Development of a condition-specific measure of quality of life for children with cerebral palsy: empirical thematic data reported by parents and children

E. Waters,*†‡ E. Maher,*†‡ L. Salmon,*†‡ D. Reddihough†‡§ and R. Boyd†‡¶

*School of Health and Social Development, Faculty of Health and Behavioural Sciences, Deakin University, Melbourne
†Department of Paediatrics, University of Melbourne,
‡Murdoch Children’s Research Institute, Melbourne,
§Department of Child Development & Rehabilitation, Royal Children’s Hospital, and
¶Department of Neonatal Neurology, Royal Children’s Hospital, Victoria, Australia

Accepted for publication 24 June 2004

Abstract

Background  Although there is increasing recognition that quality of life (QOL) and health-related quality of life (HRQOL) are important outcome variables in clinical trials for children with cerebral palsy, there are substantial limitations in existing measures of QOL. This study identify themes of QOL for children with cerebral palsy and their parents to guide the development of a new condition-specific QOL scale.

Methods  A qualitative study of parent and child views on QOL composition was conducted, using a grounded theory framework. Families participated in semistructured interviews on QOL until thematic saturation was reached (n = 28 families).

Results  Overall, 13 themes emerged from the interviews: physical health, body pain and discomfort, daily living tasks, participation in regular physical and social activities, emotional well-being and self-esteem, interaction with the community, communication, family health, supportive physical environment, future QOL, provision of, and access to services, financial stability, and social well-being.

Conclusions  Research with parents and children with cerebral palsy, representative of severity across the disease spectrum and socio-economic status, reinforced and expanded on the traditional themes that have underpinned QOL measurement development. This has implications not only for the development of a new QOL scale for children with cerebral palsy, but also for clinical interventions and community care management.

Cerebral palsy is the most common physical disability in childhood, occurring in 2–2.5 per thousand live births (Stanley et al. 2000). Cerebral palsy is a ‘disorder of movement and posture due to a defect or lesion of the immature brain’ (Bax 1964). Treatment of the motor disorder alone may involve a range of therapies and interventions such as physiotherapy, the use of orthoses, orthopaedic surgery and medications for spasticity such as Botulinum toxin A (BTX-A), and oral and intrathecal baclofen (Boyd & Hays 2001). Over the past 10 years, there have been increasing efforts to obtain empirical-based evidence to guide practice in the field. Outcome measures that have been used...
include spasticity rating scales, tests of gross and fine motor skills to assess changes in function, and gait analysis to demonstrate alterations in walking patterns. More recently quality of life (QOL) and health-related quality of life (HRQOL) have become core outcome variables in clinical trials (Bjornson & McLaughlin 2001). However, limitations in existing measures of QOL for children with cerebral palsy have spurred the development of a new condition-specific QOL scale for children with cerebral palsy (Liptak et al. 2001).

The area of QOL measurement for children remains underdeveloped and marred by a lack of consensus in the definition of QOL and HRQOL. There is consensus that the language and content of children’s QOL measures need to be appropriate to children’s experience and development, and therefore researchers have been reluctant to simply apply the principles and definitions of adult QOL to child QOL (Gerharz et al. 2003).

The major problem with many definitions of QOL and HRQOL is that they are often complex and difficult to operationalize (Eiser & Morse 2001). For example, QOL has been defined by the World Health Organization (WHO) as ‘the individual’s perception of their position in life, in the context of culture and value systems in which they live and in relation to their goals, expectations, standards and concerns’ (World Health Organization 1993). Definitions of HRQOL are similarly unclear, such as ‘a rubric, encompassing various aspects of personal experience, including physical and psychological health, cognitive factors, social role performance, and general life satisfaction’ (Warchausky et al. 2002).

A clearer conceptualization of QOL and HRQOL, developed by Spilker and Revicki (1996), is that HRQOL is a subset of QOL. HRQOL refers to an individual’s perception of his or her health (Irrgang & Anderson 2002), which as defined by the WHO, consists of physical well-being, mental well-being, and social well-being (World Health Organization 1948). QOL refers to satisfaction with a variety of domains, including health domains and non-health domains. Whilst there is little agreement on these non-health domains for children, if HRQOL is considered a subset of QOL, relevant non-health domains for children may include home environment and parental support (Juniper et al. 1996).

Two types of scales to measure QOL are commonly recommended and available: generic and condition-specific. Generic scales are designed to be applicable to all population subgroups and are useful for comparing outcomes of subgroups (Bjornson & McLaughlin 2001). Condition-specific scales are designed to be applicable to one group, and are useful to detect small changes in a condition (Bjornson & McLaughlin 2001).

Both generic and condition-specific scales have been used in the past to measure the QOL/HRQOL of children with cerebral palsy. A commonly employed generic scale is the Child Health Questionnaire (CHQ; Landgraf et al. 1996). Condition-specific scales include the Lifestyle Assessment Questionnaire (LAQ; Mackie et al. 2002), the modified Caregiver Questionnaire (CQ; Schneider et al. 2001) and the Pediatric Outcomes Data Collection Instrument (PODCI; Daltroy et al. 1998). Each of these measures, although appearing to be adequate outcome measures for children with cerebral palsy, has substantial limitations if they are used as outcome measures of QOL or HRQOL for children with cerebral palsy in clinical trials.

Generic scale: the CHQ

Many researchers have relied on the CHQ to measure QOL/HRQOL of children with cerebral palsy (Houlihan et al. 2001; Schneider et al. 2001; Vitale et al. 2001; McCarthy et al. 2002; Warchausky et al. 2002; Wake et al. 2003). The CHQ is a generic child health instrument that measures physical functioning, behaviour, mental health, general health, social and family functioning, family cohesion, self-esteem, pain and the impact of health issues on parental time and emotions (Landgraf et al. 1996). Generic measures such as the CHQ have been shown to be inordinately useful in establishing a profile of child health across multiple health domains that align with the WHO definition of health (Waters et al. 2000a; Waters et al. 2000b; Wake et al. 2003). Although it has been argued that some of the questions (i.e. playing soccer or bike riding) may be irrelevant for children with severe cerebral palsy (Schneider et al. 2001), the CHQ has
been shown to have adequate psychometric properties for Australian children with cerebral palsy (Wake et al. 2003). However, it must be recognized that the CHQ has not yet been extensively tested in intervention trials, and therefore it remains uncertain whether it is responsive to change over time or following interventions (Boyd et al. 2003; Wake et al. 2003).

Condition-specific scales: the CQ

The CQ was developed in 1990 at the Rehabilitation Institute of Chicago, USA for a defined population of children with spastic quadriplegic cerebral palsy who were to undergo selective dorsal rhizotomy. The CQ assesses the caregiver’s difficulties and satisfaction with their child’s progress in four areas: personal care, positioning/transferring, comfort and interaction (Schneider et al. 2001).

The major problem with the CQ, one that is also evident with the CHQ and PODCI, is that many of the items in the scale measure the absence of health difficulties or limitations rather than well-being. For example, the CQ assesses difficulties with daily personal care, and difficulties with participating in sports. If it is assumed that QOL is measured by a presence of well-being as opposed to an absence of ill being then measures need to clearly work within this model. Whilst this underlying assumption has not been well discussed in the child health or pediatric literature, researchers of the adult literature, propose that ill being is not low well-being, and the absence of ill being is not high well-being (Argle 1996).

Condition-specific scale: the LAQ

The LAQ is a condition-specific scale that measures physical independence, clinical burden, mobility, schooling, economic burden and social integration (Mackie et al. 1998, 2002). Little research has been conducted on this measure; however, analyses of a small sample of children with cerebral palsy suggest that the scale is reliable (over a 4-year gap), and valid (when a measure of disability is used as a benchmark; Mackie et al. 1998).

An important philosophical concern for the LAQ, but also the CQ and PODCI, is the lack of a child self-report version. QOL is inherently the subjective experience of the individual (Eiser & Morse 2001), and thus it is important to elicit children’s views of their QOL if they have the ability and desire (Eiser et al. 2000; Ronen et al. 2003). A parent proxy scale of QOL is necessary as a result of children’s level of vocabulary and language (Stewart 2000), however, it must be recognized that proxy ratings are not a substitute for individual ratings (Eiser et al. 2000).

Condition-specific scale: the PODCI

The PODCI measures comorbidity index, upper extremity and physical function, transfers and basic mobility, sports and physical function, pain/comfort, expectations and happiness (Daltroy et al. 1998). The PODCI has been shown to be reliable, valid (using the CHQ physical functioning scale as a benchmark) and sensitive to change for a large sample of children with musculoskeletal disorders (Daltroy et al. 1998). One concern with the PODCI is that it focuses on physical health and functioning and excludes other potentially important domains such as social well-being. The PODCI’s focus on physical health and functioning is warranted given that the PODCI does not claim to measure QOL or HRQOL; it is simply that other researchers have used it for that purpose. The PODCI claims to measure functional status, and assesses what children can do, rather than their level of well-being. Other scales that have been misused include the CHQ, which measures health status, and the LAQ, which measures the impact of disability. The fact that researchers are using these scales to measure QOL/HRQOL clearly suggests that: (1) there is a lack of understanding of QOL; and (2) there is a lack of scales to measure QOL in children with cerebral palsy. This had led researchers to conclude that there is an internationally recognized need for a sensitive and discriminate measure that can be used to evaluate the effectiveness of interventions (Bjornson & McLaughlin 2001).

In designing a scale of QOL, one of the questions that requires consideration is which domains constitute QOL for children with cerebral palsy and their families. Although past scales (CHQ, PODCI) have measured domains that were decided a priori
by researchers and clinicians, there is increasing recognition that families and children need to be consulted in this process (Bjornson & McLaughlin 2001). Qualitative research is the most suitable design for exploratory research, and thus for the purpose of researching the underpinning elements of QOL (Ronen et al. 2001).

This paper presents the results of a qualitative study conducted to identify the major themes of QOL for children with cerebral palsy and their parents. The study employed a grounded theory approach, which refers to the generation of theory from data (Glaser & Strauss 1967). This method is based on the proposal that pre-existing theory may influence the research process, and impede the development of new theoretical formulations (Strauss & Corbin 1990). The themes identified in this study will then be used to inform the first international condition-specific measure of QOL for children with cerebral palsy.

**Methods**

**Participants**

A sample of 42 families was purposely selected from the Victorian Cerebral Palsy Register maintained by the Department of Child Development and Rehabilitation at the Royal Children’s Hospital Melbourne (Dite et al. 1995). The sample was intended to be representative of age, functional severity, socio-economic status and geographical location. Functional severity was classified by the Gross Motor Functional Classification System (Palisano et al. 1997). Levels I and II were termed mild impairment with high level mobility, Levels III and IV were termed moderate impairment limited mobility with some antigravity postural stability and Level V was termed severe impairment without independent antigravity function. Socio-economic status was based on parental occupation collected from the Victorian Cerebral Palsy Register and other details known to the treating paediatrician. Geographic location was based on postcode and classified according to Australia Post classification of rural and metropolitan regions.

Twenty-eight families participated in the study, contributing data until saturation was reached. Three families declined to be involved in the study, and 11 families either did not respond to the initial letters sent out or were not contactable by four subsequent follow-up telephone calls. It is unlikely that this response rate will affect the validity of the responses, as there were sufficient participants within each disease category (e.g. mild and rural, severe and aged 4–8 years, etc.) and socio-economic background.

Of the 28 families who participated in the study, 16 were parents of children aged 4–8 years and 12 were parents of children aged 9–12 years. Out of 12 children that could possibly be involved in self-report, five children with mild impairment were able to be involved. The remaining were either unable to communicate because of moderate/severe impairment (n = 5), or because the times chosen by the parents prevented child participation (n = 2). Although only five children were involved in the interviews, the themes were exhausted for children with mild impairment.

In total, 10 children were classified as having mild impairment, nine were classified as having moderate impairment and nine were classified as having severe impairment. The majority of children came from two-parent families (n = 24), and had one or more siblings (n = 26; including twins n = 6). Sixteen families were classified with a high socio-economic status background and 12 were from a low socio-economic status background (based on Registry data). Half of the families were located in the metropolitan region, and half were located in rural Victorian areas.

**Materials**

Interview questions were derived from a review of the QOL literature. The questions were designed to prompt discussion and elicit new information relevant to QOL (refer to Appendix 1).

**Procedure**

Ethics approval for the study was received from the Research in Humans Ethics Committee of the Royal Children’s Hospital. Interviews were conducted at a time convenient to the parent(s), either in their home, at their workplace or at the Royal
Children's Hospital. Within the 4–8-year age group, only the parent(s) were interviewed, however, within the 9–12 years age group, both parent(s) and child were interviewed when possible. The inclusion of children in the interview depended on the parent’s consent and the child’s ability to communicate and willingness to be involved. The interviews lasted approximately 30 min. The interviews were transcribed by one researcher, and themes were extracted by three researchers. Agreement on key themes was achieved by discussion.

Results

The interviews with children and parents provided significant information about the daily lives of children with cerebral palsy, and their families. Thirteen themes of QOL were extracted from discussions with parents and children about their child’s life. According to families, to have a high QOL, children need to have the following:

- Physical health – refers to adequate gross motor skills and fine motor skills, the ability to use aids (if required), and good overall physical health.
- Body pain and discomfort – refers to absence of stiffness and soreness in joints, and pain associated with therapy.
- Daily living tasks – refers to the ability to carry out normal daily living tasks including dressing, feeding and toileting and being independent.
- Participation in regular physical and social activities – refers to participating in school activities, sporting activities and community activities.
- Emotional well-being and self-esteem – refers to being happy, being able to achieve goals, and being satisfied with one’s body and emotions.
- Interaction with the community – refers to being socially accepted, being a valued member of the community, and being treated ‘normally’.
- Communication – refers to having good communication skills with family, peers, and people in the general community.
- Family health – refers to good parental emotional health, good family relations, and few restrictions on the family to go out socially.
- Supportive physical environment – refers to a supportive school environment, family environment, and community environment. These environments need to have the required equipment and devices.
- Future QOL – refers to having the opportunities to do everything that they desire, being able to do things as well as their peers, and being able to make choices in their life.
- Provision of and access to services – refers to having access to therapy, respite, and having the support required.
- Financial stability – refers to the earning capacity of parents, and the ability to cover the expenses of equipment and treatment.
- Social well-being – refers to the ability to interact with family members, peers, and people in the general community.

Discussion

Consultations with children with cerebral palsy and their parents about their QOL provided some insight into the daily lives of such families; their good times, their bad times, their challenges and their desires. The findings confirmed but also enhanced the previous evidence-based impression of the key themes of QOL. All domains of health identified by the WHO, including physical well-being (i.e. physical health), mental well-being (i.e. emotional well-being and self-esteem) and social well-being emerged as themes.

These were complemented by themes that were specific to children, such as family health, and themes that were specific to cerebral palsy, including body pain and discomfort, daily living tasks, communication and future QOL. The remaining themes differed somewhat to the traditional QOL literature. These themes, including satisfaction with access to services, supportive physical environments, financial stability of the family, and acceptance in the broader community, appear to address the practicalities of having a child with cerebral palsy. These findings have implications for the development of a QOL scale for children with cerebral palsy, but also for clinical interventions and community management.
Implications for a QOL scale for children with cerebral palsy

The findings suggest that a new QOL scale for children with cerebral palsy should examine the traditional domains of QOL (e.g., physical well-being), the condition-specific domains of QOL (e.g., body pain and discomfort) and the more practical domains of QOL (e.g., financial stability).

One potential criticism of using the results of this study to guide the development of a QOL scale is that some of the themes mentioned by parents/children may be inappropriate for a QOL scale. This study sought the opinions of children with cerebral palsy and their parents as there is increasing recognition that their contribution is important. However, it must be recognized that there are few guidelines to determine how to incorporate the opinions of parents and children with the opinions of experts. Indeed, it is questioned whether more emphasis should be placed on the opinions of the parent/child or the opinions of the researchers/clinicians, and exactly how conflict between the two groups should be resolved.

It is possible that parents may report predictors of QOL, rather than domains of QOL. Predictors of QOL are variables that are related to QOL, such as personality, whereas domains of QOL are components of QOL, such as social well-being. For example, in this study parents and children mentioned body pain as being important to their QOL, yet researchers have suggested that domains of QOL cannot be negative (Hagerty et al. 2001). They propose that negative domains, such as pain can only have a neutral effect on QOL, and never a positive influence (Hagerty et al. 2001). However, even if this were clearly accepted, it is problematic to exclude body pain from the scale, as it is clearly important to the parents and children.

One way to handle this inconsistency is to develop items for all of the themes reported by parents and children, test them in a pilot study and examine their psychometric properties. If after the pilot study and further consultation with experts, some of the themes were deemed to be predictors of QOL rather than domains, it is recommended that the themes still be included in the scale but that the score is not aggregated into the total QOL score. If this procedure is followed, the resulting scale would include areas that are important to parents and children whilst still recognizing the theoretical developments that have been made in the QOL literature.

In developing items to measure each theme, the interview transcripts will be examined further. Based on these interviews, items will be developed to measure each theme in alignment with the social model of health (Learmonth 1985; Najman 2001). For example, for the theme of social well-being, the items may refer to relationship with parents, relationship with siblings, relationship with peers and relationship with people in the community.

In addition to considering the content of the items, there are many other areas of scale development that require significant attention. These include identifying the stem of the item (i.e. ‘happy is your child with’ or ‘how would you rate your child’s abilities’), identifying the response scale, investigating whether the items need to be weighted, and considering the administration issues that are particularly pertinent for children with intellectual disabilities (acquiescence, respondent competence; Cummins, 2004).

One further important consideration for this proposed instrument is involvement of children. In the current study, issues associated with the age of the children and the structure of the questions limited the involvement of children to those with mild to moderate impairment. Future phases of the instruments’ development will strive to ensure children across the spectrum of impairment are engaged, and an appropriate instrument ensues.

Implications for clinical interventions and community management

The findings from this study not only have implications for the development of a QOL scale, but also for clinical interventions and community management. Specifically they suggest that in order to improve the QOL of children with cerebral palsy, clinical trials need to utilize scales that consider among others, the child’s physical, emotional, social, and family health, as well as their financial stability, community acceptance and their physical environment.
It may be argued that it is unnecessary for clinical trials to measure some of the more 'practical' themes of QOL before and after intervention, as they are not easily amenable to change. For example, an intervention assessing the effectiveness of a particular medication may not be expected to alter the child's community environment. However, it must be recognized that if these 'practical themes' were excluded from a clinical study of QOL, the study cannot claim to increase total QOL. Instead, they could only report the changes in individual domains (e.g. physical well-being, emotional well-being).

It is recommended however, that rather than excluding domains of QOL that are less amenable to change, researchers, clinicians, social workers, psychologists and community workers collaborate to ensure that they can understand and address all areas of QOL. For example, they can assist parents with access to affordable and accessible respite, or they can advocate for supportive school environments.

Conclusion

There now exists an empirical evidence-base for the important aspects of QOL for children with cerebral palsy. This data can be used to develop a condition-specific measure, and concurrently, aid in clinical interventions and community management.

Acknowledgements

Funding: NHMRC grant number 284 514, Telstra communications PLC, Murdoch Children’s Research Institute Cerebral Palsy Theme grant, Murdoch Children’s Research Institute salary support grant, NHMRC postdoctoral fellowship.

References


Houlihan, C. M., O’Donnell, M., Conaway, M., Stevenson, R. D. & North American Growth in Cerebral Palsy project (2001) Bodily pain and...


Appendix 1 – focus group questions

1 Firstly, when everything is going great for you/your child, what sorts of things are happening in his/her life (and your family life)?
2 What about when things are not so good?
3 What sorts of activities make your child feel like good about him/herself?
4 What, if any limitations does your child have? If so, how do they affect his/her life? How do they make him/her feel?
5 What is a good quality of life?
6 What kinds of things do you (and your child) need for a good quality of life?
7 Tell me about cerebral palsy?
8 How does it (cerebral palsy) affect yours and your child’s life?
   • prompt: Good and bad? school? family relationships? other social relationships? general health? What kind of things keep your child healthy?
9 Thinking about the impact your child’s cerebral palsy has:
   • tell me about the relationships of family members or general family cohesion?
   • tell me about the impact of your child’s cerebral palsy on the family unit as a whole? (prompt – family activities, being able to do things as a family)
   • tell me about your child’s social relationships (prompt – at home, at school);
   • about school – does he/she have any limitations (over 5 only).
10 What about general health issues – day to day coping strategies, general feeling of wellbeing?
11 Are there any other issues you would like to raise?