

Subjective quality of life in children with intellectual impairment – how can it be assessed?

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The relationships between quality of life (QoL), participation, and environment are being studied within a multicentre European study of 8- to 12-year-old children with cerebral palsy (CP; Study of PARticipation of Children with cerebral palsy Living in Europe [SPARCLE], www.ncl.ac.uk/sparcle/). The international group was determined that children with CP who also had intellectual impairment (IIm) should not be excluded from the study, but the group realized that there was little information available on how to assess subjective QoL in children with IIm. This annotation reviews and discusses the literature on conceptual and methodological issues in relation to such assessment.

The meaning of the concept of QoL is much debated. A critical notion is that there exists an underlying universal standard of the quality of human life, even though each individual has a unique perception of the quality of their life, influenced by their cultural environment, past experience, personal values, and aspirations. QoL is a multidimensional construct encompassing several core domains, generally identified as material conditions, physical status and functional abilities, social interactions, and emotional well-being.¹ Each of these domains can be considered from objective and subjective perspectives. Broadly, an objective assessment of QoL focuses on the physical and social activity and material life conditions of an individual, whereas a subjective perspective places emphasis on each individual's perception of their quality of life. There is now ample evidence that the relation between objective living

conditions and subjective well-being is weak,² hence the importance of assessing both perspectives.

A number of specific issues arise in child QoL assessment due to age and developmental level. Compared with adults, children have less experience on which to base their interpretation of events. Their less-developed cognitive skills make comprehension and articulation of abstract concepts more difficult and affect their ability to recall behaviour and emotions from defined time periods, as required by most questionnaires. Furthermore, there is a need for instruments based on children's own interests and life values, rather than modified questionnaires developed by and for adults using adult conceptualizations of QoL.

Most recent studies aiming to assess the QoL of children with CP are mainly concerned with functional assessment of health status or objective QoL and do not pay specific attention to the difficulties of subjective assessment in children with associated IIm.³⁻⁵ Only a few explicitly discuss the latter issue and justify their choice of assessment methods.⁶⁻⁹ All the studies (except Hodgkinson et al.⁴) used proxy reports to evaluate QoL in children with CP, regardless of their intellectual ability.

Subjective QoL in children with IIm (with or without CP) has not been extensively assessed. A few studies have examined related subjective dimensions in children with IIm, such as pain¹⁰ or emotional and behavioural problems,¹¹ based on proxy reports. Self-reported fears¹² and anxiety¹³

were assessed and compared in children with and without IIM. In both studies, a formal procedure was set up to evaluate the children's cognitive ability before the assessment itself. The authors concluded that children with IIM can give valuable feedback regarding subjective aspects of their life, and that, consequently, they should be actively consulted regarding their internal state.

Methodological considerations for self-report in people with intellectual impairment

COMPREHENSION, COMMUNICATION SKILLS, AND INTERVIEWING TECHNIQUES

It is generally agreed that the order in which children without IIM master various types of question is relatively consistent (from factual 'yes–no' through pictorial and multiple-choice to abstract open-ended questions) and depends on cognitive development.¹⁴ Similarly, the responsiveness of adults and children with IIM is a stable, individual behaviour positively correlated with intelligence quotient (IQ) and highly dependent on the type of question asked.¹⁵ Some individuals have less difficulty dealing with simple formats, such as multiple-choice questions, than they have when responding to open-ended questions which require greater cognitive and linguistic capacities. For instance, 'yes–no' and pictorial questions can be answered non-verbally, by shaking the head or pointing. However, though closed questions allow high response rates, open and explorative questions obtain more valid responses.¹⁶ Content also influences responsiveness: questions involving concrete situations usually obtain greater response rates and more accurate information than abstract questions.¹⁵

Questions for children with or without IIM should be kept as basic as possible, using simple terms and syntax and active sentence constructions. Comprehension is further facilitated by keeping to a single core idea per question, rather than asking children directly to provide comparisons. In terms of readability, factors such as paragraph length, print size, clarity of instructions, and attractiveness of layout are important. QoL questionnaires should be administered in a familiar environment where the child feels at ease, is not distracted or under pressure, and is allowed ample completion time. QoL assessment has frequently been simplified by providing pictorial representations of response categories or of analogue scales (ranging from a very happy to a very sad face).^{17–20} Pictures engage children's interest and increase their understanding, and, thus, may help to obtain more meaningful responses. There is evidence that children as young as 4 years of age can provide reliable self-report data using such devices.²¹ Furthermore, visual response categories can all be viewed simultaneously, thereby avoiding premature acquiescence to the first of a list of alternatives.

The responsiveness of people with IIM often depends on external cues and care must be taken not to influence their answers by careless interviewing techniques. Antaki and Rapley²² showed that investigators frequently transform questions to adapt them to their respondents' limited cognitive abilities. One type of question distortion is the tendency to paraphrase linguistically complex terms, which introduces interrater variability and may alter the original meaning of the question. Second, for certain difficult questions, interviewers may feel the need to ask a preliminary question before coming to the 'real' question, which can be confusing for respondents with IIM. Third, questions are often followed by a list of

possible alternatives that the interviewer must enumerate, the problem being that people with IIM often speak after each alternative is given and thus give several different answers to the official question. There may also be conversational dynamics that cause interviewers to distort what they understand to be the answers to those questions. Where the respondent has correctly understood the question and answers immediately, an interviewer who continues to list the other alternatives will prompt the respondent to believe they have given the 'wrong' answer and, thus, change what they have said. Another source of distortion arises when the respondent replies with terms other than those stated in the QoL instrument or starts talking about some apparently irrelevant subject. In both cases, interviewers tend to continue to question the respondent and often distort the information offered. Antaki²³ also found that interviewers reformulate questions not only to lower the cognitive difficulty of the interview but in such a way as to increase the probability of the respondent obtaining a higher score. Although it is unintentional, this attitude goes against respondents' interests, as they may end up with high QoL scores on modified unambitious questions which would not be posed to people without IIM.

FORMAL COMPETENCY EVALUATIONS

Evaluating competency to comprehend and respond to questions is a difficult but necessary prerequisite of QoL assessment. In a comparative study of subjective QoL judgements of people with and without IIM,²⁴ interviewers explained the study procedure to each person before asking them five questions in order to establish understanding and consent: (1) What will I be talking to you about?, (2) How many times will I want to talk to you?, (3) Are there any good things about talking to me?, (4) Are there any bad things about talking to me?, (5) What can you do if you decide you don't want to talk to me anymore? People who did not respond appropriately to at least three of these questions were not considered able to participate. The issue of formal consent is different in the case of children, as it is the guardian's legal responsibility to make this decision for them. However, ensuring that the child understands and accepts the general process of the QoL evaluation is an important way of showing respect and encouraging cooperation. Cummins' Comprehensive Quality of Life Scale¹⁷ includes a formal pretesting protocol with three graded tasks designed to determine whether completion of the subjective axis is likely to be valid. First, the respondent has to order five wooden cubes according to size. The second task involves matching cube size to a visual ladder. In both tasks the protocol starts with two cubes and if the step is successfully completed, the task difficulty is increased to three then five cubes. Finally, the person is asked to identify something that is important to them and rank it on an importance scale (the ladder) in order to demonstrate their ability to use this scale in an abstract manner. Thus, the participant's ability to deal with a two-, three-, or five-step scale to measure importance is determined and the highest level of complexity successfully reached is adopted for the whole questionnaire. Only individuals who perform correctly on all three phases of the pretest are considered able to proceed with the subjective axis of the questionnaire. The possibility of adapting the scale complexity of the questionnaire according to the person's abilities is potentially problematic from a methodological point of view.

Although further research is required for paediatric

populations, this type of competency testing seems an essential procedure and should make it possible to obtain reliable and valid assessment for a significant proportion of children with IIm with sufficient communication skills. However, for children with profound IIm, proxy report remains the only realistic alternative.

Use of proxy reports

DIFFERENCES BETWEEN SUBJECT AND PROXY RATINGS

Various categories of 'significant others', such as family members, caregivers, and medical staff, have been asked to provide indirect assessments of a person's QoL. When proxy ratings are compared with those provided by the subjects, the assumption is that the subject is the 'truest' source of information regarding their QoL. Most studies find that relatives and clinicians have a tendency to underestimate a child's QoL compared with how the child rates it; also, there seems to be a lower level of agreement between proxies and children when dealing with the more subjective dimensions of QoL, e.g. social or emotional functioning and psychological domains, compared with more observable aspects, e.g. physical activity, functioning, and symptoms.²⁵⁻³⁰ Men, fathers (versus mothers), grandparents (versus parents), and older proxies report fewer chronic health conditions and fewer accidents.³¹ Compared with mothers, fathers rate their children as having fewer behavioural or psychiatric problems;³² in addition, mothers (but not fathers) who rate their own health as poor also tend to rate their child's QoL as poor.³³

Stancliffe³⁴ studied agreement between self-reports and caregiver proxy responses in 63 adults with IIm; he found substantial positive correlations and no significant difference between self and caregiver reports. However, a recent study of 93 adults with mild or moderate IIm showed that though the life-satisfaction reports of residents and caregivers were positively correlated, caregiver ratings were significantly higher than subject ratings.²⁰ As no such divergence was found between parents' and subjects' reports, it was suggested that parents should be selected in preference to caregivers when assessing life satisfaction.

One study reports that the level of concordance between child and proxy ratings of QoL tends to diminish with decreasing health status of the child.²⁷ However, a study of adults suggests that concordance of proxy ratings may be better for patients with very high or very low QoL than for patients with intermediate QoL, suggesting a U-shaped relation between the degree of subject-proxy agreement and QoL.³⁵ The authors suggest that this may also apply to those with severe impairment in whom we have to rely on proxy ratings. However, this makes the unlikely assumption that those with severe impairment necessarily have low QoL.

Measurement issues, such as the timing of assessments and varying interpretations of questions and response categories, may act as 'noise' and artificially deflate the level of subject-proxy agreement. Eiser and Morse²⁹ note that the terms used to describe social and emotional functioning domains vary greatly, whereas physical functioning is assessed with more homogeneous terms. Thus, lower levels of agreement between child and proxy ratings for subjective dimensions could be an artefact resulting from the way in which these domains are measured. Furthermore, concordance between parent and child ratings has most frequently been gauged using Pearson's product-moment correlations.

However, a good Pearson correlation does not necessarily provide an indication of actual agreement, as it disregards any systematic bias. To overcome this problem, intraclass correlation and comparison of group mean scores should also be used to determine the direction and magnitude of any such bias.

INTERPRETATION OF DIFFERENCES BETWEEN SUBJECT AND PROXY REPORTS

Interestingly, studies of people without IIm have a tendency to attribute divergence between subject and proxy reports to bias in the proxy ratings, whereas for people with IIm such differences are often interpreted as evidence of bias in the subjects' responses.²⁰

The difference between proxy and subject responses may be 'false' or erroneous and reflect biased information. Proxies may provide valid but incomplete accounts of QoL, as children's behaviour varies depending on the environment in which they operate. Professional staff may be influenced by prior hypotheses concerning children's well-being and consequently have a tendency to over-report behaviours that confirm their beliefs. In our own studies, we observed that caregivers of children with profound impairments noted changes in function and mood more precisely than staff working with children with a broad range of impairment levels, who tended to compare those with severe impairment with those with mild impairment. The impact of the subject's disabilities on the proxy respondent is also likely to have a strong influence on proxy reports;³⁶ parents' anguish may colour their perception of their child's life and lead them to over-report difficulties. However, children may also be responsible for 'false' differences. Children with chronic illnesses or long-term impairments may minimize reports of their symptoms of distress in the process of adapting to their illness or in order to avoid further medication. It has been shown that adolescents rated their own QoL substantially higher than their evaluation of a scenario describing their health status, whereas in a control group of healthy adolescents there was no significant difference between the self-rating and the scenario.³⁷ Hence coping strategies can also distort children's responses.

Discrepancies between subject and proxy ratings are not necessarily evidence of inaccuracy or bias in either data source. The differences may be 'real' and valid, reflecting each respondent's perspective. One study reported that health professionals placed greater value on children's mood, behaviour, and child and family function than on mobility and self-care, whereas parents and the general adult population had opposite views.³⁸ Therefore, it may be an advantage to obtain ratings from several proxies for an overall impression of a child's life. However, collecting multiple ratings raises methodological problems. For example, averaging scores defeats the purpose of obtaining different viewpoints, as precious information is lost. Another possibility is to determine the domains for which each respondent provides the most 'reliable' information and to combine these responses for an overall estimate of QoL. But again, how does one define reliability? Furthermore, if each person's QoL is assessed by different people for different domains, intra-individual variability is introduced and the interpretation of group differences between various domains becomes impossible. In considering 'real' differences between proxy and child, it is also a specific difficulty that children do not share the same perspectives, life standards, or expectations

as adults. Poor agreement between subject and proxy ratings should not be a criteria of rejection when assessing a QoL instrument as it might stem from a genuine difference in opinion between the two parties on certain issues.^{29,34} Eiser³⁹ has even suggested that 'very close parent-child agreement is indicative of poorer quality of life; childhood is about gaining autonomy and independence from parental views' (p 351). We are not aware of attempts to use other children as proxies but it could be an interesting way to overcome this problem.

Despite the obvious limitations and potential biases involved in proxy assessment, particularly in children with IIm, such reports provide at least a partial view of a child's QoL.

Conclusion

There is continuing debate about how to evaluate QoL in people with IIm and from whom to obtain the information. Table I summarizes the characteristics of the articles concerning subjective QoL assessment reviewed in this paper.

The history of QoL assessment in those with IIm has moved from the evaluation of exclusively objective dimen-

sions to the acknowledgement of the necessity for subjective assessment as well, reflecting the view that all individuals have their own experience of their life's quality and should, therefore, be given the opportunity to voice their perceptions. However, some authors have reservations concerning the relevance of assessing subjective aspects of QoL in people with IIm, and particularly question the use of satisfaction as a reliable indicator of QoL.⁴⁰⁻⁴² They believe assessing only subjective QoL disadvantages vulnerable people, such as those with IIm, as they may express high enjoyment of their lives within a poor environment. People with IIm may be unaware that better quality is possible or consciously aware that they must limit their expectations because of their present circumstances. Furthermore, most people maintain a feeling of satisfaction and well-being at a fairly constant level over the longer term due to various processes of adaptation, selective-attention, and social comparison.⁴³ This relative permanence of satisfaction emphasises the importance of an objective assessment to complement the subjective perspective.

We think there are fundamental conceptual and methodological reasons for applying universal standards of life quality to all people, regardless of impairment, and that both subjective and objective dimensions of QoL should be assessed to provide a complete perspective on an individual's life. The challenge is to present questions on universal concepts in a way that is understandable and answerable by all, and particularly by children with IIm.

Whenever possible the children themselves should be asked for their opinion on their life. Although QoL instruments should be chosen according to the underlying conceptual hypotheses, simplicity, clarity, length, and the use of pictures should also be taken into account, as they are likely to improve interest, comprehension, accuracy, and responsiveness in children with IIm. The implementation of a formal and systematic competency test similar to Cummins' procedure¹⁷ is an important methodological recommendation as it is a simple means of evaluating the child's comprehension and ability to respond adequately. In the case of children with insufficient communication skills, the use of proxy respondents becomes inevitable. Further research is needed on proxy-subject and proxy-proxy agreement for the assessment of subjective QoL of children with IIm, bearing in mind that different people have different perspectives and will provide valid and complementary information. However, it is important to remember and explicitly acknowledge that a proxy response only provides an indirect account of a child's well-being. Finally, if children with and without IIm are to be included in the same study, it is important to obtain proxy reports for all children, including those who can complete a self-report, in order to make valid comparisons of QoL across the ability range.

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Table I: Characteristics of subjective quality of life studies analyzed in this paper^a

References	Children	Intellectual impairment	Self(S) and/or proxy (P) report
Schneider et al. (2001) ⁶	+	+	P
Liptak et al. (2001) ³	+	+	P
Kenes et al. (2002) ⁵	+	+	P
McCarthy et al. (2002) ⁸	+	+	P
Wake et al. (2003) ⁹	+	+	P
Rosenbaum and Saigal (1996) ³⁷	+		S
Hodgkinson et al. (2002) ⁴	+		S
Ennett et al. (1991) ²⁵	+		S/P
Varni et al. (1995) ²⁶	+		S/P
Levi and Drotar (1999) ²⁷	+		S/P
Parsons et al. (1999) ²⁸	+		S/P
Eiser (1997) ³⁹	+		S/P
Eiser and Morse (2001) ²⁹	+		S/P
Britto et al. (2004) ³⁰	+		S/P
Waters et al. (2000) ³³	+		P
Antaki and Rapley (1996) ²²		+	S
Felce (1997) ⁴⁰		+	S
Antaki (1999) ²³		+	S
Schalock et al. (2000) ¹⁹		+	S
Hensel (2001) ⁴¹		+	S
Hatton and Ager (2002) ⁴²		+	S
Hensel et al. (2002) ²⁴		+	S
Cummins (1993) ¹⁷		+	S/P
Stancliffe (1999) ³⁴		+	S/P
Schwartz and Rabinovitz (2003) ²⁰		+	S/P

^aThe following databases were searched: PUBMED (Medline), Ovid, BIOSIS, EMBASE PSYCHIATRY, CURRENT CONTENTS, and PASCAL between 1977 and April 2004 using various combinations of the following keywords: quality of life, well-being, child*, mental retardation, intellectual disability*, cognitive impairment. We also searched recent contents pages (September 1999 to beginning 2003) in the following journals: 'Quality of Life Research'; 'Developmental Medicine and Child Neurology'; 'Child: Care, Health and Development'; and 'Journal of Intellectual Disability Research'. This was supplemented by cross-referencing with reference lists in identified articles.

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References

1. Schipper H, Clinch JJ, Olweny CLM. (1996) Quality of life studies: definitions and conceptual issues. In: Spilker B, editor. *Quality of Life and Pharmacoeconomics in Clinical Trials*. 2nd edn. Philadelphia: Lippincott Raven. p 11–23.
2. Edgerton RB. (1990) Quality of life from a longitudinal research perspective. In: Schalock RL, editor. *Quality of life: Perspectives and Issues*. Washington DC: American Association on Mental Retardation. p 149–160.
3. Liptak GS, O'Donnell M, Conaway M, Chumlea WC, Worley G, Henderson RC, Fung E, Stallings VA, Samson-Fang L, Calvert R, Rosenbaum P, Stevenson RD. (2001) Health status of children with moderate to severe cerebral palsy. *Dev Med Child Neurol* **43**: 364–370.
4. Hodgkinson I, d'Anjou MC, Dazord A, Berard C. (2002) Quality of life of a population of 54 ambulatory children with cerebral palsy. A cross-sectional study. *Ann Readapt Med Phys* **45**: 154–158.
5. Kennes J, Rosenbaum P, Hanna SE, Walter S, Russell D, Raina P, Bartlett D, Galuppi B. (2002) Health status of school-aged children with cerebral palsy: information from a population-based sample. *Dev Med Child Neurol* **44**: 240–247.
6. Schneider JW, Gurucharri LM, Gutierrez AL, Gaebler-Spira DJ. (2001) Health-related quality of life and functional outcome measures for children with cerebral palsy. *Dev Med Child Neurol* **43**: 601–608.
7. Beckung E, Hagberg G. (2002) Neuroimpairments, activity limitations, and participation restrictions in children with cerebral palsy. *Dev Med Child Neurol* **44**: 309–316.
8. McCarthy ML, Silberstein CE, Atkins EA, Harryman SE, Sponseller PD, Hadley-Miller NA. (2002) Comparing reliability and validity of pediatric instruments for measuring health and well-being of children with spastic cerebral palsy. *Dev Med Child Neurol* **44**: 468–476.
9. Wake M, Salmon L, Reddihough D. (2003) Health status of Australian children with mild to severe cerebral palsy: cross-sectional survey using the Child Health Questionnaire. *Dev Med Child Neurol* **45**: 194–199.
10. van Dongen KAJ, Abu-Saad H, Hamers JPF, Zwakhalen SMG. (2002) Pain assessment in the intellectually disabled child: the challenges of tool development. *The Suffering Child* **1**: 1E–5E.
11. Dekker MC, Nunn RJ, Einfeld SE, Tonge BJ, Koot HM. (2002) Assessing emotional and behavioural problems in children with intellectual disability: revisiting the factor structure of the developmental behavior checklist. *J Autism Dev Disord* **32**: 601–610.
12. Ramirez SZ, Kratochwill TR. (1997) Self-reported fears in children with and without mental retardation. *Ment Retard* **35**: 83–92.
13. Sarphare G, Aman MG. (1996) Parent- and self-ratings of anxiety in children with mental retardation: agreement levels and test-retest reliability. *Res Dev Disabil* **17**: 27–39.
14. Tyack D, Ingram D. (1977) Children's production and comprehension of questions. *J Child Lang* **4**: 211–224.
15. Sigelman CK, Winer JL, Schoenrock CJ. (1982) The responsiveness of mentally retarded persons to questions. *Educ Training Ment Ret* **17**: 120–124.
16. Atkinson D. (1988) Research interviews with people with mental handicaps. *Ment Hand Res* **1**: 75–90.
17. Cummins RA. (1993) *Comprehensive Quality of Life Scale – Intellectual Disability*. 4th edn. Melbourne: Psychology Research Centre.
18. Dagnan D, Ruddick L. (1995) The use of analogue scales and personal questionnaires for interviewing people with learning disabilities. *Clin Psychol Forum* **79**: 21–24.
19. Schalock RL, Bonham GS, Marchand CB. (2000) Consumer based quality of life assessment: a path model of perceived satisfaction. *Eval Prog Plan* **23**: 77–87.
20. Schwartz C, Rabinovitz S. (2003) Life satisfaction of people with intellectual disability living in community residences: perceptions of the residents, their parents and staff members. *J Intellect Disabil Res* **47**: 75–84.
21. Harter S, Pike K. (1984) The pictorial scale of perceived competence and social acceptance for young children. *Child Dev* **48**: 80–87.
22. Antaki C, Rapley M. (1996) Questions and answers to psychological assessment schedules: hidden troubles in 'quality of life' interviews. *J Intellect Disabil Res* **40**: 421–437.
23. Antaki C. (1999) Interviewing persons with a learning disability: how setting lower standards may inflate well-being scores. *Qual Health Res* **9**: 437–454.
24. Hensel E, Rose J, Kroese BS, Banks-Smith J. (2002) Subjective judgements of quality of life: a comparison study between people with intellectual disability and those without disability. *J Intellect Disabil Res* **46**: 95–107.
25. Ennett ST, DeVellis BM, Earp JA, Kredich D, Warren RW, Wilhelm CL. (1991) Disease experience and psychosocial adjustment in children with juvenile rheumatoid arthritis: children's versus mothers' reports. *J Pediatr Psychol* **16**: 557–568.
26. Varni JW, Katz ER, Colegrove R, Dolgin M. (1995) Adjustment of children with newly diagnosed cancer: cross-informant variance. *J Psychosoc Oncol* **18**: 23–38.
27. Levi RB, Drotar D. (1999) Health-related quality of life in childhood cancer: discrepancy in parent-child reports. *Int J Cancer Suppl* **12**: 58–64.
28. Parsons SK, Barlow SE, Levy SL, Surpan SE, Kaplan SH. (1999) Health-related quality of life in pediatric bone marrow transplant survivors: according to whom? *Int J Cancer Suppl* **12**: 46–51.
29. Eiser C, Morse R. (2001) Quality-of-life measures in chronic diseases of childhood. *Health Technol Assess* **5**: 55–63.
30. Britto MT, Kotagal UR, Chenier T, Tsevat J, Atherton HD, Wilmott RW. (2004) Differences between adolescents' and parents' reports of health-related quality of life in cystic fibrosis. *Pediatr Pulmonol* **37**: 165–171.
31. Rajmil L, Fernandez E, Gispert R, Rue M, Glutting JP, Plasencia A, Segura A. (1999) Influence of proxy respondents in children's health interview surveys. *J Epidemiol Community Health* **53**: 38–42.
32. Landgraf JM, Abetz LN. (1996) Measuring health outcomes in pediatric populations: issues in psychometrics and application. In: Spilker B, editor. *Quality of Life and Pharmacoeconomics in Clinical Trials*. 2nd edn. Philadelphia: Lippincott-Raven. p 793–802.
33. Waters E, Salmon L, Wake M. (2000) The parent-form Child Health Questionnaire in Australia: comparison of reliability, validity, structure, and norms. *J Pediatr Psychol* **25**: 381–391.
34. Stancliffe RJ. (1999) Proxy respondents and the reliability of the Quality of Life Questionnaire Empowerment factor. *J Intellect Disabil Res* **43**: 185–193.
35. Sneeuw KC, Aaronson NK, Sprangers MA, Detmar SB, Wever LD, Schornagel JH. (1998) Comparison of patient and proxy EORTC QLQ-C30 ratings in assessing the quality of life of cancer patients. *J Clin Epidemiol* **51**: 617–631.
36. Magaziner J, Simonsick E, Kashner T, Hebel J. (1988) Patient-proxy response comparability on measures of patient health and functional status. *J Clin Epidemiol* **41**: 1065–1074.
37. Rosenbaum P, Saigal S. (1996) Measuring health-related quality of life in pediatric populations: conceptual issues. In: Spilker B, editor. *Quality of Life and Pharmacoeconomics in Clinical Trials*. 2nd edn. Philadelphia: Lippincott-Raven. p 785–792.
38. Cadman D, Goldsmith C, Bashim P. (1984) Values, preferences and decisions in the care of children with developmental disabilities. *J Dev Behav Pediatr* **5**: 60–64.
39. Eiser C. (1997) Children's quality of life measures. *Arch Dis Child* **77**: 350–354.
40. Felce D. (1997) Defining and applying the concept of quality of life. *J Intellect Disabil Res* **41**: 126–135.
41. Hensel E. (2001) Is satisfaction a valid concept in the assessment of quality of life of people with intellectual disabilities? A review of the literature. *J Appl Res Intellect Dis* **14**: 311–326.
42. Hatton C, Ager A. (2002) Quality of life measurement and people with intellectual disabilities: a reply to Cummins. *J Appl Res Intellect Dis* **15**: 254–260.
43. Cummins RA. (1995) On the trail of the gold standard for subjective well-being. *Social Indic Res* **35**: 179–200.