



Brief Communication

Quality of life in childhood epilepsy: What is the level of agreement between youth and their parents?

L.H. Verhey^a, D.M. Kulik^b, G.M. Ronen^{a,*}, P. Rosenbaum^a, L. Lach^c, D.L. Streiner^d, the Canadian Pediatric Epilepsy Network¹^a Department of Paediatrics, McMaster University, Hamilton, Ont., Canada^b Department of Paediatrics, The Hospital for Sick Children, University of Toronto, Toronto, Ont., Canada^c School of Social Work, McGill University, Montreal, Ont., Canada^d Department of Psychiatry, University of Toronto, Toronto, Ont., Canada

ARTICLE INFO

Article history:

Received 5 September 2008

Revised 29 November 2008

Accepted 13 December 2008

Available online 4 January 2009

Keywords:

Health-related quality of life

Epilepsy

Childhood

CHEQOL-25

Child–parent agreement

Discrepancy

Intraclass correlation coefficient

ABSTRACT

Children and parents evaluate the child's quality of life (QOL) from their own perspectives; therefore, responses may differ, especially in abstract domains. We examined differences between self- and proxy-reported QOL of children with epilepsy. Children with active epilepsy ($N = 375$) and their parents ($N = 378$) separately completed the CHEQOL-25, a condition-specific QOL measure. The intraclass correlation coefficient was used to determine interrater agreement. Concordance on the Total CHEQOL-25 was 0.45 ($P < 0.01$). Discrepancies were greatest for the subscales of Secrecy (0.24, $P < 0.01$) and Present Concerns (0.32, $P < 0.01$). School placement correlated with discrepancy in the Intrapersonal/Emotional subscale ($r = 0.19$, $P < 0.05$), and the child's age at testing correlated with discrepancy of the Total measure ($r = 0.15$, $P < 0.01$). This study demonstrates that parent perspectives alone are insufficient to measure their child's QOL. The CHEQOL-25 is a practical tool, with complementary parent and child versions, which can be used to determine health-related quality of life in children with epilepsy.

© 2008 Elsevier Inc. All rights reserved.

1. Introduction

The concept of health outcome recognizes that the goal of health care is to help people feel and function better, as well as adapt to the social and psychological impacts and values of their impairments. The recent introduction of the WHO International Classification of Functioning, Health and Disability (ICF) framework importantly expands the focus on health to include environmental and personal factors, and to identify “activity” and “participation” as goals worthy of attention [1]. Furthermore, there is growing interest in understanding and enhancing the “life quality” of individuals with chronic medical conditions for which cure is not currently a realistic goal.

There is a significant misconception about what constitutes “quality of life” (QOL) and “health-related quality of life” (HRQOL) [2]. A generally accepted definition of QOL is the “individual's per-

ceptions of their position in life in the context of the culture and value systems in which they live, and in relation to their goals, expectations and concerns” [3]. In a nutshell, it entails “an overall assessment of wellbeing across various domains” [4]. HRQOL is considered to be either a subdomain of the more global construct of QOL, which includes health-related domains of life [5], or a closely related but independent construct [6].

“Health outcome” refers to both objective indicators of functional status, activity, and participation [7] and subjective indicators of HRQOL. For children with a chronic health condition, quality of life should incorporate the child's own perception of how his or her internal and external life and well-being are affected by the condition and its treatment. As such, self-reported evaluation of QOL is essential. However, children with limited cognitive or communication abilities may not be able to articulate their experiences, difficulties, and concerns about living with a chronic condition [8]. In these circumstances, parents may act as surrogate informants of their child's HRQOL. Parents' assessments of their child should correlate closely with self-reports when rating their child's performance on concrete, observable functioning, such as motor ability; the correlations would be expected to be lower on functional emotional measures (i.e., anxiety or depression) and lowest on their perceptions of abstract issues, such as their child's

* Corresponding author. Address: Department of Paediatrics (Neurology), McMaster University, 1200 Main Street West HSC 3A, Hamilton, Ont., Canada L8N 3Z5. Fax: +1 905 521 7914.

E-mail address: roneng@mcmaster.ca (G.M. Ronen).

¹ F. Booth, D. Buckley, C. Camfield, L. Castagna, P. Cooper, I. Elliot, D. Keene, S. Levine, D. Meek, S. Penney, A. Prasad, L. Roche, M. Shevell, B. Sinclair, O. Snead, J. Tibbles, S. Whiting.

internal life [8–11]. The reason is that, when children and parents evaluate the child's HRQOL, they may draw on different values and factors that could result in discordance between raters [12,13]. For example, we know that parents of children with epilepsy are not aware of the child's quest for normality; and, in contrast to their parents, children are not concerned about the future [14,15]. Thus, a combination of child self- and parent proxy-reported HRQOL may provide more complete information about how the child is affected by the condition or its treatment than either alone [16].

The objective of this study was to gain a better understanding of how children and their parents differentially report various issues of the child's HRQOL when responding to a measure that captures the experiential aspects of childhood epilepsy from the youths' and parents' perspectives [14] and where the conceptual components of HRQOL are closely aligned with what youth draw on to generate a representation of their HRQOL [17]. We hypothesized that concrete external domains of HRQOL, such as interpersonal-social consequences of epilepsy, correlate more highly between the child and their parent than less visible abstract domains, such as keeping epilepsy a secret. In addition, we suggest that age at epilepsy onset, gender, epilepsy duration, and seizure severity are not associated with interrater discrepancy. However, the age of the child at testing and whether or not the child receives special help in school [18] are associated with discordance between parent- and child-reported HRQOL. This is expected, in part, because of the age-dependent level of communication between children and their parents and a potentially lower level of communication between children with cognitive or behavioral problems and their parents.

2. Methods

Children and youth with epilepsy and their parents were recruited from four university hospital epilepsy centers, five pediatric neurology hospital clinics, and five pediatric neurology private practices across Canada. Criteria for inclusion were: (1) children and youth aged 8–17 years inclusive, (2) a diagnosis of active epilepsy, with at least one seizure in the previous 24 months, and (3) the ability of the child or youth and their parent(s) to comprehend the English language at a grade 3 reading level, as judged by the parents themselves. Parents were informed that they and their child would be asked to complete measures regarding their child's life with epilepsy. Relevant research ethics boards approved the study, and informed consent and assent were obtained from each participating parent or caregiver and child, respectively.

Children and parents independently completed the 25-item CHEQOL-25, an epilepsy-specific measure of QOL, on which the items follow the format of alternative paired options of forced responses [19]. Proxy raters were asked to respond the way they thought their child would score the individual items. Our current analyses included either the mother's or the father's perspective because, in a previous study [14], we found no significant differences between the groups of responding mothers and fathers, and couples showed relatively similar scores on each of the five subscales.

The conceptual model for this measure was generated by interviewing children and their parents separately, using a focus group format [17,20]. Factor analysis generated the conceptual domains constituting HRQOL, and these domains became the five subscales of the measure, each consisting of five items [14]. The subscales are: (1) Interpersonal/Social, (2) Intrapersonal/Emotional, (3) Present Worries and Concerns, (4) Secrecy, (5) Quest for Normality (child only) and Future Worries and Concerns (parent only). Each item is scored on a scale of 1 to 4. A higher score on the CHEQOL-25 reflects a more positive perception of the child's QOL in

that domain. The psychometric properties of the CHEQOL-25 were detailed in our original article [14].

Our primary interest was to determine the level of agreement between parent proxy reports and child self-reports rather than the statistical significance of correlation between the raters. To do this, in our analyses we used the intraclass correlation coefficient (ICC), which is based on a two-way repeated-measures ANOVA [21]. The strength of the ICC is that it takes both agreement and association into account. Correlational analyses between discrepancy and other variables of interest (e.g., age at epilepsy onset and seizure severity) were conducted using Pearson's *r*.

3. Results

Of this cohort of 391 children and their parents, 375 children and 378 parents had complete data on the CHEQOL-25 questionnaire. The demographics of the cohort are provided in Table 1.

All discrepancy scores were calculated by subtracting each parent's score from his or her child's score on the Total scale and four subscales common to both the parent proxy- and child self-report versions (i.e., Interpersonal/Social, Intrapersonal/Emotional, Present Concerns, and Secrecy). A positive discrepancy score means that children self-reported their HRQOL higher compared with parent proxy reports.

Agreement between child and parent on the CHEQOL-25 measure ranged from ICC = 0.24 to 0.49 (Table 2). Parents rated the HRQOL of the child lower than did the children themselves on the Total scale (Table 3). Discordance between parent proxy-report and child self-report was greatest for Secrecy (ICC = 0.24, $P < 0.01$), where the children's rating was higher than the parents', and Present Concerns (ICC = 0.32, $P < 0.01$), where the children's rating was lower than the parents' (Table 2). Children and their parents were more likely to agree on the more external measures of Intrapersonal

Table 1
Demographics.

	Median	Mean	N	SD
Age at epilepsy onset	7.0	6.9	390	3.5
Duration of epilepsy (years)	3.0	4.6	390	3.5
Age at test	11.0	11.4	391	2.1
Grade in school	6.0	5.9	382	2.2
<i>School placement</i>				
Regular classes			276	
Classes with special help			111	
Missing			4	
<i>Seizure severity</i>				
None in 12 months			45	
Low severity			134	
High severity			210	
Missing			2	
<i>Gender</i>				
Male			189	
Female			202	

Table 2
ICCs between parent and child on CHEQOL-25 (sub)scales.

	ICC	<i>P</i> value
Total	0.45	<0.001
Interpersonal/Social	0.49	<0.001
Present Worries	0.32	<0.001
Intrapersonal/Emotional	0.49	<0.001
Secrecy	0.24	<0.001

Table 3
Means and SD of (sub)scale scores on child self-reported and parent proxy-reported CHEQOL-25.

CHEQOL-25 subscale	Self-report		Proxy report		Mean difference (discrepancy score)
	Mean	SD	Mean	SD	
Total	77.02	12.93	72.18	13.53	4.8
Interpersonal/Social	16.81	3.54	15.79	3.50	1.0
Present Worries	13.04	3.70	14.43	3.09	-1.4
Intrapersonal/Emotional	14.67	5.93	13.09	3.97	1.6
Secrecy	16.23	3.24	14.14	3.37	2.1
Normality	16.82	2.87	—	—	—
Future Worries	—	—	15.12	3.89	—

sonal/Emotional ($ICC = 0.49$, $P < 0.01$) and Interpersonal/Social ($ICC = 0.49$, $P < 0.01$) (Table 2).

The child's age at time of testing was directly related to CHEQOL-25 child–parent discrepancy score on both the Total scale ($r = 0.15$, $P < 0.01$) and Interpersonal subscale ($r = 0.12$, $P < 0.05$). The level of child–parent agreement on the CHEQOL-25 Intrapersonal/Emotional subscale was greater for children enrolled in regular classes than for those in classes with special help ($r = 0.19$, $P < 0.05$).

Gender, age at epilepsy onset, duration of epilepsy, proportion of life with epilepsy, and seizure severity [22] were not significantly correlated with the level of agreement between child self-reports and parent proxy-reports on the CHEQOL-25.

4. Discussion

Agreement between parent proxy-reports and child self-reports is a function of the measure of concreteness, visibility, and externality of the variable being measured [10,11,23]. This applies when HRQOL is conceptualized as a functional and objective phenomenon that incorporates factors such as functional impairment, emotional health, social activity, and cognitive functioning, all of which are observable. When more internal and experiential factors such as self-perception and experience of social support are incorporated into the measurement of HRQOL, agreement is understandably less likely. The findings in the present study of levels of agreement between child self-reports and parent proxy reports show lower parent–child agreement on the more abstract domains of HRQOL in children with active epilepsy. This finding is in agreement with previous studies [8,10,23–26].

We found that the distributions of the level of child–parent agreement for the Interpersonal and Intrapersonal subscales were centered around zero, suggesting that these domains reflect more externally visible perceptions of life with epilepsy and, therefore, are not as vulnerable to reporting discrepancy. Perceptions of the more abstract, less concrete internal experiences of life with epilepsy correlated less strongly between parent proxies and children (i.e., Secrecy and Present Concerns). Intraclass correlations ranged from 0.24 to 0.49, with a median ICC of 0.45. Others have reported that the level of agreement between child and parent proxy-reports on multidimensional health and functional status measures ranged from a median correlation of 0.42 to 0.78 [8,11,23,24,27]. In a review of child–parent agreement when reporting on level of impairment, a clear difference is demonstrated in the magnitude of agreement on physical domains of functioning, such as motor function, activities of daily living, and overall health (median ICC ranging from 0.60 to 0.71) compared with domains of emotional, social, and cognitive functioning, such as social support, social activities, attention, alertness, and emotional behavior (median ICC ranging from 0.48 to 0.50) [8].

Why does this decreased concordance between parent proxy-reports and child self-reports exist for the abstract and phenomenological domains of QOL in childhood epilepsy? Private feelings

experienced by children and youth with active epilepsy, and the desire to keep these experiences a secret, means that parents may be unaware of the nonvisible experiences and nonexpressed feelings of their children. On the other hand, parents have more developed cognitive capabilities and life experiences than their child, enabling them to think about the future concerns of their child and how they might adapt to life with a chronic health condition later in life [14]. Another possible reason for the discordance on abstract domains is that children with epilepsy may be more prone to focus on and recall the positive experiences because they are engaged in a quest for normality, whereas parents report the negative perceptions in the life of their child in addition to positive ones, perhaps reflecting their worries about the future [13].

We and others believe that parents and children may be drawing on different experiences and different groups of underlying factors to evaluate quality of life [12]. For example, increased parental stress, rather than severity of the child's condition, was a factor contributing to decreased parent ratings of child's QOL in children with cerebral palsy [28,29]. From the children's point of view, style of coping with a health condition and resilience may affect how they think of their HRQOL [9,30].

Another question is why parents underestimate their children's overall QOL, compared with the children themselves. The fact that some children with significant health problems are highly satisfied with at least some aspects of their lives (the so-called "disability paradox") may not be apparent to their parents [31]. Emotions and attitudes can be selectively revealed by individuals with a condition. Negative feelings are more likely to be displayed or communicated than positive ones [11]. Parents may then make an inference about their child's HRQOL based on these negative emotions and feelings that their child has felt and expressed. Parents, as observers, also place more weight on negative emotions than positive ones because they are more salient and memorable [8,11,16,23,24].

Age of the child with active epilepsy (8–17 years) was associated with the child–parent discrepancy scores. This is in agreement with other studies of children with cerebral palsy (aged 7–13 years) where parents of older children rated their children's QOL lower on the Parental Relations domain than did the children themselves (odds ratio ranged from 0.9 to 2.4), and parents of younger children proxy-reported their child's QOL on the School domain lower than the children themselves (odds ratio ranged from 1 to 2.9) [18]. School placement was also weakly associated with child–parent agreement. This may suggest that, in contrast to the child who feels well-adjusted, parents are cognizant of their child's social, cognitive, and emotional challenges, when they compare their children's performance with that of the rest of the class. It is important to note that in the present analyses some of these relatively small correlations were statistically significant, presumably as a function of the large sample size, but the amount of variance explained (R^2) is very small.

The child's age at epilepsy onset, duration of epilepsy, proportion of life with epilepsy, and seizure severity did not correlate

with the level of agreement between each of the child's and parent's ratings of HRQOL. This suggests that indicators of HRQOL cannot be explained by the seizure and epilepsy variables alone. The phenomenological experience of life with epilepsy is multidimensional. Objective and subjective factors at the child, family, and societal levels extend far beyond the biomedical factors, and may act as moderators or mediators of the biomedical factors of epilepsy [32]. Less clear is the relative importance of each of these factors and the paths and feedback loops that they follow over time and at different ages. Understanding these issues requires studies designed specifically to measure these factors simultaneously.

The question remains as to whether these findings are specific to children with epilepsy or whether this discrepancy between child self-reports and parent proxy-reports can be generalized to children with other neurodevelopmental conditions. Compared with individuals with visible neurodevelopmental conditions, children with epilepsy experience unpredictability of seizures, lack of control over the body and mind, a constant sense of vulnerability and uncertainty, and a feeling of marginalization or stigma. However, children with more visible disabilities also experience feelings of fatigue, fear, sense of hopelessness, and perceived or real stigma. In these situations, it is likely that a similar level of disagreement between children and their parents on these less tangible, abstract domains of HRQOL may be evidenced.

There was generally a modest mean level of agreement between children with active epilepsy and their parents on the phenomenological and subjective domains of HRQOL, with the exception of lower agreement on the Present Worries and Secrecy domains. Any difference in the level of agreement that exceeds half of a standard deviation may be considered as clinically significant [33]. Our findings suggest the importance of capturing both child and parent evaluations of children's QOL to attain a more descriptive understanding of the perceived challenges and concerns regarding the past and present, as well as issues of secrecy experienced by children and youth with epilepsy and their families. Children lack the cognitive maturity to think about their future experiences of life with epilepsy. In these instances, parent-proxy reports contribute valuable information that would otherwise go unreported, noting of course these are parent perceptions and, therefore, cannot be attributed to children themselves. Furthermore, understanding these perspectives may provide guidance about the experience of epilepsy for counseling individual children and families.

We believe that these results are generalizable to a population of children aged 8–17 years, in comparable societies, who comprehend language at a grade 3 reading level. However, cross-sectional studies neither lend themselves to establish temporal relationships nor are appropriate to study the natural history of the phenomena. Longitudinal data are necessary to determine how the parent-child level of agreement changes over time and whether the data reveal any specific pattern that might provide insight into the relative importance of the factors underlying HRQOL.

Acknowledgments

This study was funded by the Canadian Institutes of Health Research Health Professional Student Research Award.

References

- [1] World Health Organization. International classification of functioning, disability and health (ICF). Geneva: WHO; 2001.
- [2] Waters E, Davis E, Ronen GM, et al. Quality of life instruments for children and adolescents: conceptual perspectives on how to choose the appropriate instrument for individuals with disabilities. *Dev Med Child Neurol* 2009, in press.
- [3] WHOQOL Group. Study protocol for the World Health Organization project to develop a quality of life assessment instrument (the WHOQOL). *Qual Life Res* 2003;2:153–9.
- [4] Bjornson KF, McLaughlin JF. The measurement of health-related quality of life (HRQL) in children with cerebral palsy. *Eur J Neurol* 2001;8:183–93.
- [5] Spilker B, Revicki DA. Taxonomy of quality of life. In: Spilker B, editor. *Quality of life and pharmacoeconomics in clinical trials*. Philadelphia: Lippincott-Raven; 1996. p. 25–31.
- [6] Livingston MH, Rosenbaum PL, Russell DJ, Palisano RJ. Quality of life among adolescents with cerebral palsy: what does the literature tell us? *Dev Med Child Neurol* 2007;49:225–31.
- [7] King GA, Law M, King S, et al. Measuring children's participation in recreation and leisure activities: construct validation of the CAPE and PAC. *Child Care Health Dev* 2007;33:28–39.
- [8] Sneeuw KC, Sprangers MA, Aaronson NK. The role of health care providers and significant others in evaluating the quality of life of patients with chronic disease. *J Clin Epidemiol* 2002;55:1130–43.
- [9] Barbosa J, Tannock R, Manassis K. Measuring anxiety: parent-child reporting differences in clinical samples. *Depress Anxiety* 2002;15:61–5.
- [10] Sprangers MAG, Aaronson NK. The role of health care providers and significant others in evaluating the quality of life of patients with chronic disease: a review. *J Clin Epidemiol* 1992;45:743–60.
- [11] Epstein AM, Hall JA, Tognetti J, Son LH, Conant Jr L. Using proxies to evaluate quality of life: can they provide valid information about patients' health status and satisfaction with medical care? *Med Care* 1989;27(Suppl.):387–97.
- [12] Ronen GM, Lach L, Streiner DL, et al. and the North American Pediatric Epilepsy HRQL Research Group. Exploring predictors of self-reported health-related quality of life (HRQL) among children with epilepsy [abstract]. Presented at the American Epilepsy Society (AES) annual meeting; 2008, Philadelphia.
- [13] Elliott IM, Lach L, Smith ML. Adolescent and maternal perspectives of quality of life and neuropsychological status following epilepsy surgery. *Epilepsy Behav* 2000;1:406–17.
- [14] Ronen GM, Streiner DL, Rosenbaum P. Health-related quality of life in children with epilepsy: development and validation of self-report and parent proxy measures. *Epilepsia* 2003;44:598–612.
- [15] Elliott IM, Lach L, Smith ML. I just want to be normal: a qualitative study exploring how children and adolescents view the impact of intractable epilepsy on their quality of life. *Epilepsy Behav* 2005;7:664–78.
- [16] Eiser C, Morse R. Can parents rate their child's health-related quality of life? Results of a systematic review. *Qual Life Res* 2001;10:347–57.
- [17] Ronen GM, Rosenbaum P, Law M, Streiner DL. Health-related quality of life in childhood epilepsy: the results of children's participation in identifying the components. *Dev Med Child Neurol* 1999;41:554–9.
- [18] White-Koning M, Arnaud C, Dickinson HO, et al. Determinants of child-parent agreement in quality-of-life reports: a European study of children with cerebral palsy. *Pediatrics* 2007;120:804–14.
- [19] Harter S. *The Self-Perception Profile for Children: revision of the perceived competence scale for children*. Denver, CO: Univ. of Denver; 1985.
- [20] Ronen GM, Rosenbaum P, Law M, Streiner DL. Health-related quality of life in childhood disorders: a modified focus group technique to involve children. *Qual Life Res* 2001;10:71–9.
- [21] Shrout PE, Fleiss JL. Intraclass correlations: uses in assessing rater reliability. *Psychol Bull* 1979;86:420–8.
- [22] Cramer JA, Westbrook LE, Devinsky O, Perrine K, Glassman MB, Camfield C. Development of the Quality of Life in Epilepsy Inventory for Adolescents: the QOLIE-AD-48. *Epilepsia* 1999;40:1114–21.
- [23] Theunissen NC, Vogels TG, Koopman HM, et al. The proxy problem: child report versus parent report in health-related quality of life research. *Qual Life Res* 1998;7:387–97.
- [24] Hays RD, Vickrey BG, Hermann BP, et al. Agreement between self reports and proxy reports of quality of life in epilepsy patients. *Qual Life Res* 1995;4:159–68.
- [25] Yeh CH, Chang CW, Chang PC. Evaluating quality of life in children with cancer using children's self-reports and parent-proxy reports. *Nurs Res* 2005;54:354–62.
- [26] Achenbach TM, McConaughy SH, Howell CT. Child/adolescent behavioural and emotional problems: implications of cross-informant correlations for situational specificity. *Psychol Bull* 1987;101:213–32.
- [27] Robitail S, Siméoni MC, Ravens-Sieberer U, Bruil J, Auquier P, and the KIDSCREEN Group. Children proxies' quality-of-life agreement depended on the country using the European KIDSCREEN-52 questionnaire. *J Clin Epidemiol* 2007;60:469–78.
- [28] Arnaud C, White-Koning M, Michelsen SI, et al. Parent-reported quality of life of children with cerebral palsy in Europe. *Paediatrics* 2008;121:54–64.
- [29] Ravens-Sieberer U, Gosch A, Rajmil L, et al. and the KIDSCREEN Group. The KIDSCREEN-52 quality of life measure for children and adolescents: psychometric results from a cross-cultural survey in 13 European countries. *Value Health* 2008;11:645–58.
- [30] Manassis K, Mendlowitz S, Menna R. Child and parent reports of childhood anxiety: differences in coping styles. *Depress Anxiety* 1997;6:62–9.
- [31] Albrecht GL, Devlieger PJ. The disability paradox: high quality of life against all odds. *Soc Sci Med* 1999;48:977–88.
- [32] Baron RM, Kenny DA. The moderator-mediator variable distinction in social psychological research: conceptual, strategic, and statistical considerations. *J Pers Soc Psychol* 1986;51:1173–82.
- [33] Norman G, Sloan J, Wyrwich K. Interpretation of changes in health-related quality of life: the remarkable universality of half a standard deviation. *Med Care* 2003;41:582–92.