Validation of the Danish version of the revised cystic fibrosis quality of life questionnaire in adolescents and adults (CFQ-R14+)

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Abstract

Background: Quality of life is an important parameter in the evaluation of quality and outcome of health care and treatment, especially in patients with chronic disorders. The aim of this study was to assess the validity and reliability of the Danish version of the revised disease-specific health-related quality of life questionnaire for adolescents and adults with cystic fibrosis (CFQ-R14+).

Methods: A total of 196 cystic fibrosis (CF) patients completed the CFQ-R14+ (response rate 71%). Forced expiratory volume in 1 s in percentage of predicted (FEV1%) and body mass index (BMI) were included as measures of health status.

Results: Internal consistency coefficients ranged from 0.54 to 0.95. Eight out of the twelve scales had alpha coefficients above 0.7. Test–retest correlations ranged from 0.42 to 0.88 and they were significant in eight scales. All the CFQ-R+14 scales except the digestive symptoms scale discriminated significantly (p<0.05) between patients with mild, moderate, and severe disease. Nine out of the twelve scales discriminated significantly (p<0.05) between nourished (BMI ≥19) and malnourished (BMI <19) patients. Significant differences between participants and non-responders were found for age, sex and FEV1 (higher age, more males and lower FEV1 among non-responders). All of the scales met standards for floor effects (<15% of the responders with the lowest score) but five of the scales failed to meet standards for ceiling effects (>15% of the responders with the highest score).

Conclusion: The Danish CFQ-R14+ is a reliable and valid instrument for measuring the health-related quality of life in Danish adolescents and adults with CF, though with the exception from a few of its subscales.

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1. Introduction

Cystic Fibrosis (CF) is a chronic, progressive and fatal disease. Life expectancy has increased substantially in the recent decades due to early diagnosis and improved treatment [1]. CF affects many organs; the burden of treatment is heavy for the patients and for most the burden grows with age. Traditional measures of clinical status such as lung function and body mass index do not capture all the aspects of the disease [2]. Health-related quality of life (HRQOL) questionnaires evaluate the impact of a disease on the patient’s daily life rather than just the physical status. The HRQOL questionnaires are patient centered and provide information about the patient’s own assessment of his physical, functional, social, emotional and psychological condition as well as his daily functioning and well-being. This information is essential when assessing the impact of a chronic disease on a patient’s life, in the evaluation

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of new treatments, and in the decision making concerning treatment and care. They ought to be included routinely in clinical trials and in medical cost benefit analyses. However, the questionnaires on quality of life are not sensitive enough and not designed to capture disease-specific problems [3–9]. About ten years ago disease-specific quality of life questionnaires on other chronic diseases such as asthma and diabetes were developed [10,11]. In 1997, a CF-specific quality of life questionnaire was developed in France (CFQ) [12] and in 2000 it was translated and validated in English and then modified (CFQ-R) [7]. In 2000 another questionnaire was developed in the United Kingdom (CFQol) [5]. The advantage of the CFQ-R is that versions of the questionnaire for children with CF from the age of six and their parents were developed, which provided the opportunity to conduct long-term studies. A patient’s quality of life evolves over time due to life events, illness progress, coping abilities, development of treatment, and cultural changes [13]. Therefore, it is important to monitor the patient’s quality of life over time. A specific tool like the CFQ-R can provide a broader assessment of the CF patient’s life situation and can be very useful in the collaboration between the patient and the CF team.

The revised cystic fibrosis quality of life questionnaire for adolescents and adults (CFQ-R14+) [14] is now translated into several languages [15] and widely used. It allows cross-cultural studies [16] and multicentre studies where quality of life is often used to assess health care outcome in addition to conventional measures. Only a few of the translated CFQ-R14+ questionnaires have been psychometrically validated and published [14,17,18]. The validation of the American, the Dutch and the German versions all demonstrated that the CFQ-R14+ is a reliable and valid tool for measuring HRQOL in CF patients. They demonstrated Chronbach’s_α_ (reliability) as follows: American: 0.67–0.94, German: 0.71–0.94, Dutch: 0.43–0.92 and test–retest stability as follows: American: 0.45–0.90, Dutch: 0.72–0.98. All of the studies showed good differentiation between mild, moderate and severe disease.

The aim of the current study was to assess the validity and reliability of the Danish version of the CFQ-R+14.

2. Methods

2.1. Measures

The CFQ-R+14 consists of 49 self-reported items within twelve domains: physical functioning [8], vitality [4], emotional functioning [5], eating disturbances [3], treatment burden [3], general health perception [3], social functioning [6], body image [3], role limitations [4], weight problems [1], respiratory symptoms [6] and digestive symptoms [3]. The possibilities for answer are on a four point scale rating frequency, difficulty or truth and selecting one out of four statements that would best describe the patient’s situation. The scores range from 0 to 100 and the higher the score, the higher is the patient’s quality of life.

A method of assessing whether measures can differentiate between patients with various degrees of disease severity is to divide the patients on the basis of percentage of predicted forced expiratory volume in 1 s (FEV,%) [5]. The patients were divided into three disease severity groups: mild (FEV,_% ≥ 71), moderate (FEV,_% 41–70) and severe (FEV,_% ≤ 40). Since severity of the disease is related to older age the patients were divided into age groups: adolescents (14–17 years), young adults (18–25 years) and adults (>25 years). As nutritional status might play a role in the evaluation of the disease severity of the CF patient, the patients were also categorized according to their nutritional status (BMI ≥ 19=nourished, BMI < 19=malnourished) [17,19].

2.2. Translation

Linguistic validation was made in accordance with international guidelines [20] and the specific recommendations for the CFQ-R+14 [7]. At first, two independent translators made a forward translation from English to Danish. Two CF pediatricians reviewed these translations and the translations were merged to one raw translation. Two individual translators made a backward translation from Danish to English and it was compared with the original English version of the questionnaire. The second raw translation into Danish was made. Several adult CF patients critically read the second version of the raw translation and after a few adjustments were made to form the final Danish CFQ-R+14 aimed at teenagers and adults.

2.3. Participants and procedures

The CFQ-R+14 was mailed to all Danish CF patients above the age of 13 (N=278) in August 2006. A total of 196 patients with a confirmed diagnosis of CF from the two Danish CF centers filled in the CFQ-R+14. The response rate was 71%. A group of clinically stable patients with no change in their basic treatment (N=14) completed the questionnaire again 10–14 days later. Mean age of the participants was 26 years (range 14–52) and 53.1% of the participants were female (104 females, 92 males).

Data on BMI and FEV,_% were extracted from the national CF registry. Sixteen of the participants were left out of the validity calculations because their data from the national CF registry were too old (>6 months). Mean number of days between the clinical data and the CFQ-R+14 was 16 days (range 0–182).

A total of 180 participants had relevant data from the CF registry. Their mean FEV,_% was 71.9% predicted (SD 25.4, range 24.7–132.1%), (mild N=95, moderate N=60 and severe N=24).

Mean BMI was 20.9 (SD 3.3, range 13–35), (nourished N=131 and malnourished N=49). Mean age was 25 years (SD 8.5, range 14–48) (40 adolescents, 56 young adults and 84 adults); 53.9% were female (97 females, 83 males). There were no significant differences between participants and non-responders concerning BMI (participants M=20.9, SD=3.3; non-responders M=20.9, SD=3.6; t=−0.08, p=0.94). There were significant differences in age (participants: M=25.5, SD=8.5; non-responders M=27.9, SD=10.0; t=2.00, p=0.047) and FEV,_% (participants M=71.9, SD=25.4, non-responders M=63.8, SD=26.3;
Most of the non-responders were male (70.7%) (Pearson Chi-Square 12.77, \( p < 0.05 \)).

2.4. Ethics

In accordance with the regulations of The National Danish Ethics Committee, questionnaire-based projects do not have to be notified. The project is registered at The Danish Data Protection Agency.

3. Statistical analyses

3.1. Patients

The reliability calculations were made on the sample of 196 participants filling in the CFQ-R14+ questionnaire. The validity calculations were made on the sample of 180 participants who had relevant data from the CF registry and had filled in the CFQ-R14.

Independent sample \( t \)-tests were conducted to test for differences in age, BMI and FEV1. A chi-square test for independence was conducted to explore the relationship between sex and responder/non-responder.

3.2. Reliability

The internal consistency or reliability of each scale was calculated using Chronbach’s \( \alpha \) coefficient. The purpose was to assess the association between the items and their scales, respectively. A minimum level of 0.7 is recommended [21].

Test–retest reliability was tested on a medically stable group of patients (\( N = 14 \)) using Spearman’s correlation. Their mean age was 27 years (SD 9.4, range 17–42), mean BMI 20.4 (SD 2.5, range 15–24) and mean FEV1 % was 78.6 (SD 32.3, range 18.7–103.0); 50% were males. The purpose was to assess the stability of the scores over time. For test–retest with one to two weeks interval a correlation higher than 0.80 suggests adequate stability [22].

3.3. Construct validity

Independent sample \( t \)-tests were conducted to test whether the scale scores could differentiate between sex and BMI groups. One-way analyses of variance (ANOVA) between groups were conducted to test whether the scale scores could differentiate between age groups and disease severity groups.

4. Results

4.1. Reliability

The internal consistency or reliability of each scale is shown in Table 1. The Chronbach’s \( \alpha \) coefficients ranged from 0.54 to 0.95 with the majority of the coefficients >0.70 (Table 1). The test–retest reliability with Spearman’s correlation ranging from 0.42 to 0.88 is also shown in Table 1.

Floor and ceiling effects were analyzed. The majority of the responses were in the mid-range. All of the scales met standards for floor effect but five of the scales failed to meet standards for ceiling effect (Table 1).

4.2. Construct validity

To test whether the CFQ-R14+ scales could differentiate between participants concerning disease severity, patients were divided into three groups according to pulmonary function — mild disease severity (\( N = 95 \)), moderate disease severity (\( N = 60 \)) and severe disease severity (\( N = 24 \)). Differences between the three disease severity groups are shown in Table 2. In all the CFQ-R=14 scales, except for the scale of digestive symptoms higher scores were associated with milder disease severity.

Table 1
Internal consistencies (Chronbach’s \( \alpha \)), test–retest (Spearman’s correlation) and ceiling effects on the CFQ-R14+

<table>
<thead>
<tr>
<th>No of items</th>
<th>Chronbach’s ( \alpha ) coefficient</th>
<th>Test–retest Spearman’s correlation</th>
<th>Sig. value (2-tailed)</th>
<th>Floor effects (% min score of 0)</th>
<th>Ceiling effects (% max score of 100)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Physical functioning</td>
<td>8</td>
<td>0.95</td>
<td>0.68</td>
<td>0.008 *</td>
<td>1.5</td>
</tr>
<tr>
<td>Role limitations</td>
<td>4</td>
<td>0.81</td>
<td>0.52</td>
<td>0.068</td>
<td>1.0</td>
</tr>
<tr>
<td>Vitality</td>
<td>4</td>
<td>0.78</td>
<td>0.53</td>
<td>0.052</td>
<td>2.1</td>
</tr>
<tr>
<td>Emotional functioning</td>
<td>5</td>
<td>0.76</td>
<td>0.84</td>
<td>0.00 *</td>
<td>0.0</td>
</tr>
<tr>
<td>Social functioning</td>
<td>6</td>
<td>0.54</td>
<td>0.42</td>
<td>0.139</td>
<td>0.0</td>
</tr>
<tr>
<td>Body image</td>
<td>3</td>
<td>0.67</td>
<td>0.45</td>
<td>0.11</td>
<td>2.6</td>
</tr>
<tr>
<td>Eating disturbances</td>
<td>3</td>
<td>0.91</td>
<td>0.88</td>
<td>0.00 *</td>
<td>1.5</td>
</tr>
<tr>
<td>Treatment burden</td>
<td>3</td>
<td>0.72</td>
<td>0.70</td>
<td>0.006 *</td>
<td>1.0</td>
</tr>
<tr>
<td>Health perceptions</td>
<td>3</td>
<td>0.87</td>
<td>0.84</td>
<td>0.00 *</td>
<td>3.6</td>
</tr>
<tr>
<td>Weight problems</td>
<td>1</td>
<td>0.83</td>
<td>0.70</td>
<td>0.00 *</td>
<td>9.9</td>
</tr>
<tr>
<td>Respiratory symptoms</td>
<td>6</td>
<td>0.84</td>
<td>0.70</td>
<td>0.007 *</td>
<td>0.5</td>
</tr>
<tr>
<td>Digestive symptoms</td>
<td>3</td>
<td>0.64</td>
<td>0.61</td>
<td>0.028 **</td>
<td>0.0</td>
</tr>
</tbody>
</table>

* Correlation is significant at the 0.01 level (2-tailed).
** Correlation is significant at the 0.05 level (2-tailed).
Comparisons were made between nourished \((N=131)\) and malnourished \((N=49)\) participants (Table 3). The nourished group scored significantly higher on all the CFQ-R+14 scales except on the scales of vitality, social functioning and digestive symptoms. On these scales there was a tendency towards higher CFQ-R+14 scores in the nourished group.

Comparisons were made between males \((N=83)\) and females \((N=97)\) and no significant differences in scores were found in any of the scales.

Participants were divided in age groups: adolescents \((N=40)\), young adults \((N=56)\) and adults \((N=84)\). We found no significant age-related differences.

5. Discussion

The aim of the current study was to assess the validity and reliability of the Danish version of the CFQ-R+14. Our results indicate that the CFQ-R+14 for most scales is a reliable and valid tool to measure health-related quality of life in adolescents and adults with CF.

Comparisons of participants with non-responders showed no significant differences in BMI but significant differences in FEV\(_1\), sex and age. The non-responders were older, there were more males and FEV\(_1\) was lower. The difference in sex is common and the difference in age might be explained by the possibility that the parents remind the younger patients to fill in the questionnaire. The difference in FEV\(_1\) could be explained by the fact that it requires some energy to face the problems and the limitations that some patients with low FEV\(_1\) might have. It must be considered as a weakness in this study. Comparison with similar studies was not possible because of lack of data on the non-responders.

The internal consistency was good with the majority of the scales being higher than 0.7 (Chronbach’s \(\alpha\)), two of the scales were slightly lower (body image 0.67, digestive symptoms 0.64) and the scale on social functioning was 0.54. Our results are comparable with the results of other studies\[14,17,18\] though the scale of social functioning was higher than 0.6 in those studies.

Eight of the scales did not show adequate test–retest stability. The test–retest reliability is especially important when a scale is used to assess the progress of treatment. If a scale is not stable, then it is impossible to determine whether measured change is real or represents random error in the scale\[22\]. The poor

### Table 3

Mean scales scores on CFQ-R14+ compared to nutritional status

<table>
<thead>
<tr>
<th></th>
<th>Nourished BMI ≥ 19 (mean±S.D.)</th>
<th>Malnourished BMI &lt; 19 (mean±S.D.)</th>
<th>(t)-value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Physical functioning</td>
<td>79.9±22.1</td>
<td>65.2±33.2</td>
<td>-2.86 *</td>
</tr>
<tr>
<td>Role limitations</td>
<td>84.3±16.1</td>
<td>72.0±24.9</td>
<td>-3.18 *</td>
</tr>
<tr>
<td>Vitality</td>
<td>60.2±18.6</td>
<td>54.3±23.3</td>
<td>-1.78</td>
</tr>
<tr>
<td>Emotional functioning</td>
<td>76.0±19.2</td>
<td>69.2±20.2</td>
<td>-2.04 **</td>
</tr>
<tr>
<td>Social functioning</td>
<td>75.4±15.4</td>
<td>70.4±15.9</td>
<td>-1.94</td>
</tr>
<tr>
<td>Body image</td>
<td>81.9±19.3</td>
<td>56.7±28.5</td>
<td>-5.71</td>
</tr>
<tr>
<td>Eating disturbances</td>
<td>89.5±21.5</td>
<td>80.6±25.9</td>
<td>-2.14 **</td>
</tr>
<tr>
<td>Treatment burden</td>
<td>58.7±21.9</td>
<td>49.7±26.5</td>
<td>-2.11 **</td>
</tr>
<tr>
<td>Health perceptions</td>
<td>63.0±26.3</td>
<td>51.9±30.5</td>
<td>-2.40 **</td>
</tr>
<tr>
<td>Weight problems</td>
<td>87.1±26.1</td>
<td>54.6±38.3</td>
<td>-5.37 *</td>
</tr>
<tr>
<td>Respiratory symptoms</td>
<td>71.1±19.9</td>
<td>62.0±24.1</td>
<td>-2.35 **</td>
</tr>
<tr>
<td>Digestive symptoms</td>
<td>74.4±18.5</td>
<td>73.3±19.5</td>
<td>-0.35</td>
</tr>
</tbody>
</table>

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

** Correlation is significant at the 0.05 level (2-tailed).
values could be explained by a small number of retests (N=14) or of the possibility that the patients were not clinically stable in spite of no change in their basic treatment.

Floor effects were small, but five of the twelve scales failed to meet standards for ceiling effects. Thus the scales of physical functioning, role limitations, body image, eating disturbances and weight problems had poor sensitivity for change and for patients scoring at the top of the scales. In the American validation high ceiling effects were found on three of those scales (eating disturbances 60.6%, body image 28.8% and physical function 19.7%). This might indicate that an alteration of these scales should be considered.

Similar to previous studies [14,16] we found that quality of life measured by the CFQ-R+14, except from few of the subscales was, significantly associated with FEV₁ and BMI. This is an indication of the construct validity of the CFQ-R+14 in a Danish context. The lack of association between the subscales of digestive symptoms and disease severity was expected. Patients with low FEV₁ might have no digestive problems and patients with high FEV₁ might have significant digestive problems.

Contrary to what could be expected and findings of others [14,18], we found no differences in CFQ-R+14 between age groups although disease is progressing with age. The reason might be that the oldest patients are not necessarily the most severely ill. Among the patients in our study 46.7% (N=84) were >25 years, but only 13.4% (N=24) of the patients had severe disease (FEV₁ ≤ 40% of predicted).

Gender differences in morbidity and mortality between male and female CF patients are well-documented [24,25] but our study did not corroborate these findings. Females demonstrated lower, though not significant, CFQ-R+14 scores than males in all scales except respiratory symptoms, body image and weight. This is consistent with other studies [26,27] where females demonstrated higher satisfaction with low weight than males. This is consistent with other studies [26,27] where females demonstrated higher satisfaction with low weight than males.

In conclusion the internal reliability of the Danish CFQ-R+14 was acceptable in all except three subscales. When conducting intervention studies the low test–retest correlations of four subscales have to be taken into consideration, and the sensitivity of the eating disturbances and weight problem subscales need to be further investigated.

The CFQ-R+14 can be used for many purposes. Firstly, it ought to be used in the investigation of any new treatment. Secondly, it could be used in daily clinical practice as a tool to decide on the most important topics for the patient to discuss with the physician. The CFQ-R+14 could be used routinely in connection with outpatient visits; the physician could use the patient’s CFQ-R+14 answers as a starting point for the communication with the patient. Thirdly, the CFQ-R+14 could be used to follow the patients’ quality of life over time. This would, however, require a validation of the CFQ-R for children and their parents; a process we hope to initiate in the immediate future.

Acknowledgments

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References


