Title: Exploring quality of life of children with cerebral palsy and intellectual disability: What are the important domains of life?

Running title: Quality of life in cerebral palsy

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Abstract
BACKGROUND Although it is estimated that half of all children with cerebral palsy (CP) also have comorbid intellectual disability (ID), the domains of quality of life (QOL) important for these children are not well understood. The aim of this study was to identify important domains of QOL for these children and adolescents.
METHODS Due to the children's communication impairments, qualitative semi-structured interviews were conducted with 18 parents. The children (9 males) had a median age of 12 (range seven to 17) years at interview and nearly two thirds were classified as Gross Motor Function Classification System IV or V. A grounded theory approach was used to identify domains of QOL.
RESULTS The eleven domains identified as important to QOL were physical health, body comfort, behavior and emotion, communication, predictability and routine, movement and physical activity, nature and outdoors, variety of activity, independence and autonomy, social connectedness and access to services.
CONCLUSIONS The domains of quality of life that emerged from this study will be useful for professionals who support children with CP and their families. They will also be important for developing a QOL instrument essential for informing the development of interventions and their monitoring and evaluation.

Keywords: Quality of life, cerebral palsy, intellectual disability, children, adolescents

Key messages

- Children with CP and ID face unique issues beyond those captured in current CP specific QOL instruments.
- The domains 'predictability and routines' and 'opportunity to enjoy nature and the outdoors' are not traditionally included in QOL instruments for children.

INTRODUCTION

With a prevalence of approximately 2 per 1000 births,(Reid *et al.* 2016) cerebral palsy (CP) is the most common physical disability in childhood and is considered to be a permanent disorder of movement and posture.(Rosenbaum *et al.* 2007) Many areas of the lives of children with CP, including physical, social and emotional wellbeing and participation, may be impacted by their disability.(Colver *et al.* 2014) Consequently, there is need for substantial support, especially as approximately half of these children also have an intellectual disability (ID).(Reid *et al.* 2016)

Quality of life (QOL) is broadly defined as "the individual's perceptions of their position in life in the context of the culture and value systems in which they live, and in relation to their goals, expectations, and concerns".(World Health Organization 1993) Assessment of QOL is increasingly used as a mechanism to gain insight into a child's life, to identify areas of life that are positive or challenging, and to inform and evaluate interventions. The International Classification of Functioning, Disability and Health (ICF) provides a framework for conceptualizing different aspects of life affected by disability.(World Health Organization 2001) These have been worded more contemporaneously as the F-words (Fitness, Function, Family, Friends, Fun) which together build towards the child's Future.(Rosenbaum and Gorter 2011) The adult literature regards QOL as an attribute of the reporter and therefore self-report is critical to an accurate appraisal. In the pediatric literature, child self-report is also recognized as important.(Bjornson and McLaughlin 2001, Davis *et al.* 2007) However, despite technological advances, for some children with marked communication difficulties or ID, parent-proxy reporting may be the only option given a child's inability to communicate verbally or their reduced capacity to understand and respond to questionnaires or provide narrative during interviews.

Increasing interest in measuring the QOL of children with CP has resulted in the development of five CP specific instruments over the last 10 years. These instruments vary in their purpose, origin, domains, opportunity for self-report, item wording, their length and psychometric properties,(Waters *et al.* 2009) as well as their applicability for children with comorbid ID. Specifically, the Cerebral Palsy Quality of Life Questionnaires (CP QOL-Child and CP QOL-Teen) focus on wellbeing rather than functioning or limitations and were developed based on qualitative interviews with children with CP and their parents.(Davis *et al.* 2009, Waters *et al.* 2005) In contrast, the Pediatric Quality of Life Inventory (PedsQL)-CP Module was based on a combination of previous scales, with contributions from parents and health professionals, and items focus on functioning.(Varni *et al.* 2006) The Child Health Index of Life with Disabilities (CPCHILD) does not measure QOL, but was designed to assess caregiver priorities in domains of personal care; positioning, transfer and mobility; communication and social interaction; comfort, emotions and behavior; and health.(Narayanan *et al.* 2006) The DISABKIDS-CP Module was developed to include the perspectives of children and parents, however the items are negatively worded and may threaten self-esteem (e.g. "Do people think you are not as clever as you are").(Baars *et al.* 2005)

It must be recognized that these instruments were all developed over 10 years ago and it is questioned whether the domains of QOL remain the same. In that time, there have been social changes as well as changes to disability and health care that could impact expectations and outcomes and in turn influence our understanding and perceptions of

quality of life. In addition an understanding of the QOL domains that are important for children with CP and ID has not been developed previously. The qualitative study that informed the development of the CP QOL-Child used data from 28 families of children across different levels of functioning,(Waters *et al.* 2005) but the number with ID was unknown. A later qualitative study investigated the applicability of the CP QOL measures for children in the UK and most of the children had no or mild ID (22/28).(Parkinson *et al.* 2011) Overall, there are limited data on the important areas of quality of life for children with CP and ID. There has, however, been no investigation of the areas of life that contribute to QOL specifically for children with CP and ID. In our experience some parents report that completing the CP QOL-Child, which is designed to be used for all children with CP, is challenging because they are not aware how their child feels about a particular area of their life, such as whether they feel accepted in their community.

QOL instruments developed for children with ID could be useful for children with CP and ID, but instruments measuring the QOL of children with ID are few. The parent/carer report of the Quality of Life-Profound Multiple Disabilities (QOL-PMD) was developed for use with children and adults with the greatest support needs.(Petry *et al.* 2009) A new QOL instrument, KidsLife, has been developed but is based on a predefined model of eight domains that have been shown to be important for adults rather than children.(Gomez *et al.*) We do not know whether the domains of QOL that are important for adults with ID differ from children with ID, but in the general population, QOL instruments for children include different domains to those for adults.

We have previously identified domains of QOL important to children with Rett (Epstein *et al.* 2016) and Down(Murphy *et al.* 2017) syndromes by coding parent-reported observations that indicated the child's pleasure, satisfaction or challenge with different aspects of their life. More information is needed about the areas of life that are important for children with CP who also have ID. Given communication limitations, parent-proxy reports based on ratings of what they believe their child's perceptions to be rather than their own perceptions, are the best available way of gathering this information.(Davis *et al.* 2007) In the absence of any research specifically focused on children with CP and ID, the aim of this study was to identify the domains of QOL important for children with CP and ID. Due to the children's communication impairments, this preliminary study utilizes parent-proxy reports of their children's QOL.

METHODS

Participants

Eligible participants were identified from the Victorian Cerebral Palsy Register, a population-based registry established to collect data on individuals with CP born or living in Victoria, Australia, from 1970 onwards.(Reid *et al.* 2016) Recruitment was purposive for sex, age, health issues and area of residence to optimise variability within the sample. Of 373 families who were provided information by email about the study by registry staff, 18 responded that they were interested in being contacted by the researchers and all then agreed to participate. The 18 children (9 males) had a median age of 12 (range 7-17) years at the time of interview. Nearly two thirds of children were classified as Gross Motor Function Classification System (GMFCS)(Palisano *et al.* 1997) level IV or V and more than three quarters were non-verbal. Routinely collected

information on the registry indicated that each of the children had ID and experienced at least one comorbid medical condition (e.g. epilepsy). Half of the parents reported that their child had behavioral problems (e.g. hitting or biting) (Table 1).

Procedure

Qualitative research was undertaken using a grounded theory approach, allowing the domains of QOL to evolve naturally through constant assessment and contrasting of different themes as captured in the interview transcripts.(Strauss and Corbin 1990) Semi-structured stem and leaf telephone interviews were conducted by two researchers with psychology training (NM and AE), using our previously described methods.(Epstein *et al.* 2016, Murphy *et al.* 2017) Of note, the additional leaf questions captured observable behaviors that more clearly illustrated aspects of life quality by asking parents to provide specific examples that supported their proxy report (e.g. *"How do you know this?"*).

Recordings were transcribed and given to parents to provide them with the opportunity to edit or add information, and 77% (14/18) of families provided feedback. Coding data during the data collection period confirmed that new themes were not emerging and that thematic saturation was achieved after the participation of 18 parents. A Consumer Reference Group (CRG) meeting was held with four parents who participated in the study interviews to review the domains and further inform interpretation of the findings. Ethics approval for this study was provided by the Human Research Ethics Committee at the University of Western Australia, Western Australia (RA/4/1/6931) and parents provided informed consent to participate in this study.

Analyses

The interview transcripts were read and reread, and data describing observable behaviors were coded by NM using NVivo (10th Ed, QSE International Pty Ltd, Burlington, MA). Similar segments of interview data were compiled and coded into themes until no new themes or "domains" were identified. Four researchers (ED, NM, AE and JD) reviewed and interpreted the categorization and labeling of the domains with joint discussion until a consensus was achieved. Domains were then compared with those in five parent-report QOL measures designed for CP, including the CP QOL Child and Teen,(Davis *et al.* 2009, Waters *et al.* 2005) PedsQL v3 CP,(Varni *et al.* 2006) CPCHILD(Narayanan *et al.* 2006) and DISABKIDS-CP.(Baars *et al.* 2005)

RESULTS

Quality of life domains

Eleven domains were identified as important to QOL in children with CP and ID and sample quotes are shown in Figure 1. Parents in the CRG discussed the 11 domains in the light of their experiences and concurred with data coding.

Function

"Communication" referred to verbal or non-verbal expressions. This domain included the child's ease or difficulty in making choices and conveying feelings or sharing details of their own experiences. Parents discussed their child's ability to communicate feelings in a variety of ways if they were not able to use language, or how they showed frustration when unable to verbalize or express their needs. Some parents commented

that their child showed pleasure in simply being with others and listening to their conversations.

"Predictability and routine" referred to feeling comfortable with known patterns of activity, familiar people and recognizable environments. Some parents described how their child felt more relaxed when routines were followed. Others indicated their child was happy for changes in their environment or routines, provided a clear explanation was given in advance.

"Movement and physical activity" referred to opportunities to be mobile and active. For example, walking, being active in water, and participating in physical activities such as horseback riding.

Fitness

"Behavior and emotion" referred to behaviors and associated emotional states. Observable behaviors included variations in body language that indicated emotional wellbeing and willingness to cooperate. Parents described their children as smiling and being interested in the environment and activities, or in contrast, being distressed and frustrated with moodiness, anxiety, aggression and poor impulse control.

"*Body Comfort"* referred to comfort or discomfort experienced because of the child's physical condition or sensory needs and could include sensory stimulation such as the enjoyment of listening to conversations or alternatively physical pain or discomfort.

"Physical health" referred to bodily health and wellness. This domain included energy levels and fatigue, nutrition, fitness, illnesses, comorbidities and poor saliva control. Surgical and other medical procedures were sometimes associated with improvements in a child's life, or alternatively, with set-backs in learning or community engagement.

Fun

"Nature and outdoors" referred to opportunities to be outside, to enjoy nature, gardening, or spending time with pets, farm animals, or visiting the zoo.

"Variety of activities" referred to participation in different home-based and community endeavors. This domain included activities such as listening to music, watching TV, playing sport, being a part of a cheer squad, or attending special events such as theatre and concerts.

"Independence and autonomy" referred to developing skills that provided opportunities for the child to control their own actions and aspects of their environment. Elements related to the child's choice and personal preferences, mastery and achievement, and developmental maturity. Several parents gave examples of children expressing pleasure when mastering new activities that ranged from self-care to domestic tasks and when making persistent effort to improve strength and physical capability.

Family/Friends

"Social connectedness and relationships" referred to interactions within social settings. This domain included elements such as social inclusion and acceptance, shared enjoyment and anticipation, expressions of love and affection, empathy and other

behaviors that reflected social maturity. Children's sensitivity to how others responded to them, involvement in shared activities, excitement when sharing news and playful interactions were also described.

"Access to services" referred to experiences with support services and included elements such as service accessibility and provision of necessary financial assistance. Parents discussed the importance of the right match between the services offered and their child's needs, professional sensibility, being provided with appropriate equipment and access to facilities.

Comparison of domains with those in comparative measures

Concept mapping of our data against the five existing QOL measures indicated that some of the domains within the comparative measures also emerged as domains in our dataset (e.g. physical health, behavior and emotion, body comfort and communication) (Table 2). CP QOL-Child and CP QOL-Teen included the most domains in common with ours and DISABKIDS-CP the least. None of the measures included all the currently identified domains, and the domains "predictability and routine" and "nature and the outdoors" were not included in any of the measures (Table 2). It is also acknowledged that the items within the domains for these existing tools may be very different or not applicable for children with CP and ID. For example, our finding that communication included detail such as expressing happiness when being understood was not articulated in the comparison measures.

DISCUSSION

The 11 QOL domains identified as important for children with CP and ID aligned with concepts described in the International Classification of Functioning, Disability and Health (ICF) model of disability(World Health Organization 2001) and their associated F-words (Fitness, Function, Family, Friends, Fun)(Rosenbaum and Gorter 2011) as illustrated in Figure 1. The domains that emerged from our qualitative data could be viewed as essential life domains necessary for professionals to consider when counselling and supporting these children and their families. It is interesting that 10 years after the existing questionnaires were developed, the domains observed by parents as important remain similar. Previously identified domains were also observed but there were some important differences.

Difficulties in functioning may have impacted on the child's QOL. Many of the children had substantial difficulties with communication and relied on their communication partners to be sensitive to subtle changes in their vocalisations or gestures. Communication devices such as tablets, computers or phone applications, head switches and other equipment provided a framework to enable choice-making for exchanging feelings of love and affection and for some sharing of news and events. The children displayed a sense of accomplishment with access to physical activities and independent achievement of tasks, such as indicating toileting needs or by opening their mouth during teeth cleaning. Many of the children thrived on their day to day routines and would become upset if unexpected changes occurred. The value of routines was consistent with our findings for Rett syndrome(Epstein *et al.* 2016) and Down syndrome(Murphy *et al.* 2017) and not previously reported in other QOL measures(Solans *et al.* 2008) including those for CP.(Baars *et al.* 2005, Davis *et al.* 2009, Narayanan *et al.* 2006, Varni *et al.* 2006) The presence of childhood routines affects

wellbeing in the general population(Sytsma *et al.* 2001) and was also observed to be supportive of functioning in our sample with CP.

The domains related to fitness concerned physical and behavioral wellbeing. Children with more severe motor impairment are more frequently affected by poor health including the presence of pain (Colver *et al.* 2014) and comorbidities such as epilepsy and respiratory illnesses, which were also associated with more hospital admissions compared to children with less severe CP.(Meehan *et al.* 2016) Mental health problems can also affect individuals with CP(Parkes *et al.* 2008) with some studies suggesting poorer mental wellbeing in those with comorbid ID.(Parkes *et al.* 2008) Parents reported that when children were healthy and happy, they were more likely to be involved in social or other activities. Conversely, a distressed or irritable child sometimes engaged in tantrums or other injurious behaviors detrimental to QOL.

Aspects of fun included the child's involvement in a variety of recreational activities (e.g. horse riding) as well as independent pastimes (e.g. listening to music). Children's participation in a range of activities was less common in those who were more severely affected, yet these activities remained an important source of pleasure, helping to build confidence and enhancing skills.(Majnemer *et al.* 2008) As is being increasingly recognized in the general population,(Bratman *et al.* 2012) participation in the natural environment was associated with enjoyment and relief of stress. This domain has not been articulated in other QOL measures(Baars *et al.* 2005, Davis *et al.* 2009, Narayanan *et al.* 2006, Solans *et al.* 2008, Varni *et al.* 2006) but was observed in our qualitative studies on Rett syndrome(Epstein *et al.* 2016) and Down syndrome.(Murphy *et al.* 2017)

The domain of 'social connectedness and relationships' focuses on the child's interactions with family and friends. The importance of social relationships and inclusion in activities observed by the parents has been previously documented.(Colver *et al.* 2014, King *et al.* 2006) Family and friends played an important role in the lives of our sample of children with CP and ID, and the quality of social relationships was critical to building a sense of personal worth and wellbeing. Important elements that have not been previously identified included excitement when sharing news with others, and being present and involved during mealtime conversations. New technologies and applications (e.g. video chatting with friends) helped make social connections available, which facilitated children's engagement with their peers. Access to services, such as timely replacement of equipment, may mean the difference between comfort or discomfort for some children or engagement in rewarding pastimes using communication, educational and recreational aids.

A suite of QOL measures has been developed for CP but their domains do not fully correspond to those identified in our study, possibly because their development methods did not focus on ID. Our domains most closely mapped to CP QOL-Child.(Waters *et al.* 2005) This is not surprising because the CP QOL-Child was also developed using qualitative methods with a focus on wellbeing rather than on limitations,(Baars *et al.* 2005) function,(Baars *et al.* 2005, Varni *et al.* 2006) or parent priorities,(Baars *et al.* 2005) which may not align with the concept of QOL.(World Health Organization 1993) However, it must be acknowledged that the items within the domains may need to be quite different if the child has comorbid ID. For example, the

CP QOL-Child assesses emotional wellbeing through how the child feels about themselves and their opportunities, whilst the current study suggests suitable items for a new measure would need to take into account behaviors such as frequency of smiling, and those that indicate frustration, problems with impulse control and aggression. Furthermore the CP QOL-Child omits potentially important domains of 'predictability and routine' and 'nature and outdoors'. In contrast, the domains identified in our study were remarkably consistent with those identified for children with Rett syndrome (Epstein *et al.* 2016) and Down syndrome.(Murphy *et al.* 2017) ID in children appeared to be associated with commonalities for QOL that are not restricted to a specific diagnosis. Similar to our study with Down syndrome,(Murphy *et al.* 2017) we did not observe differences in the domains reported for children or adolescents.

We relied on parent report given the pronounced communication difficulties that many of the children experienced because we did not want these children to be excluded from QOL investigations. However, we recognize that this is associated with limitations because QOL is an experiential concept and parents and children do not always agree as to the composition of a child or young person's QOL.(Davis *et al.* 2007) Some parents described ongoing observations of their child to piece together combinations of words, gestures and emotional tone to determine what was satisfying or challenging for their child. To overcome this potential limitation, our interview schedule included probing questions eliciting what was observed by parents, and it was only the reported observations that were coded, not expressed personal views or values of the parents. Although recruited from a population-based database to capture variability in child experiences, most children had severe disability and we may not have captured all relevant domains for those with CP and milder ID. It would also be important to replicate our methodology in other settings to check that our domains are replicable.

Through the observations of parents, our study revealed the children's reactions to life's challenges and rewards. It reinforced the need for attention to concepts such as adequate and timely responses to physical and emotional needs, building and maintaining communication skills with social and community involvement, and the need to aim for best possible levels of independence. Novel themes in relation to 'predictable routines' and 'opportunity for time in the natural environment' emerged, and could indicate useful ways that day to day QOL could be increased. The current CP measures do not address the unique aspects of QOL for children with CP and ID and an important task is to translate these findings into a measure that can quantify QOL for this group. Another challenge is to develop methods that can directly capture the points of view of the children with milder ID and gain insights into their views on important domains of QOL. Guided by the diversity of communication strategies that the children used, this might be possible with creative use of media and technology along with skilled delivery of tailored approaches to communicate with each child. The ultimate goal would be to capture their reactions to experiences and situations that have been raised within each of these domains.

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Table 1. Characteristics of children with cerebral palsy (n=18).

	Number (%)
Age	
Middle childhood (6 to 11 years)	8 (44.4)
Adolescence (12 to 18 years)	10 (55.6)
Sex	
Female	9 (50.0)
Male	9 (50.0)
Location of residence	
Urban	11 (61.1)
Rural	7 (38.9)
Gross Motor Functioning Classification System (GMFCS)	
Level II	5 (27.8)
Level III	2 (11.1)
Level IV	5 (27.8)
Level V	6 (33.3)
Intellectual impairment	
Severe Impairment	5 (27.8)
Mild to Moderate Impairment	3 (16.6)
Unknown Severity	10 (55.6)
Speech Impairment	
Some impairment	4 (22.2)
Non-verbal	14 (77.8)
Parent reported behavior problems[a]	9 (50.0)
Medical comorbidities	
Hearing impairment	8 (44.4)
Vision impairment	8 (44.4)
Epilepsy	7 (38.9)
Gastrostomy	7 (38.9)
Lower limb musculoskeletal problems necessitating orthopaedic surgery	7 (38.9)
Diagnosis of scoliosis	3 (16.6)

[a] These included hitting, biting, throwing objects, and threatening behaviors.

Table 2. Quality of life domains from the current cerebral palsy dataset and their presence in the five comparative measures.

Domains for children with CP and ID	CP QOL-Child[a]	CP QOL-Teen	PedsQL-CP[b]	CPCHILD[c]	DISABKIDS-CP
Physical health	X	X		X	X
Behavior and emotion	X	X		X	X
Body comfort	X	X	X	X	
Communication	X	X	X	X	X
Movement and physical activity	X	X	X	X	X
Predictability and routine					
Independence and autonomy		X			X
Social connectedness and relationships	X	X		X	X
Variety of activities	X	X	X		
Nature and outdoors					
Access to services	X	X			

[a] Cerebral Palsy Quality of Life (CP QOL).
[b] Pediatric Quality of Life Inventory (PedsQL).
[c] Caregiver Priorities and Child Health Index of Life with Disabilities (CPCHILD).

FIGURE LEGEND

Figure 1. Domains of quality of life important for children with cerebral palsy and intellectual disability, shown alongside the F-words.(Rosenbaum and Gorter 2011)

COMMUNICATION

“Unless someone has known him for a while, it is very hard for him to communicate.”

“He has got some words. He will tell you bits and pieces. You have to try and work it out yourself.”

“Her receptive language is quite good. I can say to her ‘your book is in your school bag’, and she will go to her school bag.”

PREDICTABILITY & ROUTINE

“He is not a kid that copes with a sudden change at all. And again he will vocalise, or get irritated or shut down.”

MOVEMENT & PHYSICAL ACTIVITY

“She really likes physical things. She likes jumping on the trampoline, the swing; there is a pool at her Dad’s house. She loves swimming.”

“Oh he loves it. That is why he loves the horse because it trots and bounces. He just thinks that is awesome. And he loves jumping, evidently. He can’t jump to save himself but that is what he thinks he likes. Yes he loves movement.”

NATURE & OUTDOORS

"He loves fresh air. Even when he is distressed and upset it will always be the circuit breaker.”

“He loves going for walks along the river with his dog. He has a companion dog.”

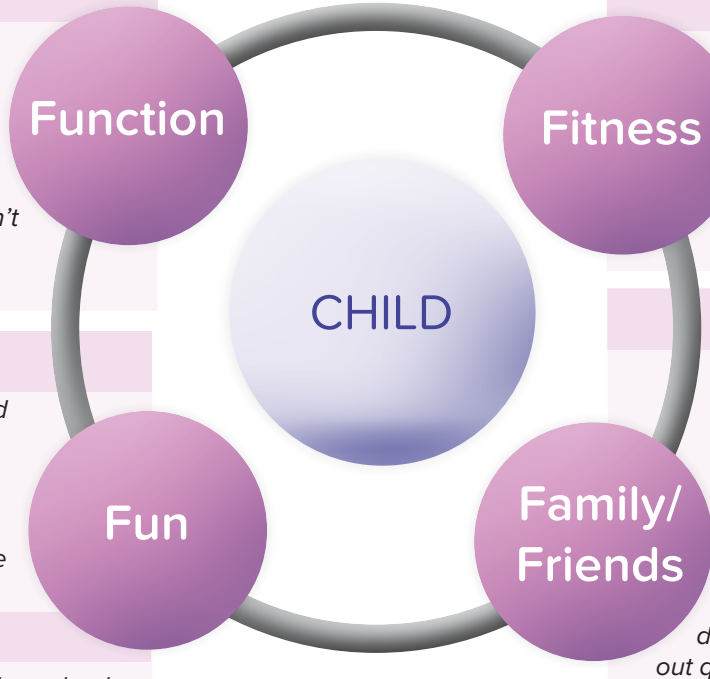
“And she loves those elements on her face, like the sun and wind and things like that.”

VARIETY OF ACTIVITY

“Enjoying listening to music players, watching TV, horseback riding, swimming, football, being in a cheer squad, special events such as musical theatre and concerts.”

INDEPENDENCE & AUTONOMY

“He took ages to be able to ride his bike. My husband used to get him and push him on the bike and then run after him. It took him a long, long time before he finally got on the bike. Now if you see him on the bike with one arm and what he can do is amazing.”



BEHAVIOUR & EMOTION

"She has big smile on her face, she is excited and her body started trembling. She was pretty happy about that.”

“I mean she will get frustrated and she will get upset. She flaps her arms, as she does not know how to tell you, so the arms are flapping, so that’s how I know there is something wrong. And crying also.”

BODY COMFORT

“Every time he is in pain he bites onto his lip and he can’t let go. It is like a muscle spasm.”

PHYSICAL HEALTH

"He has days when he is just unwell, because of these seizures or medications, there is no alertness.”

“She has had scoliosis surgery. She has had seven surgeries. She has had two spine, three jaw and two ankle surgeries. Pulled up fine after every single one of them. She is pretty remarkable really.”

SOCIAL CONNECTEDNESS & RELATIONSHIPS

“And she is really compassionate. Like she will be the first one there if you have hurt yourself. She will be right there giving me a cuddle. If someone has hurt themselves at school she will be there saying they need a Band-Aid.”

“Anyone worth their grain of salt will take time, even to just hold his hand, and he responds to that. Whereas someone who approaches, who looks down at him and talks down at him. He works people out quite quickly really. He will just shut off.”

ACCESS TO SERVICES

“They have kids with disabilities who are really not included with the other kids. They are off in their own room with their separate integration aide. And that is not inclusion. That is parallel education. I was going ‘You could do awesome stuff in the school as you have full time aiding for my child.’ And they were just busy telling me ‘This is so hard because we don’t know what she thinks because she can’t talk’”.