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BRIEF COMMUNICATION

Reliability and validity of the Cystic Fibrosis Questionnaire-Revised for children and parents in Turkey: cross-sectional study

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Abstract
Purpose The purpose of study was to translate Cystic Fibrosis Questionnaire-Revised (CFQ-R) into Turkish for children with cystic fibrosis (CF) and evaluate its reliability and validity. This is the first CF-specific health-related quality of life (HRQOL) measure validated in a Muslim country.
Methods Fifty-one children aged 6–13 years treated at four centers in Turkey and 30 parents participated in this cross-sectional study. Demographic characteristics and disease severity parameters were recorded for all participants. All participants completed the parent or child versions of CFQ-R and KINDL questionnaires at enrollment. Reliability and construct validity analysis were carried out.
Results Both children and parents endorsed a range of responses, with no evidence of floor or ceiling effects. Item-to-total correlations indicated that most items were more highly correlated with their intended scale than competing scales. Good internal consistency was found for majority of child and parent scales. CFQ-R scales correlated significantly with clinical indices of disease severity. Good evidence of convergent validity with a generic HRQOL scale was found.
Conclusion Turkish versions of CFQ-R Child and Parent instruments have demonstrated adequate reliability and validity and can be utilized in clinical trials or integrated into clinical evaluation and follow-up of Turkish children with CF.

Keywords Cystic Fibrosis Questionnaire-Revised · Cystic fibrosis · Validity · Reliability

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Background

It is critical to evaluate the impact of cystic fibrosis (CF) and its treatment on patients' health-related quality of life (HRQOL) [1, 2]. HRQOL measures, which fall under the category of patient-reported outcomes, are now being used in descriptive studies, as well as clinical trials [3–7]. Evaluation of HRQOL in patients with CF is important because it adds unique information that is not captured by traditional clinical parameters. In addition, as patients' disease trajectories improve, these traditional outcome measures may not be sensitive indicators of the effects of new treatments [8].

The Cystic Fibrosis Questionnaire-Revised (CFQ-R) is the most widely used HRQOL instrument for CF [1, 9]. No reliable and valid HRQOL instrument for patients with CF

in Turkey or any other Muslim country currently exists. Further, European Cystic Fibrosis Society (ECFS) is currently establishing a clinical trials network, with a focus on improving the health outcomes of patients with CF across Europe, making it essential to have measures that can be used in all of these countries. Thus, the major aim of this study was to translate the CFQ-R into Turkish for children with CF and their parents, and perform an initial evaluation of the instrument's psychometric properties.

Methods

Subjects and study design

The CFQ-R questionnaires for children and parents were translated into Turkish and then validated in a multicenter cross-sectional study [10]. Children with CF presenting to the centers were enrolled consecutively during the study period. Demographic characteristics and clinical indicators of disease severity were recorded. Clinical severity parameters recorded included number of hospitalizations due to a CF-related health problem, number of exacerbations related pulmonary findings that required treatment, and number of presentations to the outpatient department or emergency room due to a CF-related health problem that was not routine other than routine control during the previous 1-year period. Data on clinical severity parameters were collected over a 1-year period from a systematic chart review.

All children completed age-appropriate CFQ-R and a general HRQOL measure, the questionnaire for measuring HRQOL in children and adolescents (KINDL). Parents completed CFQ-R Parent version. All instruments were completed during a visit to the department under the supervision of the pediatrician involved in the study.

The study was approved by the Institutional Ethics Board of Celal Bayar University and informed consent was obtained from participants.

Cystic Fibrosis Questionnaire-Revised questionnaire

The CFQ-R is a disease-specific HRQOL questionnaire [2, 11]. CFQ-R Child version consists of 35 questions on the following scales: Physical Functioning, Emotional Functioning, Social Functioning, Body Image, Eating Disturbances, Treatment Burden, and Respiratory and Digestive symptoms. The CFQ-R Parent version is a self-report instrument composed of 44 items grouped into 11 domains that assess Physical Functioning, Emotional Functioning, Vitality, School Performance, Eating Disturbances, Body Image, Treatment Burden, Health Perceptions, Respiratory and Digestive Symptoms, and Weight. Both instruments have well-established reliability and validity [11, 12]. Items for both child and parent CFQ-R are rated according to a 4-point scale. Higher scores indicate better HRQOL.

KINDL questionnaire

The KINDL Questionnaire is a generic HRQOL [13–15]. It consists of 24 items rated on Likert-type scales evaluating six domains of HRQOL: physical well-being, emotional well-being, self-esteem, family, friends, and everyday functioning.

Translation of CFQ-R's into Turkish

The translation process consisted of initial forward translation from English into Turkish by two independent, native Turkish speakers. Translations were discussed with the developer of CFQ-R in several conference calls and "consensus" translation in Turkish was developed. Next, Turkish version was back-translated into English by two independent translators, followed by discussion with the developer. Minor revisions were made to the Turkish versions of CFQ-R. Finally, cognitive debriefing was performed [16].

Statistical analysis

Statistical analyses were performed using SPSS 13.0 (Chicago, IL, USA), with p values less than 0.05 considered statistically significant. Kruskal–Wallis test and Mann–Whitney U and student's t test were used to compare CFQ-R scores in different sociodemographic groups.

To assess reliability, we calculated item-to-total correlations using Pearson correlations; we evaluated internal consistency using Cronbach's alpha. We assessed construct validity by examining correlations between CFQ-R and KINDL, as well as between CFQ-R scores and clinical indicators.

Results

Sociodemographic characteristics of the participants

A total of 51 children with CF and 30 parent caregivers participated. Mean age of children was 9.8 years $\pm$ 2.6 (Table 1).

Few gender differences were found on the CFQ-R domain scores; girls scored significantly higher on Emotional Functioning, indicating better functioning, than boys (78.7 $\pm$ 13.9 vs 68.4 $\pm$ 11.9, $p = 0.003$).

Mean age of parents was 35.5 $\pm$ 5.3 years, with a range of 26–47. Most parents were mothers (83.3 %) (Table 1).

Table 1 Sociodemographic characteristics of the children and parents enrolled in the study

Variable	N	%
Children		
Age		
Child CFQ-R 6–13 years		
Mean ± SD	9.8 ± 2.6	
Median	10	
Gender		
Child CFQ-R 6–13 years		
Female	25	49
Education[a]		
Child CFQ-R 6–11 years		
Not attending school	5	18.5
Kindergarten	5	18.5
Grade 1	2	7.5
Grade 2	4	14.8
Grade 3	8	29.6
Grade 5	3	11.1
Child CFQ-R 12–13 years		
Not attending school	2	8.3
Grade 6	14	58.3
Grade 7	6	25.0
Grade 8	2	8.4
Parents		
Age		
Mean ± standard deviation	35.5 ± 5.3	
Min–max	26.0–47.0	
Gender		
Female	25	83.3
Education (last graduated school)		
Less than high school	25	83.3
High school diploma	1	3.3
Technical school	1	3.3
University–College	3	10.1
Occupation		
Looking for job	1	3.3
Housewife	21	70.0
Working full/part time	7	23.4
Cannot work for other reasons	1	3.3

[a] Education of the children enrolled in the study who filled in CFQ-R 6–11 and 12–13 years

There were no significant associations between age of the child or parent and CFQ-R scores (p's > 0.05). CFQ-R scores did not change significantly in relation to level of parental education (p's ≥ 0.05). Parents working outside the home scored significantly higher (68.6, SD = 9.2) than non-working parents (52.2, SD = 21.6) on Vitality scale ($p = 0.03$).

Reliability analysis

There was no evidence of floor or ceiling effects on any scales (defined as <10 or >90 %). Item-to-total correlations supported the item structure of the CFQ-R, with a majority of items correlating more highly with their intended scale (Table 2). Two items on the CFQ-R Child version demonstrated slightly lower correlations: item 13 ($r = 0.29$) and item 21 ($r = 0.33$). For the CFQ-R Parent version, all items correlated better with their intended scale than competing scales.

Internal consistency was adequate for most of CFQ-R Child scales (Cronbach's alpha above 0.60) exception for Treatment Burden (alpha = 0.55) (Table 2) [17]. Good internal consistency was also found for all CFQ-R Parent Scales (alphas 0.63–0.88), except School Functioning (alpha = 0.37) (Table 2).

Validity analysis

As hypothesized, scores on the CFQ-R Child instrument were correlated with several clinical parameters in the expected direction; children with lower lung function, for example, reported more Respiratory Symptoms (Table 3). For the CFQ-R Parent version, significant correlations were also found between clinical indices of disease severity and the CFQ-R scores (Table 3).

In terms of convergence between the CFQ-R, which is a disease-specific instrument, and the KINDL, scores on the Physical Functioning scale of the CFQ-R were correlated with all domains of KINDL except the School domain (r's > 0.31, $p < 0.03$ for all scales; $r = 0.27$ $p = 0.08$ for KINDL school domain). Emotional Functioning on both scales was significantly correlated ($r = -0.57$, $p < 0.001$) and Emotional Functioning scale of the CFQ-R was also significantly correlated with physical, self-esteem, friends, and chronic disease domains of KINDL ($r = 0.52$ $p < 0.001$, $r = 0.41$ $p = 0.003$, $r = 0.39$ $p = 0.005$, $r = 0.38$ $p = 0.008$, respectively). The CFQ-R scales, including Eating Disturbances, Treatment Burden, Respiratory Symptoms, and Digestion, were all significantly correlated with physical domain of KINDL ($r > 0.38$ $p < 0.008$ for all), demonstrating good convergent validity (Table 4).

Discussion

This is the first study to translate and evaluate the psychometric properties of the CFQ-R in a predominantly Muslim country. It is critically important to have reliable and valid disease-specific HRQOL instruments for all European countries, as efforts are being made to develop a

Table 2 Descriptive statistics, corrected item total correlations, and Cronbach alpha values for CFQ-R child and parent instruments

Domain [a]	Mean	Median	SD[b]	Min–max	R[c] (Min–max)	Cronbach alpha
CFQ-R child version						
Physical functioning (6)	50.5	50.0	25.5	5.6–94.4	0.25–0.72	0.78
Emotional functioning (8)	73.4	70.8	13.8	50.0–100.0	0.14–0.51	0.64
Social functioning (7)	56.2	57.1	19.1	23.8–100.0	0.38–0.60	0.69
Body image (3)	64.1	66.7	31.9	0.0–100.0	0.20–0.52	0.55
Eating disturbances (3)	57.3	55.6	29.8	0.0–100.0	0.42–0.63	0.70
Treatment burden (3)	70.8	77.8	23.8	11.1–100.0	0.44–0.56	0.72
Respiratory symptoms (4)	70.3	66.7	22.6	25.0–100.0	0.11–0.71	0.60
Digestion (1)	77.8	66.7	27.2	0.0–100.0	–	–
CFQ-R parent version						
Physical functioning (9)	53.4	55.6	28.4	3.7–100.0	0.43–0.79	0.88
Emotional functioning (8)	59.1	60.0	22.9	20.0–100.0	0.25–0.51	0.63
Vitality (5)	56.1	53.3	20.9	13.3–100.0	0.37–0.58	0.68
School performance (3)	55.9	66.7	28.9	0.0–100.0	0.10–0.33	0.37
Eating disturbances (2)	36.2	33.3	33.9	0.0–100.0	0.53–0.54	0.69
Body image (3)	50.6	55.6	36.1	0.0–100.0	0.40–0.70	0.73
Treatment burden (3)	45.6	44.4	31.9	0.0–100.0	0.48–0.68	0.75
Health perceptions (3)	64.8	77.8	27.4	11.1–100.0	0.43–0.85	0.78
Respiratory symptoms (6)	65.5	72.2	27.2	5.6–100.0	0.53–0.80	0.86
Digestion (3)	83.9	88.9	22.9	11.1–100.0	0.75–0.77	0.87
Weight (1)	37.9	33.3	34.2	0.0–100.0	–	–

[a] Number of items

[b] Standard deviation

[c] Corrected item total correlations

European Clinical Trials Network to evaluate efficacy of new treatments and conduct comparative efficacy studies. Therefore, availability of a validated HRQOL measure will enable CF Centers and clinicians to better track outcomes of their patients.

This study demonstrated that Turkish CFQ-R yields a good range of scores, with no evidence of floor or ceiling effects. Items fit better with their assigned scales than competing scales and internal consistency of both the CFQ-R Child and Parent versions was acceptable. The only scale that did not demonstrate good internal consistency was CFQ-R Parent School Functioning scale which may be due to the low percentage of children attending school. Cronbach's α coefficients for Danish CFQ-R have been reported to range between 0.54 and 0.95, with a majority of the coefficients above 0.70. Despite the similar range of internal consistency coefficients between the Danish and Turkish versions of the CFQ-R, generally higher coefficients were found in the Danish version [18]. On the other hand, Cronbach's α coefficients for child version of the German CFQ-R are closer to the Turkish values being reported [19]. This result highlights the potential differences in both the daily functioning and health outcomes of children with CF in less industrialized countries.

Strong evidence of convergent validity was found both in terms of associations between CFQ-R and a generic quality of life measure and children's health outcomes. Specifically, good convergent validity was found between the CFQ-R scores and the KINDL, a generic QoL measure, especially in domains of functioning that were similar (e.g., emotional functioning). In contrast, no associations were found between the CFQ-R Physical Functioning domain and the similarly named KINDL scale. This is likely because they ask about different physical abilities. Interestingly, significant correlations were found between CFQ-R scales that reflect physical aspects of the disease, such as respiratory symptoms and digestion, and these were significantly associated with the physical domain on the KINDL. This difference in associations may reflect the generic nature of KINDL in comparison with a disease-specific tool like the CFQ-R. Generally, the results supported the construct validity of the instrument.

Further, significant associations were found between several key CFQ-R scales (Physical Functioning, Respiratory Symptoms, Social Functioning, Treatment Burden) and clinical parameters reflecting disease severity.

New developments in the treatment of CF over the past 20 years have led to longer life spans but an increase in

Table 3 Correlations between CFQ-R child and parent versions and clinical parameters

Clinical parameter	Hospitalizations[a] *r*	Exacerbations[b] *r*	Sick visit[c] *r*
CFQ-R child version			
Physical functioning	**−0.44**	**−0.34**	**−0.41**
Emotional functioning	−0.25	**−0.29**	−0.23
Social functioning	**−0.54**	−0.21	**−0.38**
Body image	0.08	−0.05	−0.12
Eating disturbances	−0.21	0.002	−0.18
Treatment burden	**−0.39**	0.02	−0.24
Respiratory symptoms	−0.30	−0.18	−0.19
Digestion	−0.18	−0.14	−0.42
CFQ-R parent version			
Physical functioning	**−0.54**	−0.25	−0.36
Emotional functioning	−0.31	−0.14	−0.37
Vitality	**−0.40**	−0.25	**−0.51**
School performance	**−0.45**	−0.33	−0.52
Eating disturbances	−0.24	−0.12	−0.26
Body image	−0.28	−0.08	0.33
Treatment burden	−0.31	−0.06	−0.38
Health perceptions	−0.34	**−0.43**	−0.27
Respiratory symptoms	**−0.63**	**−0.40**	**−0.51**
Digestion	−0.25	−0.02	−0.27
Weight	**−0.44**	−0.32	**−0.47**

Figures in bold indicate statistical significance

[a] Number of hospitalizations due to a CF-related health problem during the previous 1 year period

[b] Number of exacerbations in CF-related pulmonary findings that required treatment during the previous 1 year period

[c] Number of presentations to the outpatient department or emergency room due to a CF-related health problem other than routine control

Table 4 Correlations between CFQ-R Child Version and KINDL Total and Domain Scores

CFQ-R domains	KINDL domains							
	Physical role	Emotional role	Self-esteem	Family	Friends	School	Chronic disease	Total
Physical	**0.31**	**0.59**	**0.46**	**0.32**	**0.65**	0.27	**0.39**	**0.62**
Emotional	**0.52**	**0.57**	**0.41**	0.04	**0.39**	0.27	**0.38**	**0.52**
Social	**0.45**	**0.72**	**0.51**	**0.43**	**0.72**	0.19	**0.65**	**0.72**
Body	**0.41**	**0.49**	**0.44**	0.17	**0.44**	**0.29**	**0.33**	**0.54**
Eat	**0.57**	**0.65**	**0.38**	**0.29**	**0.42**	0.03	**0.55**	**0.55**
Treat	**0.38**	**0.61**	**0.43**	**0.37**	**0.60**	0.15	**0.58**	**0.59**
Respiratory	**0.47**	**0.51**	**0.63**	−0.14	**0.41**	**0.32**	**0.31**	**0.54**
Digestion	**0.67**	**0.64**	0.16	**0.39**	**0.65**	−0.06	**0.37**	**0.57**

Figures in bold indicate statistical significance

treatment burden [11]. This is one reason that patient-reported outcomes are needed. Moreover, clinical trial endpoints that reflect respiratory symptoms and daily functioning from the patient/caregiver perspective are needed. Reliable and valid HRQOL measures are also being used in clinical practice to monitor patient functioning, and this may become more important as comparative effectiveness trials are performed. HRQOL measures may provide crucial evidence on which treatments are most effective from the patient's perspective in relation to the burden of their administration. The CFQ-R has now been translated into 34 languages and is being used in many of the international clinical trials, including Turkey.

A major limitation of this study was the relatively small size of both the patient and parent samples. Turkey has a relatively small population of patients with CF (estimated at 1,000 across all ages) and major efforts were made to enroll participants across 4 CF Centers in Turkey. The other limitation is the absence of pulmonary function data, which was due to the young age of the children in this sample.

Future directions for the use of the CFQ-R in Turkey include the translation and validation of the teen/adult

version of the CFQ-R, development of a national CF registry to track patients' health outcomes, including HRQOL, and use of the CFQ-R to improve clinical care. In addition, the Turkish CFQ-R can now be incorporated into ongoing clinical trials in Turkey.

In conclusion, Turkish versions of the CFQ-R Child and Parent measures have demonstrated adequate reliability and good validity, both in terms of the constructs that were measured and their convergence with generic QoL measures and health outcomes. Incorporation of this instrument into patient care as well as clinical trials should facilitate steps toward improving the management and outcomes of these patients and families.

References

1. Goss, C. H., & Quittner, A. L. (2007). Patient-reported outcomes in cystic fibrosis. *Proceedings of the American Thoracic Society, 4*(4), 378–386.
2. Quittner, A. L., Modi, A. C., & Cruz, I. (2008). Systematic review of health-related quality of life measures for children with respiratory conditions. *Pediatrıc Respiratory Reviews, 9*(3), 220–232.
3. Gee, L., Abbott, J., Conway, S. P., Etherington, C., & Webb, A. K. (2000). Development of a disease specific health related quality of life measure for adults and adolescents with cystic fibrosis. *Thorax, 55*(11), 946–954.
4. Goldbeck, L., Schmitz, T. G., Henrich, G., & Herschbach, P. (2003). Questions on life satisfaction for adolescents and adults with cystic fibrosis: Development of a disease-specific questionnaire. *Chest, 123*(1), 42–48.
5. Henrich, G., Goldbeck, L., Schmitz, T. G., & Besier, T. (2010). Measuring life satisfaction in children and adolescents–psychometric properties of the "Questions on Life Satisfaction" FLZ(KJ). *Psychotherapie, Psychosomatik, Medizinische Psychologie, 60*(6), 219–226.
6. Retsch-Bogart, G. Z., Quittner, A. L., & Gibson, R. L. (2009). Efficacy and safety of inhaled aztreonam lysine for airway *Pseudomonas* in cystic fibrosis. *Chest, 135*(5), 1223–1232.
7. McCoy, K. S., Quittner, A. L., Oermann, C. M., Gibson, R. L., Retsch-Bogart, G. Z., & Montgomery, A. B. (2008). Inhaled aztreonam lysine for chronic airway *Pseudomonas aeruginosa* in cystic fibrosis. *American Journal of Respiratory and Critical Care Medicine, 178*(9), 921–928.
8. Quittner, A. L. (1998). Measurement of quality of life in cystic fibrosis. *Current Opinion in Pulmonary Medicine, 4*(6), 326–331.
9. Quittner, A. L., Sawicki, G. S., McMullen, A., Rasouiliyan, L., Pasta, D. J., Yegin, A., et al. (Epub ahead of print). Psychometric evaluation of the cystic fibrosis questionnaire-revised in a US national sample. *Quality of Life Research*.
10. Yuksel, H., Yilmaz, O., Eser, E., Quittner, A. L. (2007) Turkish translation and validation of revised cystic fibrosis questionnaire (CFQ-R) for children [abstract]. *Journal of Cystic Fibrosis, 6*(S1), S77.
11. Quittner, A. L., Buu, A., Messer, M. A., Modi, A. C., & Watrous, M. (2005). Development and validation of the cystic fibrosis questionnaire in the United States: A health-related quality-of-life measure for cystic fibrosis. *Chest, 128*(4), 2347–2354.
12. Modi, A. C., & Quittner, A. L. (2003). Validation of a disease-specific measure of health-related quality of life for children with cystic fibrosis. *Journal of Pediatric Psychology, 28*(8), 535–546.
13. Ravens-Sieberer, U., & Bullinger, M. (1998). Assessing the health related quality of life in chronically ill children with the German KINDL: First psychometric and content-analytical results. *Quality of Life Research, 7*(5), 399–407.
14. Erhart, M., Ellert, U., Kurth, B. M., & Ravens-Sieberer, U. (2009). Measuring adolescents' HRQoL via self reports and parent proxy reports: an evaluation of the psychometric properties of both versions of the KINDL-R instrument. *Health Qual Life Outcomes., 2009*(7), 77.
15. Eser, E., Yüksel, H., Baydur, H., et al. (2008) The psychometric properties of the new Turkish generic health-related quality of life questionnaire for children (Kid-KINDL). *Türk Psikiyatri Dergisi, 19*(4), 409–417.
16. Quittner, A. L., Sweeny, S., Watrous, M., et al. (2000). Translation and linguistic validation of a disease-specific quality of life measure for cystic fibrosis. *Journal of Pediatric Psychology, 25*(6), 403–414.
17. Ware, J. E., Brook, R. H., Ross Davies, A., Williams, K. N., Stewart, A. L., Rogers, W. H., et al. (1980). *Conceptualization and measurement of health for adults in the Health Insurance Study. Vol. I: Model of health and methodology. Doc. no. R-1987/1-HEW*. Santa Monica, CA: RAND Corporation.
18. Bregnballe, V., Thastum, M., Lund, L. D., Hansen, C. R., Preissler, T., & Schiøtz, P. O. (2008). Validation of the Danish version of the revised cystic fibrosis quality of life questionnaire in adolescents and adults (CFQ-R14+). *Journal of Cystic Fibrosis, 7*, 531–536.
19. Schmidt, A., Wenninger, K., Niemann, N., Wahn, U., & Staab, D. (2009). Health-related quality of life in children with cystic fibrosis: Validation of the German CFQ-R. *Health Qual Life Outcomes, 7*, 97.