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Assessing the reliability and validity of a health-related quality of life instrument, CPADULT, in a Dutch sample of adults with severe disabilities who are non-ambulatory

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Abstract

Background: A measure to provide insight regarding health-related quality of life of adults with severe motor and intellectual disabilities was lacking. For this reason, the CPADULT was developed. This measure includes domains relating to an individual's physical, mental, and social functioning. The purpose of this study was to assess the psychometric characteristics of the CPADULT.

Method: Caregivers ($n = 47$; 77% female, 23% male) of individuals with severe disabilities who are non-ambulatory completed the questionnaire. Internal consistency, test-retest reliability and construct validity were analysed.

Results: Internal consistency was adequate with Cronbach's alpha values from 0.75 to 0.95. Test-retest reliability was good, as intraclass correlation coefficient of the total score was 0.84 (domains: 0.61–0.89). Construct validity was confirmed with significant differences between subgroups of motor or intellectual abilities.

Conclusion: The CPADULT has sufficient reliability and validity as a proxy measure of health-related quality of life for adults with severe disabilities who are non-ambulatory.

KEYWORDS

cerebral palsy, health-related quality of life, severe disabilities, validation

1 | INTRODUCTION

Adults with severe disabilities who are non-ambulatory constitute a heterogeneous group. Severe motor and intellectual disabilities can be caused by genetic, developmental, intrauterine, or neonatal disorders, (e.g., cerebral palsy), or they can be of unclear origin (Nakken & Vlaskamp, 2007). People with chronic severe motor disabilities include those with the inability to walk and/or sit without support which make these people non-ambulatory (Level V and Level IV), as

derived from the Gross Motor Function Classification used for individuals with CP (Canchild, 2015). Additional intellectual disabilities (ID) are classified according to the levels of intensity of support needs and level of adaptive behaviour (Schalock et al., 2021). Most people with severe disabilities who are non-ambulatory can be classified in severe or profound ID (Maes et al., 2021). These people require daily lifetime support across all environments. However, mild and moderate ID, with some independence in self-care, can also occur (Schalock et al., 2021).

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Adults with severe disabilities who are non-ambulatory often encounter severe secondary disabilities (e.g., sensory impairments) and health problems (e.g., epilepsy and pulmonary problems) (Hollung et al., 2020; van Timmeren et al., 2016). In addition to existing chronic disabilities and problems, adulthood can induce new physical limitations, such as musculoskeletal pain, fatigue, deterioration of functional skills and multimorbidity (Jahnsen et al., 2004). For example, one systematic review studying pain prevalence in adults with cerebral palsy (CP) estimated an overall prevalence of various types of pain in adults with CP at 70%. For the adults who are non-ambulatory the pain may be explained by musculoskeletal problems such as scoliosis, joint contractures and/or hip subluxation, prolonged sitting with an inability to change position and physical inactivity (van der Slot et al., 2021).

Another study concluded that there is a high prevalence of multimorbidity—at least two chronic conditions—amongst middle-aged adults with CP. These estimates are higher for individuals who are non-ambulatory (Cremer et al., 2017).

Each individual has a unique combination of abilities and limitations that affect their functioning. Advancing the inclusion of people with disabilities (Centers for Disease Control and Prevention, 2020) calls for a person-centred approach. A person-centred approach is characterised by seeing people as unique individuals with valuable gifts and contributions, and by focusing on what matters to the people receiving support and their families (NSW Health, 2020). Support should focus on their abilities, needs, and unique circumstances. Adults with severe disabilities who are non-ambulatory depend on substantial and lifelong support from their caregivers or health care professionals (Narayanan et al., 2006). This high level of dependency is reinforced by the fact that many are unable to communicate verbally when and how support should be given (Petry et al., 2009). It is therefore challenging to determine the best support for adults with severe disabilities who are non-ambulatory. It is imperative to find ways to identify what is important to improve quality of life for each individual.

When considered within the context of health and disease, quality of life it is commonly referred to as 'health-related quality of life' (HRQoL): the perceived physical and mental health of an individual or group over time (Healthy people, 2020). To assess unmet needs and to evaluate intervention outcomes, HRQoL is generally regarded as a valid indicator, especially for people with chronic illness or disabilities (Healthy people, 2020). This multi-dimensional concept includes domains related to physical, mental, emotional, and social functioning (de Vet et al., 2011). It is a subset of the general domains of the broader concept of quality of life (QoL), which also includes features of life that are not related to health, such as material well-being (Gómez et al., 2015; Schalock et al., 2005).

In general, HRQoL is assessed according to patient-reported outcome measures. For people with severe disabilities who are non-ambulatory, the self-evaluation of HRQoL is often highly challenging, if not impossible, due to the limited intellectual, communicative, or reflective capabilities of these individuals (Maes et al., 2021). One commonly used method involves relying on the perspective of the caregivers, with parents or direct support professionals (DSPs)

completing a proxy questionnaire (Alves-Nogueira et al., 2020). In this way, the views of those who are most familiar with the person with severe disabilities are included in the assessment (Maes et al., 2021). If possible, the person with severe disabilities can be involved in completing the questionnaire.

Existing measures of HRQoL are not suitable, as they assume higher levels of motor and/or intellectual function (Zalmstra et al., 2021). Given the lack of any HRQoL measure specifically customised for adults with severe disabilities who are non-ambulatory, there is need for a proxy HRQoL instrument that takes their severe motor disabilities and combined intellectual disabilities into account. In a previous study, we developed an instrument to assess the HRQoL specifically for this target group (Zalmstra et al., 2021). Known as the CPADULT, the instrument is based on the Dutch version of the widely used Caregiver Priorities and Child Health Index of Life with Disabilities (CPCHILD) questionnaire, a measure of HRQoL for children with severe disabilities who are non-ambulatory (Sickkids, 2021; Zalmstra et al., 2015). The CPADULT was developed by using a qualitative design involving a three-step adaptation process including a 'sensitivity assessment': (1) Focus groups of primary caregivers concerning the adaptation of the items and sensibility topics (comprehensibility, clarity of instructions, suitability of the output scales and ease of usage). (2) E-survey amongst health-care professionals (healthcare psychologists, physicians, researchers, and therapists who are professionally involved with adults with severe motor and intellectual disabilities who are non-ambulatory) concerning content validity. (3) One-to-one interviews with primary caregivers discussing the items and sensibility topics (Zalmstra et al., 2021). Throughout the process, 72% of the items of the CPCHILD-DV remained unchanged, three items were added (e.g., 'menstruation pain' and 'experiencing sexuality'), and one element ('shaving') was added to an existing item concerning personal care. For 10 items the wording was adapted to the target group of adults (e.g., 'to play' changed to 'amuse oneself'). Minor changes were made to the instruction (e.g., 'child' changed to 'client' in the version for DSPs) and response scales (e.g., adding a box 'not applicable') with regard to the sensibility topics.

Before a newly developed instrument can be used in clinical or research settings, its psychometric characteristics must be assessed (Terwee et al., 2007). The aim of this study was to assess the reliability (internal consistency, test-retest reliability, measurement error), validity (content and construct validity), and interpretability of the CPADULT in a sample of Dutch adults with severe disabilities who are non-ambulatory.

2 | METHOD

2.1 | Participants

Participants included primary caregivers (parents or DSPs) responsible for the daily support of an adult person with severe motor disabilities and intellectual disabilities who is non-ambulatory. To be eligible to participate caregivers were required to have sufficient understanding

of the written Dutch language. The inclusion criteria for the adult person (18 years of age or older) included chronic disabilities caused by cerebral palsy, genetic disorders, or other conditions consisting of severe motor disabilities (resulting in becoming non-ambulatory) and intellectual disabilities (from mild to profound). According to the quality criteria for measurement properties of health-status questionnaires, a sample size of 50 persons is assumed sufficient (Terwee et al., 2007). Because of recruitment challenges (including the COVID-19 pandemic), the sample for this study comprised only 47 parents (86% mothers) and DSPs. Demographic characteristics of both the caregiver participants and their children/clients are presented in Table 1. Of the caregivers, 36 were women (77%), with 27 parents (57%) and 14 DSPs (30%). Of the clients, 30 were men (64%) with a mean age of 29 years. With regard to living situations, 28 of the clients were living in residential care facilities or small scale group homes (60%) and 17 were living with their parents (36%).

2.2 | Measure

The CPADULT is a proxy measure of HRQoL specifically customised for adults with severe disabilities who are non-ambulatory. It consists of 42 items across six domains: (1) Activities of daily life/personal care (9 items); (2) Positioning, transferring and mobility (8 items); (3) Comfort and emotions (10 items); (4) Communication and social interaction (10 items); (5) Health (4 items); and (6) Overall quality of life (1 item). Each item is rated on an ordinal scale, reflecting on the past 2 weeks. In the first two domains, the items concern the performance of skills (e.g., 'putting on a shirt' or 'getting in and out of bed'). The degree of difficulty accomplishing each activity is rated on a 7-point ordinal scale, ranging from 'almost impossible' (0) to 'no problem at all' (6). Additionally, the level of assistance is rated on a 4-point ordinal scale, rating from 'total assistance' (0) to 'independent' (3). For the third domain, the frequency of discomfort or pain during activities is rated on a 6-point ordinal scale and the level of discomfort or pain is rated on a 4-point ordinal scale. The items of the fourth domain concern communication and social interaction (e.g., 'to understand you'). The degree of difficulty performing these activities is rated on a 7-point ordinal scale. The following is an example of items in the domain of Health (Domain 5): 'the frequency of doctor's consult in the past 2 weeks'. These items are rated on a 6-point ordinal scale. Domain 6 is not a subscale but consists of a single global item about the construct ('How would you rate your child's/client's overall quality of life?'). Standardised scores from 0 (worst) to 100 (best) are calculated for each of the six domains, as well as for the total survey.

In addition to the 42 items outlined above, the questions included demographic questions (included in Table 1). Regarding the adults who are non-ambulatory, caregivers provided information about age, gender, aetiology (cerebral palsy, genetic disorder, other, unknown), living situation (residential care facility, small-scale group home, with parents, unknown), day-care situation (no day care, external day care—outside residential facility—, internal day care—within residential facility—, unknown), level of motor function, and intellectual level. For

TABLE 1 Participant demographics.

Clients (n = 47)		Frequency (percentage)
Gender	Men: 30 Women: 17	
Age (years)	Mean: 29 y, 0 mo (SD: 8 y, 10 mo) Min: 19 y, 1 mo; Max: 53 y, 9 mo	
GMFCS level	GMFCS IV GMFCS V	8 (17%) 39 (83%)
Cognitive level	I Mild intellectual disability (IQ 50–69) II Moderate intellectual disability (IQ 35–49) III Severe intellectual disability (IQ 20–34) IV Profound intellectual disability (developmental perspective <24 month) Unknown	2 (4%) 2 (4%) 7 (15%) 35 (75%) 1 (2%)
Aetiology	Cerebral Palsy Genetic disorder Other Unknown	18 (38%) 8 (17%) 14 (30%) 7 (15%)
Living situation	Residential home Small scale home With parents Unknown	14 (30%) 14 (30%) 17 (36%) 2 (4%)
Day support	No day care External day care Internal day care Unknown	3 (6%) 31 (66%) 12 (26%) 1 (2%)
Total score CPADULT (0–100)	Mean: 47.1 (SD: 11.5) Min: 21.1 Max: 67.5	
Caregiver (n = 47)		Frequency (percentage)
Gender	Men: 11 Women: 36	
Age	Mean: 55 y, 6 mo (SD: 13 y, 1 mo) Min: 26 y 4 mo; Max: 79 y, 0 mo	
Relation	Parent Guardian Direct support professional Other Missing	27 (57%) 1 (2%) 14 (30%) 3 (7%) 2(4%)
Educational level ^a	Tertiary Secondary Primary Missing	25 (53%) 19 (40%) 2 (4%) 1 (2%)

Note: y, years; mo, months; min, minimum; max, maximum; SD, standard deviation; GMFCS, Gross Motor Function Classification System; IQ, intelligence quotient.

^aUnesco (2012).

the last two questions, respondents could choose from several categories based on a description of abilities. Adults who are non-ambulatory were categorised in two different motor-function levels (Level V and Level IV), as derived from the Gross Motor Function Classification (GMFCS) used for individuals with CP (Canchild, 2015). Level V is described as follows: restrictions in the ability to maintain head and neck position against gravity; is confined to a wheelchair; needs many adaptive equipment and physical support. Level IV is described as follows: can sit independently with support; can propel oneself with adaptive equipment with limitations (Canchild, 2015).

Intellectual functioning is measured according to the following four descriptions, as derived from the classification of intellectual disabilities (Schalock et al., 2021). For caregivers who are more familiar with IQ levels, IQ ranges were included in the descriptions as well (Resing & Jan, 2002). Mild—can learn basic self-care and home activities, support is basic or episodic (IQ 50–69); Moderate—some independence in self-care, home activities with consistent supervision (IQ 35–49); Severe—requires daily lifetime support, some may acquire basic self-care with intensive training (IQ 20–34); Profound—high intensity support needed across all environments (developmental perspective <24 month).

Caregivers provided information about themselves with regard to the following characteristics: age, gender, relationship to the person with severe disabilities, and highest educational level completed (primary, secondary, or tertiary; Unesco, 2012).

2.3 | Procedure

The local ethics committee provided a waiver for formal approval of this study, as it was not a clinical research project involving human subjects, as referred to in the Medical Research Involving Human Subjects Act (file number METc 2018/044).

Between December 2019 and May 2021, flyers recruiting eligible caregivers were distributed amongst physiatrists and physicians specialised in treating people with intellectual disabilities as well as through residential facilities for adults with intellectual disabilities in the Netherlands. The flyers were also posted on social media. Caregivers were invited to participate and to respond by email. The aim was to gather information from participants with varying motor and intellectual functioning to allow for comparisons between subgroups. Based on previous studies (Narayanan et al., 2006; Zalmstra et al., 2015), however, we expected that the group of people with mild and moderate intellectual disabilities would be small.

In all, 47 people responded to the recruitment flyer, and all met the inclusion criteria. Participants signed informed consent prior to entering the study. An information letter was sent to each of them at home (by email or by post), along with the questionnaire. Two weeks later, the questionnaire was sent for the second time, in order to assess the test–retest reliability. The second questionnaire was completed by 32 caregivers (seven of whom were DSPs), despite sending reminder emails to increase the number of responses.

2.4 | Analyses

Data were analysed using IBM SPSS version 25. The significance level was set at $p < .05$ (two tailed). The CPADULT total scores were tested for normal distribution and skewness to determine whether parametric tests could be used. The CONsensus-based Standards for the selection of health Measurement INstruments (COSMIN) aims to improve the selection of outcome-measurement instruments in both research and clinical practise by developing methodology and practical tools (Cosmin, 2011). The COSMIN taxonomy of measurement properties defines three domains of measurement properties for outcome measurement instruments: reliability, validity, and responsiveness. Interpretability is also regarded as an important characteristic for a measurement instrument (Mokkink et al., 2010). The domains of reliability and validity are addressed in this study, along with interpretability, as described in detail in the following paragraphs. Responsiveness is not addressed in this study, as it would require a longitudinal design.

2.4.1 | Assessment of reliability

The domain of reliability refers to ‘the degree to which the measurement is free from measurement error’ (Mokkink et al., 2010). It consists of the following measurement properties: internal consistency, reliability, and measurement error.

Internal consistency is defined as ‘the degree of the interrelatedness among the items’ (Mokkink et al., 2010). The internal consistency of the total score and five domain scores of the CPADULT was calculated according to Cronbach's alpha, which uses item-to-item correlations to assess the homogeneity of multi-item scales. Alpha coefficients between 0.70 and 0.95 were considered adequate for all domains (Terwee et al., 2007).

Reliability is defined as ‘the proportion of the total variance in the measurements which is due to true differences between patients’ (Mokkink et al., 2010). In this study test–retest reliability—reliability of a test measured over time—was assessed by giving the same measurement twice for the same people at different times. The two measurements were taken at an interval of 2 weeks, as commonly applied to achieve a balance between the stability of the characteristics and the independence of the repeated test (de Vet et al., 2011). Test–retest reliability was estimated according to the single-measure intraclass correlation coefficient (ICC Case 1, single measure), including 95% confidence intervals (CI). An ICC value of 0.70 or more is considered acceptable (de Vet et al., 2011).

Measurement error is defined as ‘the systematic and random error of a participant's score that is not attributed to true changes in the construct to be measured’ (Mokkink et al., 2010). The method developed by Bland and Altman was used to assess the magnitude of measurement error and calculate limits of agreement (Bland & Altman, 1986). The latter also provide information about the smallest (or minimum) detectable change (SDC), which reflects change beyond measurement error.

2.4.2 | Assessment of validity

Domain of validity refers to ‘the degree to which a health-related patient-reported outcome (HR-PRO) instrument measures the construct(s) it purports to measure’ (Mokkink et al., 2010). Validity comprises the measurement properties of content validity, construct validity, and criterion validity. Content and construct validity are addressed in this study.

Content validity is defined as ‘the degree to which the content of an HR-PRO instrument is an adequate reflection of the construct to be measured’ (Mokkink et al., 2010). The content validity of the CPADULT was assessed according to the caregivers' rating of the importance of the extent to which each item contributed to the HRQoL of their children/clients on a 5-point scale ranging from 0 (unimportant) to 4 (very important). The threshold value was set at a median of 2 (fairly important) (Narayanan et al., 2006). Content validity was further assessed by calculating the correlation between the global item (domain six) about the construct (‘How would you rate your child's/client's overall quality of life?’) and all other items. Based on clinical reasoning, we expected at least a moderate correlation (between 0.40 and 0.60).

Construct validity is defined as ‘the degree to which the scores of a HR-PRO instrument are consistent with hypotheses based on the assumption that the HR-PRO instrument validly measures the construct to be measured’ (Mokkink et al., 2010). In this study, hypotheses were formulated with regard to differences between relevant subgroups. The CPADULT was administered to caregivers of adults who are non-ambulatory with two different levels of motor function (Level IV and Level V). The mean domain and total scores of individuals with Level IV were compared to those of individuals with Level V using *t*-tests for independent samples. Those with Level V (poorest function) were expected to have significantly lower (worse) CPADULT scores than those with Level IV. A second comparison of groups was related to different intellectual levels. An analysis of variance (ANOVA) was used to compare the mean domain and total scores of four groups of individuals categorised by level of intellectual disability (mild, moderate, severe, and profound). Based on previous studies (Zalmstra et al., 2015), adults with less severe intellectual disability were expected to have significantly higher CPADULT scores, especially in the domain of ‘communication and social interaction’.

2.4.3 | Assessment of interpretability

Interpretability is defined as ‘the degree to which one can assign qualitative meaning [...] to an instrument's quantitative scores or changes in scores’ (Mokkink et al., 2010). Several issues must be considered in his regard: the distribution of the scores, floor and ceiling effects, and the minimal clinically important difference (MCID; Mokkink et al., 2010).

The distribution (mean and standard deviation) of the scores is important for a proper interpretation of the scores. The occurrence of floor and ceiling effects was tested by assessing whether more than

15% of the people achieved the lowest or highest possible scores. One measure used to determine clinical relevance is the MCID, the smallest change in score that is considered important to the individual (Terwee et al., 2003). In most circumstances, the threshold of discrimination for changes in HRQoL instruments for chronic diseases appears to be approximately one half the SD of the mean score (Norman et al., 2003).

3 | RESULTS

3.1 | Reliability

The internal consistency of five domain scores and the total score was greater than 0.70, which is considered adequate (Table 2). The ICC score for the test-retest reliability of the CPADULT total score ($n = 47$) was 0.84 which is considered good. The ICC for the domains ranged from 0.61 to 0.89 for the domains (Table 2).

The Bland and Altman plot shows the mean of the scores (*x*-axis) against the difference between the scores (*Y*-axis) for each participant. As indicated by this plot, the measurement error is random and 95% of the scores (the limits of agreement, as indicated by the dotted lines) ranged between ± 11.5 points of the mean difference (Figure 1). The SDC score was therefore calculated as 11.5 points.

3.2 | Validity

3.2.1 | Content validity

Of the 41 items evaluated, 15 had a median score of 4 (very important), one item had a median score of 3.5, and 23 had a median score of 3 (fairly important). Two items scored below the threshold of 2: ‘standing during transfers’ and ‘experiencing sexuality’. Analysis of the correlation between the item of Domain 6 (overall rating of HRQoL) and all other items together resulted in an R^2 of 0.49 ($p < .05$), which is considered moderate.

3.2.2 | Construct validity

The group of individuals with Level V (poorest motor function) had a significantly lower mean total score on the CPADULT compared with the group of individuals with Level IV motor function (mean difference 10.7 points [95% CI: 2.0–19.4]; $p < .05$). This is illustrated by the box-plots in Figure 2.

At the domain level the group of individuals with Level V motor function had significantly lower scores in the domains of ‘positioning, transferring, and mobility’ (mean difference: 13.7 points [95% CI: 0.3–27.8]; $p < .05$), and ‘communication and social interaction’ (mean difference: 17.9 points [95% CI: 3.9–31.9]; $p < .05$).

Due to the low number of individuals in the groups with mild, moderate, and severe intellectual disability, we compared the group

Domains CPADULT (42 items)	Cronbach's α	Intra observer ICC	95% CI
Personal care & ADL (9)	0.95	0.73	0.50–0.87
Positioning, transferring & mobility (8)	0.91	0.71	0.47–0.85
Comfort & emotions (10)	0.92	0.69	0.43–0.84
Communication & social interaction (10)	0.87	0.89	0.75–0.93
Health (4)	0.75	0.88	0.77–0.94
Overall quality of life (1)	†	0.61	0.34–0.79
Total score	0.95	0.84	0.70–0.92

Note: †No Cronbach's α since only one item.

Abbreviations: CI: confidence interval; ICC: intraclass correlation coefficient.

TABLE 2 Cronbach's α and Intra-observer intraclass correlation coefficients.

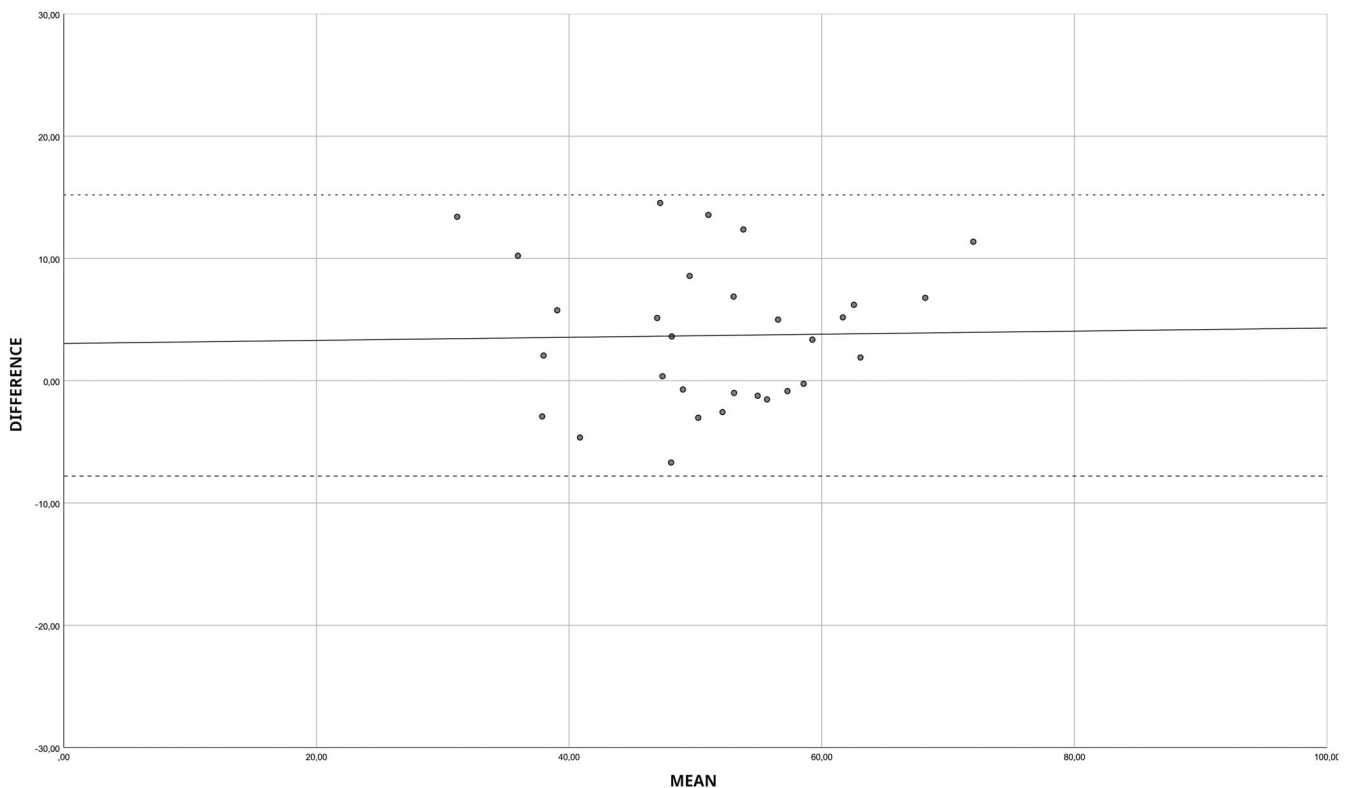


FIGURE 1 Bland and Altman plot of the test–retest reliability of the total score. The plot displays the mean of the scores (x-axis) for each participant against the difference between the scores (Y-axis).

with those with profound intellectual disability to the combined group of those with mild, moderate, and severe intellectual disability, based on *t*-tests for independent samples. Individuals with profound intellectual disability had a significantly lower mean total score (mean difference: 13.3 points [95% CI: 6.2–19.4]; $p < .05$). This is illustrated by the boxplots in Figure 3.

At the domain level, the group of individuals with profound intellectual disability had significantly lower scores in the domains of ‘activities of daily life/personal care’ (mean difference: 12.7 points [95% CI: 3.7–21.7]; $p < .05$), ‘positioning, transferring and mobility’ (mean difference: 16.0 points [95% CI: 3.9–28.1]; $p < .05$) and ‘communication and social interaction’ (mean difference: 24.3 points [95%

CI: 13.2–35.4]; $p < .05$). The results for the construct validity corresponded to our expectations (see section 2.4.2).

3.3 | Interpretability

The total score of the CPADULT was approximately normally distributed, with no skewness. The Kolmogorov–Smirnov test was non-significant, thereby confirming the normality of the data. The mean score was 47.1, with a SD of 11.5 (Table 1). The scores ranged from a minimum of 21.1 to a maximum of 67.5 within the possible range of 0–100, with no floor or ceiling effects detected. The MCID was

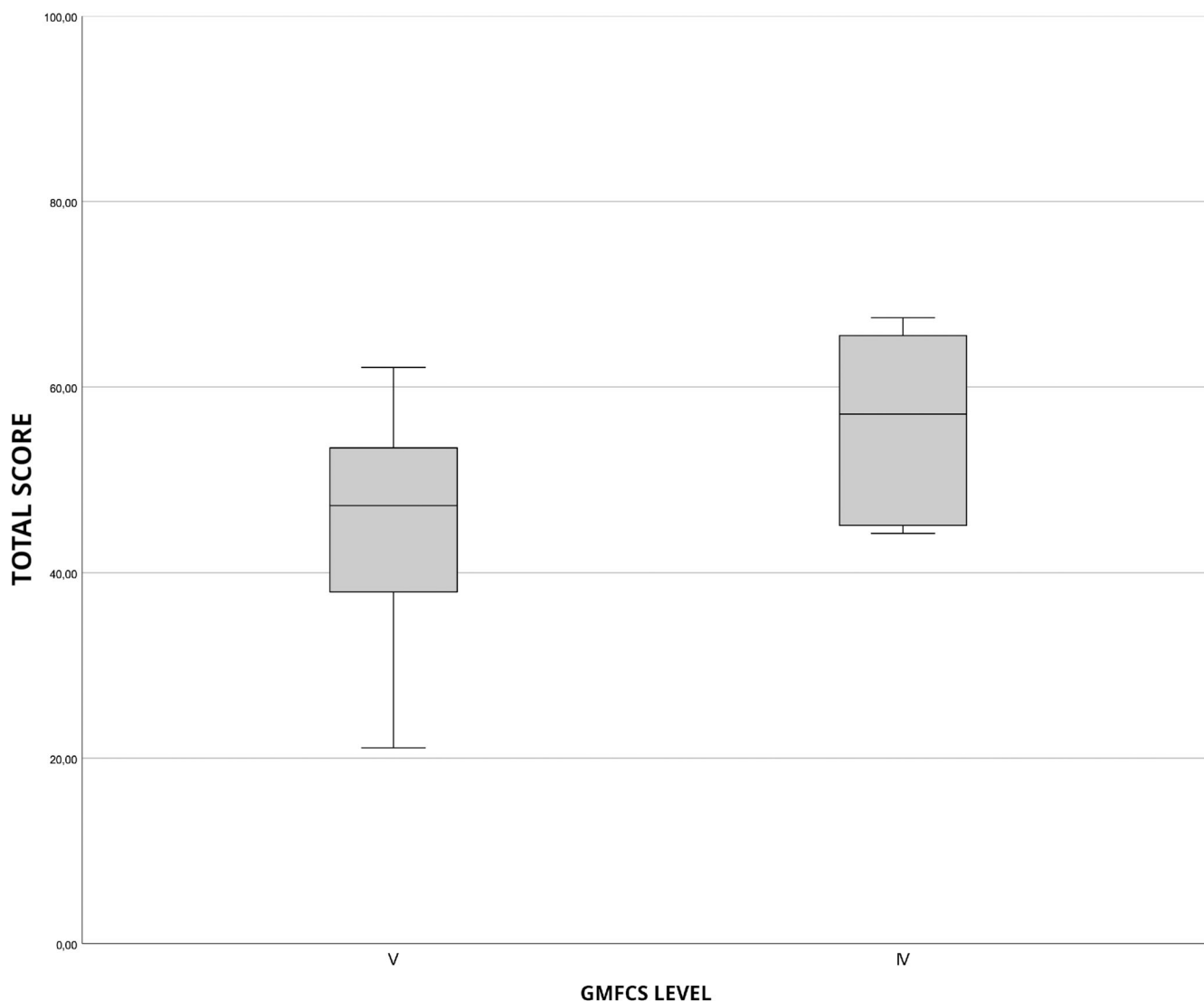


FIGURE 2 Box plots of mean CPADULT total score (y-axis) for each motor-level group (x-axis). GMFCS, Gross Motor Function Classification System.

estimated as one half the *SD* of the mean score ($11.5 \div 2 = 5.8$). All change scores between the subgroups exceeded the MCID.

4 | DISCUSSION

This study assessed the psychometric quality of a proxy HRQoL instrument, the CPADULT, which was developed specifically for adults with severe motor and intellectual disabilities who are non-ambulatory. The results indicated that the reliability and validity of the CPADULT is promising for assessing HRQoL amongst Dutch adults with severe disabilities who are non-ambulatory, based on the perspective of caregivers.

More detailed analysis of the results confirms the internal consistency of the instrument and indicates that its test-retest reliability was good. Two ICC scores were nevertheless below the recommended threshold of 0.70. The ICC score for the domain of ‘Comfort

and emotions’ was 0.69, which is just below the threshold. The ICC score for the domain of ‘Overall quality of life’ was 0.61, possibly due to the fact that this domain consisted of only one item. Change scores for single items have a greater impact on the ICC than do those based on a mean score of several items. Content validity was confirmed, as 95% of the items were rated more than sufficiently important. Only two items had a median value below the threshold of 2. One item, ‘standing during transfers’ had a score of 0 (not possible) in 60% of the participants. Another item, ‘experiencing sexuality’ had a score of 0 (not possible) in 51% of the participants. These two items were apparently appropriate for only a proportion of the participants (40% and 49%, respectively), depending on their abilities. Within these proportions, however, the median scores for these participants were 3 (fairly important) and 2 (somewhat important), respectively. Given that these two items are important for a subgroup of participants with certain abilities (Gil-Llario et al., 2018), we concluded that these items nevertheless still warrant inclusion in the questionnaire.

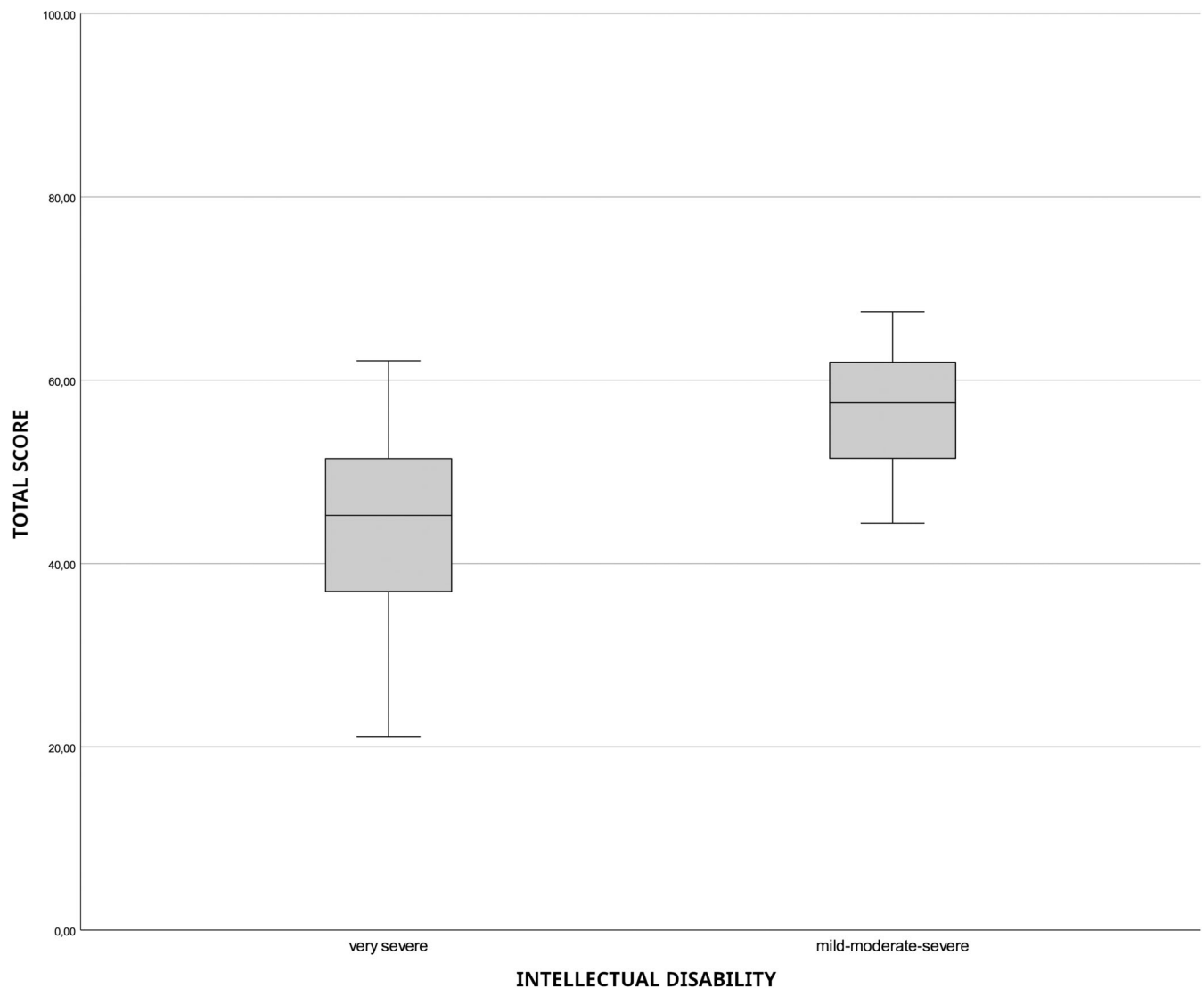


FIGURE 3 Box plots of mean CPADULT total score (y-axis) for two intellectual-disability groups (x-axis).

The hypotheses concerning construct validity were confirmed with statistically and clinically significant differences in the total score and relevant domain scores. Only the total score difference between the two motor-function groups was slightly below the SDC. In terms of interpretability, the results revealed that there were no floor and ceiling effects, thereby indicating that this measure is appropriate for the target population and that good differentiation between individuals is possible.

Similar results have been reported for the psychometric properties of the CPCHILD-DV, as compared to the CPADULT (Zalmstra et al., 2015). In this study, the Internal consistency of the CPCHILD-DV was 0.89 for the total score, with a range of 0.60–0.95 for the domains. The ICC for the test–retest reliability of the total score was 0.73, with a range of 0.55–0.80 for the domains. The limits of agreement ranged within 16.9 points of the mean difference. The CPCHILD-DV score was able to differentiate between subgroups of motor function, as well as between subgroups of cognitive function

within the domain of ‘communication and social interaction’. Given the similarity in items and psychometric characteristics, both measures can be used consecutively to follow a person from childhood to adulthood. In a cross-sectional study in persons with CP, by comparing a sample of youth ranging from 13 to 18 years old with a sample of adults from 23 to 33 years old, health and HRQoL outcomes (measured with general instruments) in persons with CP (ambulatory and non-ambulatory) appeared to be relatively stable across the transition from childhood to early adulthood (Young et al., 2010). However, it was stated that, since persons with CP have limited health status and will require health care support throughout their lives, longitudinal follow-up studies in this population are essential to enhance the understanding of patterns of health and its relationship to QoL over time (Young et al., 2010).

An example of longitudinal follow-up studies are the longitudinal decade studies of PERRIN (Paediatric Rehabilitation Research in the Netherlands; van Gorp et al., 2020). The PERRIN studies identified

childhood factors in children with CP that may predict future restrictions to participation. In this study, 67 children with CP (ambulatory and non-ambulatory) were followed over a period of 13 years, throughout the transition to adulthood. Based on the results, it was concluded that children with CP, who have limited motor capacity, limited manual ability, intellectual disability, or epilepsy are at risk of future restrictions to participation in domestic life (e.g., activities of daily life) or interpersonal relationships (e.g., social interaction) in young adulthood. However, information concerning HRQoL was lacking. Specifically for the group of children with CP who are non-ambulatory, the CPCHILD-DV and CPADULT together give opportunities to perform longitudinal follow-up studies on HRQoL.

4.1 | Methodological reflections and further research

The COSMIN group recommends collecting data from at least 50 participants when establishing the psychometric properties of a questionnaire. Although our sample of 47 participants almost reached this number, the results would have been statistically more robust with more participants. Due to the COVID-19 pandemic, it was difficult to receive enough participants, even after extending the recruitment period by 6 months. One problem could have been that the pandemic increased the burden of care for parents at home when day-care centres were temporarily closed (Mathur et al., 2023). In addition the DSPs might have encountered an increase of their workload due to illness on the part of their colleagues (Shi et al., 2022). Under such circumstances, participation in a study might not have been regarded as a priority. Furthermore, the recruitment of participants in this target group is often inherently challenging (Maes et al., 2021). Although we aimed to achieve an even distribution of the subgroups, the group with higher levels of motor function and the groups with less severe intellectual disability were small. It was therefore necessary to combine groups in order to perform statistical analyses. In addition, when comparing subgroups, the confidence intervals were wide and the results were statistically less robust. With regard to the characteristics of the participants, it is notable that the majority (77%) were women. The underrepresentation of men (fathers and male DSPs) might have influenced the results, given that the views of the caregivers were incorporated in completing the proxy-measure. As indicated by previous studies (Narayanan et al., 2006; Zalmstra et al., 2015, 2019), however, mothers are more likely to participate. It is also known that the majority of DSPs are women. This sample might therefore be a valid representation. At the same time, however, it could be equally meaningful and important to obtain the perspectives of fathers on the quality of life of their children with severe disabilities. A review of 12 studies demonstrated, in families with a child with a disability, differences in experiences of fathers compared to mothers (Boyd et al., 2019). For example, it is known that fathers might appraise the impact of raising a child with severe disabilities less negatively than mothers (Luijkx et al., 2019).

This study was based on proxy-reports. It is known that parents have unique knowledge of their child's communication, well-being and pain through intensive interactions (Kruithof et al., 2020). Professional caregivers learn to understand the non-verbal communicative behaviour of their clients (Mietola et al., 2017). However, the interpretation of an individual's non-verbal communicative behaviour has limitations. A recent review has shown inconsistent agreement between self-reports and proxy-reports, for individuals with intellectual disabilities who were capable of self-reporting (Santoro et al., 2022). For this reason, recommendations call for using self-reports, whenever possible. However, in the group of people with severe disabilities who are non-ambulatory, self-evaluation is highly challenging, if not impossible. Using proxy-reports is assumed to be the closest approximation to self-report and can give a voice to persons who are nearly unable to self-report (Alves-Nogueira et al., 2020). Nevertheless, in practise, caregivers could involve their children/clients, in completing the CPADULT questionnaire, if their communicative and cognitive skills allow.

This study is the first step in the process of validating the CPADULT. The ongoing validation process will guide future research. In the absence of a 'gold standard' for assessing HRQoL in this target group, it was not possible to assess the criterion validity. It might nevertheless be possible to examine convergent validity by comparing the instrument to other measures of HRQoL.

For further testing, future studies could assess the responsiveness of the instrument to assess the ability of the measure to detect change over time. This would require a longitudinal study, however, in which participants are followed over time (e.g., before and after an intervention). Such studies could also provide the opportunity to calculate the MCID using an anchor-based method, with the rating of improvement serving as the anchor.

The generalizability of the results of this study is limited, as all participants were Dutch. The translation and cultural adaptation of the CPADULT into other languages would create opportunities to enhance the instrument's validity and generalizability.

4.2 | Practical implications

The development and testing of a proxy instrument for measuring HRQoL that takes the severe motor disabilities and combined intellectual disabilities into account has made it possible to apply HRQoL in support services for adults with severe motor and intellectual disabilities who are non-ambulatory. It could also be applied in research, as an evaluative or discriminative outcome measure for this target group. The CPADULT can be used at the individual level to evaluate adults with severe disabilities who are non-ambulatory over time (e.g., through annual evaluation). It could also be used to assess the effects of interventions (e.g., medication or surgery) or life events (e.g., moving to another living environment, changing routines related to sleeping problems, or the loss of a relative). In research, the CPADULT could be used at the group level to assess the effects of interventions.

In conclusion, this study has demonstrated that the CPADULT has sufficient reliability and validity as a proxy measure of HRQoL for adults with severe disabilities who are non-ambulatory.

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DATA AVAILABILITY STATEMENT

Data not available, due to no consent for sharing from participants.

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