

Assessing quality of life of children with chronic health conditions and disabilities: a European approach

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Quality of life has been used as a synonym for a subject-centred or individually appraised perspective on health. Despite the increase in quality of life research in adults, quality of life in children is relatively neglected. While generic measures begin to emerge now, methods to assess the quality of life of children with chronic conditions are still in development. The design of such an assessment tool for different age groups and different levels of disabilities is the objective of a European-Union-funded study: the DISABKIDS project. In addition, it addresses the psychosocial determinants of quality of life in children with disabilities. A major aim of the project is to develop and test instruments for children and adolescents with disabilities (as well as for their families) in seven countries, to assess the impact of the chronic health conditions on quality of life and to provide a tool for systematic monitoring of the quality of care given to children with disabilities. Assessment and monitoring will allow identification of unmet health care needs and, it is hoped, ultimately, the fostering of the development of effective intervention strategies.

L'expression "qualité de vie" a été employée comme synonyme pour désigner l'appréciation subjective d'un individu sur son état de santé. La qualité de vie a fait l'objet de nombreuses études chez l'adulte mais pas chez l'enfant. Des mesures génériques d'évaluation commencent à apparaître actuellement mais les méthodes d'étude de la qualité de vie chez l'enfant présentant une pathologie chronique sont encore en cours de développement. La conception d'un outil d'évaluation pour différents groupes d'âge et différents niveaux de handicap constitue l'objectif d'une étude subventionnée par l'UE, le projet DISABKIDS. Ce projet s'intéresse en outre aux déterminants psychosociaux de la qualité de vie chez l'enfant handicapé. Un de ses objectifs majeurs est de mettre au point et tester des instruments destinés aux enfants et adolescents handicapés, ainsi qu'à leurs familles, dans sept pays, pour évaluer les répercussions des pathologies chroniques sur la qualité de vie et fournir un outil permettant un suivi systématique de la qualité des soins administrés aux enfants handicapés. Cette évaluation et ce suivi devraient permettre d'identifier des besoins encore sans réponse et de promouvoir la mise en œuvre de stratégies d'intervention efficaces.

Die "Lebensqualität" wurde als Synonym für eine personenzentrierte oder individuell eingeschätzte Gesundheitsperspektive verwendet. Zwar wurde die Lebensqualität bei Erwachsenen verstärkt untersucht, die Lebensqualität von Kindern wurde jedoch vergleichsweise vernachlässigt. Während derzeit allgemeingültige Maße aufkommen, befinden sich Methoden zur Beurteilung der Lebensqualität von Kindern mit chronischen Krankheiten noch in der Entwicklung. Die Konzeption eines Beurteilungstools für verschiedene Altersgruppen und verschiedene Grade von Behinderungen ist das Ziel einer von der EU finanzierten Studie: das DISABKIDS-Projekt. Darin geht es auch um die psychosozialen Determinanten der Lebensqualität bei Kindern mit Behinderungen. Ein wesentliches Ziel des Projekts ist die

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Entwicklung und Untersuchung von Instrumenten für Kinder und Jugendliche mit Behinderung sowie für ihre Familien in sieben Ländern, die Beurteilung des Einflusses von chronischen Krankheiten auf die Lebensqualität und die Bereitstellung eines Tools für die systematische Überwachung der Qualität der Versorgung, die Kindern mit Behinderungen zukommt. Die Beurteilung und Überwachung erlaubt die Identifikation von bisher unerfüllten Bedürfnissen nach Versorgungsmaßnahmen und wird hoffentlich letztendlich die Entwicklung von wirksamen Interventionsmaßnahmen unterstützen.

La calidad de vida se ha utilizado como sinónimo de una perspectiva de salud centrada en el sujeto o evaluada individualmente. No obstante, mientras que cada vez disponemos de más estudios sobre esta dimensión en los adultos, en los niños está relativamente olvidada. Han aparecido algunas determinaciones genéricas, pero se encuentran aún en fase de desarrollo los métodos para evaluar la calidad de vida de los niños con dolencias crónicas. En el presente artículo se detalla el diseño de una herramienta de evaluación de este tipo para diferentes grupos de edad y diferentes niveles de discapacidad realizado en el marco de un estudio financiado por la UE: el proyecto DISABKIDS. En él se estudiaron asimismo los determinantes psicosociales de la calidad de vida de los niños con discapacidades. Eran objetivos importantes del proyecto la elaboración de instrumentos de evaluación para niños y adolescentes con discapacidades y para sus familiares en siete países, la valoración del efecto de las dolencias crónicas sobre la calidad de vida y la elaboración de una herramienta para el control sistemático de la calidad de la asistencia que se presta a niños con discapacidades. La evaluación y el control permitirán identificar las necesidades de salud no cubiertas, y se espera que en última instancia estimulen el desarrollo de estrategias de intervención eficaces.

Keywords: adolescents; children; chronic condition; disability; quality of life

Introduction

Chronic health conditions constitute a challenge both for children and their families (Perrin *et al.*, 1993a). Depending on clinical characteristics of the condition, socio-economic factors associated with the life situation of the family as well as psychosocial mechanisms relevant for adaptation, these health conditions may be associated with a wide range of impairments, handicaps or disabilities. Within the international classification of handicaps, impairment and disability (ICIDH) the impact of chronic conditions on the patient has been acknowledged. While for adults these limitations have been spelled out clearly, children have not been sufficiently addressed. The relevant question for care and appropriate treatment of these children is three-fold: (1) What are the specific problem areas associated with the clinical condition, independent of aetiological or nosological considerations? (2) How are these impairments or challenges subjectively represented, that is, how do children as well as their family perceive the health state? (3) Which medical and psychosocial determinants regulate the health-related self-perception of the people concerned?

An important issue related to these three questions is the subjective perspective on health, namely health-related quality of life. Quality of life can be defined as subjective perception of health in physical, emotional, mental, social and functional domains. It is represented in terms of well being as well as functioning and should be identified via patients' self-reporting.

While in adults the definition assessment and practical use of the quality of life concept is rather advanced, quality of life research in children has developed recently. Even though defining quality of life as a concept for children, especially with regard to developmental stages, is a challenge, measures to assess quality of life in children from self-reporting as well as external reporting (parents) are emerging (Bullinger and Ravens-Sieberer, 1995; Ravens-Sieberer and Bullinger, 1998; Ravens-Sieberer *et al.*, 1999). These can be differentiated according to the scope of the quality of life assessment as generic or disease-specific measures, that is, measures that can be used independently of actual health states or that are bonded to such states. Such measures of perceived health are relevant in clinical research and practice to identify the patient's state (a descriptive perspective), the effects of treatments (a clinical perspective) and for quality assurance (an evaluative perspective).

However, not only is documenting the quality of life of patients and family important, but also the unravelling of the determinants or factors influencing the quality of life. Here psychological constructs, such as coping and adaptation, health locus of control and health beliefs, as well as social support and social networks have been identified as major factors influencing patient-perceived quality of life (Thompson and Gustafson, 1996). Despite increased research efforts, it is not yet clear how regulation of the quality of life perception of patients, especially children, occurs and which factors are relevant. This goes

beyond identification of single factors and necessitates addressing the complex interplay of resources and strains as well as the perceptions in a dynamic model of quality of life regulation in children which is (as is the case in the adult literature) still lacking.

Taking into account the dimensions of quality of life, as well as their determinants, is also expected to have practical implications. The question is, if and how assessment of quality of life and its determinants contribute to the understanding of patient and family needs and to the provision of improved care. Thus, patient needs, seen as a direct reflection of documented deficits in quality of life, would arise from corresponding assessment; they can also be addressed in terms of reflection of patient-perceived and patient-expressed problem areas. Assessment of patient needs, especially in paediatric medicine, is even more neglected than quality of life (Ravens-Sieberer *et al.*, 1999).

Assessing both quality of life and patient needs can be viewed as taking up a challenge for medicine and health care, with the aim of improving patients' well being and functioning as well as their clinical health state by defining, understanding and intervening in problem areas (strains) as well as strengthening potentials (resources) associated with the health conditions. The focus on quality of life and needs in children as well as their families is important also with regard to cultural diversity. In the European Union, depending on the organization of health care systems, different approaches to quality of life and needs assessment may be necessary, which has not been well understood so far. Adding an international perspective to assessing quality of life and patients' needs associated with chronic health conditions in childhood was a major impetus to develop and constitute a European co-operative research project. The current paper will focus on aims and tasks of the European co-operation project in this area, which has been approved and is now funded by the European Community.

The DISABKIDS Project

International Scope

Within the Fifth Framework Research Programme 'Quality of life and management of living resources' the European Community elicited proposals in six key action areas, as well as in several areas of so-called generic activities. Within the programme, one of the generic activities relates to people with disabilities. Research projects in this area were expected to focus

on assessing the quality of life of people with disabilities, taking into account special needs and perspectives of this population. Following the call for proposals for the European Community Grant Proposal, a group of researchers from seven countries co-operated to develop a research proposal oriented towards the main objectives required for grant solicitation.

The internal logic of the organization of the projects is derived from experiences in international instrument development, namely the World Health Organization quality of life (WHOQOL) questionnaire (WHOQOL Assessment Group, 1998) with the short form-36 (SF-36) health survey International Quality of Life Assessment Group (IQOLA) (Spilker, 1996) as well as European groups such as the EORTC (Bullinger *et al.*, 1996), the Nottingham Health Profile, the Sickness Impact and the FACT groups (Najman and Levine, 1981). In adapting instruments for international use these groups share a similar procedure, which involves a translation phase, a psychometric testing phase and a norming phase (Bullinger, 1991). Within the translation phase, forward/backward translations (to and from source to target language) according to defined guidelines are carried out, using in addition the quality ratings of the translation. In the psychometric testing phase the instrument is used in samples of healthy or chronically ill populations in order to test the reliability, validity and sensitivity of the measure. In a final norming phase, measures are included in representative population surveys in order to obtain information about the distribution of the scale scores in the population as well as norm-based scoring.

Within the international quality of life measures available so far, three approaches to adaptation of one instrument in one language to one or more different languages have been used. The first is a sequential approach in which an existing measure, which has been developed in a source language, is translated according to the above-mentioned protocol to other languages. This is the case with the SF-36 health survey questionnaire (Spilker, 1996), for example. A second approach is the development of an instrument in different languages, using already existing subscales or instruments from several languages. This has been the case with the EORTC quality of life questionnaire, which contains segments derived from instruments available in the countries participating in the project. The third approach is sequential in that different countries co-operate to create a new instrument simultaneously in several languages. This approach has been adapted by the WHOQOL working group (Bullinger *et al.*, 1996) who have identified relevant

dimensions from expert discussions in focus groups and then proceeded to collect items within each country and language to be accumulated, translated and reduced, thus forming the first basis for item development and data collection in participating countries.

Although instruments to assess the quality of life of children are available, in terms of generic as well as increasingly in terms of disease-specific approaches, only a few measures have addressed quality of life from a cross-cultural perspective. This is especially necessary for disease-specific measures because, due to cultural perceptions as well as existing differences in health care systems, these diseases may have different impacts on the quality of life and well being of children in different countries. In addition, perspectives not only of children but also of families and physicians are important sources for identifying the relevant dimensions of quality of life for children with chronic health conditions. Therefore the current project adopted a methodology based on the WHO-QOL work, which involved identifying relevant dimensions as well as items for condition-specific instruments both from available literature and from focus interviews with children, families and physicians. Using such a methodology, which is based on the simultaneous approach to quality of life assessment, the first phase of the project is devoted to instrument development and pilot testing and the second phase to field testing and implementation in different care contexts.

Conceptual basis

The term 'health-related quality of life' has been introduced into the health care field relatively recently, as a consequence of rising dissatisfaction with available indicators to assess health in the clinical context (Najman and Levine, 1981). Despite its relatively short history, quality of life research within the past years has made a contribution to current thinking in the medical and health care field by explicitly focusing on patient views and concerns (Bullinger, 1991, 1997; Landgraf *et al.*, 1997a).

Within the available definitions of quality of life, a consensus has emerged that quality of life reflects the subjective perceptions of physical, psychological, social, cognitive, functional and behavioural dimensions of wellbeing and function as perceived by the people concerned (Stewart and Ware, 1992). More broadly defined by the World Health Organization (WHO), health-related quality of life is seen as 'the individual's perception of their position in life in the context of the culture and value systems in which they

live, and in relation to their goals, expectations, standards, and concerns' (WHOQOL Assessment Group, 1998).

The quality of life field has developed rapidly in the past years, especially in terms of development of methods to assess quality of life and their implementation in medical and health care settings (Spilker, 1996). The majority of research in the quality of life area, however, pertains to adults. Research involving children constitutes, according to a recent literature research, only about 13% of the adult literature (Ravens-Sieberer *et al.*, 1999). Although recent attempts to understand and assess the quality of life of children with chronic conditions, for example, asthma (Juniper *et al.*, 1992), have improved, the assessment methodology is still in need of development for other health conditions (Bullinger and Ravens-Sieberer, 1999). This is especially true for children with chronic health conditions and for disabled children (Kokkonen and Kokkonen, 1995).

Parallel to the development of the quality of life field, current thinking about the classification of health conditions has also changed (Perrin *et al.*, 1993b). In addition to the biomedically based model and classification of diseases (ICD-10) the concept of disease burden (in terms of, for example, functional limitations) has evolved. This has led to the development of corresponding classification systems such as the ICIDH (currently ICIDH-2). While in adults the classification of the ICIDH-2 relating to the structure of the condition, activity and participation is advanced (the ICF), such a classification and assessment system for children is still at the developmental stage (Newacheck and Taylor, 1992; Pless *et al.*, 1993; Levine *et al.*, 1999).

Additional determinants, that is, psychosocial variables such as living conditions, social support, coping, treatment-related factors, perceptions of care and socio-economic factors are relevant determinants of patients' quality of life (Lazarus and Folkman, 1984; Thompson and Gustafson, 1996). This view has been supported by a series of studies in which, for example, by using regression analysis, the proportion of variance explained by psychosocial variables has been found to be high; even higher than that of clinical variables (Lavigne and Faier-Routman, 1992).

In addition to assessing the quality of life from the perspective of children and adolescents (using self-reporting questionnaires as well as interviews and computer-assisted forms of administration) the quality of life of the children's families should also be the focus of attention. This involves the parents' perception of children's quality of life as well as their rating

of their own quality of life. For practical reasons, the research often focuses on the main caregiver to the child, mostly mothers; however, deliberate efforts should also be made to include the perceptions of fathers and siblings (Dyson *et al.*, 1989).

Innovative methods of assessment, which are based on views of patients and parents and pertain to multiple dimensions of perceived health and self-reports and are practicable for people with disabilities, are still relatively new in the paediatric area (see, for example, Landgraf *et al.*, 1997b, 1998). Corresponding methodologies have been suggested (WHOQOL Assessment Group, 1998; Orley and Kuyken, 1994) along with procedures to translate and evaluate their results (Bullinger *et al.*, 1996, 1998) but the co-operative simultaneous development of such measures (not only the mere translation of an existing measure from one language to another) is necessary. Furthermore, the current literature relating to the quality of life of people with disabilities are in need of assessment methods that are compatible with the condition. The development of computerized (for example, touch-screen) versions of an assessment instrument with the benefits of easy application and scoring has very recently been introduced for adult health conditions but has not yet reached quality of life assessment in children (Ravens-Sieberer and Bullinger, 1998). Another major problem with the available studies on the situation of children and adolescents with disabilities is the lack of involvement or interest in patient perceptions, not only about their condition but also about their care. Assessing care as perceived by the recipients and eliciting alternative concepts of care from the people concerned is of utmost importance (Stein, 1998). It has to do with empowerment of the patient, with customer orientation in health care systems and with genuine respect for the individuals' perception of their health conditions and their views and needs of care (Baumann *et al.*, 1997; Perrin and Thyen, 1999). Such correspondence between assessment methods and goals of care based on direct patient involvement is innovative in medicine and is a prerequisite towards improving care in order to integrate people with chronic health conditions into the community (Perrin *et al.*, 1993a).

Objectives

Overall, the aim of the project is to enhance the quality of life and the independence of people with disabilities, taking into account their expectations and the greater contributions they could make to society. Central to the pursuit of this aim is the assessment of

quality of life from the perspective of people with disabilities, in terms of personal and social wellbeing, with special emphasis on health care and medical treatments. The project focuses on children and adolescents, trying to understand the views and needs of the young and very young in order to improve their chances of leading a fulfilling life at present and in the future.

Explicitly, the project aims to provide methods to assess the quality of life of children and adolescents and their families by using knowledge already accumulated in the area of adult quality of life research with different types of healthy and ill populations in the cross-cultural context. As such, the tasks within the project are oriented towards developing, with international consensus, modules for assessing quality of life in children and adolescents with chronic health conditions, to psychometrically test the quality of the instruments in different countries and to assess the value of implementing them in routine care.

Specifically, a major objective of the project is to understand quality of life as a function of the patients' and their families' views of health conditions and treatments, by using condition-specific methods to assess quality of life in chronic disease and disablement in combination with available generic instruments for quality of life assessment. These condition-specific methods are developed co-operatively in a European context and will be grounded in the perceptions of the patient and their family, will be rigorously empirically tested and will be available in computer-assisted versions for easy and practical use.

The analysis of the determinants of quality of life for these populations ranges from medical through psychosocial and socio-economic factors to perceptions of current treatment and care. Interactions and dynamics in the relationships between determinants and outcomes will be analysed, for example, the effect of the caregiver's quality of life on the child and vice versa. In a third project phase the assessment methodology will be implemented in caregiving institutions, for example, before and after an intervention aimed at improving the child's health state or perceived quality of life.

In sum, the project attempts to approach quality of life assessment in children and adolescents with disabilities in a cross-culturally sensitive manner:

1. By proposing European co-operation in the construction of an assessment instrument for quality of life, in which the people concerned (children, adolescents and their families) play a major role.

2. In that needs for care are explicitly voiced and addressed by the people concerned.
3. In that the psychosocial as well as the clinical and socio-economic determinants of quality of life are respected or addressed, by a mode of administration being developed which is practicable for the patient group concerned.
4. So that in using the instrument, current care can be evaluated and future care can be improved by correspondence with the patients needs.

Research

The DISABKIDS project consists of three phases, ranging from the development of instruments to assess quality of life in adolescents and children (as well as their families) through testing of the instrument in the out-patient clinical setting to its implementation in institutions involved in the care of this population of people with disabilities. Conceptually, it is based on recent revisions in the definition of disabilities. Methodologically, it is oriented towards rigorous testing of a quality of life assessment instrument in a field study. It follows a process of instrument development, which is based on simultaneous, co-operative effort between the European partners, following cross-cultural assessment guidelines. Practically, it takes into account assessing the quality of life of people with chronic conditions in health care settings within the European context.

In addition to a strong conceptual aspect, the current project is also oriented towards empirical testing of the assessment instruments in a pilot-test phase and to a field trial and the evaluation of its implementation in the clinical context. In its empirical phase it works with a cross-sectional design in the pilot study and the field study, in which a retest in a subset of responses for sensitivity analysis is planned. In the implementation phase, a longitudinal study design involving evaluation of the use of the new tool in institutions in conjunction with ongoing interventions or time-sensitive repeated measurements after a period of four weeks is planned.

Patients

People from the following diagnostic groups will be included in the study: children and adolescents aged 4–16, in three age groups (4–7, 8–12, 13–16) with the chronic condition of asthma and, depending on the participating centre's clinical focus, severe cerebral affections such as cerebral palsy, epilepsy, juvenile

diabetes, juvenile rheumatoid arthritis, cystic fibrosis and serious skin disease.

In addition to the diagnostic classification of the chronic conditions in the study, the degree of severity of each condition will be assessed by global clinical impression, and for high severity levels be defined according to the following criteria:

1. impairment in bodily, emotional or mental functions or activities (ICIDH, impairment level);
2. anticipated duration of the disease of <6 (or 12) months;
3. presence of at least one of the following consequences of the limitations:
 - functional limitations in daily activities (ICIDH disability level)
 - limitations in age-appropriate growth and development
 - limitations in age-appropriate social activities (ICIDH handicap level);
4. need for compensation of health-related limitations by medication, diets, medical technology, aids and health care; and
5. excessive need for medical, nursing or psychosocial care and support.

Ethical issues

The current work might raise ethical issues due to the involvement of children. Although the current project is not a clinical trial and does not involve specifically planned medical interventions, issues regarding potential 'psychological adverse effects of interviewing' have to be taken into account. Previous experience with quality research in chronically ill children suggests that such effects are rare and that they can be avoided by making sure that a therapeutically qualified person is available upon request.

Participating families are being informed about objectives and procedures within the project both in written and oral form. Informed consent to participate in the study is being obtained first from parents and then from the child. Informed consent forms and study information will be provided to the ethics committees along with the study proposal in each country. An acknowledgement of the project and all approvals of the ethics committees have been obtained prior to data collection.

Instruments and analysis plan

Available generic instruments to assess quality of life include examples such as the American child health

questionnaire (CHQ) (Landgraf *et al.*, 1998), the German KINDL (Ravens-Sieberer and Bullinger, 1998), the TACQOL (Feekes *et al.*, 2000) and the French VSP-A (Simeoni *et al.*, 2000). From these generic instruments one will be selected by the partners to be included in all countries; each country is free to choose two more. From the KIDSCREEN sister project, the newly developed European generic quality of life (QOL) scale for children will be used. Where available, psychometrically tested disease- and condition-specific measures will also be included for validation purposes.

Socio-economic and clinical data will also be collected; psychosocial data will include coping, social support and locus of control. A special emphasis is placed on identifying instruments for the evaluation of care and needs of care as expressed by the patients and their families.

Within the project, descriptive statistics as well as psychometric methods and exploratory statistical approaches will be used. Psychometric testing will include methods to assess reliability (internal consistency, test-retest reliability) and validity of questionnaires (here confirmatory factor analysis will be performed to replicate the assumed scale structure of the questionnaire). Classic and modern techniques such as item response theory will be used to examine the instruments' psychometric characteristics further. Exploratory statistics involving inferential methods will be used to identify differences between patient groups, the relationship between parent and child assessments and questions relating to the prediction of psychosocial, clinical and socio-economic data. To explain the quality of life variance, logistic as well as multiple regression analysis will be applied. The use of factor analysis will be restricted to identification of a hypothesized factor structure of the instrument (principal component analysis). Analysis across countries will use correlational and structural equation modelling for the global dataset, with individual countries subsequently taken out in the replication of the analysis.

Workplan

The project work is organized in 11 work packages, which are defined with regard to aims, co-operating partners, methods and deliverables (see Table 1). The 11 work packages pertain to three project phases, namely preparation, pilot and field study and implementation, and the first three of these work packages are already complete.

Table 1. Workpackages of the DISABKIDS project

WP 1	Literature review
WP 2	Focus Groups
WP 3	Instrument Dev.
WP 4	Translation
WP 5	Pilot Study
WP 6	Analysis of Pilot
WP 7	Field Study
WP 8	Statistical Analysis
WP 9	Implement. Plan
WP 10	Implement. Phase
WP 11	Final Results

1: Literature review and start-up workshop (months 1–3)

The results of a literature review on available quality of life research in disabled children were presented at the first workshop. Here, partners gathered to review the literature, discuss available generic questionnaires with regard to their use in the current project and come up with dimensions of quality of life to be included with newly developed condition-specific modules.

2: Focus group work (months 4–6)

In each participating country, nine families per condition (three age groups, three severity degrees, children and parents) were interviewed by the project staff at the centre with regard to their perception of quality of life issues pertaining to the health condition. The goal was to come as close as possible to item formulation in the respective dimensions. In addition, parents and experts will be consulted to provide items for the common item pool. Altogether, 196 focus-group interviews were conducted in all centres and 3027 statements were derived from the focus groups.

3: Item development (months 7–9)

The responses and items collected in each centre were translated into English and transferred back to the project partners for feedback. Here, consensus about retaining or rejecting items was reached using an iterative process including redundancy scoring, and card-sorting procedure to calmly items and a cognitive debriefing. From this, pilot versions of three age-related, severity-level-oriented questionnaires were developed in English to be pilot-tested in the different languages, along with a draft of the study protocol governing the pilot-testing procedure.

4: Translation (months 10–12)

The translation of the questionnaires will include two forward and one backward translations and be

performed according to published guidelines as well as an international harmonization session. In parallel, the study protocol for the pilot-testing will be developed and finalized at the second workshop.

5: Pilot study (months 13–15)

Here 18 patients per condition (three age groups, three degrees of severity, both sexes) and their parents will be asked to complete the pilot questionnaire. Informed consent will be obtained during routine visits to the centre in which a research assistant will inform participants about the study, distribute the questionnaires, help the children to fill the questionnaires out by themselves and interview children unable to do this. In a short cognitive debriefing session, the newly developed items will be discussed with children and parents with regard to their comprehensibility, clarity and acceptance. Preparatory work for the development of condition-specific, computer-assisted modules will also begin.

6: Analysis of pilot test results (months 16–18)

The next step involves the analysis of the pilot data including the cognitive debriefing results from the questionnaires. Here, explanatory psychometric methods will be applied to the questionnaires on the item level and on the scale level (reliability and validity). Cognitive debriefing results will serve to identify items as candidates for inclusion or modification according to international guidelines (for example, WHO). Results will be discussed at the third workshop in which the study protocol for the main phase will also be finalized.

7: Field test (months 19–24)

The main testing phase of the questionnaire will involve 180 families per country focusing on asthma, with each centre choosing at least two additional patient populations (for example, epilepsy, diabetes, arthritis, serious skin disease). As in the pilot phase, interviews and questionnaires will be completed according to procedures developed in the study protocol. Technical prerequisites for a computer-assisted version will be prepared in parallel.

8: Statistical analysis (months 25–27)

Analysis regarding psychometric properties of the newly developed instrument will use classical methods (reliability and validity) and modern psychometric methods (for example, item-response theory). Its results will be fed back to the group at the fourth workshop. During this phase the first prototype of a computer adaptation of the questionnaire will be developed.

9: Implementation plan (months 28–30)

Each country will be responsible for designing and logistically planning the implementation of the new assessment instrument by devising an implementation programme in which the quality of the instrument can be tested. Here, the use of computer-adapted versions and their installation in participating centres is planned. An evaluation programme to test feasibility and the clinical impact of the tool will be developed.

10: Implementation study (months 31–33)

In this step, the implementation of the instrument and its evaluation in the participating centres will take place. Questionnaires and computer-assisted versions will be used at selected centres over time and in a prospective manner. At least 10 participants per condition will be asked to complete the questionnaires twice at a four-week interval; clinical events during this time will be noted to assess sensitivity. Should an intervention be ongoing, children will be asked to assess their quality of life at the beginning, within the intervention and its end. A user evaluation form will accompany the questionnaire, to be filled in by children and medical staff.

11: Final analysis and report (months 34–36)

Analysis of data across countries and conditions, as well as the implementation evaluation data, will be addressed in the final step. Field-test data will be analysed from the global dataset across countries using modern statistical techniques. In the final workshop these analyses will be discussed before writing the final report, which will be a joint effort by all participants under the leadership of the study centre.

Project management and partnership

The programme management structure involves a study centre, in which the co-ordinating centre is incorporated, as well as the participating centres, forming the project consortium. Central project management lies within the study centre and is responsible for time management, financial management and the reports regarding the deliverables. It thus advances the project and, with the help of the co-ordinating centre, organizes the activities in the work packages.

The participating centres are active in that they are involved in each project step (high involvement in the data collection phases and centre-specific lower involvement in the data analysis phases). Throughout the project they provide specific expertise in clinical, statistical or organizational matters as expressed in

their role in the work packages. The partners also represent different cultural groups. They are experienced both in work with disabled people and in quality of life research. Their expertise ranges from clinical knowledge to epidemiological knowledge including statistics and psychometrics.

Each centre has one voice in the project consortium that functions as a steering committee and is necessary for scientific and policy issues. In addition, an external advisory board supports the steering committee. The advisory board includes people with experience in quality of life and rehabilitation research as well as clinicians, health officials and researchers involved in the disability area. Members of the advisory board have consented to be available for contact and to give advice to the project group at critical phases. They will be informed about the progress of the work in conjunction with the completion of work packages and the start of new work packages and they are invited to participate in telephone conferences as well as to participate in project meetings. The DISAB-KIDS project co-operates closely with its sister project KIDSCREEN, which involves the development of a generic instrument to assess the quality of life of children and adolescents and its inclusion in representative samples of young people in seven European countries.

Discussion

The development and use of standardized assessments for chronic health conditions are of increasing importance in order to minimize, in time, possible differences in health care systems in terms of what is offered and how it is evaluated. To devise an instrument to assess quality of life for children with disabilities in several European languages makes it easier on a national and international level to communicate the approaches to and experiences with care for the disabled to identify as well as possibilities for improvement. The project complies with the notion of assessing quality of life from the perspective of people with disabilities and their families by the implementation of the assessment methodology in caregiving institutions and evaluation of its contribution to improved care. It contributes conceptually by co-operative European instrument development, methodologically by testing assessments tools in a pilot and a field study, and practically by implementing these tools in caregiving institutions to enable them, on the basis of patient views and feedback, to continuously monitor, improve and evaluate their treatment options.

In providing a quality of life assessment tool, the current project goes beyond what has been achieved so far in terms of health assessment. It makes available a unique assessment of quality of life, in which children with disabilities can learn to express their current feelings through a paper-and-pencil as well as a computer-assisted, touch-screen programme. Such a computerized instrument, installed in the different clinics, can serve as a prototype for further application of quality of life research in other countries, other centres and with other conditions.

Caring for children with disabilities will be one of the important challenges for health care systems in Europe in the twenty-first century. Not only will the number of children with chronic health conditions increase, but also the resources to provide care for this population will be reduced Europe-wide due to savings in health care expenditure. The question of how optimal care for these children can be provided is most important for the people concerned, as well as for national and European health policies. In order to address this question, research, at a European level rather than national level, is necessary. Quality of life is one of the major descriptors and outcome criteria that have been discussed in health care systems in recent years. Quality of life of children and especially disabled children, however, is yet a not a well-researched area. By aiming to achieve European standards, in which generic as well as condition-specific quality of life assessments are included, as well as children's self report and parental perceptions of routine health care planning and outcome assessment, a major contribution to European Union policy is sought. Hopefully, this will lead to European Union policy in which the quality of life of children and adolescents with disabilities plays a role in the provision and evaluation of health care.

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