



Minerva Access is the Institutional Repository of The University of Melbourne

Author/s:

Epstein, A;Williams, K;Reddihough, D;Murphy, N;Leonard, H;Whitehouse, A;Jacoby, P;Downs, J

Title:

Content validation of the Quality of Life Inventory—Disability

Date:

2019-01-01

Citation:

Epstein, A., Williams, K., Reddihough, D., Murphy, N., Leonard, H., Whitehouse, A., Jacoby, P. & Downs, J. (2019). Content validation of the Quality of Life Inventory—Disability. *Child Care Health and Development*, 45 (5), pp.654-659. <https://doi.org/10.1111/cch.12691>.

Persistent Link:

<https://hdl.handle.net/11343/286882>

Epstein Amy (Orcid ID: 0000-0003-4441-0901)
Downs Jenny (Orcid ID: 0000-0001-7358-9037)

Title: Content validation of the Quality of Life Inventory - Disability (QI-Disability)

Running title: Content validation of QI-Disability

Authors:

Amy Epstein MA,¹ Katrina Williams MBBS,^{2,3} Dinah Reddihough MD,^{2,3} Nada Murphy MAppPsych,¹ Helen Leonard MBChB,¹ Andrew Whitehouse PhD,¹ Peter Jacoby MSc,¹ Jenny Downs PhD^{1,4}

Affiliations:

1 Telethon Kids Institute, The University of Western Australia, Perth, Western Australia, Australia.

2 Developmental Disability and Rehabilitation Research, Murdoch Children's Research Institute, Melbourne, Victoria, Australia.

3 Department of Paediatrics, University of Melbourne, Melbourne, Victoria, Australia.

4 School of Physiotherapy and Exercise Science, Curtin University, Perth, Western Australia, Australia.

Address correspondence to:

Dr Jenny Downs
Telethon Kids Institute
University of Western Australia
PO Box 855, West Perth
WA 6872, Australia
ph +61 8 6319 1763
E-mail: Jenny.Downs@telethonkids.org.au

Word count: 2454 words

This is the author manuscript accepted for publication and has undergone full peer review but has not been through the copyediting, typesetting, pagination and proofreading process, which may lead to differences between this version and the Version of Record. Please cite this article as doi: [10.1111/cch.12691](https://doi.org/10.1111/cch.12691)

ACKNOWLEDGEMENTS

The authors would like to thank all of the families who shared their thoughts and time with us. This study is currently funded by the National Health and Medical Research Council (#1103745). AW and HL are each supported by a Senior Research Fellowship from the National Health and Medical Research Council (#1077966, #1117105 respectively). The funders of this research have had no roles in the study design, data collection, data analysis, manuscript preparation and / or publication decisions.

ABSTRACT

BACKGROUND Focus is shifting to better understand the lived experiences of children with intellectual disability in relation to their quality of life (QOL). Yet no available QOL measures are grounded in the domains important for this population. We previously conducted qualitative parent caregiver interviews identifying QOL domains in children with intellectual disability to constitute a new measure of QOL. This study describes the content validity of the Quality of Life Inventory – Disability (QI-Disability), a parent-report measure developed for children with intellectual disability.

METHODS AND RESULTS Questionnaire items were extracted from a qualitative dataset of 77 parent caregiver interviews. To establish content validation, a draft of QI-Disability was administered to 16 parent caregivers of children with intellectual disability (Down syndrome, Rett syndrome, cerebral palsy, or autism spectrum disorder). Parents participated in a cognitive interviewing procedure known as the “think-aloud” method. The process of item generation, cognitive debriefing and refinement of QI-Disability prior to its pilot testing are described. A conceptual framework is presented.

CONCLUSIONS Satisfactory content validity is reported, where ongoing consumer feedback shaped the dataset from which the final items were selected. Use of QI-Disability for children with intellectual disability will allow for greater insight into service utility and targeted intervention.

Keywords: Quality of life, intellectual disability, children, adolescents, measurement, test validation

Key messages

- The Quality of Life Inventory–Disability addresses the need for a quality of life measure designed specifically for children with intellectual disability.
- Satisfactory content validity supports this measure’s capacity to reflect the perspectives and experiences of the group of interest.

INTRODUCTION

Approximately 1.9 per 100 children have intellectual disability (ID) where significant difficulty performing daily life skills is apparent prior to the age of 18 years. Severity ranges from mild to profound, affecting functional abilities and support needs, and approximately 15% of children have severe impairments (Leonard *et al.* 2003). Children with ID have greater exposure to the social determinants of poor health such as economic disadvantage (Bigby 2012, Leonard *et al.* 2011). The effects of ID are pervasive for both physical (e.g. (Leonard *et al.* 2017, Roizen and Patterson 2003)) and mental health (Einfeld *et al.* 2011), and pose substantial threats to quality of life (QOL).

QOL refers to an individual's satisfaction with a composite of life experiences across physical, social and emotional domains, some of which are universal (e.g. physical health) and others which are condition-specific and have relevance to particular groups of children (e.g. those with chronic illness) (Felce and Perry 1995, Verdugo *et al.* 2005). Focus is shifting to better understand the lived experiences and effectiveness of treatment for children with ID in relation to their QOL. A prerequisite is access to a QOL measure that is validated for this group, yet no previously available measure was grounded in QOL domains important for children with ID.

Using *a priori* methodology and based on previous experience developing a QOL measure for cerebral palsy (Davis *et al.* 2009, Waters *et al.* 2007, Waters *et al.* 2005), we undertook the development and validation of a QOL measure for children with ID. Initial projects included four qualitative studies with parents of six to 18-year-old children with either Down syndrome (n=17) (Murphy *et al.* 2017), Rett syndrome (n=21) (Epstein *et al.* 2016); a severe genetic neurodevelopmental disorder mainly affecting females (Neul *et al.* 2010)), cerebral palsy (n=18) (Davis *et al.* 2017) or autism spectrum disorder (ASD) (n=21) (Epstein *et al.* 2019). The emergent QOL domains described observable aspects of health and wellbeing, functioning and social immersion. This is consistent with the scope of definitions of QOL which include physical, social and emotional components (Schalock *et al.* 2008, Verdugo *et al.* 2005, Waters *et al.* 2009). Items were extracted from the interview transcripts to develop the parent-report Quality of Life Inventory – Disability (QI-Disability) (Downs *et al.* 2018) and were therefore grounded in the lived experience of those affected by a particular condition or disorder, an important component of best practice development processes (Waters *et al.* 2009).

Piloted with 253 families across the four diagnosis groups, exploratory and confirmatory factor analyses were conducted and goodness of fit of the factor structure was found to be satisfactory (Downs *et al.* 2018). Compared to Rett syndrome, children with Down syndrome had higher total QOL and “independence” subscale scores, and children with ASD and comorbid ID had lower scores for “social interactions”, illustrating expected differences between known groups (Downs *et al.* 2018). We also recently confirmed that the QOL domains were consistent with those identified for

children with the CDKL5 Deficiency Disorder (Tangarorang *et al.* Forthcoming), another severe genetic neurodevelopmental disorder caused by a pathogenic mutation on the *CDKL5* gene (Fehr *et al.* 2013), suggesting generalisability of QI-Disability to other ID groups.

Establishing reliability and validity is central to developing a scientifically sound instrument and is an ongoing process (Brod *et al.* 2009). We have previously summarized the development and initial validation of QI-Disability (Downs *et al.* 2018). The aim of this study is to provide a deeper description of the content validity of QI-Disability including item generation, cognitive debriefing, refinement prior to administration and its conceptual foundation.

METHODS AND RESULTS

Description of the content validity of QI-Disability is guided by the development process for patient-reported outcomes described by Brod and colleagues (Brod *et al.* 2009) (Figure 1). Ethical approval of this study was prospectively reviewed by the Human Research Ethics Committee at the University of Western Australia, Western Australia (RA/4/1/6931) and parents provided informed consent to participate in this study.

Item generation

The 77 interview transcripts collected from parent caregivers of children with Down syndrome (Murphy *et al.* 2017), Rett syndrome (Epstein *et al.* 2016), cerebral palsy (Davis *et al.* 2017) and ASD (Epstein *et al.* 2019) formed the basis for the development of QI-Disability. The investigators (JD, AE, NM, HL) read and reread the transcript materials, extracting 77 items that described behaviours and indicated to the parent caregiver that the child was experiencing good or poor QOL. With ongoing iterative review, this set of items was refined and condensed to 50 items. The investigators then reviewed items contained in KIDSCREEN (Ravens-Sieberer *et al.* 2007), Pediatric Quality of Life Inventory (Varni *et al.* 2006) and DISABKIDS-CP module (Narayanan *et al.* 2006), but no potentially valuable item was identified as missing from our item set.

Cognitive debriefing

Parent caregivers were recruited to review the item set and participate in “think-aloud” sessions, and were selected so that their children represented variability in age, gender, and severity of ID. The children (6 males, 10 females) were aged 7-18 years and the mean (SD) age of the child at the time of the interview was 13.2 (3.3) years. Four parent caregivers were recruited from each diagnostic group. Approximately two thirds of the children (n=11) were limited in their mobility or unable to walk, and half of the children (n=8) were nonverbal and did not use speech to communicate.

Think-aloud is a cognitive interviewing method during which participants are asked to share their thoughts out aloud with the interviewer by verbalising any words or ideas that come to mind as they complete the task (Charters 2003). This technique is an

effective and reliable way to assess unique higher-level thinking processes, whilst identifying individual differences in task performance (Ericsson and Simon 1980). During a recorded telephone interview, respondents completed the 50 items in the draft QI-Disability measure during which they 1) verbalised the response given for each item on behalf of their child; 2) provided a rationale for each response; and 3) described the item in their own words. Interviews were conducted until data saturation was reached as observed by repetition of responses. The interview schedule is shown in Box 1.

Refinement of measure based on cognitive debriefing and format of validation-ready version

After reviewing all think-aloud interview data, 41 of the 50 items remained in the draft measure: six of the original items were excluded because they did not capture the intended meaning of the item, and three items were combined with other closely related items to avoid repetition. The wording of 24 (48%) items was revised to more closely align with parent interpretations and descriptions of the items (Table 1). Subsequent factor analyses of the 41 items resulted in the exclusion of an additional nine items whose loadings were < 0.4 , resulting in our final 32-item measure (Downs *et al.* 2018).

To enhance clarity, memory cues including examples discussed during the think-aloud parent interviews were integrated into 20 of the 32 items. For instance, “Over the past month, how often has your child showed happiness through body language (e.g. **making eye contact, body facing others**)”. This strategy is believed to assist responders with understanding and evaluating potentially ambiguous items whilst also bringing events to mind that may have been forgotten or overlooked (Brod *et al.* 2009).

Most items (25 of 32 in the final set) were phrased positively and the remaining seven items were worded negatively, but this was considered appropriate by the parent caregivers. The negatively worded questions pertained to pain, discomfort and negative emotions (e.g. “Showed signs of being anxious or agitated (e.g. teeth grinding, fast breathing, avoidance)”).

Finally, a five-point Likert scale was included with categories labelled by frequency (never, rarely, sometimes, often, very often), allowing parent caregivers to indicate the frequency that they *observed* a specific behaviour (e.g. smiling) rather than asking responders to infer the emotions associated with each item (e.g. happiness) on behalf of their child. A one month recall period was nominated, comparable to other QOL measures including KIDSCREEN (Ravens-Sieberer *et al.* 2007) and Pediatric Quality of Life Inventory (Varni *et al.* 2006). The items of QI-Disability have been reported by Downs and colleagues (Downs *et al.* 2018).

Conceptual model to frame relevant items and domains

Exploratory factor analysis using principal component analysis followed by confirmatory factor analysis grouped the items into six factors (Downs *et al.* 2018), broadly consistent with the ten domains identified in the qualitative studies (Davis *et al.* 2017, Epstein *et al.* 2016, Epstein *et al.* 2019, Murphy *et al.* 2017). Items describing physical health, and positive and negative emotions as extracted from qualitative data, were contained in similar factors after factor analyses. Otherwise, items from different qualitative domains were grouped to form the factors “social interaction”, “leisure and the outdoors” and “independence”. However, these groupings also made conceptual sense. For example, items describing communication experiences in social settings loaded together to represent the child’s social interactions. Items describing the pleasures of movement and balance loaded with other items describing “leisure and the outdoors” as did spending time in the natural environment, providing a comprehensive picture of participation in a variety of activities. Items necessary for day-to-day communications, routines and everyday tasks in daily living loaded onto the factor “independence”.

The framework for the International Classification of Functioning, Disability and Health (ICF) (World Health Organization 2001) conceptualizes different aspects of life affected by disability including: Body Functions and Structure and associated impairments; Activity at the individual level and associated limitations; Participation at the societal level and associated restrictions; Environmental Factors that impact these experiences and associated facilitators or barriers; and Personal Factors, which impact how an individual experiences disability. The QI-Disability items mapped across each component of the ICF framework and some factors mapped to more than one component of the framework (Figure 2).

DISCUSSION

Qualitative methods were used to identify domains of QOL important for children with ID and inform the development of a QOL measure - QI-Disability (Downs *et al.* 2018). We have reported its satisfactory reliability and validity (Downs *et al.* 2018) and we now report satisfactory content validity. Adequate assessment of content validity provides evidence that the conceptual framework, content of items, and overall measurement approach are consistent with the perspectives and experiences of the patient group of interest (Brod *et al.* 2009).

Several fundamental characteristics have guided the construction of QI-Disability (Waters *et al.* 2009). QI-Disability was specifically designed to assess the QOL of its intended population of school-aged children with ID. Therefore, each item comprising the measure is purposefully and appropriately included to help illustrate what life is like for children with ID. QOL instruments that focus extensively on functioning and skill development may fail to recognise that although a child’s level of functioning can impact their QOL, it does not define it (Waters *et al.* 2009). A disability paradox exists, whereby individuals living with serious medical issues and complex needs do not necessarily

experience poorer QOL (Albrecht and Devlieger 1999). Accordingly, items on QI-Disability were designed to focus on QOL by capturing multiple aspects of the child's health, wellbeing and life participation (World Health Organization 2001), which have visible impacts on their overall enjoyment of life.

Consumer experience contributed to an extensive dataset from which items were selected and thereafter to the specific item wording. Parents shared their experiences and perspectives throughout to help shape the measure, including their likes/dislikes, queries and concerns. Item development took all feedback into account, which attests to the relevancy of the final set of items. The majority of items were positively phrased to protect responder self-esteem (Waters *et al.* 2009) and the negatively worded items were judged acceptable by the parent caregivers because they were frank and direct descriptions of very complex difficulties. These aspects of the development of QI-Disability ensure that the measure is relevant, understandable and acceptable to its users (Brod *et al.* 2009). Finally, the length of the measure was condensed from 50 to 32 items, enabling responder completion within approximately 10-15 minutes.

Measuring QOL is considered to be a process which requires an evaluation of a person's perceptions of their own satisfaction or happiness (Verdugo *et al.* 2005). For children with ID, limited communication and cognitive ability may impede individuals from understanding and identifying specific factors contributing to their personal happiness, and may require a parent caregiver to observe their daily lives and consequently interpret important wants and needs on behalf of their children. Due to impairments in communication and intellectual functioning, children with ID were not able to self-report during this phase of the study. A next step for this research would be to sample children from the ID population for which the instrument was specifically designed and adapt the questionnaire for the purpose of self-reporting. For example, questions addressing individual feelings and activity preferences could be selected from QI-Disability to create a streamlined tool which facilitates testing using a variety of communication methods (e.g. eye gaze technology).

Establishing content validity is part of measure development because it is the process of identifying patient-relevant issues and generating items that reflect those issues. We propose that our development process provides satisfactory evidence of the content validity of QI-Disability. Additional evaluations are necessary to continue validation and determine responsiveness to change.

REFERENCES

- Albrecht, G. & Devlieger, P. (1999) The disability paradox: high quality of life against all odds. *Soc Sci Med*, **48**, 977-988.
- Bigby, C. (2012) Social inclusion and people with intellectual disability and challenging behaviour: A systematic review. *Journal of Intellectual & Developmental Disability*, **37**, 360-374.
- Brod, M., Tesler, L. E. & Christensen, T. L. (2009) Qualitative research and content validity: developing best practices based on science and experience. *Quality of Life Research*, **18**, 1263-1278.
- Charters, E. (2003) The use of think-aloud methods in qualitative research. An introduction to think-aloud methods. *Brock Education*, **12**, 68-82.
- Davis, E., Reddihough, D., Murphy, N., Epstein, A., Reid, S. M., Whitehouse, A., Williams, K., Leonard, H. & Downs, J. (2017) Exploring quality of life of children with cerebral palsy and intellectual disability: What are the important domains of life? *Child Care Health Dev*, **43**, 854-860.
- Davis, E., Shelly, A., Waters, E., Mackinnon, A., Reddihough, D., Boyd, R. & Graham, H. K. (2009) Quality of life of adolescents with cerebral palsy: perspectives of adolescents and parents. *Developmental Medicine and Child Neurology*, **51**, 193-199.
- Downs, J., Jacoby, P., Leonard, H., Epstein, A., Murphy, N., Davis, E., Reddihough, D., Whitehouse, A. & Williams, K. (2018) Psychometric properties of the Quality of Life Inventory-Disability (QI-Disability) measure. *Qual Life Res*.
- Einfeld, S. L., Ellis, L. A. & Emerson, E. (2011) Comorbidity of intellectual disability and mental disorder in children and adolescents: A systematic review. *Journal of Intellectual & Developmental Disability*, **36**, 137-143.
- Epstein, A., Leonard, H., Davis, E., Williams, K., Reddihough, D., Murphy, N., Whitehouse, A. & Downs, J. (2016) Conceptualizing a quality of life framework for girls with Rett syndrome using qualitative methods. *American journal of medical genetics. Part A*, **170A**, 645-653.
- Epstein, A., Whitehouse, A., Williams, K., Murphy, N., Leonard, H., Davis, E., Reddihough, D. & Downs, J. (2019) Parent-observed thematic data on quality of life in children with autism spectrum disorder. *Autism*, **23**, 71-80.
- Ericsson, K. A. & Simon, H. A. (1980) Verbal reports as data. *Psychological Review*, **87**, 215-251.
- Fehr, S., Wilson, M., Downs, J., Williams, S., Murgia, A., Sartori, S., Vecchi, M., Ho, G., Polli, R., Psoni, S., Bao, X., De Klerk, N., Leonard, H. & Christodoulou, J. (2013) The CDKL5 disorder is an independent clinical entity associated with early-onset encephalopathy. *Eur J Hum Genet*, **21**, 266-273.
- Felce, D. & Perry, J. (1995) Quality of life: Its definition and measurement. *Research in Developmental Disabilities*, **16**, 51-74.
- Leonard, H., Cobb, S. & Downs, J. (2017) Clinical and biological progress over 50 years in Rett syndrome. *Nat Rev Neurol*, **13**, 37-51.

- Leonard, H., Glasson, E., Nassar, N., Whitehouse, A., Bebbington, A., Bourke, J., Jacoby, P., Dixon, G., Malacova, E., Bower, C. & Stanley, F. (2011) Autism and intellectual disability are differentially related to sociodemographic background at birth. *PLoS ONE*, **6**, e17875-e17875.
- Leonard, H., Petterson, B., Bower, C. & Sanders, R. (2003) Prevalence of intellectual disability in Western Australia. *Paediatric and Perinatal Epidemiology*, **17**, 58-67.
- Murphy, N., Epstein, A., Leonard, H., Davis, E., Reddihough, D., Whitehouse, A., Jacoby, P., Bourke, J., Williams, K. & Downs, J. (2017) Qualitative analysis of parental observations on quality of life in Australian children with Down syndrome. *Journal of Developmental and Behavioral Pediatrics*, **38**, 161-168.
- Narayanan, U. G., Fehlings D Fau - Weir, S., Weir S Fau - Knights, S., Knights S Fau - Kiran, S., Kiran S Fau - Campbell, K. & Campbell, K. (2006) Initial development and validation of the Caregiver Priorities and Child Health Index of Life with Disabilities (CPCHILD). *Developmental Medicine & Child Neurology*, **48**, 804-812.
- Neul, J. L., Kaufmann, W. E., Glaze, D. G., Christodoulou, J., Clarke, A. J., Bahi-Buisson, N., Leonard, H., Bailey, M. E. S., Schanen, N. C., Zappella, M., Renieri, A., Huppke, P., Percy, A. K. & Rettsearch, C. (2010) Rett Syndrome: Revised Diagnostic Criteria and Nomenclature. *Annals of Neurology*, **68**, 944-950.
- Ravens-Sieberer, U., Auquier, P., Erhart, M., Gosch, A., Rajmil, L., Bruil, J., Power, M., Duer, W., Cloetta, B., Czemy, L., Mazur, J., Czimbalmos, A., Tountas, Y., Hagquist, C. & Kilroe, J. (2007) The KIDSCREEN-27 quality of life measure for children and adolescents: psychometric results from a cross-cultural survey in 13 European countries. *Qual Life Res*, **16**, 1347-1356.
- Roizen, N. J. & Patterson, D. (2003) Down's syndrome. *Lancet*, **361**, 1281-1289.
- Schalock, R., Bonham, G. & Verdugo, M. (2008) The conceptualization and measurement of quality of life: implications for program planning and evaluation in the field of intellectual disabilities. *Evaluation and Program Planning*, **31**, 181-190.
- Tangarorang, J., H., L., A., E. & Downs, J. (Forthcoming) A framework for understanding quality of life domains in individuals with the CDKL5 Deficiency Disorder *American Journal of Medical Genetics Part A*.
- Varni, J. W., Burwinkle Tm Fau - Berrin, S. J., Berrin Sj Fau - Sherman, S. A., Sherman Sa Fau - Artavia, K., Artavia K Fau - Malcarne, V. L., Malcarne Vl Fau - Chambers, H. G. & Chambers, H. G. (2006) The PedsQL in pediatric cerebral palsy: reliability, validity, and sensitivity of the Generic Core Scales and Cerebral Palsy Module. *Developmental Medicine & Child Neurology*, **48**, 442-449.
- Verdugo, M. A., Schalock, R. L., Keith, K. D. & Stancliffe, R. J. (2005) Quality of life and its measurement: important principles and guidelines. *JIDR. Journal of Intellectual Disability Research*, **49**, 707-717.
- Waters, E., Davis E Fau - Ronen, G. M., Ronen Gm Fau - Rosenbaum, P., Rosenbaum P Fau - Livingston, M., Livingston M Fau - Saigal, S. & Saigal, S. (2009) Quality of life instruments for children and adolescents with neurodisabilities: how to choose the appropriate instrument. *Developmental Medicine & Child Neurology*, **51**, 660-669.

- Waters, E., Davis, E., Mackinnon, A., Boyd, R., Graham, H. K., Kai Lo, S., Wolfe, R., Stevenson, R., Bjornson, K., Blair, E., Hoare, P., Ravens Sieberer, U. & Reddihough, D. (2007) Psychometric properties of the quality of life questionnaire for children with CP. *Developmental Medicine and Child Neurology*, **49**, 49-55.
- Waters, E., Maher, E., Salmon, L., Reddihough, D. & Boyd, R. (2005) Development of a condition-specific measure of quality of life for children with cerebral palsy: empirical thematic data reported by parents and children. *Child: Care, Health & Development*, **31**, 127-135.
- World Health Organization (2001) *International Classification of Functioning, Disability and Health: ICF*, World Health Organisation, Geneva.

Box 1. The think-aloud interview schedule.

The purpose of today's call is for you [the parent] to complete a draft of our new Quality of Life questionnaire out aloud while sharing with me your thoughts and opinions about each question. We will be recording the interview for research purposes and your responses will be kept confidential.

For each item, please

1. Think out aloud when generating an answer to each question about [your daughter/son].
2. Say why you gave them that rating.
3. How would you restate this question in your own words?

After reviewing each item:

1. Was there anything about the questionnaire that you were confused about or did not understand?
2. Do you think that there was anything that the questionnaire missed that you would have liked to see included?

Table 1. Sample questionnaire items revised following think-aloud parent interviews.

Domain	Original item	Revised item
Health	Been alert during the day	Been alert and aware during the day
Comfort	Been unsettled for too long	Been unsettled without an apparent reason
Behaviour and emotions	Become withdrawn	Become withdrawn with a low mood
Communication	Expressed happiness when others understood what they wanted	Expressed happiness when they were understood
Movement	Enjoyed being in control of their body movements (e.g. sitting, walking)	Enjoyed feeling steady and stable during physical activities (e.g. sitting, standing, bike riding)
Routines	Helping to accomplish tasks (e.g. dressing, feeding)	Helped to complete routine activities (e.g. dressing, feeding)
Family and friends	Greeted people verbally or nonverbally (e.g. eye contact)	Initiated greetings with people verbally or nonverbally (e.g. eye contact)
Leisure and recreation	Enjoyed going to community events (e.g. party, sports, theatre)	Enjoyed going on outings in the community (e.g. shopping, party, sports, theatre)
Nature and the outdoors	Enjoyed spending time outdoors / Expressed pleasure when in contact with natural elements (e.g. water, grass, wind, sunshine)	Enjoyed spending time outdoors (e.g. contact with water, grass, wind, sunshine)

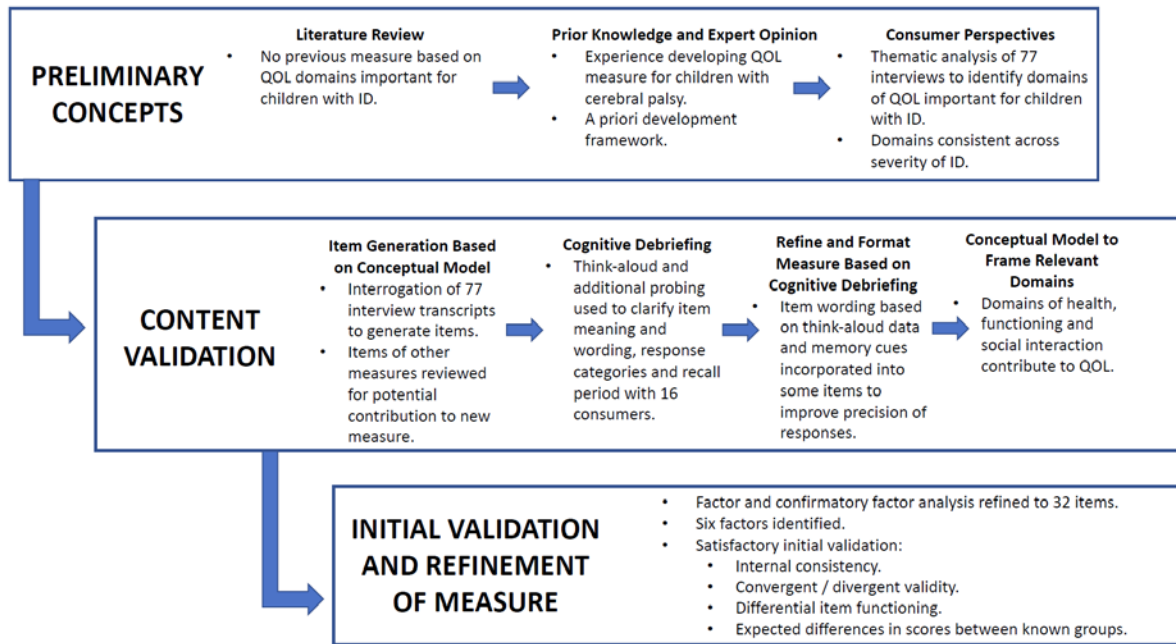


Figure 1. Patient-reported outcomes development processes including content validation procedures.

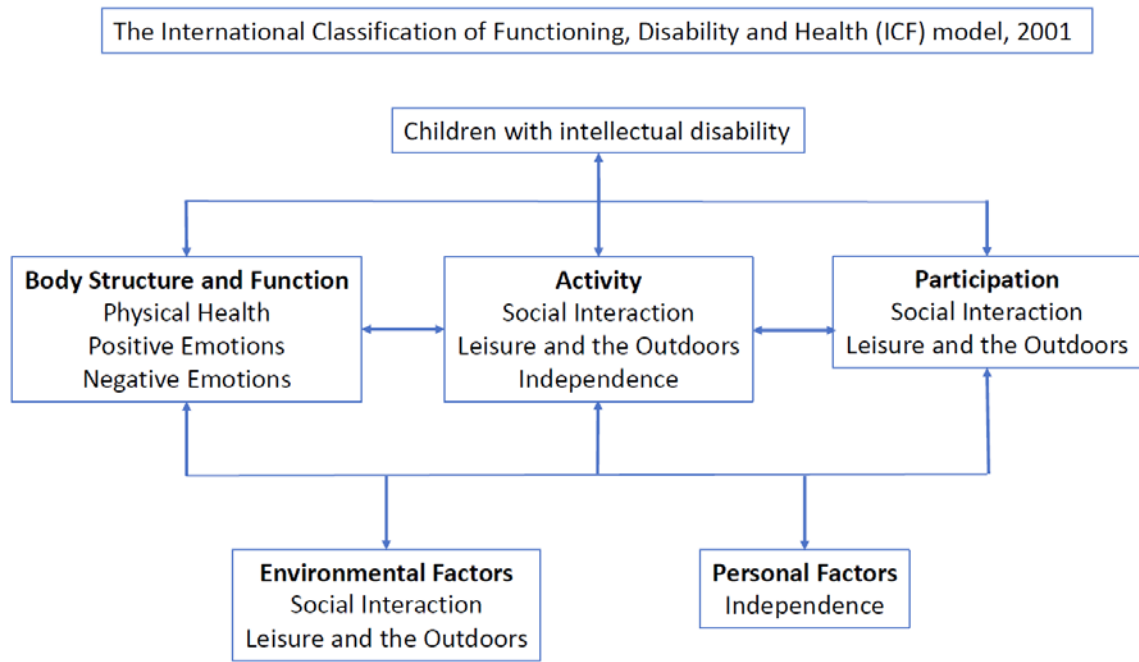


Figure 2. Mapping of the factors of Quality of Life Inventory – Disability to the International Classification of Functioning, Disability and Health framework.

PRELIMINARY CONCEPTS

- Literature Review**
 - No previous measure based on QOL domains important for children with ID.
- Prior Knowledge and Expert Opinion**
 - Experience developing QOL measure for children with cerebral palsy.
 - A priori development framework.
- Consumer Perspectives**
 - Thematic analysis of 77 interviews to identify domains of QOL important for children with ID.
 - Domains consistent across severity of ID.

CONTENT VALIDATION

- Item Generation Based on Conceptual Model**
 - Interrogation of 77 interview transcripts to generate items.
 - Items of other measures reviewed for potential contribution to new measure.
- Cognitive Debriefing**
 - Think-aloud and additional probing used to clarify item meaning and wording, response categories and recall period with 16 consumers.
- Refine and Format Measure Based on Cognitive Debriefing**
 - Item wording based on think-aloud data and memory cues incorporated into some items to improve precision of responses.
- Conceptual Model to Frame Relevant Domains**
 - Domains of health, functioning and social interaction contribute to QOL.

INITIAL VALIDATION AND REFINEMENT OF MEASURE

- Factor and confirmatory factor analysis refined to 32 items.
- Six factors identified.
- Satisfactory initial validation:
 - Internal consistency.
 - Convergent / divergent validity.
 - Differential item functioning.
 - Expected differences in scores between known groups.

Figure 1. Patient-reported outcomes development processes including content validation procedures.

The International Classification of Functioning, Disability and Health (ICF) model, 2001

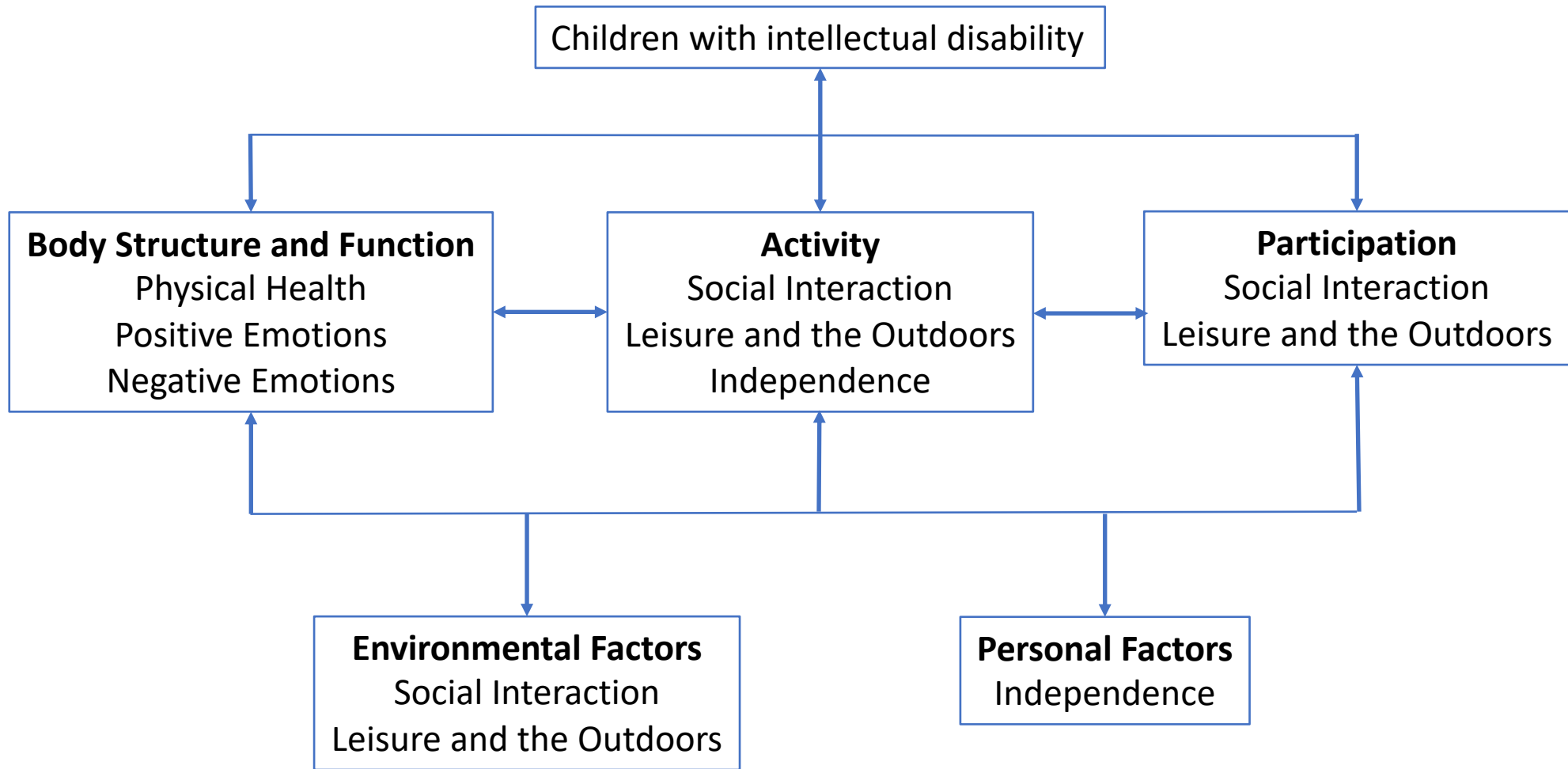


Figure 2. Mapping of the factors of Quality of Life Inventory – Disability to the International Classification of Functioning, Disability and Health framework.