Health and Quality of Life Outcomes

Review

Health-related quality of life in childhood epilepsy: Moving beyond 'seizure control with minimal adverse effects'

Gabriel M Ronen*[1], David L Streiner[2] and Peter Rosenbaum[1]

Address: [1]Department of Pediatrics, McMaster University, 1200 Main Street West, Hamilton, Ontario, Canada, L8N 3Z5 and [2]Kunin-Lunenfeld Applied Research Unit, Department of Psychiatry, University of Toronto., 734-3560 Bathurst Street, Toronto, Ontario, Canada, M6A 2E1

Email: Gabriel M Ronen* - roneng@mcmaster.ca; David L Streiner - dstreiner@klaru-baycrest.on.ca; Peter Rosenbaum - rosenbau@mcmaster.ca

* Corresponding author

Published: 28 August 2003

Health and Quality of Life Outcomes 2003, **1**:36

Received: 15 July 2003
Accepted: 28 August 2003

This article is available from: http://www.hqlo.com/content/1/1/36

Abstract

Childhood epilepsy is one of the most important and prevalent neurological conditions in the developing years. Persons with childhood onset epilepsy are at a high risk for poor psychosocial outcomes, even without experiencing co-morbidities. The goal of management of children with epilepsy should be to enable the child and the family to lead a life as free as possible from the medical and psychosocial complications of epilepsy. This comprehensive care needs to go beyond simply trying to control seizures with minimal adverse drug reactions. Seizure frequency and severity is only one important outcome variable. Other factors such as social, psychological, behavioural, educational, and cultural dimensions of their lives affect children with epilepsy, their families and their close social networks.

A number of epilepsy-specific health-related quality of life (HRQL) scales for children have been developed with the aim to include and measure accurately the impact and burden of epilepsy. Their target populations, details of the origin of the items, and psychometric properties vary significantly. Their strengths and weaknesses will be identified more clearly through their continued use in the clinical setting and in research studies. Only a few studies to date have utilized these or generic HRQL measures to assess the HRQL of specific populations with epilepsy.

Future research needs to develop theory driven models of HRQL and identify measurable factors that have important correlations with outcomes. Since biomedical variables like seizure frequency and severity have only moderate correlations with HRQL, other independent factors including the child's resilience, co-morbid conditions, parental well-being, family factors and societal/cultural variables may play a major role. We also need to learn what encompasses comprehensive patient care, define the goals of management and evaluate the impact of different interventions. Future studies need to include the children's own perspectives of their HRQL in addition to parental reports.

Finally, clinicians need to familiarize themselves with outcome measures, be able to evaluate them, and use them routinely in their day-to-day practice.

Childhood epilepsy: Pervasive impacts of a complex condition

Childhood epilepsy is among the most prevalent and therefore important neurological conditions in the developing years. Population based studies report prevalence rates of 3.6 to 4.2 per 1000 for children in developed countries [1,2], and approximately double these rates in developing countries.[3–5]

Epilepsy is characterized by its episodic and chronic nature. The seizures usually produce brief periods of disruption, which include phenomena such as loss of consciousness, bodily distortion, injuries, unusual and often frightening psychological experiences as well as urinary and bowel incontinence. The unpredictability of seizure recurrence is a constant threat to the patient with epilepsy and his or her family. Apart from the episodic seizures, there are many other ever-present factors – social, psychological, behavioural, educational, cultural and so forth – which affect the lives of children with epilepsy (CWE), their families and their close social networks. These factors vary considerably from one person to the next, but have a significant impact on the daily quality of life in every affected individual.

In the past, clinicians have tended to address the child's and family's perspectives on the impact and burden of epilepsy only marginally. The traditional medical goal in the management of epilepsy has focused almost exclusively on *seizure control with minimal or no adverse medication effects*. [6–8] Clearly, the goal should be to enable the child and family to lead a life as free as possible from the medical and psychosocial complications of epilepsy. This comprehensive care needs to go beyond the attempt simply of controlling seizures with minimal adverse drug reactions. [9–12]

In summarizing his experience with over 20,000 CWE, Livingston concluded that although the effort to control seizures is of primary importance, there remain many problems concerning the psychological management of the child and his or her parents and their relationship to the social milieu in which they live.[13] There is ample scientific evidence to confirm Livingston's contention regarding poor psychosocial outcome in childhood onset epilepsy. CWE are found to have relatively more compromised health-related quality of life (HRQL) in the psychological, social and school domains compared to children with asthma, suggesting that these problems are specific to epilepsy and not simply the result of living with a chronic condition.[14] CWE are also reported to experience academic underachievement in relation to their IQ.[15] The children often experience significant restrictions of activities, leading to lower HRQL.[16] In adults with epilepsy, outcome is negatively affected when a perceived stigma persists even in the absence of seizures.[17] Epilepsy has been felt to be a major determinant of psychosocial problems, irrespective of seizure frequency.[18] Age of onset of epilepsy has also emerged as a significant predictor: earlier age of onset has been associated with reduced likelihood of being married and older age of onset has been implicated in feelings of depression and stigma.[19]

Long-term population based outcome studies of childhood onset epilepsy do not show uniform results. A Finnish cohort of people with childhood onset epilepsy without any significant co-morbidity, followed for 35 years, showed that many patients had problems with social adjustment and competence as adults. In addition, 77% did not reveal their epilepsy to their employer or to peer workers. The authors concluded that seizure control plays only a partial role in the social adjustment and competence problems and that additional factors are likely contributors.[20] Other researchers have also concluded that although patients with frequent seizures had poorer psychosocial profiles than those with infrequent or no seizures, important predictors of psychopathology and social dysfunction seemed to exist in the patients with refractory epilepsy which could not be explained by physical or demographic data.[19] However, another study from Finland suggests that adults with childhood onset epilepsy do not have increased problems with depression or anxiety compared to controls and that people with epilepsy without co-morbid intellectual or learning disabilities do not have overt problems with social immaturity.[21] Similarly, a Japanese study of adults with childhood onset epilepsy reported more favorable prognoses than the former Finnish sample[20] in terms of educational and social aspects for those with normal intelligence, except for a lower marriage rate for the younger age group.[22] In contrast to CWE without co-morbidities, the outcome in populations with co-morbid intellectual deficits has been uniformly poor.[21,22] Intellectual deficits and mental retardation have been estimated at 33%, 37% and 39% in three population-based studies in childhood onset epilepsy.[22–24]

Families of children with frequent seizures suffer significantly more stress than families of children with infrequent seizures or of healthy controls.[25] Mothers of children with additional psychiatric problems are found to have higher rates of psychiatric disturbances themselves, although it is impossible to disentangle cause and effect in this study.[26] Siblings of children with chronic epilepsy also have increased behavioural issues,[26] mostly in externalizing behaviors.[25] Siblings of CWE report higher level of concern (1) that people will make fun of them because of their sibling's seizures; (2) not knowing how to help during a seizure; (3) feeling left out;

and (4) regarding- injury and death as a result of a seizure.[25]

Measuring HRQL in childhood epilepsy

(a) What is the current state of HRQL scale development?

There are two popular but distinct approaches to the measurement of HRQL.[27] One approach involves the application of '*generic*' HRQL tools, that provide a broad measure of HRQL irrespective of the underling disorder. Both the Child Health Questionnaire (CHQ) and the 'PedsQL' are gaining recognition as child focused, broad-based health profiles.[28,29] These instruments have been developed to address a comprehensive array of domains or attributes of psychosocial and physical functioning, and have the advantage that the data acquired can be compared across demographic or clinical populations. A potential limitation of these tools is that theoretically they might lack the sensitivity to detect subtle aspects of specific-conditions or disorders in a way that provides meaningful information to patients and professionals, although the empirical evidence in this regard is contradictory.

'*Disease* or *condition-specific*' HRQL instruments are created to assess characteristics of a particular condition. As such they are generally seen to be more relevant and sensitive to the nuances of the disorder. On the other hand, they provide data that address a narrower range of issues than generic instruments, and it is usually difficult or impossible to relate data from one disease-specific measure from another. Condition-specific instruments are less widely used than generic measures and therefore do no always have the well-documented psychometric properties that the latter generally show. The focus of this review is primarily on condition-specific measures that explore the HRQL of children and youth with epilepsy.

Adult HRQL measures are generally inappropriate for use in children.[30] In children, the HRQL measure must accommodate the changes that occur through development, and the domains in adult HRQL do not readily overlap those of children.[31] For children, HRQL is primarily about their social life, their activities and their physical appearance, and less about being economically productive and self-sufficient.[32,33] The development of HRQL measures in children therefore requires particular care on multiple specific perspectives and methods distinct from those in adults.[34,35] Furthermore, any evaluation of children's HRQL must provide for the children to rate their own HRQL.[35]

(b) How should one evaluate HRQL measures?

Clinicians are likely to use patient-reported outcome measures routinely only if these tools are as familiar to them as blood pressure assessment and other physiologic measures. This cannot occur until outcome measures become meaningful and easy to include in daily practice.[36] The health professional must have confidence that a scale measures what it intends to and that it does so with minimum of error.[37]

Any evaluation of scales should start with the information about where the items originate. Measurement tools are more likely to have content validity if the relevance of the HRQL questions is derived from a sample of the population in which the tool is to be used.[38] These should therefore be determined by the patients themselves, even if they are children, rather than solely by their parents or by health professionals.[32] It has been shown that children identify more items than health professionals or even their own parents, and contribute significantly to the wording of the questions.[39,40] Children as young as seven or eight years have been found to be consistent and accurate in their understanding of the questions and response options, and have demonstrated very good test-retest reliability.[41,42] Checking the quality of the items comes next, including comprehensibility, clarity (lack of ambiguity), opportunity to respond to a wide range of response options phrased in both the positive and negative directions, and lack of cognitive or emotional burdens.[37]

A paper which introduces a new scale and does not report its reliability is incomplete, and should be read with a healthy dose of skepticism. *Cronbach's Coefficient* α for internal consistency is a useful measure in scales that tap a single dimensional construct. However, in multidimensional measures such as HRQL instruments, where the items do not necessarily correlate closely with each other, a relative low value of alpha can lead to situations where a measure is wrongly dismissed for not being reliable. Conversely, because alpha is sensitive to the length of the instrument, a long scale may have a high value even in the absence of homogeneity. [43] Therefore, an optimal value of alpha to measure the internal consistency of a measure is a necessary but not a sufficient index of reliability. Alpha values over 0.9 most likely indicate unnecessary redundancy.[43,44] Good test-retest reliability confirms that the scale is stable over time, assuming that nothing has changed in the interim. The values should be higher than 0.6 when measured with the *intra-class correlation coefficient* (ICC). Inter-rater reliability is required when another person, such as a health professional, administers the test. The values should be over 0.6, and ideally over 0.7, when measured with the ICC.[37] Reliability is only maintained if the sample being tested resembles that in the original report.[37,43]

The reader is often confronted with the term 'construct validity'. This approach to validation of a measure is used

in the absence of a gold standard (where one would be able to establish 'criterion validity') and refers to predictions based on hypotheses. Construct validity is usually established over a number of studies, tapping various aspects of the hypothetical construct. Furthermore, the studies must show that the sample tested is similar to the groups with which it will ultimately be used, including the developmental age of the children. The measure should also provide evidence that it can discriminate within the group(s) of interest.

Finally, the clinician has to be convinced about the measure's utility. The implementation has to be feasible and acceptable to the potential user, the format user-friendly and the scoring simple.

(c) What specific HRQL measures in childhood epilepsy currently exist?

William Lennox can be credited with the first meaningful contribution to quality of life in epilepsy. Lennox recognized that the "...psychosocial obstructions in patients with epilepsy are as formidable as the seizure barriers". He encouraged clinicians to "match modern drug and surgical therapy with practical sociopsychological therapy", and concluded, "... the good physician is concerned not only with turbulent brainwaves but with disturbed emotions and with social injustice."[45] His vision was to have epilepsy and patients with epilepsy accepted socially and in the workplace. He further realized that treatment of children with epilepsy is broader in scope than that of adults in that the whole family is involved.[46]

The first significant attempt to measure psychosocial adjustment in people with epilepsy was the *Washington Psychosocial Seizure Inventory* in 1980.[47] The *Adolescent Psychosocial Seizure Inventory* is based on it and contains the same domains.[48] Comprehensive quality of life instruments for adults with epilepsy started being developed in the early 90s[49,50] and have helped assess the well-being of people with epilepsy in the clinical setting,[51] to evaluate the benefit of epilepsy surgery [52] and to examine the impact of anti-epileptic drugs (AEDs).[18]

Condition-specific scales for measuring HRQL in CWE have been developed to focus on problems relevant to CWE, to detect changes that one would like to assess including evaluation of different therapies. For most of these instruments health professional 'experts', with or without the input of families, have chosen the items and domains.[48,53–57] Only two studies have specifically attempted to identify the attributes of HRQL in children with epilepsy. Ronen et al. used separate focus groups for CWE, ages 6–10 years, and their parents, in order that each could discuss their own perceptions of life with epilepsy.[40] Textual analysis of the raw data enabled the researchers to extract, understand, explain, and categorize the HRQL elements. Five dimensions were identified: (1) the experience of epilepsy; (2) life fulfillment and time use; (3) social issues; (4) impact of epilepsy; and (5) attribution. The different HRQL elements are accompanied by sample quotations from the focus group discussions.[32] In their follow-up study, 381 CWE and their parent(s) independently completed a 67 item HRQL questionnaire. Factor analyses revealed five HRQL dimensions which the children considered most important: (1) interpersonal/social impact; (2) areas of worries and concerns; (3) intrapersonal/emotional consequences; (4) issues of keeping epilepsy a secret; and (5) quest for normality and resilience. Factor analysis of the parents' reports of their children's HRQL identified only the first four factors. In addition, the parents thought their children were worried as much about the future as about present issues whereas in fact the children worried almost exclusively about present matters.[42] Arunkumar et al. asked 80 parents of CWE, ages three months to 20 years (median 10), and 48 of the children to list their concerns about living with epilepsy in order of importance. The burdens and concerns were listed in order of frequency and helped establish questionnaires for parents and children.[58]

Details of the current HRQL childhood epilepsy measures and scales are summarized in Table 1. The psychometric properties of four of the instruments lack either reliability or validity data.[16,53,57,58] Only two of the psychometrically sound measures use a self-response questionnaire,[42,56] and only one [42] has parallel questionnaires for the children and the parents to report independently. The utility of the instruments has yet to be tested and one has still to demonstrate that any of these measures can better detect changes in the quality of life of CWE compared to generic HRQL measures.

(d) What do studies using HRQL scales tell us and not tell us?

In the past few years, there has been a progressive increase in appreciation of the importance of including patient preferences and values into healthcare management. Although there are many important studies reporting the psychosocial outcome of CWE, we have identified only a few that attempted to measure the HRQL using either generic or specific instruments. The samples in these studies were mostly relatively small and in two reports consisted of the same samples used for the development of the measures.[56,57] Using the parent-proxy generic CHQ [28] to assess outcome in 33 children following epilepsy surgery, Gilliam et al [59] found significantly lower (i.e., poorer) scores in the domains of emotional impact on parents, time impact on parents, and the general health index. There were also decreased scores for the

Table 1: HRQL measures and scales in children and adolescents with epilepsy

Scales	Purpose: to assess-	Domains	Item source	Sample size	Ages (yrs)	Items	Reliability	Validity	Respondents	Report
Batzel et al [48]	Psychosocial problems in adolescents with epilepsy	1. family adjustment 2. emotional adjust. 3. interpersonal 4. Vocation outlook 5. school adjustment 6. adjust to seizure 7. management 8. antisocial activity	Correlation of experts and patients	120	12–19	38	Internal consistency, Test-retest, Inter-rater	Face, Construct	Adolescents	Rater
Hoare & Russell [53]	Impact of epilepsy on child & family	Impact of 1. epilepsy /treatment 2. child development & adjustment 3. parents 4. family	Expert	21	6–17	30	none	Face	Parents	Proxy
Carpay et al [16]	Disability due to restrictions		Parents	122	4–16	10	Internal consistency, Test-retest	Face, content	Parents	Proxy
Camfield et al [54,55]	Impact of epilepsy/ childhood neurological. disability on family	1. outside activities 2. social 3. home life	Existing scale, Expert	97	2–16/18	11/44	Internal consistency, Test-retest	Construct	Parents	Self
Arunkumar et al [58]	HRQL in children & adolescents		Parents & children	80 p 48 c	3 months -20	20 each	none	Face, content	Parents & children	Self & proxy
Cramer et al[56]	HRQL in adolescence	1. impact 2. memory 3. attitude 4. physical 5. stigma 6. health 7. behaviour 8. social support	Expert & Focus-groups of adolescents	197	11–17	48	Internal consistency, Test-retest	Construct	Adolescents	Self
Sabez et al [57]	HRQL in children with intractable epilepsy	1. physical 2. emotional 3. cognitive 4. social 5. behaviour	Expert & families	63	4–18	77	Internal consistency	Construct	Parents	Proxy
Ronen et al[42]	HRQL in children	1. interpersonal/social 2. worries/concerns 3. emotional 4. secrecy 5. normality/resilience	Focus-groups of Children & parents	381 c 424 p	8–15	25 each	Internal consistency, Test-retest	Construct	Children & parents	Self & proxy

c – Children p – Parents

domains of self-esteem, general behaviour and the physical function index. The authors could not explain the nature and significance of the lower scores and concluded that although the HRQL may have improved following surgery, the intervention alone did not allow the children to achieve the levels of healthy controls. This study, however, did not explore which subgroups of children, if any, and what HRQL domains, improved following the surgery. Using the same parent-proxy CHQ Miller et al [60] compared 41 CWE ages 4–19 years to healthy controls. Most of the patients had severe epilepsy and 54% had co-morbid neurological impairments. Of the epilepsy related factors, co-morbid impairment and being on multiple AEDs were the best predictors for poor HRQL. Non-epilepsy related HRQL markers for comparison with the normal controls were unfortunately not included. Norrby et al.[61] compared 31 children, ages 9–13, with controlled epilepsy and without any co-morbidity with healthy controls in an attempt to assess their well-being. The Swedish well-being self-report visual analog scale of 39 items was used.[62] There were no differences between the children with epilepsy and the healthy controls. The obvious question is whether the measure used was appropriate and sensitive enough to answer the research question and whether the sample size was sufficiently large to detect any differences.

Devinsky et al.[63] attempted to assess the risk factors for poor HRQL in 197 adolescents with epilepsy. These researchers correlated AED toxicity, sociodemographic, academic and social variables, as well as epilepsy and other health-related variables, with self-reported HRQL. Older age, lower socioeconomic status, increased seizure severity, and AED neurotoxicity were associated with poorer HRQL. Potential remediable factors responsible for the lower HRQL in older adolescents and those of lower socioeconomic status, which could potentially improve following an intervention, were not identified in this study.

Sabaz et al.[64] compared the HRQL of children with refractory epilepsy with and without mild or moderate

intellectual disability. The sample consisted of 94 children, 68% of whom had normal intelligence. Using their own parent-proxy HRQL scale and two behaviour scales, the authors concluded that epilepsy and intellectual deficiency are independently associated with decreased HRQL scores.

Sherman et al.[65] tested 44 children with refractory epilepsy with three different parent-rated HRQL-impact scales: the generic *Impact of Childhood Illness Scale (ICI)*; the *Impact of Child Neurologic Handicap Scale (ICNH)*[55]; and the *Hague Restrictions in Childhood Epilepsy Scale (HARCES)* [16]. This retrospective sample, which lacked information in certain demographic and clinical data, consisted of 18 children before an intervention of surgery or vagal nerve stimulator, 11 after the intervention and 15 who were assessed for other reasons. The *HARCES* correlated best and the *ICNH* least well with the available neurological variables, whereas all three scales correlated similarly with psychosocial variables. We did not find this study helpful in guiding the clinician to choose the appropriate measure for clinical or research use.

Pal et al. [66] studied the social integration of CWE in rural India. Disability field workers listed normal day-to-day activities of children in their village. A large list of items was extracted and then condensed into five sections by sex and age group and tested on healthy controls. Mothers of CWE rated these activities. All groups of CWE had significant social deficits. The boys had limited peer group activities, and parents conferred fewer responsibilities to school age and adolescent children compared to controls. In preschoolers parental overprotection was reported. The nature of the social deficits was beyond the constraints imposed by the neurological impairment. There was also no relationship between the social integration and seizure frequency, nor was there any association with AED treatment. Parental attitudes toward their children were found to be negative in 25%. Fieldwork helped improve parents' attitudes with an increase in social opportunities for the children, such as a 50% increase in CWE attending school at the end of one year of intervention.

In another study involving the same families, the same research group measured mothers' satisfaction with social support from informal sources, and correlated it with a parental adjustment measure. They found positive independent correlation of satisfaction with social support and negative correlation with number of lifetime seizures and neurological impairment accounting for 34% of the variance. The lifetime seizures and neurological impairments were found to be dependent variables.[67] The authors argue that parental adjustment is an important target for intervention because it influences outcomes both for the CWE and the family.

In developing countries, negative attitudes and stigma appear to be more prevalent compared to the western world.[68–70] Parental adjustment is a particularly important target because of these negative attitudes toward disability in general and epilepsy in particular. For example, in surveys from India[69] and Taiwan[70] 15% and 7% of respondents, respectively, believed epilepsy to be a form of insanity; 40% and 18%, respectively, believed that CWE should not go to school or that their children should not play with them; and 66% and 72%, respectively, objected to their children marrying someone who had ever had epilepsy. In Taiwan, 31% believed that people with epilepsy should not be employed in jobs as other persons are. Support within existing social networks, and meeting informally with other families with similar predicaments, are potentially very appropriate interventions for a community based setting in developing countries. In their study Pal et al. [67] speculated that other factors as yet unknown, might be as important in the process of adjustment. These two studies by Pal and colleagues illustrate that effective interventions with innovative use of existing community resources could improve the HRQL and the psychosocial outcome in CWE, and that these interventions can be inexpensive and therefore suitable for developing world societies.[67] Furthermore the changes can be measured validly.

(e) What are the future directions in HRQL research of CWE?

Despite recent achievements in developing HRQL measures, there is a need to improve our understanding of the functional and experiential dimensions associated with complex neurodevelopmental disorders.[27] It is difficult to attribute better or poorer quality of life to the nature of epilepsy alone, when so many disparate factors play key roles in people's lives. These factors include, among others, a child's resilience, co-morbid conditions, parental well-being, family factors, attitudes and societal/cultural variables. Recent studies have shown that relationships between clinical symptoms such as seizure frequency and severity, or other biomedical markers, have only moderate correlations with HRQL.[42] Furthermore, HRQL may change over time with the development of the child and the family's accommodation to the situation. In addition, we need to learn what truly encompasses comprehensive patient care, define the goals of management, and attempt to evaluate the impact of interventions wherever possible.

Recently researchers have issued a call to develop better theory driven models of HRQL and to identify measurable factors that have important correlation with HRQL.[71] A model such as the one proposed here (Figure 1) would

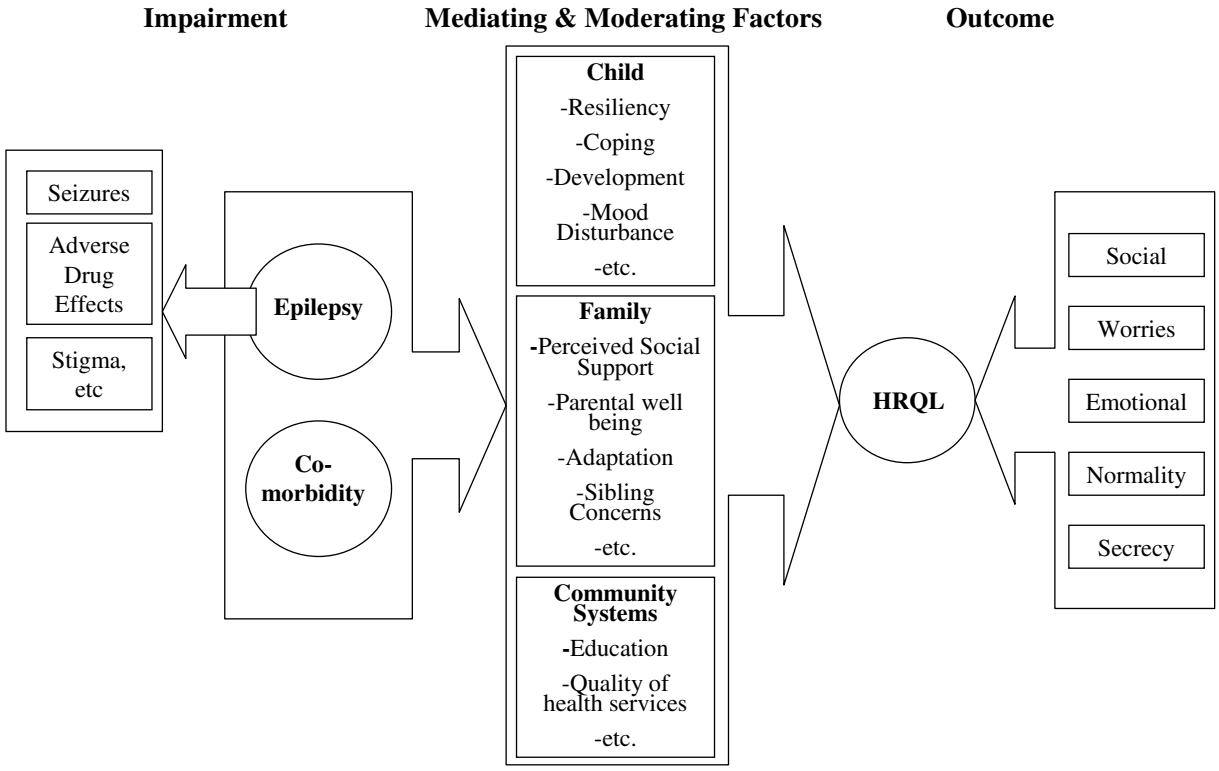

Figure 1
Conceptual model of quality of life in childhood epilepsy.

allow researchers to test how factors fit together, and make it possible to evaluate the predictive validity of that model. One should use or create systems that account simultaneously for the many factors that impinge on HRQL; test large cohorts in cross sectional, longitudinal and experimental designs; and apply contemporary scientific measurement procedures and complex statistical techniques to the evaluation of the relationships among the variables explored.

Contemporary assessments should include measures where the items originated from CWE, and provide for the children to rate their own HRQL.[34,42] However, parent-proxy report measures may prove to be a useful complement to the child's self assessment. Although the child and parent perspectives may be different, resulting in different scores, both are potentially valid.[34,42] The combination of self-report and parent responses – especially differences between the two sources of information – may give better insight into the family dynamics of coping with epilepsy, may better identify particular issues, and may lead to specific family counseling. Future research is therefore needed to examine the potential advantage of using both scales together, and to identify what factors contribute to a difference in scoring between children and parents. Whenever parents' reports are used alone, such as with children who are unable to respond independently, the clinician should be aware that although parental perspectives are important, parents are not true substitutes for reporting the HRQL of their children, and that relying on parents' reports alone may result in an incomplete HRQL assessment. This is true because certain perceptions of the children, such as their quest for normality and resilience, will be overlooked by parents.[42] One needs also to examine the emerging evidence that younger children's responses and probably those of children functioning at

younger developmental levels correlate less well with their parents' views than do those of older children, suggesting caution when interpreting proxy responses of younger age groups.[42] Parents take an important role in medical decision-making by defining what they believe to be the most appropriate treatment for their child and by evaluating the relative success of that treatment. This is another important reason for the need to be able to evaluate the validity of parent-proxy assessment of children's HRQL.[42]

On a more practical level, there is need to test the strengths and weaknesses of the different available instruments, understand what they truly measure, test their stability and sensitivity, and examine whether they provide similar or complementary information to generic HRQL and Health Status measures. Translations and cross-cultural adaptations of existing measures for use in different countries are also desirable in order to make comparisons possible across studies or to aggregate data, but this requires extensive work to establish true comparability.[72]

Innovative strategies involving qualitative research methodologies, either alone or in combination with quantitative approaches, are needed to further our knowledge of HRQL in developing countries. From the methodological point of view, involving CWE in identifying their own burdens and concerns is paramount before any truly useful interventions are planned. Modified focus groups for CWE should be considered as a potentially feasible and powerful tool to involve CWE in exploring their own HRQL.[40] Once we know the underling protective and risk factors that mediate HRQL, and the natural history of HRQL in children with epilepsy, we might be able to understand fully and to maximize HRQL of these children and their families. In addition it is very important to recognize the need to address opportunities for dissemination, translation of information and implementation of the knowledge into everyday clinical as well as research activities.

Clinical implications

To recap briefly, epilepsy is a complex neurological condition with many possible co-morbid features. Thus in addition to the need to address the etiology and treatment of seizures it has become increasingly recognized that professionals should attend to the impact of seizure disorders on the lives and well-being of children as they perceive the issues themselves. In childhood epilepsy, as in many related fields in clinical medicine, this interest has led to an effort to understand aspects of the condition beyond the biomedical dimension, and to do so by accessing the perceptions of the people who have the conditions. This expanded focus on both processes of service delivery and self-perceived outcomes is illustrated by the focus in adult health care on 'client centered practice' and in child health with the adoption of 'family centered services' as the standard for clinical practice. Without in any way diminishing the need to treat the 'impairments' associated with childhood epilepsy (the primary disorder and its associated co-morbidities) this approach broadens the scope of intervention to include concerns about the 'human dilemma' aspects of childhood epilepsy. By attending to the HRQL of CWE service providers have many additional possible 'points of entry' to support children and families. Instruments such as reliable and valid measures of the phenomena of interest provide tools to aid in the detection, assessment and follow-up of issues important to children and families. In applying these tools as professionals we will be expanding our understanding of the conditions for which we have much to offer, and still much to learn.

Acknowledgements

The authors thank Joan K Austin, Indiana University, for reviewing this manuscript and for the thoughtful and valuable comments.

References

1. Beilmann A, Napa A, Sööt A, Talvik I and Talvik T: **Prevalence of childhood epilepsy in Estonia.** *Epilepsia* 1999, **40:**1011-1019.
2. Sidenvall R, Forsgren L and Heijbel J: **Prevalence and characteristics of epilepsy in children in Northern Sweden.** *Seizure* 1996, **5:**139-146.
3. Sridharan R and Murthy BN: **Prevalence and pattern of epilepsy in India.** *Epilepsia* 1999, **40:**631-636.
4. Nicoletti A, Reggio A, Bartolini A, Failla G, Sofia V, Bartalesi F, Roselli M, Bamboa H, Salazar E, Osinaga R, Paradisi R, Tempera G and Dumas Hall AJ: **Prevalence of epilepsy in rural Bolivia. A door-to-door survey.** *Neurology* 1999, **53:**2064-2069.
5. Christianson AL, Zwane ME, Manga P, Rosen E, Venter A and Kromberg : **Epilepsy in rural South Africa children-prevalence, associated disability and management.** *S Afr Med J* 2000, **90:**262-266.
6. **Living Well With Epilepsy Conference.** *CDC & NCCPHP* 1997.
7. Pellock JM: **Antiepileptic drug therapy in the United States: A review of clinical studies and unmet needs.** *Neurology* 1995, **45(3 suppl 2):**S17-24.
8. Meador KJ: **Cognitive outcomes and predictive factors in epilepsy.** *Neurology* 2002, **58(8 suppl 5):**S21-26.
9. Tettenbaum B and Kramer G: **Total patient care in epilepsy.** *Epilepsia* 1992, **33(suppl 1):**S28-32.
10. Jones MW: **Consequences of epilepsy: Why do we treat seizures?** *Can J Neurol Sci* 1998, **25:**24-26.
11. Greenwood RS and Tennison MB: **When to start and stop anticonvulsant therapy in children.** *Arch Neurol* 1999, **56:**1073-1077.
12. Schachter SC: **Epilepsy: Quality of life and cost of care.** *Epilepsy behaviour* 2000, **1:**120-127.
13. Livingston S: **Comprehensive management of epilepsy in infancy, childhood and adolescence.** *Springfield Illinois, CC Thomas* 1972.
14. Austin JK, Smith MS, Risinger MW and McNelis A: **Childhood epilepsy and asthma: Comparison of quality of life.** *Epilepsia* 1994, **35:**608-615.
15. Mitchell WG, Chavez JM, Lee H and Guzman BL: **Academic underachievement in children with epilepsy.** *J Child Neurol* 1991, **6:**65-72.
16. Carpay HA, Vermeulen J, Stroink H, Brouwer OF, Peters AC, van Donselaar C, Aldenkamp A and Arts Wm: **Disability due to restrictions in childhood epilepsy.** *Dev Med Child Neurol* 1997, **39:**521-526.
17. Jacoby A: **Epilepsy and the quality of life findings from study of people with well controlled epilepsy.** *Soc Sci Med* 1992, **34:**657-666.

18. Smith D, Baker G, Davies G, Dewey M and Chadwick DW: **Outcomes of add-on treatment with lamotrigine in partial epilepsy.** *Epilepsia* 1993, **34:**312-322.
19. Jacoby A, Baker G, Steen N, Potts P and Chadwick D: **The clinical course of epilepsy and its psychosocial correlates: Findings from a UK community study.** *Epilepsia* 1996, **37:**148-161.
20. Jalava M, Sillanpää M, Camfield C and Camfield P: **Social adjustment and competence 35 years after onset of childhood epilepsy: A prospective controlled study.** *Epilepsia* 1997, **38:**708-715.
21. Kokkonen J, Kokkonen ER, Saukkonen AL and Pennanen P: **Psychosocial outcome of young adults with epilepsy in childhood.** *J Neurol Neurosurg Psych* 1997, **62:**265-268.
22. Wakamoto H, Nagao H, Hayashi M and Morimoto T: **Long-term medical, educational and social prognoses in childhood onset epilepsy: a population-based study in a rural district of Japan.** *Brain Dev* 2000, **22:**246-255.
23. Eriksson KJ and Koivikko MJ: **Prevalence, classification and severity of epilepsy and epileptic syndromes in children.** *Epilepsia* 1997, **38:**1275-1282.
24. Waaler PE, Blom BH, Skeidsvoll H and Mykletum A: **Prevelance, classification and severity of epilepsy in children in western Norway.** *Epilepsia* 2000, **41:**802-810.
25. Mims J: **Self-esteem, behavior, and concerns surrounding epilepsy in siblings of children with epilepsy.** *J Child Neurol* 1997, **12:**187-192.
26. Hoare P: **Psychiatric disturbance in the families of epileptic children.** *Dev Med Child Neurol* 1984, **26:**14-19.
27. Ronen GM, Rosenbaum P and Streiner DL: **Outcome measures in pediatric neurology: Why do we need them?** *J Child Neurol* 2000, **15:**775-780.
28. Langraf NJ, Abetz L and Ware JE: **The child health questionnaire (CHQ): A user's manual.** *Boston, The Health Institute, New England Medical Center* 1996.
29. Varni JW, Seid Michael and Rode CA: **The PedsQL ™: Measurement model for the pediatric quality of life inventory.** *Med Care* 1999, **37:**126-139.
30. Carpay JA and Arts WFM: **Outcome assessment in epilepsy: Available rating scales for adults and methodological issues pertaining to the development of scales for childhood epilepsy.** *Epilepsy Res* 1996, **24:**127-136.
31. Bradlyn AS, Ritchey AK, Harris CV, Moore IM, O'Brien RT, Parsons S, Patterson K and Pollock BH: **Quality of life research in pediatric oncology. Research methods and barriers.** *Cancer* 1996, **78:**1333-1339.
32. Ronen GM, Rosenbaum P, Law M and 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:**554-559.
33. Eiser C: **Children's quality of life measures.** *Arch Dis Child* 1997, **77:**350-354.
34. Rosenbaum P and Saigal S: **Measuring health related quality of life in pediatric populations.** In: *Quality of life and pharmacoeconomics in clinical trials* 2nd edition. Edited by: *Spilker B. New York: Lippincott-Raven*; 1996:785-791.
35. Eiser C and Morse R: **A review of measures of quality of life for children with chronic illness.** *Arch Dis Child* 2001, **84:**205-211.
36. Clancy CM and Eisenberg JM: **Outcome research: measuring the end results of health care.** *Science* 1998, **282:**245-246.
37. Streiner DL: **A checklist for evaluating the usefulness of rating scales.** *Can J Psychiatry* 1991, **38:**140-148.
38. McLauglin JF and Bjornson K: **Quality of life and developmental disabilities,.** *Dev Med Child Neurol* 1998, **40:**435.
39. Young NL, Yoshida KK, Williams , Bombardier C and Wright JG: **The role of children in reporting their physical disabilities.** *Arch Phys Med Rehabil* 1995, **76:**918-935.
40. Ronen GM, Rosenbaum P, Law M and Streiner DL: **Health-related quality of life in childhood disorders: A modified focus group technique to involve children.** *Qual Life Res* 2001, **10:**71-79.
41. Juniper EF, Guyatt GH, Feeny DH, Griffith LE and Ferrie PJ: **Minimum skills required by children to complete health-related quality of life instruments for asthma: Comparison of measurement properties.** *Eur Respir J* 1997, **10:**2285-2294.
42. Ronen GM, Streiner DL, Rosenbaum P and the Canadian Pediatric Epilepsy Network: **Health-related quality of life in children with epilepsy Development and validation of self-report and parent proxy measures.** *Epilepsia* 2003, **44:**598-612.
43. Streiner DL: **Starting at the beginning: An introduction to coefficient alpha and internal consistency.** *J Personality Ass* 2003, **80:**99-103.
44. Streiner DL: **Being inconsistent about consistency: When coefficient alpha does and doesn't matter.** *J Personality Ass* 2003, **80:**217-222.
45. Lennox WG and Markham CH: **The sociopsychological treatment of epilepsy.** *JAMA* 1953, **152:**1690-1694.
46. Millichap JG: **William Lennox.** In: *The founders of child neurology* Edited by: *Ashwal S. San Francisco: Norman Publishing*; 1990:758-763.
47. Dodrill CB, Batzel LW, Queisser HR and Temkin N: **An objective method for the assessment of psychological and social problems among epileptics.** *Epilepsia* 1980, **21:**123-135.
48. Batzel LW, Dodrill CB, Dubinsky BL and Queisser HR: **An objective method for the assessment of psychosocial problems in adolescents with epilepsy.** *Epilepsia* 1991, **32:**202-211.
49. Devinsky O, Vickrey BG, Cramer J, Perrine K, Hermann B, Meador K and Hays RD: **Development of the quality of life in epilepsy inventory.** *Epilepsia* 1995, **36:**1089-1104.
50. Baker G, Smith DF, Dewey M, Jacoby A and Chadwick DW: **The initial development of a health-related quality of life model as an outcome measure in epilepsy.** *Epilepsy Res* 1993, **16:**65-81.
51. Leidy NK, Elixhauser A, Vickrey B, Means E and William MK: **Seizure frequency and the health-related quality of life of adults with epilepsy.** *Neurology* 1999, **53:**162-166.
52. Vickrey BG, Hays RD, Rausch R, Engel J Jr, Visscher BR, Ary CM, Rogers WH and Brook RH: **Outcomes in 248 patients who had diagnostic evaluation for epilepsy surgery.** *Lancet* 1995, **346:**1445-1449.
53. Hoare P and Russell M: **The quality of life of children with chronic epilepsy and their families: Preliminary findings with a new assessment measure.** *Dev med Child Neurol* 1995, **37:**689-696.
54. Camfield C, Breau L and Camfield P: **Impact of pediatric epilepsy on the family: A new scale for clinical and research use.** *Epilepsia* 2000, **42:**104-112.
55. Camfield C, Breau L and Camfield P: **Assessing the impact of pediatric epilepsy and concomitant behavioral, cognitive, and physical/neurologic disability: Impact of childhood neurologic disability scale.** *Dev Med Child Neurol* 2003, **45:**152-159.
56. Cramer J, Westbrook L, Devinsky O, Perrine K, Glassman MB and Camfield C: **Development of the quality of life in epilepsy inventory for adolescents: The QOLIE-AD-48.** *Epilepsia* 1999, **40:**1114-1121.
57. Sabez M, Cairns DR, Lawson JA, Nheu N, Bleasel AF and Bye AM: **Validation of a new quality of life measure for children with epilepsy.** *Epilepsia* 2000, **41:**765-774.
58. Arunkumar G, Wyllie E, Kotagal P, Ong HT and Gilliam F: **Parent- and patient-validated content for pediatric epilepsy quality-of-life assessment.** *Epilepsia* 2000, **41:**1474-1484.
59. Gilliam F, Wyllie E, Kashden J, Faught E, Kotagal P, Bebin M, Wise M, Comair Y, Morawetz R and Kuzniecky R: **Epilepsy surgery outcome: Comprehensive assessment in children.** *Neurology* 1997, **48:**1368-1374.
60. Miller V, Palermo TM and Grewe SD: **Quality of life in pediatric epilepsy: Demographic and disease-related predictors and comparison with healthy controls.** *Epilepsy Behavior* 2003, **4:**36-42.
61. Norrby U, Carlsson J, Beckung E and Nordholm L: **Self-assessment of well being in a group of children with epilepsy.** *Seizure* 1999, **8:**228-234.
62. Wiklund I, Wiren L, Erling A, Karlberg J and Albertsson-Wikland K: **A new self-assessment questionnaire to measure well-being in children, particularly those of short stature.** *Qual Life Re* 1994, **3:**449-455.
63. Devinsky O, Westbrook L, Cramer J, Glassman M, Perrine K and Camfield C: **Risk factors for poor health-related quality of life in adolescents with epilepsy.** *Epilepsia* 1999, **40:**1715-1720.
64. Sabez M, Cairns DR, Lawson JA, Bleasel A and Bye A: **The health-related quality of life of children with refractory epilepsy: A comparison of those with and without intellectual disability.** *Epilepsia* 2001, **42:**621-628.
65. Sherman EMS, Slick DJ, Connolly MB, Steinbok P, Camfield C, Eyrl KL, Massey C and Farrell K: **Validity of three measures of health-related quality of life in children with intractable epilepsy.** *Epilepsia* 2002, **43:**1230-1238.

66. Pal DK, Chaudhury G, Sengupta S and Das T: **Social integration of children with epilepsy in rural India.** *Soc Sci Med* 2002, **54:**1867-1874.
67. Pal DK, Chaudhury G, Das T and Sengupta S: **Predictors of parental adjustment to children's epilepsy in rural India.** *Child Care Health Dev* 2002, **28:**295-300.
68. Austin JK, Shafer PO and Deering JB: **Epilepsy familiarity, knowledge, and perceptions of stigma: Report from a survey of adolescents in the general population.** *Epilepsy Behaviour* 2002, **3:**368-375.
69. Gambhir SK, Kumar V, Singhi PD and Goel RC: **Public awareness, understanding & attitudes toward epilepsy.** *Indian J Med Res* 1995, **102:**34-38.
70. Chung MY, Chang YC, Lai YH and Lai CW: **Survey of public awareness, understanding, and attitudes toward epilepsy in Taiwan.** *Epilepsia* 1995, **36:**488-493.
71. Lawford J and Eiser C: **Exploring links between the concepts of quality of life and resilience.** *Ped Rehab* 2001, **4:**209-216.
72. Bullinger M, Anderson R, Cella b and Aaronson N: **Developing and evaluating cross-cultural instruments from minimum requirements to optimal.** *Qual Life Res* 1993, **2:**451-459.