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Development and sensibility assessment of a health-related quality of life instrument for adults with severe disabilities who are non-ambulatory

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Abstract

Background: Insight in health-related quality of life (HRQoL) of adults with severe disabilities who are non-ambulatory is important, but a measure is lacking. The aim was to develop a HRQoL measure for this group.

Method: The developmental process consisted of the adaptation process of a proxy HRQoL measure for children with severe disabilities who are non-ambulatory and the assessment of the sensibility of the developed instrument. A three-step process was used: focus groups, e-survey and interviews.

Results: In total, 72% of the items remained unchanged. Three new items and one element to an existing item were added. In ten items, the formulation of the items was adapted to the target group. Concerning the sensibility, respondents suggested minor changes to the instruction and the output scales.

Conclusions: This study has yielded a proxy HRQoL measure for adults with severe disabilities who are non-ambulatory, the CPADULT, with good sensibility.

KEYWORDS

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

1 | INTRODUCTION

Health-related quality of life (HRQoL) is defined as an individual's perception of how an illness and its treatment affect the physical, mental and social aspects of his or her life. HRQoL is seen as a multidimensional construct, which can be measured indirectly using multiple items in several domains (Vet et al., 2011). These domains encompass aspects of physical, psychological and social functioning. The domains of HRQoL are a subset of the eight-core quality of life (QoL) domains that have been identified and which cover all aspects of life: emotional well-being, interpersonal relationships, material well-being,

personal development, physical well-being, self-determination, social inclusions and rights (Schalock et al., 2005) (Gómez et al., 2015). QoL is, therefore, broader than HRQoL because it includes evaluation of non-health-related features of life, whereas HRQoL is connected to an individual's health or disease status. As QoL is important to everyone (with or without disabilities), HRQoL focuses on the impact health status has on quality of life. HRQoL is especially important for people with chronic illness or disabilities since the consequences of their conditions play a prominent role in almost every aspect of their lives and are lifelong. Most interventions for these people have a common goal, not to cure but to preserve or enhance their health-related quality of

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life. Solid evidence that the intervention will make a meaningful difference is often lacking. HRQoL is generally considered a valid indicator of intervention outcomes and can be used to assess unmet needs (Health related quality of life, 2018).

For people with severe motor and intellectual disabilities who are non-ambulatory, insight into their HRQoL is, therefore, important, however difficult to measure. In the Netherlands, around 125.000 adults have such severe motor disabilities (Klerk et al., 2016) that they are characterized as non-ambulatory. The severe motor disabilities due to, for example, cerebral palsy (CP) or genetic disorders are often accompanied by moderate, severe or profound intellectual disabilities. These combined disabilities cause difficulties in activities in daily living, communication, mobility and health in such a way that these individuals highly depend on others such as caregivers or health care professionals for the main part of their daily needs (Narayanan et al., 2006). As life expectancy increases for these individuals, like the general population (Bahk et al., 2019) (Jones et al., 2015), there is a growing number of adults with severe motor and intellectual disabilities who are non-ambulatory. A small number of them still lives with their family, while others stay in residential facilities for adults with intellectual disabilities where direct support professionals (DSP's) support them in nearly all aspects of their daily lives. The focus of support for these adults—as in their childhood—is to limit the consequences of the disabilities and to improve HRQoL. In order to evaluate health status, well-being and the effects of interventions and support, HRQoL has been recognised as an important and valid outcome measure (Laporta-Hoyos et al., 2017). Ideally, a person's HRQoL should be assessed by the individual concerned. However, for adults with severe motor and intellectual disabilities who are non-ambulatory, self-report questionnaires are generally highly challenging and nearly impossible. Therefore, to assess HRQoL, the next best option is to rely on proxy rating, the perspective of the primary caregiver (parent or DSP) (Alves-Nogueira et al., 2020).

Little is known about the HRQoL of adults with severe motor and intellectual disabilities who are non-ambulatory (Jiang et al., 2016) (Petry et al., 2009). In one study concerning people with cerebral palsy with varying levels of intellectual disabilities, it was observed that adults who are non-ambulatory had significantly lower HRQoL scores than a group of adults with less severe motor disabilities (Young et al., 2010). In this study, the HRQoL was measured with a generic HRQoL instrument. Generic HRQoL instruments are applicable to everyone, but do not sufficiently focus on and are not sensitive to individuals who are non-ambulatory because of floor effects and inappropriate items (Vitale et al., 2005). A systematic review (Townsend-White et al., 2012) identified six QoL instruments for people with intellectual disabilities with acceptable psychometric properties, such as the Multifaceted Life Satisfaction Scale (Harner & Heal, 1993) and the Evaluation of Quality of Life instrument (Nota et al., 2006), which relies on proxy responses. Most instruments were suitable for people with high levels of intellectual/adaptive functioning. The QoL-PMD (in the Dutch language) is specially developed to measure QoL for

people with profound intellectual and multiple disabilities (Petry et al., 2009) and has promising validity. However, these are all QoL instruments which, as mentioned before, cover all aspects of life. The items of QoL and HRQoL instruments overlap to some degree, but HRQoL instruments have a focus on the impact health status and treatment may have on QoL. HRQoL measures enable demonstrating the impact of health on quality of life in a scientific way (Health related quality of life, 2018). A specific HRQoL measure for adults with severe motor and intellectual disabilities who are non-ambulatory is lacking. In order to use HRQoL as an evaluative and discriminative outcome for adults with severe motor and intellectual disabilities who are non-ambulatory in clinical practice or in research, there is need for a proxy HRQoL instrument that takes into account the severe motor disabilities and combined intellectual disabilities of this target group. A validated HRQoL proxy measure specifically developed for children with severe motor and intellectual disabilities who are non-ambulatory is already available: the Caregiver Priorities and Child Health Index of Life with Disabilities (CPCHILD) (Narayanan et al., 2006). The CPCHILD covers a variety of activity and participation categories of the International Classification of Functioning Children and Youth version (Schiariti et al., 2011). The CPCHILD is translated in many languages and used worldwide (CPCHILD, 2018). In previous studies, a validated Dutch version of the CPCHILD was made (Zalmstra et al., 2015) (Zalmstra et al., 2018).

The CPCHILD focusses on observation of practical everyday activities and behaviour for children who are non-ambulatory. The items concern how difficult aspects of daily life are and the level of assistance that is needed. Furthermore, pain behaviour and communication skills are observed. Also, some health aspects (visits to doctor and medications) are registered. Although the CPCHILD is not developed to be applicable to adults, it may contain many elements that are likely to be relevant for adults with severe motor and intellectual disabilities as well. The CPCHILD could, therefore, be used—with permission of the authors of the original instrument—as a starting point for developing a proxy HRQoL measure for adults with severe motor and intellectual disabilities who are non-ambulatory. It is unknown, however, which items used in the CPCHILD are also applicable to adults with severe motor and intellectual disabilities who are non-ambulatory. Furthermore, additional items related to differences in HRQoL after the transition to adulthood should be considered. Alterations of the wording, the content of the questions and instructions may be needed. Probably adaptation of the questionnaire will also be necessary to change the forms of address: from child to adult and from parent to parent/DSP.

When an instrument is adapted to another target group, it is also important to assess the sensibility of the instrument. Sensibility specifically assesses whether an instrument is meaningful to respondents. Evaluation of sensibility is often qualitative and frequently based on the judgment of clinicians and individuals for whom the questionnaire was developed (Collins, 2003) (Dermott, 2015) (O'Brian et al., 2013). With a thorough development of the instrument and evaluation of its sensibility, we aim to set a firm base for

the assessment of the psychometric evaluation of the instrument in a future study.

The purpose of this explorative study was to develop a proxy instrument in the Dutch language to assess the HRQoL for adults with severe motor and intellectual disabilities who are non-ambulatory. The instrument is called the CPADULT, as it is based on the widely used CPCHILD questionnaire.

2 | METHOD

The ethical review board at the University Medical Center of Groningen decided that formal approval of the study was not required (2018/044). Written informed consent or implied consent was received from all participants. For the development of the CPADULT we used a qualitative design with the adaptation process and sensibility assessment in three steps (Figure 1).

2.1 | Step 1 (Focus groups)

The aim of this step was firstly to adapt the CPCHILD items to adults with severe motor and intellectual disabilities who are non-ambulatory and secondly to assess the sensibility of the questionnaire. The CPCHILD consists of 37 questions and two additional items across six domains: 1. Activities of daily life/personal care (nine items); 2. Positioning, transferring and mobility (eight items); 3. Comfort and emotions (nine items); 4. Communication and social interaction (seven +two items); 5. Health (three items); 6. Overall quality of life (one global item). Each item is rated, reflecting on the past 2 weeks, on a six- or seven-point ordinal scale, and for some domains a four-point level of assistance modifier is added. Standardized scores from 0 (worst) to 100 (best) are calculated for each of the six domains, and for the total survey. As it was suggested that health and HRQoL outcomes were relatively stable across the transition to adulthood (Young et al., 2010), we aimed to stay as close as possible

to the original child-version by making only necessary changes to the adult version. In this way, comparison between both measures is possible, for example, to follow a person from childhood to adulthood.

A framework for evaluating sensibility was proposed by Feinstein (Feinstein, 1987), consisting of seven domains: 1. Purpose and framework; 2. Comprehensibility; 3. Replicability (clarity of instruction); 4. Suitability of the output scale; 5. Face validity; 6. Content validity; 7. Ease of usage. The domain 'Purpose and framework' of the CPADULT is similar to that of the CPCHILD and did not need further evaluation. The purpose of this instrument is to measure the caregivers' perspective of the activity limitations, health status, well-being and ease of care for people with severe motor and intellectual disabilities (Narayanan et al., 2006). The measure can be used as an evaluative application (to evaluate effects of interventions or longitudinal changes) or as a discriminative application (to discriminate between (groups of) persons) (Vet et al., 2011). The conceptual framework is based on a reflective model, in which the construct is reflected by and manifested in the items of the measure (Vet et al., 2011). The domains 'Face validity' and 'Content validity' were addressed during the adaptation process. The other four domains of sensibility were addressed in the sensibility assessment.

Two focus groups were formed: a group of parents and a group of DSP's. Homogeneous groups were created to enable comparison of similarities and differences in the results. We aimed to include five to ten people per focus group (Cleary et al., 2014). We approached 15 residential facilities in the Netherlands and we posted messages on social media for potential participants: primary caregivers (parent or DSP) of persons with severe motor and intellectual disabilities who are non-ambulatory due to, for example, cerebral palsy or genetic disorders, age from 18 years up. The primary caregiver (parent or DSP) had to have sufficient understanding of the written Dutch language.

Beforehand all focus group members were provided with the CPCHILD questionnaire and were asked to formulate suggestions for adaptation of the items. In the focus group meetings, the participants discussed all the proposed adaptations of the items until consensus was reached thereby addressing the face validity. In the

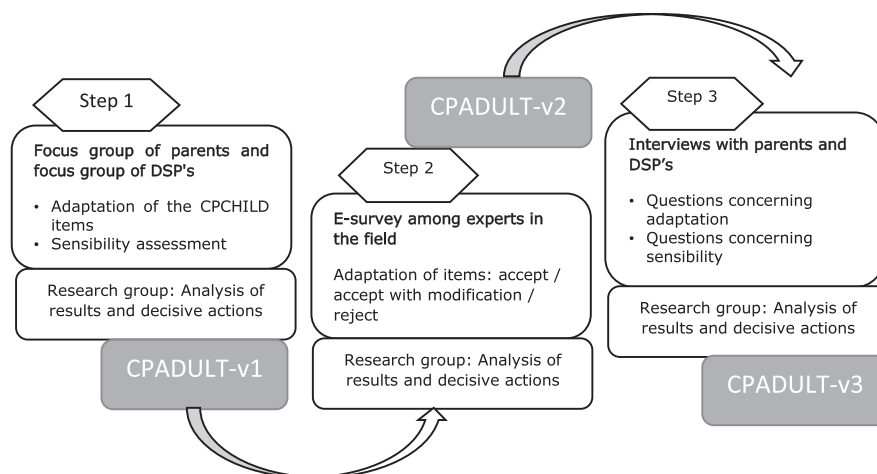


FIGURE 1 Flow chart of adaptation process and sensibility assessment

DSP=direct support professional; CPCHILD=Caregiver Priorities and Child Health Index of Life with Disabilities; v1=version 1; v2=version 2; v3=version 3

next step, the remaining four domains of the sensibility assessment were addressed: comprehensibility, clarity of instruction, suitability of the response scale and ease of usage. The members were asked to rate the sensibility items on a five-point ordinal scale (from 1 'poor' to 5 'good') and the items were further discussed during the meeting. Beforehand it was stated that the median of the rating scores should be at least 4 ('adequate') to be sufficient and that at least 75% of the respondents should consider the output scales to be suitable (Rowe & Oxman, 1993). A coordinator—one of the researchers—guided the group to ensure that the original meaning of items and the questionnaire structure were maintained. The proposed adaptations and the rationale for these were documented. The results of both focus groups were compared afterwards. When adaptations had been suggested in only one focus group, these suggestions were subsequently presented to the members of the other focus group by e-mail to endorse or reject.

The results of the focus groups were subsequently analysed by the research group. Each suggestion and its rationale were reviewed against the general principle to stay as close as possible to the original child-version and to make only those changes that were necessary to compose the adult version. In this way the first version of the questionnaire, the CPADULT-v1 was constructed.

2.2 | Step 2 (E-survey)

The second step was the evaluation of the CPADULT-v1 concerning the adaptation process and assessing content validity by means of an e-survey among experts in the field. We aimed to include a minimum of 25 participants with different professional background and various levels of experience (Dermott, 2015). The inclusion criteria were: healthcare psychologists, physicians (physiatrists and physicians working with persons with intellectual disabilities), researchers, therapists who were professionally involved with adults with severe motor and intellectual disabilities who are non-ambulatory for at least 1 year. We approached relevant networks of the respective professional groups in the Netherlands to recruit participants for the e-survey.

These experts were asked to review all items of the CPADULT-v1 questionnaire according to the following options: accept, accept with adaptation or reject with rationale. Additionally, strong and weak points of the questionnaire and its usefulness were evaluated. Subsequently, the results of the e-survey were reviewed in the research team applying the same general principle as in step 1. A question was only considered to be rejected if it was suggested by more than 10% of the experts (Dermott, 2015). The analysis of the results of the e-survey resulted into the CPADULT-v2.

2.3 | Step 3 (One-to-one interviews)

In the third and final step, the CPADULT-v2 was tested by means of one-to-one interviews with a different group of parents/DSP's of

adults with severe motor and intellectual disabilities who are non-ambulatory. We aimed to conduct five to ten interviews or until data-saturation was reached. We approached ten residential facilities in the Netherlands and we posted messages on social media for potential participants for the interviews: primary caregivers (parent or DSP) of persons with severe motor and intellectual disabilities who are non-ambulatory due to, for example, cerebral palsy or genetic disorders, age from 18 years up. The primary caregiver had to have sufficient understanding of the written Dutch language. During the interviews, the participants were invited to provide comments about each item of the questionnaire. The sensibility topics were addressed by rating them on five point ordinal scale (from 1 'poor' to 5 'good') Beforehand it was stated that the median of the rating scores should be at least 4 ('adequate') to be sufficient and that at least 75% of the respondents should consider the output scales to be suitable. The same inclusion criteria as in step 1 were applied. Following this, the responses were reviewed in the research team, leading to the CPADULT-v3 and final version.

The qualitative results were analysed using Atlas TI. IBM SPSS statistics 23 was used for descriptive statistics of the participants.

3 | RESULTS

3.1 | Step 1 (Focus groups)

In the focus group of parents, five females participated with a mean age of 41.0 years (SD 10.9). In the focus group of DSP's, one male and four females participated with a mean age of 59.4 years (SD 5.4). The suggested adaptations per domain are presented in Table 1. Table 2 represents the suggested changes concerning the sensibility topics. The median of each rating of the sensibility topics was 4 ('adequate'). The output scales were considered suitable by 90% of the respondents. All the suggestions made in only one group were subsequently endorsed by the other group. Three new items directly related to adulthood were added: menstruation pain, sexual experiences and identity. Also, the element 'shaving' was added to the item about personal care around head and face. Furthermore, a fourth new item, not specifically connected to adulthood, was suggested by the focus groups: sleeping pattern. Moreover, suggestions for rephrasing the items were made and subsequently agreed on by the research team (see Tables 1 and 2). Because of the general principle to stay as close as possible to the original child-version, some suggested changes to the items were not implemented (see the third column in Tables 1 and 2). The order of the domains remained, therefore, unchanged and the question about eating, drinking and tube feeding was not split, because that would otherwise complicate the comparison of the domain scores. The regarded time frame in which to assess the items was kept at 2 weeks, to keep the time frame uniform throughout the questionnaire. The CPADULT-v1 contained 43 items.

TABLE 1 Adaptation of items in focus groups

Domains	Suggested changes	Participants	Agreed in research team
Activities of daily life/personal care	• Reversing domain 1 and 2, because of emotional impact of domain 1	Parents	No
	• Adding an item about shaving	DSP's	Yes
	• Splitting item about eating/drinking/tube feeding into three items	Parents	No
Positioning, transferring and mobility	Minor rephrasing of the items	Both	Yes
Comfort and emotions	• Adding the item 'menstruation pain'	Both	Yes
	• Adding scores of pain scale (when applicable)	Both	No
	• Adding positive emotions	Both	No
Communication and social interaction	• Rephrasing of items: 'to play' changed into 'amuse oneself'; 'to play with others' into 'social interaction'; 'school/child day care centre' into 'day care';	Both	Yes
	• Adding to the item 'building a relationship with you': 'and significant others'.	Both	Yes
	• Adding two items about sexuality, one about experiencing sexuality, and one item about identity	Both	Yes
Health	• Adding an item about sleep pattern	DSP's	Yes
	• Regarding the past 6 months instead of past 2 weeks	Both	No
Overall quality of life	No changes	Both	Yes

DSP, direct support professional.

TABLE 2 Sensibility topics of the focus groups

Topic	Sensibility ratings	Suggested changes	Participants	Agreed in research team
Comprehensibility	Median: 4 out of 5	Regarding the past month instead of 2 weeks in which the items were to be assessed	Both	No
Replicability (clarity of instruction)	Median: 4 out of 5	Missing of framework of reference regarding the disabilities: adding to the instruction the words: 'taking into account the limitations of your child'	Parents	Yes
		For DSP's the word 'client' seemed the best option, for parents the word 'child' remained the best fit.	Both	Yes
Suitability of the output scales	Suitable: 90%	Include a box 'not applicable' in some of the questions (e.g. toilet use).	Both	Yes
		Level of assistance scale: adding another category 'largely'	Both	No
		The scale 'how often pain or discomfort' contains unclear categories	Both	Yes
Ease of usage	Median: 4 out of 5	Make a digital version of the questionnaire	Both	Yes
		Make clear that you could write comments on the line under every item.	Both	Yes

Abbreviations: DSP, direct support professional.

3.2 | Step 2 (E-survey)

Table 3 shows the characteristics of the 32 participants (mean age 43.8 [SD 13.1]) of the e-survey. The main results of the e-survey are listed in Table 4. Overall, 34 out of the 43 items of the CPADULT-v1 were accepted in their original form and 8 items were accepted with adaptations. The adaptations mentioned in the fourth column

of Table 4 were implemented in the questionnaire. The adaptation 'exercise some control over his own life' connects with the growing awareness of rights and self-determination of people with intellectual and motor disabilities. One item was rejected by more than 10% of the participants: the new question about identity. It was considered too difficult and too complex to answer and was subsequently removed from the questionnaire. In the instructions, an indication of the time needed to complete the questionnaire, namely 20–30 min,

was added. The CPADULT was considered by 73% of the participants to be potentially useful in their work field. As strong points of the questionnaire, 44% of the respondents considered it 'complete', 'multidimensional', and 22% mentioned 'clear', 'specific'. The most important weak points mentioned were: 'sometimes vague output scales', 'subjective' and 'subject to various interpretations' (16%). The CPADULT-v2 contained 42 items.

TABLE 3 Characteristics of the e-survey participants

n = 32	n (%)
Gender	
Female	26 (81)
Male	4 (13)
Missing	2 (6)
Profession	
Physiatrist	5 (16)
Intellectual disability physician	4 (13)
Health care psychologist	6 (19)
Physiotherapist	10 (31)
Occupational therapist	4 (12)
Researcher	3 (9)
Setting	
Residential facility	18 (56)
Day care centre	4 (13)
Consultancy-function	2 (6)
Missing	8 (25)
Experience with target Group	
<5 years	8 (25)
5–10 years	5 (16)
10–15 years	4 (12)
>15 years	15 (47)

TABLE 4 Results of the e-survey

Domain	Accepted	Accepted with modification	Modifications	Rejected by >10%
Activities of daily life/personal care	8	1	'Diaper' rephrased into 'incontinence pads'	0
Positioning, transferring and mobility	7	1	Delete 'exercise' in 'standing for transfers/exercise'	0
Comfort and emotions	9	1	Delete 'in bed' in 'pain while lying in bed'	0
Communication and social interaction	6	4	'To make his intentions clear' changed into 'exercise some control over his own life' Change: 'being at day care' into 'participate in day care' Change 'practice sexuality' into 'experience sexuality' Change 'to be understood' into 'to express oneself'	1
Health	3	1	'How many medication' was changed into 'impact of daily medical actions'	0
Overall quality of life	1	0		0
Total	34	8		1

3.3 | Step 3 (One-to-one interviews)

Two DSP's and two parents, three females and one male, with a mean age of 55.8 years (SD 19.4), participated in the interviews. Data-saturation concerning the adaptation process was reached after these four interviews. No adaptations of the items were suggested, all items were considered important by all participants, thereby confirming the face validity. The median of each rating of the sensibility topics was 4 ('adequate'). The output scales were considered suitable by 100% of the respondents. The comprehensibility was found good and the items were easy to understand by all participants. The time it took to complete the questionnaire was on average 20 min. Concerning the sensibility topics, there were some minor changes suggested. To improve the clarity of instruction, one participant suggested minor rephrasing of one sentence. Another participant proposed to add a comment box under each domain instead of under each item, to enhance ease of usage. These two changes were incorporated in the CPADULT-v3. For suitability of the response scale, one participant suggested a 5-point scale instead of a 7-point scale. Following the before mentioned general principle to stay close to the child-version, this suggestion was disregarded. During the interviews, it became also clear that a digital version of the questionnaire was not easy to use for everyone. So it was recommended to also have a paper version of the questionnaire. The CPADULT-v3 and final version contained 42 items across six domains: 1. Activities of daily life/personal care (nine items); 2. Positioning, transferring and mobility (eight items); 3. Comfort and emotions (ten items); 4. Communication and social interaction (ten items); 5. Health (four items); 6. Overall quality of life (one global item).

In this three-step adaptation process, overall 28 out of 39 items (72%) remained the same as compared with the original CPCHILD questionnaire. In total, three new items and one new element to an existing item were added. In ten items the phrasing was adapted, mostly in the domain 'Communication and social

interaction'. In five of these items, the wording was changed because of the transition to adulthood. In the other five items more general minor rephrasing was done to clarify the item. For further information about the items, the corresponding author can be contacted.

4 | DISCUSSION

This explorative study has set out to develop a proxy HRQoL instrument specifically for adults with severe motor and intellectual disabilities who are non-ambulatory, the CPADULT. The results show that the sensibility of the CPADULT has been well established. The comprehensibility, clarity of instruction, suitability of the output scale, and ease of usage have been discussed extensively, the median of each rating of the sensibility topics reached the threshold and minor alterations were made. Looking at the adaptation process, the CPADULT mainly consists of items of the CPCHILD, although specific items which were connected to adulthood were added and some items were rephrased to fit the target group. Face and content validity are supported by these findings. The nature and number of the domains remained unchanged during the adaptation process. Comparison between scores of the CPCHILD and CPADULT is possible, for example, when evaluating the transition of a person from childhood to adulthood. The domains that have been altered the most were the following: 'Communication and social interaction' and 'Health'.

There was one new item added to the CPADULT, which seems to be important for children as well as for adults: the sleeping pattern. Sleeping problems are found in about 15%–50% of adults with intellectual disabilities and 58% to 80% of children with intellectual disabilities (Hylkema & Vlaskamp, 2009). A negative correlation between the presence of sleeping problems and quality of life in children with CP was found previously (Horwood et al., 2019) (Zuculo et al., 2014). Therefore it might be worthwhile to add this item also to the CPCHILD.

Although there are some similarities in the items of the CPCHILD/CPADULT and the before mentioned QoL-PMD, for example, communication and emotions, the focus of the instruments is different, resp. HRQoL and QoL. Instead of the broader focus of QoL which covers all aspects of life, the CPADULT explores in greater detail the effects of the health status on daily life, for example, activities of daily living, personal care, positioning and transferring.

As mentioned in the introduction, we are compelled to use proxy rating, the perspective of the primary caregiver (parent or DSP), because of the intellectual and communicative disabilities of the target group. The agreement between the ratings of the person himself and the proxy is somewhat questioned (Gómez et al., 2015). The parents or DSP's, however, develop special skills in listening and interpreting the persons' non-verbal communicative behaviour (Mietola et al., 2017). Unfortunately, the understanding of others is always partial, especially in the case of people whose communicative behaviour is

unconventional and idiosyncratic. Still the parent's or DSP's reports are the closest possible approximation of the HRQoL of persons with severe motor and intellectual disabilities who are non-ambulatory.

An alternative to self-report of people with severe motor and intellectual disabilities can be observations of their behaviour. The CPADULT, as is the CPCHILD, focusses on observation of practical everyday activities and behaviour. Although, interpretation of behaviour can also be biased by the observers (Maes, 2020) because of their own knowledge of the individual and previous experiences with the individual in similar situations (Munde et al., 2011). When validating the Dutch version of the CPCHILD we assessed the inter-observer reliability in asking two members of the family—mostly mother and father—to complete the questionnaire. The ICC was good—0.64—but a lower score than the test–retest ICC of 0.73 (Zalmstra et al., 2015). This shows that proxy ratings will inevitably be influenced by the parent's or DSP's unique perceptions and attitudes, value judgments, as well as elements of their own quality of life.

To make observations more objective, controlled observations with or without the use of video recordings could be considered alongside the questionnaire which has also been done for observing pain behaviour (Putten & Vlaskamp, 2013) or emotions (Vos et al., 2013). There are also promising ongoing studies using bio-response systems (Frederiks et al., 2019).

It would be also useful to explore how the adults with severe motor and intellectual disabilities who are non-ambulatory can be more involved in completing the questionnaire. The parent or DSP can be encouraged to discuss the items with the adult with severe disabilities, when this is possible while completing the questionnaire. Also, an attempt can be made to develop a subset of adapted items to complete for the adult with severe disabilities who are non-ambulatory with less severe intellectual disabilities. The challenges of this study will include to determine which items are applicable for self-evaluation for this target group and how to formulate the items. Also, the type of Likert scales must be assessed (words, numbers or faces) since it has been established that even children in primary school have limited understanding of Likert response formats (Mellor & Moore, 2014). Maybe photos or pictures could be used to help understand the questions.

4.1 | Methodological reflections and further research

The development of a HRQoL instrument for a specific target group by means of a thorough three-step adaptation process and sensibility testing is a strong point of this study. Assessing the sensibility of the instrument enhances the meaningfulness of the instrument for the respondents and attributes to the psychometric quality of the measure. A limitation of this study is the relatively low number of participants in the interviews. Given the fact that the interviews provided minimal new findings, it seemed that data-saturation has arisen, especially regarding the adaptation process.

The impact of the small sample size therefore seems to be limited. Another limitation is that the CPADULT questionnaire is at this moment in a developmental stage. Therefore, reliability and validity need to be further explored in future studies. Furthermore, the questionnaire is written in the Dutch language and has, therefore, a small range of use.

Comparing the two focus groups (parents and DSP's), there were many similarities in the results as well as some small differences. Both parents and DSP's in general appeared to have the same view on the HRQoL of their child/client. This supports the face and content validity of the instrument, as both groups are proxies of the target population of the CPADULT. A disadvantage of these homogeneous groups is that there was no opportunity for interaction or discussion between parents and DSP's, which could have brought new insights. Considering the small differences between the two groups we propose that in similar future studies parents and DSP's will join in one heterogeneous focus group.

Before the CPADULT can be used in practice, the psychometric quality of the instrument has to be assessed. We are currently performing a study, in which the internal consistency, test-retest reliability, minimally clinical important difference, floor and ceiling effects will be assessed. Furthermore the known-groups validity—comparing the scores of different levels of motor function and of different levels of cognitive function—and the content validity of the CPADULT will be assessed. If we can show that the psychometric quality is acceptable, the CPADULT can be used in clinical practice as an evaluative measure of HRQoL. It can be used to evaluate adults with severe motor and intellectual disabilities who are non-ambulatory over time (e.g. a yearly evaluation) or assess the effect of an intervention or life events, for example, a move to another living environment, changing routines related to sleeping problems or the loss of a relative. In clinical care and in research it can be used to assess the effect of an intervention.

To make the CPADULT more internationally usable, a translation and validation in other languages, particularly in English, is to be recommended and part of future studies.

In conclusion, this explorative study has yielded a newly developed proxy HRQoL instrument for adults with severe motor and intellectual disabilities who are non-ambulatory, the CPADULT, with good sensibility that could serve as a measure of HRQoL after its psychometric quality is assessed.

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DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

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