority and power. For the current study, the questions were: "Can you tell me a little about yourself?", "What is something you're really good at?", "If you could have any super power, what super power would you choose?", and "If you were to be any animal in the world, what animal would you be?". This activity was introduced to each child as a game for the child to get to know the researcher a little before the interview, and because it would be unfair for only the researcher to ask questions.

4.2.4.3 Drawing materials and printed prompts

Several activity options were prepared to aid the data collection portion of the child interviews. These were introduced throughout the interview to make the questions more interesting and fun. Prepared activity props included 1) colour printed emoji faces, 2) pens, pencils, and large paper for drawing and written prompt cards for use in drawing activities, 3) printed photographs of children's everyday places and activities, and pictures of cartoon children with visible motor disabilities. All printed materials were affixed to stiff cardboard to make them easier to handle. Ultimately only the emoji, drawing materials, and prompt titles were used in interviews—this is discussed further in the following chapter.

Most of the materials were visible from the beginning of the interview, so that if the child expressed interest in specific activities, this would dictate how the interview proceeded. Otherwise, the researcher directed the child to activities that might be suited to their interests or abilities based on information from the parent. All materials involving written text were intended to be read aloud by the interviewer as they were used; in a few cases parents assisted children in reading materials aloud.

4.2.4.3.1 Emoji

The emoji faces were used initially as a representation of how the child has been feeling lately "Which one of these is most like you? ... Why did you choose that one?". These could also be combined with other drawings or discussions as prompts for eliciting the child's feelings or applied to specific areas of a child's life such as school, family, or other activities.

4.2.4.3.2 Drawing materials and prompted drawings

Drawing materials were provided primarily as a distraction during the interview rather than for visual data collection. However, they were also used to prompt conversation, for example for children to draw things they mentioned or things they enjoyed. This could be paired with photos or prompt cards, by asking the child to draw specific things representing different parts of their life. The interviewer was then able to ask questions about the drawings and their meanings.

Printed prompt cards could be used as prompts for verbal discussion or incorporated into other drawing activities. There were four prompt cards, with text including “places I go”, “things I do”, “people I meet”, and “feelings I have”. These phrases were chosen based on their successful use in previous similar QOL research (Parkinson et al., 2011), to cover a range of issues relevant to children’s experiences of QOL. As they are more directive than some of the other methods, they were intended to be used where the child appeared to have difficulty responding to other more open activities or questions, to begin discussions about these areas of life.

4.2.4.3.3 Pictures of children’s places and cartoon children with disabilities

The last two activities were planned for but not used in any child interviews; this is discussed further in the following chapter reflecting on the use of all strategies. The first of these was based on photos of children’s everyday settings and activities for general prompts for discussions of QOL. Pictures could be introduced as “some places and things that may look like places you go to and things you do”, with follow up questions focused on asking the child to choose one that is meaningful to them, why they have chosen it, whether it is something they enjoy, or how they feel about the place in the picture.

The second planned activity not ultimately used was based on the vignette activity used by Teachman and Gibson (2013). The child would be presented with a cartoon picture of a child with a visible disability, to be used as a prompt for a story telling game about the cartoon child’s life. The cartoon picture could be targeted to most closely resemble the child participant in age, gender, and visible use of motor aids such

as walker or wheelchair. The picture would be introduced with a script adapted to each child, such as:

“This is a picture of Sally. She has cerebral palsy just like you. Sally is in Grade 5 at school, and she really loves [movie, book, hobby that child participant enjoys]. Let’s make a story about what Sally is doing this week.”

4.2.5 Data analysis

Data analysis was based on interview transcripts, supplemented with interviewer notes and memos made immediately after interviews. All interviews were audio-recorded and transcribed verbatim, including verbal encouragement from the interviewer such as ‘uh huh’, ‘yeah’ etc. Most interviews were transcribed using a professional service. Four interviews were transcribed by the PhD candidate: two early interviews (1 child and 1 parent) transcribed as part of the analysis process; two additional child interviews of children with communication impairments resulting in unclear speech. The content of professional transcriptions was checked against audio recordings prior to analysis.

To allow for child self-report and parent report versions of the new CP QOL-Child to accurately represent the QOL knowledge and content of family interviews, child and parent interviews were analysed separately. However, comparisons were made between them at the level of focused coding and emerging categories. Furthermore, interviewer observations of parent-child interactions during the interviews were used for context in analysing some interview conceptual content.

Data analysis was concurrent with data collection. Initial open coding began during the transcription process. Codes which emerged in early analysis were tested in later interviews, through updated focused questioning during interviews, and in the coding process.

Transcriptions were imported into NVivo Pro 11 for Windows to facilitate coding and analysis; however, all codes were created manually by ES. Following the constructivist grounded theory method (Charmaz, 2014), interviews were initially coded line-by-line, with short, active codes. These were then recoded into focused codes, based on the

most common or most salient codes, as well as on what processes best fit the data. Categories were developed from the focused codes. Emerging focused codes and memos were discussed with supervisors, as were conceptual categories, to explore alternate interpretations of the data and ensure methodological rigour. Moving analysis from initial codes to categories occurred in an iterative process, as later interviews and new interpretations of data raised questions about the earlier codes.

A secondary aim of the analysis was to critique the interview methods used with children. This involved reflecting on multiple levels within the interviews. Firstly, there were practical considerations of applying the chosen strategies with the children—such as the ease of use, children’s interest or lack of interest, and any other similar practical consequences. Secondly, there were specific considerations of the influence of prompted activities on the content of interviews with the children—such as whether the use of emoji or prompted drawings elicited rich or detailed discussion of QOL. Lastly, there were broader reflections on the interactional process and how these strategies may have influenced the work of the interviewer and participant within the interview. For example, Rapley’s review of qualitative interview research (Rapley, 2012) exposes the interactional nature of qualitative interviews, including the active work of the interviewer in guiding participants and the position of participants in creating the narrative and their own identity. His recommendations for explicitly considering the interactional process in the analysis fits easily within the constructivist perspective, and are echoed by Potter and Hepburn in the same book (J. Potter & Hepburn, 2012). Furthermore, the recommendations of these authors together provide a way to evaluate how the specific strategies may have contributed to data produced in the child interviews. Therefore, especially in relation to the use of child-friendly materials, this level of analysis considered 1) how the interviewer’s questioning and prompting guided the children’s responses and way of responding, 2) contextual assumptions and identities children created within the interviews, 3) the roles of interviewer and child within the interview with respect to power and the co-construction of the interview content. The understanding of these factors influences the analysis of interview content, therefore before the presentation of the main

findings for the child interviews, the following chapter describes the implementation of child-specific strategies and appraises their influences on interview dynamics.

4.2.6 Use of pseudonyms

To provide anonymity and maintain confidentiality in keeping with the ethics agreement, all child and parent names were removed once transcription was complete. Similarly, references to specific services, therapists or other identifying information were removed from direct quotes where these are included in any reporting of findings. However, because of the small number of the children involved and the desire to humanise them and enhance their voice in the research, pseudonyms for each child are used to report quotes and findings. Pseudonyms were chosen based on relatively common baby names for the years the children were born. As cultural background did not appear to greatly influence the interview findings, confidentiality was considered a more important factor than adhering to ethnicity in choosing pseudonyms. However, names were chosen for pseudonyms that may be used outside of English-speaking cultures to ensure that other ethnicities are not entirely erased.

Pseudonyms were not used for parents, as the larger number (18) made this more impractical and potentially confusing in the text. Where it was relevant to quotes used, parent's words are instead contextualised with their child's age and level of functioning.

4.2.7 Procedural ethics

Ethics approval was granted from the University of Melbourne (project ID 1646922.1) in mid-September 2016. There were three main ethical issues to consider in this project: 1) ensuring consent from child participants was freely given and understood, 2) discussions about QOL might bring up distressing issues for some participants, and 3) that those invited to take part did not feel pressured to do so due to the affiliation between the Royal Children's Hospital and the Victorian Cerebral Palsy Register (VCPR). Protocols were developed to reduce these risks.

Both child consent and distress protocols were informed by other work in the Jack Brockhoff Child Health and Wellbeing Program. Parental consent was required but not

sufficient for all child participants. Children who were self-reporting were required to provide verbal assent before participating, and this involved confirming that they understood what participation would involve, and that it was entirely voluntary. A Plain Language Statement explaining the research in age-appropriate language was prepared for child participants, and parents were present during the child consent process to provide additional assistance to children if necessary to ensure the child understood what research participation entailed. All participants were advised that participation was voluntary, and they could stop the interviews at any time should they wish.

Following the planned distress protocol, if participants appeared to be distressed by the subject matter of an interview, the researcher reminded them that they could stop the interview at any time, or take a break, and asked if they wanted to continue. Parents were present or close by for all child interviews and were able to support their child as required. If children raised topics which appeared to be distressing for them, such as experiences of bullying, they were also asked if they were able to talk to their parent or another adult about them. In all cases, the children had done so and continued to seek support from other adults in their lives.

A page of support services for children and parents, such as Kids Helpline for children, Cerebral Palsy Support Network, Association for Children with a Disability, and other family support groups for parents, was developed should they require further support during or following the interview. This was separate and additional to the study Plain Language Statement with researcher contact details. At no time during the interviews did participants express significant distress such that additional support services appeared warranted. This approach to potential participant distress was explicit in the ethics application submitted to the University of Melbourne ethics committee and approved as part of the protocol. However, in most interviews both children and parents raised subjects that could be distressing, such as experiences of bullying and social exclusion, or parental trauma at the time of an infant's diagnosis of CP. As participants met with the interviewer face-to-face on only one occasion, it was possible that they experienced distress following the interviews and required follow-up support from other services. For example, during some parent interviews parents

spoke of the period of time of their children's diagnosis as a time of trauma, and suggested it was distressing to recall. Therefore, future protocols using similar methods with this population should consider providing support information to all participants with the PLS, and highlighting them in the event of participant distress during the interviews.

Although potential participants were contacted through the VCPR, they were informed in the PLS that participating or refusing to participate would not impact on services that they receive from the Royal Children's Hospital or any other health service. Participants were informed of the confidentiality of their responses prior to interviews. The recruitment protocol was developed with input from the manager of the VCPR, following procedures that had been used successfully for other studies.

4.3 Summary

This chapter has described the methods to investigate a conceptual model of QOL for children with CP. The qualitative methodology is well suited to answering questions of 'what' and 'how', while constructivist grounded theory allows researchers to build a model of the phenomena under investigation. Through the iterative process of analysis, the constructivist grounded theory method allows participants to shape not just the data produced, but the direction of later research interviews. This matches well with QOL measures' emphasis on the perspectives of respondents, in this case, children with CP and their parents.

Therefore, open-ended interviews were conducted directly with children with CP and their parents and analysed following constructivist grounded theory principles. Interview guides loosely addressed QOL domains raised in previous research but were flexible to address any topics raised by participants concerning their or their children's wellbeing. Interviews with children followed a 'child-friendly approach', utilising multiple strategies including drawing, and props, within a verbal interview structure. This approach has previously been found by other researchers to facilitate participation of children with disabilities in research, and to lead to additional insights on research questions. However, to date there is limited evaluation of the use of this approach for children with disabilities.

The following three chapters present results of these interviews, respectively: sample characteristics and reflexive critique of the implementation of the child-friendly approach, presentation and discussion of the key conceptual categories of the child interviews, and key conceptual categories of the parent interviews.

Chapter 5: Implementation and appraisal of child specific interview methods

This chapter continues from the methodology chapter by discussing the implementation of the child specific interview strategies and reflecting on the influence they may have had on data production and interactions between the researcher and participants. The appraisal of the child specific methods provides further information on the data collection process, and analysis of the interview interactions, to provide context to the main conceptual findings presented in the following chapter.

In the first section of this chapter prior to reflecting on the methods, the sample and interview setting are detailed, for both this chapter and the following main child findings chapter. The second section reflects on the implementation of the child interview strategies. This includes both practical considerations of introducing strategies to the child interviews, as well as the flow-on effects on children's engagement and potential influences on data production.

The third section follows recommendations in child interview methods literature (J. Potter & Hepburn, 2012; Rapley, 2012; Westcott & Littleton, 2005) by analysing the interactions within the interviews, between children and their parents, and between children and the interviewer. It concludes with reflections on the insider-outsider dynamic between a child with a disability and an adult interviewer without disability, and the impact this may have on children's disclosure of disability-specific experiences.

The chapter concludes by discussing links between this critique and existing methodological literature of child interviews, with implications for future work in this area.

5.1 Sample and interview setting: child interviews

All interviews were conducted in the participants' family home. Eighteen families were interviewed; eight included a direct child interview. For each of these eight families, parent and child interviews were scheduled for the same day and occurred

consecutively. In all but one, the child was interviewed first; for one family the parent was interviewed first. Regardless of order, the child left the room for the parent interview. Child interviews ranged in length from 34 to 71 minutes (mean 52.5).

5.1.1 Parent involvement

For all eight children, their mother was their primary caregiver. Therefore, each child's mother responded to the research invitation, scheduled the interviews, and provided parental consent. In one case, the child's father also joined the parent interview following the child interview. Mothers were present at the beginning of each child interview to provide consent, but only three chose to sit with their child throughout the interview; two others attended part or most of the interview while engaged in other tasks in the room; three were occupied in other parts of the house and did not appear to follow the conversation. The child's siblings were also present during three of the child interviews, though only one sibling contributed to the interview conversation—primarily by interpreting his brother's speech.

5.1.2 Characteristics of child interviewees

The children were almost evenly distributed over ages nine to 12 years, with a mean of 10.6 years. There were six boys and two girls. The children's severity of CP motor impairments varied between GMFCS levels I to III (two each at levels I and II, and four at level III). While two children had just one limb affected, three were regular wheelchair users and another used a wheelchair at times; half of the children were unable to walk without some form of mobility aid. Two children had confirmed intellectual disability, both classified as mild (IQ 55 to 70)—neither was able to read independently. One other child had significant attentional problems that caused learning difficulties at school resulting in a lower than expected literacy level for his age group. During two interviews it emerged that the child experienced significant anxiety requiring the support of a psychologist.

All the children were able to communicate verbally, though speech production was affected in three children. Two children had some slowing of their speech, which did not significantly impair their communication. One child had more significant speech impairment including challenges in pronunciation making him difficult to understand.

During the interview this child used a communication program on an iPad to produce written text to clarify some words not easily understood by the interviewer; a younger sibling who was present also interpreted this child's speech on a few occasions.

5.1.3 Family characteristics

Seven of the eight children had at least one sibling (either older or younger), including one with a fraternal twin without CP. Six of the children's families included both parents living in the family home. One family had a shared custody arrangement between the parents, with the child living primarily with mother and stepfather. One child lived with his mother and two younger siblings and had a noncustodial relationship with his father.

All eight families lived in metropolitan Melbourne ranging in location from affluent neighbourhoods close to the city centre to outer suburbs of lower socioeconomic status (SES). In respect to children's experiences discussed in their interviews, parents and children reported varied access to schooling, health services, recreation opportunities in their local community, and disability-specific recreation services based on both geographic and financial considerations. All the children attended school local to their homes. Seven children attended mainstream schools, either public or private; one attended a special school for children with intellectual disability. All the children at mainstream schools received some level of individual assistance at their school through government funded aide staff. One child attended a private mainstream school specifically to receive greater support than was available at the local public school, despite the greater financial burden of private schooling. All families relied primarily on publicly funded disability support through governmental health and education services, although the type of publicly funded services varied according to location and child's severity of CP. Some families could afford additional private support—primarily allied health and/or in-home aides; for others this was not possible.

None of the families were asked initially about their ethnic background, but it emerged in three interviews that the child's parents were recent migrants to Australia. For two children, both parents had emigrated together; one child had an Australian-born mother and father born outside Australia. In all three of these families the parents had

emigrated as adults from non-English speaking countries, however English was the first language of their children. Parental migration was discussed in respect to visiting extended family living outside of Australia; ethnic and cultural background was not otherwise either raised by children or explicitly discussed in the interviews.

5.2 Reflections on implementation of child specific interview strategies

Before considering an analysis of the interactions and content of the child interviews, it is necessary to reflect on the implementation of the child-friendly approach in the interviews. This section describes in three stages how the child-friendly approach was applied during the interviews: 1) introductions and consent, including parent collaboration; 2) implementation of specific strategies and flexible use of multiple methods; and 3) influence of the strategies on the interview process. As part of the reflexive critique of this chapter, each section describes and critiques the approach used in the interviews and then draws comparisons to relevant literature.

As detailed in the previous chapter (see Section 4.2.6 'Use of pseudonyms', page 106), individual children discussed in this and following sections are referred to by pseudonym.

5.2.1 Introductions and consent process

The early part of the interviews created the initial context and set up for the data collection process. There were two steps to this process relevant to child interviews. Firstly, parents were contacted by phone to arrange the interviews and gather initial information on their child to tailor the strategies used. Secondly, interviews in the family home began with child introductions and gaining informed consent from parents and children. The way that these steps unfolded and reflections on this process are discussed here.

5.2.1.1 Parent collaboration and introductions

Parents necessarily acted as gatekeepers to children's participation. The initial approach to recruit families relied on parental interest, as well as parent's judgement

of their children's ability to complete an interview; children did not have the opportunity to participate unless their parents first consented. While parents were provided information based on the research Plain Language Statement (PLS) prior to the interview, their understanding of this, or how and what they conveyed to their child prior to the interview was unknown. The protocol approved by University of Melbourne Human Ethics Committee relied on written informed consent from parents. Once parental consent was provided, verbal agreement (assent) was considered sufficient for children's participation, with an option for written consent from children able to provide it.

One parent interested in the research project completed an interview herself, but after discussion with the researcher, ultimately declined consent for her child to be involved. Although the child was old enough to participate and fully capable of a verbal interview, the parent reported that the child experienced a high degree of self-consciousness related to her CP to the point of refusing to discuss it except with a very few trusted people in her life. This mother reported that when asked by others about her physical impairment, her daughter usually told others that it was an injury, rather than using terms such as disability or CP. Therefore, the parent concluded that her child was unlikely to want to participate, and to ask her to do so would cause unnecessary distress. In this case the parent clearly articulated the decision process behind declining to ask her child to participate. However, the process of parental decision-making for child participation is often largely invisible and may be influenced by other aspects of child rights besides a right to participate, such as the right to protection from distress.

Beyond establishing child consent, parents also acted as gatekeepers in providing background information about their child. Background information about the child was intended to inform the use of specific strategies within the interview. Following the parent collaboration approach used by Teachman and Gibson (2013), parents were asked some brief questions about their child's interests and communication abilities during the phone calls to set up the interviews. However, there were a few drawbacks to this approach. Without having met the child, it was difficult to ask specific questions beyond the child's general interests and communication abilities. Phone calls with

parents were mostly unscheduled, meaning some parents had limited time to talk and preferred to simply make an appointment rather than discuss the research or interview protocol. Many of the parents were familiar with participating in research using conventionally structured interviewing formats or written surveys. It is therefore likely parents did not understand the context of questions about their child, and this was difficult to describe in the initial phone call. Again, the researcher was necessarily guided by parents in these discussions. During early interviews, questions to parents began with asking about child interests, such as whether they enjoyed drawing. As it was difficult to explain the non-traditional interview format to parents over the phone, this often did not lead to the type of information useful to deciding on child strategies. Therefore, later phone calls with parents focused on general parental judgements of the child's ability—such as “do they need any accommodations for the interview?”.

There is also limited practical guidance in the literature regarding the sensitivities of asking parents about their child's abilities and parental practical considerations, though a few have discussed the influence of parents or other adults who are present in child interviews (e.g. Pyer & Campbell, 2013). Parents are usually highly knowledgeable about their children's abilities and have acted in many situations as a bridge for their children to participate in a wide variety of activities. However, they may also be sensitive to suggestions that their child requires special treatment or may be unclear how these questions relate to their child's ability to participate in a research interview. Asking “Can your child read easily?” before meeting the children is straightforward and easily understood, but other questions about children's general abilities or interests are often not as clear, as children's abilities and impairments resulting from CP differ significantly. Questions which refer to specific abilities or aspects of impairment are likely to be more useful than applying a general approach to every parent and child. However, as these questions were usually asked in the first phone call between the parent and the researcher, prior to meeting either parent or child, there is a limit to how much the approach can be tailored without creating a further burden to parents or requiring another appointment for a separate phone call. This may have limited how much the child-specific methods could be adapted in the

current research, though it may also have ensure a greater degree of flexibility, as techniques could not be planned or tailored ahead of time.

One final point worth discussing with parents prior to child interviews is the way in which their child understands and talks about their condition. Throughout the interview process, it became clear that some parents avoid using terms such as CP, disability, or impairment, or have particular words or ways of discussing these with their children. This directly affects how children respond to questions about their disability. For younger children and children with mild impairments, terms such as cerebral palsy or disability were often largely meaningless. For example, one young child referred to having one weak side and did not understand the phrase ‘cerebral palsy’. Therefore, sensitive and respectful discussion with parents about how they discuss CP with their child is warranted prior to the child interview, to increase children’s ability to fully communicate their experience, improve chances for the researcher to build rapport with children, and provide richer data around children’s understanding of their condition.

5.2.1.2 Child consent process

The first introduction between the child and the researcher in all cases was the face to face meeting in the family home. All of the parents had discussed the interview with the child ahead of time, however it was unclear how much detail was passed on to children. The initial face to face introduction to the child and the reason for the researcher’s visit was led by the parent. Both the parent and the child sat with the researcher to complete the formal consent process prior to beginning the interview. In all the child interviews, this involved only the mother as the primary caregiver, though one child’s father was present during the latter part of the interview. Both parent and child were provided with the research information sheet included in ‘Appendix A: Information sheets and consent form for child and parent’ (see page 326); the child was provided the child-friendly version, and asked to read it through before signing the consent form. Both parents and children were told they could ask any questions about anything on the information sheet that was unclear before consenting—at least one

child did so. Audio recording did not begin until after the consent process was complete, so child or parent queries were not recorded.

For five of the children—older children with good reading comprehension—this consent process presented no problems. They generally took this task seriously, reading through both pages before signing the consent form. While a few of these children noted that they “don’t really have a signature”, they nevertheless happily provided their name and signature on the consent form. Two of the children had motor impairments affecting their handwriting but were willing and able to provide written consent. If the sample had included children with more severe impairments, written consent directly from the child may not have been possible for more children, though the approved ethics protocol only required verbal assent from children. Phelan and Kinsella (2013) had similar concerns working with children with physical disabilities and suggested that future research include a check box or sticker option for children for whom writing was laborious.

For the two children with confirmed intellectual disability, their mother immediately said they would be unable to read the information sheet, and she would consent on their behalf. Therefore, neither of these children provided written consent, instead their mother wrote their name in the space provided on the consent form. In this case, children’s consent was assumed from the parent conversation and they provided verbal assent to begin the interview. For a third child with learning disabilities, it was unclear how much of the written sheet he understood, though his mother offered to read through it with him. He wrote his first name in the space indicated on the consent form, and verbally assented to the interview. Early in the interview, he asked what the audio recorder was—suggesting that he at least did not understand the section on the information sheet stating that the interview would be recorded, and what would happen with the data—though he felt comfortable questioning the research process.

Reflections on the consent process led to two main conclusions. Firstly, engaging children in a written consent process increased their engagement and appeared to help break down unequal power structures by explicitly respecting their agency and right to choose their participation. Similarly, asking children during the consent process

if they wanted a final research summary and where to email it may have helped to engage them in understanding what the research means and what happens to the information they shared. One parent asked the child what email address he wanted a research summary sent to—this recognised that the research summary would go directly to him and not through his parents. By beginning the interview with an explicit recognition of the child’s agency, this may have also prepared them to question the process during the interview or refuse questions they didn’t wish to answer, reflecting more equal participation. Ongoing consent (described by Block et al., 2013) may have been beneficial though less intuitive to manage in a single interview. The interviews ended when children indicated they didn’t have anything else to say, or otherwise if they were no longer interested in participating.

Secondly, while written consent is standard in research practice, is it problematic for people with low levels of literacy. For children who cannot read, even if they are able to provide verbal assent, it is not likely to be similarly ‘informed’ if research information is provided in written form or is provided by the parent prior to the interview from the research invitation. Therefore, for these children, it was necessary to alter the planned process and inform them verbally about the study and allow them to assent prior to the interview. Furthermore, in these situations, parents play a greater role as gatekeeper in preparing children for participation and providing parent consent, and more fully mediating the child’s consent. To be fully inclusive, these children should have the same opportunities to engage in informed consent or assent as those able to read and sign the consent form. This involves not only preparing research materials and approved protocols to be inclusive, but also being flexible and reflexive to what happens in practice at the family home or in the interview setting. Similar reflections from other published research and implications for future research are described at the end of the chapter, Section 5.4.

5.2.2 Implementation of multiple methods

This section focuses mainly on the practical implementation of specific methods within the interviews. The child interviews included the use of child-friendly interview strategies including drawing, emoji, and question and answer games (discussed in the

previous chapter, Section 4.2.4 page 100). Successes and challenges in the use of the child-specific interview strategies are discussed, including decisions made during the interviews and how child-led interviews contributed to the use of some strategies above others. The section concludes by discussing how the use of multiple methods both allowed for and required flexibility to implement.

5.2.2.1 Warm-up activity

The first planned activity in the child interviews was designed as a warm-up rather than data collection exercise, and is described in the previous chapter, 'Introductions and warm-up activity' (Section 4.2.4.2, page 101). In this activity, the child was shown four cards printed with questions which they could choose to ask the interviewer, often introduced by the interviewer explaining "it's not fair for me to ask all the questions". A scan of the cards used is presented in Figure 1 on the following page; the actual size of each card was approximately 12cm by 7cm. The aim of the activity was to act as an ice-breaker, reducing child discomfort at being the focus of interview questions, and breaking down power imbalance by allowing the child to choose a question to ask and take on the interviewer role. Although this activity was awkward for children who were not necessarily interested in the interviewer disclosing details about themselves, it generally served the purpose of reducing child discomfort. The use of self-deprecating personal disclosure from the interviewer helped to shift the power imbalance and usually resulted in mutual laughter. Laughter and humour helped with engagement for most children, especially at the expense of the adult in the interview. Previous qualitative researchers have described reducing the expert status of adult interviewers to improve rapport with children (Darlington & Scott, 2002); self-deprecation in the warm-up activity provided an early opportunity for this.

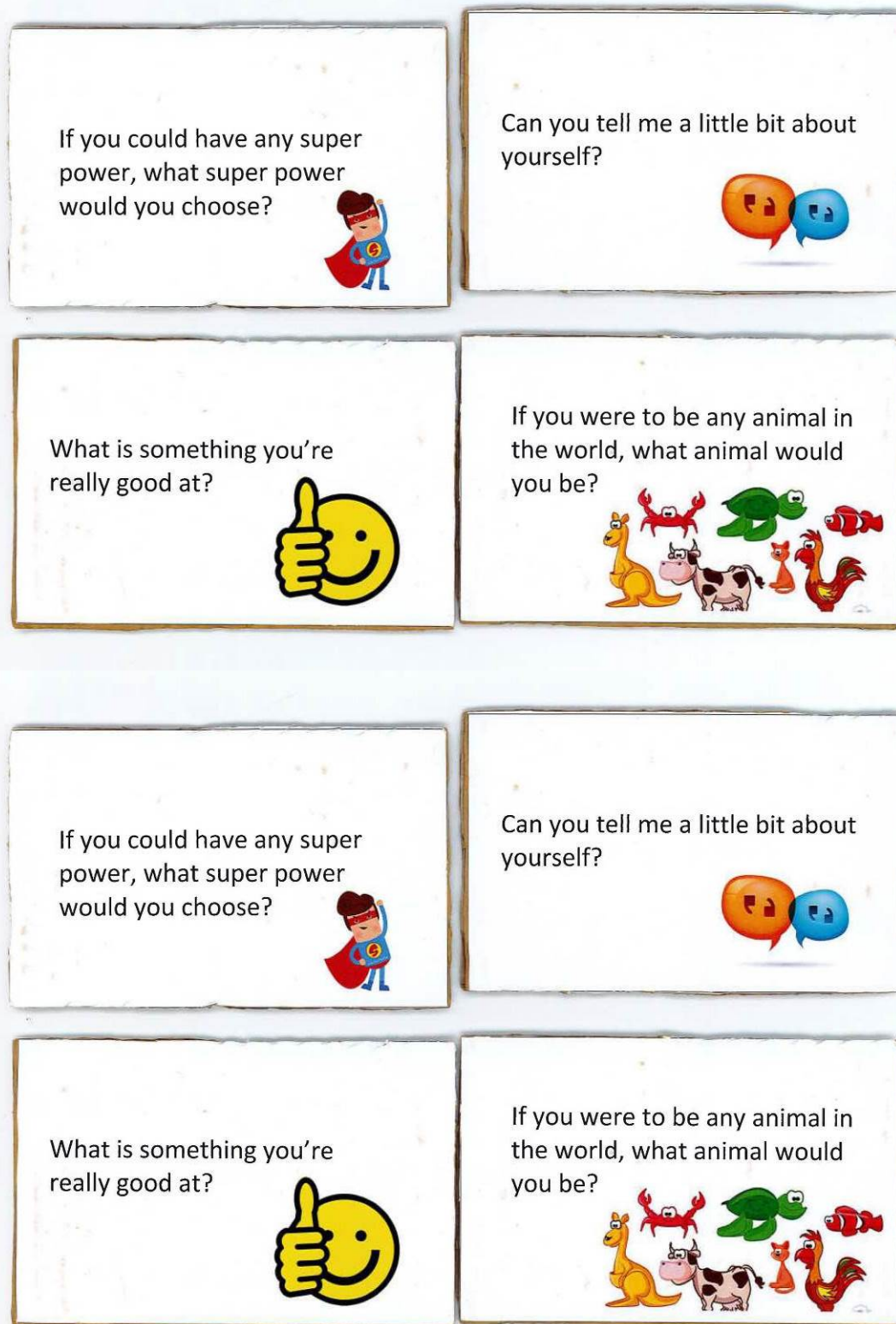


Figure 1. Printed cards for child ice-breaker activity

The main drawback of the warm-up activity used here was the fact that it relied on children being able to read the question cards. In two interviews, the parent was

present and assisted their child to read the question cards, but for one child who did not have sufficient literacy, the emoji were used instead as an icebreaker and did not provide the same benefits as the question cards. Child engagement was also reduced in children who relied on parental support to read the question cards compared to those who read independently. Future research using a warm-up activity should employ a similar activity that does not include reading, or an alternate way of employing the question cards that allows children with lower levels of literacy to participate without relying on adult support.

5.2.2.2 Emoji

Emoji were used in every interview because they were universally acceptable and familiar to children and could be used and interpreted in multiple ways. Children were eager to engage with them and did not require strong literacy skills to do so. Questions could be adapted, including asking children what they thought particular faces meant, for example those with expressions not as easily interpreted as ‘happy’ or ‘sad’. The actual emoji cards used are included below in Figure 2; the actual size of each emoji was approximately 8cm by 8cm.

The question ‘which one is most like you’ generally did not lead children to detailed answers, and they mostly immediately chose a ‘happy’ emoji. However, further probing usually led to children exploring temporary emotional states that they experienced related to the other emoji, and when or why they might feel them. One spontaneous addition to the original strategies came in adopting the emoji later in the interviews to discuss feelings, generally or specifically. When asked later in the interview about feelings they have, or how they feel about certain topics, they sometimes appeared at a loss as to how to describe abstract emotional ideas—the physical prompts provided by the emoji cards suggested options without seeming to restrict the conversation. Overall, the use of printed emoji was highly adaptable, prompted discussions of child identity, emotions, and emotional experiences, and was highly engaging for the children interviewed even when the original question did not lead to long responses.

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for copyright reasons.

Figure 2. Printed emoji for child interviews

5.2.2.3 Drawing materials and prompts

All of the interviews included drawing activities as an interview strategy, with both the child and the interviewer drawing. The way the drawing was incorporated into each interview varied with the child's interest in talking, and their interest and willingness to draw. Therefore, as a strategy, children's drawing included both spontaneous non-

directed drawing (i.e. they drew whatever came to mind) and prompted drawing such as “draw something that makes you happy”. Two children invented drawing games, requiring the interviewer to guess what their drawing was; one child then insisted on swapping the roles, so that the child and interviewer took turns to draw or guess the subject of the other’s drawing. This then led to the child directing the drawing related to the topic of the interview, such as “Let’s write down all our friends names” during a discussion about school friends. The four prepared prompt cards proved to be useful in suggesting drawing topics for those children who were initially reluctant to draw. The prompt cards are shown in Figure 3 below; the actual size of each card was approximately 12cm by 3cm.

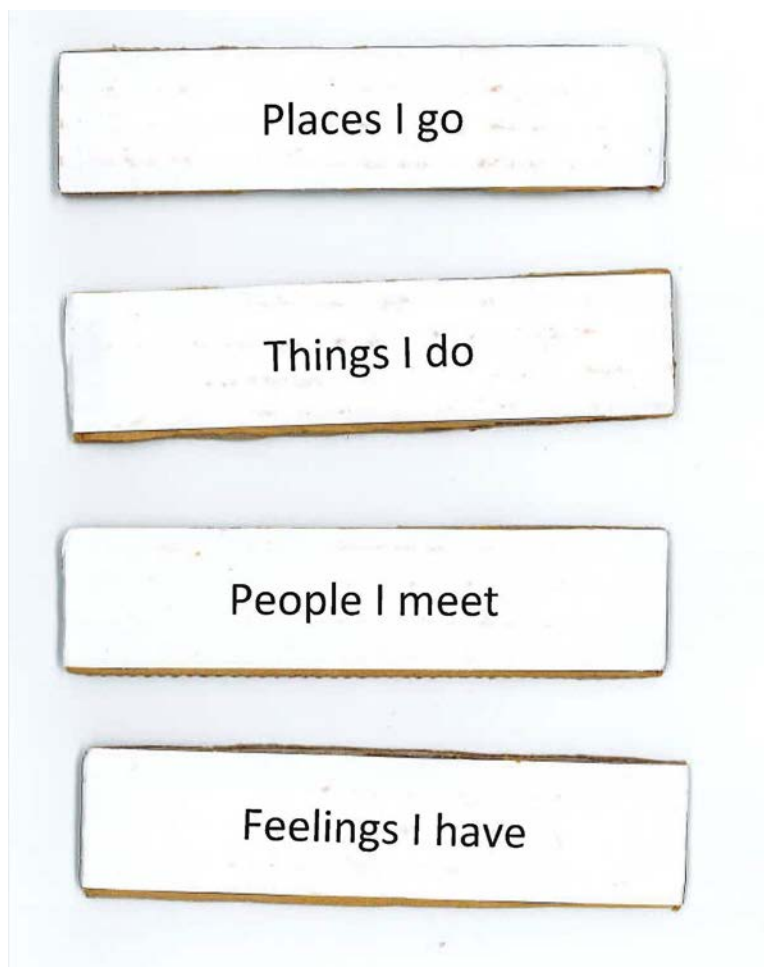


Figure 3. Prompt cards for drawing in child interviews

For a few children who expressed self-consciousness about their drawing ability, or were not overly interested, the interviewer acted as a scribe, drawing what the child described. Large drawing sheets (A3 size, 29.7cm x 42cm) could be quartered to

explore each area of the child's life indicated by the prompts. Two examples of these drawing sheets are shown in Figure 4 and Figure 5 below. In each case, the interviewer contributed the majority of drawings at the child's direction, with a few additions drawn by the child to explain particular details. Names have been redacted from the drawings to protect child privacy.



Figure 4. Prompted drawing from child interview

For those children with some impairment of fine motor control, the provision of printed materials backed by thick cardboard was helpful. The strategy of the interviewer saying, “what if I draw, and you tell me what to draw?”, made drawing engaging and accessible to all of the children, even children who did not enjoy drawing or did not wish to draw during the interview.

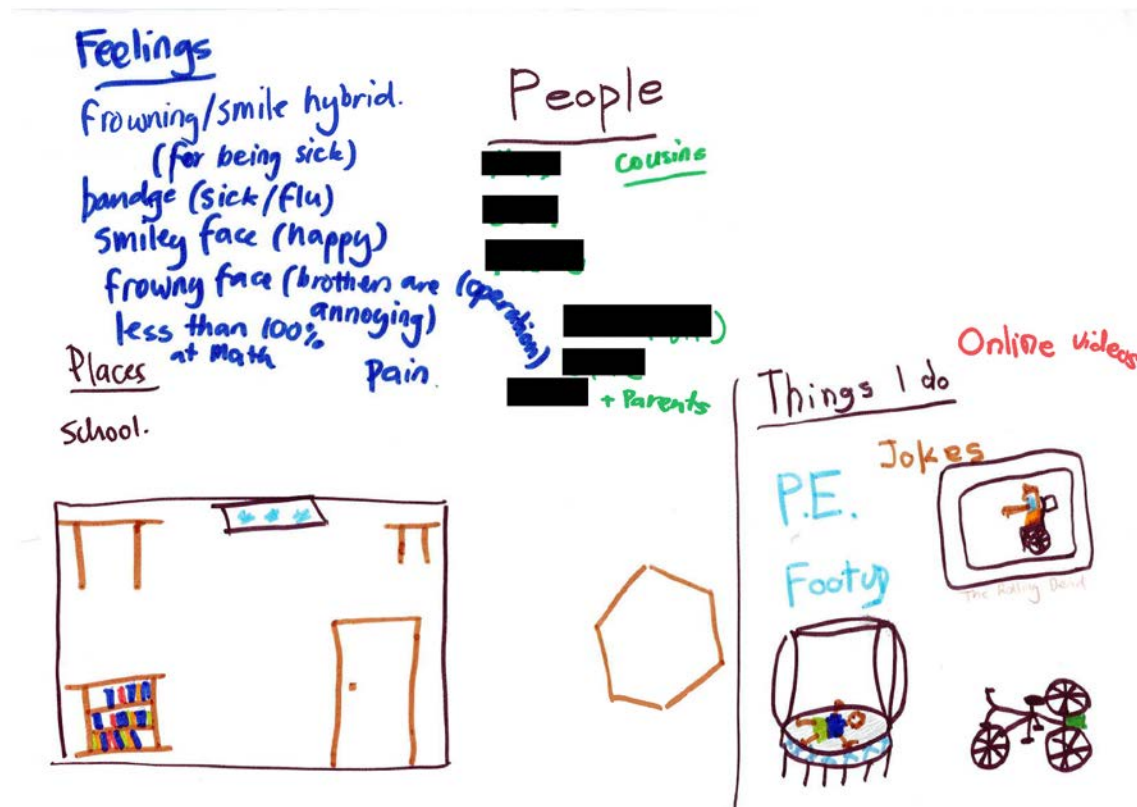


Figure 5. Prompted drawing from child interview

With the additional prompts or choosing between child or interviewer drawing, drawing was highly adaptable to individual children based on their initial engagement. Drawing was acceptable to all children due to flexibility in how the activities were used, and guiding children but allowing them to choose how they engaged. The drawings produced were not used in subsequent analysis, however, the act of drawing and explaining drawing provided topics of discussion for the child.

5.2.2.4 Flexibility in use of multiple methods

The implementation of multiple methods within the interviews required flexibility. This was related in part to parent collaboration, but also due to the number of planned strategies: not all strategies were intended to be used in each interview. Therefore, the

use of multiple strategies required flexibility dependent on child engagement. Ultimately, two types of planned strategies were not used in any of the interviews. These were prompts using printed pictures of places children frequently go such as parks, playgrounds, and classrooms, and prompted stories using cartoon images of children with visible disabilities. In the current interviews, pictures of children's places were largely unnecessary, as the children engaged instead with drawing, the written prompts, or verbally, in discussing their everyday activities and places they visited. Due to the increased time these activities added to the children's interviews, it would have been counterproductive to introduce another activity after drawing proved highly engaging in the first few interviews.

The story activity with cartoon images of children with disabilities was not included for different reasons. While it was not introduced in the first couple of interviews due to time constraints, these interviews also raised the need for sensitivity as to how children discussed their impairments. Due to the limited nature of parent collaboration, it was challenging to produce an image tailored to the child's disability, as used by Teachman and Gibson (2013), while also acknowledging how the disability was discussed in the family. The idea of writing a story "about someone like you" was introduced verbally in one child interview, and the child responded negatively, with decreased interest compared to more detailed responses to other questions. The combination of these challenges meant that this activity was last to be introduced, and ultimately, time constraints meant that it was not implemented in any other interviews. A few of the children discussed writing stories as an enjoyable school activity, suggesting this form of activity would be easily understood by children. However, given that drawing children's lives with the simple written prompts lasted the length of an interview when this activity was used, creating a story may be better suited to an interview that does not include other activities.

Age appeared to make limited difference in engagement with the various activities among the children interviewed, however, it did have some impact. The older children (11 to 12-year-olds) expressed greater concern over the quality of their drawings, and their lack of drawing skill, than younger children. This may have contributed to lesser interest in general spontaneous drawing, though they were still happy to engage with

prompted drawings especially when the drawing was completed by the interviewer. It is worth considering that older children are more likely to express preferences for or against specific creative activities—most younger children are still regularly encouraged to engage in drawing through their school, while by the time they are entering high school, drawing is often considered a specialist pastime only engaged in by particularly artistic children. This parallels children's increasing concerns over the skill in their drawing, which was more evident in those who were older. Concern over the quality of drawing may also be related to children's fine motor impairments affecting use of pens and pencils, but this was not stated explicitly by any children.

Gus, a child with communication impairment affecting his speech production, adopted his own strategy throughout the interview to describe particular experiences. Early on in the interview, his mother brought him his iPad and reminded him to use his app for typing words to help the interviewer understand him; she also commented to the interviewer that Gus preferred not to have to use it. Whilst Gus relied on his own speech and occasionally his brother's interpretations for most of the interview, he did at times type out individual words that the interviewer struggled to understand. With his iPad to hand, he was able to share other information—including videos of a museum that he visited during a school trip, and a theme park ride that he had particularly enjoyed. In contrast, some of the other children were using iPads or electronic devices on the interviewer's arrival and these were removed by their parents. However, this way of sharing information and experiences was already very familiar to these children—all of them at some point discussed using devices as a part of their everyday lives. Gus's use of his iPad during the interview not only allowed him to show his experiences without laborious and difficult spoken explanations but presented a natural display of ways he has adapted to communicate with others. This suggests that excluding technological options in strategies for child-led interviews, even in those who do not regularly use alternative communication methods, may risk missing out on information about all the ways in which children now interact with others in their lives. This may be especially true for children with more severe disabilities and communication impairments who are more comfortable using alternate means of communication, including the use of iPads or other devices in ways

that don't include formal communication software. For most parents, removing children's access to an iPad or smartphone prior to beginning the interview appeared to be a default action. Therefore, if these are desired within the child interview, this may also require parent collaboration.

The use of multiple strategies allows for eliciting different types of information and being able to choose activities and means of communication to suit each participant. However, it may be bewildering to participate in multiple games over a short period of time; and most children were happy to spend 30-40 minutes drawing and chatting. In general, these strategies take more time in the interview than just asking questions even when they are limited to a single activity. The act of having a distracting task seemed to ease children into the discussion, especially where the topic was emotional. The potential influence of the strategies is described in the following section.

5.2.3 Influence of child friendly approach

There were two main aims of how the child-specific strategies would influence the interview process. The first aim was to increase rapport building and child comfort and engagement, in an effort to make it easier for the children to share information. The second main aim was to increase data production by providing alternate ways to discuss topics relevant to QOL, elicit additional specific details in children's responses, and to explore the many different areas of life that influence QOL. This section critically examines the influence that the strategies had on these two parts of the interview process.

5.2.3.1 Rapport building and child engagement

While different activities began with varying goals, they did not always proceed as expected. The first surprising finding was the immediate and universal enjoyment children took in using the printed emoji. For some of the interviews, this activity improved engagement more than any other. Similarly, the unintended effects of the interviewer participating in the drawing activities led to far greater rapport building than expected. Children became more engaged with both drawing and discussion, perhaps because it reduced their self-consciousness of talking about their own drawings.

In general, children seemed to appreciate having an alternative activity besides talking face-to-face. When difficult topics such as bullying or anxiety were raised, they focused on drawing or another activity while talking, with reduced direct interviewer eye contact. This may have allowed children to distract themselves from the distressing subject matter or their own embarrassment while continuing to engage in the conversation. Notably, the instances where this happened were all later in the interviews, after the breakdown of the child's initial reserve, and after mutual participation in the drawing and other activities had led to an establishment of rapport—shown by greater eye contact, and more detailed responses and spontaneous conversations from children. It is possible that it was the combination of the established rapport and the possibility of distraction with another activity that allowed children to open up about these difficult topics.

There was a need to balance structure with flexibility which could be tricky to manage. For example, one child with focusing and attentional challenges came up with his own drawing game rather than following interview prompts. He became engaged through this activity but continued to not engage with the majority of the interview questions. However, he spontaneously offered information part of the way through the interview, and at that time was willing to discuss some topics related to his everyday activities. In this case, the combination of attentional difficulties and lack of structure worked against production of the data planned from the interview, but also offered the child the opportunity to demonstrate his own agency and to some extent subvert the interview agenda. Most children were willing to take direction and engage with any activity, though they showed preferences for some types of activity. When activities also involved interviewer engagement, and directions were offered as a possibility, it seemed to lead to an easier establishment of rapport.

Children benefited from the establishment of structure, especially in the limited timeframe of the single interview format, as they were generally unlikely to take the lead in directing the interview. While the ideal is to allow the interview to be child-led, without any guidance children were unwilling to engage, because they did not seem to understand the purpose of what they were doing and were wary as to what might later be required of them. This may explain why the initial warm-up activity at times seemed

to be tolerated rather than enjoyed: it was a false shifting of power in that the interviewer was still directing the conversation and could control their level of disclosure. In contrast, children might think they were expected to answer any question they were asked, without actually achieving any power to direct the interview. Again, changing the balance of power is probably easier to establish over multiple interactions—with a single interview, greater thought and effort needs to be put into how the interview itself, and the activities within it, are introduced as well as emphasising that children can refuse to answer anything they wish, or end the interview at any time. Alternative approaches could include altering the interview structure entirely and telling children that they are in charge (after laying out what the interviewer wants from the interview) or providing other physical props (such as a speaking stick). These changes could lead to a much more child-led format, with real engagement and shifting of power.

The benefit of shifting of the power balance and the use of the ‘dummy interviewer’ (Darlington & Scott, 2002) method was demonstrated as well in the drawing activities. For example, demonstration of the less than technically perfect drawing abilities by the interviewer reduced the interviewer’s default power and expert status, often humorously, and allowed children to assume a position of either reassuring the interviewer or expressing a sense of critique or superiority. For children who expressed self-consciousness or doubt over their own drawing abilities, this provided an additional bridge to rapport-building and a more level playing field in the conversation. Similarly, when the interviewer was drawing on behalf of the children who were less interested in drawing, correcting the interviewer’s drawing increased children’s immediate engagement in the activity, both in establishing their greater knowledge or competence, and in specifically correcting the details depicted in the drawing. This had the added effect of demonstrating how increased child engagement in the activities improved the interpretation of the data produced.

Overall, the strategies provided additional ways to engage children in discussing their lives, especially their emotions. They were generally all able to participate in the activities used, with or without adaptations, and several expressed their enjoyment of them. Children’s engagement in both the activities and the interview increased with

improved rapport. This was shown through increased eye contact from children, more frequent and more detailed responses, and more frequent spontaneous conversation from the children. Humour was generally appreciated by most children. Therefore, the use of these strategies not only seemed to make the interview easier for the children, but with some practice, also made the process much easier and more fun for the interviewer. Lastly, it is worth noting that interviews probably lasted a lot longer than planned (often up to one hour each) by including the activities—though they also held children’s attention longer. Again, if multiple interviews are not possible, future researchers considering these strategies would need to take note of the consequently longer interview time and plan accordingly—in the case of school evening interviews, it did at times mean the ending was rushed, as by the time the parent interview was concluded the entire process lasted over two hours.

5.2.3.2 Data production

Within the broad range of topics discussed by children, and in the subsequent analysis of the transcripts, it appeared that the use of specific strategies added to data that was collected compared to what would have been produced using a traditional interview format. There were benefits particularly in terms of the way the various strategies facilitated conversations and subsequent analysis.

Emoji provided a convenient way to discuss emotions and to reflect on self and self-image. They did not work for all children, but for most children reflections on feelings were directly prompted by the use of emoji, and it is likely that they would not have been elicited without this activity. Furthermore, by leading into this (enjoyable) activity early in the interview, the practice of reflecting on emotions was introduced, which may have made it easier later in other discussions for children to say things like “It made me feel like...” or “I’m not generally like that because of ...”. The emoji activity also helped to demonstrate fairly clearly the ways in which children choose to think of themselves, and to present themselves holistically to others. They were able to begin by showing a positive self-image, as most of them immediately chose the happy face emoji as most representing themselves. Having established the positive self-identity,

they could then point to specific instances of things that were more negative or troubling, without exposing an immediate threat to self-esteem or self-identity.

Interestingly, asking children to reflect on themselves in this way—with the question “which one is most like you?”—also prompted analysis of children’s representations of self within the interview. This was an unintended consequence of the activity but informed findings regarding children’s identity, as well as analysis of the interactional processes. The activity originally was intended only to lead to discussions of QOL-relevant emotions. Instead, the additional application demonstrated the way in which alternate strategies within interviews can lead to new, unexpected directions in the research.

Drawings, both prompted and unprompted, may have led to greater discussion of children’s creative pursuits. About half of the children when being offered the drawing activity or during the time they were drawing spoke unprompted about how much they enjoy drawing or similar creative activities, and about engaging in these activities in their spare time. It is not clear whether this would have been raised in any case, as children were asked and spoke about the recreational activities and hobbies they enjoyed. However, the activity may have prompted more discussion of these mostly solo activities—children otherwise tended to refer to either school activities or other organised community activities, such as sports clubs. Furthermore, reflecting on the drawings that children produced, the way they spoke about them, and their humour and enjoyment in drawing added some dimensions to the analysis, and clearly demonstrated aspects of the analytical categories within those interactions.

Both prompted and spontaneous drawings provided triggers for children to discuss different areas of their lives. These activities may have raised topics that would have been discussed during the interview, but the prompted drawings had the advantage of providing an easy structure to check that the main areas of a child’s life relevant to QOL had been discussed.

Furthermore, the prompted drawings helped to explain subjects that were raised, check assumptions, and provided more details—by requiring clarification for how to draw the subject. This was particularly the case for drawings where the interviewer

drew subjects raised by the child, or the child dictated what the interviewer should draw. For example, one child said he did not like PE (physical education) at school, which was added to the drawing, but then led to further discussion about what this was, and then to questions about whether there was anything he did like that could be added to another part of the drawing. Using the drawing activity in this way to advance the conversation happened similarly when children were asked to draw something that made them happy, or something they enjoyed, and by allowing them to explain aspects of it while they drew. Undirected drawings did not tend to provide this function.

Pauses in the conversation while things were drawn or written down may also have provided ways for children to consider their answers further, as most needed to pause before answering questions. However, in conventional verbal interviews, there were often awkward pauses where the child seemed to feel a pressure to answer before they were ready to do so. This frequently led to “I don’t know”, “it’s hard to answer”, or “I can’t remember”, derailing the subject and potentially reducing the child’s comfort level in the conversation. Pausing for a drawing activity, with the interviewer otherwise occupied, therefore provided natural conversational pauses.

5.3 Analysis of interactional processes in child interviews

Moving beyond the focus on the use of the child-friendly strategies, this section discusses the analysis of interactions in child interviews, and the way these processes influenced the interview, or provided examples of conceptual categories discussed in the following chapter. The aim of analysis of the interactions is to elucidate three types of process within the interviews. The first process is the way an interviewer’s questioning and prompting guided the children’s responses and way of responding, as Rapley (2012) discusses. With the child interviews, parent-child interactions played a role in shaping these responses as well. The second interactional process is the contextual assumptions and identities that children create within the interviews. This is particularly salient for children with disabilities, who have been chosen to be interviewed primarily because of their disability. The way they choose to present their identity as a child with a disability, and to minimise or defend the place of disability

within their everyday lives, has direct consequences for what they say about the impact of disability on their lives. Lastly, the analysis of interactional processes considered the roles adopted by the interviewer and the child within the interview. These roles are relevant to power and the way that interview content is co-constructed. The analysis of roles within the interview considers both the interviewer's questioning style that may have influenced roles and how an adopted role may have influenced a child's response to the questions. It follows on from reflections on the use of the child-friendly strategies, especially how these shifted the power balance.

There were three main areas of interaction which appear in this analysis: 1) parent-child interactions, 2) presenting the disability identity to the non-disabled interviewer, and 3) challenging answers: hesitating to answer and changing answers. Each is discussed below in terms of the current interviews, and how it relates to literature in the field.

5.3.1 Parent-child interactions

While child interviews involved the direct questioning of the child, parents were also involved to a greater or lesser extent. Parents were necessarily present immediately prior to the interview to provide consent, whether or not they remained throughout the interview. Before the interviewer met the child, parents had explained the research project to their child. Parents adopted different roles with respect to how they supported their child in answering the interview questions, whether they chose to be present, to leave the room, to directly contribute, or to verbally encourage their child to answer questions. Where interview content reflected on the child's independence and agency, direct observations of the parent-child interactions showed helped to inform and substantiate the conceptual analysis by providing examples of the concepts in action, especially in terms of children's family relationships.

Within the interview setting itself, managing parent-child interactions was at times challenging. While it was possible to plan to solicit answers directly from the child and de-emphasize parent input, the reality was that the interviews were taking place in the family home, and ultimately, the parent held the authority for how the child participated. Even the presence of other family members, such as siblings, was largely

out of the interviewer's control. To insist only on the child's presence and input would have been inappropriate, or at least potentially damaging to rapport with both parent and child. Furthermore, while family members present in these interviews may have hindered child interviewing by providing distractions or limiting child responses, they were just as likely to help by providing support to the child. Parental interactions were either parent initiated, or child initiated, and each of these had different outcomes.

Parent initiated interactions related to either interview content, or behaviour control. Parents responded to interview content by sometimes responding for their child or directly suggesting answers if the child appeared to struggle for a response, or sometimes providing suggestions and encouraging the child to discuss them. In a few cases, the parent elaborated on a child's response, or provided context for an experience they had described. For example, Gus spoke about wanting to re-join an afterschool activity, and his mother, who was nearby but not present, jumped in to explain that he had been unable to do so because of a long period of recovery from illness. This type of response was helpful for the interviewer in providing additional details as to why or why not things had happened, but also frequently shifted the focus to the stories or details parents provided. Most children when interrupted in this way deferred to their parent's response—when this happened repeatedly, the child's feelings or knowledge of the issue were de-emphasised as the parent completed their response. In the present example, how much Gus understood the reason he was missing afterschool activities remained unclear. These parent interruptions were more likely to explain the parents' view on the interview process, and what they thought was required—valuable, but not necessarily the same as child views.

Conversely, parent-initiated responses were the most likely to result in negative child responses, compared to either statements made by the interviewer or parent responses solicited by the child. That is, children were most likely to challenge, disagree, or otherwise indicate that they thought otherwise to new information offered unprompted by their parent, despite a general tendency to agree with adult statements. It is unlikely that this was because the parent was most likely to make incorrect statements—rather it appeared to reflect how comfortable and safe a child felt in challenging an adult, or adult views. Parents whose role in the interview

remained more authoritarian—that is, more instances of behaviour correction—were conversely less likely to have their input directly challenged by their child. Children whose parents reflected a more egalitarian parental style, with more child-led interactions, were more likely to have their input challenged or corrected. This then sometimes led to an impromptu dialogue between the parent and child about the issue at hand, temporarily excluding the interviewer, but providing potentially richer data. Understandably, these interactions were also more likely with older children (11 or 12 years), while younger children (9 or 10 years) were more likely to defer to parent opinions.

Other than contributions to interview content, parent-initiated interactions served as behaviour control or correction. This at times involved simply encouraging the child to express themselves, or to respond to the interview questions. This encouragement could be purely supportive or focused on behaviour management. In a few instances, it involved admonishments to behave or comply with the interview process—such as to listen, or to put away a phone or iPad. Nicholas’ mother said a few times “You’re being silly” as a way of telling him he was misbehaving. In a couple of interviews, the parent returned from a different part of the house, and directly asked about the child’s behaviour in front of the child, such as asking if they had been talkative—in this context Sofia’s mother commented, “She can be a chatterbox”. Barring purely supportive encouragement, children did not generally respond verbally to this form of parental input. The interview otherwise took a child-led approach. Therefore, for children who were particularly distractible or displayed challenging behaviours, this type of parental authority assisted in continuing the research process. Ultimately, these interactions restored the status quo power imbalance between child and adult, and in doing so, potentially reduced children’s engagement and ownership in the interview process. In these cases, the child usually remained engaged in the activities, i.e. continuing to draw, while (temporarily) withdrawing from responding to interview questions. Especially in children with higher levels of behavioural challenges, researchers may face difficulties in balancing child engagement and child agency in research versus behaviour management and compliance with the research processes. Such challenges have been discussed by others in critiquing participatory methods in

child research, as these methods have a strong focus on child agency (Gallacher & Gallagher, 2008).

Parent presence during the interviews not only led to parental input or behavioural control. Some children also appeared frequently and understandably to derive comfort from the presence of their parent, and at times sought verbal reassurance or encouragement. Therefore, some of the parent-child interactions in the interviews were child initiated. Child-initiated interactions with their parents were usually ostensibly about interview content. That is, children sought responses from their parent to provide more detail, or because they were unsure of how they should respond. For example, after being asked what kind of things he did with his friends, Caleb turned to his mother to say, “Mum, tell them when I had to—the bell—where I sprinted today”. At times, children appeared to seek purely factual information because they could not recall or were unsure of specific details. At other times, children appeared to seek parental support because they were either unsure of the interview expectations, or what the ‘right’ answer was. These interactions not only demonstrated ways in which interview questions made children uncomfortable, but the manner in which the children engaged in help-seeking. Despite the presence of their parents, some children responded instead with silence, “I don’t know” and “I don’t remember” to factual questions about their everyday activities that they might be expected to easily recall—others directly sought their parent’s support.

The degree to which parents and children became involved in interrupting or reassuring each other probably showed relationship dynamics that were habitual, not just in managing behaviour, but in managing the child’s disability. Parent-child interactions, especially those that were child-initiated, demonstrated directly the connection between help-seeking and child agency. Children who appeared to experience more agency in their relationship with their parent had more confidence to seek parent support, as well as to disagree with their parent’s input. Where parents took a more authoritative role, with greater behaviour correction, children seemed less likely to seek parental support for interview questions they found difficult—even though their parents were as involved, and as encouraging, as others. Instead, they waited for their parent’s intervention or for the interviewer to redirect the interview.

Necessarily these dynamics are influenced by the personality of both parent and child, among other factors; it would be simplistic and unwarranted to conclude any level of causation between, for example, authoritative parenting and child agency. Rather, these interactions provide a snapshot view of how children with different levels of agency respond to situations in which they required help, as in answering difficult interview questions. The range of input provided by parents and sought by children suggested the impact that parental presence, or absence, may have in interviews with children.

5.3.2 Child responses to difficult questions

Throughout the child interviews, there were at times questions which children were unwilling or unable to answer. Although the specific questions varied with each child, the commonality was that of answers which were in some way challenging for children to give. In these situations, children variously responded with silence, looked to a parent for input, or in a few cases provided a nonsensical response—by saying yes or no to a question that required a different response. Similar responses were “I don’t know” or “I don’t remember”—both to factual questions and questions about the child’s own preferences. These could all be considered valid responses in themselves, but they provided no further information or possibility to continue the conversation.

There were some commonalities to these responses. Children with ID and/or learning difficulties more frequently refused to answer by providing no verbal response at all to questions. This was more frequent earlier in the interviews. Giving one-word answers or easy but at times nonsensical agreement was similarly more frequent earlier in the interviews. Both these types of responses seemed to be related to children being presented with a question for which they could not articulate an answer but feeling equally unable to ask for clarification or say they either could not or did not want to answer. In these situations, the researcher response of changing the topic or asking simple, closed questions sometimes resulted in greater child engagement. When parents were present, they generally intervened after one or two questions a child did not answer.

Later in the interview some children were more likely to respond instead with “I don’t know” or ask for clarity in the question. Both these types of responses seemed to be related to developing rapport and increasing child comfort. As children became more comfortable they felt more able to say they could not answer because they did not understand or did not know, rather than simply agreeing or not responding.

A related pattern that emerged was a change in children’s answers from the beginning to the end of the interview. That is, children responded easily “yes” to questions early in the interview, but later when responding to less direct questions on the same topic, clearly contradicted themselves. For example, early in the interview with Ana, when asked “Do you enjoy your classes at school?”, she responded quickly “Yeah, mm hm. But I don’t enjoy maths very much”. Much later, in the context of missing school because of medical appointments, she said unprompted “I don’t like school very much”. All of the children were happy to say early on that they liked school, either unprompted or in response to a question. However, two others beside Ana later said that they did not enjoy school. This pattern was repeated with other topics, though less clearly. Similarly, at times they changed their initial response to a general question almost immediately after being asked for more details.

Questions could be difficult for children to answer if they required factual content that children could not recall or did not know. Questions were more likely to be factually challenging for some topics and/or for some children. Topics were generally challenging because they required recalling specific or complicated details, especially for a new subject or where there could be many possible responses. For example, the interviewer introducing a new topic by saying “What are things that make you happy?”.

Children often did not recall specific facts about things for which they were not responsible. For example, while they were obviously aware of attending varied medical appointments related to their CP, how often they attended them or their purpose was generally left to parents to remember or explain. For younger children, and those with ID, these questions were even more difficult. Children with ID were more likely to respond with nonsensical agreement when they could not either understand the

question or provide an appropriate detailed response. Alternately, Zachary, who was 12 years old, articulate and engaged, regularly attended physiotherapy but had seen a few different clinicians. When asked for details, he said “I don’t have the best memory, so I don’t know how many physios”. Whether or not children with CP have poorer memory, it was a sentiment expressed by others to similar detail-oriented questions. Children with CP may be more sensitive to perceived limitations or more ready to attribute perceived deficiencies to themselves. This suggests that researchers may need to prepare for alternative ways of questioning for children with CP who find detail-oriented questions difficult to answer.

Other than factually difficult questions, some questions touched on topics that were sensitive or emotionally loaded and difficult for the child to discuss, which presented a different challenge for the interviewer. An example from these interviews was the three children who discussed experiences of deliberate social exclusion and bullying. In all three cases the topic was raised by the child themselves later in the interview, some time after they had answered earlier questions positively about friends or other social relationships. Even later in the interview when the children appeared generally more comfortable and open to answer, they responded to these topics with limited eye contact, closed body language, and were quick to change the subject when asked for more details. While bullying is obviously a distressing topic, there are other topics common for children with disabilities that elicited similar responses. All the children interviewed had noticed receiving more attention from strangers because of their disabilities or being singled out. They all at one time had missed out on recreational or social opportunities because of their disabilities. Similarly, some of the issues of identity and disability discussed in the following section (page 143) could be very emotive for children. All three of these topics are in some ways an everyday feature of the life of a child with a disability, and children were usually open and willing to discuss them. However, it was clearly harder for them to discuss earlier in the interviews when they were less comfortable or felt a greater need to maintain a positive self-image.

Lastly, some questions required answers which contradicted children’s general tendency to agree with adults and to be seen as agreeable. A perceived power imbalances in the interview likely contributes to this tendency. This may have been

exacerbated in some ways by having a disability; that is, because of their disability children may have a greater need to be seen as easy to get along with. In the context of disability, this is exemplified by Eric's reluctance to discuss aspects of having CP which he considered "annoying"—he preferred to maintain the image of not being bothered. Children who have milder CP tended to have part of their identity focused on "I am still normal" and did not like to call attention to things that contradicted that identity, or to point out situations in which they were singled out for being different. This is explored in more detail in the following section, 'Disability knowledge and identity positioning' (Section 5.3.3, page 143). In the context of challenging answers, it may mean children were less open to questions touching on negative aspects of having CP, or less likely to recall these in discussing everyday events. The pattern of non-responses and omission suggests at some level the children attempted to portray a more positive image than they may feel, especially with regard to difficult aspects of having a disability.

This was similarly exemplified in children changing answers when asked for more details and giving contradictory answers at different times in the interview. Both of these behaviours related to several factors. If the question touched on a complex topic, or one for which the child had ambivalent feelings, it was easier at first to provide a yes/no response, especially if they did not feel comfortable to explain, or thought the interviewer may not understand a specific disability issue. They would then correct themselves as they explained, or when the reality was not what they originally wanted to present, or simply change their mind as they further considered the question. If the question was challenging, and they wished to be agreeable, it was simply easiest to agree initially, even if this did not encompass the whole truth for them. Again, when the same children instead later responded directly with "I don't know" rather than simply agreeing, the child had become more comfortable and less amenable to the need to provide answers that they felt were required even if they did not have a response.

Events in the interviews which upset the unequal power balance between the interviewer and the child often led to a decrease in children's need to be agreeable. The child-friendly interview activities and strategies could be very useful for

demonstrating children's power of choice. For example, providing the opportunity for children to disagree with minor aspects of activities, such as the subject of drawings or what they wanted to do among multiple activities, or allowing them to have control over the subject—such as what they want to draw or what they get to ask the interviewer. Similarly, when the interviewer was equally engaged in the activities, through drawing or back-and-forth questions, it provided a more egalitarian setting. Changing the interviewer's focus from the child's responses to the interviewer's drawing reduced children's expressions of self-consciousness—especially around the quality of drawing if the interviewer was similarly critical of their own drawings. These examples all decreased the tendency for the children to agree and seemed to allow them to speak more freely. When children spoke more freely, the description of their experiences was more detailed, more emotional, as they answered questions with more complexity, and engaged more with the subject matter.

Children's increasingly detailed and engaged responses were probably partly consequent of increasing rapport; it happened over the course of the interview naturally, in some cases, and often led to conflicting information provided at the beginning compared with the end of the interview. For example, early in her interview Ana said that she went swimming once a week and enjoyed it, but later she stated that in fact she didn't like going swimming because it was difficult and she was not good at it. The underlying implication suggested it was because she did not feel competent physically as a swimmer when she maintained strongly as part of her identity that she was capable of many things, especially physically. In this instance, she also allowed herself to be more vulnerable and share more things that were painful to her by focusing on her own drawings, and by refusing to make eye contact. Although she was open from the beginning of the interview, it was obvious that she became more comfortable throughout, enough to ask at the end more about the interview process (such as whether every child interview included drawing), and this indicated her increasing comfort. By maintaining attention on her drawing, she maintained control over herself and was able to share more sensitive information, including that which touched on subjects threatening to her self-identity. She no longer appeared to feel a

need to agree with everything that was said, or what she thought the interviewer wanted to hear—that she was always happy and could do many things well.

Part of agreeing with the interviewer appeared to be related to children wishing to project an image of being happy and doing many things well. While this may be more salient for children regularly called on to counteract a deficit model of disability, it may simply be the result of wishing to appear positive to a stranger. In the context of interviews about wellbeing and QOL, it may also have been easy to maintain this stance because questions focused primarily on positive aspects of their lives which enhance QOL: things they enjoy and what they do with their friends. In the interviews presented here, too great a reluctance by the interviewer to ask about things that were difficult or painful for them may have contributed to this type of miscommunication.

5.3.3 Disability knowledge and identity positioning

In this context ‘disability knowledge’ refers to a recurrent pattern of conversations that emerged in interviews with parents and the older children. All interviews in this thesis were conducted by the same interviewer, an able-bodied adult with no visible physical disability. Therefore, the knowledge in question is the interviewer’s, not the parent or child’s. In many of the interviews, the interviewee would begin recounting something specifically related to their (or their child’s) CP, only to interrupt themselves and check whether the interviewer knew what they were talking about. A simple example was a conversation with Ana, a bright and bubbly 11-year-old. She was describing what happened in her regular CP-related appointments, and stopped to ask, “Do you know what AFOs are?”—looked closely to check the interviewer’s reaction, before providing more detail about the appointment. Once the interviewer’s knowledge of AFOs (a type of foot/ankle orthosis) was confirmed, Ana could confidently continue to tell me about the purpose of her appointments. Simultaneously, it established that although the interviewer did not have CP, she knew enough about aspects of having CP that Ana didn’t need to explain everything. A similar checking process was repeated in several interviews, and may have contributed to openness about experiences of having CP. While many questions were about specific aspects of CP diagnosis, interventions and

therapy, they contributed to the positionality of interviewee and interviewer as insider or outsider in disability knowledge and culture. That is, participants observed that the interviewer was able-bodied (an outsider), but conversant with CP symptoms and treatments (holding insider knowledge), and adjusted their conversation accordingly.

A different perspective emerged in conversation with Eric. Towards the end of the interview, he responded with silence to a general question about whether there was anything else annoying or otherwise about having CP that he wished to mention. His mother suggested hospital appointments, and he responded, “yeah but the hospital thing I think goes without saying”, to which his mother replied, “it’s just that maybe she [the interviewer] hasn’t experienced what it feels like”. He began to talk about specific parts of the process that annoyed him, or particular occasions that he found very frustrating—there were several. He obviously had many negative feelings about his various necessary hospital appointments and the way these are handled by the health system—but he would not have raised this without his mother’s prompting. This was repeated with other issues of school inclusion raised by his mother—he was animated in adding details about his frustrations to his mother’s comments but did not directly raise any of the several frustrations resulting from his CP that clearly negatively affected him.

Throughout the interview, Eric shied away from presenting details of having a disability unless directly asked, and then generally downplayed the things he found “annoying” (his term for them). This is not to suggest that he was being deliberately dishonest, or even that he was not open or willing to speak—he was generally charming and chatty. However, in the context of an interview that he was specifically invited to because of his disability, he spent time and effort presenting an identity in which having a disability was not prominent. Overall, this suggested two interesting points. Firstly, there is a breadth of issues unrelated to disability which contribute to QOL for a child with CP—echoed by all the interviewees. Secondly, the importance of recognising and validating the other part of a child’s life and the conflict they may experience in continually being singled out because of the disability identity—especially to an outsider to the disability community.

These contrasts were particularly obvious in the interview with Zachary, a very articulate and self-reflective 12-year-old. From early in the interview, Zachary introduced various aspects of his experience of having CP, both positive and negative. He discussed these with some self-consciousness, but with significant detail. However, several times through the interview, he explicitly emphasised that he was not different to other children. For example, in the emoji activity at the beginning of the interview, he first picked an unhappy emoji saying:

This was in the past, I reckon, because mostly I had to wear an orthotic which is really annoying, uncomfortable. And, it was just when I couldn't do as much stuff. I mean, like, I'm still normal and everything. It still feels like I'm a bit different.

At the end of the interview, when asked if there was anything else about children with CP that would help them have better lives, he responded “There's not really, like, because you're not different so it's not like there's one specific thing now that will make your life better”. This statement emphasised not only the diversity of CP experience, but that CP itself did not signify “difference” that requires intervention.

In contrasting the interviews with Zachary and Eric, it is worth noting that Zachary may have been better at integrating these things because his impairments were less severe—at GMFCS level I, he is physically more like a typically developing child, has a less visible disability, and is able to be included in most, if not all, school activities. Unlike Eric, who is a wheelchair user with a minor speech impairment: his disability is immediately apparent to those around him, and he faces physical barriers in navigating his mainstream school, as well as specific school rules that prevent his inclusion in most outdoor/sports activities (this is discussed in greater detail in the following chapter, in ‘Negotiating (physical) limitations’, Section 6.3). Both were bright, creative, socially adept children—both had a disability—but their experience of the reactions of others to their disability was quite different. Eric may have been less willing to speak about the impact of his CP not only because he felt it was more obvious, but because he felt more need to distance himself from an identity framed solely around ‘child with disability’. The specifics of children's developing identity related to disability is

explored in the conceptual category 'Defining the self' in the following chapter (Section 6.4, page 171). Analysis of the interactional processes in the interviews discussed here informed aspects of the conceptual category.

Both Zachary and Ana showed this aspect of their self-concept throughout their interviews and explicitly in things they said. For example, Zachary stated that he was normal because he could still do everything, or Ana emphasised, "it's not like I'm an alien, I just have a disability!". They have incorporated a stable idea, and stable identity, involving having a disability, and what that means for what they are or are not able to do. However, they are still, like most children, striving to appear 'normal', and do not like to be reminded of any situation or experience that is incongruent with being 'normal'. Defending this aspect of their identity may be more prominent in a research interview specifically focused on what it means to be a child with a disability. Children's reflections on these experiences echo the duality of inclusion and difference reported in classroom practices for children with disability (Holt, 2004). In that research, the author found that settings and practices create narratives of levels of difference that were accepted, but also that children could be active in resisting these representations, or accepting positive, inclusive practices. This shows that reflecting on these interactions in the research interview setting can provide insight into children's usual patterns of behaviour in fitting both worlds of 'disability' and 'normality'.

Reflecting on the insider-outsider effect of able-bodied adult interviewer and child interviewee with a disability can provide additional context to the interview. While taking a 'dummy' stance as an interviewer can help to decrease the power imbalance and encourage children to provide more detail as an expert on the disability experience (Darlington & Scott, 2002), there can be negative consequences. It may create a divide in what is explainable if the interviewer does not appear to understand or have the basic knowledge of common frustrations and limitations in accessing disability services. By emphasising their distinction as a person with a disability in the research context, child participants may additionally feel the need to present disability-specific information in a uniformly positive light. In contrast, if too much knowledge is assumed by interviewers or interviewees, generalisations and underlying assumptions about experiences may be misinterpreted or incompletely explained.

5.4 Discussion

The approach used in this study included multiple, non-traditional interview strategies for child interview participants in a single interview within the family home. The reflections and analysis included in this chapter provide suggestions for future researchers intending to include similar methods. These include practical considerations of collaboration with parents or carers, and incorporating multiple methods, as well as broader considerations of how novel interview strategies can impact on the power balance between child participants and adult researchers.

One of the major practical considerations with child interviews is the presence of other adults, such as parents or carers, in the interview setting (Pyer & Campbell, 2013). While previous researchers were successful in engaging parents as collaborators to tailor interview materials to each child participant (Teachman & Gibson, 2013), this requires additional time with parents, and may be more suited to protocols where multiple meetings with families are possible. In the interviews for this thesis, it was difficult to prioritise the information gathering part of the parent phone call as this call was also used to organise an interview time. In future research it may be necessary to prioritise time for parental collaboration during the early part of the interview or be prepared to explain it to parents at that time, rather than including it at the end of the initial phone call scheduling the interview. For children with diverse abilities a research design which allows additional opportunities for the researcher to get to know the child directly may be preferable, rather than relying on parent collaboration. Even with ideal parent collaboration the use of the strategies still needs to be flexible and adaptable during the interview, which reflects a need to prepare multiple activities and allow the interview to be child led. In discussing challenges in conducting interviews with children in family homes, Coad et al. (2015) emphasised the need to be flexible in conducting interviews with children regardless of the use of strategies. Planning for multiple interviews scheduled with time set aside specifically for parent collaboration and time for the researcher to become familiar with the child and their abilities may assist with adapting methods, especially for children with more complex communication impairments or more severe disabilities for whom interviews generally

lasted longer in this sample. However, the single interview may be more feasible for families who must balance research participation with childcare and other commitments. Similarly, multiple methods provided additional ways to gather data and allowed children to exercise more control over how they participated, but may add more time to an interview, or require more planning on how to tailor methods to each participant within the interview even with parent collaboration.

Regardless of parents' initial involvement in the interview strategy, the presence of parents during the interview is likely to change how children respond to questions. Pyer and Campbell (2013) discuss the impact that significant adults can have on children's participation in multiple stages of a research process. As in this project, they concluded that the presence of adults often changes how children participate, from early in the process where adults act as gatekeepers, to altering the specific content children will offer as participants. They also note that not all changes are necessarily challenging for data collection, similar to the interviews analysed here: for example, parents in this project frequently encouraged children to respond through specific prompts or more general support. This raises two main points for future researchers working with children in similar interview settings. Firstly, it is necessary to build into an interview protocol how to negotiate for access with children—with or without adults present—throughout the research process. Secondly, it is doubly worthwhile to include an analysis of the interactions in the interview, as these provide insight into the impact of adult-child interactions and how they shape the data produced. This builds on the work of Rapley (2012) and others who note the way dynamics between interviewer and participant influence data.

Interactional analysis of child interviews can also help to illuminate power dynamics in research interviews. Most interview participants are influenced by social desirability however children are likely to be especially vulnerable due to the perceived authority adult researchers hold. Greene and Hill (2008) discussed children's susceptibility to coercion in research environments which to adults may not appear to be coercive. They highlight that children have been shown to give answers even to questions which have no sensible answer in research interview settings. This may be due to power differentials, or misunderstanding of the intentions of the researcher questions, or

other factors. Researchers working with children therefore need to consider processes for checking informed consent not only during the informed consent process, but throughout data collection.

The use of multiple non-traditional strategies within an interview may help to change unequal power dynamics, as discussed earlier in this chapter within the analysis of interactional processes (Section 5.3). With an alternate strategy, Anderson and Balandin (2011) discuss how a visual 'storybook' method provided a way of checking children's responses. They found particularly that the use of stories and cartoons encouraged children's participation by breaking down assumed power relations and enabling children to contradict adults. This echoes the concept of the 'dummy interviewer' raised by Darlington and Scott (2002), which subverts the adult-child power dynamic by positioning the child as an expert. Taking a different approach, Lewis (2010) cautions researchers to respect children's right to silence in interviews, where they may not feel comfortable to contradict an adult interviewer or refuse to answer.

The use of strategies such as those employed in this project may assist children to choose their participation not only by permitting them greater agency and power, but by providing distractions that facilitate silence when they choose not to answer. While employing child-specific strategies may focus methods on a process of drawing data out of children, it is essential to be flexible to allow for children's own right to refuse not only during initial consent to participation, but within the entire research process. Strategies designed to facilitate communication must also be sensitive to children's vulnerability to coercion in research settings, and respect refusals to respond. Hill and colleagues (1996) raised similar methodological considerations following research regarding children's views of their emotional wellbeing when researchers were first advocating for increased 'child voice'. They concluded that children's engagement could be increased by multiple methods for participation, but that researchers must be flexible in allowing children agency in how they choose to participate. Especially with children from potentially vulnerable populations, researchers must prepare before the interviews to respond to potential child distress: some topics may be too difficult for children to discuss even in the best of circumstances, and this must be respected.

Others working within participatory methods have suggested it is naïve to establish any type of research protocol for work with children and imagine that it can be rigidly applied to manage all ethical concerns of child engagement—without reflexivity, any method has the potential to compromise child voice (Gallacher & Gallagher, 2008).

Other researchers have reported similar challenges when interviewing children as those reflected on in this chapter in Section 5.3.2 ‘Child responses to difficult questions’. Combining the reflections in this chapter and conclusions from other authors (for example, see Coad et al., 2015; Lewis & Porter, 2004; Westcott & Littleton, 2005) suggests three possible ways to reduce these challenges in future research. Firstly, by focusing on building rapport earlier so children were able to say that they did not know an answer, rather than merely agreeing—it may additionally help to remind them before and during the interview that it is okay to respond with “I don’t know”. Secondly, the interviewer can emphasise child-led aspects of the interview, such as choosing topics but allowing them to select what to talk about. Children interviewed here were more confident and talkative when they could follow their own narrative and introduce their own topics—even if they were the same things the interviewer wanted to ask about. Dockett et al. (2009) suggested that allowing children greater control over data that is generated and included in research not only increases children’s engagement in the research process, but also improves interpretation of children’s experiences. Thirdly, being child-led also implies going at the child’s pace. In this research, this was especially necessary for those with slowed or impaired speech, as well as allowing children greater time to recall details or choose how to respond. These challenges have direct implications for content: if the interviewer is unable to overcome these challenges, children make very little contribution to the interview content; if the challenge is reduced, new, unexpected information in the child’s own voice is likely. Furthermore, the combination of these approaches reiterates Lundy’s (2007) two factors of space and voice necessary to allowing children to express their views, by facilitating opportunities where children can comfortably express themselves to the best of their ability.

5.5 Summary

In conclusion, the use of multiple methods in this research provided additional ways for the children to participate, especially for those with alternate means of communication. The strategies also may have helped to change the traditional, unequal power balance between the adult researcher and child participants, increased child engagement, and allowed greater accuracy in understanding children's perspectives by supporting children's right to contradict or disagree with adult authority. Flexibility in implementing the strategies within the interviews enabled children to maintain agency over their participation and adapt the activities to their own preferences, including communication preferences. This helped to overcome the challenges of parent collaboration, which was not largely productive within initial phone calls. The interactional analysis in this chapter provided richer insights and context for the main content analysis of child interviews, especially for understanding the impact of adult-child dynamics.

There are clear positive benefits of multiple methods and flexibility in interviewing strategies, especially when including child participants with diverse abilities, though the current sample did not include children with severe CP at GMFCS levels IV and V. Although there can be difficulties in practical implementation of the strategies included here, these difficulties do not necessarily detract from data collection. With regard to parent collaboration, it can be difficult to engage parents as collaborators prior to the interview without providing significant context if they are expecting a traditional interview. There is additional value to be gained in analysis by considering interactions between child and interviewer, and child and parent, as they can inform analysis of interview content data—especially relational aspects. Additionally, for researchers who are outsiders to the disability world, interactional analysis can inform views of how children interact in their everyday lives with those who are not knowledgeable on disability and impact interpretation of interview content. The following chapter includes the main analysis of the child interview data, including references to how the interactional analyses above impact interpretation of the main findings.

Chapter 6: Child interview findings

This chapter focuses on the findings from the analysis of the child interviews. As discussed in the methodology chapter, parent and child interviews were analysed separately. The secondary aim of the chapter is to apply reflexivity to the analysis, recognising that the findings are co-constructed between the context, the interviewer, and the interviewees. All child names used in this chapter are pseudonyms, as discussed in Chapter 4, Section 4.2.6 (see page 106).

Analysis of the interview content revealed the following five main conceptual categories described below: ‘social beings; being social’, ‘play’, ‘negotiating (physical) limitations’, ‘defining the self’, and ‘establishing agency’. Although they all relate to each other in various ways, all are underpinned by the conditions of social relationships and socialising. Therefore, ‘social beings; being social’ is described first. The connections between the main categories, and their main sub-concepts are mapped below in ‘*Figure 6. Main conceptual categories of child interview findings with sub-concepts*’.

Each of these categories are drawn from and represent facets of children’s everyday life, occurring as a process that changes over time. The resolution of each category forms a developmental goal of childhood, though resolutions are continually renegotiated as the child ages and environmental demands change. Time forms the background for how children discuss their changing experiences—contrasting contextual changes as they move through grades at school, as Dylan describes “from grade five onwards we need to do...”, or simply growing older as when Sofia clarifies her own changing behaviour with “like, when I was little I used to...”. The desired outcome for each category also changes with time—as children’s ambitions for independence and achievement change with age, as well as the different expectations others have for an eight-year-old compared with a twelve-year-old.

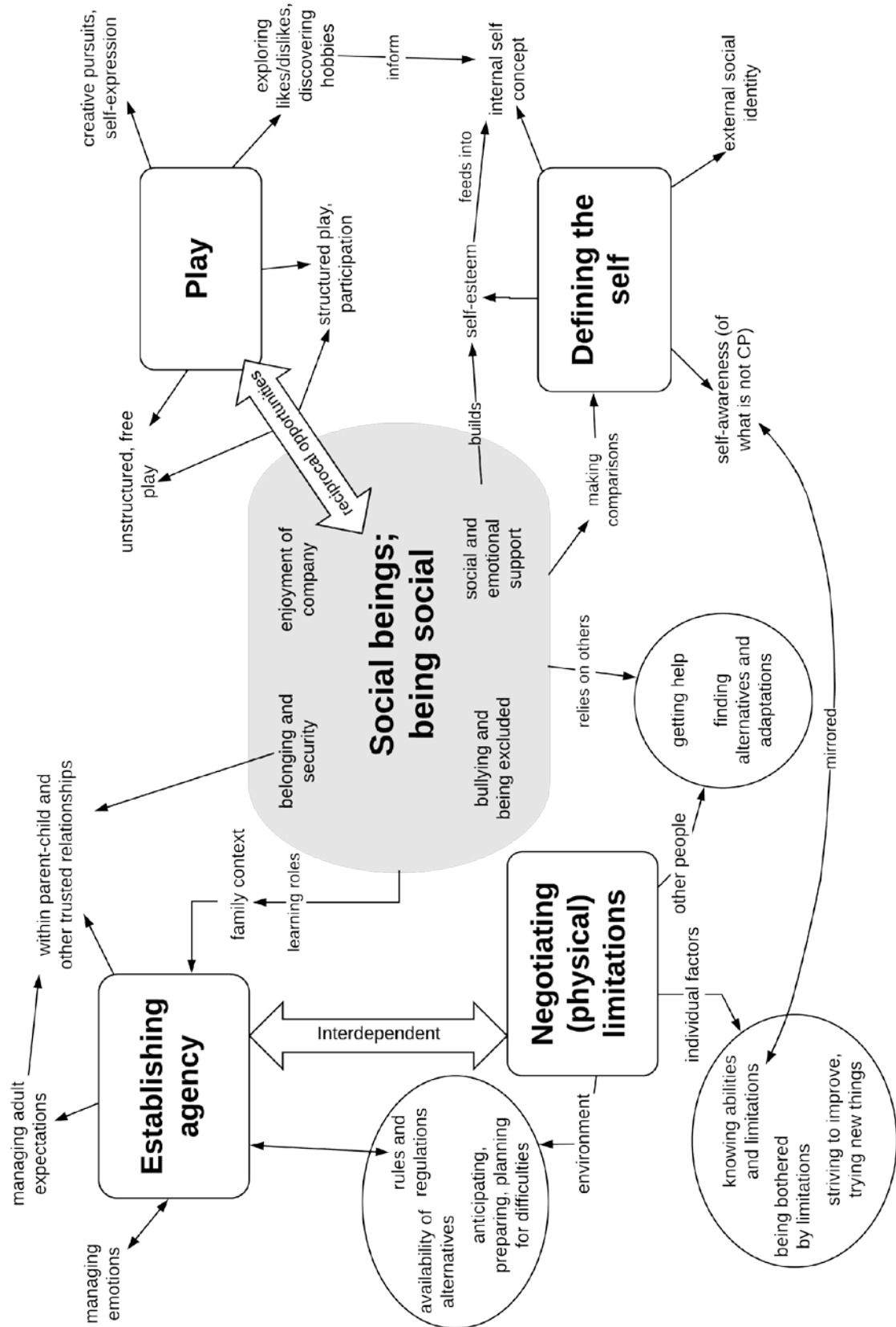


Figure 6. Main conceptual categories of child interview findings with sub-concepts

It is likely that each of the identified concepts would be developmentally relevant to children without disabilities, as necessary goals and experiences of growing up. However, the environmental interactions often differ for children with disabilities—partly because children with disabilities usually experience restricted opportunities in each of these areas of their lives. In some cases, the specific circumstances related to being a child with a disability may simply make developmental challenges more salient. In others, the disability itself adds new aspects unlikely to be faced by typically developing children. Within some families, this is shown by differences between siblings. Where children make direct comparisons between themselves and siblings, they are described in the following sections in each main conceptual category.

6.1 Social beings; being social

All the children interviewed spoke about the importance of being with people for the sake of socialising, and the specific people in their lives who were important to them. Often, they discussed the processes involved in being social (such as how to make friends, or maintain friendships), and situations and people that presented help or hindrance to their socialising. They went on to discuss how the presence and absence of social relationships and social support can create opportunities in other areas of their lives, or conversely inhibit those opportunities.

One of the aspects of being social raised most easily and most often by children was simply the enjoyment of company. This was most clearly expressed with regard to peer friendships, as Ana said, “I like hanging out with my friends”, or Nicholas agreed he was like the happy emoji “when I’m playing with my friends”. Besides their peers, children expressed enjoyment and appreciation for relationships with their parents and siblings, first and foremost. They mentioned complex extended social networks in which they singled out valued relationships with other adults, including extended family, carers, teachers, school aides, therapists, and other community members they engaged with in leisure activities. Spending time with people with whom a child has a positive relationship was directly related to a child’s general state of happiness and contentment, whether those relationships were with peers and friends, a family member, or an unrelated adult. Not all children (or adults) require large social

networks to achieve satisfaction with their social life, but those who were happiest with their social situations generally nominated positive relationships with close and extended family, school friends, additional long-term adult caregiving relationships, and peers in their neighbourhood.

A universal theme as to why specific adult relationships were valued positively was because of perceived kindness, support, encouragement, and inclusivity with regard to disability. Nicholas frequently did not engage with interviewer questions but became animated and responsive after he spontaneously raised his relationship with his school aide—overall, he preferred to discuss the people in his life, such as his aide, teacher, or brother, more than his activities or himself. In the context of pursuing physical sports, Zachary mentioned how important his physiotherapist had been because “she’s really like, helps me get through it and stuff. Like, she’s really supporting ... she was, like, very encouraging, saying “Good work” and stuff”. Similarly, Gus spoke about a valued swimming coach who had taken extra effort to prepare adapted lesson plans, as well as joking with Gus and engaging him personally. In contrast, after that coach left the local pool, the next swimming instructor was not able to be as supportive, was not interested in cultivating a personal relationship with Gus and did not provide additional lessons adapted to Gus’ abilities. Gus has since discontinued his swimming lessons until his mother is able to find a more supportive coach. Although the facilities exist, and the pool is happy to have a child with a disability attending lessons, it was the relationship with the individual coach that made participating a positive experience for Gus and his mother.

It was clear and unsurprising that children required and received social and emotional support not just from peers but from the adults in their lives. Among the common experiences of disability-related exclusion, sympathetic adults who facilitated opportunities for inclusion were highly valued by all the children. For these children, perhaps more so than typically developing children, longevity of the relationship was especially appreciated. Children generally require time to bond with an adult, but time may also be required for the adult to be trained in a child’s disability specific needs, and how best to respond to and support them. Every new adult relationship requires a new explanation of what the child’s disability is and how it affects them: most children

with CP have already transitioned through multiple health service contexts and consulted with many health professionals by the time they reach school age. Zachary was unsure exactly how many physiotherapists he had seen saying “I’ve probably seen at least, like, five maybe or I don’t know”. When asked if changing physiotherapists was a problem he responded, “Well it depends how long I’ve been with the physio...”. He mentioned the positives of the relationship that he had established with his current physiotherapist and concluded pragmatically, “Well yeah, so right now I’d stay but it’s not too bad to change. It’s not the end of the world or anything but it is—yeah”.

Especially in the context of aides, therapists and leisure activity coaches, established relationships may allow others to provide more meaningful support and encouragement to help children reach their personal functional and participation goals. Long-term relationships additionally may provide more opportunities to celebrate their ongoing improvements and achievements, by allowing for greater comparison from past performance. Children thereby have more positive experiences of their abilities even when these differ from their peers. These concepts are explored further in the category ‘Defining the self’ (Section 6.4, page 171), in terms of developing positive self-concepts and self-esteem.

Ongoing social groups also provide children with a sense of belonging—especially in the school context, which is the major social setting for children outside of their immediate family. School friends are extremely important to children’s enjoyment of schooling, and not having a trusted, supportive friendship group leaves them open to bullying and exclusion. The schoolyard at recess is the site of a significant number of children’s social interactions and opportunities to play. When Zachary was asked what his favourite subject at school was, he was quiet for a moment before he and his mother shared a joke about whether recess counted as a subject. He was not alone: several of the children when asked generally what they liked best about school replied with variations of “recess” and “playing with my friends”.

Most children, regardless of their disability, have had negative experiences of peer socialising—however other research suggests children with disabilities may be more vulnerable (Chatzitheochari et al., 2016). Three children shared experiences of being

bullied because of their disability, including taunting, deliberate exclusion, and in one case, physical bullying. These children framed the experience as the extreme of poor social experiences. In turn, they reflected that the antisocial behaviours were the result of characteristics or behaviours of the children who had bullied them, for example, Ana framed it initially by saying “sometimes there’s some annoying friends”. A few of the children then went on to emphasise their own pro-social behaviours, or appreciation for prosocial behaviours by others in their lives. For example, Sofia mentioned “some friends do help me”, while Dylan said he looked like a frowny face emoji sometimes because his two brothers could be annoying, but immediately added “But they can be pretty helpful as well sometimes.... Just help me with getting around”. It seemed children’s experiences of exclusion or bullying ultimately emphasised the value of people who were kind or helpful.

All the children, including those who had not been bullied, mentioned experiences of exclusion related to their disability, especially in situations where activities or environments were not or could not be adapted to their physical differences. While they expressed varying levels of frustration with these situations, again, they also expressed appreciation for the prosocial behaviours and efforts of others to be inclusive, supportive, and welcoming. Most children can recognise the importance of ‘playing nice’, however, children with disabilities may have these lessons reinforced earlier or more directly through experiences of disability-related exclusion.

Children with disabilities may experience social exclusion outside of the school context as well. Most children discussed visiting friends’ houses, or having friends visit their home. Much of children’s socialising occurs through unstructured play, during organised playdates, or group social occasions such as birthday parties. Dylan mentioned that many of his friends lived close to him and referred to “the things I like to do when friends come around” being a common part of his activities. However, playdates can be an issue for children with higher levels of physical impairment who require specialised transport, or assistance to navigate other children’s houses. Sofia spoke about attending few birthday parties in comparison to her siblings and reflected an awareness that this was related to her disability when she said, “some people think it’s too hard”, referring to children’s parents as much as the children themselves.

Where children were able to access disability specific services or sporting and leisure activities, these provided value not just through inclusion, but because children formed favourable peer comparisons and friendships with other children who also have experience of disability. For example, Caleb joined grade 1 initially at a mainstream school—but he did not want to talk about this at all, and his mother reported that he did not join peer groups and was not very comfortable because of his intellectual disability. He was then admitted to a special school for children with intellectual and learning disabilities and has formed a few strong bonds with his peers—he was very animated in talking about his school friends since he had changed schools, and he clearly valued these friendships as he did his family relationships. Through attending a special school, he has been able to form more positive and supportive social relationships and has a sense of belonging in his new peer group. Special schooling is not possible or desirable for all children with disabilities, but disability specific services often provide children with similar positive social experiences above and beyond the chance for leisure participation.

In talking about the conditions and consequences of being with people, children often recognised that social relationships are both a context underlying other aspects of their lives, and an end in themselves. That is, social relationships provided a background for each of the other main categories, in forming the environmental context of children's lives—but the act of socialising is a primary goal in itself. Beyond the benefits of having positive social relationships, some of the children described the process of making friends, especially in the context of a perceived lack of necessary social support. Children recognise the value of making friends with those who have similar interests and were deliberate and active in the process of choosing friends.

It was school holidays when Zachary was interviewed. During the holidays, he had not spent much time socialising with his school friends but mentioned he had been to the movies with one friend. When he was asked for more details about the movie or other movies he enjoyed, he said that he quite often goes to the movies “mostly because of my friend, because he's always getting tickets and then inviting me”. In this case, it was the shared interest, especially on his friend's part, in seeing movies that provided him with frequent social opportunities outside of school.

Counterintuitively, being excluded from more physical forms of play is used by some children as a basis for building social networks. That is, knowing that they are unable or uninterested to join peer groups focused on energetic play, children will seek companions who prefer quieter games, spending time indoors, writing, reading, or playing video games. For example, Dylan is a wheelchair user with limited capacity to ride his modified tricycle. He shares his peer group with his typically developing fraternal twin but mentions that he is often left out if they go for long bike rides in the neighbourhood. When discussing friends at school he says that he and his brother have a few friends in common “but I’ve got my own friends as well”; later in the interview he remarked on his and his friends’ shared interests in making movies and jokes.

Sofia made similar observations in comparing her typically developing brother’s friends and her own. She was speaking in the context of missing out on attending birthday parties, saying even if she attended they were very loud and involved a lot of running around—with her mobility aids she is able to walk, but could not keep up with most children running. She concluded by saying “I don’t like those type of people... running around people... I like people who are more quiet” but also mentioned “my brother likes those type of people”. Both Sofia and Dylan explained clearly their experiences of social exclusion because of their physical disabilities, as well as their deliberate choice to seek different peer groups from their siblings to engage in play and social activities within their abilities. They have tailored not only their personal play activities, but also their peer groups, to what they enjoy without restrictions due to physical impairments.

As noted in Figure 6 (page 153), the social category provided context to children’s everyday lives, and linked to each of the other categories. These links are described briefly here before moving onto each of the other categories. Socialising was most strongly linked to the conceptual category of ‘play’. Many types of play, both structured and unstructured, can intentionally and/or unintentionally exclude children with disabilities. In turn, this excludes them from social opportunities. This has both an immediate effect, in reducing their present experience of socialising, and ongoing effects, in leading to a reduced social network that continues to leave them with fewer opportunities to socialise or build social skills. While most children at times engage in

solitary play or pursuits, children report this generally as an exception rather than the norm. Sofia spoke poignantly about her lack of friends, saying “I wish I had friends in this street so I can play... we could play an activity after school”. In so saying, she directly linked opportunities for play with having peer relationships—because of her lack of local friends, she was unable to play after school. Having a wider social network created more opportunities for play, but the relationship is reciprocal, with children forming social relationships through play with like-minded peers, as discussed above.

Defining the self relies on social relationships, in that internal self-concept is usually delineated by comparisons with others, while self-image and social identity is based on considerations of other’s supposed opinions and sense of belonging with one’s peers. Positive social relationships also directly build self-esteem—and potentially, the opportunity for prosocial behaviours also builds self-esteem. Similarly, in making all sorts of comparisons between the self and others, children are disadvantaged if their social context is more restricted. Those with broader social networks are exposed to more differences and are therefore less likely to conclude that their own visible differences, such as physical disabilities, are a negative aspect of self. This is especially true for children who have experienced social exclusion or discrimination as a result of their disability—supportive social relationships are likely to mitigate the effect that these experiences will have on self-esteem and self-image.

Lastly, social relationships enable child agency, through appropriate support and encouragement, providing a safe environment in which a child can begin to explore autonomous actions and choices. Social relationships can provide support for the child to develop their internal sense of agency or can limit it by encouraging dependencies or intervening too quickly when the child attempts independent actions. Additionally, child agency is usually first negotiated in a family context, with parents or siblings—both within the family, as well as by modelling parents’ actions. This is further explored in the later section on the category ‘establishing agency’.

6.2 Play

The right “to engage in play and recreational activities” is enshrined in the CRC (p. 9, United Nations, 1989).

It is recognised, for all children, that free play provides opportunities for learning and contributes to child development (Missiuna & Pollock, 1991). Children learn many things through play, especially play with others, and if they do not have enough opportunities for play, they have fewer chances to develop essential life skills. Definitions of free play from research literature suggest, among other things, that it is: universal and central (Factor, 2009); the 'primary productive activity of childhood', fun, and self-motivated (Missiuna & Pollock, 1991); not 'serious' or 'real', and must involve freedom of choice for the child (Mindes, 2015).

To children, 'play' is an idea and activity that cannot really be explained more than that: play is simply play, universally understood by children but unexplainable. It is malleable enough to encompass almost any activity, and generally has no purpose except it is something that they value and actively engage in, often with other children such as siblings or friends. For the older children interviewed, the idea of 'play' tended to transition into something that younger children do, and they instead referred to 'doing stuff' with friends. Older children used the phrase 'doing stuff' to refer to the same type of activities: a collection of overall purposeless activities that usually involve fun, socialising, and creativity, and can be indoors or outdoors or for any level of ability or impairment. Similar exchanges are recounted by Factor (2009).

All the children interviewed introduced play as something they mostly do and enjoy with their friends but cannot fully explain to an adult. These conversations tended to follow a typical pattern, generally as the following:

Interviewer: What sort of things do you do with your friends?

Younger child: I don't know, we're just playing, running around and stuff

Older child: I don't know, just doing stuff

This type of conversation reinforces the strong link between socialising and play—play is what you do with your friends during recess at school or on weekends and holidays. In a way, children used the terms "play" and "doing stuff" as catch-all phrases to explain how they pass the time with their friends. However, especially in saying they are 'playing', there is a specific meaning behind the term. For the children, it was

obvious what they were referring to, just as it was obvious that schoolwork or adult directed hobbies are not play.

The specific activities referred to in talking about play varied greatly. Children talked about playing with siblings, friends, pets, games, and toys; play involved “running around” outside, sitting down indoors, or some combination of activities. Along with the difficulty each child had in elaborating further on what ‘playing’ with friends actually involved, it suggests the importance of spontaneous and creative play. In a sense, play can be whatever children choose to label ‘play’—with the emphasis on children’s choice, or children’s creativity. The implication in every conversation, even where they (rarely) referred to playing with an adult, is that ‘play’ occurs without adult oversight, rules, or purposes. Even where play involved video games that have predefined rules, children are in charge of the play itself. Children didn’t parse out technological play, such as playing with video games, as being any different to other types of play, except that it was easy to explain as something they do with their friends.

Having access to time, space and friends to engage in this kind of play is important regardless of level of ability or type of impairment—but for children with more severe or diverse impairments, it becomes increasingly necessary for both the children and the adults around them to create opportunities to engage in play. Play opportunities are one direct consequence of social inclusion—and conversely, the lack of play opportunities are as keenly felt by children with restricted social circles as the lack of social interactions. This is recounted earlier in Sofia’s desire for friends in her street with whom to play. Although she spoke at other times about solitary activities she enjoyed that she considered to be playing, having friends was a precondition to general ‘play’.

Solitary play was infrequently discussed by the children. Instead, when discussing positive solitary occupations, they often refer to creative pursuits, such as drawing, movie making, writing stories—often mediums for self-expression. These usually have things in common with general play—the first is that they are a source of and expression of joy, that they include an element of pretend or imagination, and lastly

that they are activities in which the process is divorced from achievement. As children age they may become increasingly concerned about the skilfulness of their creative output—but these remain activities that are accessible to anyone. The quality of a drawing may be judged, but anyone can draw; the complexity of games may change, but from a child's perspective, anyone can play.

Children with physical disabilities may require adapted or alternative toys to explore their environment, just as they require functional mobility aids. These resources can be scarce, unaffordable, or hard to access; so too can play equipment. As mentioned above, Dylan has a modified tricycle that he is able to ride independently for short distances. However, he has recently outgrown the tricycle, and has been on a waiting list to get a replacement for several months. Consequently, even if he were motivated to increase the distance and speed he can ride to keep up with his twin brother and friends, it is not possible for him to do so without significant discomfort. Therefore, he no longer joins his friends even on short rides—missing valuable play and social opportunities, as well as physical activity. There are many other adaptations and alternatives that are required or helpful for children to join in play, especially with their able-bodied peers. The conditions of enabling this participation are explored further in the concepts of 'Negotiating (physical) limitations' (Section 6.3, page 164).

Children with disabilities often face additional limitations through rules and regulations—designed principally for their safety—especially in mainstream environments, as noted by Missiuna and Pollock (1991). This was raised by a few of the children interviewed. For example, Eric uses his wheelchair at school, but is not allowed in his chair on the grassed area of the playground, “where all my friends are”, unless he is pushed there by his aide. Therefore, he is unable to join the main play area during recess without adult intervention, which is not always available. This ties into both negotiating limitations and developing child agency. Eric was clear in his understanding and disdain for the rules—“it's a stupid rule.... Apparently, it's the government's, but I don't think so. I think the school's just way paranoid”—but within the school system neither he nor his parents were able to change it. To Eric, not only does it limit his opportunities to play at recess, it demonstrates a frustrating inability to achieve his own agency over play.

Play does not just provide a teaching ground for developmental goals and social skills. Through play, children can explore the world around them, discover what they like or dislike, what interests them, as well as their own strengths and abilities, physical and otherwise. Through play, children may be initially motivated to discover new hobbies—some of which lead to structured leisure activities labelled in disability research as ‘participation’ (Imms et al., 2016). The children interviewed generally did not discuss organised activities as ‘play’, therefore differentiating play from the concept of ‘participation’ in disability literature. However, organised recreation can offer some of the same opportunities and benefits as play: building social networks, providing joy, learning likes, dislikes, strengths, and teaching how to collaborate and cooperate. It is unclear whether participation activities (hobbies, sports, organised recreation) can replace play for children who are not as able to join in spontaneous play, but it may be that even with the benefits of participation in organised recreation, children themselves will feel deprivation of restricted spontaneous play.

6.3 Negotiating (physical) limitations

The concept most directly related to disability for the children interviewed was that of negotiating (physical) limitations. As they age, all children discover their strengths and weaknesses in comparison to their peers, and within formal schooling. However, children with disabilities experience more frequent limitations both in their environment and in their own bodies. From a young age, children with disabilities must engage in additional work to achieve their full potential in a world made for the able-bodied.

Due to the physical impairments inherent in CP, the limitations children faced primarily involved issues of environmental access and assistance for physical tasks such as writing, dressing, or getting around at school. However even purely physical limitations required alterations for children to be included in social activities and, due to frustrations with motor difficulty or missing out on inclusion or regular school activities, children with CP may have more challenges with emotional regulation than typically developing children. Children and teens with CP have a higher prevalence of mental illness than their typically developing peers (Downs, Blackmore, et al., 2018),

and this echoes children's behavioural and emotional regulation problems. A few of the children interviewed experienced anxiety at a level which required psychological intervention—and therefore their everyday lives involved negotiating accommodations for fluctuating anxiety symptomatology, with people and environments around them. Comorbidities common to CP, such as intellectual disability, add another layer that may outweigh milder physical impairments—and may increase as children age and the gap between developmental expectations and cognitive abilities increases. It is the combination of these additional limitations in the context of the dominant physical impairments of CP that lead to the addition of parentheses around 'physical' in the naming of this concept.

The notion that dealing with CP and disability is a process of negotiation supports the social model of disability—that disability is a result of environments that do not accommodate alternative abilities or ways of being. The experience of being a child with CP involves an ongoing process of negotiation between personal abilities and impairments, an environment that is at times hostile or unwelcoming, and often relying on the assistance of others to find alternatives or make adaptations to expectations and activities. Successfully negotiating these hurdles results in higher levels of participation in education, play, and social activities, and a greater personal sense of satisfaction and achievement.

At the personal level, managing limitations involves children knowing specifics of their CP impairments, handling feelings of bother or frustrations about limitations and impairments, and the willingness to try new things and strive to improve in existing hobbies. The environmental aspects of limitations that must be managed include anticipating, preparing and planning for difficulties, the availability of alternatives, and the presence of rules and regulations that may provide additional restrictions because of a child's disability. Between the individual child and their environment, other people enable negotiations around limitations. From a child's perspective, this is the broad concept of getting help, which often forms a background for their everyday lives.

6.3.1 Personal factors of negotiating limitations

One of the first personal aspects of overcoming disability-related limitations is understanding personal abilities and CP related impairments and how they affect activities. Most of the children interviewed were intensely familiar with their limitations through the diagnosis and medical management of their CP throughout their lives and spoke matter-of-factly about specific limitations. For example, when asked if she missed school activities because of CP, Ana responded in part “Sometimes on triathlons we do for sport... like I can’t really jump, unless it’s a big trampoline with a net... or hockey sometimes is hard for me ‘cause I can’t run with the stick down”. Dylan spoke similarly of sports at school, saying, “there’s only one sport during PE that I can’t do though, is skipping. Skipping is really hard”.

Understanding their own limitations relates to how bothered children were by things they struggle with. The children seemed to find it easier to say definitively “I can’t do this thing”, than to talk about things they could do, but which are difficult because of their CP. For example, tasks involving fine motor control which required significant concentration and practice, and which they could not perform as well as their peers. For a couple of the children interviewed, this translated to a reluctance to draw during the interviews due to embarrassment about their level of drawing skill, though one boy later revealed he actually enjoyed drawing and was trying to improve.

One way in which children successfully negotiated limitations was by achieving and enjoying alternate or adapted participation. That is, where limitations are inflexible, such as physical impairments which definitively prevent ‘normal’ ways of participating in an activity, a successful negotiation might instead result in a child enjoying an alternate activity, or an alternate role in the mainstream activity. This requires both the assistance of others to adapt the activity/or adapt to the child’s alternative participation, but also requires the child with CP to either accept that alternative option or push for an alternative that is acceptable to them.

While children were often bothered by limitations, they discussed adapting to *not* being bothered by their limitations, and instead finding satisfaction in being able to do what was possible. Dylan was designated ‘sports reporter’ so that he could be included

during interschool team sports—instead of playing in the team sports, he provided commentary and ‘reported’ on the results of games. In this case, he was physically unable to participate in team sports and his teacher had made an effort to find an alternative for him to be part of the class activity. But he was unenthusiastic about it and would have preferred a way to actually join in—after talking about being sports reporter he said, “Next time I think I might make the t-ball team It’s easier, one of the easy sports that I can do that I think is at my level”. In this case, the negotiation for an alternative was not entirely successful from his perspective, but he had adapted his goals for inclusion to a specific sport in which his CP would create fewer limitations.

Following on from the frustration of limitations is the awareness of having to strive for improvements—both in functional outcomes and in leisure pursuits. Eric has to practice writing and cutting out because they are challenging for him, but he is also trying to improve his drawing because he enjoys it. This also relates to therapeutic goals, partly because therapists often use personal participation related goals (like achievement in sports) to achieve functional goals. A few of the children talked about the importance of the encouragement of others, including physical therapists, in striving for and achieving personal goals related to physical sports—this was especially true for the older children. Children with CP will face lifelong challenges because of their disability. Learning early in life the importance of persisting to achieve physical and other challenges is one way in which they will sometimes be able to overcome environmental limitations. There may be many things they can only do if they persist for longer than typically developing children.

6.3.2 Environmental factors of negotiating limitations

Environmental factors of negotiating limitations include basic aspects of physical mobility like surfaces and planning routes, as well as rules and regulations that may limit access. Environmental barriers usually related to reduced inclusion and participation, and for the children interviewed were mostly about physical aspects of disability, except for those with high anxiety. Negotiating environmental barriers may include knowing how far it is between places a child has to walk, or similar considerations: children may be able to walk short distances on level ground but

become quickly fatigued or unbalanced if there are long distances to travel and will instead use a wheelchair. This was the case for at least two children who were interviewed, who did not use their chair to navigate their home, but always used the wheelchair at school. Environmental factors need to be known in advance and planned for. Often this is work that parents do on behalf of their children, especially in finding out access to special places like beaches, disability-adapted playground equipment, or community recreation services. However, especially in a school environment where they do not have regular parental supervision, children must also manage this themselves. Much of children's knowledge of physical environments is taken for granted, yet they are also required to plan for and work around barriers in their everyday environments. Both Sofia and Dylan when asked "do you have any trouble getting around your school?" responded with detailed descriptions of specific areas they avoided, needed help with, or couldn't access, as well as the workarounds they used to reduce disruptions to their days.

Rules and regulations may form more difficult barriers for children—such as Eric's frustrations with school rules, first mentioned in the 'play' concept (see page 163). Similarly, Dylan recounted how a seemingly-accessible sports activity at his school left him frustrated because of rules he couldn't understand:

We had a man in a wheelchair come and teach kids at my school how to play sports in a wheelchair. And everyone thought I would be good, because I'm always in the chair. ... They brang [sic] wheelchairs for kids. And I wanted to use my own chair but they wouldn't let me. They wanted me to use one of the chairs they got. ... It was kind of harder to use the other one because ... it was harder to push. Because, my hands couldn't get like, right down to push at all. So, I could only really do tiny pushes at a time.

While he was able to participate fully in the game, because he was not allowed to use his regular chair that fitted him properly, he did not have an opportunity to excel. To him, the experience reinforced a pattern of missing out on fully achieving in physical sports because of rules that seemed to be unfair—even when it was a school-organised wheelchair sports activity.

Despite his regular participation in wheelchair basketball through a hospital program, and his general enjoyment of sports, Eric had a similar experience in organised sports at school, as he explained:

Instead of doing the fun things, like actually playing, I get to be the crappy scorer, all the time. Not even in skittles, I can play. And skittles uses arms. The one thing I'm good with! And the one time I did play skittles, I got hit in the stomach. But it didn't hurt, and so I just... the school decided I shouldn't play skittles anymore. I can't play any sports games. I can only do modified versions of what I've done millions and millions of times, over and over and over again.

The rules were created with the intention of ensuring his welfare (and/or limiting school liability) but do not recognise his abilities and therefore he finds them overly restrictive. For both children, rules imposed by their school create environmental barriers to their participation and ability to negotiate solutions to CP impairments.

6.3.3 Getting help

A few of the children mentioned the value in having identified adults they could ask for help—or the consequences of not having someone identified to ask for help. This was similar to the way they talked about valued adult relationships, and why those relationships were valuable. All children (and adults) at some point require help, but children with disabilities more frequently require more help than is typically offered to other children. This is captured by Sofia's remark "Remember how for some people they need help?".

As children age and develop greater independence from their primary caregivers, identifying when, how, and who to ask for help is necessary for them to succeed in their schooling and to participate in leisure activities. As with inclusion through alternative activities, the availability of willing help could overcome barriers children otherwise faced. Zachary had recently started at a new secondary school and said that what he liked about it was "The nice teachers. Like, if you're in trouble, they'll help you out and stuff". Sofia made direct contrasts between help available at her current school where "here the school is, like, it's psychologists... if you're feeling angry you go

to that person and they teach you strategies or have a chat with you”, and the previous school “But at that school there was no one”. She went on to talk about how difficult it had been dealing with her anxiety at her previous school because there was no help available; she often became so distressed she had to leave class and the only alternative was to spend time alone in the school office. Part of the ongoing management of her anxiety at the new school includes knowing that she is able to seek help, not only for schoolwork, but with the school psychologist.

Social relationships are necessary for help seeking and are heavily involved in negotiating limitations. In some ways, the connections are reciprocal: where a child can access alternatives or adaptations instead of being excluded because of physical impairments, this leads to additional opportunities for being social. Conversely, the presence of more supportive relationships often makes the external business of disability easier to manage, either in anticipating and planning for situations that will be challenging, sharing knowledge about alternatives, or providing physical help to enable a child to be included in social activities. Some of the children also mentioned regularly receiving help from peers and siblings, as previous researchers have reported (Schauwer et al., 2009), meaning social networks with other children provide direct benefits to negotiating limitations. Supportive and encouraging social relationships are also invaluable for children to continue striving for improvements and achievements, and to persist in goal-directed behaviours.

Negotiating limitations is closely related, and somewhat interdependent to establishing agency. Children need a level of agency to make these negotiations, but successfully negotiating for inclusions or adaptations can provide them with a sense of agency. Eric’s frustration with school rules preventing him from joining in with sports he knows that he can play leads to both exclusion and denial of his own agency. Children also learn how to negotiate with their environment from how their parents or caregivers do—where parents or caregivers were willing and able to advocate more strongly for disability inclusion, children learnt to do the same: Eric’s frustration with the perceived inequity in school participation directly reflected his mother’s expressed attitude in her own interview. These parallels are described further in Section 6.5 ‘Establishing agency’.

6.4 Defining the self

All the children interviewed, to a greater or a lesser extent, have a visible disability. Throughout their lives, they have experienced extra attention from family members, friends, health professionals, teachers, community organisations, and strangers, as a direct result of their disability (or disabilities). Seemingly as a result, they spend significant mental energy, consciously and unconsciously, on defining their self-identity with respect to their disability. Perhaps more than most children, they develop an earlier understanding of difference, and how being ‘normal’ can be socially protective. At the same time, they must face a reality that they are permanently ‘different’, and in some aspects of their lives, will always fall short in peer comparisons. Throughout their childhood their work in defining themselves is framed through the presence of their disability—and the knowledge of how this makes them different. The sub-categories of ‘defining the self’ include making comparisons, self-esteem, self-awareness, internal self-concept, and external social identity. More detail on each of these is given below.

Many of the ways in which children define or describe themselves involve making comparisons with others. Mostly these comparisons are made to siblings and peers. Ana spoke about her afterschool hobbies and activities but emphasised that her older sister “plays heaps”, saying “She plays the most, I play a few”. Missing out on participation opportunities compared to siblings was a common experience. Sofia mentioned her two siblings’ multiple hobbies, and followed with “But, I just go to swimming... because there’s not that much stuff to do for me [sic]”. Often it was clear that they were both proud and envious of their siblings. They were happy to tell me how much their beloved brother or sister could do—but at the same time, they have already realised that their physical disability has shut them out of many community organisations. The difference between the two conversations with Ana and Sofia reflects the differences in their own activities. Ana was happier to make these comparisons because she has been involved in cross-country running (including ‘handicaps’, i.e. scoring differences to provide athletes of different ability opportunities to win) and modified horse-riding—both hobbies she enjoys. In comparison, Sofia has been limited to swimming which she was not enthusiastic about

discussing and has not been involved in any other disability specific hobbies. She has accepted that she cannot join dance classes as she would like to, while her brother and sister are both pursuing their preferred hobbies: she reports missing out with a great deal more sadness. This further reflects a connection to the outcomes of negotiating limitations: successful outcomes result in more favourable comparisons with peers.

Children not only compare the number of activities that they can participate in contrasted with others, but also their experiences of activities, achievements, play, friendships, and many other aspects of their lives. Children with milder CP may be in an especially difficult position: they all have some form of disability—some of them multiple disabilities—yet the majority of their peer experiences occur in the context of typically developing children’s milestones. Perhaps more than most, their comparisons result in them evaluating themselves as falling short and missing out.

Flowing on from the outcomes of successfully negotiating limitations, these ongoing comparisons highlight the benefits of adapted activities and disability specific leisure services. Through adapted activities, not only do children experience inclusion and increased social opportunities—they make more favourable comparisons between themselves and their peers and can experience a sense of achievement not otherwise possible. To quote Zachary talking about being part of a disability specific athletics carnival: “I just like competing. It feels good when you come first”. Like Ana, this put his other experiences of missing out in a more positive context. And like Ana being given a handicap to compete in cross-country running, he would not have made his athletics achievements without disability specific adaptations. Caleb began Grade 1 at a mainstream school but did not integrate well socially and within the first year switched to a special school for children with intellectual disabilities, where his mother reports he is much happier. When talking about activities he does at his current school he mentioned, “We jump over hurdles and there was basketball. I got to win!”. Whether it is creative, physical, or other pursuits, the value of opportunities to achieve—for children used to falling behind—cannot be overstated.

Following directly from achievement experiences, a primary developmental goal of identity work is building self-esteem. Measurable achievements in physical activities

help build self-esteem—but so also do creative pursuits. Creative hobbies may have the added benefit of fewer barriers to inclusion than physical sports. Several of the children became animated in discussing their creative hobbies and reflected pride in their achievements. For example, Dylan spends time making short movies with his brother and friends. Eric took pride in his drawing during the interview and mentioned other drawing and building projects he was working on during the school holidays. Gus was anticipating a transition to secondary school and said that his main hope was that he would be able to continue being involved in theatre at the new school. All of these hobbies provided opportunities for the children to enjoy developing skills and feel a sense of competence and achievement in their results.

As well as achievements, children rely on social and emotional support to develop self-esteem. Again, children with disabilities are at a disadvantage: they already have higher risk of social exclusion and bullying from their peers. The ways in which three of the children responded to bullying in building self-esteem were interesting. Gus already seemed to have a strong positive self-identity around his CP, so with his mother's support he tackled name-calling directly with a presentation to the school about CP. He directly answered the bullying experience by advocating for disability inclusion and this became a source of pride for him. Ana has new friends who accept her, and talks about her previous school saying, "first of all when I went there, I didn't know there were bullies and I hung out with them". As she talked about the experience, she avoided eye contact and was clearly still upset but has internalised their actions as resulting from their shortcomings, not her disability. Sofia talked in detail about verbal and physical bullying she had experienced at school; her feelings about it are summarised in these quotes: "people were being so mean to me ... I get angry about being bullied... But, I don't bully or hit people.... I couldn't do that". Both Ana and Sofia have been hurt by bullying and are rebuilding their own self-esteem by externalising their experiences to the actions of others, and internalising positive ideas of themselves as people who are kind and would not bully.

Several of the children also talked about close, supportive relationships with aides and allied health professionals mentioned in 'Social beings; being social' (Section 6.1, page 154). As well as contributing to functional goals, relationships contributed to good self-

esteem through helping children to make positive self-appraisals about themselves and/or about their disability.

Just as knowing the limitations caused by CP was necessary to overcome them, self-awareness of what is not CP seemed equally important to children's developing identities. An interesting pattern developed when the children talked about themselves and their physical experiences. They frequently drew sharp distinctions between physical experiences related to the CP, and other physical sensations. This was clearest when they mentioned being sick because of illness. For example, early in his interview Dylan chose an emoji with a bandage on its head to represent himself, but emphasised in his explanation of why he had chosen this "Because I've got a bit of a flu this week ... I'm not really injured. It's just the flu". Even if they were sick more often (Sofia mentioned recurrent chest infections), being 'sick' is defined not only as outside the physical experience of their disability, but a common part of life—a 'normal' childhood experience.

It is the duality between normal and difference that is the context of developing an internal self-concept. Beyond catalogues of hobbies, friends, likes and dislikes, this is an aspect of identity children struggled to assimilate. Ana captured the feeling when talking about receiving attention from strangers in public places: "People stare at me. And I get like so annoyed, 'cause I'm like, I only have a disability—it's not like I'm an alien or something!". She humorously acknowledged both that she is visibly different, but human just like everybody else. This was a theme repeated by other children, of defending their difference. A tension developed in their self-concept between wanting to be like everyone else, while continuing to need to assure themselves and others that it is ok to be different. Other examples of the experience and presentation of difference were described in the previous chapter in Section 5.3.3 'Disability knowledge and identity positioning' on page 145.

The final sub-concept of defining the self is that of the external self-identity, or social identity, especially around being a child with a disability. This is how a child chooses to present themselves to others—which partly reflects their self-concept and self-esteem. An example of this from the interview process was children first presenting themselves

as reflecting a 'happy' emoji, and only after establishing a positive identity, providing richer reflections on their lives. This reflects some of the same aspects of agreeableness that were mentioned in children's difficulty in answering some questions (Section 5.3.2, page 138) but include the wider social context of children's lives. Presentation of a social identity necessarily relates to a child's social activities—especially in the comfort that can be derived from long-standing relationships. Children mentioned having to explain to new friends and new peer groups the basics of CP, generally as a negative experience. In contrast, in longstanding relationships a child's CP did not need explanation, and there was greater latitude to consider their abilities foremost, rather than their impairments. On the other hand, in a few cases, children and their parents discussed the child's visible disability as a useful tool in some ways—Gus' mother referred to it as him being "the golden child". In this case, strangers' reactions to the disability enabled them special access, or additional considerations that they would not otherwise receive. In this way, the inescapable visible identity of having a disability is not only used for benefit and to provide accessibility, but can provide a stronger, positive association with the social identity for the child.

The outcome of the definition of self relates strongly to how children act out their sense of personal agency. On the one hand, children in contexts that have supported a greater sense of agency are more easily able to establish 'positive' self-concepts and are likely to have higher self-esteem. On the other hand, the frustration with situations beyond their control may result partly from a higher sense of self-esteem and greater expectations of agency that are not borne out. How children establish a sense of agency over themselves and disability contexts is explored next.

6.5 Establishing agency

The final major conceptual category that emerged in the interview findings was children establishing a sense of personal agency. This is both the belief that through their personal actions, they are able to create changes in their lives, and their sense that they have the right and ability to define what happens to them, and their bodies, in their everyday lives (United Nations, 1989). Establishing a sense of agency may be

seen as a significant developmental goal. Some children appeared to have a high level of agency, and some a low one. Agency is not synonymous with autonomy or independence, especially for those children who are physically dependent on others for basic needs. For children with CP, agency may be eroded by diminished control over their own bodies, but also by reduced power over others' interactions with their bodies in the context of health care and medical interventions. The sub-concepts that inform 'establishing agency' include: the family context and roles, especially in navigating disability systems; the role of the child with parent-child and other trusted adult relationships; managing expectations of adults and others; and managing emotions.

There were examples of differing levels and development of child agency directly through interactions within the interviews. These included interactions between the child and the interviewer, as well as those observed between the child and their parent. Some of these interactions impacted on the research process, including the child consent process. For example, a child with a low sense of agency may consent to the interview despite limited interest in the research, and respond with low levels of engagement, rather than refusing outright to participate. This appeared to be the case with Nicholas, who struggled to engage with the research process, frequently declined to respond to questions, and at times appeared bored. He responded by creating new games related to the drawing activity, leaving the conversation to play with other things in the room, and otherwise 'acting out'. However, he did not directly challenge his mother's admonishments to answer or not "be silly" and did not directly suggest to the interviewer until the end of the interview that he no longer wanted to talk.

Family is especially influential in children's agency, generally and in a disability setting. The family is largely where children learn roles and model behaviour on how to deal with health care and other systems. For both parents and children, the presence of a child's disability may erode a sense of agency, as from a very young age child's functional outcomes and expectations are placed in the hands of health professionals. For parents, the first steps of this journey are often traumatic, and continue to be so through many future transitions from early intervention services, to children beginning primary school, and moving to secondary school. For young children, advocating for a

child's perspective and a child's voice in the healthcare system falls to the parent—who may or may not feel they are able to achieve this. Where parents experience frustrations and disappointment due to lack of access to appropriate services, children may experience this twofold—through missing out, and through observing their parent's inability to advocate for them. Sofia had recently been ill when she was interviewed, and raised her disillusionment with healthcare, saying:

One doctor was like “She can go to school tomorrow” ... And, then everyone believed that doctor ... doctors don't understand how much sick I was feeling... but when you go back home you get a lot sicker... They just think that you're feeling a little bit sick but you're feeling a lot sick.

Earlier in the interview she mentioned that doctors in her parent's home country were better, saying “everybody were saying, like, the doctors are not good here in Australia. Because in [other country] doctors are good because if you have a problem immediately they'll do something... they'll fix it, quickly”. Some of what Sofia said about doctors in Australia sounded like a repetition of what she had heard from her mother or other adults in her family. Her resulting conclusions were a reflection not only of her own distressing experience of lingering illness, but her mother's frustration with limited access to healthcare and other services. Frustration with lack of accommodation, lack of understanding and lack of choice through systemic bias toward the able-bodied contributes to a systematic erosion of agency that goes beyond what most typically developing children experience.

Within familial relationships, it is usually with parents or other highly trusted adults that children are first able to enact their own sense of agency in response to authority. Those who have a network of trusted and trusting relationships, and a strong sense of belonging and security, are likely to feel more able to first act in their own interests. Some of these differences were apparent in the interviews, as touched on when reflecting on parent-child interactions in the research process (see Section 5.3.1 ‘Parent-child interactions’, page 134). Through their actions in the interviews, all of the children appeared to have a valued, supportive relationship with their parents; their parents demonstrated through participation in the research that they willingly devote

additional time and effort to advocating for their child's wellbeing in relation to their CP. However, even within these limitations, there were differences in parental expectations of child agency—which in some ways reflected parents' own expressions of their sense of agency related to the child's disability, discussed in the previous page. There appeared to be multiple mechanisms impacting children's building sense of agency—children observe their environment and their parents' interactions (and frustrations) with external systems, children have varied familial relationships where they are allowed varied amounts of agency, and children learn how to assert their agency in disability-specific situations from their parents' behaviours.

In this sense, child agency was also directly related to getting help. For the children interviewed, it appeared that those with a sense that they were capable of acting actively sought help. Conversely, children who received 'too much' help, such as those with school aides who did a great deal of work for them, may have then developed more of a sense of helplessness, and correspondingly, lower agency. This has been observed in young children with disabilities, with researchers describing lower levels of engagement and greater passivity in a settings in which adult actions were more directive with limited opportunity for child individuality in response (Nind et al., 2010). Children's ways of enacting agency were nuanced and responsive to their perceived control in the setting. This may have been the case with Nicholas, who generally seemed to have a lower sense of agency but responded to this in ways that did in fact allow him some control—such as his response to research involvement. He mentioned he had an aide at school, and when asked what she helped him with he said, "All of my work". His mother responded, in a disbelieving tone, "All your work?", to which he replied, "Normally I let her do all the work". The way he reported it, it seemed he was happy that she helped him, but also ready to exploit this situation so that he didn't have to complete his own schoolwork. While he otherwise did not appear to feel able to directly challenge any authority or was used to having these challenges ignored, he may have found a way to exercise his agency by manipulating his aide to do schoolwork he didn't wish to do.

The corollary of children's ability to enact agency in their relationships with trusted adults is the way in which they manage their own response to the expectations of

those adults. These include expectations in relation to self, changes to impairments, desire for inclusion, activity preferences, and more. When Zachary was asked if he had a chance to discuss with his physiotherapist his preferred goals or activities he explained:

Well, most of the time I will do what they want to but if I'm not comfortable with doing something I will say, like, "I'm not really sure about doing that. Can we try something else?", but if I have to do it I'll just try and see how it goes, I guess. So, if I do end up liking it, I'll do it.

This example reflected his awareness of his own agency in his current relationship with a supportive physiotherapist, and the limitations to his own preferences in therapeutic goals. Not long after that he mentioned how it might be different if it was your first time visiting a physiotherapist and "you could be really nervous and you just don't know how it's going to go.... Now that I've gotten used to it I know what's [expected]". This suggested not only that he had adjusted his own expectations as he became more experienced with physical therapy, but that it allowed him to change his response to others' expectations as he became more confident.

At some levels managing adult expectations is related to CP and/or disability, but it may also be universal for children. That is, all children at different points become aware of adult expectations that in some way do not match their own personal goals or preferences. Sofia referenced these discrepancies a few times in regard to social situations: she does wish she had more friends, but she is happy with a very small social circle so long as she has opportunities for play. At the same time, she is aware that adults expect her to have a much higher level of social engagement in more diverse situations. Other children were managing adult expectations directly related to CP—for example, adults want them to participate in more physical activities when their personal preference may be to not do so much—especially where they recognise their own limitations.

Children with CP may face greater conflict between establishing personal agency and adult expectations for two disability-specific reasons. Firstly, due to additional rules and restrictions related to accessibility, such as those described in Section 6.3.2

‘Environmental factors of negotiating limitations’. Secondly, most children with CP are involved in some level of physical therapy that demands participation in additional goal-directed activities and exercises that are outside of the child’s control and may restrict participation in their preferred activities. These two conflicts then relate to a narrative of being different, continually being regarded as ‘other’, strange, or alien, usually in negative ways involving limited personal choice, which can negatively affect self-concept and self-esteem and decrease personal agency. The way that children reflect on adults’ expectations of them, and how children incorporate adult expectations into their sense of self impact both establishing agency and the previous category, ‘Defining the self’ (Section 6.4, page 171). For example, if adults expect a child to perform poorly, or be unable to contribute to an activity, because of the child’s disability, the child may in turn hold fewer expectations for themselves. If attitudes towards their disability are consistently couched as impairments to be treated and fixed, children may struggle to form positive ideas of what it means to be a child with a disability. Several of the children interviewed saw little value in repeated physiotherapy exercises at home and older children were less likely to engage in this practice—both Ana and Zachary had side conversations with their mother about not completing all their exercises. Children reported these tasks to be onerous and boring—Ana rolled her eyes at her mother over this—and felt that exercises may have little relationship to functional outcomes. Being expected to engage in disability-specific ‘work’ when they would rather play then becomes part of the narrative of the cost of being different.

Similarly, positive experiences of self are likely to build a sense of personal agency. Demonstrating again the interdependency with ‘negotiating limitations’—children whose successful negotiations have resulted in increased opportunities for achievement will have increased opportunities to build personal agency. If they can achieve, then they can also be in control of outcomes. Outside of recreational achievements, undertaking disability advocacy is likely to be an alternate opportunity to re-establish agency. For example, Gus’ presentation about CP to his school allowed him not only to present a positive image of disability to himself and others, but also to establish that he has some control over that image.

The final sub-concept in establishing agency is the most individual—children learning to manage their own emotions. All of the children indirectly mentioned this in relation to ‘stress’ or everyday emotional situations. Eric independently raised a need for relaxation if he felt too emotional or fatigued—he already knew how to manage his own emotional needs. Similarly, when Ana was asked how she dealt with some negative emotions she mentioned “having a bath or patting my cat” as her two primary strategies. For those children with high anxiety, management of the anxiety was a primary consideration before they could be in control of their external situation. Successfully managing their own emotions is likely to increase children’s agency, not only in feeling in control of themselves, but in how they are able to respond to situations. Conversely, rules and environmental barriers creating additional restrictions, without any option to enact their own agency, are likely to lead to greater frustrations and stress for those with a greater sense of agency. How greatly their bodily limitations frustrate them may prompt both additional attempts to establish a greater sense of agency—or engender a sense of helplessness.

6.6 Discussion

From interviews with children with CP, the analysis in this chapter showed that there were five areas contributing to the children’s QOL: socialising, play, negotiating limitations, defining their self-identity, and establishing agency. These five conceptual categories were interconnected, with children’s social relationships forming the context for achievement of other goals, especially play, and interdependence between how children negotiated limitations and how they establish a sense of agency over themselves and their environment. Achieving their QOL goals within each area was a result of interactions between the child and their environments, often through key relationships with family members, peers, and other adults in their community such as teachers, aides, and health professionals. Within each category, children’s CP affected their QOL, through the need for assistance or adaptations for inclusion and participation, and through how children managed their own and others’ attitudes and reactions to their differing abilities.

Overall, there are parallels between the conceptual categories developed in this chapter and those derived in two other qualitative projects on QOL in children and teens with CP. Researchers in the UK interviewed children with CP aged 8 to 12 years, and their parents, to identify their priorities for child QOL, and how these compared to two existing measures of QOL, the KIDSCREEN and the CP QOL-Child (Parkinson et al., 2011; Young et al., 2007). Their analysis yielded four overarching themes of social relationships, home and school environments, self and body, and recreational activities and resources. Although the balance of themes differs from the categories of this chapter, aspects of each were similar. Children in both published studies valued the support of wide and varied social networks, having choice in their recreational activities, fairness in participation and inclusion, and achieving their goals in a range of activities. In both published studies the authors reported in each main theme aspects of children's frustrations that mirrored the concepts described in this chapter of children's challenges in developing self-identity and establishing agency. In both papers the authors reflected on the high priority children placed on a network of social relationships within and outside their family to support their QOL. Similarly, in this chapter social networks and socialising framed the context of children's other QOL goals.

In another research project, Shikako-Thomas et al. (2009) used constructivist grounded theory methods to analyse interviews with twelve adolescents with CP (aged 12-16 years), and highlighted the interaction between person and environment in the resulting findings. There are many similarities between their model and the findings of this chapter, especially regarding the importance of activity preferences, managing limitations, and sense of self as an interaction between personal reflections and external perceptions. Social relationships were broadly defined in the same way as well, including family, peers, and other community relationships forming a broad network that provided support for good QOL. The emphasis by Shikako and colleagues on the interaction between 'intrinsic' and 'extrinsic' factors in QOL is more explicit than in many previous QOL models and is reflected in details of conceptual categories in this chapter, such as the way that children's agency results from both internal

appraisals of competency and environmental factors that support or suppress children's opportunities for decision-making.

Beyond these overall models of QOL, the links can be seen between the findings of this chapter and existing qualitative literature on children's lives. Children with disabilities, including those interviewed here, frequently face barriers to participation and inclusion. This has been widely discussed in previous research. Two articles of the CRC—Article 23 and Article 31—specifically recognise the need for support for children with disabilities to actively participate in their communities with appropriate assistance or adaptation, and to have equal recreational opportunities (United Nations, 1989). The findings presented in this chapter indicate that these obligations are not always met to the satisfaction of children with CP, as has been shown in previous research on the participation of children with disabilities in recreational activities (e.g. Majnemer, Shevell, Law, Birnbaum, et al., 2008). Researchers focusing on participation of children with disabilities have shown both strong correlations between children's participation in leisure activities and QOL (Shikako-Thomas et al., 2012), and bidirectional relationships between person and environmental factors in supporting children's participation (Imms et al., 2017). Children's perspectives described in this chapter of experiences with participation and inclusion at school and in the community echo both of these links. Another qualitative project on a small sample of children with special education needs (Mortier et al., 2011) reflected similar findings regarding children's opinions of their supports for inclusion and participation. In both that research and this thesis, children reflected on the potential negative impacts of mismatches between their desired goals and offered help and reported that accommodations at school did not always mitigate disabling environmental barriers. In a discussion of interventions for the inclusion of children with disabilities in play, Mindes (2015) emphasises not only the need to balance functional goals and leisure goals for children, but shows that if aides and interventions are too directive and do not allow children choice, they fail to replace children's spontaneous play or facilitate skills to initiate play.

Although there is very limited research on developing a sense of agency for children with disabilities, there is some support for the links established here. For example,

Mortier et al. (2011) emphasised the need for children to maintain rights to choice even with the use of school aides, as did Mindes (2015). The current analysis demonstrated that children's experiences of decision-making in their lives were directly related to their developing sense of personal agency. Other authors have drawn on research with children with various learning, behavioural, and attentional disorders to theorise that children's personal agency relies on the combination of beliefs of personal competence and perceived control (Martin et al., 2017). This is mirrored by the strong links in these findings between the concepts of 'negotiating (physical) limitations' and 'establishing agency': for children with CP, personal agency developed from achieving both personal and participation goals, reflecting a sense of competence and a sense of control over their lives. Other researchers have focused on child rights in research participation have highlighted the importance of respecting children's choices in supporting their developing sense of agency and upholding child rights (Lundy, 2007). Lastly, in a model of QOL for people with intellectual disability, Schalock et al. (2016) outlined 'enhancement strategies' for the QOL concept of 'empowerment', that included decision making, goal setting, and self-management—this is reflected by the conceptual category of agency in this chapter and the sub-concepts that contributed to establishing agency for children.

The reciprocal links between socialising and play echo ideas of Missiuna and Pollack (1991) in research on play participation for children with disabilities. In discussing the consequences of exclusion from play due to disabilities, these authors referred to primary and secondary deprivation—primary deprivation refers to missing the activities of play themselves, while secondary deprivation refers to missing opportunities for social or skills development as a part of play. The findings in this chapter extend this link by showing its reciprocal nature—which can become a downward spiral. Children with CP may be excluded from social opportunities due to their disabilities, which in turn leads to missing opportunities for play and recreation. Other authors have written eloquently on the ability of children with various disabilities to engage in active and creative play (Burke, 2012), however if children are physically incapable of joining play spaces in their school or other community settings,

as was demonstrated by some children in this thesis, they may miss opportunities for play, socialising, and other skills development.

In an extension of the idea of secondary deprivation, Mindes (2015) links play participation to developing self-identity, suggesting that if children's experiences of play and participation are usually of being 'inept' or being excluded, this is likely to negatively influence their self-concept. The findings of the chapter similarly showed that children's experiences, not only of participation but of achievement and winning at games, were tied to their self-identity. A recent project to develop a measure of self-concept for children with CP has uncovered similar links (Cheong et al., 2016). In qualitative work with children with CP aged 8-12 years and their parents, and health professionals working with this population, the researchers identified several domains which contributed to self-concept for children. These covered similar domains to self-concept for typically developing children but with an additional focus on participation, persistence, valuing supportive relationships and interpersonal skills (e.g. ability to make friends), and incorporating ideas of disability and difference into positive self-identity. Furthermore, the measure of self-concept developed from this work encompasses many of the same links as the findings of this chapter (Cheong et al., 2017), though the perspective is shifted to focus on children's reflections of self rather than feelings of wellbeing.

6.7 Summary

The analysis conducted here used constructivist grounded theory methods to analyse eight in-depth interviews with children with CP aged between eight and twelve years. The analysis derived five main conceptual categories: social beings, being social; play; negotiating (physical) limitations; defining the self; and establishing agency. Interdependencies and interactions were identified between these categories, with socialising providing context for children's experiences and strong reciprocity with play, and interdependency between negotiating limitations and establishing agency. The categories reflected children's day-to-day experiences as well as developmental goals as they approach teen years and sought increased independence and built identities and networks outside of the family home. In every category, children's

experiences and outcomes were a result of interactions between themselves and their environment. Sub-concepts within the categories reflect existing literature both about children with disabilities and typically developing children. Overall, the conceptual categories identified in the analysis have strong similarities with findings of research with adolescents with CP (Shikako-Thomas et al., 2009), while the way children presented their experiences and negotiated for their QOL goals through social interactions reflected themes of the social-relational model of disability (Thomas, 2004a). How these findings fit with broader models of disability is explored in more detail in the general discussion in Chapter 9, following the analysis of the parent interviews in the next chapter and comparison between child and parent findings in the following chapter.

Chapter 7: Parent interview findings

Following the presentation of findings from child interviews in the previous chapter, this chapter presents the findings from parent interviews. These include those whose child was interviewed, and those whose child did not complete an interview. The first part of this chapter, Section 7.1, describes some family characteristics of the sample, with reference to the descriptions of children in the previous chapter (see Section 5.1.2, page 111), followed by reflections on the interview process in Section 7.2 and how it influenced the data collected. The overall map of the conceptual findings is described briefly in Section 7.3. The larger portion of the chapter, Sections 7.4 to 7.11, describes the findings of the analysis of these 18 parent interviews. Lastly, Section 7.12 draws parallels to wider literature and theory exploring experiences of parents of children with disability.

7.1 Sample and interview setting: parent interviews

Eighteen families completed the interviews. Ten interviews were parent-only, and eight included both a child and a parent interview; a total of 26 interviews were completed. Parent interviews ranged from 33 to 75 minutes in length (mean= 51.3). The children of the parents interviewed were aged from five to 12 years old (mean=9 years 1 month), and had a range of comorbidities including epilepsy, intellectual disability, and communication impairments. The children represented all GMFCS levels; including the eight children interviewed directly, there were four at GMFCS I, four at GMFCS II, five at GMFCS III, one at GMFCS IV, and five at GMFCS V. There were 11 boys and seven girls. Three children in the sample had fraternal twins without CP.

Six parent-only interviews were conducted at the family home, another two between appointments at the Royal Children's Hospital Melbourne, one by Skype, and one by phone. All eight family interviews that included a child interview took place in the family home. The interviewer was able to meet the child during six of the parent-only interviews; in four other cases, the child was not at home at the time. Of the six parent interviews in which the child was present, the child was present throughout the interview in only three. Two of those children were at GMFCS level V with nonverbal

communication, such that parents did not appear to feel constrained in discussing their child though the child vocalised directly to the parent and the parent responded to them a few times in both interviews. In one interview the parent stated that their child had responded to talk of their favourite foods. The third interview in which the child was present throughout, again, the parent did not appear to feel constrained in discussing aspects of parenting, and in a few instances sought additional detail of clarification from her child (who was too young to interview directly). In all cases where the child was introduced but not interviewed, it appeared to facilitate the discussion in that it was easier to talk about a real person who had been met, rather than an abstract child. In one other family in which both the child and parent were interviewed separately, the parent asked the child to leave the room at the beginning of the parent interview so that it could be conducted privately. This suggests that parents probably made deliberate decisions on how comfortable they would be discussing their child with or without them present—the interviews were always scheduled at the convenience of the participants.

In sixteen families the child's mother was the primary caregiver and completed the parent interview alone; in one case the father was primary caregiver and completed the interview alone; in one case the mother (the primary caregiver) completed the interview with the child's father present and contributing for most of the time. During several of the interviews at family homes, siblings of the child involved were present during some parts of the interviews. In only one case did the sibling contribute remarks at the end of the parent interview, as he was invited to do so by his mother.

Contributions by siblings to child interviews are described in the previous chapter, Section 5.1.1 page 111. Overall, siblings did not contribute to interview content. However, their presence did illustrate the other commitments that parents balance, both to take part in the research project, and to provide a higher level of care to their child with a disability. Several parents commented explicitly that they regularly accepted invitations to participate in CP-focused research in the hope of improving the care and wellbeing not only of their child, but also of others with CP.

All eighteen families lived within the greater Melbourne metropolitan region, but their locations varied from inner city to outer suburbs, and in one case, a regional town. As

disability services in Melbourne are region specific, service access necessarily varied for families depending partly on location as well as the severity of their child's disability. All parents raised issues of cost for services or equipment at some point during the interviews. However, individual financial circumstances varied, such that some parents mentioned paying for additional private services, whereas for others these were unaffordable, and they relied on government provided services. This was related to parental employment. Several of the mothers interviewed were fulltime carers for their child and did not work. Two mothers were single parents with limited parental involvement of their child's father. One family included step-parents, with co-parenting of the child with CP. In the remaining 15 families, the child's mother and father were still partnered with each other, either married or living together. At least two families spoke English as a second language; another family was multilingual. Overall, cultural background and language differences were not raised as significant additional issues for these families in the context of caring for a child with a disability, except that these families were geographically distant from extended family and support systems.

7.2 Reflections on the interview process

Interviews with parents followed a traditional interview format and did not include any additional methods as in the child interviews. However, as in the child interviews, the process may have been influenced by the insider/outsider perspective difference between the parent and the interviewer. That is, parents identified to some extent as part of the 'disability culture', of which the interviewer was not part.

Early in the interviews, parents appeared to signify their disability knowledge with the use of medical terminology. Often the opening question in the interview was "Tell me about your child". Parent responses usually included a narrative of how their child came to have a CP diagnosis, including recitations of specific medical terms that the parent sometimes explicitly signalled they did not understand.

Most parents have significant experience in repeating their CP story and discussing various aspects of their child's health and disability with multiple health professionals. Many similarly have significant experience of research participation. In this context,

the use of specialised medical jargon appeared to serve two goals. Firstly, to show the parent's expertise and establish a level playing field with a researcher whom they assumed to have medical expertise in CP. Secondly, it may reduce the need for further explanation of their child's circumstances with new clinicians by early establishment of the known facts. Parents appeared to equate the researcher with their child's clinical health professionals and approached the research interview as they would other clinical discussions. This meant beginning the interview by displaying their knowledge about CP, thereby showing that they can be included in conversations and decisions on health care. Parents' perspectives of learning more about CP during the time of their child's diagnosis is discussed in greater detail in Section 7.4.1 (see page 194).

Unsurprisingly, several parents were clear in their own goals for participation in the research—that the findings would result directly in changes to existing disability services and policy. To that end, the research interview was an opportunity for advocacy by identifying specific areas for improvement. While this may not directly reflect a child's personal QOL, access to community and disability-specific health services is a factor in QOL, and for parents such access was directly linked to both child and family wellbeing. Many of these wider issues are therefore reflected in Sections 7.5 'The business of disability' page 200, 7.7.2 'Enabling child goals' page 218, and particularly in 7.9 'Disabling vs enabling environments: the value of knowledgeable, timely support' page 223. The inclusion of wider areas of concern to parents reflects not only parents' own agendas that they brought to the research, but the interview style that was used which was largely open-ended, with questions framed as prompts to discuss domains of life raised in previous research, such as schooling, socialising, or the future. If parents spontaneously raised additional issues, these were given equal weight in the interview and subsequent analysis to the planned topics from the interview guide. A more structured interview with parents focusing only on children's immediate QOL may have resulted in conceptual categories for parent and child data that were more aligned to each other but would have excluded a significant amount of data reported by parents to affect their child's QOL.

Aside from this wider background, parents often presented and emphasised a narrative, at times explicitly, of their child as happy, and not different from typically

developing children in their interests. For example, after a brief summary of their child's CP symptoms, one parent stated, "but aside from that, he's a very happy child". Another parent discussed their daughter's love of 'princess' movies saying, "she's a typical girl in that sense". This was often raised early in the interview, before probing led to parents providing more detailed accounts of children's day-to-day lives. At times it seemed related to a parental narrative of the acquisition of their child's CP emphasising factual elements of the medical condition as evidence that it was not a result of parental fault. Both of these narratives were provided early in the interview, as essential background information before a parent discussed greater details of their child's life. These two narratives relate to the conceptual categories 'Assimilating diagnosis' (page 194), 'The saving grace(s)' (page 196), and 'Loving my child' (page 216)—and to how parents work on building their child's self-identity, either explicitly or otherwise (discussed in the child findings Section 6.4 'Defining the self', page 171).

7.3 Mapping the conceptual categories

The focus of the parent interviews was the child's QOL, however discussions encompassed other aspects of family life. These included both practical issues, as well as social-emotional aspects of parenting, such as parental mental health, or loss of social and employment opportunities due to carer burden. Such factors are not considered to be strictly part of child QOL, but they influence and form the background for children's QOL. Conceptual categories can therefore be separated into those related directly to children's QOL, and those of parent and family QOL. A third area emerged in the analysis which links to these two but which is a dynamic, central part of life and QOL for both children and parents: valuing knowledgeable and systematic support, personalised for a family's individual circumstances. The presence or absence of support can create either disabling or enabling environments.

Parental interviews often presented a narrative of their child's life beginning from infancy and CP diagnosis and progressing to the present day and beyond – a logical biography of their child's life, and their own parenting journey. Analytical categories therefore align with a temporal context: moving from the knowns of the past, through current day-to-day concerns, to the unknowns of the future. While categories that

grace(s)', such as celebrating their child's joyful moments, were an active part of the later category 'loving my child', while others were quite separate. Positive and negative signifiers in arrows leading to 'parental mental health' indicate how the concepts of these categories were part of either positive or negative aspects of parental wellbeing. Where negative conditions were salient, parents overall described difficulties in maintaining good mental health – but this was tempered by consideration of categories with positive links to parental mental health.

One of the main points of including the parent journey in this analysis of child QOL is not just that it frames how parents discuss their parenting joys and challenges, but that the outcomes of these categories have significant consequences for child QOL. Especially around the time of child diagnosis, if parents do not have sufficient support to manage their own mental and emotional health, and the other demands of their lives, then they have fewer resources to spend on their child. However, the converse was also true. Parents who faced greater challenges in the areas directly related to child QOL may then be less able to successfully manage their own and other family goals.

7.4 The first steps

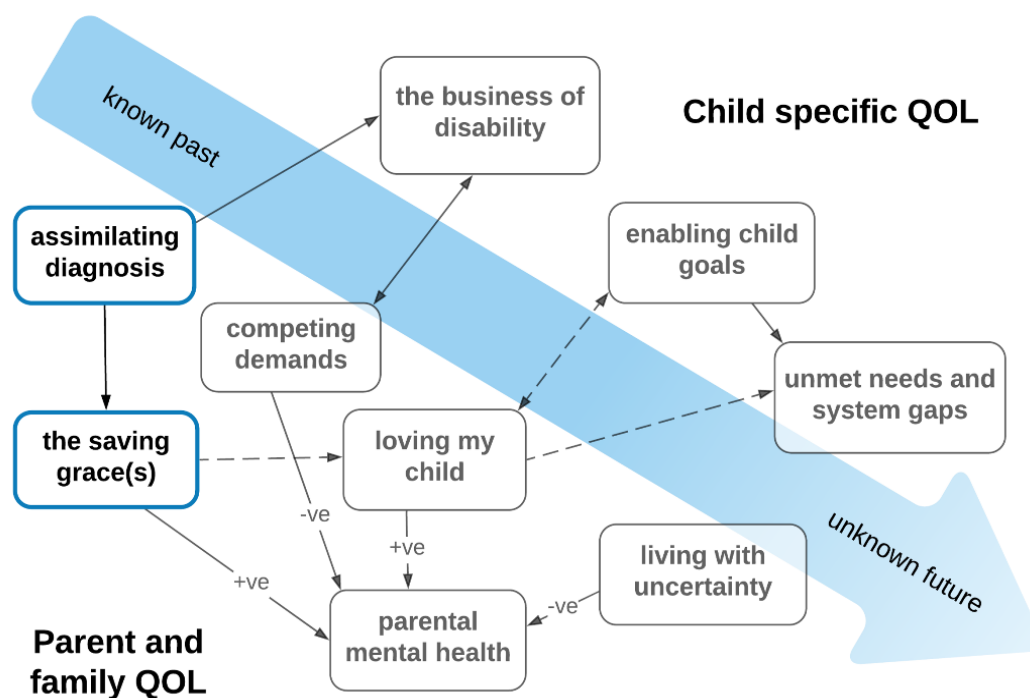


Figure 8. Parent findings conceptual map: The first steps

Following the main categories chronologically through the child's life, the first steps on the parent journey of caring for a child with a disability are around the time of diagnosis. These involve how parents understand and accept their child's permanent diagnosis, and adjustments they make to their expectations of a child's development, as described below. Both of these categories focus largely on events of the past, as shown above in Figure 8.

7.4.1 Assimilating diagnosis

As part of the diagnosis journey, parents formed a narrative of how things had happened related to normal and non-normal development, trauma, understanding why the CP occurred, and adjusting their expectations going forward. Assimilating their child's diagnosis combined three related elements: a process of understanding what CP is and why it occurs; accepting that CP is a permanent diagnosis with ongoing implications for their child's life; and lastly, the recollection of their child's diagnosis as a time of parental trauma which may continue to overshadow aspects of caring for their child.

7.4.1.1 The process of understanding what and why

In the context of research about children with CP, parents often interpreted the opening question, "tell me about your child", as "tell me about your child's CP". Parents therefore usually responded first with comments such as "well where to start" [father of a 5-year-old boy, GMFCS I], then began a seemingly well-rehearsed story about how their child was diagnosed with CP.

Several of the parents used medical terms that they had probably researched but didn't seem to fully understand (for example, they asked the interviewer if they were using the correct term, or to confirm an acronym such as GMFCS). The use of very specific medical terminology is striking. Parents were repeating terms usually several years after they had first heard them, which may only have relevance to other health professionals, or those with specific CP knowledge. Parents appeared to be repeating the diagnosis story in specific terms because they were trying to understand and assimilate what can be a devastating diagnosis—initially to understand what has happened, and later to be able to explain it in the official terms.

It is possible that parents focused on learning the formal medical language in order to break down some of the divide between themselves as parents and the hierarchy of medical professionals who have been involved in their child's life, in some cases, from birth.

7.4.1.2 Accepting the permanence of CP and finding the exceptions to impairment

The other aspect of learning medical terminology reflected the way that parents themselves have assimilated the diagnosis into their own expectations of their child—and the potentially traumatic history of discovering their child's CP, related grief or disappointments, and adjusted expectations of parenting. Accepting the permanence of a CP diagnosis moves the focus of healthcare interventions to prognosis of functional outcomes. Furthermore, it gives parents an opportunity to start noting the things their child *can* do, or to note ways in which their child can still be happy and fulfilled.

The way in which parents came to understand the diagnosis of CP also informed the goals that they developed for their child's functioning and QOL. For example, whether they push for their child to walk rather than relying on mobility aids. In a way, it was the initial understandings of what the diagnosis means that framed all the future expectations of limitations for the child themselves and for the whole family. It also related later to how parents managed parenting in situations where more independence was required from the child—for example, when they started school, or even earlier, in playgrounds or play groups. One parent discussed struggling with allowing their child to have more independence as they aged because of fears around the child injuring themselves, or others outside the family being unable to provide the care required for the child's disability.

7.4.1.3 The time of diagnosis can be a time of trauma

Lastly, but by no means least, for many parents, the time of diagnosis for their child was a time of trauma for them. This was especially so for those who experienced a difficult birth event related to the child's CP, or for those who faced the prospect of potentially losing their child at birth or early infancy. Even those who noted only mild

functional irregularities later in the child's development, if they struggled to obtain a definitive diagnosis or specialist medical attention, they found the process in some way traumatic. For many parents, the process of obtaining a diagnosis for their child was an experience that lingered long after they accepted their child's CP. Some parents retained anger over perceived flaws or mistreatment in the medical system from the time of their child's diagnosis. One mother recalled the prognosis given for her child at the time of diagnosis by saying, "they pretty much told me she was going to be a vegetable". She went on to describe the ways in which this prognosis was incorrect and echoed a sense of disillusionment with the medical system.

Often there was a strong element of bewilderment—especially because most of the children with mild CP initially showed normal development. Some of the way the story was told to the interviewer emphasised that changes to development occurred through no fault of the parents. One mother began by saying her son was "born to a completely, you know, normal pregnancy. I wasn't unwell.... Disability was picked up months after he was born. But essentially it started in utero... It wasn't because of any trauma or anything like that".

There was also an element of watchfulness by the parents as their child developed—the same mother went on to explain "as he started growing, we noticed his head wasn't growing. So, we did all these scans on his head...". Parents frequently recalled that they first noticed atypical development in their infant, followed by a period of distressing confusion after it was brought to the attention of healthcare professionals, until receiving a final diagnosis.

7.4.2 The saving grace(s)

Most of the parents mentioned at some point during the interview (or afterwards, at the front door) something that is their 'saving grace:' the positive in the midst of the challenges of parenting a child with a disability. It frequently signalled how parents helped to manage their own emotions and frustrations. This helped ensure they were able to continue to care for their child even though that care may be sometimes, frequently, or chronically, burdensome. The saving graces tended to fall into three

categories: 'it could be worse', 'it can't be helped', or a deliberate focus on joyful moments with their child.

7.4.2.1 It could be worse

A frequent refrain expressing "it could be worse" that came from several parents was referring specifically to families of children with more significant levels of impairment, or more complex needs. One parent of a child who used a wheelchair reflected on exactly how this must seem to others, as he recounted:

Sometimes I see some kid who's actually, you know, severely impacted by quadriplegia or whatever, and think how terrible must that be. That must be such a burden or whatever. And I know people will say exactly the same about [my child].

Another parent of a younger child who recently began primary school reflected on psychosocial aspects, saying, "certainly, there's other kids having a lot more problems adjusting to starting school".

One mother of a nonverbal child functioning at GMFCS level V, who had additional challenges related to social isolation, poor access to services, and English as a second language, reported that in times of stress she relied on religious faith, saying her child's disability was part of God's plan, that God knew that she could cope and "if I didn't believe in God I don't know if I'd be here today". She stated that despite the many things her son missed out on due to his impairments, "he's happy and I think he's a child of God anyway". However, she also reflected on the traumatic birth and the thought that her son almost died at that time. Even though she faced many challenges and frustrations since because of his disability, she was extremely moved to think that she may not have had him at all, because he almost passed away at birth. She summed up by saying, "Well, it could've been worse. If he had gone I'd be—oh, my God. I'd be so depressed. I would be depressed right now".

Parents seemed to need to maintain these reflections to both continue to care for their child and to remain in some way positive about the everyday frustrations. It could always be worse—so there was a sense of gratitude for what they had.

7.4.2.2 It can't be helped

Developing a firm understanding of the causes of their child's CP was similarly protective for some parents. Understanding and accepting the permanence of their child's CP removed some of the uncertainty of the diagnosis and provided the sense for parents that it was not worth wasting energy on because "it can't be helped". One parent spoke of the need to pay for private healthcare for her daughter, saying "I really can't afford it. But, I do it because she needs it".

The contrast to this view was provided by the parent of a child who developed normally to the point of speech, and then suffered an unexplained seizure without an apparent cause at several months of age. Her daughter is now considered to have CP functioning at GMFCS level V, with limited communication, producing sounds but not words. To this mother, the memory of her typically developing toddler was still clear in the forefront of her mind. She did not feel she had received an adequate explanation for why her daughter suffered a seizure which resulted in the acquisition of her global, severe disability. Although she still clearly expressed strong love for her daughter in her current condition, the comparison in her mind was not firstly with what could be worse, but how she could have been 'better'. In the background of caring for her daughter, the driver for her was that 'normal' functioning was possible for this child (whether or not this was true from a medical perspective). Therefore, this parent had sought interventions that promised significant functional improvements but were outside the conventional medical system. Her ongoing uncertainty over her daughter's original unexplained seizure contributed to a sense of insecurity over future expectations of her daughter's CP course which impacts on how she cares for her daughter, and the distress she felt over her daughter's functional skills.

For other parents, continuing to undergo genetic testing to determine the cause of their child's CP offered hope of providing an accurate prognosis, and also supported notions that "it wasn't my fault", reported earlier in parents' initial discussions of the diagnostic process.

7.4.2.3 Joyful moments

Moving away from questions of diagnosis, many parents echoed similar sentiments that their children were generally happy, reflected in the comment “if she’s not well, she’s not well, but 99 percent of the time she’s always got a smile on her face”, reflecting a deliberate focus on joyful moments with their child. Noticing moments of joy in their child, particularly for those with more severe impairment and limited communication, was especially meaningful. For those whose children experience chronic pain, even moments of peace became treasured. One mother of a boy at GMFCS V with very limited communication relayed her son’s experience of joy at a disability specific horse-riding program, saying “he loved doing that and you can tell he loved it”.

These moments not only provided peace of mind and positive memories to parents but could also provide a guide for seeking out future recreational opportunities. For parents of children with more severe impairments, recreation might include tactile experiences or spending time with specific people such as a treasured grandparent. The mother quoted above whose son loved horse riding reported “he loves vestibular movement, which I think is why he liked horse riding”. She explained that because of this they sought out an opportunity for her son to ride in a ‘super’ car on a family trip, saying “he’s gone around the racetrack... and when he pulled up he was laughing. Now, to get him to have that real joyous laughter and stuff ... it was the best ever”. From understanding the tactile sensation that her son enjoyed, she was able to plan other opportunities to see him laughing and happy. For children with milder CP, parents sought hobbies and recreational opportunities for their child to enjoy, especially those where the organisers made additional effort to see their child included.

7.5 The business of disability

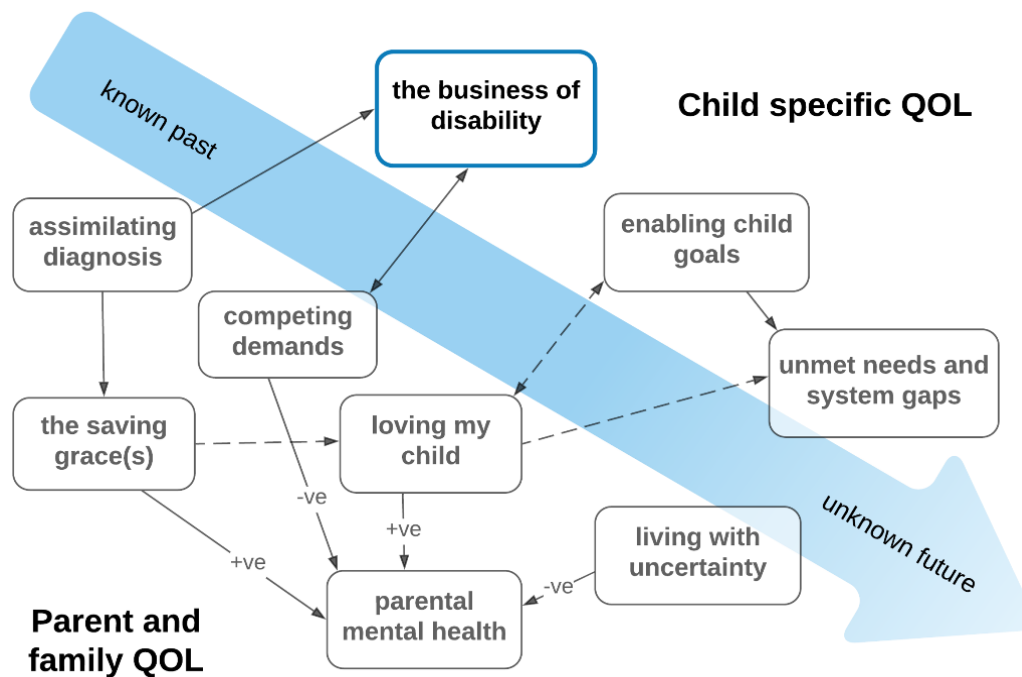


Figure 9. Parent findings conceptual map: The business of disability

As shown in Figure 9 above, parents' descriptions of child-specific QOL related mainly to two aspects of children's lives: 1) day-to-day functioning, including disability specific care needs, 'the business of disability'; and 2) providing for fulfilment of higher order QOL goals, such as psychological and emotional health, social health and participation goals, 'enabling child goals'. For both of these, children relied on their parents for both direct intervention and to 'curate the environment' that provided them with the best chance of achieving good QOL. For example, a child with limited communication may need parental assistance in having their needs understood when they are hungry or in pain. This child will also need their parent to find a school that will provide the best options for providing communication assistance devices and teaching the child to use them or promoting relationships with others who will interact with the child at their level of communication.

While both of these aspects contributed to overall child QOL, the way that they operated in family life was hierarchical. Each had attendant actions required of parents, but the ‘business’ of disability in day-to-day care must be addressed first, followed by other family care, and lastly, those for children’s higher order QOL goals.

Therefore, this and the following two sections present the conceptual categories 'the business of disability', followed by other family needs in Section 7.6 'Competing demands', and lastly, higher order child QOL goals in Section 7.7 'Curating the environment'. 'Curating the environment' is represented in the conceptual map by the parent perspective of 'Loving my child', and the child specific category of 'Enabling child goals'.

Parents universally reported that it takes work to care for a child with disability, and it can be very demanding. Existing literature recognises the additional work attendant on parenting a child with disability compared to parenting a typically developing child and the impacts this can have on parents (e.g. N. A. Murphy et al., 2006; Raina et al., 2005). However, this often focuses only on day-to-day personal care needs of children.

Parents interviewed here reported additional work that is less frequently mentioned, including balancing care for the child with CP and other aspects of family functioning as a type of project management that required organisational time and information. All parents discussed the necessity to manage relationships between themselves, their child, and their child's aides, carers, and health professionals. Therefore, this section discusses the business of disability in three parts: day-to-day demands, project management, and interceding in therapeutic relationships.

7.5.1 Day-to-day demands

A significant and obvious part of managing the business of disability was day-to-day demands of care for a child with a physical disability. These care needs were usually a reflection of the severity of the child's CP, as children with mild CP usually maintained independence in daily tasks while those with more severe physical impairments required assistance with toileting, dressing, transferring from a bed to a wheelchair, feeding and facilitating activities and entertainment. For some children, parents were required to maintain a schedule of medications or manage a child's other health problems such as epileptic seizures, pain, or gastrointestinal issues such as reflux. Many of these tasks could be physically demanding of parents, especially as children grew. Even for those families who received outside assistance with care, or respite services, these were daily tasks that had to take priority over any other family needs in

order to maintain their child's basic health and functioning. One mother framed this by saying, "when you do have that extra carer burden then ... you are limited in some sense. And you do have to put a lot of effort in. ... I don't know how to... that's work. ... But yeah, I don't think there's a way around it".

Once health and medical needs were managed, the next major job of the business of disability for parents involved delivering interventions and therapy at home. For children with CP these primarily involved ensuring children performed strength and flexibility exercises, wore orthotics or other devices, or used mobility equipment appropriately to reduce fatigue. For children with complex communication needs, practicing new communication methods was required. For older children with more independence these needs involved encouragement more than direct supervision, while for younger children or those with severe impairments, exercises sometimes required direct physical manipulation by parents.

Parents reported feeling significant pressure to maintain a schedule of interventions, and guilt when they could not manage the daily demands of delivering them. One parent reflected on the pivotal role they played in their child's functional outcomes, saying, "Us parents are the therapists". The mother of a boy who was nonverbal with probable ID and severe communication impairments discussed struggling to teach him to operate a switch to communicate yes/no, saying, "it's like he does want to communicate but we just don't have the physical ability and means to do it". While she communicated a positive perspective of her son's abilities, she also implied a level of guilt that she and his carers could not adequately support him to express himself despite practice at school and at home.

The day-to-day demands of care can be higher for children with CP than for typically developing children even where these do not directly involve health or medical needs. For example, a child who uses a wheelchair may not be able to use regular public transport or carpool with other families to go to school. Some parents made direct comparisons between the care required for their child with CP and their other children, such as this mother explaining:

It is a lot harder... I mean, we have lots of kids and it's easy for me to compare the time and effort investment. Not that I don't love all of them but what I have to allocate to make his day come together, even on a day where there's no appointments, ... I mean, the meetings I have to go to at school, you know, the— the— ... that's a lot of hours.

Several parents of children with mild-moderate impairments spoke of the ongoing vigilance involved in supervising their children in social or play environments. This was especially the case at early primary school age, as children learned to have more independence in play. For example, several parents of younger children who were mobile enough to play independently with supervision mentioned being more watchful than other parents in a playgroup situation, or more watchful than they would be for a typically developing sibling. One parent of a five-year-old reported, "when he's with his peers and I'm there—I'm keeping a closer eye on the interactions than any other parents". The way parents individually responded to the additional watchfulness varied—one parent acknowledged being more watchful but still limiting the amount of intervention they provided, while others spoke of difficulty with trusting other carers to provide the necessary intervention. Parents conveyed a sense both that they felt additional tension over allowing their child more independence because of their disability, but also that the child required a greater level of support in most or all situations because of their disability.

Day-to-day demands often changed as children aged. Parents whose children were unable to walk without assistance discussed how they had been able to travel or go out more frequently when their child was younger due to their child's size. As they grew up, care became physically challenging due to their increasing size, recurrent illnesses, or other care needs—and travel was no longer feasible. The mother of a boy who is a wheelchair user who is now entering his teens spoke about how much more restricted his and the family's activities had become because his increasing size meant wheelchair access is now a higher priority, and an increased need for physical strength and skill in transfers made it more difficult to rely on help outside the family. She explained:

when he was six, you know, we could fold him double and pack him in a suitcase. He's tiny. ... He's, you know, up to here now right, he's a big man which means that there is—you need the skills to be able to just physically get him onto the bathroom. Or out of the car or into a car even. If you don't know how to do it, it's very intimidating.

7.5.2 Project management

Beyond day-to-day demands, parents reported a need to engage in ongoing project management for their child's care. In the short-term this included activities such as acting as a case manager for medical and therapy needs, making time to deliver physiotherapy and other interventions in the home, meeting with school teachers to discuss access issues and concerns, organising transport for a child who uses a wheelchair rather than relying on carpooling with other parents, and the large time burden of seeking information about CP, interventions and therapies, and accessible recreational services. Over the longer term, parents needed to ensure that necessary equipment would be provided as their child developed, that ongoing required funding was available, that carers, aides, or respite services were organised ahead of time, that appointments were up-to-date, treatment schedules were adhered to and that information necessary for appointments was up to date. For family recreation, project management involved additional work for any family day trips or vacations to ensure accessibility, or that medical needs could be managed during travel, as well as planning for attending specific disability activities. All of this was necessary to ensure that their child's direct day-to-day care could be managed, even though much of the project management work engaged the parent away from their child.

Information seeking was a large part of project management. Parents engaged in two main types of information seeking: understanding CP and learning about CP interventions; and finding accessible facilities and service options. Although there is a large amount of information available about both CP and disability services for children, parents reported that there was no single place to source information, or that information was not specific to family needs or local services. Recent research in Australia on available information just about the changing disability services system

(i.e. the introduction of the NDIS, mentioned in Section 2.1.6.5, page 26) showed that the quality of available online information was highly variable (Simpson & Baldwin, 2017).

All the parents interviewed reported spending time on research to increase their understanding of CP and its attendant medical needs and options. Parents engaged in this research from the time of diagnosis, to ensure that their children could have the best possible outcomes, and also to be able to communicate with their child's medical team and engage in project management aspects of CP. One parent discussed the need for information that to understand about her daughter's medical needs, potential surgeries and possible outcomes because, "I don't ever want to put her under the knife, so to speak, if I don't have to or make anything ever harder for her".

Furthermore, parents spent time evaluating their child's ongoing functioning, to learn what was normal, and when to be concerned over illness or changes in function. If their child developed an illness or unusual pain, parents needed to not only distinguish between regular CP-related symptoms and the illness but be able to describe the differences to a health professional.

Parents reported spending a significant amount of time searching for information on available services, funding, additive or alternative therapies, recreational opportunities, accessible places for family outings and accommodation for vacations. One mother reflected on her initial bewilderment and how she felt her family continued to miss out because of lack of information, saying:

At the start, it was really confusing. I had no idea what to do, who to ring. Apparently, there's a lot of services out there. I still don't use those services because I still don't know what's out there.

Most parents reflected experiences of similar barriers to finding necessary services and opportunities: information required was diverse, services were fragmented, and there was no central source to direct them to where they could find out what they needed to know, whether this was for health information, seeking new equipment, or finding recreational opportunities. Frequently other parents were an invaluable resource in the information seeking process—those who had been there before were able to

provide targeted information or point out accessible opportunities that were not otherwise apparent. Some parents valued disability specific parent support groups, online or in person, partly for this reason. For others, these groups were not appealing, however they often still acknowledged the advice that they received incidentally from other parents. For example, one mother said,

I really haven't joined any groups and stuff like that. Like, when she goes to [disability service] and stuff like that I talk to the other mums, obviously. And, you know I ask them about certain things ... Or, you know advice on simple things that would help me.

For many, the specific and tailored advice provided by other parents of children with CP not only opened up new ways of managing CP, or new services, but saved significant amounts of time in research.

7.5.3 Managing therapeutic relationships

The final aspect of the business of disability involved managing and interceding in therapeutic relationships between their child and other carers, aides, or clinicians. Until children are old enough to manage these relationships and decisions about treatments or interventions for themselves, their parents are necessarily their first advocate. One mother, a single parent, discussed making decisions about timing for her young daughter's surgery and put this role starkly, saying, "It would literally just come down to me ... to do everything I can to help her fight and whatever it needs". Parents managed relationships with health professionals and doctors and, in some instances, negotiated between them, occasionally providing information from one service to another. For children with complex communication needs, parents were often their first interpreters or had the greatest skill and practice in understanding what a child wanted or was trying to communicate. Therefore, in therapeutic situations parents usually provided direct, practical intervention as interpreters.

Most children received some form of personal assistance at school, through classroom aides, or afterschool care at home from professional carers. Parents reported taking direct action in these relationships only infrequently—in the current education system

parents have very limited choice over who their child's school aide is—but usually monitored the assistance that was provided to their child, how often, and how beneficial it was. More than one parent expressed concern over the dependency that developed between their child and the aide at their school. For example, discussing their child's transition to high school, one mother commented on the new aide, saying, "He's got lots of experience but maybe almost too much experience to kind of step back to the degree that he needs to 'cause normally he's [the child] quite capable".

In some instances, monitoring and intercedence in therapeutic relationships meant changing services or advocating for changes in helpers. One mother discussed struggling to find a local recreation service that would take care of her child's unique needs, saying:

We tried a local gym place, and it was very disheartening for me watching it because we didn't have time to talk to the teachers. It's ... the same as swimming lessons—as one group finishes, the next one goes. There's no time to actually have discussions which is important for people like us.

In this case, the service was not able to develop a relationship at all that either the parent or child felt was satisfactory. Both parents and children talked about the value of positive relationships with health professionals, carers, and service providers to children achieving functional goals. The mother of a 12-year-old boy who has had the same aide at school since grade 1 said, "The aide is a really crucial thing. Having a good aide that works well with your family. I think that's very important, and he's had that". For many families, long-term aides became family friends—the same mother went on to say, "and we love her. I mean, she's a dear friend of the family now".

Managing therapeutic relationships also involved navigating their child's understanding of CP and experience of healthcare. This draws on parents' own assimilation of the diagnosis, and depended on the child's level of impairment, developmental stage, and intellectual development. For example, parents of younger children with minor impairments did not use terms such as 'disability' or 'cerebral palsy' that their child could not easily understand. One parent mentioned that their

five-year-old, who had mild CP with physical impairment mostly limited to one arm, didn't really understand CP, saying:

He's unaware of his disability as far as knowing what it is. And he knows his right hand is weaker but not necessarily applying it—probably doesn't realise that other children don't have that same thing.

The same parent went on to explain how their son found a way of describing his CP, saying, "he's called his right hand silly. There was a point where a paediatrician called it his silly hand, which we thought was a bit poorly worded". This quote suggested a way in which parents may rely on health professionals to help conceptualise their child's disability. However, it also illustrated the tension that can build if the language used by health professionals does not match a parent's own understanding and beliefs about what CP means for their child. Another parent of a five-year-old with mild ID and mild CP affecting one side of the body explained that her child did not use the term CP but understood the idea that he had "one weak side".

In both families, parents talked about "other children like you" or "other children with one weak side" to refer to others with CP or disability. For both children, the way parents chose to discuss CP and disability with their child was tailored to the child's subjective experience. This varied understanding and use of terms to refer to CP and disability can impact on how these are discussed in interviews with children, as mentioned briefly in Chapter 5 when discussing parent collaboration (see page 116).

For parents of children at GMFCS level IV or V who have global severe disability with ID, the child's conceptual understanding of disability terms may be irrelevant. Instead, there may be a more practical focus on trying to include children in conversations about themselves. For example, two of the interviews were with parents of non-verbal children at GMFCS level V, and their child was present during the interview. In both cases, parents made reference to the child during the conversation to include them in remarks even though they could not answer verbally. Both parents responded to vocalisations their child made during the interview, saying they were agreeing with the interview content, or responding to specific words or phrases. As the child was the

subject of the interview, each parent made every effort to include them in the conversation in any way possible.

Other parents discussed making similar efforts at including their children in conversations and decisions about therapeutic interventions and ensuring that the relationships they had with specific health professionals were positive and supportive. One parent was quite explicit in how she encouraged her child to participate in conversations about the consequences of different treatments, even though at seven years old her daughter was limited in the decisions she could make. She explained:

There's no point in me saying that, you know, you have to do it, when she doesn't want to do it. When she doesn't like the people she's working with or she doesn't feel comfortable. ... There's no point in me putting her in a situation where she's going to hate it. Like, if there's something that I know she's going to like and that I think she's just too scared to approach, then that's when I'll be like, 'Well, no. You're doing it.' But, I'm never emotional, you know, with it. Like, a physio, her AFO's or something like that that I know that will hit real close to home for her, I feel like those are the decisions I leave to her. Like, I talk to her about it. And, I might persuade her to think otherwise or something like that. But, yeah, the ultimate decision I leave up to her.

Therefore, some aspects of the way parents managed therapeutic relationships helped to promote other child QOL goals. They allowed children to have ownership of their own body, to learn independence and the ability to advocate for themselves in health contexts and gave children a sense of dignity.

7.6 Competing demands

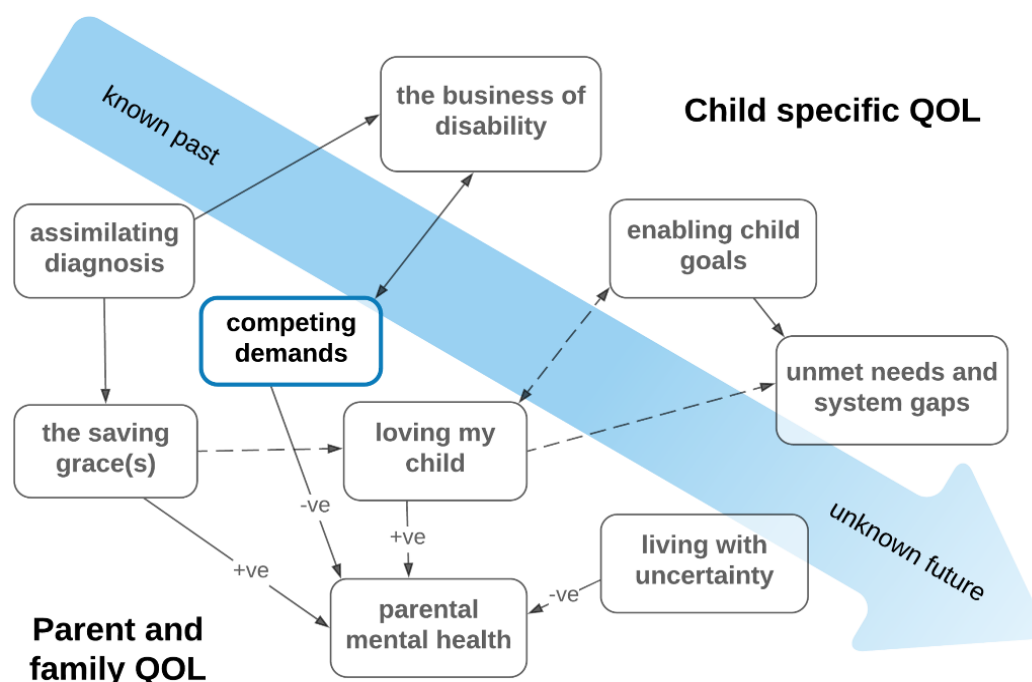


Figure 10. Parent findings conceptual map: Competing demands

Most parents discussed the difficulty of managing care for a child with disability along with making time for parenting generally and other competing demands. This formed part of the context of family life, shown in Figure 10 above. Competing demands created practical difficulties in managing day-to-day family life, which often involved making trade-offs rather than managing everything. This required a balance between not only necessary tasks, but the cost of each additional opportunity for the child with a disability or other family members. The increased demands presented personal challenges to a parent, by embodying a heightened sense of changes resulting from becoming a parent of a child with a disability, and often a sense of loss of opportunities or plans. Lastly, parents developed a variety of strategies to cope.

7.6.1 Practical difficulties

For most parents, one of the major practical difficulties was lack of time to manage both required care for their child and other aspects of their lives. As childcare was the first priority, time for other activities was lost. Some parents were unable to return to work after the birth of their child because of care needs. Others mentioned that the time spent researching CP or disability-related issues meant there was no time for

recreational pursuits, such as reading for pleasure. Some parents found they had very limited time for their relationship with their partner, as well as other family members.

Every parent with more than one child discussed the difficulty, not only of making time to spend with their other children, but of ensuring their children felt a measure of equality. The mother of young twin daughters, one with CP and one without, noted that she spent a significant amount of one-on-one time with her daughter with CP when they attended appointments and bought lunch or other treats afterwards. She then noted that the twin without CP, “misses out on stuff like that. She never actually gets one-on-one time with me, regardless if it is just sitting in a waiting room or something like that”. This mother therefore made an effort to spend time alone with her daughter who does not have CP, which not only increased the time burden on her, but caused friction between the siblings. She concluded, “so I guess, yeah. It is really hard to divide my time”. Several other parents spoke of needing to balance the time spent with siblings to trade off for time spent on therapy and appointments with their child with CP, and the strain this could place on family relationships.

Within sibling relationships, parents also faced tension between the time siblings spent supporting the child with CP, and time for each child to be independent. A different mother of twin girls who had recently started school mentioned that her daughter with CP had been facing social discrimination from her peers. She discussed not only the difficulty of managing that situation, but of the additional strain it placed on her other daughter to ensure her sister was included without disadvantaging either child. She said:

We try not to stop [twin without CP] from going off, but at the same time she has a responsibility to her sister to make sure she’s not left out ... It’s a fine line, a very fine line. You do the best that you can.

Every parent interviewed tried to ensure that their other children were not disadvantaged by the additional care required for their child with CP. However, they frequently reflected the same sentiment, of “doing the best you can”, with an implication of knowing that some things would be neglected.

Even without considering other family members, there were often competing demands just for the child with CP—for example, making a trade-off between therapy or other recreational activities. Trying to enforce interventions, such as strength or flexibility exercises, required a balance between not only parental time, but types of activities in which parents would prefer to engage with their child. One parent mentioned a sense of guilt over not requiring her daughter to wear her leg splints while on holiday. The holiday offered the chance for her daughter to take a break from the discomfort of the splints, and for her to take a break from the role of therapist.

All the parents interviewed received government assistance for aspects of the health care and equipment for their children, as well as access to public medical services. However, most found that this assistance did not cover all necessary care and equipment. They therefore faced significant financial burdens for additional services or upgrades to mobility aids as their children grew. One mother reported that as she had been unable to return to work, they relied on her husband's overtime to pay for necessary services. This had other consequences for the family, and she said, "my husband works twenty-four seven ... The kids hardly ever see him but it's the problem, you know, you try and do your best". Again, parents expressed the need to balance costs between competing family needs.

7.6.2 Personal consequences

Several parents spoke about the changes that occurred after their child's disability became apparent involving losses for either or both parents, such as losing employment or income-earning ability, losing financial status due to costs of therapy and equipment, losing friends and social circles because they physically could not maintain them and even losing confidence in themselves in dealing with the medical system. For some parents, these changes altered the division of labour and roles between themselves and their partner—especially when only one parent was able to work.

Several mothers mentioned how difficult it was to maintain social connections that they had prior to the birth of their child, due to environmental accessibility, uncertainty in future planning, and having too many other responsibilities and limited

time for personal socialising. Accessible spaces were necessary not only for social occasions where children were included, but increased care needs made it harder for parents to organise alternate care to attend events that were not accessible.

The additional difficulties involved in attending social events resulted in some parents actively avoiding additional commitments. One mother said, “sometimes I don’t have the motivation to go out, because it’s going to be hard work and sometimes it’s easier just to stay home”. She explained that she and her husband sometimes turned down social invitations partly due to the extra vigilance required to supervise her daughter. For example, when friends suggested a weekend away, both parents responded:

Like, “No”. Don’t want to go. Because it means we don’t really have a good time because we’re always concentrating on her and you can’t have conversations... and if someone’s with you, you’ve always got your eye on her. And then me and [her husband] have to tag team. Watch her, watch her.... It does sometimes mar the social occasions.

As was mentioned earlier, the nature of the competing demands usually meant that not every need could be met, even with reliance on external support and carers. This meant that most parents experienced a level of guilt at not doing more for their children. Guilt resulted from each side of the competing demands; for example, guilt for neglecting siblings or other relationships, guilt if one parent couldn’t work so the other missed out on time with the child or had to do overtime, but conversely guilt for not doing more for the child with the disability, or guilt over the need for respite. Parents of children with severe disabilities particularly spoke of guilt for not being able to provide more recreation or stimulating activities. One mother explained some of the barriers, saying:

I do feel guilty because the bigger he gets the sicker he gets... seizures and chest infections. I can’t just get up and take him like I would my other kid like, oh, let’s just go and play in the park or something.

Parents spoke of personal consequences not only to themselves but to their other children. Some emphasised that having a sibling with a disability could be a positive in

teaching their typically developing child empathy and the praise they received for supporting and helping their siblings. However, the demands between care for the child with CP and needs of other siblings could cause tension, especially between twins. Both parents of young twin girls had faced a difficult conversation in which the typically developing daughter blamed her sister for having CP and creating additional care demands. Though these were isolated incidents between the siblings, they also added to parental guilt.

Lastly, parents could experience a loss of identity, especially those who had to give up work that had been meaningful to them. Those who had previously been outgoing, active, and social, could find these changes particularly frustrating to their sense of self. Along with a sense of grief for the lost expectations of their child's life, some parents attributed later struggles with mental health to these changes. These aspects of the personal consequences of competing demands are connected particularly to how parents 'assimilate the diagnosis' (Section 7.4.1, page 194), and their ability to accept the permanence of their new family life, and find and access ways of coping.

7.6.3 Ways of coping

There were practical ways in which parents were able to cope with the competing demands. Some parents relied on the assistance of extended family, respite services, carers and aides, or in one case, home cleaners. Necessarily, the ability to rely on help varied depending on what was available to each family. Parents of multiple children mentioned relying on other families especially for their typically developing children to attend social and recreational activities. In some cases, parents were able to access services that could combine different aspects of care for their child with CP. One mother spoke of taking her son to a disability-specific horse-riding organisation. This service provided him exercise and necessary core strength building, as well as enjoyment in the activity itself and his connection with the horses. Another parent spoke positively of a school that was able to incorporate physiotherapy exercises into their class routine, reducing the need for her to ensure that her child completed the exercises at other times.

Parents also coped by maintaining flexibility in other aspects of family life, or by compromising on either ideal situations for their child or themselves. If disability-specific recreational services were too far to commute, or financially a strain, parents compromised with locally available mainstream recreation options for their child with CP so that they did not miss out entirely. Managing parental work made a significant difference. Some parents mentioned that their workplace was able to allow them to attend appointments, as one parent said, “again, we manage it because we are reasonably flexible”. For those whose work was not flexible, the decision not to return to work could be personally challenging but allowed them to attend to childcare. One mother said explicitly, “I haven’t gone back to work more than two days a week because of that”. By reducing demands of work, she was able to cope with the demands of her child’s medical needs.

7.7 Curating the environment

Parents may have limited ability to influence the environment in which their children live, in terms of availability of healthcare and medical interventions, recreational services, or schooling. However, parents exerted significant effort in finding and choosing opportunities for their child in each of these areas. In effect, parents curated their child’s environment to achieve health and wellbeing goals. This was the second level of the hierarchy of needs, once basic needs were taken care of. These were the personal goals of parents and children for fulfilling their dreams and being happy.

Parents curated the child’s environment to allow them to achieve their dreams, to participate and be included—in whatever way this was meaningful for the child and their family. From the parents’ perspective these actions were an expression of love, both in noting what made their child happy, and fighting for rights to be included in childhood activities or building their child’s social circles. Therefore, the conceptual category of ‘loving my child’ is described first here, as it leads to the following category of ‘enabling child goals’. Figure 11, below, highlights the connection between them, as well as how they fit with the overall findings map.

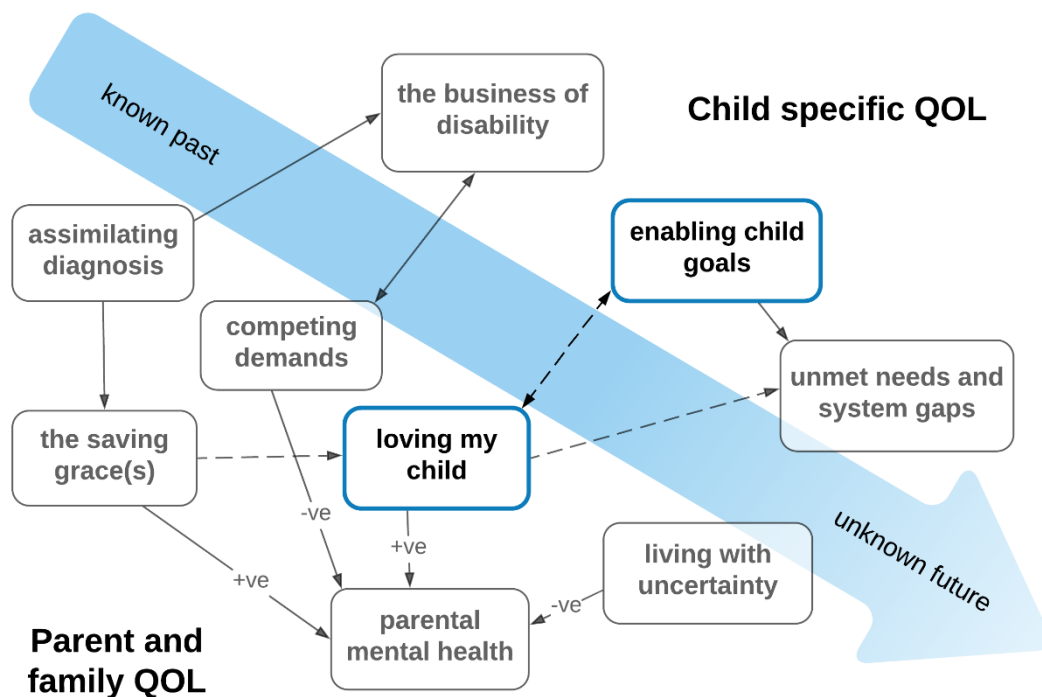


Figure 11. Parent findings conceptual map: Curating the environment

7.7.1 Loving my child

The ultimate aim of every parent interviewed was for their child to live their ideal life. They loved their children, and wanted them to be happy, healthy, well, fulfilled, and to have the best possible career or carer or independence outcomes in the future. Parents spoke explicitly about loving their child and creating a loving home as a way of building resilience for the challenges they would face as a result of their disability, such as discrimination and bullying. For them, loving their child was not only a natural part of parenting, but a deliberate action they emphasised in building self-esteem in the context of disability. For example, one parent said:

The world can give you all sorts of grief but if you've got a comfortable and lovely home to come home to with lots of love, then that becomes less significant.

Parents themselves took comfort in the idea of their children being loved and supported. One mother discussed how her daughter's significant physical limitations were frustrating for both of them, but concluded, "she still is quite happy and she's got a lot of people around her that love her and support her". The loving and supportive

environment links to the concepts from the child findings in developing positive self-identity, and positive self-esteem through supportive relationships (see page 173 of Section 6.4 'Defining the self').

Several parents spoke of negative attitudes toward disability that they or their children experienced. This was most frequently expressed in terms of unsolicited attention or questions from strangers. For example, one mother recounted, "I've had people before, I'm in a supermarket or something, turn around and go "What's wrong with her?", things like that. I go, "Absolutely nothing, she's perfect"". This parent took pride in being able to express absolute acceptance of her daughter. This aspect of the parent journey ties closely to the corresponding development of positive self-identity in children, discussed previously in Section 6.4 (see page 171). This is also tied closely to the journey parents take in assimilating the initial diagnosis of CP, and finding saving grace(s), as discussed in Sections 7.4.1 and 7.4.2. To express love and acceptance for their child may be a form of comfort for parents in the face of a sometimes-overwhelming diagnosis.

Accepting their child's CP as a natural part of the child could create a conflict for parents as they felt a need to explain their child's impairments and continue pushing for changes and improvements in their child's functioning which implicitly suggest that the child's current status is 'not enough'. For some parents, the conflict was triggered when asked specific questions about their child's disability. For example, early in most parent interviews, parents responded to the prompt "tell me about your child" by first emphasising their love for their child before describing the child's CP and the story of how they received a diagnosis. This was more evident from parents of children with milder impairments and without intellectual impairment—children who also have their own understanding of CP and may be closer to typical development. It may also relate to parents' own ongoing acceptance and understanding of CP, as well as how they focus on building their child's resilience around a disability identity. The same conflict emerged for parents dealing with the attitudes of strangers asking questions about CP, such as the mother above who emphasised to strangers that her daughter is "perfect".

As has already been mentioned, there is a significant amount of work in caring for a child with a disability, above and beyond what is required for parenting a typically developing child. The workload was not reduced by the love parents have for their child, but because of this love, parents continued to strive for the best treatments, access, and services. As expressed by the mother of an active 5-year-old at GMFCS III: “the challenge will be getting her through these couple of years before she hits teenage years. But she’s awesome so she’s worth it”. This describes the context of the second conflict of this concept for parents: doing everything possible for their child balanced with the needs for themselves and other family members. Ultimately, it is the parental love that drives the way parents ‘curated’ their child’s environment—fighting for acceptance in participation opportunities, going out of the way to provide socialising, supervising therapy and managing therapeutic relationships and becoming an expert on medical terminology. This adds additional pressure to the ‘competing demands’ discussed in the previous section.

7.7.2 Enabling child goals

Overall, the goals parents had for their child with CP were more future-facing than the day-to-day business of disability. They did not differ necessarily from goals parents have for typically developing siblings—but they required more work and intervention because of the child’s disability.

For parents, ‘enabling child goals’ involved not only improving children’s lives immediately, but also future-proofing their child—especially in the light of ongoing uncertainty, and parents’ sense of “I won’t always be here”. The implicit belief was that if parents could build supportive worlds and activities around their child now, and increase their internal resources, this would provide them with a brighter outlook for the future. At times parental goals for children’s overall QOL and psychological wellbeing conflicted with immediate therapeutic goals—between adaptations for a child’s current level of functioning against therapeutic goals for improved future function. For example, one parent recalled navigating this situation with family video games, saying:

We all play the Nintendo Wii and we have a cable tie over one side for his right hand 'cause he can't get his thumb down. ... He gets frustrated but he still tries, which is from our perspective very positive. But we were at the hospital last week or the week before for a regular visit, and they mentioned that the cable ties probably isn't a good idea. We should be trying to get him as close to his peers as possible and that sort of thing, rather than accepting that it's difficult. But then, that's a balance of he wants to play the game to a certain level, he doesn't want to be unable.

This parent faced a struggle not only between family inclusion and therapeutic goals, but between encouraging their child's persistence and managing their frustration. Several other parents similarly tied psychological or emotional outcomes to physical therapeutic goals. One parent mentioned the value for her daughter of the physical achievement of learning to ski:

We didn't actually think she'd be ever... she may not be able to do it, but we wanted to encourage her, we wanted her to try, we wanted her to have that sense of freedom, of moving unencumbered.

Both these parents, and others, showed how functional goals parents had for their children may include seeking experiences of freedom from disability-related challenges. Such experiences may be a higher priority for families than specific functional outcomes.

Child goals centred on inclusion, on education, social life, participation in recreational pursuits, and the opportunity to take family holidays and trips. Most parents aimed for some future level of independence for their child, with or without aides or support. Parents sought child goals that would provide the greatest sense of fulfilment for the child, through sense of achievement as well as functional outcomes. Specifics of these goals varied from family to family, depending on what was most valued by each child and parent. Achieving child goals relied on external supports for participation and inclusion, as well as parents' ability to manage 'the business of disability' and the 'competing demands'.

Beyond relying on external support, parents consciously worked to build their child's psycho-emotional health as part of enabling child goals. These goals explicitly focus on psychological wellbeing, in terms of self-esteem and positive self-identity related to disability, and resilience. Parents of children with milder disabilities emphasised positive character traits of their children which they encouraged as a way to help them overcome challenges of having a disability. For example, emphasising stronger social skills, or a "quick tongue", that could benefit children in social situations. Parents expressed pride in these traits and that they improved their children's future psychological resilience.

Parents were conscious of the potential negatives of additional attention that their children received due to their visible disabilities. As one parent explained, "as much as she loves being the centre of attention, she hates to be the centre of attention for, I guess, her cerebral palsy reasons." One mother spoke of the benefits of her child maintaining friendships over several years because "they're friends who don't see the disability". Parents of children with severe CP spoke similarly of the value of siblings, extended family members, and pets, who did not hesitate to interact verbally or physically (such as hugging and touching) with their child. Parents reported positive responses from their child to trusted, long-term relationships, regardless of a child's level of CP severity, or communication impairments.

Parents reflected on verbally reassuring their children in relation to their CP. This could be about their capabilities, for example, one mother said, "I always put it in her head that anything her sister could do, she's capable of doing. Like, there's no reason she can't do what her sister can do". Reassurance also related to self-esteem and sense of self, as one parent explained, "that's what I say to her: 'You're perfect in so many ways. Because of one little thing, it's only a tiny part of who you are'". Parents discussed this as an explicit strategy to help children develop an understanding of their disability to offset potential future discrimination. One parent explained:

If we can empower him to own his disability and understand it and know that if someone thinks less of him because of that, that's their problem... our challenge is to get into his psyche about that sort of thing.

In this way, parental attempts at building children's psychological health aimed not only to create immediate reassurance, but to build future resilience. For a few parents, this also included building their child's understanding of CP-related therapy, both to increase their compliance and their future independence. The mother of a 7-year-old explained:

She may be too young at the moment to actually fully comprehend what's happening or why it is the way it is. But, at the end of the day there's going to be a day when she does fully understand it. And she's going to be fully aware. And she's got to be able to make those decisions by herself.

For parents, child goals for happiness and good QOL included navigating both a current and future sense of how children understand disability and how to build positive images of themselves and their capabilities.

7.8 Parental mental health

In the context of discussing child and family QOL, several parents independently raised issues of their personal mental health, and the impact that caring for a child with a disability had on them. Previous research has shown that mothers of children with a disability have poorer mental health compared to Australian averages (Bourke-Taylor et al., 2012; Emerson & Llewellyn, 2008; Gilson et al., 2018), and that this may relate to child outcomes. It has also been shown that parents with poor mental health are more likely to report lower QOL for their child, whether or not the child experiences poorer QOL (E. Davis et al., 2012; Wiley & Renk, 2007). Given that parent personal QOL is not the focus of this thesis, and a substantial literature already exists on parental mental health, discussion of parental mental health here is confined to its links to other categories. These included 'living with uncertainty', 'competing demands', and 'assimilating diagnosis' and 'the saving grace(s)'. These links are illustrated in the conceptual map below.

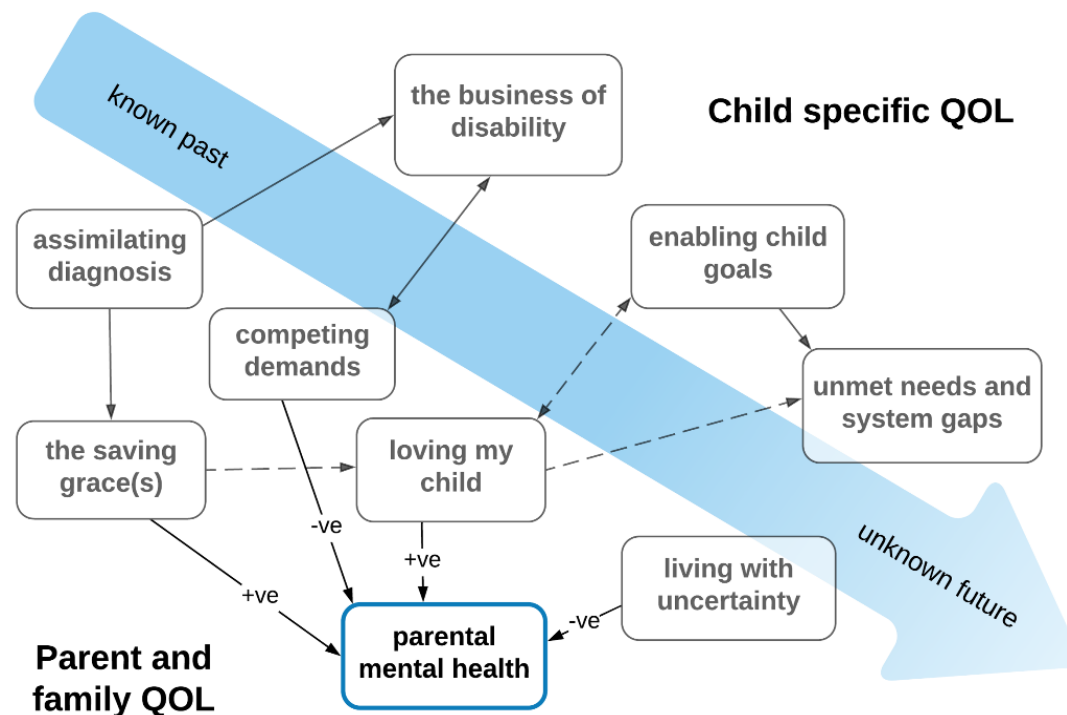


Figure 12. Parent findings conceptual map: Parental mental health

For parents, their child’s disability added an ongoing level of uncertainty to their lives (this is discussed in detail below in Section 7.10, ‘Living with uncertainty’, page 227). Living with uncertainty meant parents might neglect other areas of life because they had to change plans at a moment’s notice, or because it was not possible to make plans. One mother spoke about being glad to have travelled when she was younger because it no longer seemed possible with her child’s needs and ongoing illnesses—reflecting that what she had already achieved helped to balance what was no longer possible. Where frequently changing situations presented challenges that parents were unable to overcome, they discussed this as a direct detriment to their personal wellbeing. Missing out on social, recreational, and employment opportunities due to the inability to commit to future plans may be part of a negative impact on mental health for parents of children with a disability.

Parental mental health was related to the process of assimilating the diagnosis, with ‘the saving grace(s)’ apparently protective of good parental mental health. Several parents commented on the difficulty of care for a child with CP but immediately followed with comments on how seeing their child happy, smiling, or laughing, made the additional work worthwhile. Some parents expressed that their child’s diagnosis

had increased their own appreciation for other aspects of life, such as kindness and empathy of others. One mother felt it had changed her worldview significantly, saying:

I see the world differently too. Like, he's opened my eyes and I won't take things for granted ... It's a gift that we don't realise we have that we walk, talk, even see or hear.

7.9 Disabling vs enabling environments: the value of knowledgeable, timely support

Many parents spoke positively of key people they had met through their child's diagnosis and disability journey, who were able in some way to ease their negotiations with systems, funding, management of their child's condition, or other aspects of life. Some parents spoke of the lack of these supports, and how much of a difference they thought it could make. The common features of these discussions were those of specific targeted knowledge, empathy, understanding of the parent's individual circumstances, and practical support with tangible outcomes. These related to negotiations in the business of disability, and overall easing of burdens from competing demands, assimilating diagnosis, living with uncertainty, or addressing system gaps. In turn, this created a positive impact on parental mental health, overall child QOL and ability to achieve personal child goals, and general family functioning. The impact of the environment emerged as aspects of other categories, so is not included in the main figures of conceptual categories. Instead, links between the environment and key categories are shown in Figure 13.

One parent captured the environmental links espoused in the social model of disability when she said:

It's not the disability itself that makes you disabled in the community, it's the facilities out there. If there's no ramp, if there's no changing places, if there's no proper disabled parks, you know, what do you do?

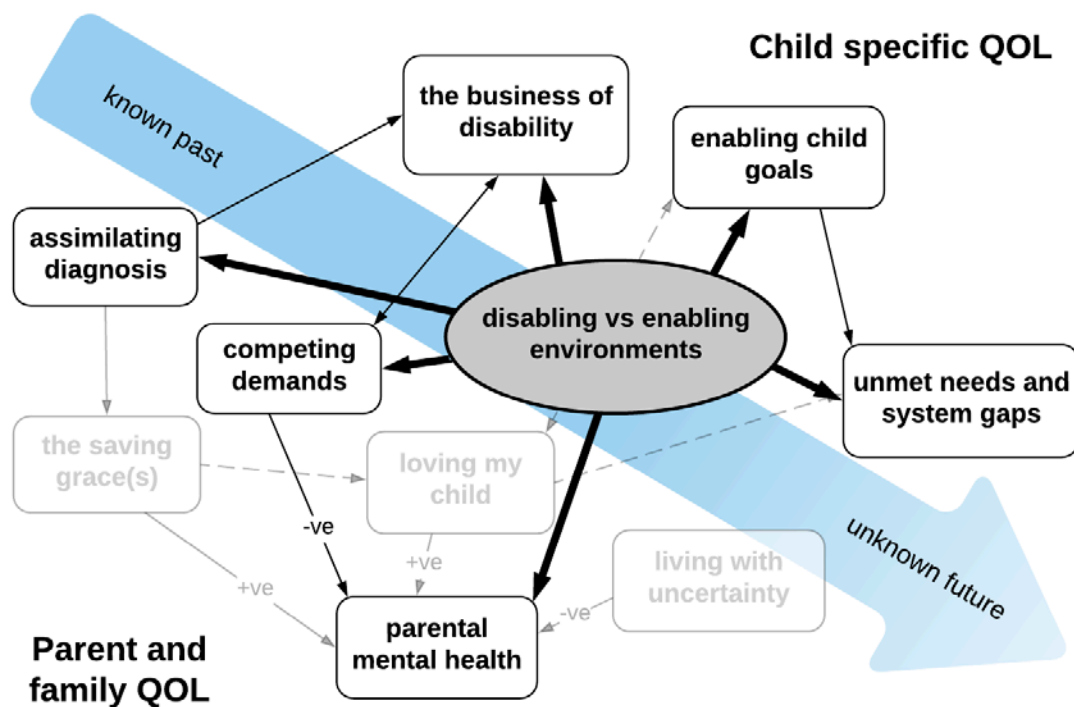


Figure 13. Links between parent conceptual categories and environmental support

Many of the environmental issues raised related in some way to access and inclusion: access to activities, funding, services, knowledge, respite, physical access to geographic locations, or inclusion for the child and family. For most of these, parents relied on support from others outside their family, including services.

One mother commented on having free buggies for wheelchair users at a few beaches, saying that “tiny little things like that make an enormous difference”, and later reflected on a different issue, “if it’s a small nuisance for 250,000 families then that’s something even if it is just a small problem. But if you fix it, wow, the benefit is just enormous”. One mother spoke positively of a disability service that helped her negotiate with her daughter’s school for additional accommodations, saying, “They were fantastic. They were going to go out to the school and have a talk”. However, after she ultimately decided to change her daughter’s school, the service did not follow up with her about negotiating with a new school. She felt “a bit disappointed about” it, saying:

I think that they should have followed up with me. ... I think, them as an organisation should understand that parents like me, you know ... you’ve got so

much going on in your life that it's the last thing that you should be remembering to do.

Additional personal touches or introductions had a similarly positive benefit. For example, in school transitions, some school staff reached out to parents to ask about additional needs for their children or offered additional walk-throughs of school grounds or services. For parents, this not only reduced anxiety over their child's school transition and the ability of schools to provide adequate additional assistance to their child but allowed both children and parents additional time for any adjustments that were needed to the environment, or equipment. One parent spoke of protracted negotiations made with her son's school for the classrooms to be made physically accessible for him. Initial building works were completed over the school holidays to the wrong specifications. An occupational therapist making a school visit for her son noticed the error. However, the school was not immediately willing to make more changes, and she thought it was lucky that due to her husband's job, her "husband had a lot of disability experience, so we had to argue with the school principal that the builder was wrong". The situation was made more difficult by the school principal changing multiple times during this period. Where able-bodied children and their parents may take for granted an ability to navigate the physical school environment, this was often an initial barrier for children with physical disabilities.

Providing families with the opportunity to navigate the child's new school environment and learn about areas that may be easier or more challenging for children to traverse, supported the child and also restored a measure of dignity that parents may otherwise feel was lost. For example, a parent stated that her son hated having to be carried to access the beach, saying "it's undignified being hauled across the sand in all kind of fashions, he's too old for that now". In the case of schools, the initial negotiations usually suggested to parents the level of understanding with which a school would approach any future disability specific challenges that arose. This encapsulates what can be the difference between a disabling and enabling environment not only for the individual child, but for individual parents, and for whole families. Making changes to a physical environment relied on supportive and understanding people to identify and implement any necessary changes.

Parents who experienced at least one key supportive health care professional early in their journey were more able later to engage with services, to seek more support, and to expand their knowledge of what was possible and what was available. Several parents identified one specific worker who guided them through their first access to services. For one mother, this was a physiotherapist, as she described:

When we first started physio and stuff like that her physio was absolutely amazing. She definitely offered a lot more support than just giving physio to [her child]. She definitely helped when it came to guiding me [for] like, payments, and other places to go and stuff like that.

Conversely, parents who identified a lack of this support within their disability health services system experience were more likely to be disengaged, isolated, and pessimistic over their child's long-term options and outcomes, and perceived a far greater level of injustice generally for children with disabilities compared to typically developing children. A few parents said explicitly that it would be a huge improvement for parents if there was a single place to find information—a mother said, “Wow - what a treasure trove that would be”. Others had found a central information source through specific services or individual health professionals. Considering the role that information seeking plays in project management for parents (see Section 7.5.2, page 204), this presents either a major help or a difficult hindrance.

This type of support linked to one of the key aspects of child QOL identified in the previous chapter: children spoke of the benefits of an identified ‘helper’ in multiple settings (see Section 6.3.3, page 169).). For children, this was often a necessary condition for negotiating physical limitations—for parents this eased one challenge of managing the business of disability. In both cases, the support provided was specific, targeted, and results in tangible outcomes. Children were able to complete tasks or participate in activities; parents were able to access necessary disability services or realise other non-disability focused aspects of their daily lives that contributed to healthy family functioning.

It was not only supportive services that could make a difference for parents. Parents able to rely on extended family, other children's families, or other members of their

local community found balancing family and childcare needs significantly easier. One mother mentioned that her parents-in-law lived in the other side of their duplex residence, and this was an invaluable support for her and her husband. She said, “it enables me and [my husband] to be able to work. Especially just with the school drop-offs and things. I only work two and a half days a week”. Without this support, she may not have been able to return to work, even at part-time hours. Conversely, other parents whose extended family was located far away usually reflected on their lack of external support.

7.10 Living with uncertainty

One aspect of the parent journey that framed the background of most decisions that parents made around caring for their child and managing other aspects of their lives was the sense of living with uncertainty. This was primarily a parental experience, related to future events, shown by its placement in the conceptual map below.

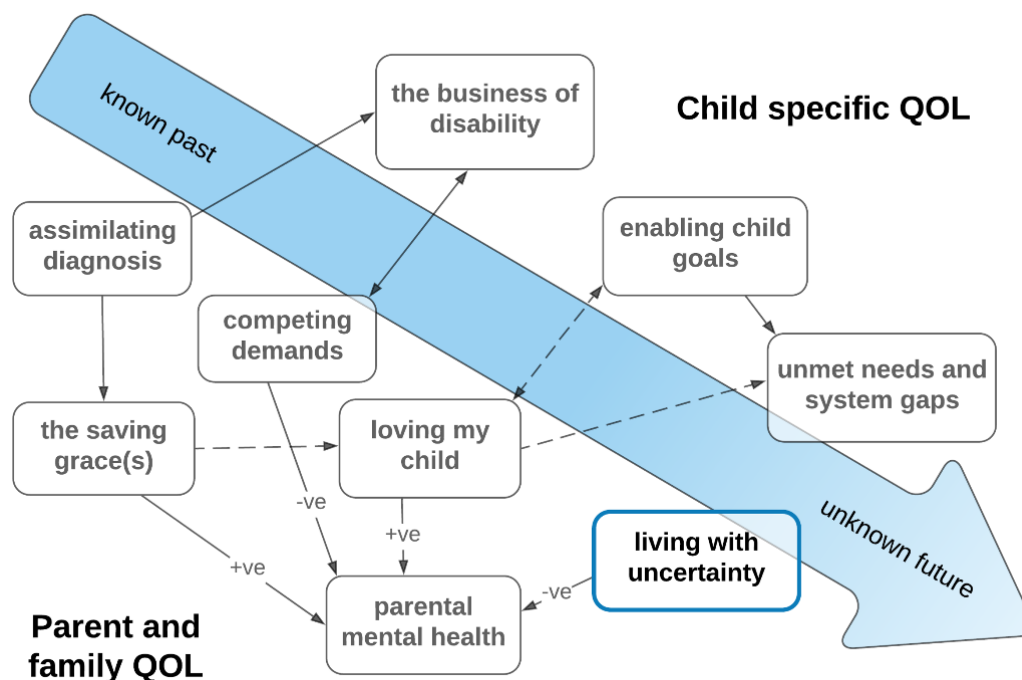


Figure 14. Parent findings conceptual map: Living with uncertainty

Parents frequently reported engaging in limited planning for the future both because it was difficult to manage the day-to-day, and because of ongoing uncertainty about the future. This was captured by the father who explained, “It’s ‘okay, what’s in the immediate future?’ and you, sort of, have a vague underlying long-term plan”. There

were two main areas of disability parenting that contributed to uncertainty, captured in this mother's statement: "we don't know what the world will look like. We don't know what his needs will be". Uncertainty came first from the course of CP, and secondly from funding and service provision. The outcomes of uncertainty were in time spent preparing for the uncontrollable, and its impact on family life. Each of these three aspects of the conceptual category of 'living with uncertainty' are described below.

7.10.1 The course of CP

CP itself is a permanent, nondegenerative condition, but its functional consequences change as children develop and age, and as a result of surgical and other therapeutic interventions. Most parents reported avoiding thinking about the future both because the day-to-day was already overwhelming, and because their child's future capabilities were unknown. Therefore, parents reported a sense of living with uncertainty, not just in terms of services being outside their control, but over their child's experience of CP itself. For parents whose children have more severe impairments, uncertainty included how or what their child is communicating, and the level of QOL they actually experience. For those with fewer impairments, uncertainty was based on anticipating changing responses to treatment, children's need for inclusion in activities or available services for recreation, as well as uncertainty over future goals for education and independence.

For many parents, uncertainty over their child's condition began with prognosis at the time of their child's diagnosis. One parent recalled "early on everyone kept saying just wait and see, just wait and see, just wait and see", but she pointed out that her 11-year-old son is now at GMFCS level V and "dependent on every single need". To her, such uncertainty in an initial prognosis now seems absurd and frustrating. For those parents who did not have a definitive cause for their child's CP, there was ongoing uncertainty over how to assimilate the diagnosis, as well as uncertainty related to family planning—some explicitly wondered if further children would carry the same disability.

As children approached primary school age, they often required surgery or surgeries with uncertain outcomes, and uncertain recovery times. The diversity of presentations of CP may make it difficult for health professionals to give accurate advice over what likely interventions a child will require—this increased the uncertainty parents felt over their child’s condition. Some children faced recurrent illnesses and infections that required medical treatment, recovery time away from usual activities or recreation, and which interfered with other ongoing treatments. One parent mentioned an ongoing fear that her child would pass away. She felt this began from the time of diagnosis when “the doctors put it in our head that also he could die... they said that pneumonia could kill him. And that is very scary”. Her son now attends a primary school for children with disabilities which in some ways maintained this fear and uncertainty as she recounted, “I get notices from the school, like so and so died, at least every year at least two kids you get notices in the school bag”. For parents for whom the time of diagnosis was a time of trauma or fear for their child’s life, recurrent illnesses continued to raise the same fears and uncertainties.

As children approached their teens, parents spoke more of worries over their future independence. Several parents also spoke of worry over their children’s psychological needs as teenagers, when they potentially faced greater stress in social inclusion. One mother of a 5-year-old said frankly, “I’m dreading the teenage years”, when discussing navigating her child’s sense of self and visible disability in the transition to primary school. Overall, parents anticipated greater changes for their children’s mental health as they aged. However, the combination of growing children, changing CP presentation, and transitions in service provision, made planning for these futures very challenging for parents.

7.10.2 Uncertainty of funding and service provision

During the time of the interviews for this thesis, disability service provision in the region where the families lived (greater Melbourne) was undergoing significant change due to staggered implementation of the National Disability Insurance Scheme (NDIS). Therefore, a few families were already receiving their funding and services through NDIS, some had been given initial information and consultation on what the changes

would involve, and some expected NDIS in their region at some future time but had not yet begun the process of switching from the previous system. For many families caring for a person with a disability across Australia, the introduction of the NDIS presented significant changes with relatively unknown outcomes to service access (Howard et al., 2015). While most of the parents interviewed here expressed uncertainty concerning what the changes would mean for them, some were optimistic that moving to the NDIS would result in better outcomes for them. For example, one mother whose child's school had not made all of accessibility changes they required with current pre-NDIS funding, was "hoping the NDIS will sort out, like, toilet seating and things like that for him". These anticipated changes provided context to discussions of service access.

Regardless of the NDIS rollout, there already existed a persistent level of uncertainty around funding and service arrangements for the parents interviewed. Many parents felt that funding and available services could change at any time, or at best, that funding was inadequate and provided only for limited services beyond hospital or paediatrician-based check-ups. Parents needed to keep track of how much money had been spent on which appointments and weigh up what may be needed in the future. Parents reported that many aspects of available funding relied on the decisions and timelines of external agencies that may have policies unknowable to families, and did not consider individual or competing needs of parents or children.

Many parents discussed having positive experiences of service access as a matter of chance, beginning with the time of their child's diagnosis. One mother described the series of events by which she came to access a disability specific service and met a specialist who provided a CP diagnosis as: "everything sort of fell into place. I've heard other horror stories, but we've just been nah, nah, nah. So, we've had support all through it. So, we've been lucky". Another parent reflected on how her family had received a significant amount of support and equipment provision through vision-specific services, saying, "if he just was disabled, and not had a vision impairment, and not had the seizures, we would have probably slipped through the cracks". Overall, parents reflected a sense that it was pure chance whether or not they met the right health professionals, or were connected to the right service, or were in a region with a

specialist who understood their child's condition who could guide them through the disability service system. The feeling that service access was a matter of luck lingered from the time of their child's diagnosis and appeared to contribute to parental perceptions of the uncertainty of ongoing provision of support services.

Following the initial diagnosis period—usually in infancy—there were major service transitions that created uncertainty for parents. Major service transitions for children with disability occur alongside school entrance – from early intervention services to primary school, and from primary school to high school. Preparation that parents could do for these transitions was often limited, or there was inadequate support for them to learn what the transitions would entail. In some cases, there are specific services to manage the transition, such as additional school support, or hospital-based information sessions. Transitions increased parental uncertainty but may be more manageable as an anticipated and acute event, rather than the chronic lower-level uncertainty of the day-to-day experience of parenting a child with disability.

Other ongoing aspects of service provision uncertainty were more frustrating. Many children with CP require some form of mobility aid or support, ranging from splints to walkers to wheelchairs, as well as other types of specialised equipment such as communication aids. These need to be replaced periodically as children grow out of them. Universally, parents reported a period of waiting after requesting new equipment, usually at least several months for major mobility aids, while their children used equipment that no longer fitted them. Parents faced uncertainty not only over when they could obtain new equipment, but over future costs for new equipment, or whether services could continue to provide the aids their children required.

Lastly, parents frequently reported a lack of control in service provision that added to the uncertainty. Regular appointments may sometimes be planned months in advance, but schedules were at the discretion of hospitals and specialists. Families had limited choice over what was provided at school, in terms of levels of funding schools could access, classroom aides and helpers, or how much a classroom teacher could adapt activities for inclusion. Ultimately, parents relied on school management to make any necessary adaptations for accessibility.

7.10.3 Preparing for the uncontrollable

Due to ongoing uncertainty, parents spent time ‘preparing for the uncontrollable’, which meant waiting and hoping for the best, and doing what could be done. The ongoing need to make decisions affecting their child’s future (and their own) without knowing what the future holds often led to parents doubting their decisions. Several parents stated that they explicitly avoided planning for the long-term because it was too challenging, logistically and emotionally, on top of the day-to-day care they needed to provide. However, uncertainty also affected day-to-day care. Preparing for the uncontrollable was often a significant source of frustration and difficulty for parents in project managing both their child’s disability and other needs of their child and family.

Some everyday activities of disability-specific care such as medications or basic physical needs followed a loose but predictable schedule. However, there were many things that parents and children had little control over that they needed to address, mentally taxing because the parent needed to prepare themselves and their child for a situation that might or might not arise, and in which they did not control the outcome. The mother of an 11-year-old at GMFCS level V discussed the difficulty of attending to his complex medical needs, especially as he had very limited ability to communicate if and why he was in pain. She mentioned that she felt “a despair feeling” at being unable to “fix” all his problems, but explained “you then go okay, what can I control? What can I, on my checklist, work on?”.

Beyond unpredictable symptomatology, parents spent time preparing for medical or therapeutic appointments, which occurred without a set schedule, at the convenience of the hospital or health service. If the appointment was a follow-up of a test or was in any way complicated, such as a visit with a specialist, the parent particularly might wish to prepare for the conversations in order to understand what the consequences were for their child. The child often wished to know why they were attending an appointment, or whether they would miss school or recreational activities. More than one parent mentioned an unexpectedly long recovery period after surgeries—during this time it was difficult to predict how much their child could engage in recreational

and schooling activities. Other parents mentioned missing social events due to illness and unexpected appointments or reducing their social plans due to the potential burden of having to change them. One parent explained:

There's many times we've had—we've been going to go away on holidays and we've had to postpone or cancel or they give you an appointment... the hospital, will give you an appointment and say "You have to attend", and if you ring up and say "I can't come this day,"—"Well the next one's not for six or 12 months".

A secondary outcome of living with uncertainty was the difficulty this presented to parents in building and maintaining an active personal and family life outside of managing their child's disability. The aspect of uncertainty added an extra layer to the challenge of managing competing demands on family time and resources.

7.11 Seeing unmet needs and addressing system gaps

All of the parents (and several of the children) expressed a certain degree of frustration because of unmet needs—not as a result of the child's CP or other conditions—but due to gaps in the current systems for services or equipment. Every parent could quickly give a list of things missing from their ideal environment for their children or had suggestions for ways to improve access and services. Even when they acknowledged limitations of public funding, or the needs of other children, or compared their situation to others who were worse off, parenting a child with a disability involved experiencing unmet needs. Families addressed these gaps after taking care of other immediate needs, and in expectation of future changes, as illustrated by the right-hand placement of this category in the conceptual map below.

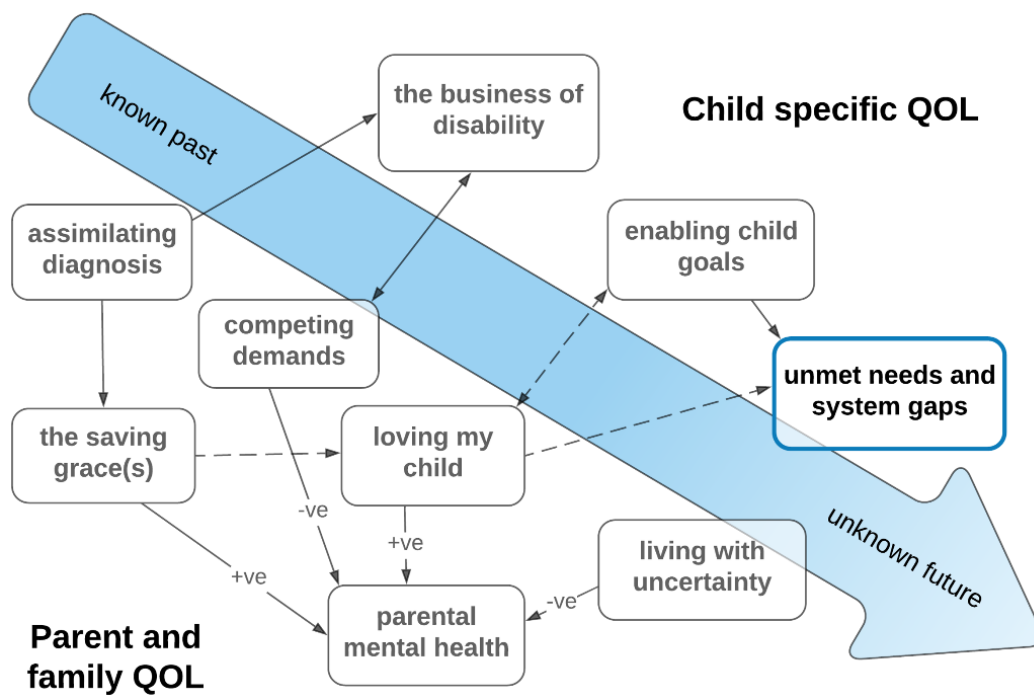


Figure 15. Parent findings conceptual map: Unmet needs and system gaps

From the time of the first diagnosis, parents acted as an advocate for their child. This was especially the case for parents of children with limited communication abilities. For example, one parent of a non-verbal child with severe CP explained, “My son can’t speak out. ... I just be his voice. We speak out for him”. Parents expended significant energy in additional advocacy activities to enable their child to achieve their recreational or social inclusion goals. Advocacy in this context refers to fighting for recognition of the rights of children with disabilities to equitable inclusion and access to physical spaces and health and recreation services that meet their needs. A certain level of advocacy was often forced on parents to enable their children to attend school and to participate in recreation. Advocacy in itself may help to build child goals of independence and self-esteem, especially where the child can be involved in the advocacy themselves. That is, parents may choose to include their child in advocacy activities in order to promote positive images of disability, or to increase a child’s resilience for future discrimination or exclusion experiences.

Some parents described the ongoing sense of injustice as a major frustration with imperfect disability access and services. For example, most parents discussed the high cost of disability specific equipment, with one mother particularly discussing a sense of injustice that not only equipment but toys and services for children with disabilities

were more expensive than those for typically developing children. She said, “Everything’s breakable. It’s just crap quality but they charge you thousands just because it’s small demand”. Another parent expressed frustration and indignation over the scarcity of wheelchair accessible places saying “it is the law. It’s equality”. Similarly, the difficulty many parents faced in navigating the complexities of disability services encompassed a sense of injustice that it was particularly challenging for those who already faced multiple challenges in caring for their children.

The feeling of injustice could also be a call to action for advocacy within the context of the wider disability community. Parents spent time advocating for additional services and access because of the unmet needs they see for their own children as well as a sense of equity and the needs of others. Several parents noted that current gaps in the system were navigable for their family but should be addressed as other families with fewer resources needed more support. Some parents viewed participation in the research interviews as a form of advocacy in order to increase the understanding of the needs of children with CP. One parent explained explicitly that she did not expect immediate benefit for herself or her son but hoped that it would help other families.

Some parents mentioned that the experience of parenting a child with disability had increased the empathy they or their other children feel for others with diverse ‘special’ needs. The sense of empathy was often linked to and evoked by the sense of injustice. However, empathy for the needs of others in the disability community may be more pronounced in those who volunteered to participate in a research project about the needs of children with disabilities. Therefore, it may be difficult to generalise this specific link to other families of children with disabilities.

All the parents interviewed had direct suggestions for ways in which their own and other children’s lives could be enhanced by improvements in service provision. However, most parents acknowledged that as a way of coping they chose when to fight for changes, and when to accept less than ideal situations. With only a limited amount of time, energy, and money with which to address gaps, parents made decisions regarding immediate priorities, and hoped for the best. One mother talked

about a situation where her daughter had three different aides who were not communicating well with each other over her daughter's care. She said:

That was quite difficult. So, I just sort of, shut off at that time... 'cause it was temporary anyway. So, and the one I wanted was very good, so I was quite happy to wait for her. But yeah, there wasn't really much we could do at that stage.

Though the immediate situation was difficult for both her and her daughter due to miscommunication between multiple carers, she decided that the amount of effort that would be required to achieve an unlikely outcome was not worthwhile. Every parent had a similar story of accepting a less than ideal situation because fighting for an alternative was overwhelming and potentially futile. For most parents, learning to navigate the disability services system also meant learning when they could effect changes and when they could not.

7.12 Discussion

The conceptual categories in this chapter reflect significantly wider issues related to child QOL than those resulting from the analysis of the child interviews. This concurs with previous literature on child QOL from parent perspectives, including the original qualitative work for the CP QOL-Child (Waters et al., 2005), and suggests that to parents, child QOL is rarely separated from broader family QOL issues. The wider issues discussed by parents also reflect the different knowledge parents and children have—children are usually only partly aware of work parents do to provide them access to services or community activities, will not remember the diagnosis journey, and are often shielded from many conversations between parents and health professionals. While a parent might reflect positively on the relationship between their child with CP and a typically developing sibling, they consider this relationship from the perspectives of both children, considering the consequences for each.

From the parent perspectives in this chapter, outcomes of QOL for children with CP begin from the time of diagnosis, echoing other research with parents of children with CP about their experiences of healthcare during their children's infancy. For example, a

recent qualitative study investigated parental perspectives of diagnosis of CP in those whose children were in neonatal intensive care (Guttmann et al., 2018). As in this thesis, parents in that study recalled the time of diagnosis as being highly emotionally charged, even while completing interviews several years later. The findings from Guttmann et al. are reflected in aspects of 'assimilating diagnosis' and 'the saving grace(s)' described in this chapter. In a very different study, a group of researchers recently conducted a systematic review of rates of CP severity and co-morbidities and published a version of the results specifically as 'clinical prognostic messages' to improve clinicians' ability to provide easily understood information to parents at the time of diagnosis (Novak et al., 2012). The authors report other research that has shown that parents often feel that health professionals do not provide adequate information on future expectations for their child which makes it difficult for the parents to care for their children.

While the conceptual category of 'living with uncertainty' arose strongly for parents in the current research, it does not appear to have been conceptualised in this way in existing QOL literature. However, other qualitative research with parents of children with disabilities has shown similar suggestions of uncertainty related to the provision of information, the future, and continuation of services. For example, another grounded theory study with parents of children with multiple sclerosis showed parents lived with uncertainty in four domains: diagnostic uncertainty, daily uncertainty, interaction uncertainty and future uncertainty (Hinton & Kirk, 2017). The concerns of parents in this chapter closely echo this work. Similarly, a mixed methods project focused on parental participation in healthcare decision making for children with disability found that parents wished to be actively involved in decision-making but often felt inadequately informed, and a lack of information was a significant barrier for parents (McNeilly et al., 2017). Both these studies relate not only to parental uncertainty over the course of CP in this sample, but to the value parents placed on timely and tailored information provision.

The impact of the environment on experiences of disabilities is enshrined in social models of disability (Bricout et al., 2004), however specific links between QOL, parental wellbeing, and environmental support have also been demonstrated in the

literature and support the concepts of this chapter. Environmental barriers, including physical, social, and attitudinal factors, have been shown quantitatively to be negatively related to the QOL of children with CP, as reported by their parents (Badia et al., 2016). Environmental barriers related mainly to the physical environment at home, at school and in the community, as well as how well services were able to support children's needs, and positive, supportive attitudes from family and school. Environmental barriers impact the whole family and may contribute to the higher parental stress levels associated with caring for a child with disability. Furthermore, the CPRD enshrines rights of people with disabilities, including children, to equal access and support their communities for education and leisure participation – suggesting a rights-based approach to supporting QOL for people with disabilities must address systemic barriers to equal rights.

A meta-analytic review of the impact of interventions focused on reducing parental stress for parents of children with a disability found positive results for programs which included specific, targeted family support (Lindo et al., 2016)—much as was reported qualitatively by parents in this chapter. Similarly, the family-centred care model uses a rights-based approach that emphasises family priorities and perspectives, and has shown to be perceived as more respectful and supportive for Australian parents accessing paediatric rehabilitation services (Dickens et al., 2011). Meanwhile, in a review of the literature on supports available for young children with disabilities and their families, Ritzema and co-authors (Ritzema et al., 2018) showed how both parental perceptions of support and actual family support impact on child wellbeing and parental perceptions of child wellbeing. These authors further highlight the need to consider supports and resources for all family members, and the limited research to date on this topic. This relates not only to the concepts of 'disabling vs enabling environments' in this chapter, but to the discussion of consequences of insufficient family support discussed in the section 'competing demands'.

Overall, the parent categories in this chapter align with the ideas found within the family QOL literature, which focuses not only on the QOL of individuals, but on the relationships between them and the interconnections of each family member's QOL. For example, Poston et al. (2003) reported a model of family QOL based on qualitative

work with families of children with a disability including ten domains that were split between those with an individual focus and those with a family focus. Individually focused domains included productivity, advocacy, emotional, physical, and social well-being; while family focused domains were daily family life, family interactions, financial well-being, and parenting (Poston et al., 2003). These are closely aligned with the conceptual categories of the parent findings in this chapter and, to some extent, reflects the split of parent identified issues relating to child QOL and parent/family QOL shown in Figure 7 in the early part of this chapter (see page 192). However, in this chapter individual parent goals and parent wellbeing were de-emphasised; in family QOL, individual goals of all family members are given equal weight.

7.13 Summary

Parents provide the foundations that allow children to thrive and achieve good QOL. This includes personal challenges parents face in their own journey to understanding and coping with the tasks of caring for a child with a disability, as well as navigating the often-complex world of service provision for their child. There are a range of positive supports that provide real meaningful assistance to parents and families that can influence these outcomes, including family, friends, community services, and broader services and policy initiatives that improve access for those with disabilities. Feeling time-poor and resource-poor in managing aspects of their child's disability may be common for parents, but isolation from services, information, and psycho-social support results in negative impacts for the whole family, including the child with a disability. On the other hand, parents who experience support in their own journey, as well as in day-to-day management of disability care, report greater capacity to meet their child's and family needs.

These findings suggest some parallels to those of the child interviews, as well as points of divergence. For example, the concept of living with uncertainty emerged strongly in parent discussion but was not a concern shared by children. The following chapter compares and contrasts these differences between child and parent perspectives in greater detail, as well as the implications this has for a future revision of the CP QOL-Child.

Chapter 8: Comparison and integration of child and parent perspectives on QOL

Not all of the findings that emerged from child and parent interviews directly referenced children's QOL. However, they all have implications for both QOL measurement content and its interpretation. As the findings and analyses have been presented separately for children and parents, the first section of this chapter compares and contrasts child and parent perspectives and the implications of the different perspectives for conceptual models of QOL. Current measures of QOL for children with CP have largely replicated domains and items across both child self-report and parent report. In contrast, the findings of this thesis suggest children and parents have some differences in perspective which lead to both overlap and some differences in conceptual models of QOL. This chapter relates differences between child and parent findings to wider literature, especially concepts of family QOL. The last section of the chapter discusses specific implications for the content of an updated version of the CP QOL-Child.

8.1 Comparing child and parent findings

Overall, children and parents expressed corresponding QOL goals that are consistent with previous literature. Themes emphasised a balance between socioemotional domains, physical domains, and environmental interactions. However, the perspectives of children and parents differed at multiple levels, not only in additional information parents provided regarding community service access and parenting issues, but at a micro level regarding how children perceive their social networks and their developing identity. Therefore, after outlining corresponding and overlapping domains of QOL for children and parents in Section 8.1.1, Section 8.1.2 describes unique aspects of children's findings, and Section 8.1.3 presents aspects of parent findings that were not captured in child data. The section concludes by discussing the differences between child and parent perspectives, and how this compares with the existing literature.

8.1.1 Overlapping domains for children and parents

There was a substantial overlap between the main categories identified in the parent findings and the child findings. The overlap reflects that parents and children share many goals for the child's QOL, physically, socially, psychologically, and in accessibility in their local community—though often their perspective on these goals differs. By comparing the content of the categories, specific parallels between the child and parent findings were identified, in which child and parent outcomes are linked. These links are shown in Figure 16 below. To some extent, these links reflect the role parents play in shaping the everyday context of children's lives, and how they are able to provide for their children's care and recreational activities. Thus, the outcomes for parents lead to some aspects of children's outcomes in the corresponding category.

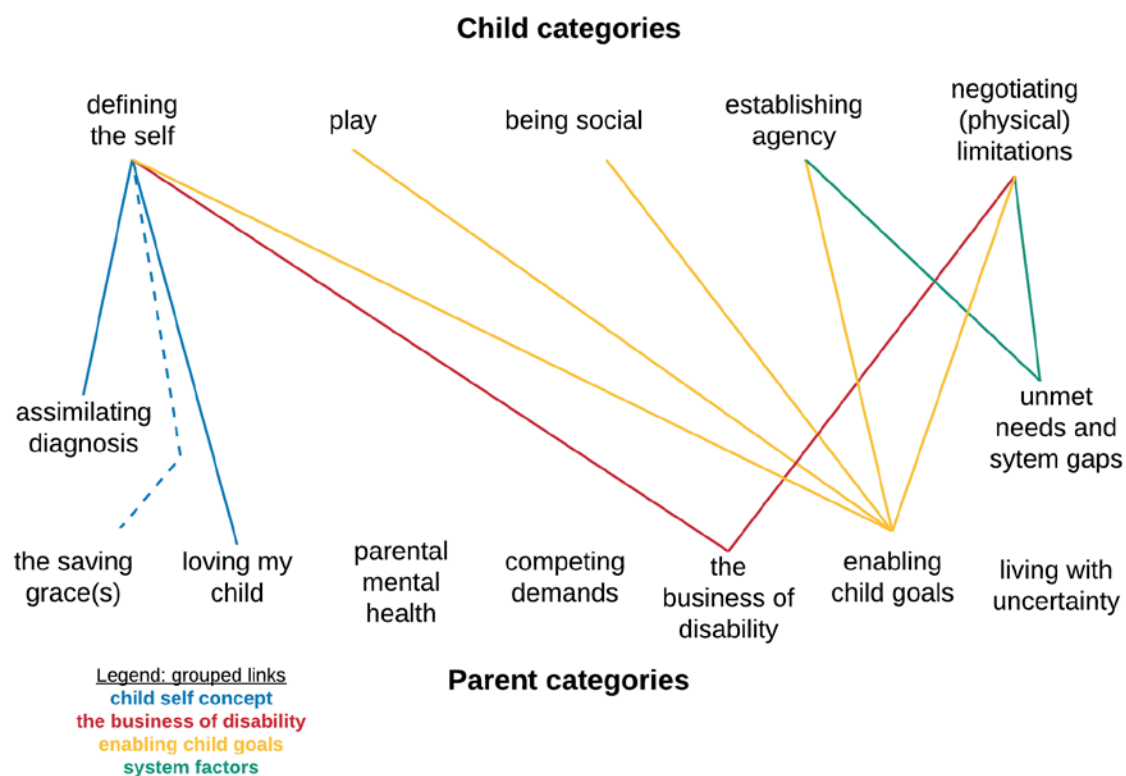


Figure 16. Connections between child and parent findings

The parallels illustrated in Figure 16 are grouped by their common major element. Three of these are based on a main parent category, while one group relates to children's developing self-concept as part of the child category of 'defining the self'. The overlap relates to how both parents and children view the CP diagnosis and the ability to develop a positive view of the concept of disability. For parents, this begins

with the assimilation of the diagnosis, as discussed in Section 7.4.1 (see page 194). Parents' initial view of what it means for their child to be diagnosed with CP, and their changing expectations of their child's future abilities and limitations, are early aspects of defining the context of children's own understanding of their disability. While children learn about their abilities and overcoming their limitations through health professionals as well, most of this, especially during early childhood, is mediated through their parents' perspectives. Even the way in which children name their condition, such as referring to a "weak side", or use of the words "condition", or "cerebral palsy", or "disability", will mostly be a result of parents' decisions on how to talk about it in the family. From an early age, children are likely to perceive positive and negative implications of their parents' own understanding of what it means for them to have CP and to incorporate these understandings into their self-concept. Similarly, the way that parents focus on positives of the CP diagnosis, through the saving grace(s) and by explicitly focusing on love for their child, feed into children's development of self-esteem. The saving grace(s) are less strongly linked, as they more clearly represent parents' own ways of coping internally, although they may be conveyed to the child as well.

The parental concept of 'loving my child' links directly to children's self-esteem. Several parents discussed explicitly that a disability diagnosis does not reflect a deficit in either their child's worth, or their love for their child. This was often offered in the context of discussing attention from others outside the family and dealing with questions from strangers about their child's disability. Even in healthcare, in the context of disability, discussions frequently focus on limitations or deficits. As discussed in the previous chapter Section 7.7.1 (see page 216), some parents cultivated a loving home environment as a deliberate defence to external perceptions of disability as a deficit. In this way, they not only build a secure attachment to develop their child's self-esteem in the family, but model how children can maintain positive self-esteem and counteract negative views of disability outside the family home. Parents built on this further through positive social relationships, and opportunities for achievement, described in Section 7.7.2 'Enabling child goals', page 218.

Attachment theory in developmental child psychology has long linked secure and positive parental relationships with positive self-concept in children (Cassidy, 1988), however there is very little research on how this relates to disability identity and wellbeing in children with disabilities. Recent research has investigated fathers' positive experiences of parenting a child with autism, including building strong emotional bonds, but did not consider how this related to child outcomes (C. A. Potter, 2016). Another recent Australian study focused on maternal emotional availability, among other relationship factors, in mother-child dyads of children with CP, but found only maternal depressive symptoms related to the child outcomes of poorer peer relationships, consistent with the literature for typically developing children (Barfoot et al., 2017b). Barfoot and colleagues found that implementing some maternal mental health strategies disrupted the parent-child bond by reducing mothers' emotional openness, which reduced their responsiveness to emotional signalling in interactions with their child. In another article, the same authors discuss how Australian services can incorporate family collaboration and parent-child relationships into treatment for children with CP under the NDIS, which preferences child outcomes rather than holistic family functioning—despite evidence for the benefits of family-centred treatment (Barfoot et al., 2017a). These studies suggest that health services could work with parents both to relieve parental barriers to positive parenting and improve this aspect of family functioning to improve child outcomes. Furthermore, disability services may be able to use family centred planning to leverage parent-child relationships for better functional outcomes for children. Parents interviewed in this thesis were often already making links between positive parenting and functional outcomes for children, both implicitly and explicitly, in the ways they parent and the services they seek for their children.

The links between parent and child outcomes and attachment style have been researched from the perspective of parental emotional resources. Notably, in a series of related publications, Al-Yagon (2007, 2010, 2015) explored parent-child attachment and parent emotional resources such as positive and negative affect and coping style in families of children with learning disabilities. These studies showed that not only children's socioemotional and behavioural outcomes were linked to parental

attachment style and coping ability, but that children's own socioemotional resilience acted as a mediator. This echoes the links found in this thesis between children's emerging self-identity and self-esteem, and parents' experiences of creating positive bonds with their children in the context of assimilating the CP diagnosis. The model proposed by Al-Yagon (2010), in which child resilience acts as a mediator, demonstrates how the child categories of 'establishing agency' and 'negotiating (physical) limitations' link to 'defining the self'. Children who have positive experiences of coping with adult expectations and accessing alternative activities outside the family home are likely to have stronger coping resources and personal resilience to either build on positive family relationships or mitigate parental distress. In turn, these lead to the greater positive child outcomes as reported by Al-Yagon and included in the 'defining the self' section earlier in this thesis (Section 6.4, page 171).

Children's goals relied directly on how parents managed aspects of parenting a child with CP through two categories of the parent findings. These were 'The business of disability' (Section 7.5, see page 200) and 'Enabling child goals' (Section 7.7.2, see page 218). For example, one aspect of children's developing self-identity in the child category 'defining the self' was derived from making comparisons with peers and siblings, not only with regard to abilities but in opportunities for inclusion and recreation. Children's experiences of participation relied directly on whether parents were able to access recreational opportunities in the community on their child's behalf—based not only on the existence of inclusive facilities, but other factors including parents' ability to find information on available services, cost, time and balancing other therapeutic goals or family activities. Similarly, while children, at times, managed their own access to alternate activities or feelings around limitations and inclusion, much of children's ability to successfully negotiate limitations relied on intervention from their parents, from early childhood through the school years. Even in families with paid carers, children rely primarily on parental care for basic needs and the provision of accessible activities and alternatives. Furthermore, before children are old enough to negotiate for themselves, or in situations in which they do not have authority to do so, parents continue to model how to negotiate barriers to inclusion or other QOL goals, as well as helping children to be included or find alternative activities.

Parental success in accessing disability-specific or disability-inclusive services in ‘the business of disability’ provided the counterpart to children’s QOL goals in ‘negotiating (physical) limitations’.

This leads to the last grouped links between parent and child categories—the parent category of ‘unmet needs and system gaps’ with child categories of ‘establishing agency’ and ‘negotiating (physical) limitations’. As with the links just described, these are both direct and indirect relationships. Parents with the resources and ability to identify gaps in current services and who successfully advocate for change provide immediate benefits to what their children can access. However, they additionally model ideals of inclusion to their children—such as one mother discussing the lack of accessible beaches who not only called the situation inequitable but pointed out that “it’s the law” for all children to have equal access. Parents who are able to show that advocacy leads to better outcomes, model higher expectations for their children and are more likely to encourage them to resist barriers. For example, the child who protested peer bullying at his school by presenting other students with information on CP did so with his mother’s full support. During her interview, his mother spoke about pushing for his rights on public transport and in other situations where she knew he was entitled to assistance that he had not received. Therefore, she not only ensured he was able to access transport but showed that advocating for entitlements or rights required by law was likely to lead to greater inclusion—despite both the child and family continuing to face barriers in access to disability services.

One of the common factors in both child and parent discussions was the impact of environments, specifically the extent to which the environment either enabled or hindered achievement of child and parental goals. This arose as a separate category for parents as part of their ongoing management of family and child dynamics, and as a sub-concept of the child category ‘negotiating (physical) limitations’. However, the impact of enabling or disabling environments was not limited to these categories for either children or parents. Figure 17, below, demonstrates which of the categories for both child and parent findings linked most closely with this aspect of the environment. Only three parent categories were largely unrelated to these supports—loving my child, the saving grace(s), and living with uncertainty. All five of the child categories

were related, though defining the self was less strongly linked than the others, demonstrated by a dotted line in the figure below.



Figure 17. Environmental connections to both child and parent domains

Both parents and children offered insights into the usefulness of different types of support that they currently do and do not receive. These insights do not seem to be well captured elsewhere in the literature nor are they reflected in existing disability services in Australia. This is especially so for children’s perspectives, which are rarely considered in developing healthcare provision policy and only infrequently in educational or recreational participation. A recent qualitative project exploring children’s perspectives of support provided by aides not only found that children had specific preferences for how support was provided, but that currently there were only limited accounts of children’s perspective in disability literature (Mortier et al., 2011). All the children in the current project discussed barriers to social and activity inclusion, and ways that people in their lives assisted them or failed to assist them to overcome them. Some barriers resulted from conflict between inclusion or participation goals and goals of other types of therapy. Conflicts occurred at multiple levels. For example, at micro levels, a therapist recommending against making functional changes to toys to allow a child to use it now and, instead, aiming for ‘normal’ functioning later conflicted with a family goal of the child’s immediate participation in games. At a higher level,

one child was unable to join in games with other children at recess as they instead spent the lunch hour with their fulltime (adult) school aide. While parents speculated on the impacts these had on children, children's views were not systematically consulted even when they expressed opinions about these situations and had clear insight into the contextual limitations. Similarly, parents perceived that their experiences of navigating the healthcare system or seeking supports for themselves and their family were rarely considered in policy that dictated what supports were available to them. Most parents reported that they frequently had to search for information on available services and supports or had only discovered available options through chance or conversations with other families of children with disabilities, rather than through services themselves. Several parents commented that their motivation for participation in research was the hope that by sharing their own experiences other families would benefit, eventually resulting in changes to services, supports or community accessibility.

Researchers in Canada have recently developed a constructivist grounded theory model investigating parent participation in family-centred treatment for children with CP, including parents' own perspectives and experiences (Phoenix et al., 2019a, 2019b). Many of the barriers that parents identified in that study included specific logistics of family functioning as well as environmental supports and the value of communication through therapeutic relationships. While the focus of the research by Phoenix and colleagues was to increase parent participation and improve child outcomes, the model identifies almost entirely the same issues that have been raised by both children and parents in this thesis in discussing environmental support and navigating the service environment. Environmental barriers for both children and parents include not only physically accessible and inclusive services, but information and outreach to enable families to connect with these services.

Lastly, parent and child findings demonstrated parallels relating to the concept of children being 'normal'. Parents presented this early in the interviews (as discussed in Section 7.2, page 189), emphasising that their children had normal interests or personalities beyond their CP. Children variously presented this as a struggle between recognising the differences created by their disability and others' reactions to it, and

the desire to be considered 'normal' by their peers and treated accordingly. For both parents and children, the desire to be considered normal appeared to relate strongly to inclusion. Children who had experienced social exclusion as a result of their disability equated being included as the default state of being 'normal': to be normal is to be included. Qualitative research with adolescents with CP found a similar relationship between inclusion and positive QOL, with participation acting as an external validation of internal self-perceptions (Shikako-Thomas et al., 2009). Proponents of the social relational model of disability have explored how the concept of normality and abnormality impacts on self-identity for children with disability (Thomas, 2004a), whereas others reflect on the ambivalence between a positive disability identity with the inherent difference this creates with 'normal' ideals of able-bodied identity (Bridgens, 2009). Both parents and children reflected some of this ambivalence in wishing to be considered 'normal' rather than being marginalised, but also seeking to achieve a positive view of functional differences inherent in CP.

8.1.2 Children's perspectives not included in parent findings

The previous section demonstrated strong links between most of the parent and child categories, both directly and indirectly. However, there were some aspects of the child findings that were either not represented, or not prioritised, in parents' conceptions of providing for child QOL. These were children's developing self-identity, the value of peer support and support from adults outside the family, the central role of play in children's lives, and the discomfort children experience in healthcare settings. This section highlights how each of these were and were not included in parent perspectives.

Parents frequently began the interview by discussing the way in which their child was first diagnosed with CP and, in the process, discussed their own understanding and feelings towards their children's disability. Some parents shared specific terminology they used with their child to refer to the child's condition. However, not all parents raised the issue of their children's understanding of the diagnosis, functional goals children may set for themselves, or the impact a disability diagnosis may have on a child's self-concept—or had limited insight into their child's perspective. Parents

mentioned how they had or planned to deal with bullying and exclusion that their child experienced related to CP, but often had limited insight into the nuances of the child's overall experience of being a person with a disability. One parent raised the issue of her daughter's past experiences of bullying at a previous school but admitted that she was not sure if her daughter had experienced bullying at the current school because her daughter did not routinely tell her everything that happened at school. For the parent who refused consent for a child interview on behalf of her child because of how distressing the child found discussions of CP, much of the subsequent parent interview focused on the mother's bewilderment in the face of her daughter's severe distress over her appearance and functional limitations due to CP. Older children approaching adolescence typically seek greater independence from their parents and family identity, and place greater emphasis on peer acceptance. As they develop a stronger individual identity, they are less likely to share their experiences with their parents. Most parents have experienced exclusion and discrimination on behalf of their child, but do not personally identify with having a disability themselves. Therefore, they may have limited insight into how a permanent disability shapes the child's identity.

Similarly, parents may underestimate the value of peer support and acceptance for their children, or the role that other adult relationships play in children's lives. Children drew support not only from peer acceptance and friendships, but close and supportive relationships with healthcare providers, teachers, and teachers' aides and carers. Parents reflected varied perspectives especially on relationships with teachers' aides or school carers, where they thought children became too dependent or aides did not give them an opportunity to complete school activities independently. However, at least one child reflected greater awareness of this situation than his mother expected him to show, and children universally remarked on the difference a supportive adult at school made for them. Similarly, while parents recognised children's need for friendships with other children, as the majority of children's peer activities happen outside of the family home, parents may not realise how central peers are to children's activities, or how great an impact they can have on children's QOL.

Within the child findings category of 'play', evidence of the concept was mainly demonstrated in research about typically developing children (e.g. Factor, 2009; Glenn

et al., 2013), though there is a growing body of qualitative literature on play and children with disabilities. In the disability literature about children's recreation, the more recent focus on 'participation' seemingly prioritises organised recreation above spontaneous play. A recent review of participation literature found that while definitions of participation focus on involvement, it still primarily includes concepts of frequency and diversity of activity—suggesting that 'active' involvement in organised leisure is the primary goal (Imms et al., 2016). Parents viewed access to recreation services as the main means of child inclusion in leisure activities. However, for younger children especially, 'play' is central to their daily lives, while for older children approaching teens, leisure is more likely to be informal socialising with peers—the mismatch between child and adult meanings of leisure and play has been shown before in qualitative research with typically developing children (Glenn et al., 2013). In either case, it tends to be subsumed by functional or goal-oriented activities by both parents and clinicians when considering child QOL, as discussed by other researchers (Missiuna & Pollock, 1991). This mismatch between the need for play and emphasis on functional goals may be related to the difficulty in facilitating spontaneous play opportunities, as adult interference is often seen to be counterproductive (Factor, 2009). However, it also raises the value of child participatory approaches not only to QOL research, but in developing intervention goals, and in allowing child agency in those goals.

The need for therapeutic goals to reflect child and parent priorities is regularly recognised in best practice literature related to family centred therapy (e.g. Palisano, 2006), but in the literature these collaboratively derived child goals still usually reflect parent goals, or are limited to functional outcomes. In this thesis, children's discomfort in healthcare settings was identified as being a result of both physical therapies and limited agency in decisions about therapeutic interventions. For children with CP, lack of agency in therapeutic settings may be compounded by reduced physical control over their own bodies due to aspects of CP—this was also suggested by the child interviewees in this project. In contrast, adult expectations of therapeutic benefits may recognise children's physical discomfort but regard it as secondary to functional gains. This represents a distinctly different perspective of both functional goals and children's

agency and autonomy in healthcare settings that has ongoing impact for treatment adherence and QOL.

In summary, children provided insight about complex issues of social inclusion specific to CP. This ranged from negative experiences of bullying and exclusion, to positive experiences with disability specific services, and adaptations during school and community activities. Children generally reflected more than parents on the support they received from their peers and its contribution to positive self-concept. While children expressed fewer practical concerns than parents in accessing treatment, they raised the need to be included in treatment goals and plans and nervousness over changing treatment schedules and expectations. Lastly, although parents reflected significant concerns over children's inclusion in organised recreation and activities within the school environment, children focussed on spontaneous play as a factor in their wellbeing and QOL.

8.1.3 Parent perspectives not captured by children

Perhaps not surprisingly, a greater portion of parent findings were not included in child perspectives. The perspectives not shared by children related mainly to either parent's own QOL (including parental mental health and the saving graces) or broader family functioning (including competing demands and living with uncertainty). Figure 18 on the following page depicts the links between child and parent categories shown in Figure 16, but highlights the parent categories that do not link to child findings.

Children did not generally reflect concerns related to parental mental health, parents' competing demands, or things parents considered the saving graces. However, these factors all impact on child QOL, both through family QOL mechanisms as well as practical considerations. Especially in the clinical context, useful information about parental issues can shed light on child QOL, and impact how interventions in the family can be implemented for the best outcomes.

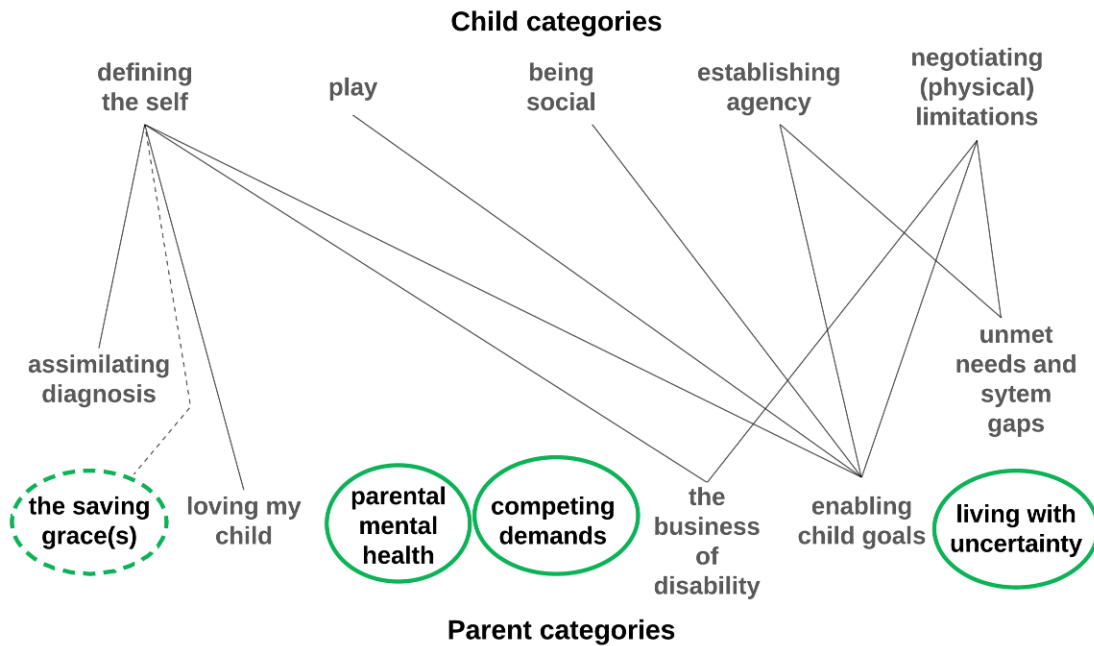


Figure 18. Parent concerns not included in child findings

Living with uncertainty was often an ongoing concern for parents. They wondered what the future might hold, particularly if their child has a severe disability. By contrast, children rarely mentioned future concerns, and seemingly accepted changing conditions day-to-day. This is probably the result of two factors. Firstly, adults are generally much more likely to consider the future than children. This difference in focus has been shown in the discordance between child and adult responses to child QOL questionnaires, with child responses generally based on a significantly smaller timeframe (E. Davis et al., 2007). Secondly, in Australia most children are diagnosed with CP during infancy (Novak et al., 2017); as such, they have no memory of not having a diagnosis of CP. By contrast, most of the parents interviewed described in detail the period of time leading to their child's CP diagnosis—usually beginning with an assumption of a healthy pregnancy or infant without CP. For parents, the narrative of their child's CP therefore begins with change, whereas children experience CP diagnosis as a stable fact.

Some of the differences discussed here may also be related to differences in severity of CP. That is, the children interviewed directly were limited to GMFCS I, II, III, with sufficient communication and intellectual ability to complete an interview with limited assistance. This contrasted with the parent interviews, which included parents of

younger children and those with more severe CP. While some uncertainty parents experienced was related to service provision, attending appointments, and other factors common to all the children, some factors related to CP, such as comorbidities and frequent illnesses. These CP-specific factors increase with increasing severity of CP diagnosis. Similarly, future concerns may be more prominent for parents of children who will require ongoing assistance into adulthood for activities of daily living, employment, or independent living. However overall, severity of their child's CP was not related to the parent interview findings. This suggests that differences between child and parent findings reflect child and parent perspective differences rather than aspects of CP severity.

8.1.4 Conclusions regarding differences in perspective between children and parents

Overall, there were three major insights arising from the comparison of differences between the child and parent findings in this qualitative study. Firstly, parent report of child QOL does not represent or equate to proxy report. Secondly, children's perspective provides insights into the social impacts on children's QOL that parents do not accurately report. Lastly, the parent perspective reflects wider issues of family functioning that influence and provide context to individual child QOL. Children are less likely to discuss family functioning, as many aspects are outside of their sight, such as how parents manage time and financial resources.

There is limited qualitative literature comparing child and parent experiences of disability, however other researchers have found similar differences in perspective. Garth and Aroni (2003) showed differences in how children with CP and their parents experienced communication in healthcare visits, as well as their priorities for communication from healthcare professionals—although both parents and children reported the importance of communication with healthcare professionals. This shows a similar perspective difference to the QOL findings reported here: both children with CP and their parents wished for supportive environments, social networks, inclusion and participation, but how they experienced these differed. Similar differences in perspective have been shown in qualitative research on wellbeing with typically

developing children (Sixsmith et al., 2007) and children with CP (Parkinson et al., 2011). Both of these studies found overall agreement in many areas of life but differences in details, such as the value of pets or alone time in children's social life.

Some of the differences in perspective between children and parents may contribute to differences in quantitative reports of QOL. For example, while parents and children with CP may report similar scores on many domains of QOL, some researchers have found larger differences particularly in psycho-emotional domains (White-Koning et al., 2007). Further detail on quantitative differences in QOL between child and parent report was discussed in Section 2.2.1.3, page 33. The findings of this thesis suggest that differences in quantitative QOL may also be related to differences in the perspectives on the underlying QOL model.

Qualitative research on how children with CP and their parents respond to QOL scales found differences in how children and parents considered events which contributed to their scores (E. Davis et al., 2007). Children were more likely to rely on single events to determine their responses, whereas parents included events over a longer timeframe. This may contribute both to how children respond to quantitative scales, and their overall perspective of QOL factors discussed in the interviews. Parents generally report lower QOL scores than their children, which may also reflect differences in expectations of what is 'normal' or desired for a child with CP if parents use a frame of reference that includes challenges to family functioning and expectations of their child's life pre-diagnosis. Other researchers have suggested this may be a source of difference in quantitative measures of QOL in combination with differences in response styles (H. Smith, 2011).

Considering both family QOL and individual QOL may be the best way to improve measurement of outcomes for children with CP in terms of QOL and wellbeing, and to target interventions. Family QOL theory reflects the findings of the parent interviews and could be incorporated into parent reports. For example, measures of family QOL can identify barriers to the pursuit of family goals, such as need for additional supports to enable better family functioning (Brown et al., 2006). Qualitative research has shown that conceptualisations of family QOL can incorporate both individual QOL of

separate family members and whole of family concerns, and interactions between them (Poston et al., 2003). Parent and child reports of child QOL do not occur in isolation—however, relying on parents’ perspective alone misses a lot of valuable information that children can contribute, at least for those who can self-report. Incorporating family QOL into parent report while maintaining conceptually different child self-report emphasises the unique perspective and information that each can offer.

For services to address both child and parent QOL, it may be necessary to further develop the current models into a hierarchy of priorities—if parents need sibling support or respite, an intervention focussed on increasing the child’s participation in leisure may not address the family’s needs but may instead place additional pressure on an already stressed family. Although many of the findings of the child analysis related to aspects of children’s lives outside of the family home, below the age of 13, children are highly dependent on their parents for their access to most interventions. Unless interventions are school-specific, therefore, priority setting may necessarily need to begin with parental priorities. School-based interventions may be more worthwhile to focus on child goals that were not addressed in the parental model, as these are less likely to be addressed in interventions involving parents and whole families.

Lastly, a study comparing family functioning of families with two different neurodevelopmental disabilities found parallels in the impact of family and contextual factors on QOL (Brown et al., 2006), suggesting commonalities in how context interacts with disability and impacts on QOL for children and their families. This suggests that a family QOL perspective may also generalise to families of children with disabilities other than CP.

8.2 Comparisons with domains of current CP QOL-Child

The findings of this thesis are ultimately aimed to lead to improved measurement of QOL in children with CP. Of the existing CP-specific measures of QOL for children evaluated in Section 2.2.3 (page 42), the CP QOL-Child presents the best measurement properties based on the QOL definition, involvement of children and parents in the

development, and focus on wellbeing. Its main drawbacks are the long length and potential changes in children's lives since its development. Therefore, this section reviews the current CP QOL-Child and its development work in comparison with the findings of this thesis, to highlight potential changes to a future revised version of the measure.

The original qualitative work for the development of the CP QOL-Child, which also used grounded theory approach to analysis, was based on parent interviews from 28 families of children with CP aged 4-12 years, including five interviews with children aged 9-12 years (Waters et al., 2005). Recruitment methods and inclusion/exclusion criteria used by Waters et al. were replicated in this thesis, however there were some differences in the samples studied. Although CP severity varied over the 28 families included in the original study, all five of the children who participated directly were mildly impaired at GMFCS level I or II—whereas half of the children interviewed in this thesis were at level III. Half of the families interviewed by Waters et al. lived in rural Victoria and half in metropolitan Melbourne; in contrast, only two families in this thesis were located outside metropolitan Melbourne. Interviews by Waters et al. were shorter, lasting approximately 30 minutes, whereas both child and parent interviews in this research averaged 50 minutes. Overall, though the sample in this thesis was predominantly located a metropolitan area, it is a larger sample of children with more diversity of CP, with much more comprehensive interviews than those included in the original qualitative work for the CP QOL-Child (Waters et al., 2005).

The main themes reported in the original CP QOL-Child study (Waters et al., 2005) included personal child factors such as physical health and comfort, and emotional and social wellbeing, as well as wider family factors, and community level aspects of life such as access to services, environmental accessibility and community acceptance. The authors highlighted that these domains may be broader than traditional conceptual measurement models of QOL, especially HRQOL, which focus only on person-level factors. The multi-level concepts, which mirror the socio-ecological models of health, were also identified in the analysis in this thesis. The original study also highlighted that many of the issues raised by families related to practicalities of care for children with CP, including environmental accessibility and support. These reflect both child

and parent findings in this thesis, especially the child concepts of ‘Negotiating (physical) limitations’, and parent concepts of ‘The business of disability’, ‘Competing demands’, and the impact of environments (Section 7.9). The main themes of the original Waters’ study to develop the CP-QOL (Waters et al., 2005) are more closely repeated by the concepts of the parent findings of this thesis. This may reflect the comparatively greater inclusion of parent perspectives in the original study, as well as the separation of child and parent analysis in this thesis. Some concepts raised in the separate analysis of child findings in this thesis were not clearly represented in either the parent findings or the original CP QOL-Child work, such as children’s emphasis on inclusion in spontaneous play, and children’s developing identity, or how children developed a sense of personal agency in the context of CP impairments. Similarly, parents’ understanding of CP and the influence of diagnosis or the process of learning to accept their child’s impairments and changed expectations was not reflected in the current child findings or the original CP QOL-Child work. However, although these issues impacted on parents’ approach to support children’s QOL, they reflect a broader understanding of child QOL than is usually included in conceptual models of QOL.

There are also some differences between the findings of this thesis and the content of the items of CP QOL-Child instruments themselves (Waters et al., 2013). There is very little difference overall in the domains, however children and parents in this thesis raised some topics that are not included in the current CP-QOL items. For example, while the CP QOL-Child asks about how children feel about “being handled by other people”, and “the way they get along with their teachers and/or carers”, there is no other consideration of the impact of therapeutic relationships, or children’s agency over therapeutic decisions. The original CP QOL-Child qualitative work refers to children’s pain “associated with therapy” (Waters et al., 2005, p. 131)—an issue raised by children in this thesis—but the items reflect only general levels of pain and discomfort. Similarly, both children and parents in this thesis reflected a desire for greater play time or a greater balance between recreation and functional goals, but this is not addressed in the CP QOL Child. The item on children’s feelings about “their ability to play with friends” may be interpreted to focus on social inclusion as much as play itself, however its inclusion in the domain of participation and physical health

suggests psychometrically this was not the case. Lastly, the parent report version of the CP QOL-Child asks for parents' satisfaction about advice they receive from paediatricians, whereas in the current thesis parents referred to the need to find information on a wide range of issues from a wide range of sources as a significant part of caring for their child.

Some of the difference between the qualitative findings and CP QOL-Child items may reflect changes made to items as a result of psychometric validation (i.e. some initial items performed poorly and were removed). However, these differences have implications for any revision of the content of the CP QOL-Child inventory. The current CP QOL-Child parent report version has been developed for children at all levels of functioning, which means that several items are not relevant to children at either end of the spectrum of impairment. While overall the current analysis found concepts for all parents were similar, some issues varied across functional levels. For example, environmental accessibility for wheelchair users, or developing self-identity for children with higher functioning. It may be appropriate to develop slightly varied versions of the parent report for children with more or less severe CP to highlight these issues. Secondly, it may be appropriate to draw a clearer distinction between the content and structure of child self-report and parent report measures, in order to represent the difference in perspectives between children and parents. This would allow the parent report version to reflect wider issues of family functioning and concepts of family QOL that are of great concern to parents and impact children's QOL but are of less concern to children. Lastly, explicitly structuring both child and parent report measures on a socio-ecological model, or social-relational model of disability, may clarify priority areas for personal, family, and community level factors of children's QOL. Links to each of these models are discussed in further detail in the following chapter.

8.3 Summary

Findings from analysis of the child and parent interviews showed both overlapping and contrasting areas, with distinct differences in the overall perspectives of children and their parents. These differences should be considered in using existing measures of

QOL for children with CP, in which items and domains of child self-report instruments are largely identical to parent report. In future revisions of the CP QOL Child and any other QOL instrument developed for use with children with CP, it would be worthwhile to consider developing separate versions of the questionnaire for children and for parents, with parent report reflecting wider family QOL concerns as well as individual QOL for the child, and child report including more detailed questions on social networks and support, play, and self-identity.

Chapter 9: General discussion and conclusions

Following the individual discussions of findings for child and parent analyses, this chapter synthesises the findings and their relationship to wider theory introduced in the previous chapter. There are four main sections to this chapter. The first section reviews how the relationship between individuals and their environmental context in QOL applied to the analyses of the thesis. It highlights the relevance of considering family and wider environments for children with disability due to the impact they have on outcomes for both children and their families.

Many of the barriers to achieving high QOL and goals for good QOL perceived by both children with CP and their parents in this study were managed through relationships with others. Therefore, the second section considers ways to integrate social models of disability into conceptual models of QOL, and the implications for ongoing QOL work with children with disabilities. The third section expands on the role of children with disabilities in research about them and the need to enhance child-centric research methods on QOL for children with disabilities. Children frequently reported about the perceived value of a wide network of relationships outside of their immediate family, from which parents were often largely excluded. Without children's own perspectives, the impact that these relationships can have on children's QOL is lost.

Lastly, the fourth section of this chapter details limitations to the current work and what this means for the wider interpretation of the thesis.

9.1 Individual goals and their environmental context

Socioecological models of child development posit that a child's health and wellbeing results from the interaction of factors at the levels of child, family, community, and wider political and social environment (Irwin et al., 2007). The WHO definition of QOL emphasises that an individual's level of QOL depends on their appraisal of their personal circumstances in relation to their cultural context (WHOQoL Group, 1995).

The analysis of both child and parent interviews showed how individual goals children have for their own QOL and those that parents have for their child, themselves, and for

their family QOL, relied on both the actions of children and parents themselves and the environment in which they lived. Children relied on their parents and carers to find them accessible services for therapy and recreation, while parents relied on services being accessible in their local community, and on the actions of others to make existing services and activities accessible for themselves and their children.

A common theme for both children and parents in considering the context of QOL goals was the comparison between achieving personal goals for the child with CP and achieving goals for a similar child without a disability. For children with disabilities, the cultural context of their appraisals is usually a community which prioritises able-bodied children. This is reflected in existing literature that considers how children with disabilities develop self-identity, and how they navigate notions of difference with positive self-concept (Connors & Stalker, 2007). This formed an aspect of children's QOL in the current analysis, as discussed in Section 6.4 (see page 171). However, this also impacts other areas of QOL, such as children's satisfaction with school activities, or available recreational opportunities in their community—situations in which they are likely to be disadvantaged in comparison to their siblings and peers who do not have disabilities. Environmental barriers affected everyday activities and social relationships for both children and parents, as well as parents' ability to access care and opportunities for their child's wellbeing, impacting the QOL for all members of the family. One mother referred to this as being trapped in the 'disability bubble', reflecting not only the segregated and restricted nature of opportunities available to her family, but the comparison with a diverse and unrestricted environment available to families who do not have a child with a disability. The children interviewed also reflected on differences in availability of recreation services between themselves and their typically developing siblings and how this impacted their activities and social networking. However, for many family-oriented activities, if the environment is not accessible for one child, the whole family misses out. For both children and parents, access to dedicated disability services, especially for children's recreation, helped to bridge some of these gaps, but for most families, the ongoing comparisons served to highlight the disparity between contexts for children with and without disabilities.

For parents, the context of other support for their family care had a direct relationship with their ability to provide for the wellbeing of their child with CP. The additional reliance on parental and carer support for a child with CP affected the QOL of parents and wider family functioning. Family context, including extended family, is a factor in the QOL and wellbeing for all children, as reflected in the socioecological model (Irwin et al., 2007), however, for children with disabilities it is more likely to impact on more areas of QOL. This is not well reflected in current QOL models for children with CP, which prioritise personal factors rather than context. For example, the DISABKIDS CP module focuses on the child's feelings about their individual level of functioning in the majority of questions, with only one item focused on physically navigating their environment including context (Baars et al., 2005). Measure instruments of individual QOL for children with CP may need to expand the scope of their content to include issues of family QOL if they are to fully capture the QOL of children with CP.

Children with disabilities have a higher reliance on those around them for care and provision of educational and recreational opportunities. The ability of parents, carers, extended family, and communities to provide not just child services, but appropriate and accessible recreational services, has a direct impact on children's QOL. The enormous influence that the environment has on the experience of disability specifically for children with CP and their families suggests that models of children's QOL which do not include contextual factors, both within and outside the family, do not fully capture children's QOL. The following section discusses in greater detail how existing social models of disability may fit with models of children's QOL.

9.2 Applying disability models to QOL

Both child and parent findings demonstrated the importance of environmental context in shaping the experience of disability, as distinct from impairment. Previous researchers have suggested that QOL outcomes encompass the entire ICF model for adults, with individual QOL impacted by the outcome of interactions between individual personal functioning and environmental context (McDougall et al., 2010). The social relational model of disability emphasises that it is through social relationships that these outcomes are negotiated (Thomas, 2004a), and Connors and

Stalker (2007) argue that this model applies more strongly to children than adults due to children's greater reliance on social relationships for achieving developmental goals and identity formation.

Both child and parent findings reflected the social relational model of disability in negotiations to achieve good QOL. Children's insights reflected the relational component of disability in how children form their identities against a background of not being 'normal' as discussed by Connors and Stalker (2007), while also reflecting learning through varied social relationships. For both children and parents, navigating accessible services frequently relied on negotiation, either for accommodations to existing services, or for the provision of information on accessible or disability specific services. However, this relationship was bidirectional, with families connected to more accessible services in their local community finding increased opportunities for social connections. Children with more severe or complex disabilities necessarily relied more heavily on their parents' social connectedness, while children with milder CP more frequently relied on their own ability to navigate social relationships at school or in their peer groups, with the support of carers external to their immediate family.

The application of the social relational model of disability to QOL additionally highlights the need for dedicated models of QOL for children, rather than adult-based models, with the inclusion of child perspective in the research process. The differential expectations of social relationships from children and their parents, with children mentioning broader and more diverse social networks than their parents considered, shows the need for child insight rather than relying solely on parent report. Many aspects of child-specific QOL, such as developing agency and building self-identity, were a direct result of their child's ongoing social relational experiences specific to their disability. Parents offered only limited understanding of their child's experiences with regard to these processes. The analysis of child's perspectives of QOL in this thesis shows how the social relational model of disability can help to explain how children with CP achieve developmental goals through both social learning and disability specific social negotiations. The combination of children's own insights and parents' reflections on family context illuminates the interactions between child impairments,

family and community context, and demonstrates how social relationships can bridge contextual disparities in accessibility.

For children with disabilities, conceptual models of QOL need to include not just considerations of context, but considerations of disability in context. This research highlights the way children's unique perspective can enhance our understanding of how children with CP achieve their QOL goals outside of their immediate family relationships. Therefore, ongoing improvement of conceptual models of QOL for children with CP requires greater focus on child-centric research in this field; this is discussed in the following section.

9.3 Enhancing child-centric research

From a rights-based perspective, as introduced in Section 3.4 (page 67), it is imperative that researchers include the voices of children with disabilities in research about them. This is also the case in QOL research, which is by definition based on a person's individual and subjective judgement. From a practical standpoint, the results of the analysis of child and parent interviews in this thesis show that considering the child perspective separately from a parent perspective offers both conceptual differences to QOL models and additional opportunities to improve the wellbeing of children with CP.

Interviews with children raised three issues impacting their QOL about which parents provided only limited reflections. Firstly, children provided additional insights into the support for good QOL they found through social networks outside of their immediate family, and ways they built these networks. Secondly, children reflected specific concerns related to their developing identities and building self-esteem. Lastly, children discussed specific concerns and discomforts related to CP therapies, as well as their own understanding of the benefits from these therapies. For children in the 9-12-year age group, all three of these issues may be related to children's developing sense of personal agency, and preparation for increasing independence during their teens. In this context, children at times explicitly reflected reticence in discussing these concerns with their parents, reducing the scope of parent report of children's QOL. Methodologically, the conceptual differences between child and parent findings

emphasise the need for future research to continue to include child participants who can voice their own QOL goals.

The methods used in the interviews conducted in this thesis confirm earlier research that visual methods are highly acceptable to children, including children with disabilities (for example: Feldner et al., 2018; Gill & Liamputtong, 2014; Teachman & Gibson, 2013). The activities offered children more opportunities to discuss abstract or emotional topics, especially with the use of emoji. The flexibility of the multiple methods also encouraged the participation of children with communication impairments by allowing for children to choose their preferred alternative methods of communication. Some researchers in the field of participatory research with children have suggested that ensuring children provide assent and have opportunities to exercise choice in data collection methods, constitutes a minimum standard of research to achieve child rights' goals (Groundwater-Smith et al., 2015). However, especially in the context of disability research, some authors have stressed that researchers should be sensitive to the fact that the use of child-specific methods for data collection can be based on assumptions of a deficit model of childhood and disability. That is, blanket assumptions that children, and especially children with disabilities, do not have capacity to participate in research without specific and directive management designed by adult researchers (Cuskelly, 2007). Other researchers have emphasised the need to use research methods that not only engage children, but build their capacity to form views and allow them to meaningfully contribute (Lundy & McEvoy, 2012). Rather than recognising children as 'deficient' adults, this highlights child developmental needs while recognising their right to hold and express views on research about them and exercise choice over their participation in research.

Lewis (2010) argues for the use of reflexive practice in recognising children's right to express their views, and their right to silence in research participation. Lewis notes that children's preferred methods of communicating with adults about disability may not match a pattern of 'ideal' participation for researchers seeking to include child voice—especially children with alternate ways of communicating such as those with disabilities. Therefore, recognising children's silence in research is necessary to fulfil

goals of child agency and consent in research participation. Other researchers have discussed that multiple methods specifically can not only produce data that researchers would not otherwise have found (Darbyshire et al., 2005), but may also offer children additional opportunities to direct their own participation in the research. By engaging in activities throughout the interview, they can more easily avoid questions they don't wish to answer, while maintaining a sense of their own agency, or through activities, raising topics they would prefer to discuss. Researchers working with children using similar strategies to those of this project should maintain reflexivity from the earliest stages of data collection to recognise and allow children the opportunity not only to refuse participation directly, but how they may choose to subvert direct participation throughout the process. Other researchers have raised the need for researcher reflexivity and sensitivity to children's responses throughout the interview to ensure children's consent to participation is ongoing (Phelan & Kinsella, 2013). This better approximates a child-rights focused research process, while including children's views in research about them.

The secondary analysis of the methods in the child interviews, focusing on the interactional processes (Section 5.3, page 133) provided additional information on children's experiences both of disability and the research process. Noting interactions between parent and child in the interview setting helped to provide context to the major child interview findings especially in establishing agency. Involving parents in the research process offered ways for children to be supported to participate, particularly in the time-limited setting of a single interview. As others have discussed (Pyer & Campbell, 2013), the ubiquitous presence of parents and other adults in research with child participants creates both challenges and opportunities to both the data collection process and data analysis. Omitting consideration of how adult presence can influence qualitative data potentially omits not only additional data, but additional insight into data collected from children. The constructivist stance to qualitative methodology used in this thesis supports this additional analysis, with the contention that both what data are created and their meaning are impacted by interactions between interviewers and participants (Charmaz, 2014).

Increased opportunities for child participation in research and greater efforts to include flexible participation methods for children with disabilities offer the chance of improving the QOL of children with CP. Further information on the context of children's QOL experiences can be provided by reflecting on the interactional components of children's research participation, both in the presence of other adults and in how children choose to present their disability. In combination with the use of social models of disability adapted for children, this may provide the conditions for developing a child-centric conceptual model of QOL for children with disabilities.

9.4 Strengths and limitations of the project

Within grounded theory research, and wider qualitative methodology, there has been long-standing debate as to when and how qualitative research can be considered generalisable. One well-cited framework for evaluating qualitative research evidence in health contexts is provided by Daly and colleagues (2007). This paper defines a hierarchy of strength of qualitative evidence based on theoretical, sampling, and analytic methods, and defined relationships to existing literature. The strongest level of evidence comes from studies which have a defined conceptual basis for the initial questions and methods, include theoretical sampling methods, extend analysis beyond thematic descriptors to explain phenomena under investigation, and generalise by situating findings in the relevant literature (Daly et al., 2007).

It is asserted that the child interviews and findings of this thesis meet these criteria in the following ways. The initial research questions and sample of were defined by conceptual frameworks, including existing QOL research, with age range limited by children's ability to understand concepts of wellbeing, and variation in severity of CP. Through the data collection process, questions and prompts were altered in response to earlier interview data, which is a form of theoretical sampling. The conceptual categories that arose in the analysis were fully described in this sample and took into account the depth and breadth of data children provided. The findings are clearly related to existing literature, while extending on that knowledge. The findings presented can be broadly generalised to other children in this age group with similar severity of CP.

Findings of the parent interviews may need to be interpreted with a greater degree of caution. The research was designed with a similar conceptual framework however sampling was based on child characteristics rather than parent characteristics—this was deemed appropriate with the topic of child QOL but may have limited applicability to parent experiences. Theoretical sampling in these interviews followed a similar method as the child interviews, with even more targeted questioning and follow-ups in later interviews based on experiences encountered in the early interviews. This is a type of theoretical sampling described by Charmaz (2014). However, some aspects of the findings reflect descriptive themes, rather than deeper explanations. This suggests that some categories may not be fully saturated and would benefit from greater exploration, such as benefits and barriers in the parent process of assimilating a child's disability diagnosis. Some categories are well supported by existing literature, suggesting they may be more broadly generalisable. For example, previous authors have highlighted the notion of parents of children with disability 'living with uncertainty' (e.g. Hinton & Kirk, 2017), supporting its generalisation to other samples, and application to QOL in this thesis.

There are a few specific limitations related to the sample. Firstly, the diversity of impairments within CP itself means that in the small sample there may have been limited representation of specific comorbidities or symptoms of CP. Some types of impairments may create significant impact on QOL in ways that were not fully captured in the current analysis. For example, while efforts were taken to include children using alternative communication, the majority of the children interviewed were able to participate in a verbal interview with only limited assistance. This may downplay the impact of the use of assistive communication technology on QOL factors. Similarly, there was limited consideration of the impact of intellectual disability in the child analysis, as this was not a factor for the majority of children who completed a direct interview. However, previous researchers have suggested that the impact on families of child disability is more strongly related to adaptive and disruptive behaviour than diagnostic designation (Gardiner et al., 2018). Therefore, while this thesis focused on children with CP, the overall concepts and their relation to QOL and family function may apply to children with other disabilities.

The age limits (8-12 years) of children participating in direct interviews was deliberately chosen based on previous research related to children's ability to complete self-report QOL scales. However, the narrow age range may have limited insights into the impact of developmental aspects of children's QOL. In the context of QOL, the variation in disability severity and related experiences may be more salient for children with CP than some developmental differences, or contextual factors related to disability may be more relevant. Achievement of developmental milestones is likely to be more varied in children with CP as a direct result of CP impairments, especially for those with learning or intellectual disabilities. Other published conceptual models of QOL have not reported developmental changes, therefore developmental differences among children with disabilities may not relate to these models either. Future research spanning younger and older children may explain how developmental changes across childhood affect QOL conceptual models.

Lastly, in the sample of families interviewed there was limited cultural and economic diversity, and universally a strong, supportive bond between mother and child. Families may self-select in this regard. Barriers that reduce the likelihood of participating in voluntary research include language barriers, parents struggling with childcare and other requirements, or parents struggling to come to terms with their child's diagnosis. While there were three families for whom English was not a first language, this was the parents only, and all parents spoke fluent English. There was some variation in the economic circumstances of families which was reflected primarily in access to leisure services or private healthcare. While for the most part participating families were fairly privileged: Australian governmental support for families of children with disabilities reduces some inequities in access to healthcare, though less so for access to leisure. In addition, families who participated were from a limited geographic area, so these contextual factors may see greater variation if, for example, there were families from rural and remote communities. Lastly, 15 of the 18 families interviewed included both parents who were in stable, long term relationships, and all children demonstrated significant trust in emotional and other support from their mother. Some research suggests families of children with disabilities have higher than average rates of parental separation (Risdal & Singer,

2004), although this is debated. Additional sampling including children with more disrupted relationships between their parents, or between child and parent, may change findings related to parental and family support and child agency, especially from children's perspectives. Given the impact of context to QOL for children with disabilities, it would be worthwhile to consider including perspectives of families with more diverse contextual factors in future research.

9.5 Implications for QOL measurement

The concepts and issues discussed in this chapter based on the child interview findings can only be generalised to children who are able to self-report; how they relate to QOL for children with more severe CP or more severe communication impairments is less clear. Existing research focused on QOL differences between children with mild and severe disabilities has been limited to quantitative studies. These have usually been for scale validation with the assumption that greater severity of impairments relates to lower QOL scale scores (e.g. see review of construct validity in Carlon et al., 2010). Research exploring conceptual models of QOL has included limited consideration of the impact of disability severity. Current literature assumes that children with more severe disabilities will report lower QOL on the same measures but that the same underlying conceptual model of QOL applies across the disability spectrum. Previous qualitative research has considered parent report for children with all levels of CP together—as has this thesis—and based child QOL on the combination of parent report and child self-report for only children with mild CP. There is limited literature on whether and to what extent severity of impairment affects the QOL conceptual model—despite many validation studies of quantitative QOL measures having considered inverse correlations between QOL scores and CP severity to be evidence of construct validity, conflating functional ability with QOL.

Many of the findings of the current qualitative study have appeared in the wider literature examining children's various experiences of disability, participation, and inclusion. However, these concepts do not seem to be well incorporated into current QOL research, which has relied on adult perspectives of children's QOL. Existing measures are largely replicated between children and adults with some concessions to

language or removal of contextual items for children, but this does not accurately reflect children's understandings of context, or what children themselves nominate as impacting on their QOL. The content of existing measures reflects in large part the replication of conceptual models developed from adult perspectives transplanted directly onto children's QOL. As discussed in Section 8.1.4 of the previous chapter (page 253), parent perspectives of children's QOL more accurately reflect family QOL concepts in addition to considerations of children's individual QOL. None of the existing conceptual models of QOL for children with disability are based entirely on children's perspectives, nor are the measurement models that inform QOL scale content. Current QOL scales for child self-report are already limited to children who are able to self-report their QOL. Therefore, there seems to be limited theoretical basis for not developing conceptual and measurement models for these measures from qualitative reports of children only, rather than combining both child and parent report. While it is common to include expert or literature review in the development process for patient reported outcomes (PROs), it is rare for PRO content of any other adult population to be based on perspectives of outsiders when intended respondents are able to self-report.

The PedsQL team has developed an additional module to measure family QOL, the Family Impact Module, to be used with the core PedsQL scales (Varni et al., 2004). This scale adds evaluation of parent individual QOL, including 'worry', as well as daily family functioning and relationships to the items on child QOL of the core module. The addition of 'worry' to the parent items closely reflects the concepts of 'living with uncertainty' explored in this chapter. Both these and other measures of family QOL recognise not only the different perspectives of children and parents, but the changes to family functioning that occur to manage the care of a child with disability. From a clinical perspective, recognising the interconnectedness of family functioning and its context to child QOL is necessary to understand how to best implement interventions for improving child functioning and wellbeing.

The current study was limited in considering only the experiences of those children able to participate in largely verbal interviews. All of these children were able to report on diverse issues affecting their QOL, however, three of the eight children would be

unable to complete any of the existing self-report QOL measures for children without significant assistance. This suggests that not only is further work on this topic required to include a broader range of children with CP with more complex communication needs, but future work may need to consider alternate ways for children to complete QOL measures that do not rely on significant adult assistance—at least existing measures should be more extensively validated to understand how this assistance affects children’s responses. This research suggests that there should perhaps be greater differences in content between child and adult reports of children’s QOL; furthermore, the format for self-completion may need greater consideration for child respondents.

There are broader questions on the usefulness of subjective measures such as QOL for groups who cannot self-report or may not understand the meaning of QOL items—such as children with severe CP and/or ID. For these children as well, parent report which focuses on family QOL may be more meaningful and useful in a therapeutic context. Recently, a group of researchers have developed condition-specific measures of QOL for children with ID and another disability, for parent report only (E. Davis et al., 2017; Downs, Jacoby, et al., 2018; Epstein et al., 2016). In qualitative interviews during the development of the measures, researchers asked parents to focus only on objective indicators of children’s wellbeing or emotional state, such as visible signs of distress or pleasure. This approach was intended to lead to an underlying model of QOL with reduced assumptions of subjective states for children who cannot report QOL themselves. For those with severe CP and ID, this may be a preferable approach, however, some children at GMFCS level IV and V are able to complete a QOL scale with appropriate assistance or with accessible response options. Conceptual models may need to be developed or tested to ensure they are appropriate for each group.

QOL measures for children with disability could incorporate a wider conceptual framework for disability generally—this may help in making the distinction between ‘functional’ measures and those addressing multi-domain QOL, incorporating subjective experience of functioning and other areas of life. This is explored further in the final chapter of the thesis.

9.6 Conclusions

CP is the most common physical disability in children in Australia with impacts across many areas of children's lives (Reid et al., 2016). QOL has been recognised as a useful conceptual model for measuring the impact of health conditions across multiple areas of life, however there are shortcomings in current understandings and measurement of QOL for children with CP. This thesis aimed to develop a constructivist grounded theory of QOL for children with CP including the views of Australian children with CP and their parents, and to compare this to the content of CP QOL-Child, a current condition-specific measure of QOL for children with CP.

Constructivist grounded theory methods were used to analyse a series of individual interviews with children with CP and their parents. Critically, the child interviews included the use of multiple child-specific strategies, including drawing activities and printed picture prompts. Evaluation of the strategies showed that multiple visual methods and flexibility in the use of strategies offered clear benefits for increasing child engagement and data production. Visual methods may be especially useful for children to discuss emotional topics, or for children who use alternative methods of communication. Analysis of interactional processes in the interviews contributed to the interpretation of the main findings, especially relational aspects of disability identity.

From interviews with children with CP, five conceptual categories related to QOL were identified: socialising, play, negotiating limitations, defining the self, and establishing agency. Children negotiated for better QOL outcomes through social relationships for each of these areas. Children's wellbeing was derived from developing a positive sense of self and having fulfilling experiences of participation—both of which required a balance between disability specific adaptations and opportunities for 'normal' childhood experiences.

Parents discussed their child's QOL as part of a temporal narrative that began with diagnosis and continued through the day-to-day management to planning for an uncertain future and recognising gaps for improvement in existing services. From a parental perspective, individual child QOL was not limited only to one child but

resulted from a complex interaction of factors within the family and wider systemic considerations which require continual balance and negotiation between costs and benefits. Throughout their interviews, parents reflected on the way in which small adjustments in their community could have ongoing impacts, both positive and negative.

Both child and parent analyses in this thesis reflect the WHO definition of QOL as the combination of personal judgement in relation to cultural context. For children with CP, the cultural context is complicated by the interaction between environmental context and disability—outcomes of which are often negotiated through social relationships as in the social relational model of disability. The QOL theory developed from parent interviews reflected elements of family QOL models in addition to considerations of individual QOL. Both parent and child findings therefore supported socioecological models of child development and of disability, emphasising interactions between individual QOL goals of children, their family needs, and environmental factors in the community and wider social systems. The unique contributions to QOL concepts from child interviews highlights the value of a rights-based approach that centres the inclusive participation of children with disabilities in research about them.

The analysis of both interactions and the content of the child interviews contributes new knowledge about children's perspectives of developing identities as a person with disability, and what enhances and inhibits good QOL in children with CP. The analysis of parent interviews explores a new way of understanding parent perspectives of QOL, the temporal nature of parents' developing understanding of disability, and the nature of parent and family roles in caring for a child with disability. By comparing and contrasting child and parent perspectives with the existing CP QOL-Child, the thesis presents implications for a CP-specific measure of QOL for children with CP and their families which could be implemented in a future revision of the CP QOL-Child. The final discussion shows that future research into conceptual models of QOL for children with CP may be improved by including the social relational model of disability, as well as developmental considerations specific to children, and emphasises the need to increase child-centric research in QOL.

In conclusion, this thesis extends understandings of QOL for children with CP, and contributes new knowledge of how child and parent perspectives differ, and how both fit into wider conceptual frameworks of disability and health.

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Appendix A: Information sheets and consent form for child and parent

Information Statement: Child Copy
School of Population and Global Health



Hello my name is Elena Swift

I am from the University of Melbourne doing a research study to find out what you and other children with cerebral palsy like to do and what it means to be healthy and happy. Dr Elise Davis, Professor Dinah Reddihough, and Professor Andrew Mackinnon are also helping me with the project.

A research study is a project that people called researchers do, so that they can find out more about something.

What is an Information Sheet?

- These pages contain information about the study, please read all the pages.
- This information will help you to decide whether or not you would like to take part.
- You can ask us or your family any questions about this information sheet.
- You can decide if you want to take part in the study or not. You don't have to - it's up to you.
- If you say yes and then you change your mind later, that's ok just let us know.

What is the study about?

We want to know what makes you feel happy, healthy and safe.

This information will help us make a questionnaire that doctors and therapists can use to help people your age with cerebral palsy.

Why am I being asked to be in this study?

- Because you are aged between 9 – 12 years and you have cerebral palsy.

What will happen if I say that I want to be in the study?

- We will have a chat with you about your life, and your feelings about different things you do.

- We will audio-tape the chat, but only the researchers will be able to listen to the tapes if that is ok with you.
- The researchers might use some of the things you say in a report about the study, but no-one will know that it's you. We won't say your name in the report and no one will know that you were in the study, unless you tell us that it's ok for us to say your name.
- All of the information that we have about you from the study will be stored in a safe place and we will look after it very carefully for 5 years, then we will destroy it.



Are there any good things about being in the study?

The information we find out from this study might also help other young people with cerebral palsy.

Are there any bad things about being in the study?

We don't think the questions will make you upset. But if you become upset you can stop the chat, or talk with your mum or dad or another adult you trust.

Will you tell me what you learnt in the study at the end?

Yes, we will if you want us to. There is a question on the consent form if your mum or dad ticks Yes, when we finish the study we will tell you what we learnt.

What if I am not happy with the study or the people doing the study?



If you are not happy with the study, then you or the person who looks after you can call:

The Executive Officer, Human Research Ethics,
The University of Melbourne, on ph: (03) 8344 207

This sheet is for you to keep.

How do I agree to take part in the study?

Your parents can contact: Ms. Elena Swift at eswift@student.unimelb.edu.au , 0403 177 131 or Dr Elise Davis eda@unimelb.edu.au

Funding is from a PhD scholarship from the Centre for Research Excellence in Cerebral Palsy (CRE-CP).

Information Statement: Parent/Guardian Copy

School of Population and Global Health



Project: Developing a version of the CP QOL-Child for clinical settings

Dr. Elise Davis (Responsible Researcher)

Tel: +61 3 8344 0921 Email: eda@unimelb.edu.au

Ms Elena Swift (PhD student) Email: eswift@student.unimelb.edu.au

Prof Andrew Mackinnon, Co-Supervisor; Prof Dinah Reddihough, Co-Supervisor

Introduction

You are invited to participate in the above project, which is being conducted by Dr Elise Davis (Supervisor) and Elena Swift (PhD student) of the School of Population and Global Health at The University of Melbourne. Your name and contact details have been selected from the Victorian Cerebral Palsy Register as you have a child with cerebral palsy between 9 and 12 years old, and indicated you would be happy to be invited to take part in cerebral palsy research.

Purpose of the research

The aim of this study is to investigate what is important for children with cerebral palsy to have a good quality of life. Quality of life means feelings about different areas of life, such as family, school life, social activities, physical health and pain, or anything else that is important to their wellbeing. This information will be used to develop a short questionnaire about children's quality of life that can be used by researchers, doctors, physiotherapists, psychologists, and others working with children with cerebral palsy. This will help them to understand what your child needs to have their best possible life. This research has been approved by the Health Sciences Human Ethics Sub-Committee at the University of Melbourne.

What will I be asked to do?

If you agree to participate, you will be asked to take part in an individual interview with the researchers. We will ask questions about the kinds of things your child usually does, what they enjoy, and what is important for them to be well. We may also ask about some of the things that have been challenging for your child.

What is my child being asked to do?

As the research is about what is important to children's wellbeing, we also want to ask children directly about what matters to them. If you think your child would like to take part in an interview with us, please read through with them the separate information sheet provided. They will be asked similar questions about what they do and their feelings about different areas of their life. We may also ask them to look at some pictures such as emojis or children's activities. If you or your child require communication assistance or an interpreter this can be arranged.

How long are the interviews expected to take?

We estimate that each interview will take 30 – 60 minutes. If you or your child wish to end an interview earlier for any reason, you are free to do so.

How will my confidentiality be protected?

We intend to protect your anonymity and the confidentiality of your responses to the fullest possible extent, subject to any legal requirements. We will discuss or publish the results but this will be done so that participants are not identified. Given the small number of families who will be taking part we cannot guarantee anonymity. Any information we collect that can identify you will remain confidential, except as required by law. Your name, your child's name, and your contact details will be kept in a separate, password-protected computer file from any data supplied. Only researchers on this project and the Ethics committee will have access to the data, including audio-tapes. The data will be kept securely in the Melbourne School of Population and Global Health for 5 years from the date of publication, and may be destroyed after this time.

Do I have to take part?

Participation is completely voluntary. Should you wish to withdraw at any stage, or to withdraw any unprocessed data you have supplied, you are free to do so without prejudice.

What happens after the project is finished?

We will discuss or publish the results at conferences and in academic journals. This research will also form part of Elena Swift's PhD thesis. A brief summary of the research findings will be made available to you and your child; there is a space on the consent form to indicate if you would like this sent to you. The quality of life questionnaire that we develop will also be available online for use by researchers and health professionals.

What if I have any concerns about the project?

Should you have any concerns or complaints about the conduct of the project, please contact Ms Kate Murphy, Manager, Human Research Ethics - Office for Research Ethics and Integrity, the University of Melbourne VIC 3010. Tel: +61 3 8344 2073 or HumanEthics-complaints@unimelb.edu.au.

How do I agree to participate or find out more?

If you would like to participate in this project, please complete the following page with your contact details, and return it to the research team in the envelope provided. Alternately, you can contact the researchers directly: Dr Elise Davis on (03) 8344 0921, or Elena Swift on 0403 177 131.

What if I don't want to participate?

If you don't want to participate in the project, please complete the second part of the following page and return it to the Victorian Cerebral Palsy Register.

This project is funded through a PhD scholarship from the Centre for Research Excellence in Cerebral Palsy (CRE-CP).

Consent Form: Parent/Guardian and Child

School of Population and Global Health, University of Melbourne



Project: Developing a version of the CP QOL-Child for clinical settings

Primary Researcher: Dr Elise Davis

Additional Researchers: Elena Swift, PhD Candidate; Prof Andrew Mackinnon, Co-Supervisor;
Prof Dinah Reddihough, Co-Supervisor

1. I consent to participate in this project, the details of which have been explained to me, and I have been provided with a written plain language statement to keep.
2. I understand that the purpose of this research is to investigate what is important for my child to feel well and have a good quality of life.
3. I acknowledge that the possible effects of participating in this research project have been explained to my satisfaction.
4. I understand that participation in the research will involve participation in an interview. I understand that my interviews may be audio-taped and I agree that the researcher may use the results as described in the information sheet.
5. I understand that my participation is voluntary and that I am free to withdraw from this project anytime without explanation or prejudice and to withdraw any unprocessed data that I have provided.
6. I understand that the data from this research will be stored at the University of Melbourne and will be destroyed after 5 years.
7. I have been informed that the confidentiality of the information I provide will be safeguarded subject to any legal requirements; my data will be password protected and accessible only by the named researchers.
8. I understand that after I sign and return this consent form, it will be retained by the researcher.

Child Consent

Your Name : _____

Signature: _____

Date: _____

Parent/Guardian Consent

The study has been explained to the child who has signed above, and I believe that they understand the purpose, extent and possible effects of their involvement in this study.

Your Name : _____

Signature: _____

Date: _____

I would like a summary report on research findings: ☐ Yes ☐ No

If yes, please provide your email or home address so we can send you a copy of the summary report

Email: _____

Address: _____

HREC Number: 1646922.1 Project Start Date: 30 June 2016 Version: 2

Appendix B: Information sheet and consent form for parents only

Information Statement: Parent/Guardian Copy

School of Population and Global Health



Project: Developing a version of the CP QOL-Child for clinical settings

Dr. Elise Davis (Responsible Researcher)

Tel: +61 3 8344 0921 Email: eda@unimelb.edu.au

Ms Elena Swift (PhD student) Email: eswift@student.unimelb.edu.au

Prof Andrew Mackinnon, Co-Supervisor; Prof Dinah Reddihough, Co-Supervisor

Introduction

You are invited to participate in the above project, which is being conducted by Dr Elise Davis (Supervisor) and Elena Swift (PhD student) of the School of Population and Global Health at The University of Melbourne. Your name and contact details have been selected from the Victorian Cerebral Palsy Register as you have a child with cerebral palsy between 4 and 8 years old, and indicated you would be happy to be invited to take part in cerebral palsy research.

Purpose of the research

The aim of this study is to investigate what is important for children with cerebral palsy to have a good quality of life. Quality of life means feelings about different areas of life, such as family, school life, social activities, physical health and pain, or anything else that is important to their wellbeing. This information will be used to develop a short questionnaire about children's quality of life that can be used by researchers, doctors, physiotherapists, psychologists, and others working with children with cerebral palsy. This will help them to understand what your child needs to have their best possible life. This research has been approved by the Health Sciences Human Ethics Sub-Committee at the University of Melbourne.

What will I be asked to do?

If you agree to participate, you will be asked to take part in an individual interview with the researchers. We will ask questions about the kinds of things your child usually does, what they enjoy, and what is important for them to be well. We may also ask about some of the things that have been challenging for your child. If you require communication assistance or an interpreter this can be arranged. Your interview will be audio-taped, but only the researchers above will be able to listen to the recording.

How long is my participation expected to take?

We estimate that the interview will take 30 – 60 minutes, but if you would like to end it sooner for any reason you are free to do so.

How will my confidentiality be protected?

We will protect your anonymity and the confidentiality of your responses. We will discuss or publish the results but this will be done so that participants are not identified. Given the small number of families who will be taking part we cannot guarantee anonymity. Any information we collect that can identify you will remain confidential, except as required by law. Your name, your child's name, and your contact details will be kept in a separate, password-protected computer file from any data supplied. Only researchers on this project and the Ethics committee will have access to the data. The data (including audio-tapes) will be kept securely in the Melbourne School of Population and Global Health for 5 years from the date of publication, and may be destroyed after this time.

Do I have to take part?

Participation is completely voluntary. Should you wish to withdraw at any stage, or to withdraw any unprocessed data you have supplied, you are free to do so without prejudice.

What happens after the project is finished?

We will discuss or publish the results at conferences and in academic journals. This research will also form part of Elena Swift's PhD thesis. A brief summary of the research findings will be made available to you; there is a space on the consent form to indicate if you would like this sent to you. The quality of life questionnaire that we develop will also be available online for use by researchers and health professionals.

Where can I get further information?

If you would like more information about the project, please contact the researchers; Dr Elise Davis on (03) 8344 0921, or Elena Swift on 0403 177 131.

What if I have any concerns about the project?

Should you have any concerns or complaints about the conduct of the project, please contact Ms Kate Murphy, Manager, Human Research Ethics - Office for Research Ethics and Integrity, the University of Melbourne VIC 3010. Tel: +61 3 8344 2073 or HumanEthics-complaints@unimelb.edu.au.

How do I agree to participate?

If you would like to participate in this project, please complete the following page with your contact details, and return it to the research team in the envelope provided. Alternately, you can contact the researchers directly: Dr Elise Davis on (03) 8344 0921, or Elena Swift on 0403 177 131.

What if I don't want to participate?

If you don't want to participate in the project, please complete the second part of the following page and return it to the Victorian Cerebral Palsy Register.

This project is funded through a PhD scholarship from the Centre for Research Excellence in Cerebral Palsy (CRE-CP).

Consent Form: Parents

School of Population and Global Health, University of Melbourne



Project: Developing a version of the CP QOL-Child for clinical settings

Primary Researcher: Dr Elise Davis

Additional Researchers: Elena Swift, PhD Candidate; Prof Andrew Mackinnon, Co-Supervisor;
Prof Dinah Reddihough, Co-Supervisor

1. I consent to participate in this project, the details of which have been explained to me, and I have been provided with a written plain language statement to keep.
2. I understand that the purpose of this research is to investigate what is important for my child to feel well and have a good quality of life.
3. I acknowledge that the possible effects of participating in this research project have been explained to my satisfaction.
4. I understand that participation in the research will involve participation in an interview. I understand that my interviews may be audio-taped and I agree that the researcher may use the results as described in the information sheet.
5. I understand that my participation is voluntary and that I am free to withdraw from this project anytime without explanation or prejudice and to withdraw any unprocessed data that I have provided.
6. I understand that the data from this research will be stored at the University of Melbourne and will be destroyed after 5 years.
7. I have been informed that the confidentiality of the information I provide will be safeguarded subject to any legal requirements; my data will be password protected and accessible only by the named researchers.
8. I understand that after I sign and return this consent form, it will be retained by the researcher.

Your Name :

Signature:

Date:

I would like a summary report on research findings: ☐ Yes ☐ No

If yes, please provide your email or home address so we can send you a copy of the summary report

Email: _____

Address: _____

Appendix C: Qualitative interview schedule for child participants

I would like to know about different parts of your life, like your family, your friends, your health, or your school.

Tell me what are the things you enjoy?

Tell me about the things you find challenging in your life.

[showing child series of different emoji faces] which one of these do you think is most like you lately? what made you choose that one?

further prompts depending on response – e.g. what made you feel like that?

[showing the child a series of age-appropriate images of children's activities and settings] I have a lot of pictures here of things that might be important or enjoyable. Do you think any of them look like things that are important to you or things you enjoy? – follow with further prompts as above

For children capable of self-reporting on the CP QOL-Child questionnaire: Do you think you would be comfortable to answer a questionnaire about these sorts of things, like your feelings about your life, if your physio/doctor/[other health service child has mentioned] asked you to?

Appendix D: Qualitative interview schedule for parent participants

- Tell me about your child's life – how they spend their time, what things they enjoy?
- What do you think are the most important things for your child to be well and to have a good life?
 - o Prompts if not covered by initial answers: e.g. at home, at school, in the community, in terms of health, in the future...
 - o possible additional prompts: CP specific questions – eg is this different because of your child's impairments?, if they don't raise limitations – are there things that prevent your child having a good life?
- Present parents with the domains of the current CP QOL-Child (these are family and friends, participation, communication, health, special equipment, pain and bother, access to services, parent/carer health); ask if they agree or disagree to the inclusion of these themes in a version for use with their health professionals – also use these are prompts if they haven't raised up any of these in earlier open questions
- We are planning to use this research project to create a new survey form for health professionals to learn about the QOL of children with CP who they are treating. How do you feel about your child's health professionals asking you and them about your child's QOL?
 - o Are there things you would not want to discuss with your child's health professionals? prompt – such as family finance or difficulty accessing other services, or your own health and QOL