

# CPCHILD

## **Caregiver Priorities and Child Health Index of Life with Disabilities (CPCHILD<sup>®</sup>) Questionnaire**

*An instrument to assess the health status, comfort, wellbeing and ease of  
caregiving of children with severe cerebral palsy*

### **Manual and Interpretation Guide**

The CPCHILD<sup>®</sup> Manual & Interpretation Guide: Narayanan, Weir, Fehlings, 2007.

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# Preface

This manual describes the Caregiver Priorities and Child Health Index of Life with Disabilities (CPCHILD<sup>®</sup>) Questionnaire. The CCHILD<sup>®</sup> measures caregivers' perspectives on the health status, comfort, well being, functional abilities and ease of caregiving of children with severe developmental disabilities. It was developed to measure the effectiveness of interventions intended to improve or preserve these outcomes for children with severe disabilities, including non-ambulant children with severe cerebral palsy, and traumatic or other acquired brain injuries.

The CCHILD<sup>®</sup> Manual & Interpretation Guide contains information about the rationale for, and development and validation of the CCHILD<sup>®</sup> questionnaire, as well as basic information about the administration and scoring of the questionnaire.

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# 1. Introduction

## 1.1 Cerebral Palsy

Cerebral palsy (CP) describes a group of permanent disorders of the development of movement and posture, causing activity limitation, that are attributed to nonprogressive disturbances that occurred in the developing fetal 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 <sup>1</sup>. The cerebral palsies are among the most common cause of chronic childhood disability, with overall rates in the developed world estimated to be between 2 and 2.5 per 1000 live births <sup>2,3,4</sup>. Although the primary lesion in the brain is non-progressive (static encephalopathy), the pathology is permanent and many of the clinical manifestations, including the musculoskeletal consequences, are acquired and progressive over time. Between 25% & 35% of these children are severely involved and experience difficulties with their activities of daily living, communication, mobility, and their health, and are dependent on their caregivers for most of their needs. These conditions have a significant and lifelong impact on the children, their caregivers and families, and to the agencies responsible for their well being <sup>5,6,7</sup>.

Children with severe cerebral palsy are often subjected to a number of interventions including different forms of therapy, bracing and medications in addition to major surgical procedures. These include orthopaedic surgery to address joint contractures, hip instability, and spinal deformities; neurosurgical procedures to alleviate hypertonia; and gastro-intestinal procedures to address feeding difficulties and gastro-esophageal reflux. These interventions share the common goals of relieving or preventing pain, facilitating

caregiving and improving the quality of life of these children. However, we have no reliable means to measure these phenomena in children with severe disabilities. Instead we rely on radiographic outcomes (e.g. hip development, or spinal alignment) or measures of impairment (e.g. range of motion, spasticity) which may or may not be associated with functional or health related quality of life improvements for these patients. Many of these interventions are resource intensive and expensive, and the evidence that they are effective in making an impact in these areas is elusive. There is an imperative to evaluate these interventions using outcomes that are more meaningful to patients and their caregivers, such as health related quality of life<sup>8</sup>.

## **1.2 Outcomes That Matter**

In 1948, the World Health Organization (WHO) defined health as “a state of complete physical, mental and social well-being, not merely the absence of disease or infirmity”<sup>9</sup>. The conceptualization of the complexity of health has led to the incorporation of a more holistic account of the human experience that recognizes individual rights and empowerment<sup>10</sup>. This evolution has been accompanied by changes in the way we measure health and disease. Interest in patients’ perspectives and the concepts of patient-centred care have grown with our understanding that the impact of health care interventions are more meaningfully assessed using patient based outcome measures<sup>11</sup>. Equally important, are patients’ perspectives prior to, and during their care to allow the incorporation of their values or preferences during the process of medical decision making<sup>12</sup>. The growth of the outcomes research movement was fuelled by the need “to sort out what works in medicine and to learn how to make clinical decisions that reflect more truly the needs and wants of the individual patients”<sup>13</sup>. This has led to the development of instruments that measure a wide range of health related phenomena, including physical and psycho-social function, patient

satisfaction and quality of life <sup>14</sup>. Reiser has described this new focus on the illness experience as the beginning of “the era of the patient” <sup>15</sup>.

### **1.3 Health Related Quality of Life (HRQL)**

While it may never be possible to assess or measure complete wellbeing, health related quality of life (HRQL) comprises the *health related aspects* of life that contribute to the goodness and meaning of life, including one’s perception of wellbeing, and one’s perceived ability to fulfill certain life roles. HRQL is a multidimensional construct that incorporates physical, mental, and social wellbeing as well as role attainment, daily functioning, and participation in community life <sup>16</sup>. HRQL measures are believed to provide a more complete picture of an individual, and are complementary to traditional biomedical measures and functional assessments <sup>17</sup>.

### **1.4 Importance of Measuring HRQL in Children with Disabilities**

It is particularly important to measure the quality of life (QOL) of children with significant chronic disabilities such as cerebral palsy <sup>16,18</sup>. In the absence of a cure, the consequences of these conditions are significant and lifelong. Most interventions share the common goals of preserving or improving the quality of life of these children, but are often associated with significant costs in terms of time, stress and resources to patients, families and to society in general, and not always with good evidence that these interventions are making meaningful differences to these children or their caregivers and families <sup>8</sup>. Reliable measurement of HRQL or quality of life can help identify individual priorities that are problematic, so that therapeutic objectives, programs and policies may be aligned with needs of patients and caregivers <sup>16,19,20,21</sup>. This information might assist medical decision-making, and may be used in clinical practice to evaluate the patient’s response to interventions that can guide on-going treatment or alteration in management

<sup>22,23</sup>. Consideration of HRQL is therefore crucial to designing and maintaining a system of patient-centered care <sup>16</sup>. Since the goal of most interventions for these children is to preserve or improve quality of life, these outcomes must be included in clinical trials of these interventions whenever possible <sup>24</sup>.

In keeping with this imperative, the American Academy of Cerebral Palsy and Developmental Medicine (AACPDMD) used the model of disablement constructed by the National Center for Medical Rehabilitation Research (NCMRR) to serve as a conceptual framework to analyze the effects of interventions on persons with chronic illness or disability <sup>25,26</sup>. More recently the AACPDMD has adopted the World Health Organization's International Classification of Function, Disability and Health (ICF) model <sup>27,28</sup>. Despite the recognition of the importance of HRQL there are no suitable measures of these outcomes for children with severe cerebral palsy.

## **1.5 Challenges in Measuring HRQL in Children with Disabilities**

There are inherent challenges to measuring QOL, health-related or otherwise. QOL is a difficult concept to define and even more difficult to measure. QOL may mean different things to different people and may mean different things to the same person at different times. Indeed, QOL may be a dynamic phenomenon, evolving as priorities change over time. Whose perspective should be considered becomes paramount in considering measurement of QOL. The medical community, general society, and much of quality of life research tends to assume that life is better if illness, disability, and pain are absent, but this may not always be entirely true for those experiencing these health states. There is sometimes a disconnect between children's objective physical wellbeing and their own perceptions of their psychosocial wellbeing <sup>29,30,31</sup>. Those with disabilities tend to rate their own HRQL much higher than members of the general public would rate the same health

state. A restructuring of values seems to occur, suggesting that quality of life is both dynamic and highly personal<sup>22</sup>. Consequently, the very low values attached to the lives of the elderly and those with permanent disabilities by utility-based instruments might be misleading when applied to individual patients or for clinical decision making for these persons, because these instruments assign values to health states based on population norms. When we are interested in making therapeutic recommendations or wish to evaluate the effectiveness of these recommendations for individuals, we should use measures that evaluate the perspective of these individuals, and not the perspectives of society as a whole<sup>22,32</sup>.

This creates special challenges in attempting to measure HRQL in children with severe developmental disabilities. These children are often unable to communicate their perspective on quality of life. We are compelled to rely on the parents or caregivers not only to report their perception of their child's quality of life but also to determine which dimensions of HRQL to measure in the first place. Such reports are no longer truly a measure of the child's HRQL but a proxy that inevitably will be influenced by the parent's or caregiver's unique perceptions and attitudes, value judgments, as well as elements of their own quality of life. Nevertheless, such reports are the closest possible approximation of severely disabled children's HRQL and have generally proven to be reliable and valid<sup>33,34,35</sup>. Fortunately, this is consistent with real life practice – where health care providers (ought to) respond to the concerns of parents or caregivers and their perception of their children's needs to arrive at recommendations that address these priorities. While some aspects of the caregiver's quality of life and issues pertaining to the ease of caring for a child with severe disabilities are inseparable from any proxy measure of the child's quality

of life, addressing these issues is valuable and beneficial to both the caregivers and their children<sup>36</sup>.

## **1.6 Brief Overview of Established Measures**

A few condition specific or generic measures exist which attempt to measure HRQL or aspects of HRQL of children with severe disabilities. These have either not been well validated or are inappropriate for children at the severe end of the spectrum of disability, who are non-ambulatory and are dependent on caregivers for most of their daily needs.

The Caregiver Questionnaire (CQ), developed at the Rehabilitation Institute of Chicago, was intended as a disease specific health related quality of life measure for children with spastic quadriplegic cerebral palsy undergoing selective dorsal rhizotomy<sup>19</sup>. It has been modified by the developers of the questionnaire and by others. One modified version, renamed the Care and Comfort Caregiver Questionnaire, was used to evaluate a cohort of children and adults with cerebral palsy who were treated with intrathecal baclofen for management of spasticity<sup>37</sup>. Another version, named the Care and Comfort Hypertonicity Questionnaire (CCHQ) was reported to be responsive to increases in dosage of intrathecal baclofen<sup>38</sup>. Neither the original Caregiver Questionnaire nor its modifications have been formally evaluated for their reliability or validity. Furthermore, several items pertinent to the quality of life of severely disabled children, such as health, emotional and behavioural issues are missing from these questionnaires.

Generic health status measures developed for children, such as the Child Health Questionnaire (CHQ)<sup>39</sup> and the Pediatric Quality of Life Inventory (PedsQL)<sup>40</sup>, and functional measures such as the Pediatric Evaluation of Disability Inventory (PEDI) do not adequately represent the HRQL in children with severe cerebral palsy. The CHQ measures the physical and psychosocial well-being of children above the age of 5 years<sup>39</sup>.

It has been extensively validated in children with conditions such as asthma, juvenile rheumatoid arthritis, epilepsy and attention deficit disorder. When applied to children with cerebral palsy, caregivers of children who were more severely involved indicated that the “questions were too high level for their children”<sup>19</sup>. Others have also shown significant floor effects when the CHQ was used in more severely involved children with cerebral palsy<sup>41,42</sup>. More recently, a disease specific module of the PedsQL has been developed. However, many of the items in the PedsQL do not apply to non-ambulatory children with severe cerebral palsy, and some items that might contribute to quality of life in these children are not represented<sup>43</sup>. Consequently, these instruments are unlikely to be sensitive or responsive outcome measures for this population.

In the absence of a suitable instrument to measure HRQL in children with severe cerebral palsy, we set out to develop a disease specific measure that could measure the effectiveness of various interventions that apply to this population.

## 2. Development of the CPCHILD<sup>®</sup>

Based on the recommendations from caregivers, health care professionals experienced in the management of children with severe cerebral palsy, and a review of other questionnaires we constructed a new measure that we called Caregiver Priorities and Child Health Index of Life with Disabilities (CPCHILD<sup>®</sup>). Specifically, we set out to establish that the CPCHILD<sup>®</sup> is a reliable and valid proxy measure of functional and health status, caregiver burden and health related quality of life in children with severe cerebral palsy. The following section summarizes the initial development and validation of the CPCHILD<sup>®</sup>, the results of which were published in *Developmental Medicine & Child Neurology*<sup>36</sup>.

### 2.1 Initial Development of the Questionnaire

In the first phase of development, items for the new questionnaire were generated from recommendations of parents and caregivers of children with severe cerebral palsy, health care professionals across multiple disciplines experienced in the management of these children, and a review of other outcome measures<sup>19,39,44</sup>. A pilot version of the new questionnaire was constructed, allowing caregivers the opportunity to suggest additional items and provide feedback. Their recommendations were incorporated into subsequent versions of the questionnaire. During multiple iterations, new items were added, and others deleted or modified. The new questionnaire was called the Caregiver Priorities and Child Health Index of Life with Disabilities (CPCHILD<sup>®</sup>) Questionnaire. The initial questionnaire was further tested in a broader cross-sectional survey of caregivers of children with cerebral palsy and traumatic brain injury, and solicited written feedback about

the overall content and structure of the questionnaire and about the wording, relevance and importance of each item, and suggestions for additional items in each domain.

The initial version of the CPCHILD<sup>®</sup> consisted of 36 items across six domains (number of items): 1) Personal care (8); 2) Positioning, transfer and mobility (8); 3) Communication and social interaction (7); 4) Comfort, emotions and behaviour (9); 5) Health issues (3); and 6) Overall quality of life (1). For items pertaining to the performance of specific tasks or activities, the *degree of difficulty* of accomplishing each task was rated on a 7-point ordinal scale from 0 (“No problem at all”) to 6 (“Impossible”) in addition to a 6-point *level of assistance* modifier (from “independent” to “total assistance”) for each task/activity to quantify the degree of dependence on the caregiver to accomplish the activity. For the items about specific experiences, the frequency of symptom/experience was rated on a 6-point ordinal scale from 0 (“None of the time”) to 5 (“All the time”) plus a 3-point “Intensity” scale to quantify the magnitude or severity of the symptom/experience. In each section respondents were allowed to rate additional items that they felt were pertinent to that domain. In Section 7 (Importance of Items), caregivers were asked to rate the importance of each of the items’ (in the questionnaire) contribution to their child’s overall quality of life, using a 6-point ordinal scale, from 0 (“Least important”) to 5 (“Most important”). The purpose of this section was to determine whether the items were relevant to individual caregivers and their children, and to what extent the items in the questionnaire were related to the caregivers’ perception of their child’s HRQL. The importance ratings quantified the relevance of the items in the questionnaire, and were used as a measure of face validity. The importance score rated below the threshold level of “slightly important” by caregivers could be used to drop items from the questionnaire. Variation in the importance ratings between items might provide the basis for differential

weighting of individual items, while significant variation in importance ratings between caregivers might justify the generation of a caregiver-child specific index score <sup>22</sup>. Standardized scores from 0 (best) to 100 (worst) were calculated for each of the 6 domains, and for the total survey <sup>36</sup>.

## **2.2 Initial Validation of the CPCHILD<sup>®</sup> Questionnaire**

Primary caregivers (n=77) of 45 boys and 32 girls between 5 and 18 years old (mean age: 13 yrs; SD 3 yrs) with cerebral palsy, categorized by the Gross Motor Function Classification System (GMFCS) level completed the CPCHILD<sup>®</sup>. Caregivers of children with severe cerebral palsy (GMFCS levels IV & V) also completed a second administration of the CPCHILD<sup>®</sup> two weeks after the first. The mean CPCHILD<sup>®</sup> scores for children with severe cerebral palsy (GMFCS IV & V) was 56.2 (range: 24-93). The mean CPCHILD<sup>®</sup> scores for children in GMFCS levels I to V were 22.0, 38.2, 23.0, 44.5 and 59.3 respectively (p<0.0001). Reliability was tested in the 41/52 caregivers who reported no health status change between the 2 administrations of the CPCHILD<sup>®</sup>. The intra class correlation coefficient was 0.94 (95% CI: 0.90-0.97). The CPCHILD<sup>®</sup> was found to be a reliable and valid measure of caregivers' perspectives on the health status, functional limitations, and well-being of children with severe non-ambulatory CP <sup>36</sup>. (Please Refer to Appendix 1 for details).

## **2.3 The Revised CPCHILD<sup>®</sup> Questionnaire**

The initial validation work established the reliability, face and content validity, and construct validity of the CPCHILD<sup>®</sup>. Based on caregivers' ratings of importance of the individual items and their feedback on the content, wording and format of the questionnaire, and a

survey of 34 health care professionals involved in the care of children with severe disabilities (developmental pediatricians, pediatric physiatrists, pediatric neurologists, orthopaedic and neurosurgeons, physiotherapists, occupational therapists, orthotists, seating specialists, and nurses) the CPCHILD<sup>®</sup> questionnaire was revised. These modifications included revision of the initial instruction page with clearer examples of responses, minor changes to the format, and revision of the wording of some items to ensure consistent interpretation and improve readability. Descriptive labels were added to each of the levels of the ordinal rating scales.

The CPCHILD<sup>®</sup> currently consists of 37 items distributed over among 6 sections representing the following domains:

1. Activities of Daily Living/Personal Care (9 items)
2. Positioning, Transferring & Mobility (8 items)
3. Comfort & Emotions (9 items)
4. Communication & Social Interaction (7 items)
5. Health (3 items)
6. Overall Quality of Life (1 item)

The direction of scoring was reversed. For the two sections involving participation in an activity and/or performance of a particular task or skill (Sections 1. & 2.), the *degree of difficulty* of accomplishing each task or activity was rated on a 7-point ordinal scale anchored by 0 ('Impossible') and 6 (No problem at all). In addition, the *level of assistance* required to accomplish each task was rated on a 4-point ordinal scale (reduced from the original 6 point scale) from 0 ('total assistance') to 3 ('independent'). Each item could therefore receive an aggregate score from a minimum of 0 (for a task that was impossible to accomplish even with total assistance) to a maximum of 9 (for an activity that was

accomplished with no problem at all entirely independently). The level of assistance modifier was dropped for Section 4. (Communication and Social Interactions) as some respondents seemed to have difficulty interpreting the “level of assistance” required for day to day communication. In Section 3. (Comfort and Emotions) the *frequency* of discomfort was rated on a 6-point ordinal scale from 0 (‘Everyday’) to 5 (‘None of the time’). An additional 4-point scale of *intensity* quantified the magnitude of the discomfort or emotional/behavioural problem from 0 (‘severe’) to 3 (‘None’). Each item could therefore receive an aggregate score from a minimum of 0 (for severe discomfort every day) to a maximum of 8 (No pain at any time). Items in the Sections on Health and Quality of life were rated on a 6-point ordinal scale.

Each section allowed respondents to rate additional items that caregivers felt were pertinent to that domain. Raw item scores were transformed to a scale from 0 (worst) to 100 (best) by dividing the raw item score by the maximum possible item score and multiplying by 100. Standardized scores were generated on a scale of 0 (worst) to 100 (best) in keeping with convention of other measures of HRQL, and were calculated for each of the domains (by taking the arithmetic mean of all items within the corresponding section), as well as for the total (mean of all 36 items in the questionnaire). Missing values were handled by removing the missing items from the calculation, provided that no more than half of the items from any domain were incomplete.

The questionnaire takes approximately 20 to 30 minutes to complete, though there is wide variation between caregivers.

## 3. Measurement Properties of the CPCHILD<sup>®</sup>

### 3.1 Overview

The CPCHILD<sup>®</sup> team has conducted a number of validation studies to establish the reliability and validity of the CPCHILD<sup>®</sup> Questionnaire. The participants of these studies were the primary caregivers of children with severe developmental disabilities either due to cerebral palsy or traumatic brain injuries. To be eligible, patients had to be between 5 years and 19 years old and the caregiver(s) must have lived with the child for at least the previous 6 months. Primary caregivers of children with ambulatory cerebral palsy or age matched temporarily non-ambulant children with bilateral lower extremity trauma were used as controls. Whenever possible, children who were able to communicate were invited to complete the CPCHILD<sup>®</sup> Questionnaire as well as their parents or caregivers. To date only a small proportion of children have been able to complete the questionnaire, since the vast majority of the sample tested were cognitively impaired.

### 3.2 Reliability

Reliability is a fundamental attribute of any valid measure. The reliability of the CPCHILD<sup>®</sup> was assessed in three ways.

- 1) Test-retest reliability was evaluated by re-administering the CPCHILD<sup>®</sup> two weeks after the first administration.
- 2) Inter rater reliability was tested by inviting two caregivers of the same child (whenever possible) to complete the questionnaire.
- 3) Internal consistency was tested using Cronbach's alpha.

Reliability of the domain and total scores were estimated using single measure intraclass correlations coefficients (ICC) including 95% confidence intervals using a fixed-

effects model <sup>45</sup>. In the initial validation, 52 caregivers of children completed the second administration of the CPCHILD<sup>®</sup> two weeks after the first, of which 41 reported no interval health status change in their children. The mean of the absolute differences in total scores between first and second administration was 4.1 points (SD 3.2; Range: 0 to 11 points). The intraclass correlation coefficient for the total questionnaire score was 0.97 (95% CI: 0.95-0.99). The intraclass correlation coefficient (ICC) ranged from 0.88 to 0.96 for the 6 domain scores.

The revised version of the CPCHILD<sup>®</sup> Questionnaire underwent reliability testing again. (See Table 1.) The absolute differences in CPCHILD<sup>®</sup> total scores between test-retest administrations two weeks apart ranged from 0.51 to 14.17 with a mean of 5.89 for the total score (SD 4.01). Overall reliability was excellent with an ICC for CPCHILD<sup>®</sup> total score of 0.85 (95% CI: 0.68-0.93). The ICC ranged from 0.63 to 0.87 for the six domain scores.

The internal consistency of the CPCHILD<sup>®</sup> was assessed for each of the domains/sections using Cronbach's  $\alpha$ , which uses item-to-item correlations to assess the homogeneity of multi-item scales<sup>46</sup>. Internal consistency was high with Cronbach's alpha exceeding the minimum threshold of 0.70 for all domains (0.74 to 0.93).

**Table 1. Reliability Statistics**

<b>CPCHILD<sup>®</sup> Scores</b>	<b>Absolute differences Mean (SD)</b>	<b>ICC<sup>a</sup> (95% CI)</b>	<b>Cronbach's alpha</b>
<b>1. Personal care &amp; ADLs</b>	9.41 (9.55)	0.63 (0.31 - 0.82)	0.77
<b>2. Positioning, transferring and mobility</b>	8.97 (7.73)	0.77 (0.54 - 0.89)	0.87
<b>3. Comfort and emotions</b>	7.57 (5.49)	0.87 (0.73 - 0.94)	0.93
<b>4. Communication and social interaction</b>	10.58 (10.13)	0.84 (0.46 - 0.87)	0.84
<b>5. Health</b>	8.06 (8.56)	0.81 (0.63 - 0.92)	0.90
<b>6. Overall quality of life</b>	14.17 (15.01)	0.59 (0.26 - 0.80)	0.74
<b>Total Score</b>	5.89 (4.01)	0.85 (0.68 - 0.93)	0.91

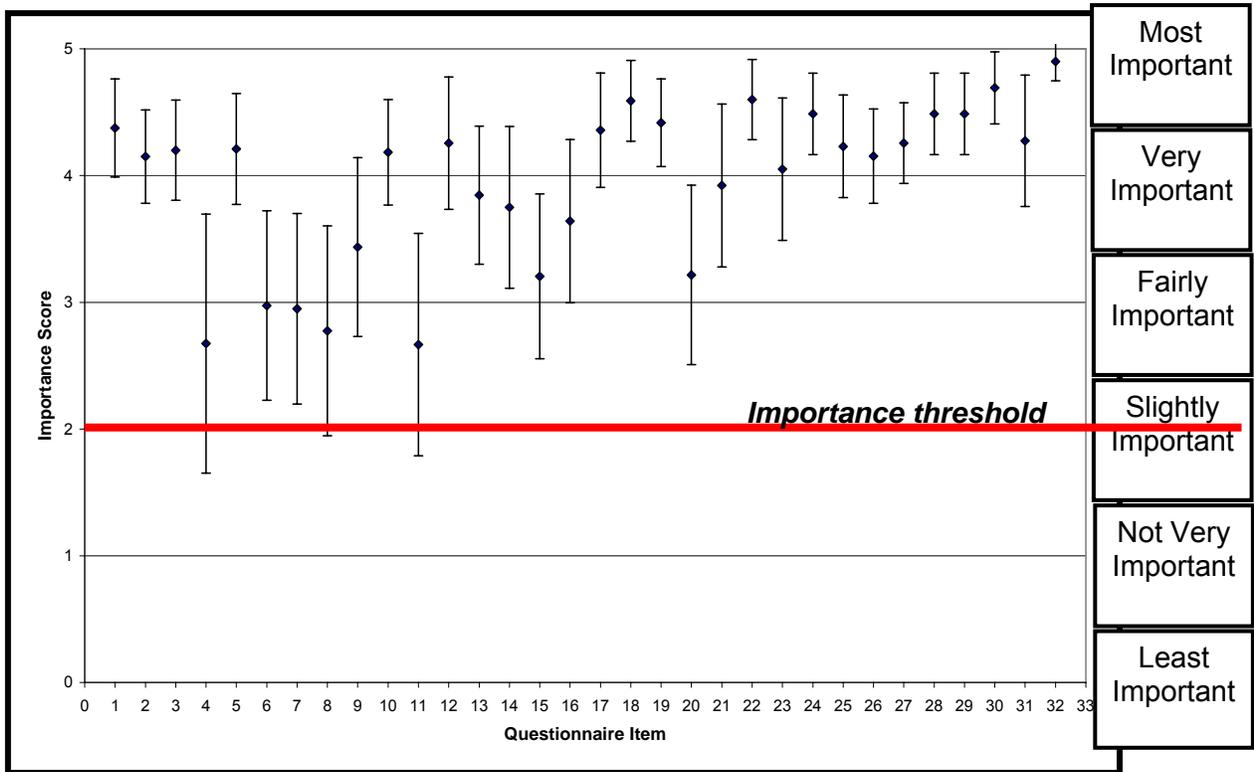
<sup>a</sup>Two-way mixed effect model

### 3.3 Face and Content Validity

A measure is valid when it fully and accurately captures the attribute that it is intended to measure. Face validity is the most basic kind of validity. It simply examines whether an instrument appears to be measuring what it is meant to measure<sup>47</sup>. Content validity examines the extent to which the attribute of interest is comprehensively sampled by the items or questions in the instrument<sup>47</sup>. To ensure that both face and content validity existed, each item in the CPCHILD<sup>®</sup> was examined by a group of experts and professionals who work closely with children with severe disabilities, including physical therapists, occupational therapists, orthotists, nurses, physicians, and caregivers themselves. The panel members scored each item on a 3 point scale: 0 (reject), 1 (accept with modifications), or 2 (accept). They also provided reasons and recommendations where applicable, which led to modifications of the questionnaire. This consensus exercise critically evaluated the CPCHILD<sup>®</sup> for its content, wording, format, ease of administration and scoring.

Additionally, Section 7 of the questionnaire incorporates the caregivers' perspective on the importance of each item in the CPCHILD<sup>®</sup> questionnaire. This importance rating, which quantifies the relevance of the items in the questionnaire, was used as a measure of face validity. Caregivers rated the importance of each item's contribution to their child's quality of life (QoL) on a 6-point ordinal scale anchored by 0 (least important) and 5 (most important). The mean importance rating for all CPCHILD<sup>®</sup> items was 3.63 (SD = 0.54, Range: 2.54 to 4.74). No items were rated below the threshold level of importance (2.0 = slightly important). Items relating to comfort, emotions, and communication were considered more important contributors to quality of life than items relating to personal care and mobility. The most important items not surprisingly related to happiness (4.74),

ability to be understood by the caregiver (4.39) and emotional status and behaviour (4.34).  
 (See Figure 1.)



**Figure 1. Mean importance scores with standard deviations.** (Importance score of 2 = “slightly important” was arbitrarily chosen as the threshold level of importance, below which items would be dropped or modified.)

### 3.4 Construct Validity

Construct validity is generally used when dealing with abstract variables, such as quality of life, and is the most rigorous test of validity. The proposed or hypothetical underlying factors are referred to as constructs. Construct validity examines the logical relations that should exist between a measure and characteristics of patients and patient groups<sup>47</sup>. This test tries to address the question of whether or not the scores of the questionnaire correlate with other related constructs in the anticipated manner. Construct validation is an

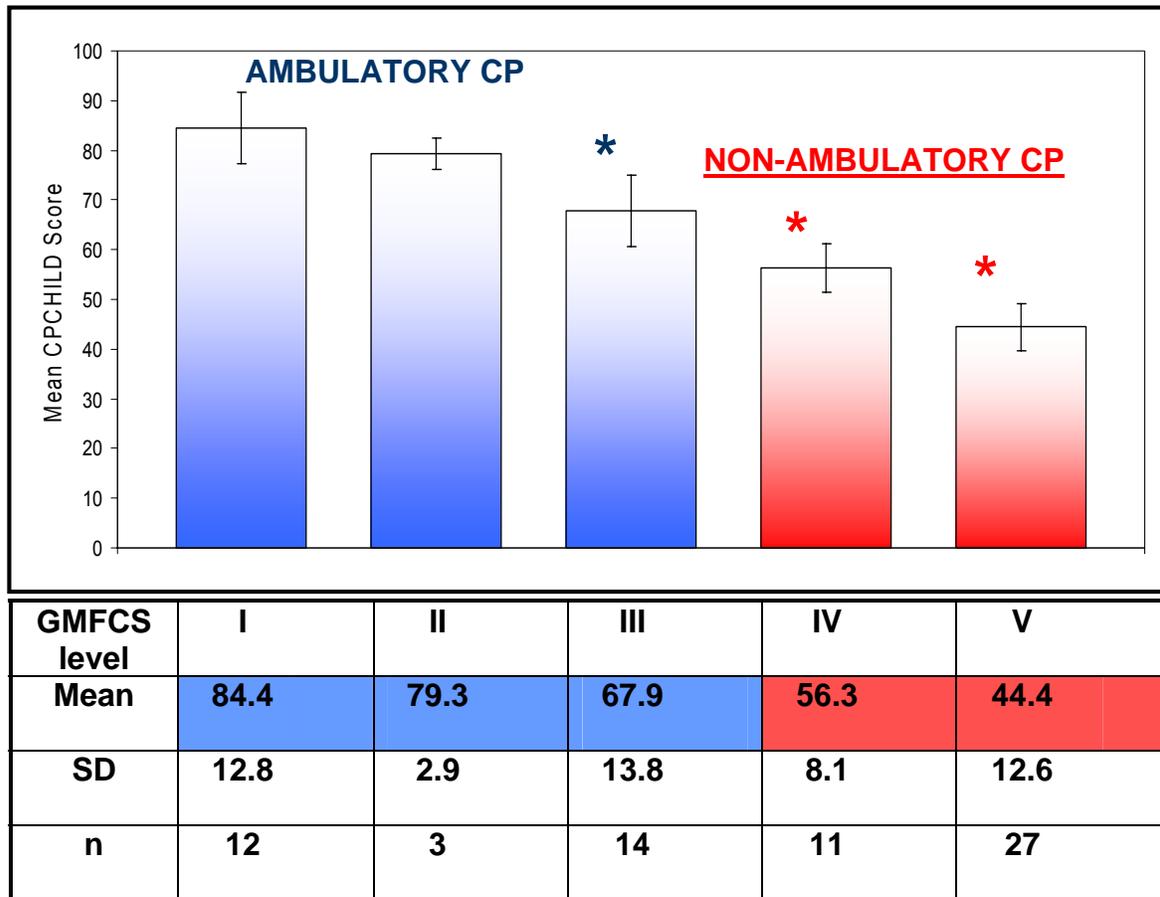
on-going process of learning more about the construct, making new predictions, and then testing them <sup>49</sup>.

Construct validity of the CPCHILD<sup>®</sup> was assessed using known-groups validity, convergent validity, and sensitivity or discriminatory ability.

### **3.4.1 Known Groups Validity**

Known-groups validity involves evaluating whether children's scores on a measure are as expected given some already established grouping variable.

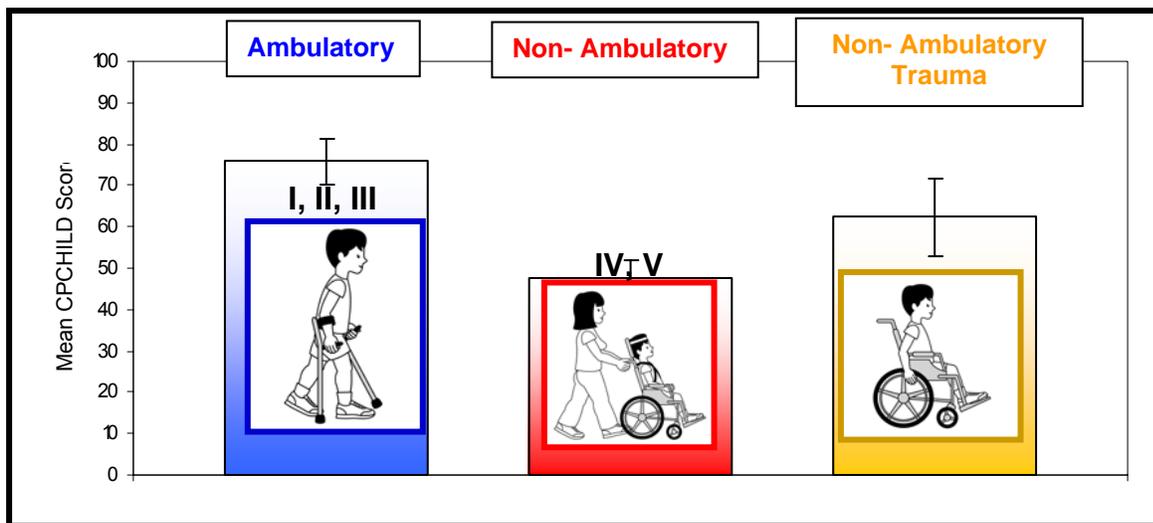
For children with cerebral palsy, known-groups validity can be evaluated based on the Gross Motor Function Classification System (GMFCS), which categorizes children into five different levels of functional ability <sup>50</sup>. The CPCHILD<sup>®</sup> was administered to the parents/caregivers of a wide range of children with cerebral palsy, who were categorized according to their GMFCS level. We hypothesized that children with higher GMFCS levels (poorer function) would have lower (worse) CPCHILD<sup>®</sup> scores. We also hypothesized that CPCHILD<sup>®</sup> scores for non-ambulatory patients (GMFCS IV & V) would be significantly higher than for ambulatory patients (GMFCS I, II, & III). We also anticipated that the CPCHILD<sup>®</sup> scores among ambulatory patients may not be significantly different across GMFCS levels I, II, and III. Analysis of variance (ANOVA) was used to compare the mean CPCHILD<sup>®</sup> scores across the five GMFCS levels. As hypothesized, the CPCHILD<sup>®</sup> was able to distinguish the non-ambulatory from the ambulatory children and within the non-ambulatory group mean CPCHILD<sup>®</sup> scores were significantly higher for the GMFCS level IV than for the GMFCS level V children. (See Figure 2.)



**Figure 2. Mean CPCHILD<sup>®</sup> scores across GMFCS levels.** Mean CPCHILD<sup>®</sup> scores decreased with increasing GMFCS levels ( $p < 0.001$ ).

As a further test, a convenience sample of children with significant lower extremity fractures were recruited and matched by age with the children with cerebral palsy. These children were temporarily non-ambulatory and dependent on their parents or caregivers for many of their daily needs. We hypothesized that the CPCHILD<sup>®</sup> scores obtained from parents of children with severe lower extremity fractures (non-ambulatory trauma group) would be higher than those from caregivers of children with severe non-ambulatory CP, but lower than those from children with ambulatory CP. Mean scores were compared using analysis of variance (ANOVA). The mean (SD) CPCHILD<sup>®</sup> total scores were 47.8 (12.6) for

the non-ambulatory CP group compared with 75.9 (14.7) for the ambulatory CP group and 62.4 (19.8) for temporarily non-ambulatory group due to trauma. Mean scores for caregivers in the non-ambulatory CP group were significantly worse than those in the ambulatory CP (adjusted  $p < 0.001$ ) and non-ambulatory trauma (adjusted  $p = 0.011$ ) groups. (See Figure 3).



	Ambulatory CP	Non-ambulatory CP	Trauma
mean	75.9	47.8	62.4
SD	14.7	12.6	19.8
n	29	38	17

**Figure 3.** Mean CPCHILD<sup>®</sup> scores comparing ambulatory, non-ambulatory children with CP and non-ambulatory children due to trauma. ( $p < 0.001$ ).

### 3.4.2 Convergent Validity

Convergent validity, another type of construct validity, is shown when the scales of a measure correlate as expected with the related scales of another measure.

In our first test of convergent validity, the Child Health Questionnaire (CHQ) was administered to caregivers in conjunction with the CPCHILD<sup>®</sup>. The self-administered 28-item parent report version (PF-28) of the CHQ was used, and both the total score and 6 domain scores of the CPCHILD<sup>®</sup> were compared with the Physical and Psychosocial summary scores of the CHQ using Spearman’s rank correlation coefficient. In addition, the total scores of the CPCHILD<sup>®</sup> measures and the 6 domain scores were compared with the Caregiver assistance scales of the Pediatric Evaluation of Disability Inventory.

Primary caregivers of 38 children with severe (non-ambulatory) CP completed the CHQ-PF28 and the PEDI. The CHQ-PF28 responses of caregivers of 13 children (34% of respondents; 2 GMFCS level IV and 11 GMFCS level V) could not be analyzed due to too many missing data. These caregivers reported that the questions were “too high level” or “not applicable” to their child, and chose to leave these items unanswered. Of the remaining 25 respondents the CPCHILD<sup>®</sup> total scores demonstrated positive Spearman’s rank correlation coefficients with the physical health ( $r=0.63$ ) and psychosocial health ( $r=0.48$ ) summary scores of the CHQ-PF28. See Table 2.

**Table 2: Spearman’s rank correlation coefficients between CPCHILD<sup>®</sup> scores and CHQ summary scores**

<b>CPCHILD<sup>®</sup> Scores</b>	<b>CHQ Summary Scores (n=25)</b>	
	<b>Physical summary</b>	<b>Psychosocial summary</b>
1. Personal care	<b>0.401<sup>a</sup></b>	<b>0.425<sup>a</sup></b>
2. Positioning, transferring and mobility	<b>0.538<sup>b</sup></b>	0.218
3. Comfort and emotions	0.295	0.307
4. Communication and social interaction	<b>0.671<sup>b</sup></b>	<b>0.403<sup>a</sup></b>
5. Health	0.231	0.321
6. Overall quality of life	0.387	0.376
Total Score	<b>0.628<sup>b</sup></b>	<b>0.480<sup>a</sup></b>

<sup>a</sup>Correlation is significant at 0.05 (two-tailed)

<sup>b</sup>Correlation is significant at 0.01 (two-tailed)

All 38 caregivers of children with severe (non-ambulatory) CP were included in the analysis of the PEDI. Spearman’s rank correlation coefficients were positive and significant between CPCHILD<sup>®</sup> total scores and the self-care (r=0.61), mobility (r=0.62), and social function (r=0.52) domains of the caregiver assistance scales of the PEDI. Correlations were highest between the “mobility” domain of the PEDI and the “positioning, transferring and mobility” domain of the CPCHILD<sup>®</sup> (r=0.61); and the “social function” function domain of the PEDI and the “communication and social interaction” domain of the CPCHILD<sup>®</sup> (r=0.71). See Table 3.

**Table 3. Spearman’s rank correlation coefficients between CPCHILD<sup>®</sup> scores and PEDI caregiver assistance scales**

<b>CPCHILD<sup>®</sup> Scores</b>	<b>PEDI Caregiver Assistance Scales (n=38)</b>		
	<b>Self-care</b>	<b>Mobility</b>	<b>Social Function</b>
1. Personal care	<b>0.461<sup>b</sup></b>	<b>0.524<sup>b</sup></b>	0.293
2. Positioning, transferring and mobility	0.510 <sup>b</sup>	<b>0.607<sup>b</sup></b>	0.304
3. Comfort and emotions	0.358 <sup>a</sup>	0.360 <sup>a</sup>	0.179
4. Communication and social interaction	0.434 <sup>b</sup>	0.449 <sup>b</sup>	<b>0.712<sup>b</sup></b>
5. Health	0.443 <sup>b</sup>	0.505 <sup>b</sup>	0.513 <sup>b</sup>
6. Overall quality of life	0.312	0.395 <sup>a</sup>	0.360 <sup>a</sup>
Total Score	<b>0.607<sup>b</sup></b>	<b>0.619<sup>b</sup></b>	<b>0.518<sup>b</sup></b>

<sup>a</sup>Correlation is significant at 0.05 (two-tailed)

<sup>b</sup>Correlation is significant at 0.01 (two-tailed)

Further testing is near completion involving comparisons of the CPCHILD<sup>®</sup> with the Pediatric Quality of Life Inventory (PedsQL) Generic Core Scales and PedsQL CP Module; and with the Health Utilities Index Mark 2 and Mark 3 (HUI2 and HUI3). The CPCHILD<sup>®</sup> will also be compared with the CP QOL-Child <sup>52</sup>.

### **3.4.3 Sensitivity (Discriminatory Ability)**

Sensitivity is the ability to detect clinically relevant differences between groups or subgroups of patients. A discriminative instrument is sufficiently sensitive to detect small differences between patients. We compared the sensitivity of the CPCHILD<sup>®</sup>, the CHQ, and the PEDI, and are in the process of comparing the CPCHILD<sup>®</sup>, the HUI2 and HUI3, and the PedsQL. We hypothesized that the CPCHILD<sup>®</sup> would be more sensitive than the CHQ, PEDI, PedsQL and HUI in detecting differences between patients, since the other measures are more likely to suffer from floor effects in this population.

When comparing the sensitivity of the CPCHILD<sup>®</sup> with the CHQ, the caregiver assistance scales of the PEDI, we reported the “relative efficiency” (RE), which is the ratio of the squares of the t-statistics<sup>51</sup>, and found that the CPCHILD<sup>®</sup> was more sensitive than the other two measures, with the exception of the PEDI self-care domain. The same is being done for the PedsQL and HUI. In addition, the dispersion (or spread) of the CPCHILD<sup>®</sup> scores is being compared with the dispersion of the CHQ, PEDI, PedsQL, and HUI scores for the same patients.

### **3.5 Responsiveness or Sensitivity to Change**

The CPCHILD<sup>®</sup> is intended to be an evaluative instrument to measure change over time or following interventions. In order to assess the property of responsiveness or sensitivity to change, prospective longitudinal cohort studies are currently underway. Responsiveness will be measured using the standardized response mean (SRM), which is the ratio of the mean change to the standard deviation of that change; and the effect size (ES), which is the ratio of the mean change to the standard deviation of the initial measurements<sup>51</sup>.

NOTE: The CPCHILD<sup>®</sup> cannot be recommended for use as an outcome measure until this work has been completed and it has been shown to be responsive.

## 4. Does the CPCHILD Measure HRQL?

The Caregiver Priorities & Child Health Index of Life with Disabilities (CPCHILD<sup>®</sup>) questionnaire has been demonstrated to have excellent reliability and appears to be a valid measure of caregivers' perspectives on the health status, functional limitations, and well-being of children with severe non-ambulatory cerebral palsy. As we cannot access the perspective of the child, one is still left uncertain as to whether the CPCHILD<sup>®</sup> is a reasonable proxy measure of the child's HRQL. Caregivers' ratings of the importance of the items of the questionnaire to their children's quality of life, and convergent validity established between sectional/domain scores of the CPCHILD<sup>®</sup> and the corresponding domains of other generic or condition specific measures of HRQL will provide some assurance that the CPCHILD<sup>®</sup> is a valid measure of HRQL for this population.

Caregivers of 38 children (24 males, 14 females; mean age 9 years, 6 months; range 4y;7m to 17y;10m) with severe CP completed the CPCHILD<sup>®</sup> and the Child Health Questionnaire (CHQ-PF28). Caregivers rated the importance of each item's contribution to their child's quality of life (QOL) on a 6-point ordinal scale from 0 (least important) to 5 (most important). The mean importance rating for all CPCHILD<sup>®</sup> items was 3.63 (SD = 0.54, Range = 2.54 to 4.74). "Comfort & Emotions" and "Communication & Social Interaction" were rated more important domains than "Positioning, Transferring & Mobility" and "Personal Care" ( $p < 0.01$ ). The global QOL item in Section 6 of the CPCHILD<sup>®</sup> correlated strongly with the total CPCHILD<sup>®</sup> score ( $r = 0.64$ ). The total CPCHILD<sup>®</sup> score correlated with the summary scores of the CHQ-PF28 ( $r = 0.48$  to  $0.63$ ). The items in the CPCHILD<sup>®</sup> are relevant to caregivers and are considered important contributors to the HRQL of children with severe cerebral palsy. These data combined with convergent

validity of the CPCHILD<sup>®</sup> with the CHQ, suggests that the CPCHILD<sup>®</sup> is indeed a valid measure of the caregiver's perception of the HRQL of children with severe CP <sup>53</sup>.

# Chapter 5: Using the CPCHILD<sup>®</sup>

## 5.1 Administration of the CPCHILD<sup>®</sup>

### **What does the CPCHILD<sup>®</sup> measure?**

The CPCHILD<sup>®</sup> Questionnaire measures the caregiver's perspective about the child's health status, comfort, wellbeing, functional abilities and ease of caregiving. It is a useful proxy measure of health related quality of life of children with severe disabilities.

### **Who is the CPCHILD<sup>®</sup> designed for?**

The CPCHILD<sup>®</sup> Questionnaire was designed to measure caregivers' perceptions of the HRQL of children with severe disability. The CPCHILD<sup>®</sup> has been validated for use for caregivers of children with severe developmental disabilities such as those with non-ambulatory cerebral palsy and traumatic brain injury, who would be categorized in level IV or V of the Gross Motor Function Classification System <sup>50</sup>. The CPCHILD<sup>®</sup> is not intended for children with ambulatory CP. The CPCHILD<sup>®</sup> is currently being assessed in children with other severe disabilities that affect a child's mobility and ability to perform activities of daily living, such as acquired brain injury and progressive neuromuscular disorders.

### **How is the CPCHILD<sup>®</sup> administered?**

The CPCHILD<sup>®</sup> Questionnaire is intended to be self-administered. Parents or caregivers can either complete the questionnaire at home and return it by mail or complete it at a clinical visit. It can also be completed by the caregiver or parent on behalf of the child if he or she is able to understand it and communicate his or her responses to the parent. When a child is able to respond both the child's and the parent/caregiver's report/s are desirable.

### **How long does the CPCHILD<sup>®</sup> take to complete?**

The questionnaire takes between 20 and 30 minutes to complete.

## 5.2 Scoring the CPCHILD®

### Basic Scoring Procedure

Scores for each domain and for the total survey are standardized and range from 0 (worse) to 100 (best). There are 4 stages to scoring the CPCHILD®; the scoring procedure is outlined in the following table.

Stage	Description	Instructions
1	Raw Score for Item	Sum of base rating and modifier rating
2	Standardized Score for Item	Raw score for item divided by total possible score for item, times 100
3	Standardized Score for Domain	Average of standardized scores for all items in the domain
4	Standardized Score for Total Survey	Average of standardized scores for all items in the survey

### Important Notes

1. *The highest possible score for Comfort & Emotions items is 7, not 8 as it appears.* It is assumed that if a caregiver selects 5 (none of the time), there can be no associated level of intensity, and they should select 3 in the modifier column. For the purposes of the scoring algorithm, however, a rating of 3 for intensity carries a value of 2. The 3 is provided solely to prevent confusion for respondents selecting 5 (none of the time).

2. Item 34 is scored from 0 to 5:

Response	Score
Admitted > 7 days	0
Admitted < 7 days	1
3 or more times	2
Twice	3
Once	4
None	5

3. Item 36 is scored in reverse order:

# of Medications	Score
0	5
1	4
2	3
3	2
4	1
≥ 5	0

4. *Do not average the standardized scores for the domains to get the standardized score for total survey.* The number of items in each domain is not consistent, and this would cause false section weighting.

### **Missing Values**

If any value is missing from an item (either the base rating or the modifier), that item is removed from the analysis completely. It is not treated as zero. The only exception is when a score of 5 has been selected for frequency in the Comfort & Emotions section and the modifier has been left blank. In this scenario, a total score of 7 for that item can be assumed, provided that the respondent filled out modifiers for other items in the questionnaire (see above).

### **Added Items**

Additional items provided by caregivers at the end of each section are currently not incorporated into the scoring, though they may provide helpful information.

### 5.3 Interpretation of Scores

The data on the following page is provided to give users a general idea of the scores (Means and Standard deviations) that can be expected for children with cerebral palsy.

Ambulatory CP is defined as GMFCS Levels I, II, and III, while severe (non-ambulatory) CP is defined as GMFCS Level IV and V.

#### **CPCHILD<sup>®</sup> Total Scores by Ambulatory Status**

<b>Ambulatory CP</b> (n = 29)	<b>Non-ambulatory CP</b> (n = 38)
75.9 (14.7)	47.8 (12.6)

#### **CPCHILD<sup>®</sup> Total Scores by GMFCS Level**

<b>GMFCS Level</b> (n = 67)				
<b>I</b>	<b>II</b>	<b>III</b>	<b>IV</b>	<b>V</b>
84.4 (12.8)	79.2 (2.9)	67.9 (13.8)	56.3 (8.1)	44.4 (12.6)

#### **CPCHILD<sup>®</sup> Total Scores for GMFCS Levels IV and V**

##### **Sample A:**

<b>GMFCS IV</b> (n = 11)	<b>GMFCS V</b> (n = 27)
56.3 (8.1)	44.4 (12.6)

##### **Sample B:**

<b>GMFCS IV</b> (n = 18)	<b>GMFCS V</b> (n = 35)
61.9 (11.5)	44.4 (12.5)

#### **CPCHILD<sup>®</sup> Domain Scores for GMFCS Levels IV and V**

	<b>GMFCS IV</b> (n = 18)	<b>GMFCS V</b> (n = 35)
<b>Personal Care/ADLs</b>	41.6 (13.3)	31.0 (15.2)
<b>Positioning, Transferring &amp; Mobility</b>	45.5 (11.3)	28.4 (14.2)
<b>Comfort &amp; Emotions</b>	81.0 (14.5)	67.9 (22.6)
<b>Communication &amp; Social Interaction</b>	74.9 (22.2)	43.4 (23.7)
<b>Health</b>	83.7 (15.0)	57.0 (16.9)
<b>Quality of Life</b>	72.5 (21.8)	55.4 (24.8)

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## Glossary

**Acquired brain injury:** an injury to the brain occurring after the age of 1 year and resulting in physical, functional, mental, and/or emotional deterioration.

**Cerebral palsy:** a group of permanent disorders of movement and posture resulting from a non-progressive injury to the developing brain, and often accompanied by disturbances of sensation, perception, cognition, communication, and behaviour, as well as seizures and secondary musculoskeletal problems.

**Disease-specific measure:** a questionnaire designed for patients with a particular illness or condition; sometimes preferred because it highlights the concerns of a unique population.

**Functional status:** the degree to which an individual is able to perform socially allocated roles with freedom from physical or mental limitations.

**Gross Motor Function Classification System:** a method of stratifying children with cerebral palsy according to the severity of the condition, based on motor ability and postural control of children. Levels range from I (best) to V (worst).

**Health profile measure:** a descriptive questionnaire that assesses different aspects of well-being across multiple health domains

**Generic measure:** a questionnaire intended for patients with any condition; sometimes preferred because it allows comparison across conditions.

**Health related quality of life:** focuses on the health-related components judged to be associated with life satisfaction, such as self-care, mobility, and communication.

**Health status:** medical and functional well-being. This is sometimes reported in terms of impact of disability.

**Hypertonia:** Abnormal increase in skeletal or smooth muscle tone.

**Preference-weighted measure:** a questionnaire that is based on population norms and designed to provide a single unitary expression of health. This expression is stated as a global index or utility score.

**Quality of life:** refers to the notion of holistic well-being, including the perceived state of physical health, satisfaction with where one lives and spends time, social fulfillment and integration, and access to education and work. Many variations of this definition exist.

**Spasticity:** involuntary muscle tightness and stiffness; this is a common problem for children with cerebral palsy and can cause a variety of difficulties in daily living.

**Utility theory:** an econometric understanding of health related quality of life that can be used to calculate quality-adjusted life years. Utility scores are typically measured on a 0.0 to 1.0 scale, with 0.0 representing death and 1.0 perfect health.

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