Development and Pilot-Testing of a Health-Related Quality of Life and Coping Inventory for Children and Adolescents with Chronic Health Conditions

A European Perspective

Dissertation
zur Erlangung der Würde des Doktors der Philosophie
der Universität Hamburg

vorgelegt von
Corinna Petersen
aus Husum

Hamburg, 2003

Referentin: Prof. Dr. Monika Bullinger, Universität Hamburg

Koreferent: Prof. Dr. Mick Power, University of Edinburgh

Tag der mündlichen Prüfung: 07.07.2003

Acknowledgements

Within the 5th Framework Programme "Quality of Life and Management of Living Resources" the European Commission supported the DISABKIDS project on which this thesis is based. The study could not have been conducted without this cross-national cooperation. Furthermore, the DAAD (Deutscher Akademischer Auslandsdienst) funded my research time in Edinburgh in 2001. My thesis benefited very much from the experience I gained in the United Kingdom.

This study would have been impossible to carry out without the support of a large number of people. First and foremost, I would like to thank the children and parents for allowing the project team to conduct the interviews and questionnaire assessment. Secondly, special thanks to the whole DISABKIDS Group for supporting my work on coping and allowing me to use the data on health-related quality of life for the psychometric testing reported in this thesis. I would like to thank my thesis supervisors, Monika Bullinger and Mick Power, for their encouragement and helpful suggestions. I wish to express sincere appreciation to Monika Bullinger for her guidance throughout the last years. Thanks to my colleague and friend, Anja Mehnert, for her constant reassurance and valuable input. Christiane Ewert and Peggy Cooke made excellent suggestions for improving my English grammar. I would also like to thank my parents for always being there for me. Finally, heartfelt thanks to Karsten for his support and understanding.

Corinna Petersen Hamburg, March 2003

Table of Contents

1 Introduction

As the prevalence of paediatric chronic health conditions is increasing, a significant proportion of children and adolescents are affected by chronic health conditions. A child or adolescent with a chronic illness has to cope with psychological, social, and physical consequences related to having a chronic health condition. The assessment of such consequences and their effect on the young peoples' health-related quality of life is a major task for medical research. Historically, the emphasis in medical research was oriented towards cure and survival. With increasing criticism and growing acceptance of new health outcome parameters, such as health-related quality of life, the focus of health outcome measurement shifted. Health-related quality of life is increasingly considered as an important health outcome parameter in medicine. However, while theory and research on children with chronic health conditions and disabilities has grown in recent decades, adequate assessment methods for outcome measures still need to be provided. Furthermore, predictors of health-related quality of life need to be identified. Concerning this, the role of coping strategies with regard to the adjustment process to a chronic health condition has been the focus of research over the past years. A relatively new area of research is the investigation of the relationship between coping and health-related quality of life. The way children or adolescents cope with their illness might be responsible for a great variation in their subjective health. This thesis is based on a project funded by the European Commission ("DISABKIDS") which aims at the cross-national understanding of children's and adolescents' health-related quality of life. Within this project the thesis focuses on the development and testing of a coping inventory for children/ adolescents as well as on the psychometric testing of a chronic generic HRQOL measure which was developed by all project partners conjointly. It is hoped that the development of a coping and a health-related quality of life instrument will facilitate assessment of health-related quality of life and coping with a chronic health condition in future studies and will help to understand the relationship between coping and health related quality of life. For the ease of reference throughout this thesis health-related quality of life is referred to as HRQOL.

2 Health-Related Quality of Life and Coping in Chronic Paediatric Health Conditions

2.1 Chronic Health Conditions in Children and Adolescents

Advances in medical care have changed the focus of paediatric medicine from the treatment of infectious diseases to the management of chronic health conditions (Eiser & Morse, 2001a). While technological advances have allowed an increasing number of children with chronic health conditions to reach adulthood, research on the long-term consequences of chronic health conditions is needed. Such results could contribute to improve care by taking the needs of young people into account. Children and adolescents with a chronic health condition require co-ordinated care that involves multiple health care providers. An appropriate transition to adult health care is certainly one of the major tasks for health care providers in the future. Research and theory on the adjustment of young people with chronic health conditions has grown considerably in recent years addressing the impact of a chronic disease on emotional, social, physical well-being or psychosocial risks (Lavigne & Faier-Routman, 1992). Chronic conditions can confront the whole family with extra demands and specific worries. These demands can rule the families' life and can have an impact on the parental well-being, the financial security, and interfamily relationships (Eiser, 1993). Family dynamics in return may influence treatment outcome.

Several factors have to be taken into account conjointly when investigating children with chronic health conditions in order to explain adjustment or maladjustment to chronic health conditions. Research so far has concentrated on maladaptive factors with regard to the adjustment process. For example, Barbarin (1990) found that frequent hospitalisation, intrusive medical procedures, and the uncertainty of survival have a negative influence on the adjustment process in childhood development. Interestingly, physical dysfunction has not necessarily had a negative impact on childhood development adjustment (Drotar et al., 1981). In sum, adjustment to a chronic disease is a complex process and a number of factors contribute to the adap-

tation process. To ensure comparability of studies a clear definition of chronic health conditions is necessary.

2.1.1 Definition and Classification of Chronic Health Conditions

Paediatric chronic health conditions are the topic of a large number of studies. With regard to the concept, the terms “condition”, “illness”, and “disease” are often used interchangeably in paediatric literature. According to Perrin et al. (1993) the term “illness” implies physical symptoms such as pain, fever or fatigue whereas “disease” is sometimes associated with health problems of an infectious origin or with discomfort and pain. These authors suggest using the term “condition” preferably.

Chronic health conditions can be described in various ways and no existing definition is exhaustive (Perrin et al., 1993). Chronic health conditions are often defined as a condition lasting for an extended period, at least three months, often for life, and cannot be cured (Eiser, 1990; Midence, 1994). In addition, Pless and Pinkerton (1975) defined a chronic physical disorder as one that:

- interferes with daily functioning for more than three months in a year or
- causes hospitalisation lasting more than one month in a year or
- is thought to do either (a) or (b) at the time of diagnosis.

The three months criterion has proven to be useful (Pless & Satterwhite, 1975). Nevertheless, the exact identification of the disease duration can be complicated if the time of onset or diagnosis is unknown. Traditionally, chronic health conditions were categorised according to the affected organ or rather organ system (e.g. heart disease) and each chronic condition was viewed as a distinct entity. This condition-specific or categorical approach presented several problems in planning efficient health policies. For example, due to the rare prevalence of each individual condition, intervention programs had to be planned and conducted for different conditions conjointly. As a consequence, awareness was growing that children with different chronic health conditions have several problems in common. These commonalties

might be as important as condition-specific aspects.

Criticism defining chronic health conditions according to their diagnostic label has led to the development of a non-categorical, generic approach. This approach was influenced by the work of Stein and colleagues (1982; 1984; 1993; 1997) and is based on the observed thematic overlap between chronic health conditions. According to these authors, different chronic health conditions may share the nature of onset and course, visibility of the disorder, degree of life threat, pain of treatment or functional impairment. Perrin and colleagues (1993) suggest that children and adolescents should be classified according to fourteen aspects placed on continua: duration, age of onset, limitation of age-appropriate activities, visibility, expected survival, mobility, physiologic/ emotional/ social/ sensory functioning, cognition, communication, course, and uncertainty. The issues patients with different conditions have in common seem to reflect the chronicity itself rather than aspects of a specific disease.

2.1.2 Prevalence of Chronic Health Conditions

In the industrialised countries the percentage of children with severe chronic health conditions has more than doubled in the past years (Perrin & Shonkoff, 2000). While in adults physicians usually have to deal with a small number of frequent types of chronic health conditions (e.g. diabetes, coronary artery disease), the range of chronic childhood diseases is broader. According to Perrin & Shonkoff (2000) only allergic and neurological health conditions can be viewed as “common” and frequent disorders. Apart from the rarity of individual childhood diseases, the cumulative number of children with chronic health conditions is high. Epidemiological surveys show that 10-20% of children in the industrialised countries have a chronic health condition. Most of these conditions can be classified as mild or moderate, only 1-2% of the child population have severe chronic health conditions (Gortmaker & Sappenfield, 1984). According to the National Health Interview Survey conducted in the USA in 1988 an estimated 31% of young people were affected by a chronic health condition. Of these 66% were classified as a mild, 29% as a moderate and 5% as a severe

chronic health condition (Newachek & Taylor, 1992). Other authors found a prevalence of up to 10% for chronic health conditions in childhood (Gortmaker et al., 1990).

Results of the National Health Interview Survey (1992-1994) indicate that about 6.5% of children and adolescents experienced some degree of disability (Newacheck & Halfon, 1998). In 1986 it was reported that 3.8% of children and adolescents were affected by chronic health conditions that caused some limitation of activity (Newachek et al., 1986a). Data from the National Health Interview Survey indicate that the prevalence of activity-limiting chronic conditions among children and adolescents doubled from 1.8 to 3.8% between 1960 and 1981 (Newachek et al., 1986b). About 30% of the children in question suffer from two or more conditions (Newacheck & Taylor, 1992).

Discrepancies can arise with regard to the applied definition of chronic health conditions or different sources of information (e.g. parent vs. clinician judgement). However, the alarming increase in the proportion of children and adolescents with limitations of activity due to chronic illness can only be partially explained by the survey design or changes in the awareness of parents and physicians with regard to chronic health conditions (Newacheck et al., 1984).

2.1.3 Psychosocial Consequences of Chronic Health Conditions

Chronic childhood health conditions are a challenge for medical care not only in terms of the clinical task, but also because of their association with psychosocial problems (Lavigne & Faier-Routman, 1992). Childhood chronic illness can be a stressor that affects the children and families' well-being in physical, emotional, social, and functional domains. In research, psychosocial consequences of chronic health conditions have been studied with regard to effects on the child and on the family (Wallander & Thompson, 1995). According to Eiser (1990) much research in the paediatric field is still based on the assumption that chronic illness has a negative impact on the development of a child. Concepts such as "adjustment", "adaptation", "cop-

ing", "stress", and "competence" have been used to describe components of the process of dealing with a chronic health conditions (Eiser, 1990). Constructs like self-esteem, social isolation, behaviour problems and achievement at school have been included in theoretical frameworks as indices of adjustment to chronic illness.

Theoretical Concepts

Pless and Pinkerton (1975) and Moss and Tsu (see Moss, 1984) formulated the first theoretical concepts explaining psychosocial consequences and identifying contributing factors. The latter describe the diagnosis of a chronic health condition as a crisis. The way of coping with an illness is an important component in their theoretical reflections. In line with these assumptions, Pless and Pinkerton (1975) viewed chronic health conditions as stressors and proposed that self-concept and the way of coping with a chronic health condition are connected and influenced by personal factors, such as intelligence. Steinhausen (1994) formulated a model which consists of five central determinants: personality, family and social environment (which can be either a protective or risk factor) and two disease-related determinants, namely life events and stress. Depending on the way these factors interact, their influence can be positive or negative. More recently, two models are discussed in international literature: the transitional stress-coping model (Thompson et al., 1994; 1996) and the disability-stress-coping model of Wallander and Varni (1992; 1995; 1998).

The transitional stress-coping model views chronic disorder as a stressor to which the child and the family must adapt. Psychosocial adaptation processes such as coping behaviours, maternal adjustment, regulation of self-esteem, health locus of control as well as biomedical and developmental processes are assumed to influence the impact of a disease according to the model. It is hypothesised that the stress perceived by the family influences the psychosocial adaptation of the child. The model has been tested in children with sickle cell disease and cystic fibrosis (Thompson et al., 1993; 1994).

The disability-stress-coping model formulated by Wallander and colleagues (1992; 1995; 1998, see figure 1) views chronic health conditions as an ongoing strain for the

whole family. The families with a chronically ill child are at a greater risk of maladjustment because they are exposed to a higher number of stressors (Varni & Wallander, 1988). In their model, adaptation is defined as a multidimensional construct that compromises mental, social, and physical components. The framework is composed of general and condition-specific stressors. Risk factors include disease characteristics, functional impairment, and psychosocial stressors like daily hassles. Resistance variables in the model are intrapersonal factors, social ecological factors like social support, and stress processing factors such as coping strategies. The outcomes are presented by social, psychological, and physical adaptation. The model focuses on the reciprocal nature of the relationships between the variables. The risk variables have a direct effect on the adjustment process and interact with each other. The resilience variables moderate the relationship between impact and coping with stress. In addition, they have a direct effect on psychological stress and adaptation.

Due to the complexity of the model, components have only been tested separately so far (Wallander & Varni, 1998). For example Varni et al. (1995, 1999a) investigated children with cancer and found that disease-related parameters like the diagnosis (leukaemia versus other cancer types) did not have an important influence on adjustment. In another study, psychological stress had an influence on adjustment (Wallander & Varni, 1998). Even though these authors started to address important but so far neglected issues of chronic childhood disease, the one-sided assignment of either resilience or risk factors neglects situational components.

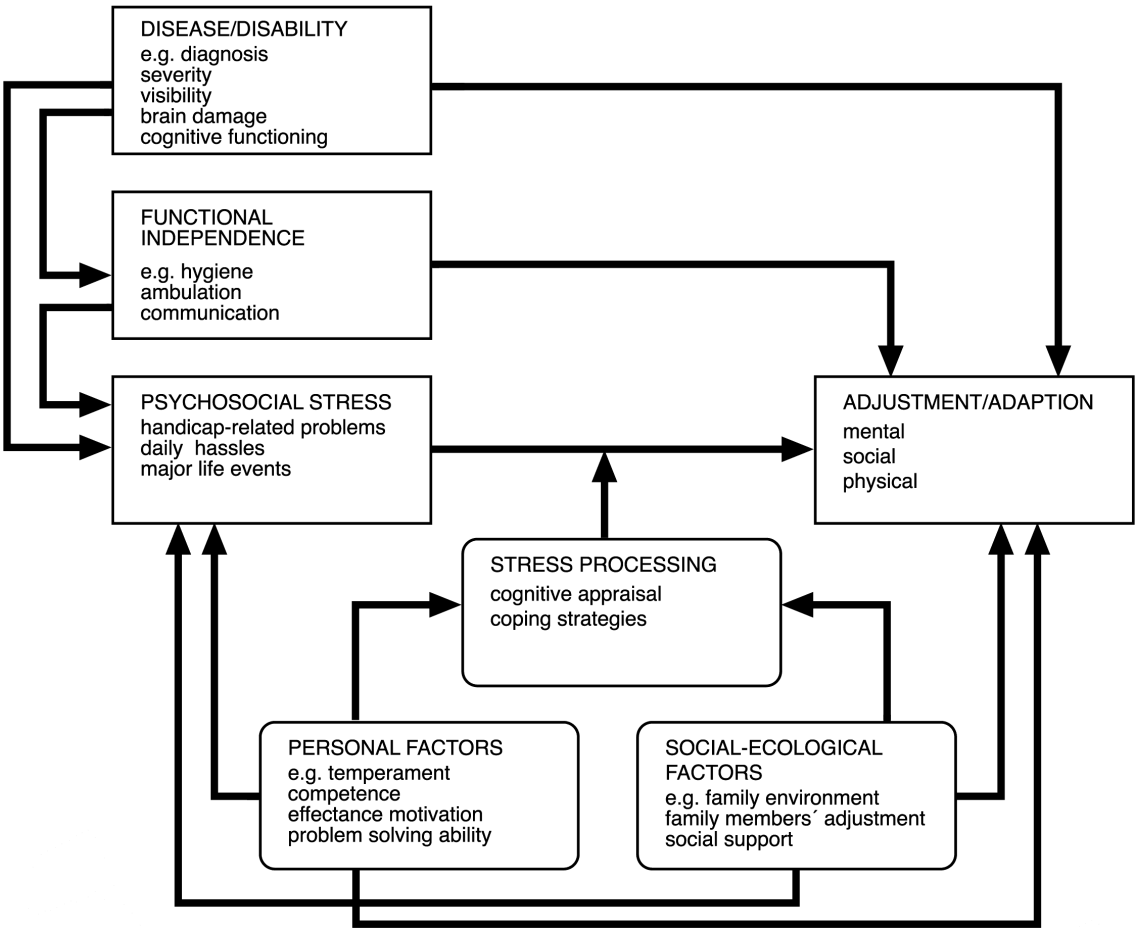


Figure 1

The disability-stress-coping model (Wallander & Varni, 1998). Round corner boxes indicate resistance factors, square-corner boxes indicate risk factors/ outcome variables.

Effects on the Child

The question of whether the prevalence of psychological problems in chronically ill children is higher has been addressed in some studies which confirmed this hypothesis. Cadman and colleagues (1987) found 22-31% of children with a chronic disorder to have a psychiatric disorder, compared to 14% in healthy children. Lavigne and Faier-Routman (1992) analysed 87 studies in a meta-analysis and concluded that children with a chronic health condition had more psychosocial problems than healthy children. A higher prevalence was especially evident for emotional, conduct, and hyper kinetic disorders. The latter was mainly found in North America. It might also be possible that the way of diagnosing this disease is different in North America

and consequently leads to a higher prevalence. According to Eiser (1990), children and adolescents with chronic conditions are more likely to exhibit signs of psychosocial maladjustment than their healthy peers. They are at a greater risk of psychiatric disorders and behavioural as well as emotional disturbances. In general, evidence supports the view of an increased vulnerability in terms of emotional and behavioural problems. Furthermore, the variability of findings indicates individual differences (Wallander & Varni, 1998).

With regard to consequences in school academic functioning, operationalised as school absenteeism or performance at school, has been examined. Up to now results underline no lower performance of chronically ill children, but more absenteeism at school (Boekaerts & Röder, 1999). However, the type of disease has a differential impact. For example, Children with epilepsy, sickle cell disease or spina bifida showed a lower performance at school compared with children with other chronic health conditions (Fowler et al., 1985). In terms of psychosocial consequences it has been suggested that restrictions to physical activity, unusual physical appearance, interruptions of daily activities, and changes in lifestyle might have an effect on peer relationships (La Greca, 1990).

Effects on the Family

The role of the family has been scrutinised in several studies (Kazak, 1989; Perrin et al, 1989; Thompson et al. 1993; Varni et al., 1996a). In recent years the focus of research has shifted from the investigation of negative effects towards a greater emphasis of family coping resources (Eiser, 1990). Although chronically ill patients and their families might be confronted with financial, physical, and psychological consequences, a lot of families adapt very well to their situation. Nevertheless, research showed that uncertainty of the child's future, marital disruption, problems with siblings, parental anxiety, family dysfunction, restrictions, and negative attitudes towards a chronic health condition may occur and affect everyday life (Eiser, 1990; Mitchell et al., 1994). Therefore, researchers suggest that the family should be the unit of intervention if psychosocial problems are observed (Patterson & Garwick,

1994). Most of the studies investigated the role of the mother and mother-child interaction (Eiser, 1990). Studies provided evidence that parents of chronically ill children are not more likely to divorce than other couples (Perrin & MacLean, 1988). Research that focused on the psychosocial consequences for healthy siblings (Eiser, 1990) brought no final conclusions, but underlined the need for further studies.

2.2 Health-Related Quality of Life (HRQOL) Research

Parallel to the paradigm shift in medical outcome criteria the World Health Organisation (WHO, 1999) reformulated the guiding principle "Add years to life" to "Add life to years". This turning point underscores the necessity to include HRQOL as an outcome criterion in medical research.

2.2.1 HRQOL- Definition and Conceptual Issues

The concept of quality of life has been of great interest during the last decades. The term “quality of life” has been widely used, but no universally accepted definition so far is available (Baker & Intagliata, 1982; Spilker, 1990; Aaronson, 1992; Felce & Perry, 1995). During the 1960s and 1970s mainly politicians and social scientists addressed quality of life to chart the well-being of populations. As an example in politics, US-President Lyndon Johnson used the term “quality of life” in one of his speeches to emphasise the importance of quality of life as compared to the quantity of goods (Campbell, 1981). The enhancement of quality of life became a major political goal in those years. In terms of measuring and evaluating quality of life researchers focussed on what they viewed as objective or external indicators such as housing, income and education (Campbell et al., 1976). It soon became clear that quality of life was not only constituted by objective factors. Consequently, Campbell and Rodgers (1976) highlighted the importance of psychological factors (e.g. satisfaction) which were taken into account in future research.

Furthermore, the definition of “health” provided by the World Health Organisation

as “a state of complete physical, mental, and social well-being, and not merely the absence of disease or infirmity” (WHO, 1948) was a milestone for quality of life research and influenced its definition. In addition, the need for a new medical model delineated by Engel (1977) provided impetus to the assessment of quality of life in medicine. In his review, Engel highlighted the disadvantages of a biomedical model and stated the need for a bio-psychosocial model of diseases.

Quality of life was no longer viewed to be the same as the standard of living. Instead it was regarded as a social construct about important aspects of life (Skevington, 2002). However, the distinction between quality of life and related concepts such as life satisfaction is difficult due to a conceptual overlap of both constructs (Schalock, 1996). The judgement of both constructs, i.e. rating one’s own quality of life or life satisfaction necessitates a comparison of the actual versus the expected state of a specific life domain.

Quality of life is often distinguished from the more specific concept of **health-related quality of life**. In medicine, quality of life clearly relates to health and the subjective well-being of a patient with regard to e.g. a treatment. For this reason, the term health-related quality of life (HRQOL) has been introduced to medicine (e.g. Feldmanet al., 2000; Guyatt et al., 1996). More specifically, HRQOL is a component of the more general construct quality of life which also includes a broader range of aspects such as political freedom and economical issues. However, it should be noted that some researchers do not support this differentiation. Koot and Wallander (2001) argue that HRQOL gives only information about the impact of a disease, but not about quality of life. From their point of view, a distinction would be arbitrary.

The World Health Organisation (WHO) and the International League Against Rheumatism (ILAR) reached a consensus on the definitions of HRQOL and quality of life. Quality of life is defined according to a needs-based model that identifies quality of life as the degree to which most human needs are met (Patrick et al., 1988; McKenna, 1994) similar to the definition of the World Health Organisation Quality of Life Assessment (WHOQOL) group which defined quality of life as:

"… individuals' perceptions on their position in life in the context of their culture and value systems in which they live, and in relation to their goals, expectations, standards, and concerns."

(The WHOQOL Group, 1995, p. 1405)

Currently, the term HRQOL is widely used when referring to a specific impact of an illness or injury, medical treatment, or health care policy on an individual's quality of life. HRQOL is defined as the physical, emotional, and social aspects of HRQOL influenced by an individual's disease and/ or its treatment (Strand & Russell, 1997). HRQOL is viewed as a **multidimensional construct.** Consensus has been reached with regard to the four core domains of quality of life, namely disease state, functional status, psychological, and social functioning (Eisen et al., 1979; Aaronson et al., 1991; Spieth & Harris, 1996, see figure 2).

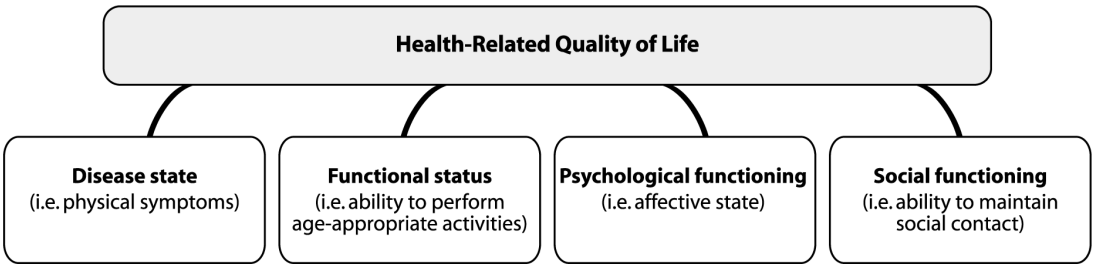


Figure 2
Domains of health-related quality of life

The self-assessment of the individuals concerned is important to capture their perception of health conditions and treatment regimes. Therefore, **subjective health** is often used as a synonym for HRQOL. HRQOL focuses on the patients' perceptions of their diseases and measures impairments that have significant impact on the patients. The constructs health status, functional status and well-being are often used interchangeably with HRQOL without taking the differences between these concepts into consideration (Guyatt et al., 1993; Patrick & Bergner, 1990). Generally,

HRQOL measures take the patient's views into account and accordingly do not focus only on functional capacity.

The objectives of HRQOL research can be described with regard to three different perspectives. The epidemiological perspective focuses on the description of the well-being and functioning of the population. For the clinical perspective, the evaluation of treatment effects is central. The health economical or health system perspective concentrates on in the analysis of the quality and costs of care (Bullinger, 1997a).

HRQOL research in medicine went through different stages of development. In general, four phases can be identified. During the first phase in the 1970s, concepts of HRQOL were discussed and established. In the 1980s the question of how to assess HRQOL was the main research interest. Instead of assessing HRQOL on an individual level a more pragmatic procedure was preferred in order to have adequate standardised assessment tools at hand. Subsequently, the application of the assessment tools began (Bullinger, 1997a). Nowadays, the phase of investigating the impact and clinical significance of HRQOL outcome has started (Symonds et al., 2002). The interpretability of HRQOL scores needs to be improved, especially in terms of their clinical significance. Although psychometrically tested questionnaires exist, it is still unclear whether most of these measures capture meaningful changes for groups or individuals. It is a challenge for research to determine the significance of any differences observed, and to disseminate this knowledge to clinicians.

HRQOL instruments have been developed for different purposes, e.g. for outcome assessment or program evaluation. Basically, two different models of HRQOL research exist: the utility and the health status measurement concept. The utility model developed by Kaplan (1989) is economically oriented and aims at the appropriate allocation of financial resources. It is derived from economic decision theory. During the assessment respondents are asked to state their preference, e.g. between a longer life with and a shorter life without a dysfunction. To quantify the results, responses are transformed into quality-adjusted years (QALY's). A very frequently used questionnaire for this approach is the Quality of Well-Being Scale (QWB, Kaplan et al., 1978). One of the main criticisms of this approach is the non-applicability for a paediatric population (Hinds, 1990; Richards & Hemstreet, 1994). In contrast, the health

status measurement model (Ware, 1984a; 1984b) focuses on the assessment of the impact of health care policies on public health and hence is also useful for paediatric populations. According to Drotar (1998) potential applications of HRQOL measures are health surveys, needs assessment, monitoring health status, evaluating care, and assessing a patient's experience with care.

2.2.2 Measurement of HRQOL in Children and Adolescents

With regard to the different objectives of HRQOL assessment, HRQOL measures vary in terms of their conceptualisation. According to Guyatt and Jaeschke (1990) as well as Spieth and Harris (1996) HRQOL measures can be classified across three dimensions: the type of report, scores, and population (see table 1). This taxonomy provides a useful classification of HRQOL instruments based on their scope and applicability.

Table 1
Dimensions of HRQOL instruments

Dimension	**Variation**
Conceptualisation	self-report vs. proxy-report
Classification of scores	single indicator, profile or battery approach
Population assessed	generic vs. condition-specific

The World Health Organisation provided a guideline for the development of quality of life instruments (WHO Division of Mental Health, 1993). The following general instrument criteria are:

- Instruments should be child-centred.
- Subjective self-report has priority.
- Instruments should be related to age and developmental stage.
- Results should be cross-culturally comparable.

- Instruments should have a generic core and specific modules.
- Positive health-enhancing aspects of HRQOL should be stressed.

In general, HRQOL measures can be divided into generic and condition-specific measures (Guyatt & Jaeschke, 1990). They can be further categorised into health profiles or preference-based measures. While generic instruments measure HRQOL across health conditions, condition-specific measures do so with regard to a specific disease, treatment or symptom. The disadvantage of generic measures may be that small changes in HRQOL might not be detected. On the other hand, condition-specific instruments may provide clinically relevant information, but comparison across illnesses is not possible (Bullinger, 1997a). Moreover, children may have more than one condition and this co morbidity complicates the development of condition-specific measures. Both types of measures have strengths and weaknesses; the choice of one type of measure depends on study aim and sometimes a combined approach is appropriate as well. Information can be obtained of the children or adolescents themselves (self-report) or of significant others like the parents (proxy-report). Self-report and proxy-report can differ from each other (Eiser & Kopel, 1997).

In a review about HRQOL instruments for children and adolescents, Eiser and Morse (2001a) included 137 papers. 43 of these papers involved the development of a new measure (19 generic and 24 condition-specific instruments). The authors concluded that there is still a lack of condition-specific measures for self-completion by children. Furthermore, the measurement of HRQOL in children as well as in adolescents should be examined with psychometric properties and whether the measures adequately relate to important domains of HRQOL in children and adolescents. Nevertheless, the development of HRQOL measures for children and adolescents is a rapidly growing area. Table 2 depicts only the most frequently used generic HRQOL measures for healthy and sick children.

Table 2

Generic HRQOL measures

Name & Author		Report	Age (y)	No. of Items (Domains)	Origin
AUQEI	Autoquestionnaire Qualité de Vie-Enfant-Imagé Magnificat et al. (1997)	Self	4-12	26	F
CHQ	Child Health Questionnaire Landgraf et al. (1998)	Self Parents	5-18	CF-87, PF-28, 50, 98 (14)	USA
CHIP-AE	Child Health and Illness Profile Starfield et al. (1995)	Self	11-17	153 (6)	USA
CQOL	Child Health-Related Quality of Life Graham et al. (1997)	Self Parents	9-15	15 (3 levels) (15)	UK
17D	Measure of Health-Related Quality of Life Apajasalo et al. (1996)	Self	8-11	17 (11 + index)	FIN
GCQ	Generic Child Quality of Life Measure Collier et al. (2000)	Self	6-14	25 (5)	UK
HAY	How Are You? Bruil, 1999	Self Parent	7-13	80 (10)	NL
HUI	Health Utilities Index Mark 3 System Feeny et al. (1998)	Self	4-18	15 (15)	CAN
KINDL	Questionnaire for Measuring HRQOL Ravens-Sieberer & Bullinger (1998)	Self Parent	4-7 8-12 13-16	24 + modules (6)	GER
PedsQL	Pediatric Quality of Life Questionnaire Varni et al. (1999b)	Self Parent	2-18	15 (core) (3)	USA
TACQOL	TNO-ACL Questionnaires Vogels et al. (1998)	Self Parent	6-15	108 (7)	NL
VSP-A	Perceived Health of Adolescents Siméoni et al. (2000)	Self	11-17	40 (9)	F

2.2.3 HRQOL and Paediatric Chronic Health Conditions

In comparison to adults, children's HRQOL assessment is a more recent area of research. The development of HRQOL measures is necessary for identifying children at risk and guiding efforts to improve HRQOL of disadvantaged populations. HRQOL assessment in children and adolescents is also a step towards patient participation when it is based on self-reports. Moreover, HRQOL assessment is necessary for understanding how young people perceive their situation and which aspects of their life affect their subjective health. The concept of HRQOL was initially developed with adult populations without considering younger populations. However, the type of activities and interactions of a child or adolescent compared with that of adults is different, so that a new or modified framework is necessary for meeting the needs of children and young people. Work in the children's area started with a debate. First, if this construct is relevant for children and second if children have the ability to reflect and express their own well-being and functioning. Similar to research into adults, early studies focussed predominately on functional assessment (Eiser & Morse, 2001b).

HRQOL research in young people was much influenced by the work in the field of paediatric oncology and neonatal intensive care. The increasing interest in HRQOL issues is reflected in the rising number of publications. Bullinger and Ravens-Sieberer (1995) conducted a literature search and reviewed research activities concerning HRQOL into children and adolescents. The authors identified over 20.000 HRQOL publications in medicine, of which 13% investigated HRQOL in children underscoring the rapid growth of this research area. Most of the identified publications were related to condition-specific topics, especially concerning oncology or transplantation medicine. Interestingly, publications about asthma, the most prevalent chronic paediatric health condition, were only ranked third place. Overall, in most of the studies, parent or staff report was obtained to provide information about children's' HRQOL and not self-report.

Overall, progress in paediatric HRQOL research was slow due to conceptual and operational difficulties (Drotar, 1998). These difficulties refer to age particularities,

proxy-report, and cognitive ability. With increasing age, for example the contents and importance of questionnaire dimensions change. In most cases, mothers or other caregivers provided information about the HRQOL of their children. Also experts, such as physicians, were questioned. This conceptualisation has been known as proxy-report. Discrepancies found between self- report and proxy rating raised the question of the value of proxy ratings (Eiser & Jenney, 1996). Parents and their children, although sharing the same environment, make different experiences. It is not clear whether children and parents share similar dimensions for describing their own HRQOL. Another important measurement characteristic of HRQOL is its multidimensionality. According to Schor (1998) different HRQOL domains are difficult to investigate in children because they are more interconnected than are the domains of adults' health. Furthermore, problems in the assessment of HRQOL can occur because children's competence in verbal comprehension and the understanding of time differences are not fully developed (Wallander et al., 2001). Questions often refer to a certain time frame (e.g. two weeks), but for children it is often not easy to remember the week before. Finally, age appropriate norms are often difficult to determine, i.e. a comparison between study populations and norm data is questionable. Furthermore, when measuring HRQOL with a questionnaire, a language that is appropriate to a child's age and developmental stage has to be used.

The range of the studied diseases with regard to HRQOL is wide but most studies are on asthma and epilepsy. Eiser, Vance and Seamark (2000) found lower HRQOL for children with asthma compared to their healthy peers (*n*=127). The authors assumed that HRQOL is the result of discrepancies between an individual's actual and ideal self. Sawyer et al. (2000; 2001) found a relationship between self-reported HRQOL of children and family functioning (*n*=236). Children with asthma showed a poorer HRQOL than healthy children. According to a study from Sabaz et al. (2001) children with epilepsy had a poor HRQOL regardless of their intellectual ability level (*n*=63). In addition, findings showed a decrement in HRQOL with increased seizures severity. Austin et al. (1994) found that children with epilepsy had a more compromised HRQOL in the psychological, social and school domain, whereas children with asthma showed limitations in the physical domain (*n*=270). The authors suggest that paying

attention only to seizure control is not sufficient for the full range of HRQOL problems in children with epilepsy. Ravens-Sieberer and Bullinger (1998) investigated HRQOL in children with diabetes or asthma compared to healthy children (*n*= 90). Findings supported the important role of coping strategies. Lindström and Eriksson (1993) investigated children with cystic fibrosis and myelomeningocele (*n*=951) in five Nordic countries and compared their HRQOL to a random sample (*n*= 10.290). They found that the psychological domain was impaired for those children. In sum, the described studies found an impaired HRQOL for young people with chronic health conditions.

2.3 Coping Research

The identification of risk and resistance factors that may explain individual differences in the way of adjusting to a chronic health condition is of importance. Psychological constructs, such as coping, health locus of control, and health beliefs, as well as social support and social network have been identified as major factors influencing patient perceived quality of life (Thompson & Gustafson, 1996). Coping with stress is a basic concept within psychology and has received much attention in research.

2.3.1 Coping- Definition and Conceptual Issues

Clinical coping research into adults started with investigating coping strategies of cardiac or cancer patients (Heim, 1998). Historically, models of coping are rooted in different scientific or therapeutic movements of psychosomatic medicine. In 1950 psychoanalysis dominated the thoughts of clinical research and the concept of defence mechanisms influenced the views on coping research. First roots can be traced to the psychodynamic model formulated by Sigmund Freud (1926), viewing coping as a defence mechanism when dealing with sexual or aggressive conflicts. In contrast to Freud, who described defence mechanisms as a protection of the ego against internal, instinctual forces, his student Alfred Adler (1929) described them as a protec-

tion of the self from external, environmental threats. Anna Freud (1948) continued the work and summarised the ten defence mechanisms formulated by her father Sigmund Freud: repression, regression, isolation, reaction formation, projection, identification, introjection, turning against the self, undoing, and sublimation. She added new important mechanisms, such as intellectualisation. However, although there are certain similarities between defence mechanisms and coping strategies, the two constructs also show different features. According to Haan (1977) coping strategies are more flexible and largely conscious whereas defence mechanisms are more concerned with issues from the past; they are unconscious and distort reality.

In the 20th century, the stress concept entered the field of the life sciences. Walter Cannon (1929) conducted physiological research which resulted in his description of the stress response as a "fight or flight response". A pioneer in research into stress was Selye (1956; 1979). The author named the stress process "general adaptation syndrome" and defined biological stress as the sum of non-specific changes in the body caused by function or damage. Selye gained his knowledge from experiments with animals and transferred his observation to humans. The predominately physiological perspective of stress formulated by Selye was soon questioned and other models focussing on psychological and cognitive constructs were formulated.

Another movement rooted in behavioural psychology was guided by the work of the Berkeley group around Richard Lazarus, who investigated the cognitive components of the coping process. The pioneering book "Psychological stress and the coping process" (Lazarus, 1966) moved the focus away from the biological and psychodynamic-rooted models towards the importance of active appraisal processes. According to the conceptualisation of Lazarus and Folkman (1984) coping is defined as the use of cognitive and behavioural strategies to help overcome a stressful situation. Stress is defined as a particular interaction between person and environment that is appraised as taxing or exceeding the resources of a person. In their model the first stage in the coping process involves the cognitive appraisal of the situation. The primary appraisal assesses the perception of a stressor as positive, irrelevant or stressful by a person. Situations appraised as stressful can be further subdivided into benefit, challenge or threat/ harm. The primary appraisal of having a chronic health

condition can be threatening and will then result in negative emotions. Secondary appraisal refers to the evaluations of one's own resources when dealing with the stressor. The person would evaluate e.g. his or her competence, social support or physical ability. If the resources of a person were sufficient, then a situation would be re-appraised as for example not threatening. The authors further discriminated between emotion-focussed and problem-focussed coping strategies. Emotion-focussed strategies aim at the regulation of the emotional state, whereas problem-focussed strategies aim at modifying a specific stressor. Emotion-focussed strategies involve crying or palliating feelings, while problem-focussed efforts involve strategies such as discussing solution alternatives. Although this model is a milestone in coping research, its complexity makes the empirical evaluation difficult. In addition, the emphases on cognitive processes within the Lazarus model nevertheless leave no explanation of the effect of more subtle stressors below the awareness level or physiological mechanisms that might moderate or mediate the coping process (Snyder & Dinoff, 1999).

Early within coping research it was discussed whether coping could be viewed either as a state or a trait variable. Lazarus' work placed the emphasis on situational determinants of the coping process. Both, the stability as well as the variability of the coping process have recently been assumed to be existent (Heim, 1998). The question if the type of disease is a predictor for the use of a specific coping strategy has been investigated by a number of researchers (e.g. Felton & Revenson, 1984; Muthny & Koch, 1997). Only minor differences were found. However, Feifel et al. (1987) could identify differences between patients with life threatening diseases compared to patients with arthritis. Patients with life threatening diseases (cancer or heart attack) preferred more confrontive coping strategies; whereas patients with arthritis coped emotionally better when applying avoidant strategies.

Although no nominal definition of coping with a chronic condition so far exists, attempts to define this construct have been made. Lazarus and Folkman (1984) defined coping as:

"… constantly changing cognitive and behavioural efforts to manage specific external and/ or internal demands that are appraised as taxing or exceeding the resources of the person"

(p. 141).

Coping is viewed as a dynamic process that changes with the demands of the environment and the appraisal of an individual. Lazarus (1991; 1993) additionally defined coping as being goal-directed with the aim to resolve the source of impact. Coping styles or strategies, goals and outcome have been distinguished to differentiate the coping process (Rudolph et al., 1995). A coping style refers to a person's manner to respond to a specific stressful situation and is a physical or mental action in reaction to a stressor. Coping styles are relatively stable personality characteristics. Coping strategies include all attempts to deal with stress, regardless of the effectiveness. They are situation-dependent. Coping goals are the underlying objectives and coping outcomes reflect the consequences of the efforts. Most researchers refer to coping as the individuals' ability to manage external and internal demands, conflicts, and feelings of distress.

Skinner and Wellborn (1994) defined coping as the way of how people regulate behaviour, orientation and emotion under stress. According to Skinner (1995) and Eisenberg et al. (1997) and in contrast to other definitions of coping (e.g. Lazarus & Folkman, 1984) the coping process includes volitional and automatic (involuntary) responses to regulate stress. Compas et al. (2001) argued against this perspective. He defined coping as:

"…conscious volitional efforts to regulate emotion, cognition, behaviour, physiology, and the environment in response to stressful events or circumstances" (p. 89).

The authors distinguish between voluntary and involuntary responses to stress. Voluntary responses are within conscious awareness and involuntary responses include reactions that are not under conscious control, such as rumination or emotional arousal. Thus coping refers to volitional responses to stress and therefore restricts the definition of coping. Following these authors, volitional and automatic responses are two levels of processing. Individuals experience them differently.

Compas and colleagues (2001) think that the investigation of these two types of processing with experimental methods will improve the knowledge about the coping process. It might also lead to a more precise questionnaire construction. According to these authors emotion-focussed coping items and symptoms of psychological distress are often confounded. This is also the case for HRQOL and coping items. Thus, a conceptual distinction is necessary.

Finally, the question of what adaptive or maladaptive coping strategies are was addressed in adult research, but it has not yet been answered. Heim (1998) made a suggestion to find an operational definition of "good" and "poor" coping strategies. Adaptive strategies are composed of active behaviour combined with the ability to mobilise social and emotional resources and the ability to accept unchangeable circumstances. Maladaptive strategies are composed of passive behaviour like resignation, hopelessness or rumination as well as prolonged avoidance and distraction behaviour.

Theoretical models of coping have been developed for adults but rarely for children. As a consequence, framework used in child research has been derived from adult coping work. However, children's stressors might be different from adults' stressors. They are often related to situation with parents or other people like teachers (Ryan-Wenger, 1992). In addition, children are restricted in their freedom to avoid certain stressors. The importance of developmental constraints and their influence on the coping process has recently been approached in research (Compas et al., 2001). The models applied to healthy children and adolescents or clinical populations so far allow the categorisation of coping strategies into mostly two contrasting ways of coping. With regard to the dimensions included in the models, there has been little consensus. The most commonly used dimensions are problem- vs. emotion-focussed coping, approach (engagement) vs. avoidance (disengagement), and primary vs. secondary control. Factor analytically derived dimensions of coping have been suggested as well (see Fields & Prinz, 1997). These classification approaches will subsequently be described.

Problem- vs. Emotion-Focussed Coping

Lazarus (1993) viewed coping as a dynamic process in which a person interacts with the environment. He underscored the need to concentrate on the nature of coping strategies in specific situations. One of the most widely accepted approaches classifies coping responses with regard to their focus, namely whether they are emotion- or problem-focussed. Studies have found that problem solving was associated with fewer symptoms and resulted in more positive and less negative emotions (Folkman et al., 1986a; 1986b).

Approach vs. Avoidance Coping

Another conceptualisation is the differentiation of approach vs. avoidance strategies which has often been applied in medical research (Miller & Green, 1985). People tend to either evade or avoid a stressor. Similar is the distinction between active vs. passive coping strategies. In general, these classifications suggest that coping strategies are relatively consistent across situations, for example Miller and Managan (1983) hypothesised personality traits, namely "monitors" (people who do better with a lot of information) and "blunters" (people who do better with less information). However, trait theories of coping have not been very successful in predicting coping in specific situations. Studies have shown that adults and adolescents are often inconsistent in their use of coping strategies across different stressors (Compas et al., 1988).

Primary vs. Secondary Control Coping

Rothbaum, Weisz and Snyder (1982) have established the primary-secondary control model. According to their perspective the controllability of a situation plays an important role for the coping process. Three different types of control are distinguished within this model: primary, secondary, and relinquished control. Primary control strategies aim at influencing objective conditions or events. Secondary strategies deal with the conditions as they are. Relinquished control is defined as the absence of coping including reactions like giving up. Primary control strategies are

targeted towards modifiable circumstances, whereas secondary control strategies are used when circumstances cannot be changed, for example when dealing with the situation that a person is going to die. The degree of the consistence of subjective control perceptions and the use of coping strategies are described as "the match". A further distinction has been made between four control strategies when analysing parental reactions to childhood cancer: predictive, vicarious, illusory, and interpretative (Grootenhuis et al., 1996). The different types of control are explained in table 3.

Table 3

Control strategies

Type of Control	**Content**	**Item Example**[1]
Predictive	Being optimistic about the situation	"I am sure everything will work out fine for me"
Vicarious	Attribute power to others	"I think I should do as I am told by a physician"
Illusory	Chance, wishful thinking	"After falling ill, I make a wish more often"
Interpretative	Searching for information to derive meaning from problems and accept them	"I want them to explain everything to me"

[1]Examples have been taken from the Cognitive Control Strategy Scale (Grootenhuis & Last, 2001)

Factor Analytic Coping Dimensions

Factor analyses of coping responses have resulted in different structures, especially when applying exploratory factor analysis (e.g. Dise-Lewis, 1988; Spirito, 1988). Children generated coping strategies and the responses were analysed. For example Spirito et al. (1988) derived their coping questionnaire structure from factor analysis. Interesting findings resulted especially from confirmatory factor analysis applied to test conceptual models of coping. The factors found show some consistency with the models described above. Ayers et al. (1996) found four factors, named as active coping, social support, distraction, and avoidance. Walker et al. (1997) identified three factors, named as active, passive and accommodative coping.

In general, the described models show some similarities, e.g. the assumed dichotomy of strategies. However, problems still remain in the conceptualisation and measurement of coping in children and adolescents. Especially the aspect of development needs to be further investigated. Coping strategies are likely to be dependent on age and cognitive abilities. With increasing age and growing cognitive ability, children employ different sets of coping strategies (Fields & Prinz, 1997). Primary school children use emotion-focussed and cognitive coping strategies. With adolescence the preferences for a specific strategy becomes less extreme and the choice can be more flexible. Generally, the variety of coping strategies gets wider and children become more flexible as they grow up.

Fields and Prinz (1997) pointed out that current classification systems do not distinguish coping strategies that promote or limit adjustment. These authors consider the coping-competence model by Blechmann et al. (1995) as an interesting conception. In this model, antisocial, asocial, and pro-social responses are distinguished. An important aspect of this theory is the role of language for acquiring pro-social coping strategies. New theoretical approaches should define coping as a multidimensional construct, involving cognitive, behavioural and emotional strategies to reduce stress.

2.3.2 Measurement of Coping in Children and Adolescents

According to Eiser (1993), the different approaches to categorise coping strategies are a major problem for drawing conclusions across studies. Coping can be measured with a variety of methods. There are a number of instruments available. Three methods of assessment have been used to measure coping processes specifically: questionnaires (self- and proxy-report), interviews, and observational methods.

Questionnaires

Questionnaires have been developed to assess coping with general stress, health-related stress or coping with specific symptoms such as pain. Most of the measures represent a variety of coping strategies and comparison of findings assessed with

different measures is therefore difficult. Compas et al. (2001) have written a comprehensive overview. The KIDCOPE (Spirito et al., 1995) and the Coping Responses Inventory (Ebata & Moos, 1991) are measures which were assessed in different clinical populations. Connor-Smith et al. (2000) have developed a questionnaire for adolescents in accordance with their theoretical conceptualisations. It reflects a model that includes volitional, goal-directed coping responses and involuntary responses to stress. The development of this measure is the most comprehensive approach in measuring coping so far. Voluntary and involuntary responses are further distinguished on a second domain pertaining to engagement and disengagement. Voluntary response moreover, can be either primary or secondary. The measure has been psychometrically tested and confirmatory factor analysis revealed three factors concerning voluntary responses to stress: primary control engagement coping (e.g. problem solving), secondary control engagement coping (e.g. positive thinking), and disengagement coping (e.g. wishful thinking). Table 4 depicts the type of report, age group and number of items/ domains of frequently used generic coping measures for children and adolescents.

Table 4

Coping measures for children/ adolescents

Name & Author	Report	Child Age (y)	No. of Items (Domains)
Adolescent Coping Scale Frydenberg & Lewis (1990; 1993)	Self	12-16	88 (18)
Coping Health Inventory for Children Austin et al. (1991)	Parent	8-12	45 (5)
Children's Coping Strategies Checklist, How I Coped Under Pressure Scale Ayers et al. (1996); Sandler et al. (1994)	Self	9-13	54 (11)
Cognitive Control Strategy Scale for Children Grootenhuis et al. (1996)	Self	8-18	36 (4)
Coping Scale for Children and Youth Brodzinsky et al. (1992)	Self	10-15	29 (4)
Coping Responses Inventory Ebata & Moos (1991)	Self	12-18	48 (2)
Kidcope Spirito et al. (1988; 1995)	Self	7-12 13-18	10/ 15 (10 strategies)
Modified Ways of Coping Checklist Halstead et al. (1993)	Self	12-17	68 (4)
Responses to Stress Questionnaire Connor-Smith et al. (2000)	Self	11-19	57 (3)
Schoolagers' Coping Strategies Inventory Ryan-Wenger (1990)	Self	8-12	25 (13)

The development of coping instruments has mainly received attention in the context of dealing with chronic pain (Jensen et al., 1991). Questionnaires have primarily been developed from a cognitive-behavioural perspective (Varni et al., 1996b; Robinson et al., 1997; Reid et al., 1998). One example is the Paediatric Pain Coping Inventory which compromises 41 items and has been developed for children aged 5 to 17. This instrument consists of five coping scales; namely cognitive self-instruction,

problem solving, distraction, seeking social support, and catastrophizing. Cronbach's alpha of these scales ranges from .55 to .74. Walker et al. (1997) developed the Pain Response Inventory for Children which is a self-report measure containing 65 items. Gil et al. (1991) developed a condition-specific measure to assess coping strategies in children with sickle cell disease. The Coping Strategies Questionnaire for Sickle Cell Disease contains 80 items and is a self-report measure for children and young people aged 7-17.

The validation methods of coping instruments applied need to be critically reviewed, especially with regard to the questionnaire content. Correlations between coping instruments and emotional problems are often circular and therefore results are confounded (Compas et al., 2001). According to them, future instruments should reduce the overlap between coping and emotional distress variables. In addition, research has only recently started to examine the relationship between coping and clinical variables, health status or HRQOL.

Interviews and Observational Methods

Just a small number of semi-structured interviews have been used to assess child and adolescent coping. For example Band and Weisz (1990) interviewed children about diabetes-related stressors. The advantage of interviews is to get detailed information about specific stressful situations, but they might still be influenced by interviewer bias.

Observational methods are important approaches when analysing coping responses with regard to specific situations, such as medical procedures. They are useful to validate self-reports. An instrument to assess coping behaviour of children is the Child-Adult Medical Procedure Interaction Scale-Short Form (CAMPIS-SF) which is a behaviour rating scale of children's acute distress and coping (Blount et al., 1997; 2001). This instrument can be used to assess behaviours of parents and medical personnel promoting distress. Dunn-Geier et al. (1986) investigated adolescents with chronic pain. The mother-child interactions have been videotaped and rated using a

variation of the Mash and Terdal's response class matrix (1981) which gives information about antecedents and consequences of an observed behaviour. Children with a higher school absenteeism rate tended to express more pain.

The comparison of the three different approaches (questionnaire, interview and observation) will be a major task for future research. However, the short description of available instruments stresses the need for a new coping module adequate for children and adolescents with a chronic health condition. For a comprehensive analysis of coping strategies it is important to compare results also across countries, otherwise coping strategies might appear to be specific to a particular language and culture.

2.3.3 Coping with a Chronic Health Condition

The way in which children and adolescents cope with their chronic health condition is increasingly considered as an important question for research. Until a few years ago, research into paediatric health status assessment has focussed on the impairments or deficits of children and adolescents with a chronic health condition. This perspective neglects the increasing evidence of resilience of those children and adolescents and what they think, feel, and do with regard to their health condition. An increasing number of studies are available about coping strategies (Compas et al., 1992; 2001).

Developmental Aspects

Coping strategies are influenced by the developmental stage of a child, the development of language and cognitive abilities (Fields & Prinz, 1997). Nevertheless, most conceptualisations for children and young people were based on models of coping in adults. Consequently they lacked a developmental component. Yet the understanding of developmental processes that are specific for certain periods of childhood is fundamental to the assessment of coping strategies of children. Conceptions

of illness and health depend on the cognitive development of a child. According to Jean Piaget (1928) children's thinking proceeds through a discrete series of stages towards greater differentiation. Piaget's theoretical framework provided a basis with regard to different concepts. Within the Piagetian framework four stages are differentiated:

- sensumotoric phase (0-2 years)
- preoperational phase (2-7 years)
- concrete operations phase (7-11 years)
- formal operations phase (ages 12 and up)

Each stage is described by the cognitive ability of a child of that phase. Children in the sensumotoric phase build up their knowledge about their environment through their actions starting with applying reflexes to a variety of objects. For Piaget (1928) the processes of assimilation and accomodation are major features of development. According to his theory, infancy ends with the capacity of mental representation. The reasoning of the next stage, the preoperational phase, is primarily described as irreversible and egocentric. It is based on obvious features. Children at the stage of concrete operations become less egocentric and can understand more than one dimension of a situation. Nevertheless, they are limited to concrete (here and now) experiences. With the onset of the period of formal operations, children begin to think abstractly. A sequence of different stages has been found for many topics, such as the development of moral or concepts about death and reproduction (Lohaus, 1993). Schmidt & Lehmkuhl (1994) concluded that a number of studies based on the Piagetian model have been conducted. Findings support the stage concept, suggesting that illness concepts progress from pre-logic explanations to a complex understanding of health. For example, children in the concrete operational phase begin to realise that disease is reversible. However, they often think that diseases are caused by contamination (Bibace & Walsh, 1980). The age-dependency of coping strategies was addressed in some studies, e.g. it was found that cognitive distraction was more often applied by older children (Altshuler & Ruble, 1989). Although Piaget's

cognitive developmental theory has been criticised, it provides a basis for understanding age-differences in paediatric health outcome research.

Disease as a Stressor

In general, the concept of coping represents a self-regulation process and is historically tied to the concept of stress. Researchers paid particular interest to stressors as major life events, for example the death of a family member or parental divorce. Subsequently, minor events or "daily hassles", such as being bullied by peers, raised the attention of researchers. According to Fields and Prinz (1997) the most frequently reported stressors by children and adolescents are:

- fear of negative evaluation by peers or adults,
- parental conflicts or loss,
- conflict with an adult, and
- feeling socially excluded.

In terms of chronic paediatric health conditions having a chronic health condition was either viewed as a major life event (e.g. the time point of diagnosis) or as a daily hassle (e.g. problems with the daily treatment regimes). Children and adolescents with a chronic disease must learn to comply with medical treatments, parental concerns and their self-image among other problems. Midence et al. (1993) pointed out that illness itself, the personality of a child, family, social environment, and medical support are important factors which influence the coping process. Following them, the identification and investigation of such specific stressors and coping skills will contribute to improve service to the chronically ill population. Perrez and Reicherts (1992) defined five characteristics of a stressor which might influence the coping process: changeability, reoccurrence, valence (stressfulness), controllability, and ambiguity (degree of information lack) of the situation. For stress reduction, the valence, controllability, and ambiguity of a chronic health condition can be regarded as important features for interventions.

Despite increased efforts to understand the complex relationships and interac-

tions between coping and potential correlates, it is yet not clear how adaptation to a chronic condition occurs. The disease might not be a stressor per se because coping strategies may lessen the risk of maladjustment. This view is supported by Filip (1995), who defines the state of being happy as a result of successful adaptation to a chronic condition, i.e. to interpret a disease as a positive experience and to cope with it. This necessitates addressing the complex interplay of resources and strains as well as their perceptions in a dynamic model in children. So far, approaches to study coping strategies with regard to adjustment of chronically ill children systematically are rare (Spirito et al., 1995). Most of the studies have focussed on coping during invasive medical procedures (e.g. Smith et al., 1990) and coping with pain (e.g. Varni et al., 1996b). Depending on the focus of their theoretical approach researchers have highlighted different issues concerning the coping process of children. Thompson et al. (1994) accentuated the stress perceived by the family as a central variable for the psychological adaptation of a child. Pless and Pinkerton (1975) focussed on the self-concept and coping style as central components of adaptation.

Research concentrated on certain conditions such as leukaemia (Kupst et al., 1985; Kupst & Schulman, 1988) and diabetes (Kovaks et al., 1985). Importantly, as Midence (1994) pointed out, coping skills e.g. identified for diabetes might not be applicable for the adjustment process to other chronic conditions. The effectiveness of coping strategies has been studied in adults, but has received little attention in children (Olson et al., 1993). Still, the ability to employ specific coping strategies is especially vital for short-term and long-term adaptation to chronic illness (Band & Weisz, 1990; Ellerton et al., 1994). Results suggest that avoidance seems to be more effective when dealing with a short-term stressor and approach when being confronted with long-term stressors (Boekaerts & Röder, 1999). In an overview of children's coping models, Compas and colleagues (1992) re-asserted the view that coping strategies have either protective or negative effects on the experience of paediatric chronic health conditions.

Furthermore, researchers examined the differences in children's coping strategies with regard to the range of coping strategies and aggressiveness (Hardy, Power & Jaedicke, 1993). Aggressive strategies often lead to a short time resolution, but are

less optimal for long-term outcome. Overall, it cannot be assumed that a specific coping strategy is universally helpful across different situations. The effect of a coping strategy depends on the match between the demands of a situation and the employed strategies.

Boekaerts and Röder (1999) reviewed the literature on the consequences of having a chronic disease on functioning in daily life. They concluded that coping strategies used by children with a chronic disease appear to be similar to those of healthy children and that children with a chronic health condition do not experience more stress in terms of frequency or intensity compared to healthy children. However, other researchers found some differences between healthy and chronically ill children. For example, Brook and Tepper (1997) questioned children with asthma (*n*= 51) about their self-image, coping, and family interaction and compared the results with findings in healthy children (*n*= 32). According to these authors, children with asthma coped worse with stressful situations than healthy children.

Many children can adapt very well, for example Zeltzer and colleagues (1980) compared healthy children with children with cancer. The latter experienced less disruption in their daily life. Interestingly, the relationship between the severity degree of a disease and coping is not linear, e.g. children with a mild or severe form of asthma showed poorer coping strategies compared to children with a moderate form (Perrin et al., 1989). The severity degree of an illness seems not alone to account for variation in adjustment. The relationship between coping strategies and the burden of a disease has been reflected in a study by Manus & Killeen (1995). Obese children who underestimated their weight showed a higher self-esteem. This is just one example how coping strategies can influence the perceived burden of a disease.

Schanberg et al. (2001) investigated the relationship between coping, family pain history, the child's pain, and the physician-rated degree of illness severity in children with chronic rheumatic diseases (*n*= 100). They found that the use of the pain coping strategy „catastrophizing" is associated with family pain experiences, pain ratings of the child and the physicians' disease severity rating. The authors suggest that intervention programs should focus on reducing children's use of catastrophizing strategies. Manne et al. (1992) examined adult-child interaction during stressful medical

procedures in the course of cancer treatment (*n*= 43). Findings indicate that adult distraction activities enhanced adjustment to stressful situations; explanations did not reduce momentary distress and crying. Attempts to give the child control proved to be helpful for adjustment. Milousheva et al. (1996) investigated coping strategies used by children and adolescents with diabetes mellitus (*n*= 43). Younger children showed mostly "instrumental action", "emotional expression", and "catastrophizing" as coping strategies. Older boys showed mainly "behavioural avoidance", whereas the most prominent coping strategy for girls was "talking to peers". This stresses the need to investigate gender differences. In sum, studies examined coping strategies from different perspectives. The results are promising.

Coping and HRQOL

While a lot of studies analysed children's coping strategies, the relationship between coping strategies of chronically ill children and adolescents and their HRQOL has not been thoroughly investigated yet. A Medline search from 1985 to 2002 entering the term "quality of life AND coping" (in the title field) resulted in 58 articles. Only nine of these articles pertained to research into children. This small number of articles found reflects the importance to address the relationship between coping and HRQOL in future research. This necessity was also supported by Bandell-Hoekstra et al. (2000), who aimed at reviewing findings concerning the relationship between coping with pain and HRQOL in children with headaches. In their literature search only two studies were identified. The authors conclude that more research on coping and HRQOL in paediatric headache is needed to identify the strategies that children use when having headaches.

Goldbeck (2001) investigated parental coping with the diagnosis of childhood cancer, diabetes or epilepsy (*n*= 108) at two time points: 1-2 and 10-12 weeks after diagnosis. The author found that parental dissimilarity in information seeking was correlated with a decrease of the child's quality of life, whereas parental dissimilarities in social support seeking and religion correlated with an improved parental HRQOL. Makipernaa (1989) examined long term HRQOL and coping after treatment

of solid tumours in childhood (*n*= 94). Most of the participants of the study lived a well-balanced life and showed an adequate capacity to cope with their situation.

Ravens-Sieberer et al. (2001a) investigated the relationship between HRQOL and coping after in-patient rehabilitation in children with obesity, asthma or atopic dermatitis (*n*= 1019). Coping, the lack of emotional support and poor global health explained 37% of the variance of the HRQOL total score of the KINDL measure in the obesity group. In another study, the training of coping skills, such as problem solving, cognitive behaviour modification, and conflict resolution had an effect on metabolic control and HRQOL measured with a condition-specific instrument in adolescents with diabetes mellitus (Grey, 2000, *n*= 77). Fuggle et al. (1996) investigated the impact of pain on HRQOL and coping strategies in sickle cell disease (*n*=25). Results suggested that the assessment of pain in the home environment is essential for pain management service and enhances children's HRQOL. Rose and Clark-Alexander (1998; 1999) explored HRQOL and coping strategies of caregivers of children with HIV/ AIDS (*n*= 79). The authors point out that HRQOL of caregivers can be improved if better coping mechanisms are trained.

The investigation of parents of children with cerebral palsy (Sjobu, 1994) showed that HRQOL was related to the spouses' satisfaction with each other, the degree of openness and the well-being of the partner. According to the author, these findings should be taken into consideration when rehabilitation plans are made. Staab et al. (1998) examined the relationship between HRQOL and coping in adolescents/ adults with cystic fibrosis (*n*= 89) as well as of parents of younger patients (*n*= 125) with regard to their own HRQOL and coping efforts. The results showed that the ways of coping were significantly correlated with HRQOL. The coping style of parents proved to be the most important factor in explaining variance of HRQOL.

The challenge is now to find out how the enhancement of HRQOL can be supported by interventions designed for children with chronic health conditions with regard to their coping strategies.

2.4 Measuring Psychosocial Consequences of Chronic Health Conditions Cross-Culturally

A prerequisite to examine the relationship between HRQOL and coping mechanisms is the availability of appropriate measures. These measures should be psychometrically tested, applicable across health conditions and countries as well as cultural backgrounds.

2.4.1 Cross-Cultural Research Approaches

Concerning HRQOL and coping, there are different reasons to design cross-cultural measures. An important reason is to identify variables that influence health or HRQOL across countries or within a certain cultural background. Another important motive is to collect cross-cultural data for evidence-based medicine. Evidence-based medicine and healthcare is looked upon as a new paradigm, replacing the traditional medical paradigm. It is dependent on good clinical practice and the use of randomised controlled trials, as well as systematic reviews (of a series of trials) and meta-analysis, although it is not restricted to these. The comparison of evidence-based results across different countries provides crucial information for policy-makers.

When developing questionnaires in different languages, several barriers have to be overcome. The need to achieve conceptual equivalence between measures across countries demands a thoughtful translation procedure which then ensures cross-cultural comparability of questionnaire versions in the target languages. The provision of equivalence across cultures is a complex task.

According to Hui & Triandis (1985) the following aspects have to be taken into consideration:

- **Functional Equivalence**
  Are items similar in meaning cross-culturally?
- **Operational Equivalence**
  Are the procedures used to obtain information comparable?
- **Scale Equivalence**
  Do individuals respond similarly to items?
- **Metric Equivalence**
  Are individuals across countries ordered on the measure in a comparable way?

Whereas cross-cultural work has played a predominant role during the last years in HRQOL research, only a few studies could be identified for the coping field into children and adolescents. Therefore the cross-cultural approaches in HRQOL research will be delineated.

As a consequence of the growing interest in HRQOL assessment and its important role for clinical, epidemiological and political issues, the demands for international HRQOL research has increased. The need to assess HRQOL in a range of cultures and languages has received attention (Bullinger et al., 1993; Guillemin et al., 1993; Guillemin, 1995). The WHOQOL group as stated in their definition of quality of life proposed that the person's culture plays a major role for the quality of life perception. There is still little known about how the cultural context relates to children's and adolescents' HRQOL.

Most HRQOL assessment tools have been developed in English speaking countries. Instead of developing new questionnaires, existing measures were modified in a cross-cultural adaptation process. Several authors (e.g. Guilllemin et al., 1993) proposed guidelines to standardise this procedure. In recent years, there has been an increasing interest in the assessment of HRQOL across countries. Prior adaptation work was mainly focussed on translation issues, more recent work has begun to rely

on the suggested methodology for translation with the goal to produce valid and reliable questionnaires in a multiplicity of languages (Anderson et al., 1996). In her overview Bullinger (1997b) distinguished between international and cross-cultural HRQOL research. The author pointed out that the term "international" refers to activities of different countries, such as comparing HRQOL results with regard to specific health conditions, whereas the term "cross-cultural" denotes an effort to study the comparability of HRQOL instruments across countries.

Before comparing HRQOL results across countries and cultures, the question of comparability of HRQOL domains arose. It is a prerequisite for cross-cultural studies to have a core set of domains about HRQOL that are identical across countries. The WHOQOL Group (1993) addressed this question in their study. The results suggested that national items which could be included by every participating country, did not significantly increase the explained variance of the questionnaire. An overlap of nationally produced items across countries was noted. The merit of this working group is the provision of a body of evidence underscoring a substantial consensus concerning the components of the HRQOL construct across countries all over the world. However, other researchers who work with an idiographic approach underline the uniqueness of HRQOL of each individual, such as the SEIQOL group (Cook, 1993; McGee et a., 1991).

Diverse working groups have been active to develop cross-cultural measures. Historically, the first group who set about this research challenge was The European Group for Quality of Life and Health Measurement (1992) working with the Nottingham Health Profile, a generic quality of life questionnaire. A similarity of the different working groups is that their work mostly started with the aim of evaluating the adequacy of an existing HRQOL measure across countries.

A second working group to mention is the International Quality of Life Assessment (IQOLA) who in 1991 initiated a project to translate and adapt the Medical Outcome Study Short Form 36 Health Survey (SF-36) in 15 countries (Ware et al., 1995). The work of the IQOLA group resulted in a number of papers. The construction and scoring of multi-item scales from the SF-36 Health Survey have been investigated in general population samples in eleven countries (n=1483 to n=9151, Gandek et al.,

1998a). The results supported the construction and scoring of the SF-36 questionnaire. Ware et al. (1998) investigated the factor structure of the SF-36 in ten countries and the findings supported the construct validity as well. In addition, the IQOLA Group investigated the equivalence of SF-36 summary health scores (Ware et al., 1998) and the psychometric properties of the short form SF-12 (Gandek et al., 1998b). Furthermore, structural equation modelling (Keller et al., 1998), the application of the Rasch model (Raczek et al., 1998), and translation issues (Bullinger et al., 1998; Gandek & Ware, 1998c) have been studied.

A third group working on cross-cultural measurement development is the already mentioned WHOQOL Group (The WHOQOL Group, 1998a). The WHOQOL-100 was developed simultaneously in fifteen centres across the world. In contrast to a sequential or a parallel measurement development approach, the simultaneous approach allowed all centres to contribute to the content of the questionnaire. In each centre, focus groups were conducted in order to generate items. The focus group work resulted in a global item pool of 1000 items. The use of focus groups as proposed by Satorius and Kuyken (1994) proved to be a useful way to easily test the appropriateness of items for a new cultural group. Altogether, 236 items were included in the preliminary version of the WHOQOL. In a pilot version n= 300 subjects per centre had to participate in the study. After analysing the data set, the measure could be reduced to 100 items. A short form (WHOQOL-Bref) contains 26 items derived from the long version. Structural equation modelling supported the hypothesis of cross-culturally important facets and domains of the WHOQOL-100 (The WHOQOL Group, 1998b; Power et al., 1999).

Cross-cultural HRQOL has been studied for diverse diseases, such as erectile dysfunction (Parkerson, Willke & Hays, 1999), peripheral occlusive arterial disease (Marquis, Comte & Lehert, 2001), cancer (Forjaz & Guarnaccia, 2001), depression (McKenna et al., 2001), onychomycosis (Drake et al., 1999), epilepsy (Buck et al., 1999; Cramer et al., 1998) or genital herpes (Doward et al., 1998). A working group to mention is the International Breast Cancer Study Group (IBCSG) which aims at establishing HRQOL as an outcome in randomised clinical trials in patients with early breast cancer who received adjuvant therapy (Bernhard et al., 1997; Hurny, Bernhard &

Coates, 1998). Since 1998 the IBCSG has been working on the development of a longitudinal HRQOL database to investigate the impact of adjuvant therapy. The group documented their work in a number of publications (e.g. Hurny et al., 1994; Bernhard et al., 1998a; 1998b; Gelber et al., 1998; Coates et al., 2000). The European Organization for Research and Treatment of Cancer (EORTC) developed a modular approach for assessing HRQOL in cancer patients and the core questionnaire (EORTC QLQ-C30) was tested in 13 countries (Aaronson et al., 1993). In general, cancer research is the most progressive area with regard to cross-cultural approaches. Furthermore it has to be noted that different instruments exemplify different models for developing and adapting HRQOL measures cross-culturally. However, researchers attempted to standardise procedures.

2.4.2 Cross-Cultural Research in Children and Adolescents

The children's area of cross-cultural HRQOL research is an underinvestigated field. To advance knowledge about **cross-cultural HRQOL in children and adolescents**, appropriately translated and validated instruments are still needed. So far, mainly international comparison studies have been conducted. Alternatively different culture groups in one country have been compared with regard to HRQOL outcome. Only a few studies on cross-cultural research into HRQOL in children have been conducted. The cross-national adaptation and psychometric evaluation of the Childhood Health Assessment Questionnaire (CHAQ) and the Child Health Questionnaire (CHQ) in 32 countries deserves a special credit for its comprehensive approach (Ruperto et al., 2001). The international network of the Paediatric Rheumatology International Trials Organisation (PRINTO) initiated this study. The study is part of an international effort supported by the European Union to evaluate HRQOL in children with juvenile idiopathic arthritis. A total number of n= 6.644 subjects was enrolled. The results of the study suggest that the cross-cultural adaptation is a valid method to develop reliable instruments.

A multinational effort in the field of HRQOL assessment in children with haemophilia was made by Bullinger et al. (2002a). A measure was developed in six European

countries simultaneously. A stepwise procedure was applied with pilot testing the instrument, conducting a cognitive debriefing, and finally field-testing the newly developed measure. This resulted in three different questionnaire versions.

Some smaller studies also aimed at a cross-national comparison of HRQOL. For example, Richardson et al. (2001) compared English and Canadian children with inflammatory bowel disease (*n*= 53) to determine whether children in these countries have the same concerns with regard to their condition. Indeed, health-related concerns were similar and therefore correlated closely. Tapsoba, Deschamps and Leclercq (2000) designed a questionnaire to measure oral HRQOL in adults and children which includes the social and psychological impact of oral diseases (e.g. emotional functioning associated with smiling). Three countries were included in the assessment (*n*=1171 children and *n*=1062 adults). Results provided evidence for the cross-cultural stability of the questionnaire. French, Carroll and Christie (1998) adapted the Childhood Asthma Questionnaires (CAQs) for Australia. Focus groups and psychometric analyses were used to investigate and ensure equivalence. The study consisted of three phases and the sample size ranged with regard to the specific phase from *n*= 49 to *n*= 784. Manificant et al. (2000) reported a European validation study conducted in Belgium, France, Italy, Luxembourg, Spain, and Switzerland to psychometrically test an instrument to assess HRQOL of infants (under the age of three) through parent report (*n*=1412). The developed questionnaire is called QUALIN. The items have been derived from statements of parents or caregivers.

Cross-cultural comparison is a method to test the value of paradigms that emerged in national studies for other countries. Nevertheless, **cross-cultural approaches to measure coping strategies in children and adolescents** are rare so far. McCarty and colleagues (1999) addressed the interesting question which role cultural values and traditions play in the development of coping strategies. They interviewed 6-14 year-old children in Thailand and the United States (n= 141). Their self-reports of coping were compared with regard to specific stressors. The theoretical framework of this study was the primary-secondary model (Rothbaum et al., 1982). Additionally, the authors distinguished between overt (visible) vs. covert cop-

ing methods. In spite of several similarities across nations, differences in coping behaviour could be identified. Thai children reported more covert coping than American children with regard to stressors like "adult anger" and "injection in a doctor's office". American children adjusted more often than Thai children did when having to cope with an injury. This study stresses the necessity for more attention to be paid to cultural or national similarities or differences in coping behaviour.

Dolgin et al. (1997) studied the influence of culture on coping behaviour with regard to siblings' adaptation to childhood cancer in Israel and the United States. Similarities concerning parental coping and family relations variables were found. Greater family support and emotional expressiveness as well as fewer conflicts were associated with less behaviour problems in the sibling. Friedman and Mann (1993) conducted a cross-national study and compared decision-coping patterns of Australian and Israeli adolescents (*n*= 1456). Israeli adolescents scored higher on self-confidence and vigilance. It was found for both samples that decision-coping pattern contain two kinds of strategies, namely a vigilant and a maladaptive strategy (e.g. panic).

Seiffge-Krenke (1992) stressed the similarities in the coping behaviour of German, Finnish and Israeli adolescents. Watson and Sinha (1998) compared Australian (*n*= 388) and Canadian (*n*= 635) students in respect to the defence-style questionnaire. The Canadian sample showed higher means on nine scales of the defence style questionnaire. According to the authors the identified differences might be due to cultural influences. Olah (1995) examined cross-culturally coping behaviour of adolescents across different anxiety-provoking situations. Adolescents *(n*= 721) from Italy, India, Hungary, Sweden, and Yemen were included in the study. Similarities across countries were found in that adolescents preferred avoidant strategy in high anxiety level situations, whereas at a low and medium anxiety level assimilative and constructive coping strategies were preferred. Adolescents in Europe reported the use of assimilative coping strategies more frequently compared to adolescents from India and Yemen who preferred emotion-focussed strategies. The authors concluded that culture directs coping behaviour of adolescents, but experiences with special stressors have a stronger influence on the choice of coping strategies.

It has to be noted that no coping measure has been developed in a simultaneous cross-cultural approach so far to investigate coping strategies of chronically ill children and adolescents. In summary new, psychometrically sound measures to assess HRQOL and coping are needed. These new instruments must be sensitive to cognitive development. It is crucial to involve children as well as adolescents in order to understand the factors that increase or decrease HRQOL. As Lindstrom (1994) has stated in his report about the HRQOL of children in the Nordic countries, the direct dialogue with children is unfortunately often missing.

2.5 The DISABKIDS Project

The understanding and perception of disability is presently changing. Researchers recently began to address the importance of the cross-cultural subjective perspective of health. A European group dealing with this topic is the DISABKIDS Group (Bullinger et al, 2002b; 2002c, see appendix A). The thesis is based on this project and will therefore be described in detail. The acronym DISABKIDS stands for "Quality of Life in Children and Adolescents with Disabilities and their Families-Assessing Patient Views and Patient Needs for Comprehensive Care". A group of researchers from six European countries with backgrounds in medicine, psychology, and sociology initiated the project. It started in February 2001 with project duration of three years. The European Commission funded the project within the 5th framework programme "Quality of Life and Management of Living Resources".

The main aim of the project is to enhance HRQOL in children and adolescents with chronic health conditions by developing a European instrument for HRQOL assessment from the perspective of children, adolescents and their parents. More specifically, the DISABKIDS project aims at developing a **chronic generic** inventory as well as **condition-specific** questionnaire modules. It is planned to implement the measures in routine medical care. The HRQOL instruments ought to be applicable in different national and cultural contexts, comply with quality standards in instrument development, and be practical, i.e. short and easy to use and score.

In sum, the overall objectives of this project are:

- to develop and promote the use of standardised instruments to assess HQOL in children with chronic health conditions,
- to assess HRQOL from the patients' perspective by addressing the needs of care, and
- to enhance HRQOL and the independence of children with chronic health conditions.

Within the project the perception of children and adolescents with regard to important dimensions of their HRQOL is assessed. The DISABKIDS Group has defined HRQOL for their research work as a multidimensional construct with social, physical, emotional and functional domains. Furthermore, the DISABKIDS Group cooperates closely with the sister project called KIDSCREEN (see figure 3) which aims at developing a generic HRQOL instrument as a means for monitoring the health of children and adolescents in the European Community (Ravens-Sieberer et al., 2001b).

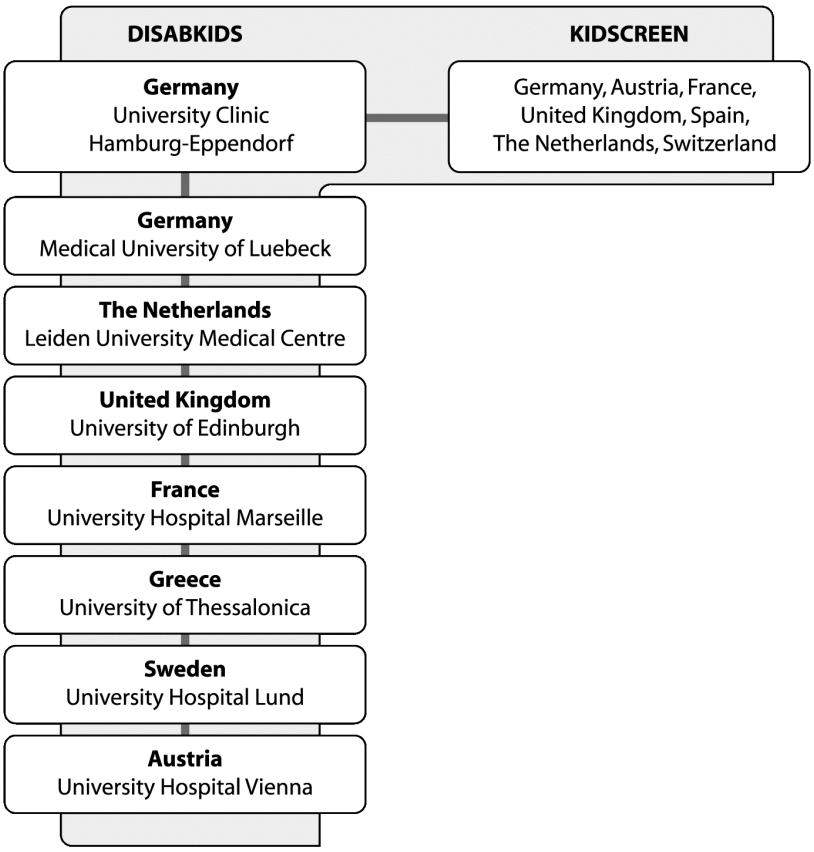


Figure 3

Participants of the DISABKIDS Group

The focus of the DISABKIDS project is on seven chronic health conditions which are asthma, rheumatic diseases (arthritis), epilepsy, cerebral palsy, diabetes mellitus, atopic dermatitis, and cystic fibrosis.

Asthma is a lung disease and causes breathing problems (Sly, 2000). It is the most frequent chronic childhood disease and nearly twice as many boys compared to girls are affected by asthma. Symptoms include cough, wheezing, tachypnea, and dyspnea. Acute episodes are often caused by exposure to irritants (e.g. smoke or perfume) or allergens. The treatment of asthma is based on the avoidance of allergens and medication and normally satisfactory control is possible.

Rheumatic diseases are caused by abnormally regulated immune responses and can lead to inflammation of target organs (Miller, 2000). The immune system reacts to molecules of the host's own tissues. The possible reasons for this self-reactivity are the similarity between foreign and self-molecules on the one hand and on the other

hand viral infections which exaggerate immune responses. Products of the immune system might affect the function of other organs. Also genetic factors may play a role by increasing the risk of developing rheumatic disease. Juvenile rheumatoid arthritis (JRA) is one of the most common rheumatic diseases in childhood. It is regarded as a disease category with three types of onset: oligoarthritis, polyarthritis and systemic-onset disease (Miller & Cassidy, 2000). Symptoms are morning stiffness and gelling, ease of fatigue, joint pain and swelling. Treatment includes physical and psychosocial intervention as well as medication.

Epilepsy is defined as recurrent seizures unrelated to fever or an acute cerebral insult (National Information Centre for Children and Youth with disabilities, 2000). A seizure occurs when there is a sudden disturbance of brain function. A patient's consciousness and motoric abilities may be affected as well. The type of seizures can be classified into partial, generalized and unclassified seizures. In most children, a cause of the seizure cannot be determined. The seizures may be controlled, but behavioural as well as psychosocial problems are more likely to occur in children with epilepsy compared to healthy children.

Cerebral palsy is a disorder of movement and posture due to a defect or lesion of the immature brain (Haslem, R., 2000). It is often associated with epilepsy and impairment of speech, vision, hearing and mental retardation. The risk of cerebral palsy increases with intrauterine exposure to maternal infections. In most cases, the cause of cerebral palsy is difficult to identify. Cerebral palsy can be classified by describing the motor impairment or the functional capacity level. The treatment includes a multi-professional approach, i.e. occupational therapists, physiotherapists, speech pathologists, social workers, psychologists, physicians, and educators working together.

Diabetes mellitus is a syndrome of metabolic disease (Sperling, 2000). It is caused by deficiency of insulin secretion or insulin action which results in abnormal metabolism of carbohydrate, protein, and fat. Three forms of diabetes have been identified, type I, type II and secondary diabetes. Type I diabetes is characterised by the dependence on exogenous insulin, pancreatic islet β-cell destruction mediated by immune mechanisms, and onset in childhood. Type II diabetes is characterised by not

being insulin dependent. Onset occurs in adulthood, although in the United States the prevalence in childhood is increasing. Persons with type II diabetes are often obese. The common manifestation of diabetes in children (type I) is a history of polyuria, polydipsia, polyphagia, and weight loss. Treatment first of all includes the infusion of insulin and the prevention of visual, renal, neuropathic complications.

Atopic dermatitis is an inflammatory skin disorder characterised by sore and itchy skin (Sly, 2000). The disease often begins in infancy and becomes less frequent by the age of 5 years. Sometimes eczema may persist. Therapy includes medical and behavioural aspects. Factors that can trigger itching and scratching should be avoided, e.g. extreme temperatures, special detergents or clothes

Cystic fibrosis or mucoviszidosis is a genetic disease affecting approximately one of 2.500 newborn babies (Boat, 2000). The disease affects the exocrine glands and causes the production of mucus. It is characterised by obstruction and infection of airways and digestion problems. The treatment includes physical therapy, especially to dislodge the thick mucus from the lungs, and antibiotics. It can be very time intensive as well as stressful. Patients with mucoviszidosis often feel exhausted and have breathing difficulties. Cystic fibrosis is a life-limiting disorder. Although survival has improved during the last decade of years, the shortened life expectancy has a great impact on patients. Statistics now indicate a median cumulative survival of 30 years.

The tasks or developmental steps of the DISABKIDS project were described in eleven work packages: literature review, focus groups, item development, translation, pilot-testing, analyses of the pilot test, field test preparation, field test, analyses of field test, implementation phase, and final analyses. The aim of the first work package, the literature search, was to review the literature on HRQOL assessment and to identify dimensions of quality of life in disabled or chronically ill children and adolescents as well as available assessment instruments. The literature search was carried out in MEDLINE according to specific criteria (see figure 4).

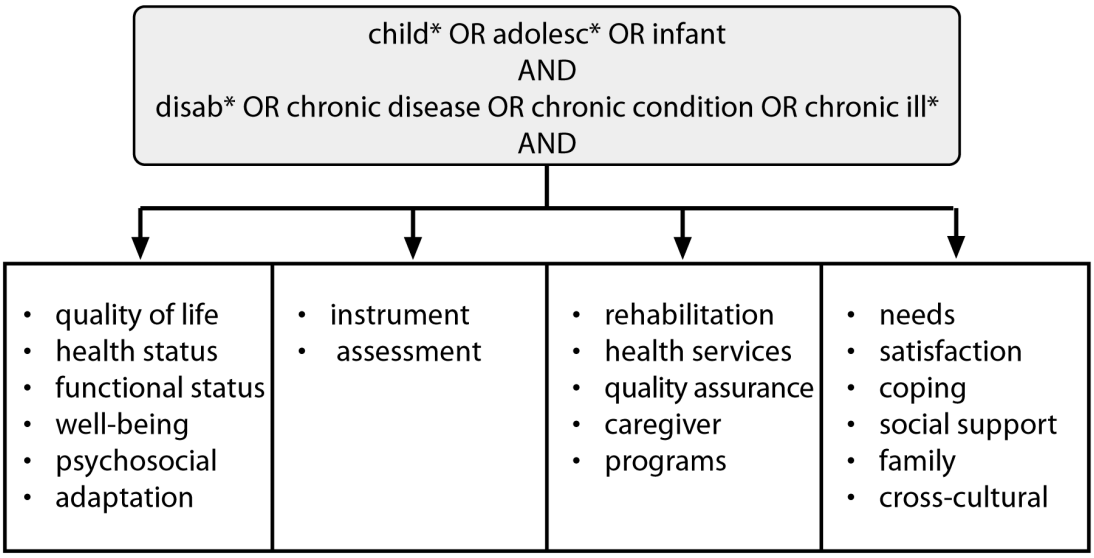


Figure 4

Keywords for the literature search

Altogether 8233 articles were identified. First of all abstracts were evaluated by the participants of the DISABKIDS project. After a first round of evaluation, 19% of the studies were found to be relevant for the project. Most of the studies were cohort (32%) or instrument validation studies (17%). 45% of the studies had a cross-sectional and 15% a longitudinal design. Only a few studies (12%) had a research aim concerning HRQOL and described HRQOL instruments. In 34% of these studies, HRQOL was considered as a main aspect. Mostly generic HRQOL instruments were used (14%). The reported sample sizes ranged from single case studies to health surveys (*n*= 99.513), but in most of the studies age groups were not reported. The evaluation revealed that 71% of the studies included chronically ill populations. Brain injuries (24%), rheumatoid arthritis (22%) and asthma (18%) were the most frequently analysed health conditions. The other finalised work packages (focus groups, item development, translation, pilot-testing, analyses of the pilot test) will be described in the method section.

In sum, the overall aim of the DISABKIDS project is to find ways of bringing basic and applied research programs together so that the newly developed modules will be disseminated to interested clinics. It will be for the first time that a simultaneous questionnaire development approach will be applied in paediatric research.

2.6 Aims of the Present Study and Research Questions

The thesis represents a European effort to develop new measures for children and adolescents with chronic health conditions. The present study was designed to provide a cross-national **coping** measure and to psychometrically test it together with a **chronic generic HRQOL** for children and adolescents with chronic health conditions. The development and psychometric testing of both measures will be described in detail. The advantage of connecting the current thesis with the DISABKIDS project is that it is a multi-centre collaborative study. The specific objectives of the thesis are

- to develop a coping in parallel to a HRQOL questionnaire applicable for different diseases from the children's and adolescent's perspective,
- to pilot test the new measures in seven different countries,
- to determine the item characteristics of both measures in terms of mean, standard deviation, percentage of missing values and "not applicable" answers, skewness, age and gender differences as well as item-scale correlations,
- to determine the scale characteristics of both measures in terms of mean, standard deviation, floor- and ceiling effects, reliability, and scale fit,
- to analyse open questions with regard to relevance and difficulty of items,
- to explore the scale structure of both measures with exploratory factor analyses,
- to reduce the number of questions and to define their structure,
- to exploratory investigate age, gender, condition, and country effects, and finally,
- to analyse the relationship of both constructs with multiple regression analyses.

These steps should ideally result in a selection of the best questions without impairing the adequate coverage of areas elicited by children and adolescents themselves. With regard to the relationship between HRQOL and coping strategies, first ideas should be formulated for future research.

3 Methods

The thesis is based on the DISABKIDS project in which cross-culturally usable HRQOL measures for children and adolescents with chronic health conditions have been developed. All project partners did the work regarding the development of a chronic generic HRQOL measure conjointly. The author of this thesis performed the psychometric analysis reported in the result section. As an ancillary part of the DISABKIDS project, the author of this thesis solely conducted the development and psychometric testing of the coping questionnaire. The Ethics Committees of the participating countries approved the study. The developmental steps conducted so far are the result of a systematic effort since February 2001. First of all, each developmental step of the HRQOL and coping questionnaire will be described. Subsequently, information about the design, conduct and analyses of the pilot test will be given.

3.1 Questionnaire Development

In the development of the chronic generic HRQOL and the coping measure care was taken to comply with existing guidelines regarding translation methods (Guillemin et al., 1993). These guidelines are important in cross-national work to ensure the equivalence of items and scales across countries. The same stepwise procedure was applied for both measures. The questionnaire development started on the basis of a simultaneous bottom-up approach. The different developmental steps of both measures are shown in figure 5.

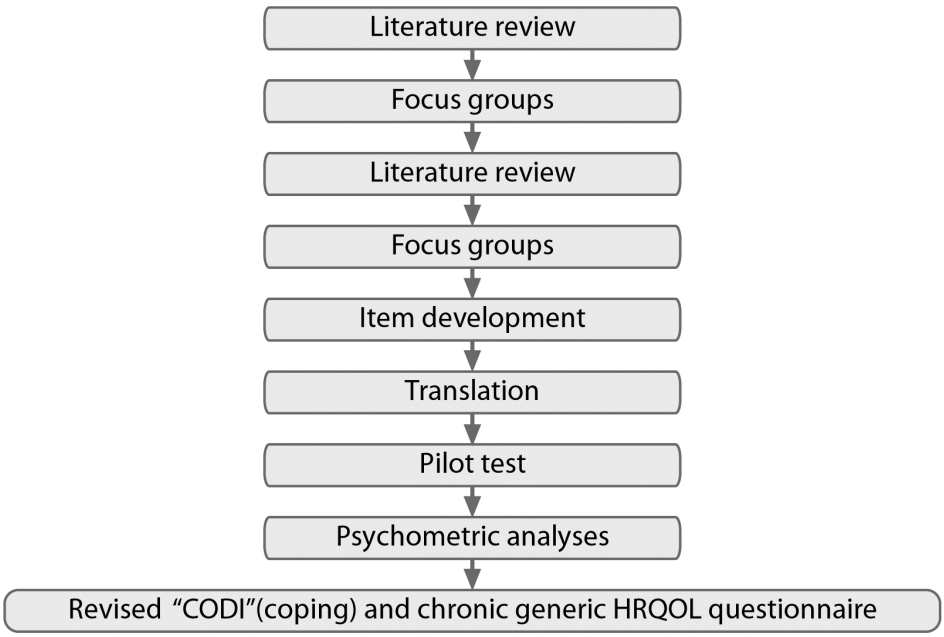


Figure 5

Instrument development

After reviewing the literature (see 2.4.2) and conducting focus groups with children, adolescents and their parents, items were written and translated. Following that, a pilot test was conducted and the items were psychometrically tested.

3.1.1 Focus Groups

The aim of the focus group work was to collect information about children's and adolescents' perception of their illness. Focus groups were conducted with children and adolescents as well as their parents and caregivers. A focus group manual delivered a general outline of the procedure in each country. The inclusion criteria for taking part in a focus group were:

- a diagnosis of asthma, arthritis, epilepsy, cerebral palsy, diabetes mellitus, atopic dermatitis or cystic fibrosis,
- age between four and sixteen years,
- the ability to understand questions, articulate thoughts, and maintain a conversation.

The participating children and adolescents were stratified by age (4-7, 8-12, 13-16 years), severity degree of the disease as rated by a clinician (mild, moderate, severe), and by the type of disease. Each country was assigned one medical condition and an additional condition if time and resources permitted. In addition to the DISABKIDS project members an Italian centre also conducted focus groups. Using medical records or contacts to special self-help groups generated a list of potential participants. Clinicians and nurses were involved in this process in order to find out whether the child or adolescent had the cognitive ability to take part in a discussion group.

A letter and a patient information sheet as well as a consent form were sent to the participants. In some countries, participants were first contacted via telephone and received an information pack afterwards. Separate focus groups were established for children, adolescents, parents and other caregivers. The groups were made up of children of similar ages. Individual interviews instead of focus groups could be conducted as a second option. In case the parents were interested in the project but could not make an appointment, a questionnaire which contained the questions of the individual interview was sent. The focus groups were conducted in a quiet room and moderated by one or two researchers experienced in working with children. Each group warmed up by getting to know each other. The procedure and aim of the discussion were again explained. Each interview took about 30 minutes up to 90 minutes and was audio taped.

Each country filled out a focus group documentation sheet. The sheet contained three sections in which statements could be grouped: a) generic, b) chronic generic and c) condition-specific. Furthermore, statements were given domain/ dimension names. If an item represented a different concept (e.g. coping) this was indicated. All statements were then collected and merged into one statement pool.

3.1.2 Item Development

The objective was to develop from the focus-item-pool items for the HRQOL and coping instrument. The preliminary pilot test versions were derived from an expert consensus which was achieved in a multistage process including a redundancy scoring, item writing, card sorting, and rereading.

Redundancy Scoring

A redundancy scoring was carried out by the German, Scottish and Dutch centres according to specific rules. Items were removed if two or more countries indicated:

- Duplicate statements
- Semantically equivalent statements
- Statements related to other constructs
- Sub-standard statements

Item Writing

After the redundancy scoring, the statements were formulated and written as items by the scientists working in each participating centre. The wording was to conform to the following guidelines:

- Use short and simple sentences.
- Ask about a single subject.
- Avoid double negatives.
- Frequency is more appreciated than intensity as an answer scale.

Card Sorting

A card sorting process was conducted. For the HRQOL instruments, chronic generic and disease-specific items were printed out and affixed on pieces of card. These cards were separated for each item pool into three different dimensions of HRQOL (psychological, social, and physical) piles.

At a project meeting in Hamburg (January 12-13, 2002) the DISABKIDS project partners were subdivided into three groups. Each group chose one of these dimensions to work with. The cards were laid out on the table. A native English speaker read the items out aloud. Items that shared a common feature were put on the same pile. This procedure was continued until all the items from the dimension were put on a particular pile. While the card sorting was taking place, any items felt to be in the wrong dimension according to the dimension definition were moved to the appropriate dimension for sorting. Whenever piles were notably small the group checked whether the items could be reassigned to another pile. Items that did not make sense or did not fit into any category were rejected as sub-standard. For each pile a name was chosen which characterised the content of the items. Once all the items had been sorted, a list of the categories (facets) within the dimensions was created.

Rereading

At the project meeting, items were read out loud. Items were removed or reformulated when they had a double meaning, were unethical or relevant only for a specific culture.

For the coping instruments all items of the focus group statement pool which were defined as coping items were included in the item development phase and a card sorting procedure was performed as well. The coping strategies were defined as deliberate actions, feelings or thoughts that occur when a child or adolescent is confronted with health-related stress. This taxonomy is non-hierarchical and the strategies are meant to be exclusive.

3.1.3 Translation

The primary objective was to forward and backward translate the HRQOL and coping items into the languages of the participating centres of the DISABKIDS project. A second objective was to harmonise translations across countries. Finally, a third objective was to finalise the pilot version of the questionnaires in each country.

For the pilot test, the English questionnaire versions had to be translated into Dutch, French, German, Greek, and Swedish. First two independent translators translated the English pilot draft version into each of the target languages. The two forward translators reviewed the translations for conceptual equivalence and decided upon a reconciled forward translation. A native English speaker performed the backward translation into English.

In the next step two project members as well as the forward translator compared the respective backward translation with the pilot draft, thus reviewing the reconciled forward translation and thereby generating the respective final forward translation. The international harmonisation took place during a meeting in Thessalonica, Greece (April 4-7, 2002) with all DISABKIDS participants and served to ensure cross-national equivalence of items. A comparison of the final forward translation across the languages was performed. Items were either modified or deleted in order to ensure the conceptual equivalence across countries.

3.2 The Pilot Test

As part of the developmental work within the DISABKIDS questionnaires, a pilot test of the newly developed HRQOL as well as the coping measure in seven countries has been conducted. The data of the thesis thus has been collected within this pilot test phase. For the pilot test procedure an agreed-upon standardised manual was followed in the participating centres (see appendix B).

3.2.1 Subjects

The preliminary version of the HRQOL and coping questionnaire were given to children and adolescents treated in a participating centre in seven different countries (Austria, France, Germany, Greece, the Netherlands, Sweden, and the United Kingdom). Patients were enrolled between May and August 2002. The inclusion criteria were:

- a chronic health condition (diagnosis of asthma, arthritis, epilepsy, cerebral palsy, diabetes mellitus, atopic dermatitis or cystic fibrosis),
- available consent form,
- age between eight and sixteen years,
- the ability to understand questions, articulate thoughts, and maintain a conversation.

With regard to the criterion of having a chronic health condition, the applied definition is in concordance with the definition as an illness that can last for an extended period, at least three months, often for life, and cannot be cured (Eiser, 1990; Midence, 1994). Per participating centre and per condition it was planned to include as a minimum 12 families in the DISABKIDS study (representing both genders, see table 5). One of the two conditions studied was asthma in every centre in order to be able to compare the questionnaire across the countries. Children aged 4-7 were also included in the pilot test of the DISABKIDS study. They received a different set of questions.

Table 5

Number of patients per conditions to be included in the pilot test

Centre	Asthma	Arthritis	Atopic Dermatitis	Dia-betes	Epilepsy	Cystic Fibrosis	Cerebral Palsy	∑
Edinburgh (UK)	24	-	-	-	12	-	12	48
Hamburg (Germany)	-	12	24	-	-	-	-	36
Leiden (Netherlands)	24	24	-	12	-	-	-	60
Luebeck (Germany)	24	12	-	-	-	-	12	48
Lund (Sweden)	24	-	-	12	24	-	-	60
Marseille (France)	24	-	24	-	12	-	-	60
Thessalonica (Greece)	24	-	-	-	-	12	12	48
Vienna (Austria)	12	-	-	24	-	12	-	48
∑	156	48	48	48	48	24	36	408

3.2.2 Procedure

The pilot study followed a cross sectional design in each participating centre. Possible participants for the DISABKIDS study were contacted in advance with an introductory letter that informed families and children about the study and included consent forms. Others were contacted during a visit at the clinic. In case the response rate with regard to the mailed letters was low, it was recommended to phone the people addressed. Informed consent was sought when the interviewer contacted the possible participants for the first time. The parent and the child or adolescent signed the consent form if he or she was able to write. Usually mothers were the respondents on the parent side unless the child routinely came to the clinic with another person. Subjects were assessed in two different settings, during clinic visit or at home.

The pilot test can be divided in three parts. The first part (A) involved the filling out of questionnaires or being interviewed, the second part (B) involved a cognitive interview and the final part (C) take home questionnaires. The cognitive interview

(part B) was not conducted with all participants of the sample depending on the time the patients and their parents were able to bestow on the study. Assistants of the participating centre conducted the pilot testing. Interviewers were trained and experienced. They had to follow the instructions in the manual. The coping questionnaire was assessed either at the clinic or distributed as a take home questionnaire (part C).

Part A: Filling out the Questionnaire/ Interview

The pilot test was preferably conducted in the clinics, but interviews at home were an alternative possibility. When parent and children arrived, they were informed about the aim of the study and the procedure. If possible, they filled out the questionnaires in different rooms. Participants willing to join in but with too little time were given the possibility to fill out a short form of the questionnaire.

Part B: Cognitive Debriefing

The children and adolescents evaluated the questionnaires. Cognitive interviews were conducted, using think-aloud technique and structured debriefing questions just for the HRQOL questionnaire. Respondents were asked to think aloud when hearing the question once again. The cognitive debriefing questions were designed to measure acceptance, relevance, appropriateness of answer categories, and the need for reformulation. Because of time exposure and burden for the child/ adolescents, just a subset of items permuted in each centre was applied to each child. The interviewer went through the subsets of questions and checked whether:

- the child/ adolescent found the question important in connection with his/her illness,
- the question was difficult to understand or answer (if so, why?),
- the response choices were clear and consistent with the question,
- the child/ adolescent would formulate the question in another way, and
- the underlying concept was interpreted correctly i.e. there were no ambiguous formulations that made more than one interpretation possible.

For the coping questionnaire, these questions were added as open questions to be filled out by the child or adolescent alone.

Part C: Take Home Questionnaires

After filling out the questionnaire and performing a cognitive debriefing in the centre, the parent received a prepaid envelope and two questionnaires to fill out at home. One was for the child and one for the parents. The questionnaire for the child was the coping questionnaire. The questionnaires had to be sent back to the respective centre within two weeks. If the questionnaire had not been sent back within two weeks the parents were phoned and politely reminded.

3.2.3 Instruments

According to the different parts of the pilot test the instruments assessed will be specified. For the ease of understanding, table 6 first of all gives an overview of the pilot test instruments with regard to the sample and modes of administration.

For part A of the pilot test, the content of the children's questionnaire (8-16 years), the caregivers' questionnaires and the medical documentation will be described followed by an account of the cognitive debriefing and take home questionnaires. In general, the questionnaire packages contain newly developed as well as standardised questions. Short versions of the questionnaires were only assessed if participants were willing to take part, but did not have enough time to do so.

Table 6

Pilot test instruments

Part	Respondent	Administration	Questionnaire Parts	No. of Items
A	• children	questionnaire	general questions	7
			anchor items	15
			generic items	119
			condition-specific items	30-48
	• caregiver	questionnaire	socio economic status	16
			generic clinical variables	11
			health status questions (FS-II-R)	20
			generic items	119
			condition-specific items	30-48
			condition-specific clinical variables	3-9
			screening questions (CSHCN)	5
	• physician	questionnaire	generic clinical variables	4
			condition-specific clinical variables	1-6
B	• children	interview	subsets of the DISABKIDS items	30-48
	• children	questionnaire	general impression	7
	• caregiver			7
C	• children	questionnaire	coping	50
			general questions	6
	• caregiver	questionnaire	health care needs	101
short versions	• children	questionnaire	coping	50
			general questions	6
			anchor items	40
			general questions	8
	• caregiver	questionnaire	socio economic status	16
			generic clinical variables	11
			health status questions (FS-II-R)	20
			screening questions (CSHCN)	5
			health care needs	101

Children's Questionnaire (8-16 years)

The children's questionnaire II (see appendix C-1) contained seven general questions about gender, age, day of birth, number of siblings, years of schooling, class/ grade and the type of school. Five items derive from the generic HRQOL measure KINDL (Ravens-Sieberer & Bullinger, 1998) and ten items from the Child Health Questionnaire (Landgraf et al., 1998). A five-point Likert response scale was utilised (1= never, 2= seldom, 3= quite often, 4= very often, 5= always). The next part of the questionnaire contains the newly developed 119 chronic generic HRQOL items. The condition-specific modules contain 42 items for cystic fibrosis, 40 items for atopic dermatitis, 32 items for diabetes, 36 items for asthma, 32 items for epilepsy, 48 items for arthritis, and 30 items for cerebral palsy.

Caregivers' Questionnaires

The caregivers' questionnaire started with 16 socio economic status variables about the relationship to the child, age, date of birth, number of persons living in the household, type of school, profession, country, language, and current economic situation. Eleven generic clinical variables about child age at onset of disease, diagnosis and treatment start, co-morbidity, development of the child, school absence, physical, social, emotional or behavioural problems were included. The clinical variables were followed by questions concerning the health status of the child/ adolescent assessed by the FS-II-R. The FS-II-R (Stein & Jessop, 1990) is a parental-report measure to inventory behavioural manifestations of illness that interfere with a child's performance of age-appropriate activities. Subsequently, the DISABKIDS items were assessed as a parent proxy-report. Finally, for caregivers of the older age group the Children with Special Health Care Needs screener (CSHCN, Bethell et al., 2002) was included which is a parent self-administered set of five questions to identify children with special or chronic health care needs. The screener is a short version of the Questionnaire for Identifying Children with Chronic Conditions (QuICCC, Stein et al., 1997). It assesses dependency on prescription medications, service use, and functional limitations.

Medical Documentation

The medical documentation contained different sets of clinical variables with regard to the respective disease. The clinicians working in the DISABKIDS GROUP suggested the variables. Four generic clinical variables were included (diagnosis, cognitive abilities, emotional/ behavioural problems) followed by specific questions about disease symptoms.

Cognitive Debriefing

According to the different dimensions of the DISABKIDS module, subsets of items were tested and a three-point scale was used to quantify the responses during the interview. The subsets were (a) medical, physical, and overall health perception domain (n= 30), (b) psychological domain (n= 38), (c) social domain (n= 52), (d) condition-specific modules (n= 30 to 48). Each item was answered with regard to its relevance (yes, sometimes, no), difficulty (yes, no), answer categories (yes, no), reformulation suggestions and associations with the question (think aloud part). To assess the general impression children and caregivers got a questionnaire containing seven open questions, four of them to be rated on a three-point scale with regard to the quality and relevance of the questions as well as the difficulties with answer categories.

Take Home Questionnaires-The Coping Module

The parent's questionnaire contained 101 items derived from the focus groups and from individual interviews asking about health care needs and quality of care. The children and adolescents take home questionnaire included the newly developed coping module called **CODI** (see appendix C-2).

3.2.4 Statistical Analyses

The focus group work was described with regard to the number of participants included in each country. For the item development process, statements were counted and sorted. Data analyses for the pilot test were carried out using the SPSS (Windows) Version 11 and the Multitrait Analysis Programme for scale structure testing (Hays et al., 1988).

A plausibility check was conducted. If out of range values or implausible values were entered into the database, they were coded as missing values. If two answers were coded, one answer was randomly picked. Each country received a list of questions with regard to detected missing values and out of range data. The centres had to respond in writing and the study centre corrected the data in the merged international data set. Each country checked again if the number of patients in the data set was correct, if the data set was completed and if the person identification numbers were identical across different data sets. For the analyses, missing values were only replaced when the scales of both questionnaires were calculated. The answer category "not applicable" of the chronic generic items of the HRQOL module was treated as missing value for the scale calculations.

The entire sample was described in terms of socio economic status variables and medical characteristics of the children and adolescents. The completed chronic generic HRQOL questionnaire and coping data was analysed both quantitatively and qualitatively. Classical multi scaling methods were applied on an item as well as on a scale level. The following analysis steps were conducted for the questionnaire development:

Item Characteristics

Descriptive statistics including range, means, percentage of missing items, and standard deviations of each item were calculated. Chi-square, Fisher and Mann-Whitney tests were used to explore differences between groups (age and gender). Item-scale correlations were calculated with the Pearson coefficient.

Scale Characteristics

Descriptive statistics including range, means, and standard deviations of the scales were calculated. The item endorsement rates were analysed and items that demonstrated floor or ceiling effects identified. The reliability of the coping scales was estimated using Cronbach's α coefficient (internal consistency) which represents the average of all possible split-half reliability estimates. Scale intercorrelations were examined. The dimensionality of the two questionnaires was explored with exploratory factor analyses. The use of the exploratory factor analysis allowed the investigation of possible alternative factors. The principal component analysis was employed as the component extraction method. The number of factors to be extracted was not specified. In order to facilitate the interpretability of factors each component matrix was rotated using the varimax with the Kaiser normalisation method. For the chronic generic HRQOL item pool, the items with a smiley answer scale were excluded from this analysis.

Open Questions

Answers for open-ended questions were reviewed for commonly occurring themes. Results concerning difficulty with understanding an item and clarity of the answer choices were examined.

Together with the information of the open questions the psychometric results were used to decide on retention, modification or rejection of items using the following criteria:

- Missing values: The percentage of missing items was treated as estimation for acceptance as well as feasibility of items.
- Item difficulty: The discriminant power of a measure is partly determined by the distribution of scores. The more the scores are spread across a continuum the more likely it is to detect differences. Skewed items were not desired.
- Item total correlation and changes in alpha: Each item in a hypothesised scale ought to correlate substantially with the construct measured; other-

wise the item does not contribute to the quality of the scale. Reliability is a function of the correlation among items of a scale. The convention is that scales with internal consistencies of at least 0.70 and higher are sufficiently reliable. Since deleting an item may lead to an increase in the alpha coefficient, the items were candidates for deletion.

- Cognitive debriefing: If children or adolescents had problems in understanding an item, this was used as an indication for deletion.
- Expert consensus: If the majority of a selected group of experts (ideally one representative per country) consented to omit or keep a certain item, this decision will be accepted.

For the revised versions of the questionnaires reliability (Cronbach's alpha) was assessed. Furthermore, age, gender, health conditions and country differences were explored with non-parametric tests. An exploratory correlation analysis of the relationship between the HRQOL and coping items was conducted.

A multiple regression analysis (stepwise procedure) with the item: "Overall, how well do you cope with your illness" as the dependent variable and the HRQOL and coping scales as the independent variables was performed. Finally, a total score of the revised HRQOL measure was calculated (=the sum of all facet scores) and included as dependent variable in a multiple regression analysis. The revised coping scales as well as age and gender were defined as independent variables.

4 Results

First of all, the results of the focus group work and item writing step will be presented. Following that, the pilot test analyses for the chronic generic HRQOL and the coping module will be described.

4.1 Focus Groups

A total of n=154 children/ adolescents, n= 142 parents and n= 26 experts took part in focus groups or interviews.

In **Austria** 3 focus groups with 16 children suffering from diabetes mellitus were carried out and 11 interviews with parents were conducted. In **France**, 11 interviews with children and adolescents with epilepsy and 9 interviews with parents were conducted. In Hamburg, **Germany**, 9 focus groups with 27 children and 1 interview with a child were conducted for atopic dermatitis. In addition, 2 experts were interviewed. In the Luebeck centre, 4 focus groups with 14 children, 3 groups with 9 parents, 1 focus group with 4 experts were carried out for arthritis as well as 4 interviews with parents. For cerebral palsy, 3 focus groups with 10 children, 3 focus groups with 12 parents, and 1 focus group with 4 experts as well as 3 interviews with experts were conducted. In **Greece**, 2 focus groups with 6 children with cystic fibrosis and 3 focus groups with 17 parents were carried out. In addition, 4 interviews with children were conducted. For asthma, 2 focus groups with 13 children and 2 focus groups with 16 parents were performed. In addition, 4 children were interviewed. In **the Netherlands**, 2 focus groups with 6 children suffering from arthritis, 3 focus groups with 16 parents and 1 focus group with 3 experts were carried out. In addition, 3 children were interviewed. For asthma, 2 focus groups with 9 children, 2 focus groups with 10 parents and 2 focus groups with 7 experts were carried out. In addition, 5 interviews with children were conducted. In **Sweden** 2 focus groups with 5 children with epilepsy and 3 focus groups with 6 children with asthma and 1 group with 2 children with diabetes were organised. 6 focus groups with 15 parents were organised as well. There were 2 interviews with children with epilepsy and 10 interviews with parents.

In the **United Kingdom**, 2 focus groups with 6 children and 3 interviews parents were carried out for cerebral palsy. In addition, 8 interviews with parents and 2 with children were conducted. For epilepsy, 1 focus group with 2 children and 1 focus group with 2 parents as well as 3 interviews with experts were conducted were carried out.

In sum, 37 children with asthma, 18 children with diabetes, 20 children with epilepsy, 28 children with atopic dermatitis, 23 children with arthritis, 10 children with cystic fibrosis and 18 children with cerebral palsy contributed to the statement generation. Table 7 gives an overview of the number of participants involved across countries with regard to their individual health condition.

Table 7

Total number of participants: focus groups and interviews

Health Condition	**Children**		**Parents**		**Experts**		∑
	FG*	**Interview**	**FG***	**Interview**	**FG***	**Interview**	
Arthritis	20	3	25	4	7	-	59
Asthma	28	9	26	-	7	-	70
Atopic Dermatitis	27	1	-	-	-	2	30
Cystic Fibrosis	6	4	17	-	-	-	27
Cerebral Palsy	16	2	12	11	4	3	48
Diabetes	18	-	-	11	-	-	29
Epilepsy	7	13	27	9	-	3	59
∑	122	32	107	35	18	8	322

*Focus Group

Altogether 1647 chronic generic statements were derived from the focus group work. With regard to the different countries involved in the statement collection, Austria collected 77, France 29, Germany 421 (Hamburg 70 and Luebeck 351), Greece 220, the Netherlands 392, Sweden 349, and the United Kingdom 99 statements. The additional Italian centre collected 60 statements. 310 statements of the total 1647 statements were rated as belonging to the concept of coping. Therefore, they were chosen for the coping questionnaire development.

4.2 Item Development

4.2.1 The Chronic Generic HRQOL Measure

The redundancy rating resulted in a reduction to 583 chronic generic statements. After the item writing process, 307 items were left. The card sorting procedure resulted in 148 chronic generic items which finally were reduced in the rereading step to 119 items. These items were then selected for the pilot test questionnaire representing 19 facets and five domains of HRQOL. Figure 6 depicts the different item reduction steps.

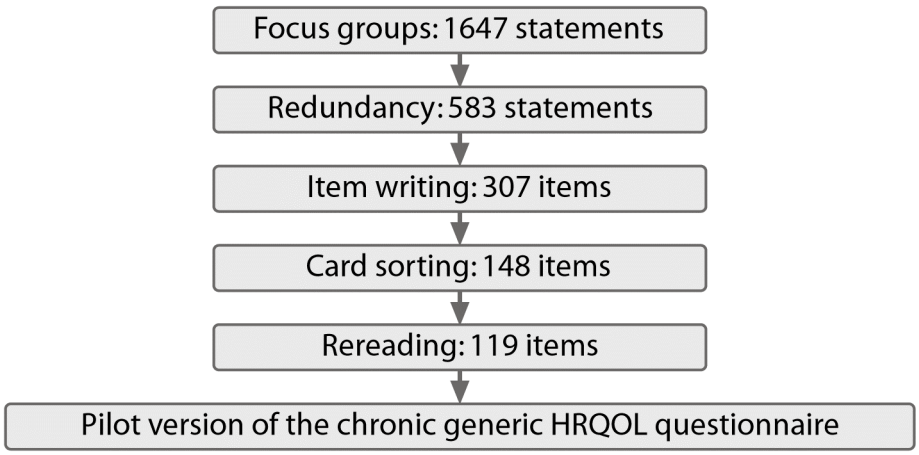


Figure 6

Item reduction process of the HRQOL chronic generic module

The items are expressed as questions in present tense. A six-point Likert response scale is utilised (1= "never", 2= "seldom", 3= "quite often", 4= "very often", 5= "always", 6= "not applicable"). The answer choice "not applicable" was only included in the pilot test version of the questionnaire in order to gain information about the applicability of items. Each facet block contains one general smiley question using a five-point scale. If necessary, items were reversed. A higher score is associated with a better HRQOL.

The content of the five dimensions of the DISABKIDS questionnaire is:

- Dimension 1 "**Psychological**": This dimension explores the psychological well-being of the child/ adolescent. The questions examine whether the child/ adolescent feels worried, unhappy, embarrassed, or anxious or whether he/ she is self-confident, enjoys life or feels independent. It also covers optimism or pessimism about the future and feelings such as loneliness and how the disease creates impacts on daily life. Five facets are included in this dimension.
- Dimension 2 "**Physical**": This dimension explores the level of the child/ adolescent's physical limitations. The mobility is examined with reference to the child/ adolescent's ability to run, move and take part in school sports. Items on sleep and the general impact of the disease are included as well. Three facets are included in this dimension.
- Dimension 3 "**Overall Health Perception**": This dimension explores the overall well-being of the child/ adolescent. General feelings about living with a disease and worries about health are considered.
- Dimension 4 "**Medical**": This dimension inspects the attitudes towards medication and treatment. It considers the acceptance of taking medicine, fear of forgetting to take it and worries about side effects. Two facets are included in this dimension.
- Dimension 5 "**Social**": This dimension examines the child/ adolescent's relationships with peers, parents and other family members. It explores social activities, the quality of interactions and whether the child/ adolescent feels supported by family and friends. In addition, this dimension scrutinises perceived stigma, general acceptance and feelings of being different because of the disease. Eight facets are included in this dimension.

The social dimension contains the largest item pool, followed by the psychological dimension. The overall health perception dimension contains of only four items. The items are depicted in table 8. These items were included in the pilot test.

Table 8

Domains, facets and item number of the chronic generic HRQOL module

Domains and Facets		Item
Psychological (38)		
▪ Future (5)	1.	Do you have fears about the future because of your condition?*
	2.	Are you confident about your future?
	3.	Do you wish your illness would go away?*
	4.	Do you feel that you will get better?
	5.	When I grow up I will be*☺
▪ Perceived Impact (10)	6.	Do you feel lonely because of your condition?*
	7.	Do you enjoy your life?
	8.	Do you feel under pressure because of your condition?*
	9.	Does your condition get you down?*
	10.	Does your condition restrict your life?*
	11.	Do you forget your condition when you do certain things (e.g. when meeting friends)?
	12.	Do you have less free time because of your condition?*
	13.	Does it bother you that your life has to be planned?*
	14.	Are you able to do everything you want to do even though you are ill?
	15.	About the restrictions in my life I feel*☺
▪ Self-Confidence (7)	16.	Does your condition make you feel bad about yourself?*
	17.	Has your illness made you feel confident about yourself?
	18.	Do you feel like everyone else even though you are ill?
	19.	Has your condition made you more grown up than other children your age?
	20.	Has your illness made you stand up for yourself?
	21.	Are you shy because of your condition?*
	22.	About myself I feel*☺
▪ Emotion (11)	23.	Are you unhappy because your are ill?*
	24.	Do you worry about your condition?*
	25.	Do you have fun in spite of your condition?
	26.	Does your condition make you angry?*
	27.	Do you hate having your condition?*
	28.	Do you think it is unfair that you are ill?*
	29.	Do you feel nervous because of your condition?*
	30.	Do you feel embarrassed that you have an illness?*
	31.	Are you ashamed that you have an illness?*
	32.	Does your condition make you moody?*
	33.	I feel*☺
▪ Autonomy (5)	34.	Do you hate having to depend on other people because of your condition?*
	35.	Are you free to lead the life you want even though you are ill?
	36.	Do you feel independent in managing your condition?
	37.	Are you able to do things without your parents?
	38.	When I do things on my own I feel*☺
Physical (11)		
▪ Limitation (4)	39.	Are you able to run and move, as you like?
	40.	Are you limited in physical activities i.e. sports, biking, running?*
	41.	Do you feel tired because of your condition?*
	42.	About the things I can do I feel*☺

*= reversed item
☺= smiley item

Table 8 continued

Domains and Facets		Item
▪ General Impact (4)	43.	Are you able to live with your condition the way it is?
	44.	Is your life ruled by your condition?*
	45.	Does it bother you that you have to explain to others what you can and can't do?*
	46.	Having my illness makes me feel*☺
▪ Sleep (3)	47.	Do you have bad dreams or nightmares because of your condition?*
	48.	Is it difficult to sleep because of your condition?*
	49.	About my sleep I feel*☺
Overall Health Perception (4)		
	50.	Is it okay for you to live with your condition?
	51.	Do you feel that everyone is healthy apart from you?*
	52.	Do you worry more than your friends about staying healthy?*
	53.	Being ill makes me feel*☺
Medical (15)		
▪ Treatment (3)	54.	Is it a problem for you to go to the doctor?*
	55.	Do you have enough time for yourself in spite of the treatment?
	56.	About the treatment of my condition I feel*☺
▪ Medication (12)	57.	Are you bothered by others watching you take your medicine?*
	58.	Are you bothered by the side effects of the medicine?*
	59.	Has your schoolwork suffered because you have been on medication?*
	60.	Does having to get help with medication from others bother you?*
	61.	Are you worried that you will forget your medicine?*
	62.	Is it annoying for you to have to remember your medication?*
	63.	Are you worried about your medication?*
	64.	Do you accept that you need medication?
	65.	Does taking medication bother you?*
	66.	Do you hate taking your medicine?*
	67.	Does taking medication disrupt everyday life?*
	68.	Taking medicine makes me feel*☺
Social (51)		
▪ School (5)	69.	Do your teachers behave differently towards you than towards others?*
	70.	Are your teachers understanding your condition?
	71.	Do you have problems concentrating at school because of your illness?*
	72.	Do you have difficulties with keeping up with the course?*
	73.	About school I feel*☺
▪ Acceptance (6)	74.	Are your friends protective of you?
	75.	Are your friends supportive?
	76.	Do your friends accept you the way you are?
	77.	Are others considerate to you?
	78.	Do other kids understand your illness?
	79.	Others make me feel*☺

*= reversed item
☺= smiley item

Table 8 continued

Domains and Facets	Item
▪ Stigma (8)	80. Do you feel that others have something against you?*
	81. Do you think that others stare at you?*
	82. Do you like it when people look at you?
	83. Are you the target of jokes?*
	84. Are you upset by other children teasing you?*
	85. Are you bothered by other people talking about you?*
	86. Do you feel excluded?*
	87. Other people treat me*☺
▪ Activities (6)	88. Do you sleep over at a friend's house?
	89. Do you go out with your friends?
	90. Are you able to play with other children?
	91. Do you take part in school sports despite having your condition?
	92. Does your condition bother you when you play?*
	93. Playing with my friends makes me feel*☺
▪ Family Support (7)	94. Do your parents argue over things to do with your condition?*
	95. Does your family bother you?*
	96. Do your parents stop you from doing some things because of your condition?*
	97. Do others in your family have complaints about your condition?*
	98. Do you get everything you want because of your illness?*
	99. Do your parents support you in your treatment?
	100. The help of my family makes me*☺
▪ Differences (6)	101. Do you think that you can do most things as well as other children?
	102. Are you one of the group?
	103. Do you feel different from other children?*
	104. Do you feel left out of things?*
	105. Do you worry that you will have problems finding a friend because of your condition?*
	106. Comparing myself to others I feel*☺
▪ Contact (6)	107. Do you get enough attention from other people?
	108. Do your friends enjoy being with you?
	109. Is it difficult for you to make friends because of your condition?*
	110. Dou you like being with other children with the same condition?
	111. Do you find it easy to talk about your illness to other people?
	112. Having friends makes me feel*☺
▪ Family Functioning (7)	113. Does your mother/father make too much of a fuss about you?*
	114. Does your condition affect the family?*
	115. Do you think that you are a worry to your parents because of your condition?*
	116. Do your parents encourage you?
	117. Are your brothers/ sisters nice to you when you are ill?
	118. Do your parents talk to you about your condition?
	119. About my family I feel*☺

*= reversed item
☺= smiley item

4.2.2 The Coping Measure (CODI)

310 coping statements were identified and included in a card sort procedure. The card sorting process resulted in a selection of altogether 50 statements for the coping questionnaire. These statements were grouped a priori to different coping strategies. The **CODI** is composed of eight coping strategies and one general question. The items are expressed as statements in the present tense and first person. The CODI questionnaire is designed for children and adolescents (aged 8-18). Respondents are asked to think of situations when they have been bothered or stressed because of their illness. The response format is a frequency five-point Likert scale ranging from "never" to "always". The overall rating ranges on a five-point scale from "very well" to "not very well at all". The items of the CODI pilot test version are depicted in table 9. The coping strategies are:

- Strategy 1 "**Spiritual Support**": includes two coping strategies focussed on religious behaviour.
- Strategy 2 "**Optimism**": includes four coping strategies focussed on either thinking positively or being pessimistic.
- Strategy 3 "**Acceptance**": includes ten coping strategies focussed on getting used to the illness or wanting to stop having it.
- Strategy 4 "**Activities**": includes three coping strategies focussed on doing special things or eating healthy food.
- Strategy 5 "**Self Disclosure**": includes five coping strategies directed at efforts to learn about the illness, to be in contact with other people and talk openly about the illness with them.
- Strategy 6 "**Expressing Negative Feelings**": includes nine coping strategies focussed upon the expression of anger, shame and tensions such as yelling and crying.
- Strategy 7 "**Distancing**": includes eleven coping strategies involving thoughts of avoidance and the feeling that everything is all right.
- Strategy 8 "**Cognitive Restructuring**": includes five coping strategies focussed upon that it could be worse and that other people have the same illness as well.
- General Question "**Overall Coping**": includes one overall rating about how well the child deals with the illness.

Table 9

Coping strategies and item numbers of the CODI questionnaire

Coping Strategy		Item
▪ Spiritual Support (2)	1.	I believe that faith in God helps me
	2.	I pray that my illness will go away
▪ Optimism (4)	3.	I think that research will help me
	4.	I am optimistic about my illness
	5.	I think positively
	6.	I don't sit in a corner and look for pity
▪ Acceptance (10)	7.	I accept my illness
	8.	I have got used to my illness
	9.	I try to do everything as normally as possible
	10.	I want to stop having my illness*
	11.	I don't want to believe that I will have my illness in the future*
	12.	I hope that my illness disappears*
	13.	I wish I were healthy*
	14.	I find it hard to carry on*
	15.	I am able to manage my illness
	16.	I cope well with my illness
▪ Activities (3)	17.	I do risky things
	18.	I eat healthy food
	19.	I do things that make me happy
▪ Self Disclosure (5)	20.	I talk openly with others about my illness
	21.	I talk with other people about my illness
	22.	I learn as much as possible about my illness
	23.	I read about my illness
	24.	I meet other kids who have the same illness
▪ Expressing Negative Feelings (9)	25.	I try to be calm*
	26.	I don't complain about my illness*
	27.	I am frustrated
	28.	I cry
	29.	I am angry
	30.	I am ashamed of being ill
	31.	I keep in mind that my illness might get worse
	32.	I wake up at night and think of terrible things
	33.	I think it is unfair that I am ill
▪ Distancing (11)	34.	I try to forget my illness
	35.	I pretend to be all right
	36.	I try to ignore my illness
	37.	I try to keep my feelings to myself
	38.	I don't care about my illness
	39.	I think my illness is no big deal
	40.	I forget about my illness
	41.	I don't think about my illness
	42.	I take my illness easy
	43.	I face my situation with humour
	44.	I think my illness is not so serious
▪ Cognitive Restructuring (5)	45.	I think it could be worse
	46.	I think there are people who suffer more than I do
	47.	I think that I am not alone with my illness
	48.	I tell myself that even famous people have illnesses
	49.	I think of worse situations
▪ Overall Coping (1)	50.	Overall, how well do you think you cope with your illness?*

*= reversed item

After conducting focus groups and developing items, the new chronic generic HRQOL and the CODI measure were pilot tested. The pilot test sample will be described. Following that the psychometric properties of the chronic generic HRQOL will be presented. Subsequently the results of the CODI testing will be described. For the ease of reference, the tables of the psychometric results will refer to the item numbers in table 8 or 9.

4.3 The Pilot Test

4.3.1 Demographic and Medical Characteristics

The sample was composed of 380 children or adolescents and 345 parents or other caregivers. In accordance with the study plan, a large majority of the sample (38.4%) had the diagnosis of asthma. The majority of the children were boys (52.7%). With regard to the parents' rating 82.6% of the children and adolescents are normally developed. The range of siblings is 0-5. Table 10 gives a demographic and medical profile of the total sample.

Table 10

Demographic and medical characteristics of the children/ adolescents (n= 380)

Characteristic		***n***	**%**
Main diagnosis	arthritis	54	14.2
	asthma	146	38.4
	atopic dermatitis	29	7.6
	cystic fibrosis	29	7.6
	cerebral palsy	21	5.5
	diabetes mellitus	64	16.8
	epilepsy	37	9.7
Sex	female	177	47.3
Co-morbidity		99	28.9
Relatives with the same condition		109	31.8
Child development (parent rating)	normal	284	82.6
	slow	49	14.2
	mental retardation	11	3.2
Characteristic		**Range**	***M (SD)***
Age		6-19	12.39 (2.59)
Child age at diagnosis		0-17	4.78 (3.96)
Years of schooling		1-13	6.68 (2.73)
Numbers of brothers		0-4	1.29 (0.91)
Numbers of sisters		0-5	1.16 (0.96)
Days of school/ pre-school kindergarten absence (during the previous year)		0-150	12.72 (25.09)
Parents rating	physical problems	1-5	1.98 (1.13)
	emotional problems	1-5	1.96 (1.02)
	social problems	1-5	1.63 (0.97)
	behavioural problems	1-5	1.61 (0.97)

Although only 8-16 year old children and adolescents should have been included for this part of the pilot test, some centres made exceptions. Mostly girls and boys aged 12 or 15 years participated. Figure 7 shows the age distributions in percent.

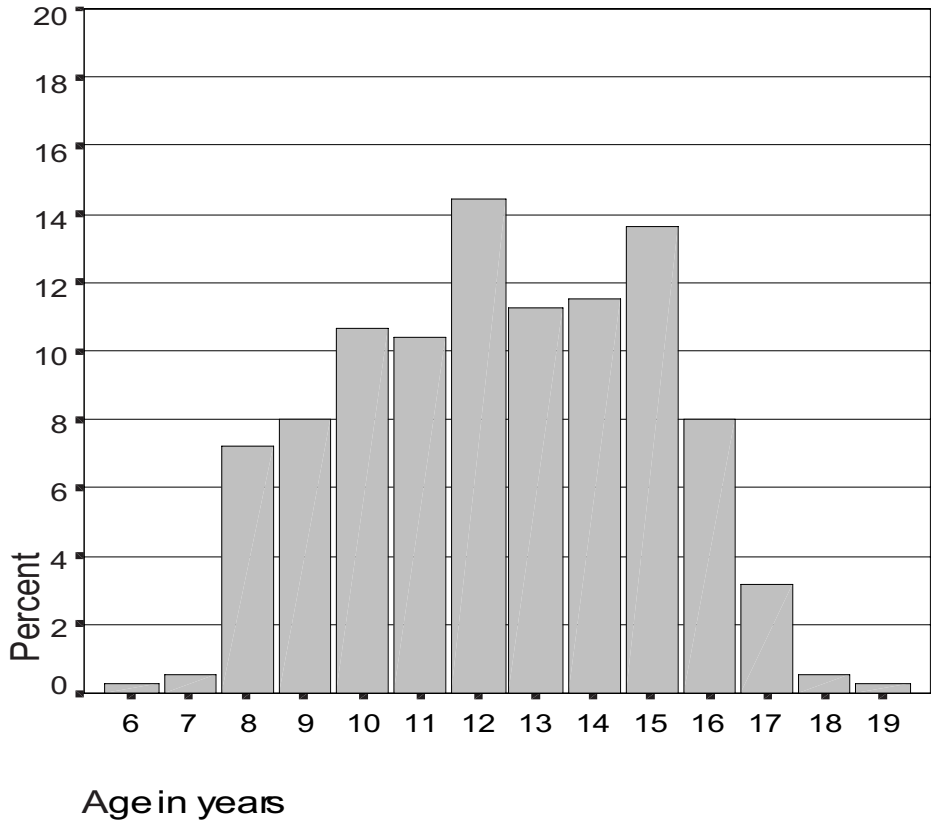


Figure 7

Age distribution in percent

With regard to the parents, mainly mothers completed the questionnaire (88.4%). The mean age of the parent who answered the questionnaire is 41.3 years. Most of the parents (81.9%) are married. 17.4% of the respondents did not answer the question of their current form of employment. Of those parents who responded, only 34.6% are currently employed full time. Table 11 gives a demographic profile of the parents' sample.

Table 11

Demographic characteristics of the parents (n=345)

Characteristic		***n***	**%**
Marital status	single	18	5.3
	married	280	81.9
	living with a partner	21	6.1
	divorced/ separated	20	5.8
	widowed	3	0.9
Employment of parent	full time	118	34.6
	part time	122	35.8
	other form of employment	21	6.1
	not employed	80	23.5
Current form of employment	worker	19	6.7
	salaried employee	139	48.8
	self-employed	28	9.8
	civil servant	41	14.4
	other	58	20.4
Economic situation (self-rating)	very well off	12	3.5
	quite well off	60	17.7
	average	222	65.5
	not very well off	24	7.1
	not at all well off	21	6.2
Characteristic		**Range**	***M (SD)***
Age		29-61	41.3 (5.56)
Days of absence at work because of child's conditions (during the previous year)		0-100	2.65 (10.41)

The parents reported a good health status of their children; only questions concerning the mood (item number 10-14) were answered with "sometimes" more often. According the parents, the mobility of most of the children and adolescents was not restricted. 90.6% of the parents answered that their child always got around the house without assistance (item number 15). Table 12 shows the distribution of responses for the questions regarding the health status of the child.

Table 12

Percentages and mean for the parents rating of the health status questions

During the last two weeks how often did your child …	**Never** %	**Sometimes** %	**Always** %	***M (SD)***
1. eat well	3.5	12.6	83.9	2.80 (0.48)
2. sleep well	2.9	15.4	81.7	2.79 (0.48)
3. seem contented and cheerful	1.2	21.7	77.1	2.76 (0.45)
4. communicate what he or she wanted	2.0	16.9	81.1	2.79 (0.46)
5. occupy himself or herself	2.9	22.1	75.0	2.72 (0.51)
6. seem lively and energetic	3.5	24.9	71.6	2.68 (0.54)
7. sleep through the night	3.5	12.0	84.5	2.81 (0.47)
8. respond to your attention	1.8	17.9	80.4	2.79 (0.45)
9. seem interested in what was going on	1.5	14.1	84.4	2.83 (0.41)
10. act moody	12.8	64.7	22.4	2.10 (0.59)
11. seem to feel sick and tired	34.0	45.9	20.1	1.86 (0.72)
12. seem unusually irritable or cross	35.6	45.8	18.7	1.83 (0.72)
13. seem unusually difficult	52.5	29.6	17.9	1.65 (0.77)
14. react to little things by crying	55.3	29.2	15.5	1.60 (0.74)
15. get around the house without assistance	5.9	3.5	90.6	2.85 (0.50)
16. go up and down stairs without assistance	5.3	2.1	92.6	2.87 (0.47)
17. communicate with words so others can understand	4.2	2.7	93.1	2.89 (0.43)
18. dress himself or herself	4.1	1.8	94.1	2.90 (0.42)
19. get undress without help	4.1	1.8	94.1	2.90 (0.42)
20. need more help with eating than other children his or her age	67.3	5.9	26.8	1.60 (0.88)

The centres assessed different numbers of patients. The highest sample size for children and adolescents was reached in Leiden, the Netherlands and the lowest sample size in Vienna, Austria. Table 13 shows the number of children and adolescents, the mean age and the percentage of females of the sample for each centre. With regard to the gender distribution more boys than girls took part in the pilot test, especially in Hamburg, Germany.

Table 13

Description of the sample from each of the seven centres (n= 380)

Centres	Country	*n*	Age: *M (SD)*	% Female
Edinburgh	UK	35	11.71 (2.64)	48.6
Hamburg	GER	67	11.79 (2.53)	37.3
Luebeck	GER	46	12.41 (2.50)	50.0
Leiden	NL	78	12.26 (2.47)	47.4
Lund	SW	30	13.63 (2.04)	50.0
Marseille	F	48	12.51 (2.83)	41.7
Thessalonica	GR	49	12.82 (2.51)	46.9
Vienna	AUS	27	12.81 (3.05)	50.0

Figure 8 depicts the gender distribution and the different types of chronic health conditions included in the pilot test. The highest difference in the gender distribution was found in the asthma population. 87 boys compared to 58 girls with asthma were included in the pilot test.

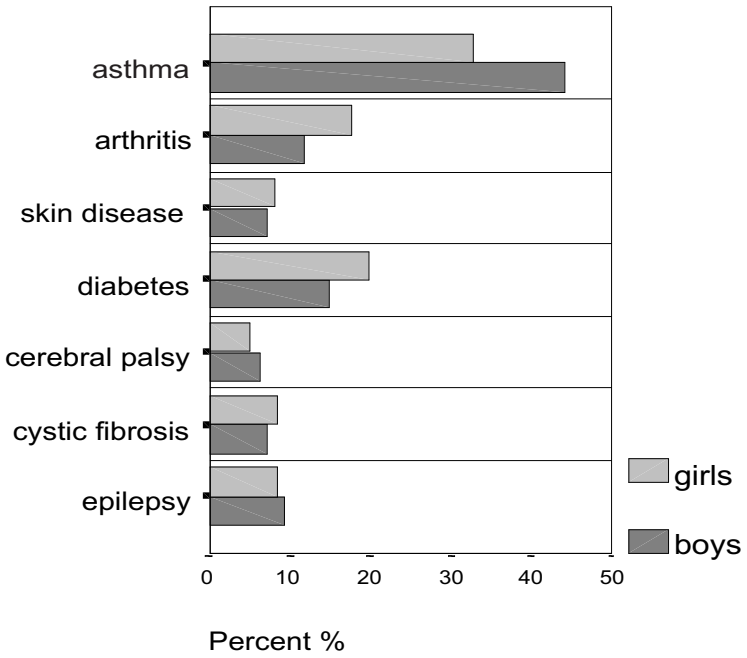


Figure 8

Gender distribution in percent with regard to the type of illnesses

188 children and adolescents filled out the coping questionnaire. With regard to gender and age 86 girls (45.7%) and 102 boys (54.3%) at the mean age of 12.69 years were included in the data analysis. 20 children and adolescents completed only the coping questionnaire. In sum, *n*=168 children and adolescents filled out the coping and the HRQOL measure.

Most of the respondents were from Germany. 93 questionnaires were administered in Hamburg, Luebeck and co-operating centres in Germany, 29 in Leiden, 5 in Edinburgh, 29 in Thessalonica, 13 in Lund, and 19 in Vienna. The centre in Marseille did not include the coping questionnaire in their pilot test. With regard to the different health conditions table 14 gives an overview of the distribution. The most frequent types of illnesses in the coping sample were asthma and arthritis. The coping strategies of only 7 children and adolescents with epilepsy were assessed.

Table 14

Included health conditions for the coping questionnaire (n= 188)

Type of Health Condition	***n***	**%**
Arthritis	38	20.2
Asthma	69	36.7
Atopic dermatitis	16	8.5
Cystic fibrosis	24	12.8
Cerebral palsy	13	6.9
Diabetes mellitus	21	11.2
Epilepsy	7	3.7

4.3.2 Instrument Performance: The Chronic Generic HRQOL Module

4.3.2.1 Item Characteristics

Table 15 shows the item characteristics for the chronic generic item pool. The item numbers correspond with **table 8**, where the items are listed. The item means are shown after reversing the respective items.

On the facet level items were identified as candidates for deletion because of their poor item total correlation. In sum, 24 items showed a poor performance (item numbers 3, 11, 19, 25, 34, 43, 47, 50, 51, 54, 55, 61, 64, 70, 82, 88, 95, 98, 99, 102, 110, 113, 114, and 118). Also items 21 and 109 showed a poor performance, but on the other hand provide clinically important information. Overall, the item distribution was skewed. The respondents predominantly scored at the ceiling.

With regard to the remaining items, five items (item numbers 17, 20, 84, 97, 117) showed a high percentage of "not applicable" responses. The items 58, 59, 60, 62 and 67 of the medical scale had a high rate of "not applicable" answers indicating the need to assess this scale in a different way (e.g. with a filter question). Eight items (item numbers 36, 52, 77, 85, 105, 107, 115, 116) showed a high percentage of missing values.

Gender effects were found for the items 5, 22, 23, 28, 34, 75, 85, 98, 100, and 112. Age effects were found for the items 3, 10, 12, 34, 36, 37, 38, 41, 44, 47, 56, 72, 76, 78, 82, 83, 88, 89, 92, 94, 95, 98, 103, 105, 107, 109, 113, 114, 115, and 118. They provided a first indication to take age and gender effects into account. The age effects were especially dominant in the social dimension. The gender effects were equally distributed across the dimensions.

Table 15

Descriptive statistics of the chronic generic item pool (n=360)

No*	M (1-5)	*SD*	Not Appl. %	Miss-ing %	Skew-ness	α**	Corr.**	Age Dif	Gender Dif	Freq.	
										1	5
1.	4.11	1.00	5.8	2.8	0.92		.44				
2.	3.94	1.09	3.3	4.4	-0.95		.33				
3.	1.77	1.17	2.5	3.1	-1.34	⇧	.15	++		•	
4.	3.56	1.29	4.7	4.2	-0.62		.22				
5.	4.41	0.72	-	4.7	1.64	-	-		+		•
6.	4.48	0.88	5.0	2.2	1.79		.58				•
7.	4.45	0.79	1.4	1.9	-1.77		.38				•
8.	4.11	1.08	3.1	3.6	1.05		.62				
9.	4.02	1.15	3.6	3.3	1.08		.64				
10.	3.88	1.23	2.5	4.2	0.93		.56	++			
11.	3.89	1.30	1.7	3.9	-0.98	⇧	.19				
12.	4.07	1.22	5.0	3.1	1.14		.40	+			•
13.	3.68	1.33	11.1	4.2	0.71		.49				
14.	3.80	1.21	3.9	3.6	-0.79		.45				
15.	3.34	1.09	-	6.1	0.32	-	-				
16.	4.19	1.05	3.6	3.3	1.29		.21				•
17.	2.80	1.35	11.7	4.7	0.14		.38				
18.	4.12	1.20	3.3	3.6	-1.28		.31				•
19.	2.40	1.37	11.1	3.9	0.52		.21				
20.	2.96	1.40	9.2	3.9	0		.38				
21.	3.80	0.61	5.6	3.6	1.98	⇧	.11				
22.	4.28	0.82	-	5.3	1.12	-	-		+		
23.	4.10	1.14	3.3	2.5	1.20		.58		+		
24.	3.82	1.15	2.8	2.8	0.87		.60				
25.	4.42	1.00	1.1	2.8	-2.06	⇧	.22				•
26.	3.89	1.27	1.9	2.8	0.93		.71				
27.	2.93	1.49	3.3	2.5	0		.56				
28.	3.54	1.48	5.3	2.8	0.56		.56		+		
29.	4.27	1.08	4.2	2.5	1.50		.59				•
30.	4.35	0.97	4.4	1.7	1.40		.63				•
31.	4.65	0.86	4.7	2.8	2.92		.48				•
32.	4.00	1.07	6.4	3.9	0.85		.58				
33.	4.21	0.87	-	5.0	1.23	-	-				
34.	3.81	1.23	13.6	3.3	0.79	⇧	.06	+	+		
35.	3.82	1.31	3.3	3.1	-0.91		.39				
36.	3.38	1.40	6.4	5.0	-0.41		.21	++			
37.	4.10	1.11	0.3	3.9	-1.31		.30	++			
38.	4.34	0.78	-	4.4	1.31	-	-	+			
39.	4.13	1.17	1.7	2.5	-1.27		.52				
40.	3.58	1.44	5.0	3.1	0.58		.44				

*= The item numbers correspond with table 8, where the items are listed.
⇧= Alpha increases if item will be deleted of that facet.
+= p≤ 0.05
++= p≤ 0.001
• = ≥50% of the answers in answer category "1" or "5"
**= Smiley items have not been included in the analyses.

Table 15 continued

No*	M (1-5)	SD	Not Appl. %	Miss-ing %	Skew-ness	α**	Corr.**	Age Dif	Gender Dif	Freq.	
										1	5
41.	3.98	1.13	5.0	2.8	0.99		.50	+			
42.	4.35	0.81	-	5.3	1.58	-	-				
43.	4.25	1.03	2.8	3.9	-1.52	⇧	.37				•
44.	3.94	1.14	5.0	4.4	0.79		.50	+			
45.	3.49	1.42	6.4	3.9	0.46		.51				
46.	2.95	1.10	-	6.1	0.22	-	-				
47.	4.73	0.67	5.3	2.8	2.79		.27	+			•
48.	4.38	0.96	4.2	3.1	1.64		.27				•
49.	4.29	0.86	-	5.3	1.10	-	-				•
50.	3.76	1.41	2.5	4.2	-0.86	⇧	.14				
51.	4.26	1.08	3.1	4.2	-1.50		.19				•
52.	3.65	1.32	6.1	5.3	-0.66		.25				
53.	2.55	1.11	-	7.8	0.17	-	-				
54.	4.10	1.18	1.9	3.6	1.30		.23				•
55.	4.32	0.93	2.5	4.2	-1.55		.23				•
56.	3.53	1.07	-	6.1	0.59	-	-	+			
57.	4.18	1.17	10.3	3.1	1.38		.37				•
58.	4.04	1.22	17.5	4.4	1.23		.39				
59.	4.42	1.05	12.2	3.6	1.88		.27				•
60.	4.19	1.12	23.9	1.7	1.46		.39				•
61.	3.24	1.77	9.4	3.1	0.20	⇧	.11				
62.	3.44	1.45	10.6	3.6	0.46		.48				
63.	4.22	1.04	9.7	4.2	1.32		.40				•
64.	3.77	1.54	8.9	3.6	-0.87	⇧	.11				•
65.	3.60	1.49	8.1	5.3	0.63		.54				
66.	3.60	1.48	9.2	4.2	0.65		.53				
67.	4.32	1.05	10.3	4.4	1.65		.52				•
68.	3.09	1.07	-	10.0	0.30	-	-				
69.	4.25	1.06	6.7	3.9	1.38		.37				•
70.	3.75	1.35	11.1	3.9	-0.79	⇧	.15				
71.	4.23	1.10	5.6	4.2	1.36		.45				•
72.	4.30	1.09	4.7	3.6	1.52		.55	+			•
73.	3.76	1.17	-	5.6	0.82	-	-				
74.	3.35	1.35	10.0	4.7	-0.31		.52				
75.	3.90	1.18	6.7	3.3	-0.84		.56		+		
76.	4.66	0.79	1.9	4.7	-2.70		.32	+			•
77.	3.86	1.20	6.7	5.6	-0.96		.41				
78.	3.64	1.22	8.1	4.7	-0.63		.40	+			
79.	4.32	0.75	-	7.2	1.12	-	-				
80.	4.37	0.84	2.8	5.0	1.24		.48				•
81.	4.42	0.89	3.9	4.4	1.64		.48				•
82.	2.19	1.24	9.7	4.7	0.84	⇧	.12	+			
83.	4.26	1.08	4.7	5.0	1.56		.38	+			•
84.	3.41	1.47	11.4	4.7	0.46		.51				

*= The item numbers correspond with table 8, where the items are listed.
⇧= Alpha increases if item will be deleted of that facet.
+= $p \leq 0.05$
++= $p \leq 0.001$
• = ≥50% of the answers in answer category "1" or "5"
**= Smiley items have not been included in the analyses.

Table 15 continued

No.*	*M* (1-5)	*SD*	Not Appl. %	Miss-ing %	Skew-ness	α**	Corr.**	Age Dif	Gender Dif	Freq.	
										1	5
85.	3.52	1.38	6.4	5.3	0.61		.47		+		
86.	4.40	0.95	3.9	4.4	1.77		.61				•
87.	4.17	0.84	-	6.1	1.20	-	-				
88.	2.71	1.18	0.8	3.1	0.05		.22	+			
89.	3.28	1.33	2.5	3.3	-0.31		.32	+			
90.	4.49	0.85	3.1	3.6	-1.87		.47				•
91.	4.40	1.10	1.9	3.6	-1.91		.28				•
92.	3.97	1.20	3.9	3.3	0.93		.25	+			
93.	4.72	0.54	-	5.3	2.38	-	-				•
94.	4.59	0.81	8.6	5.0	2.38		.30	+			•
95.	4.39	0.88	4.4	3.9	1.47		.17	+			•
96.	3.98	1.11	5.0	4.2	0.97		.18				
97.	4.74	0.59	7.5	4.2	2.66		.31				•
98.	4.02	1.19	7.5	5.0	1.14	⇧	.03	+	+		
99.	4.36	1.04	3.9	5.0	-1.70	⇧	-.10				•
100.	4.48	0.71	-	5.8	1.66	-	-		+		•
101.	4.40	0.92	1.9	6.1	1.68		.61				
102.	4.18	1.04	1.9	5.0	-1.31		.41				
103.	4.22	1.12	3.9	6.1	-1.54	⇧	.37				
104.	4.14	1.05	3.1	6.1	1.19		.53	+			
105.	4.46	0.97	6.1	5.6	1.90		.53	++			•
106.	3.84	0.94	-	6.9	0.75	-	-				
107.	3.69	1.16	3.3	5.3	-0.74		.36	+			
108.	4.41	0.77	2.5	5.0	-1.34		.32				•
109.	4.61	0.84	5.0	5.3	2.41	⇧	.07	+			•
110.	3.30	1.31	20.6	6.7	-0.19	⇧	.15				
111.	3.30	1.42	4.7	5.3	-0.33		.36				
112.	4.81	0.44	-	4.4	2.15	-	-		+		•
113.	3.91	1.17	5.0	6.4	0.91		.30	+			
114.	4.04	1.16	8.6	4.7	1.05		.19	++			
115.	3.79	1.31	5.0	5.3	0.81		.12	+			
116.	4.14	1.14	4.7	5.3	-1.34		.14				•
117.	3.96	1.20	15.0	6.7	-1.03		.20				
118.	3.17	1.23	4.2	5.3	-0.05	⇧	.06	+			
119.	4.55	0.66	-	5.6	1.71	-	-				•

*= The item numbers correspond with table 8, where the items are listed.
⇧= Alpha increases if item will be deleted of that facet.
+= $p \leq 0.05$
++= $p \leq 0.001$
• = ≥50% of the answers in answer category "1" or "5"
**= Smiley items have not been included in the analyses.

4.3.2.2 Scale Characteristics

The results for the exploratory factor analysis were obtained by using a principal component factors analysis with varimax rotation and Kaiser normalization. Smiley items were not included in the analyses. The rotation converged in 52 iterations. The analysis revealed 27 factors with eigenvalues ranging from 21.73 to 1.02, accounting for 84.78% of the variance. The first factor alone accounts for 21.73% of the variance. The factor loadings (≥ .40) are depicted in table D-1 in the appendix. The first factor extracted is about feelings and emotional states. The second factor extracted is about social integration whereas the third factor is concerned with physical issues. The fourth factor is mainly about medication. The matrix shows that the items 47, 116, 25, 118, 99, 64, 17, 98, 110, 113, and 61 only account for the last factors.

Facets

Descriptive statistics including range, means, and standard deviations are depicted in table 16. The number of items varies from 2 to 11. The lowest standard deviation was detected for the “Sleep” facet.

Table 16

Descriptive statistic: facet level (n=360)

Facets	No. of Items*	Range	*M*	*SD*
Future	4	4-20	13.39	2.69
Perceived Impact	9	9-45	36.38	5.76
Self-Confidence	6	6-30	20.28	3.54
Emotion	10	10-50	40.05	7.18
Autonomy	4	4-20	15.12	2.92
Limitation	3	3-15	11.70	2.79
General Impact	3	3-15	11.67	2.62
Sleep	2	2-10	9.11	1.26
Overall Health Perception	3	3-15	11.67	2.38
Treatment	2	2-10	8.42	1.61
Medication	11	11-55	43.03	6.90
School	4	4-20	16.53	2.86
Stigma	7	7-35	30.30	4.73
Acceptance	5	5-25	19.40	3.51
Activities	5	5-25	18.86	3.23
Family Support	6	6-30	26.01	2.51
Differences	5	5-25	21.40	3.29
Contact	5	5-25	19.32	2.89
Family Functioning	6	6-30	23.01	3.19

*smiley items were excluded

Domains

Descriptive statistics including range, means, and standard deviations of the scales are depicted in table 17. Minor ceiling effects were noted for the social domain. The internal consistency reliabilities (Cronbach's α coefficients) for the domains range from 0.45-0.89.

Table 17

Descriptive statistics and reliabilities: domain level

Domain	No. of Items	*M*	Range	*SD*	Floor %	Ceiling %	α	Scale Fit*
Medical	15	57.88	15-75	9.57	0	0	0.81	86.7
Overall Health Percept.	4	14.18	4-20	3.04	0.3	2.30	0.45	25.0
Physical	11	44.17	11-55	6.94	0	1.7	0.81	84.1
Psychological	38	146.35	38-190	20.09	0	0	0.91	88.8
Social	51	206.07	51-255	20.88	0	0	0.89	91.2

The intercorrelations between facets and domains are depicted in table D-2 in the appendix. The highest correlation was found for the "Psychological" domain and its facets "Perceived Impact" as well as "Emotion". The "Medical" domain highly correlates with the medical facet and the "Social" dimension with the facets "Stigma" and "Differences". The lowest correlations are between "Family Functioning" and "Autonomy", "School" and "Sleep", "Overall Health Perception" and "Sleep" and between "Family Support "and "Future".

4.3.2.3 Open Questions

The cognitive debriefing was performed for the items of the "Psychological" domain (item numbers 1 to 38) by *n*= 49 children and adolescents. 60 children and adolescents were cognitively debriefed for the "Medical", "Physical", and "Overall Health Perception" domain (item numbers 39 to 68). 51 children and adolescents answered the questions with regard to the "Social" domain (item numbers 69 to 119). A high percentage of respondents (≥ 30%) that had difficulties in understanding an item was detected for the items 2, 5, 10, 13, 36, 43, 46, 50, 52, 54, 55, 56, 59, 63, 67, 73, 74, 80, 82, 83, 84, 87, 89, 90, 93, 95, 98, 100, 104, 106, 113, and 119.

A high percentage of respondents (≥15%) that had difficulties with regard to the answer categories was noted for the items 5, 15, 19, 28, 42, 46, 49, 73, 79, and 87. Most of these items are smiley items.

With regard to the relevance of the items, only a low percentage of respondents (≤40%) that rated items16, 21, 29, 44, 59, 60, 81, 83, 94, and 98 as relevant. Item 83 was the item with the lowest percentage of children/ adolescents (28.0%) rating it as relevant. The items that the children and adolescents rated as "not relevant" are mostly items with a negative content (e.g. item 83: "Are you the target of jokes?") or were about medication. Table D-3 in the appendix shows the percentages of cognitive debriefing answers for each item.

4.3.2.4 Item Reduction

As a first reduction step 24 items with a low item total correlation per facet were omitted. After that the remaining items with a high percentage of missing values (*n*= 8) or not applicable answers (*n*= 5) were selected for omission. For the medical scale the "not applicable" answers were not taken into account as a criteria for deletion. The smiley items were not included in the reduction procedure.

In addition, a meeting with eight members of the DISABKIDS group from Germany, the Netherlands, Sweden, and the United Kingdom resulted in a further deletion of 8 items (expert consensus). The experts, a group of clinicians and statisticians, reviewed at a meeting in Hamburg (December 13-14, 2002) the remaining items and decided if any item should be kept or omitted because it may not provide any important clinical information.

The final version with 56 items (without the smiley items) is shown in table 18. At the expert meeting it was decided that the **smiley items** should form a separate scale and would appear as an extra module in the chronic generic questionnaire.

Table 18

Item reduction of the chronic generic item pool

No.*	Item-Total Correlation*	Not Applicable %	Missing Values %	Expert Consensus	Retain
1.					✓
2.					✓
3.	x				
4.					✓
5.	smiley item				
6.					✓
7.					✓
8.				x	
9.					✓
10.				x	
11.	x				
12.				x	
13.					✓
14.					✓
15.	smiley item				
16.					✓
17.		x			
18.					✓
19.	x				
20.		x			
21.					✓
22.	smiley item				
23.					✓
24.					✓
25.	x				
26.					✓
27.					✓
28.					✓
29.				x	
30.					✓
31.				x	
32.				x	
33.	smiley item				
34.	x				
35.					✓
36.			x		
37.					✓
38.	smiley item				
39.					✓
40.					✓
41.					✓
42.	smiley item				
43.	x				
44.					✓
45.					✓
46.	smiley item				

*= The item numbers correspond with table 8, where the items are listed.
**= First reduction step
✓= Item will be included in the revised questionnaire version.
x= Item will be omitted.

Table 18 continued

No.*	Item-Total Correlation*	Not Applicable %	Missing Values %	Expert Consensus	Retain
47.	x				
48.					✓
49.	smiley item				
50.	x				
51.	x				
52.			x		
53.	smiley item				
54.	X				
55.	X				
56.	smiley item				
57.					✓
58.					✓
59.					✓
60.					✓
61.	x				
62.					✓
63.					✓
64.	x				
65.					✓
66.					✓
67.					✓
68.	smiley item				
69.					✓
70.	x				
71.					✓
72.				x	
73.	smiley item				
74.				x	
75.					✓
76.					✓
77.			x		
78.					✓
79.	smiley item				
80.					✓
81.					✓
82.	x				
83.					✓
84.		x			
85.			x		
86.					✓
87.	smiley item				
88.	x				
89.					✓
90.					✓
91.					✓
92.					✓
93.	smiley item				
94.					✓

*= The item numbers correspond with table 8, where the items are listed.
**= First reduction step
✓= Item will be included in the revised questionnaire version.
x= Item will be omitted.

Table 18 continued

No.*	Item-Total Correlation**	Not Applicable %	Missing Values %	Expert Consensus	Retain
95.	x				
96.					✓
97.		x			
98.	x				
99.	x				
100.	smiley item				
101.					✓
102.	x				
103.					✓
104.					✓
105.			x		
106.	smiley item				
107.			x		
108.					✓
109.					✓
110.	x				
111.					✓
112.	smiley item				
113.	x				
114.	x				
115.			x		✓
116.			x		
117.		x			
118.	x				
119.	smiley item				

*= The item numbers correspond with table 8, where the items are listed.
**= First reduction step
✓= Item will be included in the revised questionnaire version.
x= Item will be omitted.

4.3.2.5 Structure of the Final HRQOL Questionnaire

The explanatory factor analysis with the reduced 56-item set revealed 16 factors that explain 71.78% of the variance. With regard to the factor analysis 4 strong factors were derived: “Emotion” (e.g. “Does your condition make you angry?”), “Social Exclusion” (e.g. “Do you feel that others have something against you?”), “Physical” (e.g. “Are you able to run and move as you like?”) and “Treatment” (e.g. Is it annoying for you to have to remember your medication?”). Although the items 94, 69, 59 do not load on its factor with regard to the content these items will remain in this facet for further testing.

In addition, the dimensions "Social Inclusion" (e.g. "Do your friends accept you the way you are?") and "Independence" (e.g. "Are you free to lead the life you want even though you are ill?") were included in the final revised HRQOL questionnaire version (see table 19).

Table 19

Final facets of the chronic generic HRQOL module

Facet	**Item No.***	**Factor Loading**
Emotion	1	.77
	26	.77
	16	72
	24	.67
	28	.64
	23	.64
	6	.63
	30	.59
	9	.54
	27	.52
	21	.44
	13	.39
Physical	39	.76
	40	.67
	41	.61
	44	.41
	48	.24
	45	.18
Social Exclusion	80	.75
	81	.72
	86	.59
	109	.54
	104	.53
	83	.38
	103	.32
	71	.31
	115	.16
	92	.14
	96	.12
	69, 94	-
Treatment	66	.77
	65	.77
	62	.76
	57	.50
	67	.35
	63	.31
	60	.31
	58	.18
	59	-
Social Inclusion	75, 76, 78, 89, 90, 91, 101, 108, 111	-
Independence	2, 4, 7, 14, 18, 35, 37	-

*= The item numbers correspond with table 8, where the items are listed.

The number of items per facet ranges from 6 to 13. The reliability coefficients (internal consistency) of the final facets range from .71 to .90 and the scale fit values from 90% to 100% (see table 20). Ceiling effects were detected for the "Physical" scale. The scale fit reaches 100% for the "Treatment" and "Emotion" facet.

Table 20

Descriptive statistics and reliabilities

Facet	No. of Items	*M*	Range	*SD*	Floor %	Ceiling %	α	Scale fit*
Emotion	12	46.81	12-60	9.54	0.6	0.3	.90	100.0
Independence	7	27.69	7-35	4.94	0	3.9	.73	97.1
Physical	6	23.58	6-30	5.04	0	13.3	.79	90.0
Social Inclusion	9	36.21	9-45	5.42	0	3.0	.71	97.8
Social Exclusion	13	55.10	13-65	8.24	0	4.2	.87	98.5
Treatment	9	35.90	9-45	7.21	0.3	8.2	.83	100.0

4.3.2.6 Gender, Age, Condition, and Country Differences

The following analyses should only be interpreted as a first exploratory approach to identify differences in the HRQOL facets due to gender, age, and type of health condition or country effects.

With regard to gender differences, the sum scores on the "Emotion" facet were significantly lower for girls than those for boys. No differences were found for the other HRQOL facets. Table 21 shows the result of the Mann-Whitney Test.

Table 21

Mann-Whitney Test for gender differences

Facet	Boys (*n*=183)		Girls (*n*=171)		*z*	*p*
	M	*SD*	*M*	*SD*		
Emotion	47.62	9.30	46.35	8.06	-2.101	0.036
Independence	23.72	4.52	23.67	3.78	0.537	0.537
Physical	23.79	4.72	23.18	4.65	0.175	0.175
Social Inclusion	35.91	4.95	36.63	4.95	0.125	0.125
Social Exclusion	55.41	7.43	55.41	6.86	-0.376	0.707
Treatment	36.50	5.98	35.52	6.37	0.161	0.161

In order to explore age differences, the sample was divided into two age groups: 6-12 years and 13-19 years. The means of these two groups were compared. Significantly higher HRQOL sum scores were found in the younger age group on the "Social Exclusion" facet. No differences were found for the other facets (see table 22).

Table 22

Mann-Whitney Test for age differences

Facet	**6-12 years (*n*=176)**		**13-19 years (*n*=178)**		***z***	***p***
	M	***SD***	***M***	***SD***		
Emotion	47.41	8.62	46.61	8.85	-0.833	0.405
Independence	23.51	4.32	23.88	4.03	-0.799	0.425
Physical	23.77	4.57	23.23	4.80	-1.065	0.287
Social Inclusion	35.89	4.84	36.62	5.06	-1.896	0.058
Social Exclusion	56.14	7.25	54.68	7.01	-2.371	0.018
Treatment	36.12	6.16	35.93	6.23	-0.142	0.887

With the exception of scores on the „Independence" scale, significant health condition differences were found for all HRQOL scales. The arthritis group showed the lowest means in the ´"Emotion" and "Social Inclusion" scales. The diabetes and asthma group showed the highest sum scores for most HRQOL facets (see table 23).

Table 23

Kruskal-Wallis Test for health condition differences (n= 360)

Facet	Health Condition	*n*	*M*	*SD*	χ^2	*p*
Emotion	Arthritis	54	42.67	9.02	32.064	0.000
	Asthma	132	49.78	7.17		
	Atopic Dermatitis	29	45.40	7.90		
	Cystic Fibrosis	28	45.07	9.43		
	Cerebral Palsy	21	43.83	9.99		
	Diabetes	59	47.83	7.81		
	Epilepsy	37	46.60	10.20		
Independence	Arthritis	54	23.31	3.98	5.525	0.478
	Asthma	132	23.95	4.04		
	Atopic Dermatitis	29	23.68	3.80		
	Cystic Fibrosis	28	23.49	4.13		
	Cerebral Palsy	21	22.94	4.72		
	Diabetes	59	24.44	3.96		
	Epilepsy	37	22.76	4.89		
Physical	Arthritis	54	21.26	4.73	32.673	0.000
	Asthma	132	24.09	4.49		
	Atopic Dermatitis	29	24.90	4.03		
	Cystic Fibrosis	28	22.66	5.61		
	Cerebral Palsy	21	20.02	4.63		
	Diabetes	59	24.36	4.07		
	Epilepsy	37	24.76	3.72		
Social Exclusion	Arthritis	54	51.96	8.50	32.209	0.000
	Asthma	132	57.47	5.97		
	Atopic Dermatitis	29	54.90	6.44		
	Cystic Fibrosis	28	53.68	7.27		
	Cerebral Palsy	21	51.38	7.31		
	Diabetes	59	56.45	6.70		
	Epilepsy	37	55.40	6.71		
Social Inclusion	Arthritis	54	33.67	5.08	33.431	0.000
	Asthma	132	36.56	4.95		
	Atopic Dermatitis	29	38.26	4.44		
	Cystic Fibrosis	28	36.16	4.12		
	Cerebral Palsy	21	34.01	5.01		
	Diabetes	59	38.16	3.82		
	Epilepsy	37	35.69	5.04		
Treatment	Arthritis	54	34.01	6.29	17.852	0.007
	Asthma	132	37.42	5.59		
	Atopic Dermatitis	29	35.12	5.94		
	Cystic Fibrosis	28	33.54	7.54		
	Cerebral Palsy	21	34.47	6.77		
	Diabetes	59	36.90	5.22		
	Epilepsy	37	36.04	6.50		

With the exception of scores on the "Emotion" and "Treatment" facet, all HRQOL facets showed significant country differences (see table 24). Scores reported by the Swedish group were the highest for the "Emotion", "Social Exclusion", "Physical", and "Treatment" scales.

Table 24

Kruskal-Wallis Test for country differences (n= 360)

Facet	Country	*n*	*M*	*SD*	χ^2	*p*
Emotion	AUS	27	47.02	8.33	7.312	0.293
	F	48	47.42	8.21		
	GER	93	45.35	9.15		
	GR	49	48.31	8.96		
	NL	78	47.16	7.96		
	SW	30	49.61	6.72		
	UK	35	46.36	10.37		
Independence	AUS	27	25.52	2.95	20.671	0.002
	F	48	22.84	4.92		
	GER	93	24.73	3.90		
	GR	49	22.60	3.70		
	NL	78	23.52	3.82		
	SW	30	23.19	3.68		
	UK	35	23.06	5.16		
Physical	AUS	27	24.39	3.81	28.693	0.000
	F	48	24.79	3.97		
	GER	93	24.01	4.14		
	GR	49	23.43	5.62		
	NL	78	21.23	4.55		
	SW	30	25.33	3.21		
	UK	35	23.26	5.51		
Social Exclusion	AUS	27	54.14	6.60	25.730	0.000
	F	48	57.10	6.31		
	GER	93	53.94	7.59		
	GR	49	57.36	7.92		
	NL	78	54.40	6.71		
	SW	30	58.34	5.77		
	UK	35	54.96	6.47		
Social Inclusion	AUS	27	37.60	4.32	18.551	0.005
	F	48	34.33	5.81		
	GER	93	36.72	5.01		
	GR	49	35.85	4.93		
	NL	78	35.58	3.93		
	SW	30	37.29	4.97		
	UK	35	37.83	4.88		
Treatment	AUS	27	34.65	6.77	4.931	0.553
	F	48	36.29	5.65		
	GER	93	35.71	6.00		
	GR	49	35.55	6.77		
	NL	78	36.10	6.07		
	SW	30	37.91	6.15		
	UK	35	36.42	5.89		

4.3.3 Instrument Performance: The Coping Questionnaire

4.3.3.1 Item Characteristics

Item characteristics of the coping questionnaire are shown in table 25. The item numbers correspond with **table 9**, where the items are listed. The item means are shown after reversing the respective items.

12 items were identified as candidates for deletion because of their poor item total correlation (item numbers 3, 5, 6, 11, 17, 18, 19, 24, 25, 26, 37, 47). With regard to the remaining 38 items, one item (item number 31) showed a high percentage of missing values. The distribution of the items was skewed, especially of items 6, 8, 9, 10, 14, 30, 32, and 47.

Mann-Whitney Tests were performed to compare means with regard to gender and age. Age differences (8-12 vs. 13-18 years) could predominantly be found in the "Self-Disclosure" scale (items 20-24). With regard to gender differences it emerged that females report higher levels of "Expression Negative Feelings" (items 28, 30, 33) and are more likely to talk about their illness (item 21). Girls compared to boys more often thought of worse situations (item 49) and had the thought that their situation could be worse (item 45). Boys did more risky things than girls (item 19).

Table 25

Descriptive statistics of the coping items (n= 188)

Item No.*	M (1-5)	SD	Missing %	Skewness	α	Corr.	Age Dif	Gender Dif	Frequency 1	Frequency 5
1.	2.85	1.56	1.6	0.11		.74	+			
2.	2.44	1.58	1.1	0.57		.74				
3.	3.32	1.30	3.2	-0.33		.14	+			
4.	3.70	1.09	3.7	-0.62		.33				
5.	3.90	1.02	2.7	-0.67		.13				
6.	2.00	1.56	1.6	1.21	⇧	-.12			•	
7.	3.72	1.30	1.6	-0.66		.56				
8.	4.25	1.02	1.6	-1.30		.46				•
9.	4.55	0.75	1.1	-1.86		.29				•
10.	1.70	1.06	1.6	1.44		.40			•	
11.	3.05	1.50	6.4	-0.68	⇧	.36				
12.	2.02	1.15	1.6	0.77		.53				
13.	2.19	1.19	1.6	0.55		.55				
14.	4.07	1.19	3.7	-1.14		.58				•
15.	4.23	0.91	1.6	-1.21		.53				
16.	4.12	1.00	1.1	-1.13		.54				
17.	2.20	1.19	1.1	0.68	⇧	-.05	+	+		
18.	3.75	1.02	1.1	-0.78	⇧	-.04				
19.	4.25	0.78	2.1	-1.25		.09				
20.	2.88	1.37	1.6	0.20		.45	++			
21.	2.59	1.16	1.1	0.36		.58	++	+		
22.	3.15	1.24	1.6	-0.07		.32	+			
23.	2.05	1.07	1.1	0.84		.31	+			
24.	2.55	1.31	2.1	0.53	⇧	.23	+			
25.	2.22	1.19	4.3	0.75	⇧	.14				
26.	3.17	1.54	4.3	-0.13	⇧	-.10	+			
27.	1.97	1.09	4.8	1.01		.59				
28.	1.92	1.01	4.3	1.02		.66		++		
29.	2.16	1.14	2.7	0.72		.65				
30.	1.66	1.06	2.1	1.67		.45		+	•	
31.	2.17	1.21	5.9	0.80		.39				
32.	1.58	1.06	2.1	2.00		.39			•	
33.	2.46	1.43	2.7	0.59		.49		+		
34.	3.29	1.34	1.1	-0.30		.36				
35.	3.07	1.50	3.2	-0.07		.42	+			
36.	3.23	1.43	2.7	-0.34		.45	+			
37.	3.09	1.32	3.7	-0.18	⇧	.14				
38.	2.48	1.40	2.1	0.41		.49				
39.	2.87	1.33	2.7	0.11		.50				
40.	3.05	1.28	2.1	-0.15		.58	+			
41.	3.14	1.17	1.1	-0.06		.54				
42.	3.43	1.28	2.7	-0.34		.55				
43.	2.97	1.37	2.7	0.01		.36				
44.	3.17	1.34	1.6	-0.11		.41				
45.	3.14	1.33	0.5	-0.23		.41	+	+		
46.	3.89	1.16	1.1	-1.00		.48				
47.	4.11	1.19	1.1	-1.29	⇧	.13				•
48.	2.89	1.41	2.1	0.06		.42				
49.	2.60	1.36	2.7	0.34		.50		++		
50.	4.04	0.95	1.6	0.78		-				

***=** The item numbers correspond with table 9, where the items are listed.
⇧= The alpha coefficient increases if the item will be deleted of that facet.
+= $p \leq 0.05$
++= $p \leq 0.001$
• = ≥50% of the answers in answer category 1 or 5

4.3.3.2 Scale Characteristics

The results for the exploratory factor analysis were obtained by using a principal component factors analysis with varimax rotation. The analysis revealed fifteen factors with eigenvalues ranging from 8.09 to 1.03 that account for 70.41% of the variance. Table 26 shows the factor loadings which are ≥ .40. The first factor extracted is about emotional reactions. The second factor extracted is about avoiding thoughts concerning the illness whereas the third factor is concerned with the acceptance of the health condition. The fourth factor is mainly about cognitive strategies.

Table 26

CODI rotated component matrix: factor loadings (n=188)

Item No.*	Component														
	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15
28	.83														
27	.79														
29	.77														
33	.74														
14	-.63														
50	-.48		.41												
32	.47														
30	.43														
31	.41														
38		.79													
39		.72													
42		.59	.48												
41		.55													
26		-.54													
40		.52													
44		.50													
15			.75												
16			.73												
8			.68												
7			.49												
1				.77											
23				.73											
2				.65											
22				.62											
10					.71										
12					.71										
13					.65										
3					-.56										
11					.55										
36						.75									
34						.71									
35						.69									
46							.80								
45							.75								
49							.58								
21								.87							
20								.87							
4									.74						
5									.72						
18										.74					
19										.45					
24											.78				
48											.49				
47											.41				
6												.81			
25													-.78		
37													.68		
17														.80	
43															.58
9															-.57

*= The item numbers correspond with table 9, where the items are listed.

Strategies

Descriptive statistics including range, means, and standard deviations of the scales are depicted in table 27. Floor and ceiling effects were detected for the "Spiritual Support" scale. The internal consistency reliabilities (Cronbachs α coefficients) for the eight coping strategies of the CODI range from 0.00 to 0.86. The "Activities" and the "Optimism" scale shows the lowest reliability coefficients and a poor scale fit.

Table 27

CODI scales descriptive statistics and reliabilities

Scale	No. of Items	Range	*M*	*SD*	Floor %	Ceiling %	α	Scale Fit
Spiritual Support	2	1-10	5.26	2.92	28.5	12.9	.86	100.0
Optimism	4	1-20	12.85	2.82	1.1	0	.27	57.1
Acceptance	10	1-50	33.99	6.51	0	0	.78	98.6
Activities	3	1-15	10.18	1.74	0	1.6	.00	9.5
Self-Disclosure	5	1-25	13.17	3.91	3.2	0	.63	94.3
Negative Feel.	9	1-45	19.24	5.82	2.7	0	.70	85.7
Distancing	11	1-55	33.75	8.22	0	1.1	.78	100.0
Cogn. Restruc.	5	1-25	16.56	4.15	0.5	4.3	.64	88.6

Scale intercorrelations are shown in table 28. The highest but negative correlation was found between the "Negative Feelings" and the "Acceptance" scale, the highest positive correlation between "Cognitive Restructuring" and "Self-Disclosure".

Table 28

Pearson's correlation matrix of the eight scales in the CODI (n= 188)

Scale		1	2	3	4	5	6	7
1	Spiritual Support							
2	Optimism	0.12						
3	Acceptance	-0.30**	0.06					
4	Activities	-0.01	0.10	0.13				
5	Self-Disclosure	0.15*	0.21**	0.13	0.12			
6	Negative Feelings	0.22**	-0.05	-0.54**	-0.02	-0.02		
7	Distancing	0.03	0.15*	0.23**	0.15*	0.01	-0.22**	
8	Cogn. Restructuring	0.07	0.20**	0.06	-0.16*	0.28**	-0.05	0.20**

*= Correlation is significant at the 0.05 level (2-tailed).

**= Correlation is significant at the 0.01 level (2-tailed).

4.3.3.3 Open Questions

45 (25%) children and adolescents thought that the coping questionnaire is "very good", 119 (66.1%) that it was "good" and 16 (8.9%) children thought it was "not good" in general. 95 (53.1%) children and adolescents said that the questions were "easy to understand", only 3 (1.7%) participants remarked that the questions were "not understandable". With regard to the items 11 (5.9%) children and adolescents had problems with the statement "I am frustrated". 7 (3.7%) children and adolescents found the statements "I don't sit in a corner and look for pity" and "I am optimistic about my illness" difficult to understand, whereas 6 (3.2%) participants had problems with the questions "I think positively" and "I think my illness is no big deal". With regard to the answer categories 112 (64%) children and adolescents had "no difficulties", only 6 (3.4%) participants reported "a lot of difficulties". 4 (2.1%) children and adolescents found it difficult to apply the answer categories to "I don't sit in a corner and look for pity".

4.3.3.4 Item Reduction

As a first reduction step items with a low item total correlation per facet were omitted. After that the remaining items with a high percentage of missing values were selected as candidates for deletion. Finally, an expert consensus decided on the further omission of eight items. Item 41 will be reformulated. The item reduction resulted in a final version of 29 items as shown in table 29.

Table 29

Item reduction of the CODI

No.*	Item-Total Correlation**	Missing %	Expert Consensus	Retain
1.				✓
2.				✓
3.	x			
4.			x	
5.	x			
6.	x			
7.				✓
8.				✓
9.			x	
10.				✓
11.	x			
12.				✓
13.				✓
14.			x	
15.				✓
16.				✓
17.	x			
18.	x			
19.	x			
20.			x	
21.			x	
22.				✓
23.			x	
24.	x			
25.	x			
26.	x			
27.				✓
28.				✓
29.				✓
30.				✓
31.		x		
32.				✓
33.				✓
34.				✓
35.				✓
36.				✓
37.	x			
38.				✓
39.				✓
40.				✓
41.				✓
42.				✓
43.				✓
44.				✓
45.			x	
46.			x	
47.	x			
48.				✓
49.				✓
50.				✓

*= The item numbers correspond with table 9, where the items are listed.
**= First reduction step
✓= Item will be included in the revised questionnaire version.
x= Item will be omitted.

4.3.3.5 Structure of the Final Coping Questionnaire

A second exploratory factor analysis was conducted with the reduced item pool. The final structure of the coping questionnaire and the items with the factor loadings are depicted in table 30. The explanatory factor analysis revealed 8 factors that explain 67.1% of the variance. The final scales of the CODI are: "Acceptance" (e.g. "I am able to manage my illness"), "Avoidance" (e.g. "I try to ignore my illness"), "Cognitive-Palliative (e.g. "I believe that faith in God helps me"), "Distance" (e.g. "I don't care about my illness"), "Emotional Reaction" (e.g. "I cry"), and "Wishful Thinking" (e.g. "I want to stop having my illness").

Table 30

Final scales of the coping questionnaire

Scale	Item No.*	Factor Loading
Acceptance	15	.78
	8	.77
	16	.71
	7	.68
	42	.55
	43	.54
Avoidance	36	.81
	35	.74
	34	.62
	41	.44
Cognitive-Palliative	1	.90
	2	.85
	22	.42
	48	.16
	49	-
Distance	38	.81
	39	.78
	44	.55
	40	.53
Emotional Reaction	28	.83
	27	.75
	29	.78
	32	.65
	30	.60
	33	.61
Wishful Thinking	10	.84
	12	.73
	13	.66

*= The item numbers correspond with table 9, where the items are listed.

The number of items per facet ranges from 3 to 6. The reliability coefficients (internal consistency) of the final strategy scales range from .69 to .82 and the scale fit values from 95% to 100% (see table 31). The scale fit reaches 100% for "Emotional Reaction", "Cognitive Palliative", "Acceptance" and "Wishful Thinking" strategy. Ceiling effects can be detected for the "Wishful Thinking" scale and floor effects for the "Emotional Reaction" scale.

Table 31

Revised CODI scales: descriptive statistics and reliabilities

Scales	**No. of Items**	***M***	**Range**	***SD***	**Floor %**	**Ceiling %**	α	**Scale Fit**
Acceptance	6	22.66	6-30	5.15	0.0	9.7	.83	100.0
Avoidance	4	12.74	4-20	4.01	1.6	4.9	.72	95.0
Cognitive-Palliative	5	13.90	5-25	4.75	2.7	1.6	.69	100.0
Distance	4	11.60	4-20	3.87	2.2	3.8	.70	95.0
Emotional Reaction	6	11.78	6-30	4.91	15.1	0.5	.82	100.0
Wishful Thinking	3	12.09	3-15	2.87	0.0	33.0	.81	100.0

4.3.3.6 Gender, Age, Condition, and Country Differences

The following analyses should only be interpreted as a first exploratory approach to identify differences due to gender, age, and type of health condition or country effects.

With regard to gender differences, the sum scores on the "Emotion Reaction" scale were significantly lower for boys than those for girls. No differences were found for the other coping strategies. Table 32 shows the result of the Mann-Whitney Test.

Table 32

Mann-Whitney Test for gender differences

Scales	**Boys (*n*=102)**		**Girls (*n*=86)**		***z***	***p***
	M	***SD***	***M***	***SD***		
Acceptance	23.02	5.23	22.35	4.72	-1.101	0.271
Avoidance	12.74	4.15	12.71	3.66	-0-007	0.995
Cognitive-Palliative	13.52	4.64	14.42	4.72	-1.264	0.206
Distance	11.79	3.96	11.34	3.67	-0.642	0.521
Emotional Reaction	10.88	4.41	12.76	5.13	-2.612	0.009
Wishful Thinking	11.95	2.96	12.25	2.69	-0.630	0.630

In order to explore age differences, the sample was divided into two age groups: 8-12 years and 13-18 years. The means of these two groups were compared. Significantly higher sum scores were found in the younger age group on the "Avoidance" scale. No differences were found for the other facets (see table 33).

Table 33

Mann-Whitney Test for age differences

Facet	**6-12 years (*n*=87)**		**13-19 years (*n*=101)**		***z***	***p***
	M	***SD***	***M***	***SD***		
Acceptance	22.60	5.44	22.81	4.61	-0.031	0.975
Avoidance	13.61	3.96	11.97	3.74	-2.946	0.003
Cognitive-Palliative	14.53	4.58	13.42	4.74	-1.799	0.072
Distance	11.75	3.83	11.44	3.84	-0.475	0.635
Emotional Reaction	11.25	4.31	12.16	5.22	-0.957	0.338
Wishful Thinking	12.51	2.62	11.72	2.98	-1.874	0.061

With the exception of sum scores on the "Emotional Reaction" and "Distance" scale, significant health condition differences were not detected (see table 34).

Table 34

Kruskal-Wallis Test for health condition differences (n= 188)

Scale	Health Condition	*n*	*M*	*SD*	χ^2	*p*
Acceptance	Arthritis	38	20.84	5.87	10.211	0.116
	Asthma	69	23.69	4.53		
	Atopic Dermatitis	16	22.12	4.19		
	Cystic Fibrosis	24	22.19	4.45		
	Cerebral Palsy	13	21.58	5.24		
	Diabetes	21	24.05	4.39		
	Epilepsy	7	24.62	6.84		
Avoidance	Arthritis	38	13.40	3.36	12.142	0.059
	Asthma	69	12.86	4.26		
	Atopic Dermatitis	16	14.31	2.12		
	Cystic Fibrosis	24	12.98	3.72		
	Cerebral Palsy	13	11.44	3.37		
	Diabetes	21	10.25	4.21		
	Epilepsy	7	13.19	4.76		
Cognitive-Palliative	Arthritis	38	14.08	4.69	6.171	0.404
	Asthma	69	13.53	5.06		
	Atopic Dermatitis	16	14.00	4.95		
	Cystic Fibrosis	24	15.42	4.48		
	Cerebral Palsy	13	13.61	3.07		
	Diabetes	21	14.45	4.46		
	Epilepsy	7	10.95	3.70		
Distance	Arthritis	38	11.29	3.37	15.636	0.016
	Asthma	69	12.78	4.07		
	Atopic Dermatitis	16	11.25	3.42		
	Cystic Fibrosis	24	10.87	3.82		
	Cerebral Palsy	13	10.90	3.21		
	Diabetes	21	9.29	3.07		
	Epilepsy	7	12.84	4.58		
Emotional Reaction	Arthritis	38	13.46	4.57	23.534	0.001
	Asthma	69	10.54	4.79		
	Atopic Dermatitis	16	12.99	5.41		
	Cystic Fibrosis	24	13.47	5.14		
	Cerebral Palsy	13	12.34	3.54		
	Diabetes	21	9.89	3.54		
	Epilepsy	7	9.86	5.40		
Wishful Thinking	Arthritis	38	12.47	2.57	6.328	0.387
	Asthma	69	11.54	3.18		
	Atopic Dermatitis	16	13.44	1.50		
	Cystic Fibrosis	24	12.57	2.59		
	Cerebral Palsy	13	12.12	2.60		
	Diabetes	21	11.90	3.06		
	Epilepsy	7	11.14	2.91		

Significant country differences were found for the "Wishful Thinking", "Avoidance", and "Cognitive-Palliative" scales (see table 35).

Table 35

Kruskal-Wallis Test for country differences (n= 188)

Scale	Country	*n*	*M*	*SD*	χ^2	*p*
Acceptance	AUS	19	22.47	4.19	8.570	0.073
	GER	93	22.88	5.56		
	GR	29	21.06	4.52		
	NL	29	22.67	4.44		
	SW	13	24.49	3.70		
	UK	5	25.88	3.34		
Avoidance	AUS	19	12.37	4.67	15.206	0.004
	GER	93	13.64	3.89		
	GR	29	11.54	3.48		
	NL	29	12.38	3.02		
	SW	13	9.95	3.72		
	UK	5	13.35	5.18		
Cognitive-Palliative	AUS	19	14.53	4.11	30.710	0.000
	GER	93	14.46	4.27		
	GR	29	16.34	4.73		
	NL	29	11.41	4.56		
	SW	13	9.25	3.44		
	UK	5	14.70	5.07		
Distance	AUS	19	9.84	3.75	2.286	0.683
	GER	93	11.66	3.96		
	GR	29	12.27	3.76		
	NL	29	12.03	2.99		
	SW	13	11.26	4.02		
	UK	5	11.12	5.53		
Emotional Reaction	AUS	19	11.89	4.11	4.327	0.364
	GER	93	11.68	4.90		
	GR	29	12.15	5.18		
	NL	29	12.14	4.81		
	SW	13	9.30	3.26		
	UK	5	13.95	7.33		
Wishful Thinking	AUS	19	12.92	2.40	11.556	0.021
	GER	93	12.49	2.80		
	GR	29	11.98	2.74		
	NL	29	10.66	2.70		
	SW	13	11.46	2.88		
	UK	5	12.00	4.24		

4.4 Relationship between the HRQOL and Coping

The highest correlation was found between the "Emotion" and the "Social Exclusion" scale of the HRQOL questionnaire. The highest correlation between the chronic generic HRQOL scales and the coping strategies was detected between the "Emotional Reaction" and the "Emotion" scale. In general, the "Acceptance" and the "Emotional Reaction" scale showed the highest correlation relationship with the HRQOL facets (see table 36).

Table 36

Correlation between the HRQOL and coping scales (n= 168)

	Scale	1	2	3	4	5	6	7	8	9	10	11
HRQOL	1 Social Exclusion											
	2 Physical	.66**										
	3 Treatment	.58**	.42**									
	4 Social Inclusion	.54**	.43**	.33**								
	5 Independence	.57**	.55**	.36**	.56**							
	6 Emotion	.70**	.65**	.63**	.41**	.50**						
Coping	7 Emotional Reac.	-.51**	-.42**	-.52**	-.32**	-.32**	-.60**					
	8 Acceptance	.40**	.31**	.38**	.44**	.45**	.55**	-.44**				
	9 Distance	.25**	.28**	.14	.25**	.28**	.38**	-.19*	.50**			
	10 Wishful Thinking	-.34**	-.23**	-.41**	-.10	-.20**	-.46**	.33**	-.25**	-.21**		
	11 Avoidance	-.04	.03	-.11	.12	.11	-.05	.16*	.16*	.37**	.20**	
	12 Cognitive-Palliat.	-.25**	-.17*	-.21**	-.10	-.11	-.24**	.27**	.01	.02	.41**	.16*

*= Correlation is significant at the 0.05 level (2-tailed).
**= Correlation is significant at the 0.01 level (2-tailed).

For a multiple regression the item "Overall, how well do you cope with your illness?" was defined as the dependent variable. The HRQOL scales ("Social Exclusion", "Physical", "Treatment", "Social Inclusion", "Independence", and "Emotion"), the coping strategy scales ("Emotional Reaction", "Acceptance", "Distance", "Wishful Thinking", "Avoidance", and "Cognitive-Palliative") as well as gender and age of the children and adolescents were defined as independent variables.

The suggested model by the multiple regression analysis included the "Acceptance", "Physical", "Emotional Reaction", "Social Inclusion", "Cognitive-Palliative", and "Wishful Thinking" scale. All the other scales as well as the variables age and gender were excluded. The model explains 45.4% of the variance (see table 37).

Table 37

Multiple regression analysis: Coping (n=168)

Independent Variables	Standardised Beta	*p*	Explained Variance % (R^2)
Acceptance	0.388	0.000	45.4
Physical	0.303	0.000	
Emotional Reaction	-0.191	0.013	
Social Inclusion	-0.137	0.038	
Cognitive-Palliative	0.201	0.002	
Wishful Thinking	-0.190	0.005	

To explain the variance with regard to HRQOL, a total score was calculated (the sum of the HRQOL facet scores). For a multiple regression the total score was defined as the dependent variable. The coping strategy scales ("Emotional Reaction", "Acceptance", "Distance", "Wishful Thinking", "Avoidance", and "Cognitive-Palliative") as well as gender and age of the children and adolescents were defined as independent variables.

The suggested model by the multiple regression analysis included the "Emotional Reaction", "Acceptance", and "Wishful Thinking" scale. All the other scales as well as the variables age and gender were excluded. The model explains 48.2% of variance (see table 38).

Table 38

Multiple regression analysis: HRQOL (n=168)

Independent Variables	Standardised Beta	*p*	Explained Variance % (R^2)
Emotional Reaction	-0.395	0.000	48.2
Acceptance	0.322	0.000	
Wishful Thinking	-0.197	0.001	

Both multiple regression models included the "Acceptance", the "Emotional Reaction", and the "Wishful Thinking" scale. The variables age and gender as well as the coping strategy scales "Distance" and "Avoidance" were excluded in both models.

5 Discussion

5.1 Summary of Main Findings

A high proportion of children and adolescents are affected by chronic health conditions. Children's and adolescents' ways of dealing with their illness and their health-related quality of life has increasingly been acknowledged to be important for the understanding of chronic childhood health conditions. While a number of empirical studies have been conducted and theoretical papers have been written, adequate assessment tools for HRQOL and coping are still lacking.

The thesis has attempted to develop a cross-national **coping** questionnaire for children and adolescents with different chronic health conditions and to psychometrically test it together with a **chronic generic HRQOL** measure. The thesis has been closely connected with the multi-centre European project "DISABKIDS". Through the combination of various methods it has been possible to provide reliable measures. The developmental steps have included focus group work, item development, translation, pilot test, and analyses.

A standardised procedure has been carried out in seven European countries. Focus group work has provided a comprehensive starting point for the questionnaire development. The focus group procedure has allowed to use a bottom-up approach for developing the facets and the items of both questionnaires. A strong argument for focus group work is that the patients' perspective of their medical condition can be taken into account for the questionnaire development. Altogether 154 children and adolescents were involved in the focus group work. Overall, 1647 statements were collected. 310 of the statements were used for the development of the coping measure. The redundancy check and card sorting procedure has been a useful method for reducing the item pool and defining a first questionnaire structure. In conclusion, the stepwise approach to questionnaire development resulted in a 119-item chronic generic HRQOL and a 50-item coping measure (CODI). The chronic generic HRQOL measure was composed of five domains ("Psychological", "Physical", "Overall Health Perception", "Medical", and "Social") and 19 facets. The coping questionnaire consisted of eight scales ("Spiritual Support", "Optimism", "Acceptance",

"Activities", "Self Disclosure", "Expressing Negative Feelings", "Distancing", and "Cognitive Restructuring") and one general question.

The CODI and HRQOL questionnaire were piloted in a sample of 380 children and adolescents with asthma, arthritis, diabetes, epilepsy, cystic fibrosis, cerebral palsy or atopic dermatitis. A detailed manual for the pilot study and pilot analyses allowed a standardised procedure across countries. 188 children and adolescents responded to the coping questionnaire. Psychometric testing and cognitive debriefing results were used to select items for the HRQOL and coping questionnaire.

The analysis of the quantitative and qualitative data gained from the pilot test showed that for the chronic generic HRQOL measure 24 items had to be omitted because of their poor item total correlation. Five items had to be omitted because of a high percentage of not applicable answers. Eight items were excluded because of a high percentage of missing values. In addition, experts consented to further omit eight items. At the expert meeting of DISABKIDS group members it was decided that the smiley items would form an additional module. Psychometric analyses of the reduced item pool with the remaining 56 items suggested a six-dimension solution ("Emotion", "Social Exclusion", "Physical", "Treatment", "Social Inclusion", and "Independence"). First exploratory analyses with regard to gender, age, health condition, and country differences were carried out. The results revealed significantly lower scores on the "Emotion" facet for girls compared to boys. Age differences were found for the "Social Exclusion" facet. The younger age group had significantly higher scores. Differences in the scores with regard to the health conditions were found for all except the "Independence" scale. With the exception of the "Emotion" and the "Treatment" facets all HRQOL scales showed significant country differences.

For the coping measure the analyses of the quantitative and qualitative data gained from the pilot test suggested to exclude 12 items because of their poor item total correlation. One item had to be omitted because of a high percentage of missing values. Again experts decided to further exclude eight items. Analyses of the reduced item pool with the remaining 29 items resulted in a six-dimension solution ("Cognitive-Palliative", "Emotional Reaction", "Acceptance", "Distance", "Wishful Thinking", and "Avoidance"). In parallel to the development of the HRQOL question-

naire, first exploratory analyses with regard to gender, age, health condition, and country differences were carried out. The results revealed significantly higher scores on the "Emotion Reaction" scale for girls compared to boys. Age differences were found for the "Avoidance" scale. The younger age group had significantly higher scores. Differences in the scores with regard to the health conditions were found for the "Emotional Reaction" and "Distance" scale. Significant country differences were detected for the "Wishful Thinking", "Avoidance", and "Cognitive-Palliative" scale.

The exploratory investigation of the relationship between HRQOL and coping revealed the highest correlations between the coping scales "Acceptance" and "Emotional Reaction" and the HRQOL facets.

A multiple regression analysis showed that the HRQOL facets "Physical" and "Social Inclusion" were included in the regression model to explain the variance of the dependent variable "Overall Coping". A second multiple regression analysis showed that the coping strategies "Emotional Reaction", "Acceptance", and "Wishful Thinking" were included in the regression model to explain variance of the dependent variable "HRQOL total score".

In conclusion, the item reduction process for both measures resulted in final questionnaire versions with sufficient reliability coefficients (internal consistency). The reliability coefficient of the final chronic generic HRQOL facets ranged from .71 to .90. The reliability coefficient of the final coping strategy scales ranged from .69 to .82. In general, the newly developed questionnaires were largely considered acceptable. This research has shown that a cross-cultural simultaneous development approach is applicable to children and adolescents.

5.2 Comparison with Other Investigations

The topics of HRQOL and coping are addressed by a large group of professionals from various fields. The centrepiece for both constructs is the perspective of the person judging the way of coping or well-being. According to a review of Eiser and Morse (2001b) there is a need to assess HRQOL in children and adolescents with

measures especially developed for this population. Adult measures may not be appropriate because they may fail to tap HRQOL aspects which are important for young people. In addition, they may pose an extra burden in terms of wording and length on them. In their report the authors formulated criteria for the development of new measures. According to them, measures should (Eiser & Morse, 2001b, p.4):

- follow established procedures for the development,
- take into account theoretical knowledge of children's understanding of illness, emotion, and ability to complete rating scales,
- include facility for child and proxy-report,
- include developmentally sensitive age-appropriate sections, and
- include generic core and disease-specific modules.

With regard to the first criteria, the newly developed HRQOL and coping measure followed established guidelines for the development. Item development through focus groups proved to be a useful procedure in prior studies (e.g. Bullinger et al., 2002). The item selection process was treated as an important part in the development of the questionnaires. According to Guyatt et al. (1986) the importance and frequency of items rated by a sample of patients should be considered as selection criteria. The importance of items rated by the children and adolescents was taken into account in the current study. Cognitive interviewing techniques as employed in this thesis are increasingly being used in questionnaire development (e.g. The WHOQOL Group, 1995). Conducting cognitive interviews with children and adolescents is a special challenge because of their developmental ability and motivation to provide information. However, research showed that children and adolescents are able to handle the demands of a cognitive interview and provide important information (Bullinger et al., 2002). The results of the current study support this point of view. Children may assign a different meaning to a wording an initially intended by the developers. This can lead to a misunderstanding of which the parties involved are unaware. To gain better understanding of concepts of respondents, these techniques

are a helpful method. However, the amount of time necessary for conduction and analysis is a weakness of this approach.

Secondly, children's knowledge was taken into account by developing the measure with a "bottom-up" approach. The statements of children and adolescents were the basis of questionnaire development. To assess children's comprehension and performance several methods have been applied in the pilot test. Thirdly, the chronic generic HRQOL and the coping questionnaires are subjective measures. The child or the adolescents describes his or her HRQOL or coping strategy. For the HRQOL chronic generic measure also a proxy-report form will be provided. With regard to the coping questionnaire future research will show if such a version might be needed. Fourthly, developmentally sensitive, age-appropriate sections for both measures need to be further considered. For example, the HRQOL measure contains several questions pertaining to the future or relationship with the opposite gender which might be assessed only in adolescents. Finally, concerning the last criteria the aim of the DISABKIDS project is to provide condition-specific HRQOL measures as well. Thus, such measures have been developed by the DISAKIDS project partners for arthritis, asthma, atopic dermatitis, cystic fibrosis, cerebral palsy, diabetes, and epilepsy. The thesis focussed on the part of developing a generic measure.

The simultaneous approach in this thesis has been applied for questionnaire development only in the adult area so far (e.g. The WHOQOL Group, 1995). Although the simultaneous approach is a complex method for questionnaire development, it certainly improved the content of both measures. Furthermore, it provided the basis for first hypothesis with regard to cultural differences. The dimensions derived for the HRQOL measure are comparable to the dimensions of the KINDL (Ravens-Sieberer & Bullinger, 1998). Nevertheless, the new measure emphases the impact of a certain chronic health condition and is not applicable for healthy children.

Coping instruments have been developed by a number of researchers (see Compas et al, 2001). The measures developed in this thesis are based on the perspectives of children and adolescents as well as on classical psychometric analyses. The strategies derived of the statements made by concerned children and adolescents are similar to the strategies identified by other authors (e.g. Spirito et al., 1995). However,

the newly developed coping questionnaire depicts the special circumstance of having a chronic health condition and is therefore an important new measure for paediatric coping research.

Researchers already addressed the question of whether culture influences HRQOL and coping strategies (e.g. McCarty et al., 1999; Ruperto et al., 2001). Studies so far focussed on adults. Nevertheless, HRQOL and coping strategies of children and adolescents in relationship with their cultural background is an important field of research. Extending research across national borders might lead to different theoretical implications. When developing questionnaires the comparability of results is of great importance. To ensure that the contents of questionnaire items can in fact be considered equivalent in different language versions requires a comprehensive methodological procedure. The extent to which concepts and dimensions are valid across countries should be examined (Anderson et al., 1993). Confirmatory as well as exploratory factor analysis has been used to test the scale structure equivalence across countries (Bullinger et al., 1993). However, a large sample size is required in order to use factor analysis. With regard to the different types of diseases in the paediatric field, this is often difficult to achieve.

In the current thesis a first attempt was made to investigate countries differences exploratory. Country differences were found for the HRQOL facets “Social Exclusion”, “Social Inclusion”, “Physical”, and “Independence”. Country effects were identified for the strategies “Wishful Thinking”, “Avoidance”, and “Cognitive-Palliative” of the coping questionnaire. At this stage these results should not be interpreted due to sample size and methodological reasons. Nevertheless, the identified differences serve as a first step toward taking country differences into account. In line with these findings McCarty et al. (1999) reported cross-cultural similarities as well as dissimilarities in coping strategies of Thai and American adolescents. The authors emphasise the necessity to take into consideration the socio cultural context for the interpretation of coping strategies. Therefore, adequate measures are needed. The findings of the thesis indicate that the chronic generic HRQOL and coping measure CODI can be assessed in cross-cultural research.

The relationship between coping and HRQOL has been approached in this thesis.

Although it is difficult to determine causalities, one suggestion is to regard coping strategies as predictors or determinants of HRQOL. The results of the multiple regression analyses suggest that the strategy "Acceptance" is positively and the strategy "Emotional Reaction" and "Wishful Thinking" are negatively related to HRQOL. Nevertheless, there are still a lot of open questions to be answered. Coping strategies can either have a direct or an indirect influence on HRQOL. A number of other factors might mediate or moderate the relationship between HRQOL and coping. Both constructs are dynamic and vary for example with age and health condition.

The examination of the relationship between HRQOL and coping might be helpful to understand certain phenomena. For example, the paradox that very ill patients describe a better HRQOL than healthy person has been delineated and summarised under the topic "response shift" in the literature (Schwartz & Sprangers, 1999; Schwartz & Meir, 1999; Sprangers & Schwartz, 1999). The authors view this paradox as a result of a shift of values and internal standards. They integrated their assumptions in a theoretical model. Coping mechanisms play an important role for the response shift of a patient. The strategies "Acceptance" and "Emotional Reaction" of the CODI measure can possibly be viewed as an important determinant for a response shift of a patient. However, the relationship between coping and HRQOL dimensions needs to be investigated in depths. Especially, the confoundation of both concepts should be avoided.

5.3 Limitations of the Study

The study has attempted to avoid a number of methodological criticisms, however, the study has several limitations. First, the number of patients across health conditions is reasonably, but with regard to the different types of health conditions rather small. Due to a small sample size per chronic health condition and differences in sample sizes per country, psychometric data have to be carefully interpreted. Second, the study was conducted in a limited number of clinics in each country. The settings of these clinics might reflect a special approach to care for a single hospital and may therefore limit the generalisation of research findings. Third, the cross sectional

design of this research with the predominant aim to develop measures of HRQOL and coping does not allow a causative conclusion to be drawn from the results reported with regard to gender, age, health condition, and country differences. The results with regard to the relationship between both constructs also have to be interpreted with caution. Furthermore, the study relies on a cross sectional sampling of children's perception. The information about the coping strategies of the children and adolescents are based on their self-report which may differ from their actual behaviour. Especially for the coping questionnaire additional information assessed with different methods could have been worth obtaining. Finally, the children and adolescents filled out the coping questionnaire at home, maybe with the help of their parents.

5.4 Research as a Process

Different points of discussion and questions pertaining to the design and nature of the questionnaire accompanied the developmental steps of both measures outlined in this thesis. This study has shown that selection of items for a questionnaire and its chosen format depend upon group decision processes. A group consensus was often not easy to obtain because arguments could be found for both controversial opinions. The whole decision making process was guided by conceptual thoughts on how to measure HRQOL in children and adolescents. On the other hand how researchers attempt to measure HRQOL indicates how they conceptualise HRQOL (Dijkers, 1999). Arguments for or against one perspective were exchanged and evaluated by all the participants of the DISABKIDS Group. The points of discussion addressed during the different development phases might be of interest for other research groups involved in questionnaire development.

The process of decision-making began with the choice of a questionnaire format. The development of questionnaires suitable for children and adolescents posed distinctive problems. Following the Piagetian framework, children in the age group 7-11 (phase of concrete operations) might have a limited cognitive ability to fill out a questionnaire. It was the aim of DISABKIDS Group to make the new questionnaires

understandable also for these children. This objected caused several problems:

With regard to the tense of the questions it was debated whether young children would be able to think of the past four weeks, if it might be necessary to have a time frame, or if the questions should be formulated in the present tense. Studies of developmental psychology showed that young children have a limited capability to think back over a long period of time (Paul, 1971). The development of time comprehension is often described as a multistage process. With entering school researchers found that children are able to understand the concept of a week (e.g. Bradley, 1947). For the DISABKIDS project it was decided to formulate questions in the present tense. In addition, it was discussed to use either a statement or a question format for the items. Concerning this issue, a consensus could not be found. Therefore, the HRQOL measure contains questions and the coping measure statements.

Another important discussion point was whether the five-point Likert answer scale was sufficient enough. Especially across chronic health conditions, the symptoms vary. As a solution the answer category "not applicable" was used in the pilot test. The information was then used for item selection. For the final version the "not applicable " category was omitted. The items of the coping questionnaire did not evoke the wish to apply the "not applicable" answer category. Concerning age differences, it was discussed among the researchers to develop versions for different age groups. A further issue related to the use of the terms "illness", "disease" or "health condition". Interestingly, a lot of children, who participated in the pilot test, did not feel different from healthy children in any way. Instead, they viewed their condition as a normal part of their life. The group decided to be very cautious using these terms in order to avoid any kind of stigmatisation for the final questionnaire versions.

5.5 Implications for Future Research

The measurement of HRQOL changes has recently been considered as being important for clinical research also in the paediatric field. It is increasingly acknowledged that children and adolescents themselves should be involved in decisions about health care. It might improve the communication between health care

providers and their patients if paediatricians, nurses, psychologists, and others involved in the treatment become more familiar with the way children deal with their disease. The implications for future research can be described from a theoretical, methodological, and practical perspective.

From a **theoretical perspective** the topic of coping with a chronic disease and HRQOL in chronically ill populations has been explored in a multitude of research populations. However, the investigation of the relationship between both constructs started only recently. Thus, more studies are warranted. With regard to the correlation between coping strategies and HRQOL scores identified in a first exploratory approach in this study, it will be useful in future research to identify causality. This causality may vary with regard to the type of disease. An interesting question for future research will be to describe the role of coping as a predictor variable for HRQOL. It can be hypothesised that special coping strategies might have an influence on certain HRQOL domains.

From a **methodological perspective** this study provided a method for assessing chronic generic HRQOL and coping strategies in children and adolescents. Nevertheless, a number of open questions remain to be resolved. For example, the relationship between the newly developed measure and other assessment methods such as proxy-report and observation needs to be examined.

From a **practical perspective** the next step will involve a field test of the new measures. The HRQOL measure will be validated and retested. The coping measure will be assessed in a larger sample. In this upcoming study, cultural differences will be further examined. A computer version of the HRQOL measure is in development and will be used for the field trial. The correspondence of paper-and-pencil and computer versions needs to be addressed in the future. After the field test an implementation phase will follow. During this phase, applicability and acceptance for clinical research will be investigated. The aim is to implement the measures in clinical intervention studies. With the help of special designed programs, skills could be taught and maladaptive strategies identified. Children and adolescents should be encouraged to generate alternative strategies and evaluate their strengths and weaknesses.

6 Summary

The growing interest of health care providers and researchers in valid and sensitive HRQOL measurement has been a driving force for the development of such instruments in the paediatric area. Historically, HRQOL has been used as a synonym for a subject-centred perspective on health. Recently, researchers started to address the relationship between HRQOL and the way of how children deal with their disease. Coping strategies may play an important role for the adaptation process and for a better HRQOL. Within the 5th Framework Programme on "Quality of Life and Management of Living Resources" the European Commission funded a project for three years starting February 1st, 2001. The aim of the project is to enhance HRQOL of children and adolescents with disabilities and their families by developing, testing, and implementing European instruments for the assessment of HRQOL. The current thesis was connected to this project and focussed on the development of two measures: a chronic generic HRQOL as well as a coping questionnaire. Using literature searches, expert consulting and focus groups with the children/ adolescents and families, items of the instruments were developed and translated into the respective languages. A pilot test with 380 children and adolescents was conducted. Children and adolescents (8-12,13-16 years) with different chronic health conditions (asthma, epilepsy, diabetes, arthritis, atopic dermatitis, cerebral palsy, and cystic fibrosis) as well as their families were included. Data was analysed according to predefined psychometric and content criteria. Analyses resulted in a 56-item version of the chronic generic HRQOL questionnaire with six domains ("Treatment", "Physical", "Emotion", "Independence", "Social Inclusion", "Social Exclusion"). The final coping questionnaire CODI contains 29 items and six coping strategies ("Emotional Reaction", "Cognitive-Palliative", "Acceptance", "Distance", "Wishful Thinking", "Avoidance"). The results of the thesis will be helpful in discussing challenges and possible solutions of European cooperation within the HRQOL field. The thesis has provided sound measures for the assessment of chronic generic HRQOL and coping. The potential implementation of the new tools in the clinical settings has to be planned, tested, and evaluated in the near future.

7 References

Aaronson, N. K., Meyerowitz, B. E., Bard, M., Bloom, J. R., Fawzy, F. I., Feldstein, M., Fink, D., Holland, J. C., Johnson, J. E., Lowman, J. T., Patterson, W. B. & Ware, J. E. (1991). Quality of life research in oncology: past achievements and future priorities. *Cancer Nursing*, *67,* 839-843.

Aaronson, N. K. (1992). Assessing the quality of life of patients in cancer clinical trials: common sense problems and common sense solutions. *European Journal of Cancer*, 28, 1304-1307.

Aaronson, N. K., Ahmedzai, S., Bergman, B., Bullinger, M., Cull, A., Duez, N. J., Filiberti, A., Flechtner, H., Fleishman, S. B. & de Haes, J. C. (1993). The European Organization for Research and Treatment of Cancer QLQ-C30: a quality-of-life instrument for use in international clinical trials in oncology. *Journal of the National Cancer Institution*, *85* (5), 365-376.

Adler, A. (1929). *Problems of neuroses: A book of case histories*. London: Kegan Paul, Trench, Treubner.

Altshuler, J. L. & Ruble, D. N. (1989). Developmental changes in children's awareness of strategies for coping with uncontrollable stress. *Child Development*, *60* (6), 1337-1349.

Anderson, R. T., Aaronson, N. K., & Wilkin, D. (1993). Critical review of the international assessments of health-related quality of life. *Quality of Life Research*, *2*, 369-395.

Anderson, R. T., Aaronson, N. K., Bullinger, M. & McBee, W. L. (1996). A review of the progress towards developing health-related quality-of-life instruments for international clinical studies and outcomes research. *Pharmacoeconomics*, *10* (4), 336-355.

Apajasalo, M., Rautonen, J., Holmberg, C., Sinkkonen, J., Aalberg, V., Pihko, H., Siimes, M. A., Kaitila, I., Makela, A., Erkkila, K. & Sintonen, H. (1996). Quality of life in preadolescence: a 17-dimensional health-related measure (17D). *Quality of Life Research*, *5* (6), 532-538.

Austin, J. K., Patterson, J. M. & Huberty, T. J. (1991). Development of the coping health inventory for children. *Journal of Pediatric Nursing*, *6* (3), 166-174.

Austin, J. K., Smith, M. S., Risinger, M. W. & McNelis, A. M. (1994). Childhood epilepsy and asthma: comparison of quality of life. *Epilepsia*, *35* (3), 608-615.

Ayers, T. S., Sandler, I. N., West, S. G. & Roosa, M. W. (1996). A dispositional and situational assessment of children's coping: testing alternative models of coping.

Journal of Personality, *64* (4), 923-958.

Baker, F. & Intagliata, J. (1982). Quality of life in the evaluation of community support systems. *Evaluation and Program Planning*, *5,* 69-79.

Band, E. B. & Weisz, J. R. (1990). Developmental differences in primary and secondary control: Coping and adjustment to juvenile diabetes. *Journal of Clinical Child Psychology*, *19,* 150-158.

Bandell-Hoekstra, I., Abu-Saad, H. H., Passchier, J. & Knipschild, P. (2000). Recurrent headache, coping, and quality of life in children: a review. *Headache*, *40* (5), 357-370.

Barbarin, O. A. (1990). Adjustment to serious illness. In B. B. Lahey & A. E. Kadzin (Eds.), *Advances in clinical child psychology* (377-403). New York: Plenum Press.

Bernhard, J., Hurny, C., Coates, A. S., Peterson, H. F., Castiglione-Gertsch, M., Gelber, R. D., Goldhirsch, A., Senn, H. J. & Rudenstam, C. M. (1997). Quality of life assessment in patients receiving adjuvant therapy for breast cancer: the IBCSG approach. The International Breast Cancer Study Group. *Annals of Oncology*, *8* (9), 825-835.

Bernhard, J., Hurny, C., Coates, A. S., Peterson, H. F., Castiglione-Gertsch, M., Gelber, R. D., Galligioni, E., Marini, G., Thurlimann, B., Forbes, J. F., Goldhirsch, A., Senn, H. J. & Rudenstam, C. M. (1998a). Factors affecting baseline quality of life in two international adjuvant breast cancer trials. International Breast Cancer Study Group (IBCSG). *British Journal of Cancer*, *78* (5), 686-693.

Bernhard, J., Peterson, H. F., Coates, A. S., Gusset, H., Isley, M., Hinkle, R., Gelber, R. D., Castiglione-Gertsch, M. & Hurny, C. (1998b). Quality of life assessment in International Breast Cancer Study Group (IBCSG) trials: practical issues and factors associated with missing data. *Statistics in Medicine*, *17* (5-7), 587-601.

Bethell, C. D., Read, D., Stein, R. E., Blumberg, S. J., Wells, N. & Newacheck, P. W. (2002). Identifying children with special health care needs: development and evaluation of a short screening instrument. *Ambulatory Pediatrics*, *2* (1), 38-48.

Biface, R. & Walsh, M. E. (1980). Development of children's concepts of illness. *Pediatrics*, *66*, 912-917.

Blechmann, E. A., Prinz, R. J. & Dumas, J. E. (1995). Coping, competence, and aggression prevention: Developmental model. *Applied & Preventive Psychology*, *4,* 211-232.

Blount, R. L., Cohen, L. L., Frank, N. C., Bachanas, P. J., Smith, A. J., Manimala, M. R. & Pate, J. T. (1997). The Child-Adult Medical Procedure Interaction Scale-Revised:

An assessment of validity. *Journal of Pediatric Psychology*, *22* (1), 73-88.

Blount, R. L., Bunke, V., Cohen, L. L. & Forbes, C. J. (2001). The Child-Adult Medical Procedure Interaction Scale-Short Form (CAMPIS-SF): validation of a rating scale for children's and adults' behaviours during painful medical procedures. *Journal of Pain and Symptom Management*, *22* (1), 591-599.

Boat, T. F. (2000). Cystic Fibrosis. In R. E. Behrman, R. M. Kliegman & H. B. Jenson (Eds.), *Textbook of Pediatrics* (1315-1327). Philadelphia: W. B. Saunders Company.

Boekaerts, M. & Röder, I. (1999). Stress, coping, and adjustment in children with a chronic disease: a review of the literature. *Disability and Rehabilitation*, *21* (7), 311-337.

Bradley, N. C. (1947). The growth of the knowledge of time in children of school age. *British Journal of Psychology*, *38*, 67-77.

Brodzinsky, D. M., Elias, M. J., Steiger, C., Simon, J., Gill, M. & Hitt, C. (1992). Coping Scale for children and youth: scale development and validation. *Journal of Applied Developmental Psychology*, *13*, 195-214.

Brook, U. & Tepper, I. (1997). Self image, coping and familial interaction among asthmatic children and adolescents in Israel. *Patient Education and Counselling*, *30* (2), 187-192.

Bruil, J. (1999). *Development of a quality of life instrument for children with chronic illness*. Leiden University: Health Psychology.

Buck, D., Jacoby, A., Baker, G. A., Ley, H. & Steen, N. (1999). Cross-cultural differences in health-related quality of life of people with epilepsy: findings from a European study. *Quality of Life Research*, *8* (8), 675-685.

Bullinger, M., Anderson, R., Cella, D. & Aaronson, N. (1993). Developing and evaluating cross-cultural instruments from minimum requirements to optimal models. *Quality of Life Research*, *2* (6), 451-459.

Bullinger, M. & Ravens-Sieberer, U. (1995). General principles, methods and areas of application of quality of life research in children. *Praxis Kinderpsychologie, Kinderpsychiatrie*, *44* (10), 391-399.

Bullinger, M. (1997a). Gesundheitsbezogene Lebensqualität und subjektive Gesundheit. *Psychotherapie, Psychosomatik, Medizinische Psychologie*, *47*,76-91.

Bullinger, M. (1997b). The challenge of cross-cultural quality of life assessment. *Psychology and Health*, *12,* 815-825.

Bullinger, M., Alonso, J., Apolone, G., Leplege, A., Sullivan, M., Wood-Dauphinee, S., Gandek, B., Wagner, A., Aaronson, N., Bech, P., Fukuhara, S., Kaasa, S. & Ware, J.

E., Jr. (1998). Translating health status questionnaires and evaluating their quality: the IQOLA Project approach. International Quality of Life Assessment. *Journal of Clinical Epidemiology*, *51* (11), 913-923.

Bullinger, M., von Mackensen, S., Fischer, K., Khair, K., Petersen, C., Ravens-Sieberer, U., Rocino, A., Sagnier, P., Tusell, J. M., van Den Berg, M. & Vicariot, M. (2002a). Pilot testing of the 'Haemo-QoL' quality of life questionnaire for haemophiliac children in six European countries. *Haemophilia*, *8* (Suppl. 2), 47-54.

Bullinger, M., Schmidt, S., Petersen, C. & the DISABKIDS Group (2002b). Assessing quality of life of children with chronic health conditions and disabilities: a European approach. *International Journal of Rehabilitation Research*, *25*, 197-206.

Bullinger, M., Petersen, C., Schmidt, S. & DISABKIDS Group (2002c). European Paediatric Health-Related Quality of Life Assessment. *Quality of Life Newsletter*, *29*, 4-5.

Cadman, D., Boyle, M., Szatmari, P. & Offord, D. R. (1987). Chronic illness, disability, and mental and social well-being - findings of the Ontario child health study. *Pediatrics*, *79,* 805-813.

Campbell, A., Converse, P. E. & Rodgers, W. L. (1976). *The quality of American life*. New York: Russel Sage.

Campbell, A. & Rodgers, W. L. (1976). *The human meaning of social change*. New York: Russel Sage.

Campbell, A. (1981). *The sense of well-being in America: recent patterns and trends*. New York: McGraw-Hill.

Cannon, W. (1929). *Bodily changes in pain, hunger, fear and rage: an account of recent researches into the function of emotional excitement*. New York: Appleton-Century Company.

Coates, A. S., Hurny, C., Peterson, H. F., Bernhard, J., Castiglione-Gertsch, M., Gelber, R. D. & Goldhirsch, A. (2000). Quality-of-life scores predict outcome in metastatic but not early breast cancer. International Breast Cancer Study Group. *Journal of Clinical Oncology*, *18* (22), 3768-3774.

Collier, J., MacKinlay, D. & Phillips, D. (2000). Norm values for the Generic Children's Quality of Life Measure (GCQ) from a large school-based sample. *Quality of Life Research*, *9* (6), 617-623.

Compas, B. E., Forsythe, C. J., Wagner, B. M. (1988). Consistency and variability in causal attributions and coping with stress. *Cognitive Therapy and Research*, *12*, 305-320.

Compas, B. E., Worsham, N. L. & Ey, S. (1992). Conceptual and developmental issues in children´s coping with stress. In A. M. La Greca, L. J. Siegel, J. L. Wallander & C. E. Walker (Eds.), *Stress and Coping in Child Health* (7-24). New York, London: The Guilford Press.

Compas, B. E., Connor-Smith, J. K., Saltzman, H., Thomsen, A. H. & Wadsworth, M. E. (2001). Coping with stress during childhood and adolescence: problems, progress, and potential in theory and research. *Psychological Bulletin*, *127* (1), 87-127.

Connor-Smith, J. K., Compas, B. E., Wadsworth, M. E., Thomsen, A. H. & Saltzman, H. (2000). Responses to stress in adolescence: measurement of coping and involuntary stress responses. *Journal of Consulting and Clinical Psychology*, *68* (6), 976-992.

Cook, M. (1993). *Levels of Personality*. London: Cassell.

Cramer, J. A., Perrine, K., Devinsky, O., Bryant-Comstock, L., Meador, K. & Hermann, B. (1998). Development and cross-cultural translations of a 31-item quality of life in epilepsy inventory. *Epilepsia*, *39* (1), 81-88.

Dijkers, M. (1999). Measuring quality of life. *American Journal of Physical Medicine & Rehabilitation*, *78*, 286-300.

Dise-Lewis, J. E. (1988). The live events and coping inventory: an assessment of stress in children. *Psychosomatic Medicine*, *50*, 484-499.

Dolgin, M. J., Blumensohn, R., Mulhern, R. K., Orbach, J., Sahler, O. J., Roghmann, K. J., Carpenter, P. J., Barbarin, O. A., Sargent, J. R., L.K., Z. & Copeland, D. R. (1997). Sibling adaptation to childhood cancer collaborative study: Cross-cultural aspects. *Journal of Psychosocial Oncology*, *15* (1), 1-14.

Doward, L. C., McKenna, S. P., Kohlmann, T., Niero, M., Patrick, D., Spencer, B. & Thorsen, H. (1998). The international development of the RGHQoL: a quality of life measure for recurrent genital herpes. *Quality of Life Research*, *7* (2), 143-153.

Drake, L. A., Patrick, D. L., Fleckman, P., Andr, J., Baran, R., Haneke, E., Sapede, C. & Tosti, A. (1999). The impact of onychomycosis on quality of life: development of an international onychomycosis-specific questionnaire to measure patient quality of life. *Journal of the American Academy of Dermatology*, *41,* 189-196.

Drotar, D., Doershuk, C. F., Stern, R. C., Boat, T. F., Boyer, W. & Matthews, L. (1981). Psychosocial functioning of children with cystic fibrosis. *Pediatrics*, *67,* 338-343.

Drotar, D. (1998). *Measuring health-related quality of life in children and adolescents-Implications for research and practice*. New Jersey: Lawrence Erlbaum.

Dunn-Geier, B. J., McGrath, P. J., Rourke, B. P., Latter, J. & D'Astous, J. (1986). Adolescent chronic pain: The ability to cope. *Pain*, *26,* 23-32.

Ebata, A. & Moos, R. (1991). Coping and adjustment in distressed and healthy adolescents. *Journal of Applied Developmental Psychology*, *12,* 35-54.

Eisen, M., Ware, J. E., Donald, C. A. & Brook, R. H. (1979). Measuring components of children's health status. *Medical Care*, *17,* 575-579.

Eisenberg, N., Fabes, R. A. & Guthrie, I. (1997). Coping with stress: The roles of regulation and development. In J. N. Sandler and S. A. Wolchik (Eds.), *Handbook of children's coping with common stressors: linking theory, research, and intervention* (41-70). New York: Plenum.

Eiser, C. (1990). Psychological effects of chronic disease. *Journal of Child Psychology and Psychiatry*, *31* (1), 85-98.

Eiser, C. (1993). *Growing Up with a Chronic Disease: The Impact on Children and Their Families*. London: Jessica Kingsley Publishers Ltd.

Eiser, C. & Jenney, M. E. (1996). Measuring symptomatic benefit and quality of life in paediatric oncology. *British Journal of Cancer*, *73* (11), 1313-1316.

Eiser, C. & Kopel, S. J. (1997). Children's perception of health and illness. In K. J. Petrie, J. A. Weinman (Eds.), *Perceptions of health and illness: current research and applications*. Singapore: Harwood Academic Publishers.

Eiser, C., Vance, Y. H. & Seamark, D. (2000). The development of a theoretically driven generic measure of quality of life for children aged 6-12 years: a preliminary report. *Child Care Health Development*, *26* (6), 445-456.

Eiser, C. & Morse, R. (2001a). A review of measures of quality of life for children with chronic illness. *Archives of Disease in Childhood*, *84* (3), 205-211.

Eiser, C. & Morse, R. (2001b). Quality-of-life measures in chronic diseases of childhood. *Health Technology Assessment*, *5* (4), 1-157.

Ellerton M. L., Ritchie, J. A., Caty, S. (1994). Factors influencing young children's coping behaviours during stressful healthcare encounters. *Maternal Child Nursing Journal*, *22* (3), 74-82.

Engel, G. L. (1977). The need for a new medical model: A challenge for biomedicine. *Science*, *196,* 129-136.

Feeny, D., Furlong, W. & Barr, R. D. (1998). Multiattribute approach to the assessment of health-related quality of life: Health Utilities Index. *Medical Pediatric Oncology*, (Suppl.1), 54-59.

Feifel, H., Strack, S. & Tong, V. (1987). Coping strategies and associate features for

medically ill patients. *Psychotherapie, Psychosomatik, Medizinische Psychologie*, *49,* 616-627.

Felce, D. & Perry, J. (1995). Quality of life: its definition and measurement. *Research in Developmental Disabilities*, *16* (1), 51-74.

Feldman, B. M., Grundland, B., McCullough, L. & Wright, V. (2000). Distinction of quality of life, health related quality of life, and health status in children referred for rheumatologic care. *Journal of Rheumatology*, *27* (1), 226-233.

Felton, B. J. & Revenson, T. A. (1984). Coping with chronic illness: a study of illness controllability and the influence of coping strategies on psychological adjustment. *Journal of Consulting and Clinical Psychology*, *52,* 343-353.

Fields, L. & Prinz, R. J. (1997). Coping and adjustment during childhood and adolescents. *Clinical Psychology Review*, *17* (8), 937-976.

Filip, S. H. (1995). Sind kranke Kinder glücklicher als kranke Erwachsene? Antworten zwischen Empirie und Spekulation. In U. Wahn, R. Szczepanski and M. Bullinger (Eds.), *Chronisch kranke Kinder: Krankheitsbewältigung und Lebensqualität* (3-22). Hamburg: EuMeCom.

Folkman, S., Lazarus, R., Dunkel-Schetter, C., DeLongis, A. & Gruen, R. (1986a). Dynamics of a stressful encounter: Cognitive appraisal, coping, and encounter outcomes. *Journal of Personality and Social Psychology*, *50,* 992-1003.

Folkman, S., Lazarus, R., Gruen, R. & DeLongis, A. (1986b). Appraisal, coping, health status, and psychological symptoms. *Journal of Personality and Social Psychology*, *50,* 571-579.

Forjaz, M. J. & Guarnaccia, C. A. (2001). A comparison of Portuguese and American patients with hematological malignancies: a cross-cultural survey of health-related quality of life. *Psychooncology*, *10* (3), 251-8.

Fowler, M. G., Johnson, M. P. & Atkinson, S. S. (1985). School achievement and absence in children with chronic health conditions. *Journal of Pediatrics*, *106* (4), 683-687.

French, D. J., Carroll, A. & Christie, M. J. (1998). Health-related quality of life in Australian children with asthma: lessons for the cross-cultural use of quality of life instruments. *Quality of Life Research*, *7* (5), 409-419.

Freud, S. (1926). Hemmung, Symptom und Angst: In S. Freud (Ed.), *Gesammelte Werke, XIV* (111-205). London: Imago.

Freud, A. (1948). *The ego and the mechanisms of defense*. London: Hogarth Press. (originally published in 1936).

Friedman, I. A. & Mann, L. (1993). Coping patterns in adolescent decision making: an Israeli-Australian comparison. *Journal of Adolescence*, *16* (2), 187-199.

Frydenberg, E. & Lewis, R. (1990). How adolescents cope with different concerns: the development of the Adolescent Coping Checklist (ACC). *Psychological Test Bulletin*, *3,* 63-73.

Frydenberg, E. & Lewis, R. (1993). *Adolescent Coping Scale: Administrator's Manual*. Victoria, Australia: The Australian Council for Educational Research.

Fuggle, P., Shand, P. A., Gill, L. J. & Davies, S. C. (1996). Pain, quality of life, and coping in sickle cell disease. *Archives of Disease in Childhood*, *75* (3), 199-203.

Gandek, B., Ware, J. E., Jr., Aaronson, N. K., Alonso, J., Apolone, G., Bjorner, J., Brazier, J., Bullinger, M., Fukuhara, S., Kaasa, S., Leplege, A. & Sullivan, M. (1998a). Tests of data quality, scaling assumptions, and reliability of the SF-36 in eleven countries: results from the IQOLA Project. International Quality of Life Assessment. *Journal of Clinical Epidemiology*, *51* (11), 1149-1158.

Gandek, B., Ware, J. E., Aaronson, N. K., Apolone, G., Bjorner, J. B., Brazier, J. E., Bullinger, M., Kaasa, S., Leplege, A., Prieto, L. & Sullivan, M. (1998b). Cross-validation of item selection and scoring for the SF-12 Health Survey in nine countries: results from the IQOLA Project. International Quality of Life Assessment. *Journal of Clinical Epidemiology*, *51* (11), 1171-1178.

Gandek, B. & Ware, J. E., Jr. (1998c). Methods for validating and norming translations of health status questionnaires: the IQOLA Project approach. International Quality of Life Assessment. *Journal of Clinical Epidemiology*, *51* (11), 953-959.

Gelber, R. D., Bonetti, M., Cole, B. F., Gelber, S. & Goldhirsch, A. (1998). Quality of life assessment in the adjuvant setting: is it relevant? International Breast Cancer Study Group. *Recent Results Cancer Res*, *152,* 373-389.

Gil, K. M., Williams, D. A., Thompson, R. J., Jr. & Kinney, T. R. (1991). Sickle cell disease in children and adolescents: the relation of child and parent pain coping strategies to adjustment. *Journal of Pediatric Psychology*, *16* (5), 643-663.

Goldbeck, L. (2001). Parental coping with the diagnosis of childhood cancer: gender effects, dissimilarity within couples, and quality of life. *Psychooncology*, *10* (4), 325-335.

Gortmaker, S. L. & Sappenfield, W. (1984). Chronic childhood disorders: prevalence and impact. *Pedatric Clinics of North America*, *31,* 3-18.

Gortmaker, S. L., Walker, D., Weitzman, M. & Sobol, A. M. (1990). Chronic conditions, socio-economic risks, and behavioural problems in children and adolescents.

Pediatrics, *85* (3), 267-276.

Graham, P., Stevenson, J. & Flynn, D. (1997). A new measure of health-related quality of life for children: preliminary findings. *Psychology and Health*, *12,* 655-665.

Grey, M., Boland, E. A., Davidson, M., Li, J. & Tamborlane, W. V. (2000). Coping skills training for youth with diabetes mellitus has long-lasting effects on metabolic control and quality of life. *Journal of Pediatrics*, *137* (1), 107-113.

Grootenhuis, M. A., van der Wel, M., de Graaf-Nijkerk, J. & Last, B. F. (1996). Exploration of a self-protective strategy in pediatric oncology staff. *Medical Pediatric Oncology*, *27* (1), 40-47.

Grootenhuis, M. A. & Last, B. F. (2001). Children with cancer with different survival perspectives: Defensiveness, control strategies, and psychological adjustment. *Psychooncology*, *10* (4), 305-314.

Guillemin, F., Bombardier, C. & Beaton, D. (1993). Cross-cultural adaptation of health-related quality of life measures: literature review and proposed guidelines. *Journal of Clinical Epidemiology*, *46,* 1417-1432.

Guillemin, F. (1995). Cross-cultural adaptation and validation of health status measures. *Scandinavian Journal of Rheumatology*, *24,* 61-63.

Guyatt, G. H. & Jaeschke, R. (1990). Measurements in clinical trials: choosing the appropriate approach. In B. Spilker (Eds.), *Quality of life assessments in clinical trials* (37-46). New York: Raven Press.

Guyatt, G. H., Feeney, D. H. & Patrick, D. L. (1993). Measuring health related quality of life. *Annals of Internal Medicine*, *8* (118), 622-629.

Guyatt, G. H., Jaeschke, R., Feeney, D. H. & Patrick, D. L. (1996). Measurement in clinical trials: Choosing the right approach. In B. Spilker (Eds.), *Quality of life and pharmacoeconomics in clinical trials* (41-48). Philadelphia: Lippincott-Raven.

Haan, N. (1977). *Coping and defending: Processes of self- environment organisation*. New York: Academic Press.

Halstead, M., Johnson, S. B. & Cunningham, W. (1993). Measuring coping in adolescents: an application of the Ways of Coping Checklist. *Journal of Clinical Child Psychology*, *22,* 337-344.

Hardy, D. F., Power, T. G. & Jaedicke, S. (1993). Examining the relation of parenting to children´s coping with everyday stress. *Child Development*, *64,* 1829-1841.

Haslem, R. (2000). The nervous system. In R. E. Behrman, R. M. Kliegman & H. B. Jenson (Eds.), *Textbook of Pediatrics* (1793-1865). Philadelphia: W. B. Saunders Company.

Hays R. D., Hayashi T., Carson S. & Ware J. E. (1988). *Users Guide for the Multitrait Analy-*

sis Program (MAP). Rand Co-operation Report No. N-2786-RC 1988

Heim, E. (1998). Coping- Erkenntnisstand der 90er Jahre. *Psychotherapie, Psychosomatik, Medizinische Psychologie*, *48,* 321-337.

Hinds, P. (1990). Quality of life in children and adolescents with cancer. *Seminars in Oncology Nursing*, *6,* 285-291.

Hui, C. & Triandis, H. C. (1985). Measurement in cross-cultural psychology: a review and comparison of strategies. *Cross-Cultural Psychology*, *16,* 131-152.

Hurny, C., Bernhard, J., Coates, A., Castiglione, M., Peterson, H. F., Gelber, R. D., Rudenstam, C. M., Goldhirsch, A. & Senn, H. J. (1994). Timing of baseline quality of life assessment in an international adjuvant breast cancer trial: its effect on patient self-estimation. The International Breast Cancer Study Group. *Annals of Oncology*, *5* (1), 65-74.

Hurny, C., Bernhard, J. & Coates, A. (1998). Quality of life assessment in the International Breast Cancer Study Group: past, present, and future. *Recent Results in Cancer Research*, *152,* 390-395.

Jensen, M. P., Turner, J. A., Romano, J. M., Karoly, P. (1991). Coping with chronic pain: a review of the literature. *Pain*, *47*, 249-283.

Kaplan, R. M., Bush, J. W. & Berry, C. C. (1978). The reliability, stability, and generalizability of a health status index. In (Eds.), *Proceedings of the American Statistical Association* (704-709). Washington, D.C.: Social Statistics Section.

Kaplan, R. M. (1989). Health outcome models for policy analysis. *Health Psychology*, *8,* 723-735.

Kazak, A. E. (1989). Families of chronically ill children: a systems and social-ecological model of adaptation and challenge. *Journal of Consulting and Clinical Psychology*, *13*, 171-182.

Keller, S. D., Ware, J. E., Jr., Bentler, P. M., Aaronson, N. K., Alonso, J., Apolone, G., Bjorner, J. B., Brazier, J., Bullinger, M., Kaasa, S., Leplege, A., Sullivan, M. & Gandek, B. (1998). Use of structural equation modeling to test the construct validity of the SF-36 Health Survey in ten countries: results from the IQOLA Project. International Quality of Life Assessment. *Journal of Clinical Epidemiology*, *51* (11), 1179-1188.

Koot, H. M. & Wallander, J. L. (2001). Future challenges in child and adolescent quality of life research. In H. M. Koot and J. L. Wallander (Eds.), *Qualitiy of life in children and adolescents: concepts, methods, and findings* London, UK: Harwood Academic Press.

Kovacs, M., Feinberg, T. L., Paulauskas, S., Finkelstein, R., Pollock, M. & Crouse-Novak, M. (1985). Initial coping responses and psychosocial characteristics of children with insulin-dependent diabetes mellitus. *Journal of Pediatrics*, *106,* 827-842.

Kupst, M. J., Schulman, J. L., Henig, G., Maurer, H., Morgan, E. & Fochtman, D. (1982). Family coping with childhood leukemia: One year after diagnosis. *Journal of Pediatric Psychology*, *7,* 157-174.

Kupst, M. J. & Schulman, J. L. (1988). Long-term coping with pediatric leukaemia: a six-year follow-up study. *Journal of Pediatric Psychology*, *13,* 7-23.

La Greca, A. (1990). Social consequences of pediatric conditions: Fertile area for future investigation and intervention. *Journal of Pediatric Psychology*, *15*, 285-307.

Landgraf, J. M., Maunsell, E., Speechley, K. N., Bullinger, M., Campbell, S., Abetz, L. & Ware, J. E. (1998). Canadian-French, German and UK versions of the Child Health Questionnaire: methodology and preliminary item scaling results. *Quality of Life Research*, *7* (5), 433-445.

Lavigne, J. V. & Faier-Routman, J. (1992). Psychological adjustment to pediatric physical disorders: A meta-analytic review. *Journal of Pediatric Psychology*, *17,* 133-157.

Lazarus, R. S (1966). *Psychological stress and the coping process*. New York: Mc Graw-Hill.

Lazarus, R. S. & Folkman, S. (1984). *Stress, appraisal, and coping*. New York: Springer.

Lazarus, R. S. & Folkman, S. (1991). The concept of coping. In A. Monat and R. S. Lazarus (Eds.), *Stress and Coping: An Anthology* (189-206). New York: Columbia University Press.

Lazarus, R. S (1993). Coping theory and research: Past, present, and future. *Psychosomatic Medicine*, *55,* 234-247.

Lindstrom, B. & Eriksson, B. (1993). Quality of life among children in the Nordic countries. *Quality of Life Research*, *2* (1), 23-32.

Lindstrom, B. (1994). *The essence of existence: on the quality of life of children in the Nordic countries*. Gothenburg, Sweden: Nordic School of Public Health.

Lohaus, A. (1993). Illness concepts of children: An overview of current research. *Zeitschrift für Klinische Psychologie, Psychopathologie, Psychotherapie*, *41* (2), 117-129.

Makipernaa, A. (1989). Long-term quality of life and psychosocial coping after treatment of solid tumours in childhood. A population-based study of 94 patients 11-28 years after their diagnosis. *Acta Paediatrica Scandinavica*, *78* (5), 728-735.

Manificat, S., Dazord, A., Cochat, P. & Nicolas, J. (1997). Evaluation of the quality of life in pediatrics: how to collect the point of view of children. *Archives of Pediatrics*, *4* (12), 1238-1246.

Manificat, S., Dazord, A., Langue, J., Danjou, G., Bauche, P., Bovet, F., Cubells, J., Luchelli, R., Tockert, E. & Conway, K. (2000). Evaluation of the quality of life of infants and very young children: validation of a questionnaire. Multicenter European study. *Archives of Pediatrics*, *7* (6), 605-614.

Manne, S. L., Bakeman, R., Jacobsen, P. B., Gorfinkle, K., Bernstein, D. & Redd, W. H. (1992). Adult-child interaction during invasive medical procedures. *Health Psychology*, *11* (4), 241-249.

Manus, H. E. & Killeen, M. R. (1995). Maintenance of self-esteem by obese children. *Journal of Child and Adolescents Psychiatric Nursing*, *8* (1), 17-27.

Marquis, P., Comte, S. & Lehert, P. (2001). International validation of the CLAU-S quality-of-life questionnaire for use in patients with intermittent claudication. *Pharmacoeconomics*, *19* (6), 667-677.

Mash, E. & Terdal, L. (1981). *Behavioral assessment of childhood disorders*. New York: Guilford Press.

McCarty, C. A., Weisz, J. R., Wanitromanee, K., Eastman, K. L., Suwanlert, S., Chaiyasit, W. & Band, E. B. (1999). Culture, coping, and context: primary and secondary control among Thai and American youth. *Journal of Child Psychology and Psychiatry*, *40* (5), 809-818.

McGee, H. M., O'Boyle, C. A., Hickey, A., O'Malley, K. & Joyce, C. R. (1991). Assessing the quality of life of the individual: the SEIQoL with a healthy and a gastroenterology unit population. *Psychological Medicine*, *21* (3), 749-759.

McKenna, S. (1994). A new theoretical approach to the measurement of quality of life. *Drug Information Journal*, *28,* 13-18.

McKenna, S. P., Doward, L. C., Kohlmann, T., Mercier, C., Niero, M., Paes, M., Patrick, D., Ramirez, N., Thorsen, H. & Whalley, D. (2001). International development of the Quality of Life in Depression Scale (QLDS). *Journal of Affective Disorders*, *63* (1-3), 189-199.

Midence, K., Fuggle, P. & Davies, S. C. (1993). Psychosocial aspects of sickle cell disease (SCD) in childhood and adolescence: a review. *British Journal of Clinical Psychology*, *32* (3), 271-280.

Midence, K. (1994). The effects of chronic illness on children and their families: an overview. *Genetic, social, and general psychology monographs*, *120* (3), 311-326.

Miller, S. M. & Mangan, C. E. (1983) Interesting effects of information and coping style in adapting to gynaecological stress: should a doctor tell all? *Journal of Personality and Social Psychology*, *45*, 223-236.

Miller, S. M. & Green, M. L. (1985). Coping with stress and frustration: origins, nature and development. In M. Lewis & C. Saarni (Eds.), *The socialization of emotions* (263-314). New York: Plenum.

Miller, M. L. (2000). Evaluation of the patient with suspected rheumatic disease. In R. E. Behrman, R. M. Kliegman & H. B. Jenson (Eds.), *Textbook of Pedatrics* (698-700). Philadelphia: W. B. Saunders Company.

Miller, M. L. & Cassidy, T. (2000). Juvenile rheumatoid arthritis. In R. E. Behrman, R. M. Kliegman and H. B. Jenson (Eds.), *Textbook of Pediatrics* (704-09). Philadelphia: W. B. Saunders Company.

Milousheva, J., Kobayashi, N. & Matsui, I. (1996). Psychosocial problems of children and adolescents with a chronic disease: coping strategies. *Acta Paediatrica Japonica*, *38* (1), 41-45.

Mitchell, W. G., Scheier, L. M. & Baker, S. A. (1994). Psychosocial, behavioral, and medical outcomes in children with epilepsy: a developmental risk factor model using longitudinal data. *Pediatrics*, *94* (4), 471-477.

Moss, R. H. (1984). *Coping with physical illness. New perspectives.* New York: Plenum Press.

Muthny, F. A. & Koch, U. (1997). Spezifität der Krankheitsverarbeitung bei Krebs. In U. Koch and J. Weis (Eds.), *Integrative Evaluation des Förderschwerpunkts "Rehabilitation von Krebskranken"*. Projektabschlußbericht BMBF Nr. 0706886.

National Information Center for Children and Youth with Disabilities (2000). General information about epilepsy. *Fact Sheet Number 6*, *Issue*.

Newacheck, P. W., Budetti, P. P. & McManus, P. (1984). Trends in childhood disability. *American Journal of Public Health*, *74* (3), 232-234.

Newacheck, P. W. Halfon, N. & Budetti, P. (1986a). Prevalence of activity limiting chronic conditions among children based on household interviews. *Journal of Chronic Diseases*, *39* (2), 63-71.

Newacheck, P. W., Budetti, P. & Halfon, N. (1986b). Trends in activity limiting chronic conditions among children. *American Journal of Public Health*, *76* (2), 178-184.

Newacheck, P. W. & Taylor, W. R. (1992). Childhood chronic illness: Prevalence, severity, and impact. *American Journal of Public Health*, *82* (3), 364-371.

Newacheck, P. W. & Halfon, N. (1998). Prevalence and impact of disabling chronic

conditions in childhood. *American Journal of Public Health*, *88* (4), 610-617.

Olah, A. (1995). Coping strategies among adolescents: A cross-cultural study. *Journal of Adolescence*, *18* (4), 491-512.

Olson, A. L., Johansen, S. G., Powers, L. E., Pope, J. B. & Klein, R. B. (1993). Cognitive coping strategies of children with chronic illness. *Journal of Developmental and Behavioral Pediatrics*, *14* (4), 217-223.

Parkerson, G. R., Jr., Willke, R. J. & Hays, R. D. (1999). An international comparison of the reliability and responsiveness of the Duke Health Profile for measuring health-related quality of life of patients treated with alprostadil for erectile dysfunction. *Medical Care*, *37* (1), 56-67.

Patrick, D. L., Danis, M., Southerland, L. I. & Hong, G. (1988). Quality of life following intensive care. *Journal of general internal medicine*, *3* (3), 218-223.

Patrick, D. L. & Bergner, M. (1990). Measurement of health status in the 1990s. *Annual Review of Public Health*, *11,* 165-183.

Patterson, J. M. & Garwick, A. W. (1994). The impact of chronic illness on families: A family systems perspective. *Annals of behavioral medicine*, *16* (2), 131-142.

Paul, J. (1971). Entwicklungspsychologie und –psychiatrie des Zeitbewußtseins bei Kindern und Jugendlichen. *Praxis der Kinderpsychologie und Kinderpsychiatrie*, *20*, 241-248.

Perrez, M. & Reicherts, M. (1992). *Stress, coping, and health: a situational-behavior approach-theory, methods, applications*. Seattle: Hogrefe & Huber.

Perrin, J. M. & MacLean, W. (1988). Children with chronic illness: the prevention of dysfunction. *Pediatric Clinics of North America*, *35*, 26-30.

Perrin, J. M., MacLean, W. & Perrin, E. (1989). Parental perception of health status and psychological adjustment of children with asthma. *Pediatric*, *83* (1), 26-30.

Perrin, E., Newacheck, P., Pless, I. B., Drotar, D., Gortmaker, S. L., Leventhal, J., Perrin, J. M., Stein, R. E. K., Walker, D. K. & Weitzman, M. (1993). Issues Involved in the Definition and Classification of Chronic Health Conditions. *Pediatrics*, *91* (4), 787-793.

Perrin, J. M. & Shonkoff, J. P. (2000). Developmental disabilities and chronic illness: An overview. In R. E. Behrman, R. M. Kliegman & H. B. Jenson (Eds.), *Textbook of Pediatrics* (121-129). Philadelphia: W. B. Saunders Company.

Piaget, J. (1928). *Judgement and Reasoning in the Child*. New York: Harcourt Brace.

Pless, I. B. & Pinkerton, P. (1975). *Chronic childhood disorder: promoting patterns of adjustment*. Chicago, IL: Year Book Medical Publishers.

Pless, I. B. & Satterwhite, B. B. (1975). Chronic Illness. In R. J. Haggerty, K. J. Roghmann and I. B. Pless (Eds.), *Child Health and the Community* New York: John Wiley and Sons.

Power, M., Harper, A. & Bullinger, M. (1999). The World Health Organization WHO-QOL-100: Tests of the universality of Quality of Life in 15 different cultural groups world-wide. *Health Psychology*, *18* (5), 495-505.

Raczek, A. E., Ware, J. E., Bjorner, J. B., Gandek, B., Haley, S. M., Aaronson, N. K., Apolone, G., Bech, P., Brazier, J. E., Bullinger, M. & Sullivan, M. (1998). Comparison of Rasch and summated rating scales constructed from SF-36 physical functioning items in seven countries: results from the IQOLA Project. International Quality of Life Assessment. *Journal of Clinical Epidemiology*, *51* (11), 1203-1214.

Ravens-Sieberer, U. & Bullinger, M. (1998). Assessing health-related quality of life in chronically ill children with the German KINDL: first psychometric and content analytical results. *Quality of Life Research*, *7* (5), 399-407.

Ravens-Sieberer, U., Redegeld, M. & Bullinger, M. (2001a). Quality of life after inpatient rehabilitation in children with obesity. *International journal of obesity and related metabolic disorders*, *25* (Suppl. 1), 63-65.

Ravens-Sieberer, U., Gosch, A., Abel, T., Auquier, P., Bellach, B. M., Bruil, J., Dur, W., Power, M. & Rajmil, L. (2001b). Quality of life in children and adolescents: a European public health perspective. *Sozial- und Präventivmedizin*, *46* (5), 294-302.

Reid, G. J., Gilbert, C. A. & McGrath, P. J. (1998). The Pain Coping Questionnaire: preliminary validation. *Pain*, *76* (1-2), 83-96.

Richards, J. M. & Hemstreet, M. P. (1994). Measures of life quality, role performance, and functional status in asthma research. *American Journal of Respiratory and Critical Care Medicine*, *16,* 31-39.

Richardson, G., Griffiths, A. M., Miller, V. & Thomas, A. G. (2001). Quality of life in inflammatory bowel disease: a cross-cultural comparison of English and Canadian children. *Journal of Pediatric Gastroenterology and Nutrition*, *32* (5), 573-578.

Robinson, M. E., Riley, J. L., 3rd, Myers, C. D., Sadler, I. J., Kvaal, S. A., Geisser, M. E. & Keefe, F. J. (1997). The Coping Strategies Questionnaire: A large sample, item level factor analysis. *Clinical Journal of Pain*, *13* (1), 43-49.

Rose, M. A. & Clark-Alexander, B. (1998). Caregivers of children with HIV/AIDS: quality of life and coping styles. *Journal of the Association of Nurses in AIDS Care*, *9* (1), 58-65.

Rose, M. A. & Clark-Alexander, B. (1999). Coping styles of caregivers of children with HIV/AIDS: Implications for health professionals. *AIDS Patient Care*, *13* (6), 335-342.

Rothbaum, F., Weisz, J. R. & Snyder, S. S. (1982). Changing the world and changing the self: a two-process model of perceived control. *Journal of Personality and Social Psychology*, *42,* 5-37.

Rudolph, K. D., Dennig, M. D. & Weisz, J. R. (1995). Determinants and consequences of children's coping in the medical setting: conceptualisation, review, and critique. *Psychological Bulletin*, *118* (3), 328-357.

Ruperto, N., Ravelli, A., Pistorio, A., Malattia, C., Cavuto, S., Gado-West, L., Tortorelli, A., Landgraf, J. M., Singh, G. & Martini, A. (2001). Cross-cultural adaptation and psychometric evaluation of the Childhood Health Assessment Questionnaire (CHAQ) and the Child Health Questionnaire (CHQ) in 32 countries. Review of the general methodology. *Clinical and Experimental Rheumatology*, *19* (4 Suppl. 23), 1-9.

Ryan-Wenger, N. M. (1990). Development and psychometric properties of the Schoolagers' Coping Strategies Inventory. *Nursing Research*, *39,* 344-349.

Ryan-Wenger, N. M. (1992). A taxonomy of children's coping strategies. *American Orthopsychiatric Association*, *62* (2), 256-263.

Sabaz, M., Cairns, D. R., Lawson, J. A., Bleasel, A. F. & Bye, A. M. (2001). The health-related quality of life of children with refractory epilepsy: a comparison of those with and without intellectual disability. *Epilepsia*, *42* (5), 621-628.

Sandler, I. N., Tein, J. Y. & West, S. G. (1994). Coping, stress, and the psychological symptoms of children of divorce: a cross-sectional and longitudinal study. *Child Development*, *65* (6), 1744-1763.

Sartorius, N. & Kuyken, W. (1994). Translation of health status instruments. In J. Orley and W. Kuyken (Eds.), *Quality of Life Assessment: International Perspectives* (3-19). Berlin: Springer.

Sawyer, M. G., Spurrier, N., Whaites, L., Kennedy, D., Martin, A. J. & Baghurst, P. (2000). The relationship between asthma severity, family functioning and the health-related quality of life of children with asthma. *Quality of Life Research*, *9* (10), 1105-1115.

Sawyer, M. G., Spurrier, N., Kennedy, D. & Martin, J. (2001). The relationship between the quality of life of children with asthma and family functioning. *Journal of Asthma*, *38* (3), 279-284.

Schalock, R. L. (1996). *Quality of Life Volume I: Conceptualisation and measurement*. Washington, DC: American Association on Mental Retardation.

Schanberg, L. E., Anthony, K. K., Gil, K. M., Lefebvre, J. C., Kredich, D. W. & Macharoni, L. M. (2001). Family pain history predicts child health status in children with chronic rheumatic disease. *Pediatrics*, *108* (3), 47.

Schmidt, A. & Lehmkuhl, G. (1994). The illness concept of children-review of the literature. *Fortschritte der Neurologie, Psychiatrie*, *62* (2), 50-65.

Schor, E. L. (1998). Children's health and the assessment of health-related quality of life. In D. Drotar (Eds.), *Measuring health -related quality of life in children and adolescents* (25-39). New Jersey: Lawrence Hillbaum.

Schwartz, C. E. & Meir, R. S. (1999). Helping others helps oneself: response shift effects in peer support. *Social Science & Medicine*, *48*, 1563-1575.

Schwartz, C. E. & Sprangers, M. (1999). Methodological approaches for assessing response shift in longitudinal health-related quality of life research. *Social Science & Medicine*, *48*, 1531-1548.

Seiffge-Krenke, I. (1992). Coping behaviour of Finnish adolescents: remarks on a cross-cultural comparison. *Scandinavian Journal of Psychology*, *33* (4), 301-314.

Selye, H. (1956). *The stress of life*. New York: Mc Graw-Hill.

Selye, H. (1979). *Stress - mein Leben*. Zürich: Kindler.

Simeoni, M. C., Auquier, P., Antoniotti, S., Sapin, C. & San Marco, J. L. (2000). Validation of a French health-related quality of life instrument for adolescents: the VSP-A. *Quality of Life Research*, *9* (4), 393-403.

Sjobu, L. (1994). Parents of children with cerebral palsy in Nordland (county in the north of Norway); factors connected to their quality of life and coping of the circumstances around the handicapped child. *Arctic Medical Research*, *53* (Suppl. 1), 1-30.

Skevington, S. M. (2002). Advancing cross-cultural research on quality of life: Observations drawn from the WHOQOL development. *Quality of Life Research*, *11*, 135-144.

Skinner, E. A. & Wellborn, J. G. (1994). Coping during childhood and adolescence: A motivational perspective. In D. Featherman, R. Lerner and M. Perlmutter (Eds.), *Life-span development and behavior* (91-133). Hillsdale, NJ: Erlbaum.

Skinner, E. A. (1995). *Perceived control, motivation, and coping*. Thousand Oaks, CA: Sage.

Sly, R. M. (2000). Allergic Disorders. In R. E. Behrman, R. M. Kliegman & H. B. Jenson

(Eds.), *Textbook of Pediatrics* (1793-1865). Philadelphia: W. B. Saunders Company.

Smith, K. E., Ackerson, J. D., Blotcky, A. D. (1990). Reducing distress during invasive medical procedures: relating behavioral interventions to preferred coping style in pediatric cancer patients. *Journal of Pediatric Psychology*, *14* (3),405-19.

Snyder, C. R. & Dinoff, B. L. (1999). Coping: Where have you been? In C. R. Snyder (Ed.), *Coping: The psychology of what works* (3-19). Oxford: University Press.

Sperling, M. A. (2000). Diabetes mellitus. In R. E. Behrman, R. M. Kliegman and H. B. Jenson (Eds.), *Textbook of Pediatrics* (1767-1791). Philadelphia: W. B. Saunders Company.

Spieth, L. E. & Harris, C. V. (1996). Assessment of health-related quality of life in children and adolescents: an integrative review. *Journal of Pediatric Psychology*, *21* (2), 175-193.

Spilker, B. (1990). Introduction. Quality of life assessments in clinical trials. In B. Spilker (Eds.), *Quality of life and pharmacoeconomics in clinical trials* (3-9). New York: Raven.

Spirito, A., Stark, L. J. & Williams, C. (1988). Development of a brief coping checklist for use with pediatric populations. *Journal of Pediatric Psychology*, *13,* 555-574.

Spirito, A., Stark, L. J., Gil, K. M. & Tyc, V. L. (1995). Coping with everyday and disease-related stressors by chronically ill children and adolescents. *Journal of the American Academy of Child & Adolescent Psychiatry*, *34* (3), 283-290.

Sprangers, M. & Schwartz, C. E. (1999). Integrating response shift into health-related quality of life research: a theoretical model. *Social Science & Medicine*, *48*, 1507-1515.

Staab, D., Wenninger, K., Gebert, N., Rupprath, K., Bisson, S., Trettin, M., Paul, K. D., Keller, K. M. & Wahn, U. (1998). Quality of life in patients with cystic fibrosis and their parents: What is important besides disease severity? *Thorax*, *53* (9), 727-731.

Starfield, B., Riley, A. W., Green, B. F., Ensminger, M. E., Ryan, S. A., Kelleher, K., Kim-Harris, S., Johnston, D. & Vogel, K. (1995). The adolescent child health and illness profile. A population-based measure of health. *Medical Care*, *33* (5), 553-566.

Stein, R. E. K. & Jessop, D. J. (1982). A noncategorial approach to chronic childhood illness. *Public Health Reports*, *97,* 354-362.

Stein, R. E. K. & Jessop, D. J. (1984). Relationship between health status and psychological adjustment among children with chronic conditions. *Pediatrics*, *73,* 169-174.

Stein, R. E. K. & Jessop, D. J. (1990). Functional Status II (R). A measure of child health status. *Medical Care*, *28,* 1041-1055.

Stein, R. E. K., Bauman, L. J., Westbrook, L. E., Coupey, S. M. and Ireys, H. T. (1993). Framework for identifying children who have chronic conditions: The case for a new definition. *Journal of Pediatrics*, *122* 342-347.

Stein, R. E. K., Westbrook, L. E. & Bauman, L. J. (1997). The questionnaire for identifying children with chronic conditions: A measure based on a noncategorical approach. *Pediatrics*, *99* (4), 513-521.

Steinhausen, H.-C. (1994). Workshop II: Psychosocial Aspects of Puberal Development - Psychosocial aspects of chronic disease in children and adolescents. *Hormone Research*, *41* (Suppl. 2), 36-41.

Strand, C. V. & Russell, A. S. (1997). WHO/ILAR Taskforce on quality of life. *Journal of Rheumatology*, *24* (8), 1630-1633.

Symonds, T., Berzon, R., Marquis, P., Rummans, T. & The Clinical Significance Consensus Meeting Group (2002). The clinical significance of quality of life results: practical considerations for specific audience. *Mayo Clinics Proceedings*, *77*, 572-583.

Tapsoba, H., Deschamps, J. P. & Leclercq, M. H. (2000). Factor analytic study of two questionnaires measuring oral health-related quality of life among children and adults in New Zealand, Germany and Poland. *Quality of Life Research*, *9* (5), 559-569.

The European Group for Quality of Life and Health Measurement (1992). *European Guide to the Nottingham Health Profile*. Montpellier: ESCUBASE.

The WHOQOL Group (1993). Study protocol for the World Health Organization project to develop a Quality of Life assessment instrument (WHOQOL). *Quality of Life Research*, *2* (2), 153-159.

The WHOQOL Group (1995). The World Health Organization Quality of Life assessment (WHOQOL): position paper from the World Health Organization. *Social Science & Medicine*, *41* (10), 1403-1409.

The WHOQOL Group (1998a). The World Health Organization Quality of Life Assessment (WHOQOL): development and general psychometric properties. *Social Science & Medicine*, *46* (12), 1569-1585.

The WHOQOL Group (1998b). Development of the World Health Organization WHOQOL-BREF quality of life assessment. The WHOQOL Group. *Psychology and Medicine*, *28* (3), 551-558.

Thompson, R. J. J., Gil, K. M., Burbach, D. J., Keith, B. R. & Kinney, T. R. (1993). Psycho-

logical adjustment of mothers of children and adolescents with sickle cell disease: the role of stress, coping methods and family functioning. *Journal of Pediatric Psychology*, *18,* 549-559.

Thompson, R. J., Gustafson, K. E., George, L. K. & Spock, A. (1994). Change over a 12-month period in the psychological adjustment of children and adolescents with cystic fibrosis. *Journal of Pediatric Psychology*, *19*, 189-203.

Thompson, R. J. & Gustafson, K. E. (1996). *Adaptation to chronic childhood illness*. Washington DC: American Psychological Association.

Varni, J. W. & Wallander, J. L. (1988). Pediatric chronic disabilities: hemophilia and spina bifida as examples. In D. Routh (Ed.), *Handbook of pediatric psychology* (190-221). New York: Guilford Press.

Varni, J. W., Katz, E. R., Colegrove, J. R., & Dolgin, M. (1995). Adjustment of children with newly diagnosed cancer. *Journal of Developmental Oncology*, *12*, 1-16.

Varni, J. W., Katz, E. R., Colegrove, J. R., & Dolgin, M. (1996a). Family functioning predictors of adjustment in children with newly diagnosed cancer. *Journal of Child Psychology and Psychiatry*, *13*, 23-38.

Varni, J. W., Waldron, S. A., Gragg, R. A., Rapoff, M. A., Bernstein, B. H., Lindsley, C. B. & Newcomb, M. D. (1996b). Development of the Waldron/ Varni pediatric pain coping inventory. *Pain*, *67* (1), 141-150.

Varni, J. W., Rode, C. A., Seid, M., Katz, E. R., Friedman-Bender, A. & Quiggins, D. J. (1999a). The Pediatric Cancer Quality of Life Inventory-32 (PCQL-32). II. Feasibility and range of measurement. *Journal of Behavioral Medicine*, *22* (4), 397-406.

Varni, J. W., Seid, M., Rode, C. A. (1999b). The PedsQL: measurement model for the pediatric quality of life inventory. *Medical Care*, *37* (2), 126-139.

Vogels, T., Verrips, G. H., Verloove-Vanhorick, S. P., Fekkes, M., Kamphuis, R. P., Koopman, H. M., Theunissen, N. C. & Wit, J. M. (1998). Measuring health-related quality of life in children: the development of the TACQOL parent form. *Quality of Life Research*, *7* (5), 457-465.

Walker, L. S., Smith, C. A., Garber, J. & Van Slyke, D. A. (1997). Development and validation of the Pain Response Inventory for Children. *Psychological Assessent*, *9,* 392-405.

Wallander, J. L. & Varni, J. W. (1992). Adjustment in children with a chronic physical disorder: Programmatic research on a disability-stress-coping model. In A. M. La Greca, L. J. Siegel, J. L. Wallander and C. E. Walker (Eds.), *Stress and coping with pediatric conditions* (279-298). New York: Guilford Press.

Wallander, J. L. & Thompson, R. J., Jr. (1995). Psychosocial adjustment of children with chronic physical conditions. In M. C. Robert (Eds.), *Handbook of pediatric psychology* (124-141). New York: Guilford Press.

Wallander, J. L. & Varni, J. W. (1995). Appraisal, coping, and adjustment in adolescents with a physical disorder. In J. L. Wallander and L. J. Siegel (Eds.), *Adolescent health problems: behavioural perspectives.* New York: Guilford Press.

Wallander, J. L. & Varni, J. W. (1998). Effects of pediatric chronic physical disorders on child and family adjustment. *Journal of Child Psychology and Psychiatry*, *39* (1), 29-46.

Wallander, J. L., Schmitt, M. & Koot, H. M. (2001). Quality of life measurement in children and adolescents: issues, instruments, and applications. *Journal of Clinical Psychology*, *57* (4), 571-585.

Ware, J. E., Jr. (1984a). Conceptualising disease impact and treatment outcomes. *Cancer Nursing*, (15), 2316-2323.

Ware, J. E., Jr. (1984b). Methodology in behavioral and psychosocial cancer research. Conceptualising disease impact and treatment outcomes. *Cancer*, *53* (10 Suppl.), 2316-2326.

Ware, J. E., Jr., Keller, S. D., Gandek, B., Brazier, J. E. & Sullivan, M. (1995). Evaluating translations of health status questionnaires. Methods from the IQOLA project. International Quality of Life Assessment. *International Journal of Technology assessment in health care*, *11* (3), 525-551.

Ware, J. E., Jr., Kosinski, M., Gandek, B., Aaronson, N. K., Apolone, G., Bech, P., Brazier, J., Bullinger, M., Kaasa, S., Leplege, A., Prieto, L. & Sullivan, M. (1998). The factor structure of the SF-36 Health Survey in 10 countries: Results from the IQOLA Project. International Quality of Life Assessment. *Journal of Clinical Epidemiology*, *51* (11), 1159-1165.

Watson, D. C. & Sinha, B. K. (1998). Gender, age, and cultural differences in the Defence Style Questionnaire-40. *Journal of Clinical Psychology*, *54* (1), 67-75.

World Health Organization (1948). *Constitution of the World Health Organisation.* Geneva.

World Health Organization (1993). *Measurement of quality of life in children*. Division of mental health, World Health Organisation.

World Health Organization European Region (1999). The health for all. Policy framework for the European Region. *Health, 21*, Kopenhagen.

Zeltzer, L., Kellerman, J., & Ellenberg, L. (1980). Psychological effects of illness in adolescents: II. Impact of illness in adolescents - crucial issues and coping styles. *Journal of Pediatrics*, *97,* 132-138.

8 List of Tables

9 List of Figures

10 Appendix

A Members of the DISABKIDS Group

B Pilot Manual

C Questionnaires

C-1: Children and Adolescents 8-16 years

C-2: Take Home Questionnaire 8-16 years (coping measure)

D Additional Tables

D-1: HRQOL rotated component matrix: factor loadings

D-2: Pearson correlation matrix: facets and dimensions

D-3: Results of the cognitive debriefing for each item

Members of the DISABKIDS Group

University of Edinburgh
Prof. Dr. Mick Power
Dr. Claire Atherton
Dr. Peter Hoare

University Clinic Hamburg-Eppendorf
Prof. Dr .Monika Bullinger
Dipl.-Psych. Corinna Petersen
Dr. Silke Schmidt

Hippocratio Hospital
Dr. Athanasios Vidalis
Ass. Prof. John Tsanakas
Dr. Voula Karagianni
Dr. Elpis Hatziagorou
Pela Elefteriadou

Leiden University Medical
Dr. Hendrik M. Koopman
Dr. Rolanda Baars

Medical University of Luebeck
Dr. Ute Thyen
Dipl.-Psych. Esther Mueller Godeffroy

University Hospital Lund
Dr. John Eric Chaplin

University Hospital of Marseille
Dr. Marie-Claude Simeoni

University of Vienna
Dr. Michael Quittan
Dr. Nilouparak Hachemian
Dr. Othmar Schuhfried

PILOT TEST MANUAL

Corinna Petersen for the DISABKIDS Group

April, 2002

Table of Contents

1 Aim of the Pilot Testing

The systematic pre-testing of a questionnaire is central to planning a good field test. Much of the accuracy and interpretability of the field test results hinge on the pilot-testing step. Pre-testing is especially critical for identifying questionnaire problems. The objectives of the pilot test are twofold, firstly to analyse the content of the DISABKIDS questionnaire and secondly to simulate the field test.

Content-related objectives are:

- to find out more about the adequacy as well as relevance of the items and the need for modification,
- to reveal problems with question content (e.g. misinterpretation of individual terms),
- to define the structure of the questionnaire and
- to reduce the number of questions and revise question wording.

Methodology-related objectives are:

- to check what kind of problems might occur in the field,
- to test major steps of data collection in the field study and
- to collect data with the pilot test questionnaires.

The pilot test should ideally result in a reduction of questions without impairing the adequate coverage of the areas elicited by the focus group work. The pilot data will also be used to gather first information about reliability and validity for the draft instrument.

2 Methodology

Pilot- or pre-testing are broad terms that incorporate many different methods with different strengths and weaknesses. Three methods/ modes of administration have been chosen to check the appropriateness of the DISABKIDS pilot questionnaire and therefore the pilot test can be divided in three parts:

Part A: Filling out the questionnaire or Interview

Part B: Cognitive debriefing

Part C: Take home questionnaires

Assistants of the respective centre will conduct the pilot testing. Interviewers have to be trained and experienced and have to follow the instructions in the manual.

2.1 Sample

Children and adolescents treated in the centre as well as their parents form the sample. Possible participant should be contacted **in advance** with an introductory letter informing families and children about the study and including consent forms (appendix A). If the response rate is low, it will be recommended to phone the addressed persons. To calculate the response rates, contacts per phone/ mail have to be recorded on a sheet (appendix B). Criteria for inclusion in the pilot-test are:

- available consent form,
- child / adolescent fits age requirements of the study (4-17 years of age)

Per participating centre and per condition **36 or 18 families** (depending on the number of conditions the centre will investigate) should be included in the study and therefore an adequate number of patients should be identified and contacted (representing both gender and 3 age groups). One of the two conditions studied must be asthma in order to be able to compare the questionnaire across the countries. Table 1 depicts the composition of the sample.

Table 1: Children/ Adolescent Sample

	Condition I (Asthma)		Condition II	
	female	male	female	male
Age Group I: 4-7	6 (3)	6 (3)	6 (3)	6 (3)
Age Group II: 8-12	6 (3)	6 (3)	6 (3)	6 (3)
Age Group III: 13-16	6 (3)	6 (3)	6 (3)	6 (3)

Each participating centre decided to test the following conditions with the sample size stated in table 2. It is much appreciated, if a centre includes a third condition (if the respective items were translated), but a large sample size of two conditions should be prioritised.

Table 2: Number of patients per conditions to be included in the pilot test

Centre	Asthma	Arthritis	Dermatitis	Diabetes	Epilepsy	Cystic Fibrosis	Cerebral Palsy	Obesity*	Total
UK	36				18		18		72
France	36		36		18				90
Greece	36					18	18	36	108
Austria	18			36		18		36	108
GER (LUE)	36	18					18		72
GER (HH)	-	18	36						54
Sweden	36			18	36				90
NL	36	36		18					90
Total	234	72	72	72	72	36	54	72	684

* Obesity will be investigated by Greece and Austria as an ancillary project. The centres will be responsible for the conduction and analysis of this extra module.

2.2 Instruments

The present pilot-test questionnaire is available for children and their parents. In the following paragraphs the content of the questionnaires will be shortly described.

2.2.1 Questionnaire Children

The children's questionnaire consists of the DISABKIDS items generated so far which are divided into a chronic generic and a disease-specific part. The chronic generic part contains items that are applicable to children with any chronic health condition. Altogether two age versions for children are available: a short version for children of the age group I with smiley items and a long version for the other two age

groups. At this stage the same questionnaire will be applied for the age-groups II and III. This procedure will give information about age-specific dimensions of health-related quality of life. Respondents should mark the most appropriate box on a five-point Likert scale. If a question is **not applicable** the child should tick the box in the respective column.

In each version, ten items have been added to have at hand anchor items as a frame of reference. The items were chosen from the item bank of John Ware and are items from the KINDL questionnaire and the Child Health Questionnaire. A global health question, open questions and a minimal number of socio economic status variables suggested by the KIDSCREEN group will be assessed as well. The English versions of the questionnaires for children can be found in appendix C and D.

2.2.2 Questionnaire Parents

For parents two questionnaire versions have been developed, one for parents of younger children and one for parents of older children. The different parts of the parents' questionnaire are listed in table 3. The parents get the same items as their children but have additionally the possibility to comment on every item (a specific columne has been added). The English version of the questionnaires for parents can be found in appendix E, F.

Table 3: Questionnaire Parents

Part	Module	Description
A	Clinical Variables	A generic and a disease specific part
B	Screener	Children with special health care needs (CSHCN)
C	DISABKIDS	Chronic generic module Disease-specific module
D	Health Status	SF2R
E	Socio economic status	Minimal number of socio economic status variables

2.2.3 Medical Documentation

The medical documentation has to be filled out by a physician, who knows the child. As former studies showed, physicians might forget to fill out the medical documentation. Please explain to them the aim of the study! This questionnaire is just available in English and can of course be translated in other languages by the centre itself (appendix H).

2.3 Preparation

For the pilot-test please make sure that you have two separate rooms with a chair and a table available. You will need paper, pencil and a small gift for the child to be given after completion of the task (money for a gift has been included in your budget). In addition, you will need the respective questionnaire and documents which are listed in checklist I (appendix G). Please use this checklist to make sure that you haven't forgotten anything. Something to drink and some snacks should be available for the pilot test. Next, please record all the patients participating in the study on a list of participants by address and identification number (appendix A). Ensure that the boxes at the top of each questionnaire are filled out in the following way:

Coding of each questionnaire

- Box 1: Country code number (GER/HH:1, GER/LUE=2, NL=3, UK=4, F=5, GR=6, SW= 7, AUS=8, I=9)
- Box 2-4: family number of the participant which will be assigned consecutively by each centre from 001.
    - It is absolutely necessary that t**he child and the parents get the same family number** in terms of comparing the results afterwards!
- Box 5: Respondent code of the person being questioned (1= child, 2= mother, 3= father, 4= stepmother, 5= stepfather, 6= other caregiver).
- Box 6: Group (1= asthma, 2= arthritis, 3= Dermatitis, 4= diabetes, 5= cerebral palsy, 6= cystic fibrosis, 7= epilepsy, 8= obesity).

2.4 Procedure

The following paragraphs describe subsequently the procedure to complete the pilot testing.

Part A: Filling out the questionnaire

The pilot test should preferably be conducted in the clinics but interviews at home are an alternative possibility.

Don't forget to ask the physician to complete the medical documentation (appendix H). When the parent and the children arrive, please inform them about the aim of the study and the procedure. Parent and children should be interviewed or fill out the questionnaires in different rooms (if the child feels comfortable with that).

Children from age group I will be questioned in a one-on-one interview with smiley questions. In order to help the child please prepare a "ruler": On this ruler the answer categories of the questionnaire should be affixed so that the child just has to point his/ her finger on an answer category when being questioned.

Children of the age- group II and III fill out the questionnaire on their own. Additionally, the father or the mother will be asked to fill out the parents' questionnaire.

Example for an introduction:

Hello....(child's name), I'm....(interviewer's name). Thank you for coming to help us with our study. We've developed a questionnaire especially for children and teenagers with xxx. The questionnaire contains questions about symptoms, treatment as well as about friends and family and I would like to know what you think of the questionnaire. First of all we like you to fill out the questionnaire. Please take as much time as you need. There's no need to hurry in filling out the questionnaire, but don't ponder too long. Important is your personal experience. There are no right and no wrong answers. On the first page of the questionnaire you find some instructions how to fill out the questionnaire. If you have any questions or need assistance, I'll be happy to help you. When you've finished filling out the questionnaire, I'd like to find out what you think about our questions, whether there were any difficult questions if something important has been forgotten.

Take the children and the parent into the rooms, so that they can fill out the questionnaire without any interruptions. Stay in the room and assist them if they need help. Please check, if the questionnaires have been fully completed and go quickly through the questionnaire and check if items have been marked as not applicable. Please clarify if this was just a misunderstanding or really the case.

Part B: Cognitive Debriefing

After a short break a "cognitive debriefing" will be performed with the children **aged 8-17** in order to assess the clarity, cultural relevance and appropriateness of wording of the questionnaire. Cognitive debriefing techniques are increasingly being used in questionnaire development to assess respondents' comprehension. The aim of this cognitive debriefing is to determine whether concepts and questions are understood by respondents in the same way that it is intended and to record proposed solutions in order to clarify questions which are problematic. The results will be applied to revise the existing questions. The cognitive debriefing will be divided in two parts, namely a general and a specific part.

General Part of the Cognitive Debriefing

After filling out the questionnaire, children/ adolescents should openly state their opinion about the questionnaire and say in general if they understood the questions or something was funny. The focus here is the **general impression** of the questionnaire. Please write the answers on the "general impression sheet" (appendix I). Enquire about the child's general feeling about the questionnaire and follow the questions on the document. Please find out if the questionnaire is:

- globally clear, easy to understand, easy to answer to?
- adapted to the condition?
- and are the instructions/ answer categories clear?

Parents received the general impression sheet already with the questionnaire in Part A and fill it out by themselves.

Specific Part of the Cognitive Debriefing

For children of the age group I the specific part will be very short. The interviewer should read out loud each smiley question and ask if the child likes the question. Give the child some time and note down the answers. After that the small children should state how much they like each question by pointing at a smiley.

The children of the age groups II and III will be queried to assess comprehensibility, importance and acceptance of the questionnaire. Because of time exposure and burden for the child/ adolescents, we will not be able to go through the questionnaire and check each question. Therefore, just a subset of items will be applied to every child. The idea is to perform the cognitive debriefing per facets/ disease-specific module, i.e. subset, and permute these subsets, so that the first child you test gets subset A, the second child subset B and so on. The subsets differ in their number of items, but this classification seems to be the best solution for different subsets. The following item subsets should be tested in the cognitive debriefing as a whole.

- Subset A: Chronic generic items: medical, physical and overall health perception domain (n= 30)
- Subset B: Chronic generic items: Psychological domain (n= 38)
- Subset C: Chronic generic items: Social domain (n= 51)
- Subset D: Disease-specific (n=25 to 48, 1= asthma, 2= arthritis, 3= dermatitis, 4= diabetes, 5= cerebral palsy, 6= cystic fibrosis, 7= epilepsy, (8= obesity)

If 36 children per condition will be tested in the pilot study, 24 children per condition should be between 8-16 (i.e. in age group II or III), so at least 4 children per subset and per condition should be tested in order to gather appropriate information (please remember: just older kids will be included in this part, see table 4). The number of children for Subset D (disease-specific part) should be 12 in order to collect enough information about this item pool. Please remind the child/ adolescent that we are not interested in his or her responses, but the formulation of the questions.

Table 4: Children/ Adolescent Sample for the cognitive debriefing (n=24)

	Condition I	
	8-12	13-16
Subset A	2	2
Subset B	2	2
Subset C	2	2
Subset D	6	6

Go then through the questions, using the specially prepared cognitive debriefing sheet (appendix I) and check whether:

- the child/ adolescents finds the question important in connection with his/her illness
- the question was difficult to understand or answer to. If so, why?
- the response choices are clear and consistent with the question
- the subject would ask the question in another way
- the underlying concept is interpreted correctly i.e. there are no ambiguous formulation that would make more than one interpretation possible.
  Therefore: Please let the child/ adolescent think aloud and write down the thought in keywords.

The comments should be recorded in the respective cognitive debriefing, where the questions have been listed in a table. If you e.g. chose subset A for the cognitive debriefing, please use the cognitive debriefing form with an A on the front page. One form should be used per participant. Once all participants are interviewed, subjects' comments should be summarized in another form that should be returned to the Hamburg Coordination Centre (appendix K). On the summary sheet, the interviewer has the opportunity to describe his/ her perception, prevalence and proposed solutions to a problem (e.g. if respondents refused to answer specific questions).

Part C: Take Home Questionnaires

After filling out the questionnaires and performing the cognitive debriefing in the centre, the parent receives two take-home-questionnaires (one for the child, just for the age- groups II and III, and one for the parents, all age- groups, see appendices L

and M) in a replied paid envelope to fill out at home. The questionnaires within this envelope are again in two separate envelopes. **Please make sure that the boxes in the heading of each questionnaire have been filled out!** This is absolutely necessary in order to identify and compare the questionnaires with the other parts of the pilot test.

The children/ adolescents' questionnaire contains a coping module derived from the focus group work package. **The parents' questionnaire** contains health care needs items. Please shortly explain the filling out to the parents and children/ adolescents. The questionnaires should be sent back to the respective centre within two weeks. Finally, don't forget to give the child a small thank you present. If you don't receive the take home questionnaire within 2 weeks, please phone the parents and remind them politely. Table 5 depicts the time flow of the pilot testing which will presumably take about 90-120 minutes.

Table 5: Time flow of the pilot test

		Minutes approx.
	Arrival and Introduction	30-40
Part A	Collection of consent form Filling out the questionnaire or interview	
	Break	10
Part B	Cognitive debriefing -general impression -structured questions	50
Part C	Handing out a present and explaining take home questionnaires	10
If necessary: Remind the parents to send back the take home questionnaire per phone!		

2.5 Analysis of the Results

All data of the pilot test have to be entered into the respective data files (all SPSS files) provided by the Hamburg coordination centre. SPSS data files will be provided, including variable name, labels, and value labels. Data files (n= 11) will be provided separately for:

- Questionnaire children I / Questionnaire parents I / General impression sheet children I / General impression sheet parents I
- Questionnaire children II / III / Questionnaire parents II/ III/ General impres-

sion sheet children II / III / General impression sheet parents II/ III

- Cognitive debriefing subset A
- Cognitive debriefing subset B
- Cognitive debriefing subset C
- Cognitive debriefing subset D
- 1= asthma, 2= arthritis, 3= dermatitis, 4= diabetes, 5= cerebral palsy, 6= cystic fibrosis, 7= epilepsy
- Take home children/ Take home parents
- Medical Documentation
- Interviewers Summary

No imputations for missing values should be made during data entry. Please send everything mentioned on checklist II (appendix N) to the HH co-ordination centre until **the end of June 2002.**

The files will be matched with the gross sample by the unique respondent identification number as key variable. The data will then be analysed from all centres collectively to examine the structure for the instrument and possibly reduce the number of items. On the basis of these data, the DISABKIDS questionnaire will be further refined. The strategy for the selection of questions will be discussed in detail with all participants.

Correspondence

Dipl.-Psych. Corinna Petersen
Department of Medical Psychology
University Hospital Hamburg-Eppendorf
Martinistr. 52, S 35
20246 Hamburg/ Germany
Telephone:+49/40/42803-8845
Fax: +49/40/42803-4940
E-mail: copeters@uke.uni-hamburg.de

Centre No.: |__|__| **Family Code:**|__|__|__| **Respondent:** |__| **Group:** |__|

Questionnaire for Children and Adolescents

Hi,

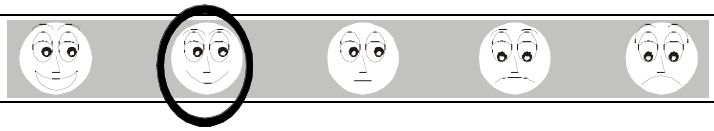
We would like to ask you some questions about how you have been feeling during the past four weeks. These questions ask about some problems that children like you might have. We would like you to answer all the questions below. Please

- ⇨ think back over the **past four weeks** when answering the questions and
- ⇨ choose the answer that fits you best and tick the appropriate box.

If you play with your friends 'very often' you would tick the box as shown in this example:

For example:	**never**	**seldom**	**quite often**	**very often**	**always**
Do you play with your friends?	☐	☐	☐	☒	☐

If you like ice-cream you would circle the face that fits best:

... Do you like ice-cream?	

There are no right or wrong answers. It's what you think that matters.

Date of completion: __ / __ / __ (day / month / year)

A) Some questions about yourself

1.	Are you a girl or a boy?	☐ girl ☐ boy
2.	How old are you?	☐☐ years
3.	What is your date of birth?	☐☐. ☐☐. ☐☐☐☐ day month year
4.	Do you have sisters or brothers? If yes, how many?	☐☐ brothers ☐☐ sisters
5.	How many years have you been at school (without pre-school or kindergarten)?	☐☐ years
6.	What class/grade are you in?	☐☐ class/ grade
7.	What kind of school do you attend?_____	

B) We would like to ask you about your feelings during the past four weeks

	never	seldom	sometimes	often	all the time
1. ... During the past 4 weeks I had fun and laughed a lot.	❑	❑	❑	❑	❑
2. ... During the past 4 weeks I felt scared or unsure of myself	❑	❑	❑	❑	❑
3. ... During the past 4 weeks I felt on top of the world	❑	❑	❑	❑	❑
4. ... During the past 4 weeks I felt pleased with myself	❑	❑	❑	❑	❑
5. ... During the past 4 weeks I felt fine at home	❑	❑	❑	❑	❑

	never	seldom	sometimes	often	all the time
6. ... During the past 4 weeks, how much of the time did you: feel sad?	❑	❑	❑	❑	❑
7. ... During the past 4 weeks, how much of the time did you: feel afraid or scared?	❑	❑	❑	❑	❑
8. ... During the past 4 weeks, how much of the time did you: worry about things?	❑	❑	❑	❑	❑
9. ... During the past 4 weeks, how much of the time did you: feel lonely?	❑	❑	❑	❑	❑
10. ... During the past 4 weeks, how much of the time did you: feel unhappy?	❑	❑	❑	❑	❑
11. ... During the past 4 weeks, how much of the time did you: feel happy?	❑	❑	❑	❑	❑
12. ... During the past 4 weeks, how much of the time did you: feel cheerful?	❑	❑	❑	❑	❑
13. ... During the past 4 weeks, how much of the time did you: enjoy the things you do?	❑	❑	❑	❑	❑
14. ... During the past 4 weeks, how much of the time did you: have fun?	❑	❑	❑	❑	❑
15. ... During the past 4 weeks, how much of the time did you: like yourself?	❑	❑	❑	❑	❑

C) Now we would like to know how you think about the future…

		never	seldom	quite often	very often	always	not applicable
1.	... Do you have fears about the future because of your condition?	❑	❑	❑	❑	❑	❑
2.	... Are you confident about your future?	❑	❑	❑	❑	❑	❑
3.	... Do you wish your illness would go away?	❑	❑	❑	❑	❑	❑
4.	... Do you feel that you will get better?	❑	❑	❑	❑	❑	❑
5.	.. When I grow up I will be						

		never	seldom	quite often	very often	always	not applicable
6.	... Do you feel lonely because of your condition?	❑	❑	❑	❑	❑	❑
7.	... Do you enjoy your life?	❑	❑	❑	❑	❑	❑
8.	... Do you feel under pressure because of your condition?	❑	❑	❑	❑	❑	❑
9.	... Does your condition get you down?	❑	❑	❑	❑	❑	❑
10.	... Does your condition restrict your life?	❑	❑	❑	❑	❑	❑
11.	... Do you forget your condition when you do certain things (e.g. when meeting friends)?	❑	❑	❑	❑	❑	❑
12.	... Do you have less free time because of your condition?	❑	❑	❑	❑	❑	❑
13.	... Does it bother you that your life has to be planned?	❑	❑	❑	❑	❑	❑
14.	... Are you able to do everything you want to do even though you are ill?	❑	❑	❑	❑	❑	❑
15.	... About the restrictions in my life I feel?						

And what do you think about yourself…

		never	seldom	quite often	very often	always	not applicable
16.	... Does your condition make you feel bad about yourself?	❑	❑	❑	❑	❑	❑
17.	... Has your illness made you feel confident about yourself?	❑	❑	❑	❑	❑	❑
18.	... Do you feel like everyone else even though you are ill?	❑	❑	❑	❑	❑	❑
19.	... Has your condition made you more grown up than other children your age?	❑	❑	❑	❑	❑	❑
20.	... Has your illness made you stand up for yourself?	❑	❑	❑	❑	❑	❑
21.	... Are you shy because of your condition?	❑	❑	❑	❑	❑	❑
22.	... About myself I feel						

And what about your feelings…

		never	seldom	quite often	very often	always	not applicable
23.	... Are you unhappy because your are ill?	❑	❑	❑	❑	❑	❑
24.	... Do you worry about your condition?	❑	❑	❑	❑	❑	❑
25.	... Do you have fun in spite of your condition?	❑	❑	❑	❑	❑	❑
26.	... Does your condition make you angry?	❑	❑	❑	❑	❑	❑
27.	... Do you hate having your condition?	❑	❑	❑	❑	❑	❑
28.	... Do you think it is unfair that you are ill?	❑	❑	❑	❑	❑	❑
29.	... Do you feel nervous because of your condition?	❑	❑	❑	❑	❑	❑
30.	... Do you feel embarrassed that you have an illness?	❑	❑	❑	❑	❑	❑
31.	... Are you ashamed that you have an illness?	❑	❑	❑	❑	❑	❑
32.	... Does your condition make you moody?	❑	❑	❑	❑	❑	❑
33.	... I feel						

		never	seldom	quite often	very often	always	not applicable
34.	... Do you hate having to depend on other people because of your condition?	❑	❑	❑	❑	❑	❑
35.	... Are you free to lead the life you want even though you are ill?	❑	❑	❑	❑	❑	❑
36.	... Do you feel independent in managing your condition?	❑	❑	❑	❑	❑	❑
37.	... Are you able to do things without your parents?	❑	❑	❑	❑	❑	❑
38.	... When I do things on my own I feel						

		never	seldom	quite often	very often	always	not applicable
39.	... Are you able to run and move as you like?	❑	❑	❑	❑	❑	❑
40.	... Are you limited in physical activities i.e. sports, biking, running?	❑	❑	❑	❑	❑	❑
41.	... Do you feel tired because of your condition?	❑	❑	❑	❑	❑	❑
42.	... About the things I can do I feel						

		never	seldom	quite often	very often	always	not applicable
43.	... Are you able to live with your condition the way it is?	❑	❑	❑	❑	❑	❑
44.	... Is your life ruled by your condition?	❑	❑	❑	❑	❑	❑
45.	... Does it bother you that you have to explain to others what you can and can't do?	❑	❑	❑	❑	❑	❑
46.	... Having my illness makes me feel						

		never	seldom	quite often	very often	always	not applicable
47.	... Do you have bad dreams or nightmares because of your condition?	❑	❑	❑	❑	❑	❑
48.	... Is it difficult to sleep because of your condition?	❑	❑	❑	❑	❑	❑
49.	... About my sleep I feel						

		never	seldom	quite often	very often	always	not applicable
50.	... Is it okay for you to live with your condition?	❑	❑	❑	❑	❑	❑
51.	... Do you feel that everyone is healthy apart from you?	❑	❑	❑	❑	❑	❑
52.	... Do you worry more than your friends about staying healthy?	❑	❑	❑	❑	❑	❑
53.	... Being ill makes me feel						

		never	seldom	quite often	very often	always	not applicable
54.	... Is it a problem for you to go to the doctor?	❑	❑	❑	❑	❑	❑
55.	... Do you have enough time for yourself in spite of the treatment?	❑	❑	❑	❑	❑	❑
56.	... About the treatment of my condition I feel						

		never	seldom	quite often	very often	always	not applicable
57.	... Are you bothered by others watching you take your medicine?	❑	❑	❑	❑	❑	❑
58.	... Are you bothered by the side effects of the medicine?	❑	❑	❑	❑	❑	❑
59.	... Has your schoolwork suffered because you have been on medication?	❑	❑	❑	❑	❑	❑
60.	... Does having to get help with medication from others bother you?	❑	❑	❑	❑	❑	❑
61.	... Are you worried that you will forget your medicine?	❑	❑	❑	❑	❑	❑
62.	... Is it annoying for you to have to remember your medication?	❑	❑	❑	❑	❑	❑
63.	... Are you worried about your medication?	❑	❑	❑	❑	❑	❑
64.	... Do you accept that you need medication?	❑	❑	❑	❑	❑	❑
65.	... Does taking medication bother you?	❑	❑	❑	❑	❑	❑
66.	... Do you hate taking your medicine?	❑	❑	❑	❑	❑	❑
67.	... Does taking medication disrupt everyday life?	❑	❑	❑	❑	❑	❑
68.	... Taking medicine makes me feel						

		never	seldom	quite often	very often	always	not applicable
69.	... Do your teachers behave differently towards you than towards others?	❑	❑	❑	❑	❑	❑
70.	... Are your teachers understanding your condition?	❑	❑	❑	❑	❑	❑
71.	... Do you have problems concentrating at school because of your illness?	❑	❑	❑	❑	❑	❑
72.	... Do you have difficulties with keeping up with the course?	❑	❑	❑	❑	❑	❑
73.	... About school I feel						

		never	seldom	quite often	very often	always	not applicable
74.	... Are your friends protective of you?	❑	❑	❑	❑	❑	❑
75.	... Are your friends supportive?	❑	❑	❑	❑	❑	❑
76.	... Do your friends accept you the way you are?	❑	❑	❑	❑	❑	❑
77.	... Are others considerate to you?	❑	❑	❑	❑	❑	❑
78.	... Do other kids understand your illness?	❑	❑	❑	❑	❑	❑
79.	... Others make me feel						

		never	seldom	quite often	very often	always	not applicable
80.	... Do you feel that others have something against you?	❑	❑	❑	❑	❑	❑
81.	... Do you think that others stare at you?	❑	❑	❑	❑	❑	❑
82.	... Do you like it when people look at you?	❑	❑	❑	❑	❑	❑
83.	... Are you the target of jokes?	❑	❑	❑	❑	❑	❑
84.	... Are you upset by other children teasing you?	❑	❑	❑	❑	❑	❑
85.	... Are you bothered by other people talking about you?	❑	❑	❑	❑	❑	❑
86.	... Do you feel excluded?	❑	❑	❑	❑	❑	❑
87.	... Other people treat me						

		never	seldom	quite often	very often	always	not applicable
88.	... Do you sleep over at a friend's house?	❑	❑	❑	❑	❑	❑
89.	... Do you go out with your friends?	❑	❑	❑	❑	❑	❑
90.	... Are you able to play with other children?	❑	❑	❑	❑	❑	❑
91.	... Do you take part in school sports despite having your condition?	❑	❑	❑	❑	❑	❑
92.	... Does your condition bother you when you play?	❑	❑	❑	❑	❑	❑
93.	... Playing with my friends makes me feel						

		never	seldom	quite often	very often	always	not applicable
94.	... Do your parents argue over things to do with your condition?	❑	❑	❑	❑	❑	❑
95.	... Does your family bother you?	❑	❑	❑	❑	❑	❑
96.	… Do your parents stop you from doing some things because of your condition?	❑	❑	❑	❑	❑	❑
97.	... Do others in your family have complaints about your condition?	❑	❑	❑	❑	❑	❑
98.	... Do you get everything you want because of your illness?	❑	❑	❑	❑	❑	❑
99.	... Do your parents support you in your treatment?	❑	❑	❑	❑	❑	❑
100.	... The help of my family makes me						

		never	seldom	quite often	very often	always	not applicable
101.	... Do you think that you can do most things as well as other children?	❑	❑	❑	❑	❑	❑
102.	... Are you one of the group?	❑	❑	❑	❑	❑	❑
103.	... Do you feel different from other children?	❑	❑	❑	❑	❑	❑
104.	... Do you feel left out of things?	❑	❑	❑	❑	❑	❑
105.	... Do you worry that you will have problems finding a friend because of your condition?	❑	❑	❑	❑	❑	❑
106.	... When I compare myself with others I feel						

		never	seldom	quite often	very often	always	not applicable
107.	... Do you get enough attention from other people?	❑	❑	❑	❑	❑	❑
108.	... Do your friends enjoy being with you?	❑	❑	❑	❑	❑	❑
109.	... Is it difficult for you to make friends because of your condition?	❑	❑	❑	❑	❑	❑
110.	... Dou you like being with other children with the same condition?	❑	❑	❑	❑	❑	❑
111.	... Do you find it easy to talk about your illness to other people?	❑	❑	❑	❑	❑	❑
112.	... Having friends makes me feel						

		never	seldom	quite often	very often	always	not applicable
113.	... Does your mother/father make too much of a fuss about you?	❑	❑	❑	❑	❑	❑
114.	... Does your condition affect the family?	❑	❑	❑	❑	❑	❑
115.	... Do you think that you are a worry to your parents because of your condition?	❑	❑	❑	❑	❑	❑
116.	... Do your parents encourage you?	❑	❑	❑	❑	❑	❑
117.	... Are your brothers/ sisters nice to you when you are ill?	❑	❑	❑	❑	❑	❑
118.	... Do your parents talk to you about your condition?	❑	❑	❑	❑	❑	❑
119.	... About my family I feel						

Centre No.: |__|__| **Family Code:**|__|__|__| **Respondent:** |__| **Group:** |__|

Take Home Questionnaire for Children and Adolescents

Hi,

We would like to ask you some questions about how you deal with your illness. We would like you to answer all the questions below. Please

⇨ choose the answer that fits you best and tick the appropriate box.

If you meet your friends 'often' you would tick the box as shown in this example:

For example:	**never**	**seldom**	**quite often**	**very often**	**always**
Do you meet your friends?	☐	☐	☐	☒	☐

There are no right or wrong answers. It's what you think that matters.

Date of completion: __ / __ / __ (day / month / year)

A) How do you deal with your illness?

Think of situations, when you have been **bothered or stressed** because of your illness. Below you find a list of things how kids may deal with their illness in these situations.

Please tell us, how often **you usually do the things or have this kind of thoughts related to your illness.**

		never	seldom	quite often	very often	always
1.	... I wish I were healthy	❑	❑	❑	❑	❑
2.	... I hope that my illness disappears	❑	❑	❑	❑	❑
3.	... I think that research will help me	❑	❑	❑	❑	❑
4	... I talk openly with others about my illness	❑	❑	❑	❑	❑
5.	... I talk with other people about my illness	❑	❑	❑	❑	❑
6.	... I learn as much as possible about my illness	❑	❑	❑	❑	❑
7.	... I read about my illness	❑	❑	❑	❑	❑
8.	... I believe that faith in God helps me	❑	❑	❑	❑	❑
9.	... I pray that my illness will go away	❑	❑	❑	❑	❑
10.	... I think positively	❑	❑	❑	❑	❑
11.	... I am optimistic about my illness	❑	❑	❑	❑	❑
12.	... I don't sit in a corner and look for pity	❑	❑	❑	❑	❑
13.	... I find it hard to carry on	❑	❑	❑	❑	❑
14.	... I keep in mind that my illness might get worse	❑	❑	❑	❑	❑
15.	... I accept my illness	❑	❑	❑	❑	❑
16.	... I have got used to my illness	❑	❑	❑	❑	❑
17.	... I try to do everything as normally as possible	❑	❑	❑	❑	❑
18.	... I want to stop having my illness	❑	❑	❑	❑	❑

		never	seldom	quite often	very often	always
19.	... I don't want to believe that I will have my illness in the future	❑	❑	❑	❑	❑
20.	... I do risky things	❑	❑	❑	❑	❑
21.	... I eat healthy food	❑	❑	❑	❑	❑
23.	... I am able to manage my illness	❑	❑	❑	❑	❑
24.	... I cope well with my illness	❑	❑	❑	❑	❑
25.	... I think that I am not alone with my illness	❑	❑	❑	❑	❑
26.	... I meet other kids who have the same illness	❑	❑	❑	❑	❑
27.	... I try to forget my illness	❑	❑	❑	❑	❑
28.	... I pretend to be all right	❑	❑	❑	❑	❑
29.	... I try to ignore my illness	❑	❑	❑	❑	❑
30.	... I don't complain about my illness	❑	❑	❑	❑	❑
31.	... I try to be calm	❑	❑	❑	❑	❑
32.	... I try to keep my feelings to myself	❑	❑	❑	❑	❑
33.	... I am frustrated	❑	❑	❑	❑	❑
34.	... I cry	❑	❑	❑	❑	❑
35.	... I am angry	❑	❑	❑	❑	❑
36.	... I think it is unfair that I am ill	❑	❑	❑	❑	❑
37.	... I am ashamed of being ill	❑	❑	❑	❑	❑
38.	... I face my situation with humour	❑	❑	❑	❑	❑
39.	... I wake up at night and think of terrible things	❑	❑	❑	❑	❑
40	... I think my illness is not so serious	❑	❑	❑	❑	❑
41.	... I don't care about my illness	❑	❑	❑	❑	❑
42.	... I take my illness easy	❑	❑	❑	❑	❑
43.	... I think my illness is no big deal	❑	❑	❑	❑	❑
44.	... I forget about my illness	❑	❑	❑	❑	❑
45.	... I don't think about my illness	❑	❑	❑	❑	❑
46.	... I think it could be worse	❑	❑	❑	❑	❑

		never	seldom	quite often	very often	always
47.	... I think there are people who suffer more than I do	❑	❑	❑	❑	❑
48.	... I tell myself that even famous people have illnesses	❑	❑	❑	❑	❑
49.	... I think of worse situations	❑	❑	❑	❑	❑

Overall, how well do you think you cope with your illness?		1	2	3	4	5
50.	1= very well 5= not well at all	❑	❑	❑	❑	❑

B) Now we would like to know what you think of the questions above. Please write down what you think and tick a box as well!

1.	What do you think about this questionnaire in general?	**Please tick a box**
		☐very good ☐good ☐not good
2.	Are the questions understandable? If not which questions:	☐easy to understand ☐sometimes difficult ☐not understandable
3.	What about the answer categories? Did you have any difficulties to use them? Please specify:	☐no difficulties ☐some difficulties ☐a lot of difficulties
4.	Would you like to change something in the questionnaire?	
5.	Would you like to add something in the questionnaire?	
6.	Were there any questions you did not want to answer? If so, why?	

Table D-1

HRQOL rotated component matrix: factor loadings

Item	Component and Factor Loading																										
	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15	16	17	18	19	20	21	22	23	24	25	26	27
16	.82																										
29	.80																										
26	.76																										
9	.74																										
1	.71																										
27	.68																										
8	.67																										
10	.66																										
32	.62																										
24	.59																										
28	.59			.49																							
30	.54																										
21	.51	.45																									
31	.49																										
44	.48		.42																								
51	-.45																										
48	.44						.41																				
18	.43													.41													
2	.42																										
3	.42																										
80		.83																									
81		.76																									
105	.45	.76																									
102		.65																									
104		.60																									
86		.58																									
108		.57																									
109		.57																									
6	-.41	-.53																									
95		.50																									
90		.41																									
40			.77																								
39			.72																								
101			.59																								
91			.52																								
41			.51																								
14			.49																								
92	.42		.44																								
66				.77																							
65				.72																							
60				.59									.41														
13				.59																							
62				.50																							
115				.40																							
35					.77																						
107					.74																						
50					.73																						
12					.56																						

*only factor loadings ≤.40 are depicted in the table

Table D-1 continued

Item no	Component and Factor Loading																										
	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15	16	17	18	19	20	21	22	23	24	25	26	27
59						.86																					
72	.40					.52																					
94							.85																				
96							.60																				
34							.42																				
74								.84																			
75								.63																			
77								.62																			
85									.79																		
55										.85																	
76										.55																	
78										.43																	
4										-.41																	
88											.85																
43											-.42																
11												.71															
97												.65															
82												-.45															
11													.86														
67													.45														
70														.86													
69															.84												
11															.51												
11																.80											
37																.65											
52																-.57											
20																	.83										
19																	.58										
83																		.81									
23	.53																	.53									
57																			.76								
54				.41															.59								
47																				.82							
71	.40																			.48							
11																					.83						
25																					.47						
11																						.84					
99																						.48					
64																							.72				
17																								.66			
98																											
11																									.78		
11																										-.46	
61																											.86

*only factor loadings ≤.40 are depicted in the table

Table D-2

Pearson correlation matrix: facets and dimensions (n=360)

Facets/ Dimensions*		1	2	3	4	5	6	7	8	9	10	11	12	13	14	15	16	17	18	19	20	21	22
1	Future																						
2	Perceived Imp.	.54																					
3	Self-Confidence	.36	.39																				
4	Emotion	.58	.68	.40																			
5	Autonomy	.24	.50	.37	.41																		
6	Limitation	.31	.55	.34	.39	.44																	
7	General Impact	.45	.63	.33	.65	.49	.53																
8	Sleep	.31	.37	28	.39	.32	.33	.36															
9	Overall Health	.36	.45	.24	.50	.40	.25	.46	.20														
10	Treatment	.31	.40	.18	.40	.30	.30	.43	.14	.38													
11	Medication	.38	.44	30	.50	.31	.30	.45	.30	.39	.35												
12	School	.27	.44	.32	.40	.40	.31	.38	.37	.34	.25	.42											
13	General Accept.	.18	.24	.37	.20	.33	.21	.25	.11	.30	.19	.24	.30										
14	Stigma	.33	.41	.28	.51	.42	.30	.45	.30	.39	.26	.33	.46	.41									
15	Social Activity	.34	.45	.38	.37	.42	.54	.38	.23	.28	.27	.28	.32	.32	.33								
16	Family Support	.17	.36	.19	.27	.22	.26	.31	.22	.26	.26	.28	.33	.20	.34	.20							
17	Differences	.41	.55	.33	.49	.35	.48	.51	.31	.38	.28	.41	.47	35	.59	.50	.35						
18	Contact	.14	.31	.29	.24	.38	.24	30	.15	.37	31	.26	.30	.50	.36	.29	.28	.43					
19	Family Function.	.27	.31	.17	.31	.08	.19	.29	.13	.24	.23	.23	.30	.21	.36	.18	.40	.35	.23				
20	Medical	.40	.48	.30	.52	.34	.33	.49	.31	.42	.48	.99	.44	.27	.36	.31	.31	.42	.30	.24			
21	Physical	.45	.69	.41	.61	.54	.86	.85	.58	.40	.40	.44	.43	.26	.45	.52	.34	.57	.31	.27	.48		
22	Psychological	.68	.86	.65	.87	.63	.54	.69	.44	.52	.42	.53	.49	.33	.52	.51	.33	.57	.35	.31	.56	.73	
23	Social	.41	.59	.44	.55	.51	.47	.57	.35	.50	.39	.46	.63	.63	.79	.59	.57	.77	.64	.57	.50	.61	.67

*= All correlations are significant at the 0.05 level (2-tailed).

Table D-3

Results of the cognitive debriefing for each item

Item No.*	Difficulty understanding "yes" (%)	Answer categories "not adequate" (%)	Relevance of the item "yes" (%)
1.	15.6	9.1	40.8
2.	30.4	10.0	53.2
3.	17.8	9.8	63.3
4.	17.8	12.5	59.2
5.	35.6	18.6	50.0
6.	22.2	9.3	49.0
7.	23.4	11.6	71.4
8.	25.0	10.6	36.2
9.	6.5	7.5	51.1
10.	31.1	7.5	54.2
11.	26.7	7.5	59.6
12.	27.3	12.2	50.0
13.	35.6	12.2	47.8
14.	18.6	12.2	63.8
15.	25.0	20.5	56.1
16.	29.5	11.1	40.0
17.	18.2	10.0	55.6
18.	22.7	10.0	62.2
19.	25.6	16.7	55.6
20.	25.0	10.0	62.2
21.	18.6	7.5	39.1
22.	18.6	10.3	62.2
23.	24.4	9.5	52.2
24.	22.7	7.3	56.5
25.	23.3	5.0	82.2
26.	20.9	10.0	55.6
27.	20.9	10.0	53.3
28.	20.9	15.4	54.5
29.	23.3	5.0	40.0
30.	20.9	7.5	55.6
31.	19.0	7.5	48.9
32.	20.9	5.0	53.3
33.	20.0	7.7	75.0
34.	24.3	8.6	55.3
35.	15.8	14.3	77.5
36.	36.8	9.7	60.5
37.	13.2	8.6	60.0
38.	16.2	14.3	72.7
39.	25.4	8.3	71.7
40.	31.0	8.8	64.9
41.	27.6	7.1	52.6
42.	25.9	19.1	75.5
43.	31.2	1.9	75.9
44.	24.6	5.7	36.2
45.	26.8	8.9	50.9
46.	30.4	19.6	80.2
47.	24.6	1.8	29.8
48.	26.8	1.8	52.6
49.	28.6	15.8	68.0
50.	31.2	5.4	68.4
51.	29.8	9.1	42.1

*= The item numbers correspond with table 8, where the items are listed.

Table D-3 continued

Item No.*	Difficulty understanding "yes" (%)	Answer categories "not adequate" (%)	Relevance of the item "yes" (%)
52.	33.3	1.9	55.4
53.	26.8	7.5	70.6
54.	31.6	0	64.3
55.	33.3	0	75.0
56.	30.4	9.3	76.0
57.	26.8	5.7	47.4
58.	29.4	6.1	42.3
59.	30.9	1.9	35.7
60.	28.3	3.8	30.4
61.	26.8	1.8	52.6
62.	27.3	7.3	53.6
63.	34.5	1.9	54.4
64.	29.1	7.3	63.2
65.	26.4	5.7	59.3
66.	25.5	8.0	57.4
67.	32.1	3.9	48.1
68.	24.5	8.2	63.3
69.	29.4	4.1	41.2
70.	27.5	2.1	66.7
71.	27.5	0	56.9
72.	27.5	4.0	41.2
73.	31.3	20.4	74.4
74.	35.3	0	62.7
75.	27.5	0	74.5
76.	23.5	6.0	82.4
77.	27.5	2.0	60.8
78.	28.0	2.0	70.6
79.	22.2	18.4	79.7
80.	36.0	0	44.0
81.	28.0	2.0	34.0
82.	32.0	2.0	52.0
83.	30.0	0	28.0
84.	30.0	0	48.0
85.	28.0	2.0	52.0
86.	26.0	2.0	28.0
87.	31.9	18.3	79.1
88.	28.0	0	74.0
89.	30.6	0	70.0
90.	34.0	2.0	84.0
91.	28.0	0	82.0
92.	28.0	0	54.0
93.	31.9	12.5	74.4
94.	28.6	0	38.8
95.	30.6	0	42.9
96.	28.6	2.1	59.2
97.	28.6	0	40.8
98.	30.6	0	36.7
99.	28.6	4.6	83.7
100.	30.4	14.6	88.1
101.	29.2	0	87.5
102.	27.1	13.0	55.3
103.	27.7	2.1	58.3

*= The item numbers correspond with table 8, where the items are listed.

Table D-3 continued

Item No.*	Difficulty understanding "yes" (%)	Answer categories "not adequate" (%)	Relevance of the item "yes" (%)
104.	30.4	4.3	53.2
105.	25.5	0	45.8
106.	32.7	17.0	64.3
107.	27.7	2.1	64.6
108.	25.5	4.4	66.7
109.	25.5	2.1	62.5
110.	25.5	4.3	51.1
111.	25.5	2.1	79.2
112.	28.9	12.8	76.7
113.	30.4	0	59.6
114.	25.5	2.1	66.7
115.	23.4	0	75.0
116.	29.8	2.1	70.2
117.	22.2	0	62.2
118.	23.9	0	74.5
119.	31.1	14.9	79.1

*= The item numbers correspond with table 8, where the items are listed.

Eidesstattliche Erklärung

nach § 3 Abs. 2 Nr. 9 der Übergangsordnung für die Promotion zum Doktor der Philosophie der Universität Hamburg vom 17. September 1969:

Hiermit erkläre ich an Eides statt, dass ich die vorliegende Arbeit selbständig und ohne fremde Hilfe verfasst sowie andere als die von mir angegebenen Quellen und Hilfsmittel nicht benutzt und die wörtlich und inhaltlich übernommenen Stellen als solche kenntlich gemacht habe.

Hamburg, den 03.03.2003

_____

Dipl.-Psych. Corinna Petersen

Eidesstattliche Erklärung

nach § 3 Abs. 2 Nr. 7 der Übergangsordnung für die Promotion zum Doktor der Philosophie der Universität Hamburg vom 17. September 1969:

Hiermit erkläre ich an Eides statt, dass ich mich nicht schon anderwärts der Doktorprüfung unterzogen oder um Zulassung zu ihr beworben habe.

Hamburg, den 04.03.2003

_____

Dipl.-Psych. Corinna Petersen