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Translation and Linguistic Validation of a Disease-Specific Quality of Life Measure for Cystic Fibrosis

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Objective: **To develop a conceptually and semantically valid English version of a French disease-specific measure of quality of life for children, adolescents, and adults with cystic fibrosis (CF).**

Methods: **Following a backward and forward translation of the measure, 60 participants, including 20 children, 20 parents, and 20 adolescents/young adults completed the Cystic Fibrosis Questionnaire (CFQ) and a series of cognitive probes evaluating their understanding of the items and response choices.**

Results: **Semantic and conceptual problems with the items were identified and modified for the second set of cognitive interviews. Response distributions across items and ages were adequate, and the predicted associations between disease severity and quality of life were obtained.**

Conclusions: **The English version of the CFQ appears to be a linguistically valid measure of quality of life for patients with CF. A national validation study is now under way to test the psychometric properties of the measure.**

Key words: *cystic fibrosis; quality of life; methodology; pediatric psychology.*

The measurement of health-related quality of life (HRQOL) provides unique information about the impact of an illness and the effectiveness of various treatments, particularly for chronic conditions, such as cystic fibrosis (CF), in which treatment aims to improve daily functioning and overall well-being (Drotar, 1998). Although traditional measures of physical functioning (pulmonary and nutritional indicators) are critical, they do not adequately capture the broader impact of the disease on the patient's physical, social, and psychological functioning (Quittner, 1998).

Significant progress has been made over the last two decades in defining and measuring HRQOL (Guyatt et al., 1997; Spilker, 1996). Although initially considered vague and difficult to define, HRQOL has become a multidimensional construct that includes several core domains, such as physical functioning and symptoms, psychological and emotional state, and activities of daily living. Furthermore, it reflects an individual's subjective evaluation of his or her daily functioning and well-being because it is generally centered on the patient rather

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than the physician (Levi & Drotar, 1998; Spilker, 1996). Rigorous standards for the development and psychometric evaluation of QOL measures have been published (Juniper, Guyatt, & Jaeschke, 1996), and reliable and valid measures of HRQOL for several chronic conditions, such as asthma, are now widely used (Juniper, Guyatt, Ferrie, & Griffith, 1993).

There are several reasons for developing a HRQOL tool for CF. First, the discovery of the genetic defect for CF in 1989 has led to dramatic advances in our understanding of the pathophysiology of the disease. This, in turn, has led to the development of new treatments that have shown promise for increasing life span and improving patients' quality of life. For example, new drug treatments, such as rhDNASE and inhaled antibiotics, have recently been evaluated in a series of short-term safety and efficacy trials, with some indication of improvement in pulmonary functioning and HRQOL (Fuchs, Borowitz, & Christiansen, 1994; Ramsey, Astley, & Aitken, 1993). In these studies, however, ad-hoc QOL items were administered in the absence of a well-validated measure of HRQOL for CF, making it difficult to ascertain how these treatments affected quality of life.

Another reason for assessing HRQOL is that for chronic diseases, in particular, some treatments may yield benefits in activities of daily living that are not reflected in conventional medical outcomes. Anecdotal reports from families in our nutrition intervention study, aimed at increasing calorie intake and weight gain in young children with CF, have suggested that the children's increased energy level and ability to participate in sports was as exciting for families as the changes in weight and height percentiles (Stark et al., 1998). Further, a patient's perception of improvements in daily functioning may play a critical role in increasing adherence to complex and time-consuming treatment regimens (Abbott, Bilton, Dodd, & Webb, 1994; Quittner et al., 2000).

Finally, the assessment of HRQOL provides another basis for evaluating the effectiveness of different treatments. Many of the new therapies being introduced for CF are time-consuming and use significant health care resources. The QOL data may be useful for comparing the cost-effectiveness of different treatments (e.g., shorter hospital stays), which may influence health care policy and reimbursement (Feeney, Torrance, & Labelle, 1996; Kaplan et al., 1989). In a recent review of outcome measures for clinical trials in patients with CF, the Consensus Group recommended that HRQOL measures be incorporated into phase 3 clinical trials for both children and adults with CF (Ramsey & Boat, 1994).

More than a decade ago, the National Institutes of Health sponsored a conference on the psychological and behavioral aspects of CF and recommended that a disease-specific measure of HRQOL be developed and included in future studies along with conventional health outcomes (Eigen, Clark, & Wolle, 1987). Despite this recommendation, efforts to develop a reliable and valid CF-specific measure have just begun (Henry, Aussage, Grosskopf, & Launois, 1996; Henry, Grosskopf, Aussage, Goehrs, & Launois, 1997; Quittner, Sweeny, Watrous, Munzenberger, & Henry, 1998).

Prior studies of HRQOL in CF have relied on generic measures, which ask general questions about symptoms and functioning that can be completed by patients with a variety of chronic conditions. Two major types of generic QOL measures have been used with CF patients: (1) utility measures, such as the Quality of Well-Being Scale (Kaplan et al, 1989; Orenstein & Kaplan, 1991; Orenstein, Nixon, Ross, & Kaplan, 1989; Orenstein, Pattishall, Nixon, Ross, & Kaplan, 1990), and (2) health profiles, such as the Nottingham Health Profile (Caine et al., 1991; Dennis et al., 1993; Buschbach, Horiks, van den Bosch, de la Riviere, & de Charro, 1994). Although a complete review of these measures is beyond the scope of this article, their major limitation is a lack of sensitivity to the specific challenges posed by CF (Czyzewski, Mariotto, Bartholomew, LeCompre, & Sockrider, 1994; Munzenberger, Van Wagner, Abdulhamid, & Walker, 1999), which makes it more difficult to quantify changes in HRQOL that result from new interventions (e.g., lung transplantation), or the natural progression of the disease (see Quittner, 1998, and Tullis & Guyatt, 1995, for reviews).

Another significant limitation of current HRQOL research is the relatively small number of measures for children and adolescents (Landgraf, Abetz, & Ware, 1996; Varni, Seid, & Rode, 1999). Instruments designed for these age groups must be developmentally appropriate and, therefore, must take into account the psychosocial and cognitive differences between age groups. Health-related quality of life measures for children, for example,

should include modules that assess daily functioning at school and social relationships with peers (Quittner et. al., 1996). The QOL measures designed for adolescents should contain items that reflect developmental shifts in independence (e.g., planning for college) and increased responsibility for disease management (e.g., remembering nebulizer treatments) (DiGirolamo, Quittner, Ackerman, & Stevens, 1997; Quittner et al., 2000). In sum, there is a great need for a reliable and valid CF-specific HRQOL measure that can be used with children, adolescents, and adults.

To date, the work of Henry et al. on the Cystic Fibrosis Questionnaire (CFQ) in France represents the only published effort to develop a disease-specific HRQOL measure for patients with CF (Henry, Aussage, et al., 1996; Henry, Aussage, Grosskopf, & Goehrs, 1998; Henry, Aussage, Staab, Prados, Grosskopf, & Goehrs, 1997; Henry, Grosskopf, et al., 1997). A major advantage of the CFQ is its developmental approach, which has yielded three separate QOL measures, one for school-age children 6 to 13 years, one for parents of school-age children, and one for adolescents and adults (ages 14 and older). Initial item development began with an extensive review of the literature, interviews with experts and CF associations, and qualitative interviews with 44 children and adults with CF and their parents (Henry, Aussage, et al., 1996). Five generic (e.g., physical symptoms, role functioning) and four CF-specific (e.g., eating disturbances, treatment burden) HRQOL domains were identified. Three initial questionnaires were constructed: a child version with 20 items, a parent version with 81 items, and an adolescent/adult version with 90 items. In all three versions of the measure, higher scores indicate better quality of life. Pilot testing with 30 participants led to minor wording changes and the addition of several new items of the child version of the CFQ (Henry, Aussage, et al., 1998).

In the item-reduction phase, 534 questionnaires were completed at 24 hospital sites in France with patients whose illness severity differed. Descriptive statistics and factor analyses provided support for the initial scale structure. Internal consistency coefficients of .46 (social) to .71 (physical) were reported for the CFQ-Child version, .60 (body image) to .89 (physical) for the CFQ-Parent version, and .61 (embarrassment) to .92 (physical) for the CFQ-14+. Moderate convergence was found between the CFQ 14+ and the Nottingham Health Profile (correlations ranging from .43 for social limitations to .75 for physical functioning). Significant associations were also found between greater illness severity, as measured by Schwachman scores, and lower scores on the CFQ-14+. Short-term test-retest reliability (7 to 10 days) yielded significant intraclass correlation coefficients ranging from .66 to .97 (Henry, Aussage, et al., 1998; Henry, Grosskopf, et al., 1997).

The CFQ is currently undergoing extensive development and psychometric evaluation in other countries. Because CF is found throughout the world, and the medical and behavioral treatment approaches are fairly consistent and shared in several conference and publication venues, it is important to have a well-validated, international measure of HRQOL for CF. As new medical treatments become available, such as gene therapy, and others, such as lung transplantation, become more widely available, it would be extremely beneficial to have an international tool to evaluate the impact of these treatments on HRQOL. Psychometric validation of the French CFQ is nearly complete, and the German and Spanish translations of the CFQ are now being evaluated with larger samples (Henry, Aussage, Staab, et al., 1997). More recently, analyses testing the conceptual equivalence of the items across all three languages (French, German, and Spanish) using Rasch modeling techniques were presented at a national conference (Henry, Staab, et al., 1998).

Given the importance of developing a disease-specific measure of HRQOL for CF, the strong psychometric properties of the original French CFQ and its potential as an internationally validated tool, the central aim of this study was to develop a conceptually equivalent English version of the French CFQ. The specific objectives of this study were (1) to complete independent forward and backward translations of the French CFQ into English following international guidelines; (2) to complete an initial qualitative evaluation of the English versions of the CFQ with a small sample of patients with CF and parents to identify semantic, conceptual, and psychometric problems with the initial translations; (3) to modify the measures based on results of the first evaluation, and then conduct a second round of qualitative interviews to identify any remaining semantic or conceptual problems with the items; and (4) to produce English versions of the CFQ that are conceptually and semantically valid and can be tested in a national psychometric evaluation.

Translation Process

For researchers developing and validating quality of life measures in different countries, a consensus has emerged that four levels of cross-cultural equivalence must be achieved: (1) conceptual equivalence, (2) construct or item equivalence, (3) operational equivalence, and (4) metric equivalence (Anderson, Aaronson, Leplegè, & Wilkin, 1996; Hui & Triandis, 1985). *Conceptual equivalence* is the extent to which the items in the target language (in this case, English) are similar in meaning to the source version (in this case, French). This form of equivalence includes both the semantic meaning and formulation of the items (e.g., wording of questions), as well as the underlying concept being assessed (Brice & Kalimo, 1971). This is achieved through both the translation process and the qualitative testing that follows. *Construct or item equivalence* is the extent to which individuals in different cultural groups respond to the same items in similar ways, which is evaluated with classical test theory (e.g., response distributions, test-retest reliability). *Operational equivalence* refers to the relative performance of the instrument using various modes of administration (e.g., self-report, interview). Finally, *metric equivalence* is the extent to which individuals are ranked similarly along a continuum of HRQOL across cultures (e.g., patients with CF in different countries who have similar pulmonary function scores will have similar values on the CFQ) (Acquadro, Jambon, Ellis, & Marquis, 1996; Bullinger, Anderson, Cella, & Aaronson, 1993). The purpose of this study is to complete the first step of cross-cultural adaptation by developing a conceptually equivalent English version of the French CFQ.

International guidelines have been established for translating quality of life measures from one language into another and then demonstrating cross-cultural validity (Bullinger, Anderson, Cella, & Aaronson, 1993; Ware, Gandek, Keller, & the IQOLA group, 1996). This is an iterative process that began in this study with conference calls between the French developer (B. Henry) and the U.S. research teams to clarify important concepts and medical terms. Next, two independent forward translations were conducted with native French speakers living in the United States (i.e., from Wayne State University and Research Triangle Institute). Once the translations from French to English were complete, discussions were held between the translators to resolve semantic and conceptual discrepancies and to develop a consensus forward translation. The English versions of the measure were sent to France for backward translation (English to French), with Dr. Henry as the consultant. Conference calls between the French and U.S. research teams focused on the conceptual and rating scale issues that had been identified (e.g., what is the equivalent of "pleine forme" in English?). Following these discussions, a consensus version of the three CFQ measures in English was finalized (Quittner et al., 1998). These measures were then qualitatively evaluated and modified in two separate studies to assess their conceptual equivalence.

Study 1: Initial Qualitative Evaluation: Method

Participants

Thirty participants were recruited for the initial qualitative evaluation: 10 children with CF ages 8 to 13, 10 parents of the children completing the CFQ-Child version, and 10 adolescents and adults with CF ages 14 and older. Participants were recruited from CF Centers in Indiana (Riley Hospital for Children) and North Carolina (University of North Carolina at Chapel Hill and Duke University). Patients with CF and their parents were contacted prior to a routine clinic visit and asked if they would be willing to complete a quality of life measure along with follow-up questions. Efforts were made to recruit even numbers of male and female patients, and a representative range of illness severity as measured by FEV_1 (forced expiratory volume in one second).

In study 1, the mean age of children completing the CFQ-Child was 10.56 (SD = 1.67); more female than male children participated in the CFQ-Child interviews (70%), but an even number of boys and girls completed the CFQ-14+. Mean age of participants completing the CFQ-14+ interviews was 23.3 (SD = 8.17). The majority of CFQ-Parent respondents were mothers (70%); however, two fathers and one grandmother, who were primary caregivers, also completed the interviews. An adequate range of illness severity was represented, with most younger children falling into the mildly ill category (range of FEV_1 73% to 101%; M = 89.67, SD = 10.31), and adolescents and adults falling into the mildly to moderately ill categories (range of FEV_1 59% to 93%; M = 69.71, SD = 11.63). (FEV_1 values for the three children and adults in study 1 were

not available.) Normal pulmonary functioning values range from 80% to above 100% predicted (Knudson, Slatin, Lebowitz, & Burrows, 1976). One child's protocol was not used because it became apparent during the interview that she was developmentally delayed and could not understand the questions.

Measures

Cystic Fibrosis Questionnaire-Child Version (CFQ-C; Henry, Grosskopf, et al., 1997; Quittner et al., 1998). CF-specific quality of life was measured using the CFQ-Child version. This measure is administered directly to children ages 6 to 13 and consists of 36 items. Twenty-one items require a frequency response ranging from "always" to "never" on a 4-point scale and 15 items require a true-false rating on a 4-point scale. These items assess the following seven QOL generic and disease-specific domains: physical functioning (eight items), energy and well-being (six items), emotional state (five items), social limitations (four items), body image (three items), eating disturbances (three items), and treatment constraints (two items). Two additional symptom scales, respiratory (four items) and digestive (one item), are also included (see Table I). Two practice questions are used with colored rating cards to train the child on the two types of response choices. This procedure ensures that the child understands the response choices. The cards are used as reminders during administration of the questionnaire. To further evaluate the child's understanding of the frequency and true-false scales, analogue thermometer scales were constructed to represent the response choices. After choosing a response (e.g., "always" to "never" or "very true" to "very false"), the child was instructed to show the interviewer where that response would fall on the thermometer. These selections were noted on the protocol. When not accompanied by additional qualitative questions, the CFQ-C takes 15–20 minutes to complete.

Cystic Fibrosis Questionnaire-Parent Version (CFQ-P; Henry, Grosskopf, et al., 1997; Quittner et al., 1998). Parents' report of their child's disease-specific quality of life was measured using the CFQ-Parent version, a self-report measure consisting of 44 items. Items require either a frequency response on a 4-point scale ("all the time" to "never"), a difficulty rating on a 4-point scale ("a lot of difficulty" to "no difficulty"), a true-false rating on a 4-point scale, or the selection of a statement that describes the child (on a 4- or 5-point scale). These items assess the following seven QOL domains: physical functioning (nine items), energy and well-being (six items), emotional state (four items), performance at school (four items), body image (three items), eating disturbances (two items), and treatment constraints (two items). Three symptom scales, respiratory (seven items), digestive (two items), and weight (one item), and an overall health perception scale (four items) are also included (see Table I). This measure takes approximately 15–20 minutes to complete on its own.

Table I. Domains Measured by the Three Versions of the CFQ

	CFQ-Child	CFQ-Parent	CFQ-14+
9 QOL dimensions			
Physical functioning	X	X	X
Energy, well-being	X	X	X
Emotional state	X	X	X
Social limitations	X		X
Role limitations/ school performance		X	X
Embarrassment			X
Body image	X	X	X
Eating disturbances	X	X	X
Treatment constraints	X	X	X
Symptom scales			
Respiratory	X	X	X
Digestive	X	X	X
Weight		X	X
Health perceptions		X	X

Cystic Fibrosis Questionnaire-14+ (CFQ-14+; Henry, Grosskopf, et al., 1997; Quittner et al., 1998). An adolescent/adult form of the CFQ was developed to measure perceptions of quality of life for teens and young adults with CF. This self-report measure consists of 48 items. Items are rated in terms of frequency, difficulty, true-false categories, or weighted statements using the same response choices described for the CFQ-P form above. In this version, however, nine QOL dimensions are assessed: physical functioning (eight items), energy and well-being (four items), emotional state (five items), social limitations (four items), role limitations (three items), body image (three items), eating disturbances (two items), treatment constraints (two items), and embarrassment (three items). Three symptom scales, respiratory (seven items), digestive (two items), and weight (one item), and an overall health perception scale (four items) are also included (see Table I). The CFQ-14+ takes 15–20 minutes to complete.

Procedure

The study's procedures were reviewed and approved by an institutional review board; consent was obtained from parents and adults; assent was obtained from children. The study protocol consisted of face-to-face interviews with each participant, during which they completed the CFQ with a series of cognitive probes following each question. The interviews were conducted in families' homes or in a private room adjoining the clinic and lasted approximately 45 minutes for children and between 60 and 90 minutes for parents and adolescent/adults. Children were reimbursed $30 for their time and effort; parents were compensated $50 for their time and effort. Recent pulmonary functioning test results were obtained, when possible, from medical charts.

Four sets of cognitive probes were developed, using well-validated cognitive testing procedures (Lessler & Forsyth, 1996), to evaluate the participant's understanding of the items and response choices. The first set of questions asked whether the directions for completing the measure were clear. The second set followed each item and served to clarify what the item meant to the participant, how the response choice was made, and what circumstances would require a different response. For example, on the CFQ-Child, after the item "You felt full of energy," the child was asked the following questions: (1) What did you think of when answering this question? (2) How do you act when you are feeling this way? (3) How did you choose your answer? (4) What would have made you choose a different answer? (5) Are there other words that mean the same thing, but are clearer? Probes for the CFQ-P also targeted the parent's reference point when answering the questions (e.g. "How well do you think your answer matches what your child might say?"). The third set of probes appeared after each major section and consisted of open-ended comments about the previous set of items, questions about the difficulty and relevance of the previous questions and answer choices, and comments on any difficulties the respondent had in distinguishing among the response choices. Finally, the interview ended with a series of open-ended questions assessing their overall evaluation of the measure, suggestions for topics that were not covered, and comments about items that seemed irrelevant or were particularly difficult to answer. On the CFQ-C, specific probes were also inserted periodically to determine whether children were attending to the 2-week time frame in choosing their responses. (Cognitive protocols are available upon request from the authors).

The qualitative interviews for each version of the CFQ were transcribed from audiotapes or session notes and collated across participants. In a series of group meetings that included several of the authors, as well as graduate and undergraduate students, the responses and comments to each item were read and discussed in an effort to identify ambiguities in the wording of items, trends in the distribution of responses, and common themes that emerged from the open-ended comments (e.g., meaning of the item, relevance). These qualitative data were then collated and tabulated across participants for each version of the measure.

Study 1: Results

The analyses of the qualitative interviews had four primary foci: (1) to examine the range of responses for each item; (2) to identify semantic problems with the items, (3) to identify conceptual problems with the items, and (4) to evaluate the scope and relevance of the measures through responses to the open-ended interview prompts.

First, we checked the numeric range of responses to each item for adequacy of the distribution. Given the small sample size of this study, a liberal criterion was set for the response distribution. Items that did not elicit responses in three of the four rating categories were flagged and reworded, as necessary.

Second, we examined semantic problems with the three versions of the CFQ. Two types of wording and language issues were documented. First, we noted problems with the translation of medical terms from French into English. As can be seen in Table II, two French terms, when translated literally into English (i.e., "sprays" and "spit"), were reported to be confusing and awkward for respondents on all three versions of the CFQ. Participants were asked to generate alternative terms for these items, and the consensus across groups was to change "spray" to "aerosol or mist treatments" and "spit" to "cough up mucus." Participants also had difficulty rating two of the items that contained double-barreled adjectives. Parents on the CFQ-P

Table II. Item-Level Semantic and Conceptual Issues in Study 1

Issues	Modifications	CFQ-C	CFQ-P	CFQ-14+
Translation of medical terminology				
A. "Sprays"	"Aerosol or mist treatment"	X	X	X
B. "Spit"	"Cough up mucus"	X	X	X
C. Mucus color				
1. "Closer to green"	"Yellowish-green"		X	X
2. "With traces of blood"	"Green with traces of blood"		X	X
Double-barreled items				
D. "Sad and depressed"	"Sad"			X
E. "Worried and anxious"	"Worried"		X	
Semantic and conceptual meaning				
F. "Felt very fit"	"Felt well emotionally and physically	X	X	X
G. "Felt full of energy"	"Felt energetic"	X	X	X
H. "Felt short-tempered"	"Felt mad"	X		
I. "Forced to eat"	"Pushed to eat"	X		
J. "Visit my friends a lot"	"Get together with my friends a lot"	X		X
K. "Spend a lot of time at home (reading, TV)"	"Have to stay at home more than I want to"			X
L. "My child works as usual (without more difficulty) at school"	a) "My child participates as usual at school"		X	
	b) "My child has typical days at school"			

noted that they would rate the two adjectives "worried and anxious" quite differently. Similarly, adolescents and adults stated that they would give different ratings to the adjectives "sad and depressed." For both of these items, a restricted range of responses was noted (i.e., only "sometimes" and "never" were endorsed). The less intense adjective was chosen for both items (i.e.,"worried," "sad").

Conceptual problems with the items (interpreting their meaning) were also identified in study 1 (see Table II). The French phrase, "pleine form," was translated into English as "felt very fit." However, the cognitive testing revealed that participants interpreted this as exclusively referring to *physical* fitness, rather than the broader French meaning of "well-being" as a whole (physical, social, and emotional). After considerable discussion among the French and U.S. teams, the item was changed to "felt well emotionally and physically." A different problem with the phrase "full of energy" emerged in all three versions of the CFQ. Both children, parents, and adults interpreted this item as meaning *too much* energy, as in "bouncing off the walls," behaving in a hyperactive manner. This item consequently was changed to "energetic." Intensity was also a problem in the item "forced to eat" in the CFQ-C and was changed to "pushed to eat." The words "short-tempered" were difficult for younger children, so this item was changed to "felt mad." Cultural differences were noted in items that reflected the socialization patterns in France and the United States. The item "visit my friends a lot" generated responses indicating that "visiting" is considered a more formal activity here than in France, and that the phrase "get together" better reflected a casual, American approach to socializing. In addition, "spend a lot of time at home" (reading and watching TV) was considered a social preference, rather than a consequence of having a chronic illness; therefore, this item was changed to "stay home more than I want to." One item on the CFQ-P was interpreted to mean how well the child performed in school, in terms of grades and achievements, rather than more general functioning. Consequently, two items reflecting typical school functioning were written and tested in the second cognitive evaluation.

Finally, all of the children reported that the directions for the CFQ-C were clear; several children liked the practice instructions, and none of the children had suggestions for additional items. On the CFQ-P, 90% of the parents reported that the instructions were clear and the items were highly relevant. Additional items for the CFQ-C were suggested to measure the child's acceptance by peers, comfort in

Table III. Item Level Semantic and Conceptual Issues in Study 2

Issues	Modifications	CFQ-C	CFQ-P	CFQ-14+
Translation of medical terminology				
A. "Green with traces of blood"	"With traces of blood"		X	X
Semantic and conceptual meaning				
B. "Felt energetic"	"Felt you had enough energy"	X		
C. "Felt well emotionally and physically"	"Felt well"	X	X	X
D. "My child works as usual at school	"My child has typical days at school"		X	

telling others about CF, and stigma associated with taking enzymes, as well as items covering sexuality, fertility issues, and job limitations for the CFQ-14.

Study 2: Second Qualitative Evaluation: Method

Participants

An additional 30 participants were recruited from the same locations and in a similar manner for the second qualitative evaluation: 10 children with CF ages 8 to 13, 10 parents of those children, and 10 adolescents and adults with CF ages 14 and older. The mean age of the children completing the CFQ-C was 10.1 (SD = 2.03); more female than male children participated in the CFQ-Child interviews (70%); however, slightly fewer females (40%) than males completed the CFQ-14+. Mean age of participants completing the CFQ-14+ interviews was 22.9 (SD = 6.24). All of the parent respondents were mothers. In the child sample, illness severity as measured by FEV_1 ranged from 75% to 104% predicted (M = 90.4, SD = 9.29), with most younger children falling into the mildly ill category. Illness severity in the adolescent/adult sample ranged from 23% to 135% predicted (M = 79.3, SD = 35), representing mildly to severely ill patients.

Procedures

The revised Cystic Fibrosis Questionnaires (CFQ-Child, CFQ-Parent, CFQ-14+; Henry, Grosskopf, et al., 1997; Quittner et al., 1998) were administered along with a shorter list of cognitive probes. Follow-up questions were written to specifically address the problematic items or response formats identified in the first evaluation. One additional item was added to the CFQ-P in order to test the phrasing of a school performance question. Parents responded to both versions of the item and were then queried about their understanding of the item and how their response might differ between the two choices.

Study 2: Results

The goal of the second round of qualitative interviews was to identify any remaining problems with the measure. Thus, we conducted the following analyses: (1) identification of remaining semantic and conceptual problems with the revised versions of the questionnaires; (2) inspection of the response distributions across participants in studies 1 and 2; (3) scoring of the CFQ measures across both studies, with an analysis of the means and standard deviations of the scales; (4) correlational analyses between illness severity and quality of life scores for the CFQ-C and CFQ-14+; and (5) review of the clarity, scope, and relevance of the measures via open-ended comments elicited at the end of the interviews.

First, the item-specific revisions of medical terms ("aerosol or mist treatment," "cough up mucus") were reviewed. Responses across all three versions of the CFQ indicated that these changes were beneficial, with little ambiguity or confusion reported. The only problem that remained was the description of mucus; several respondents noted that they may cough up blood in the absence of green mucus; thus, this item was changed back to "traces of blood" (see Table III).

Second, the changes made to clarify the semantic and conceptual meaning of the items following the first evaluation appeared to resolve many of the problems. An item that remained problematic referred to emotional and physical health as a whole ("pleine forme" in French). This item was still interpreted as referring primarily to physical health and

probably reflects the cultural value in the United States placed on physical fitness. After consulting with Dr. Henry, a decision was made to revise this item to "felt well" for subsequent studies. Factor analytic procedures with larger samples will be needed to determine which scale this item loads on, Physical Functioning or Energy and Well-Being. Rewording the item "full of energy" to "energetic" worked well for the parent and adult samples but was too difficult a word for children. Therefore, this item for the CFQ-C was changed to "felt you had enough energy."

All of the children and most of the adolescents, adults, and parents reported that the instructions and items were clear and that the items were highly relevant and easy to understand. Two parents commented on the redundancy of some items, and two adults found the true-false rating scale difficult to complete. A few of the questions related to school functioning were also described as problematic, since two of the children (20% of the sample) were home-schooled. Additionally, one parent noted that it was difficult to answer questions about her child's school behavior since she is not there to observe it. Suggestions were made by both parents and adolescents/adults for additional items. These included sibling issues, teacher lack of awareness, concerns about prognosis and mortality, marital relationships, and financial difficulties.

The samples from both evaluations were combined to examine the response distributions. In general, a majority of items on all three versions of the CFQ elicited responses in all four response categories (all categories endorsed for 50%–69% of the items, three categories endorsed for 24%–45.5% of the items, two categories endorsed for 4.5%–7% of the items). One other item distribution issue was noted. A gender difference was found on both the CFQ-C and CFQ-14+ for one item appearing on the Body Image scale. Girls tended to rate the statement "you think you're too thin" as "false" or "very false," whereas boys tended to endorse this as "true" or "very true."

Next, the three versions of the CFQ were scored for the respondents in both studies. Given the small sample size, these results should be considered descriptive and preliminary. Means and standard deviations were calculated, and those that appeared in all three versions of the CFQ are shown in Figure 1. As expected, because illness severity is related to older age in CF, adolescents and adults had lower HRQOL scores than children. Significant differences

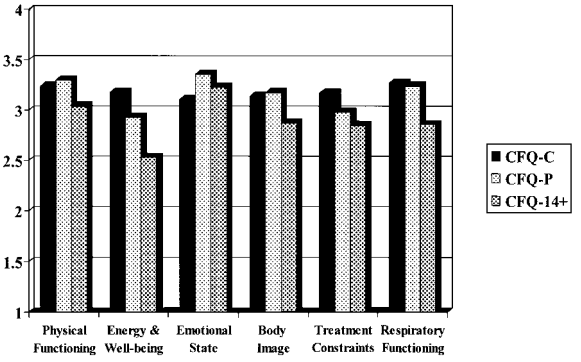

Figure 1. Average scores on the CFQ respondents.

were found on the Energy and Well-Being (t [37] = 3.50, $p < .01$), Social Limitations (t [37] = 2.15, $p < .05$)(child and adult versions only), and Treatment Constraints (t [37] = 2.0, $p = .05$) scale, and a marginal difference was found on the Respiratory Scale (t [37] = 1.86, $p < .07$). Consistent with the literature on emotional adaptation to CF (DiGirolamo et al., 1997; Quittner et al., 1996), we found no differences between children and adolescents/adults on the Emotional State scale.

Significant associations were also expected between illness severity (FEV_1 values) and quality of life scores on the CFQ-14+. Correlational analyses revealed positive associations between pulmonary functioning values (e.g., better lung function) and higher HRQOL on the Physical Functioning (r [17] =.62, $p < .01$), Body Image (r [17] =.56, $p < .05$), and Emotional State (r [17] =.54, $p < .05$) scales. (We did not compute correlations between FEV_1 scores and the CFQ-Child version because of a restricted range of illness severity in this group.)

Discussion

In general, the major objective of this study, to conduct a linguistic validation of a French, disease-specific measure of HRQOL for children, parents, and adults with CF was met. The systematic forward and backward translation process yielded measures that were generally clear to the respondents and highly relevant to their assessment of quality of life. Most important, children as young as 7 years of age were able to complete the measure with little difficulty, and adults at the older end of the developmental spectrum (41 years of age) found the CFQ-14+ to be face valid and applicable to their daily

functioning. Results from the first qualitative evaluation, however, did reveal problems with the wording of certain items, particularly medical terms, and minor problems with conceptual equivalence. Corrections of terminology (e.g., "sprays" to "aerosol treatments") were fairly easy to make, and the second phase of cognitive testing indicated that these problems were resolved.

Most of the semantic and conceptual problems were also easy to address by slightly modifying the wording of specific items and retesting them in the second qualitative evaluation. One phrase in French (i.e., "pleine forme"), which likely reflects a culturally specific view of health as both physical and emotional, remained difficult to convey in English. Although this item loaded on the Energy and Well-Being scale in the French version, feedback during both phases of cognitive testing indicated that it may load more strongly on the Physical Functioning scale in the English version. Factor analyses with larger samples will be needed to test this.

The response categories were also reported to be clear and easy to use. Adequate response distributions were found in the combined study samples across all three versions of the measure. One gender difference was noted for an item on the Body Image scale. Females tended to rate "thinness" as less of a problem than males. This is consistent with cultural pressures for females in the United States to be thin and has been noted clinically as problematic in the CF literature. This potential gender difference will have to be re-examined in a larger sample.

Although the combined samples sizes for the two studies were small, results on the scaled scores were in the expected direction. Higher quality of life scores were obtained for children, who are healthier and have fewer medical problems, than for adolescents and adults. Significant associations were also found between pulmonary function scores and the CFQ. Ratings of HRQOL were also similar for parents and their children, although a statistical test of this association will require a greater number of parent-child dyads.

Two methodological limitations should be noted. First, a small number of children, parents, and adults participated in both rounds of cognitive testing. Although this was appropriate for the purposes of evaluating the adequacy of the translation and identifying problems with conceptual equivalence, it precludes the use of most traditional psychometric tests (e.g., internal consistency). In addition, we recruited samples of volunteers who were willing to participate in a lengthy and intensive interview process, and these samples may not be representative of the larger pool of patients with CF and their family members. Efforts were made, however, to recruit participants from several CF centers to increase the diversity and generalizability of the sampled responses.

We encountered several challenges in conducting a multinational measurement study of this type that are worth noting. First, the research team represented several different disciplines (e.g., psychology, pharmacy, health economics), who brought different views and training backgrounds to the research enterprise. This was a strength in terms of adding the perspectives of different social science fields, but it added a layer of complexity to our communication and decision-making process (e.g., traditional rules of survey development vs. observations of how individuals responded to the items). Second, we experienced difficulties in trying to balance our goal of producing a "pure" translation of the original French measure into English that still captured the substance and meaning of the items for patients and family members living in the United States. These tensions are not unique to our development of the English CFQ and have been well-documented in other efforts to develop multinational measures (Cella, Lloyd, & Wright, 1996). The extent of our success in reaching this goal will be evident once we complete the national psychometric validation of the CFQ in this country. Finally, there were pragmatic difficulties involved in communicating with multiple investigators in the United States and France. We were able to arrange two long-distance conference calls to discuss potential changes in wording and question formats. This level of communication was necessary to maintain the integrity of the process, but posed significant financial and practical difficulties.

Future Directions

In the United States, the linguistic validation of the CFQ has now been completed, and the next step is to conduct a large-scale, psychometric evaluation of the measure. Funding has recently been obtained to conduct a national assessment of the measure at 22 different CF centers. One hundred and fifty respondents will complete each version of the measure. This will allow us to apply classical test theory to examine internal consistency, convergent and discriminant validity, and factor structures (Quittner

et al., 1999). Convergent validity will also be calculated between CFQ scores for parent-child dyads and with a generic HRQOL measure. Finally, in a subset of patients, a short-term (7 to 10 days) test-retest reliability study will be completed. Once this national database is available and analyzed, a manual will be developed containing the three CFQ measures, means and standard deviations, and other psychometric data. This manual will be made available to researchers and clinicians in the CF community.

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