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Original Article

Development and electronic validation of the revised Cystic Fibrosis Questionnaire (CFQ-R Teen/Adult)
New tool for monitoring psychosocial health in CF

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Abstract

Background: The Cystic Fibrosis Questionnaire-Revised (CFQ-R + 14) is a disease-specific, health-related quality of life instrument for cystic fibrosis (CF) patients ≥ 14 years. We have developed a Spanish electronic version of the CFQ-R (e-CFQ-R + 14 Spain). Our aim was to compare the paper and electronic versions and to validate the electronic version.

Methods: Fifty CF patients completed the study. All answered the paper and electronic versions on day 1 and repeated the e-CFQR version 15 days later.

Results: Concordance between the electronic and paper copy versions was high, with correlations above 0.9 in all domains. Test-retest reliability of the e-CFQ-R results was strong, with coefficients ranging from 0.8 to 0.9.

Conclusions: The e-CFQ-R version is reliable and valid and can replace the paper copy, thus simplifying the assessment of quality of life. It also provides immediate results with no errors in scoring. It is a useful new tool in CF care.

Keywords: Cystic Fibrosis Questionnaire Revised; Patient reported outcomes; Cystic fibrosis; Diagnostic tool

1. Introduction

Patient-reported outcomes (PROs) are questionnaires completed by patients that detect changes in symptoms, perceptions of health, and daily functioning as perceived by the patient. They range from a single-item rating of pain to multi-dimensional, health-related quality of life instruments. PROs are important endpoints in clinical trials of new medications and can serve as outcome measures in clinical research and to improve clinical

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Abbreviations: CF, Cystic fibrosis; CFQ-R+14 Spain, Revised Cystic Fibrosis Quality of Life Questionnaire in Adolescents and Adults. Spanish version; e-CFQ-R, Electronic Revised Cystic Fibrosis Quality of Life Questionnaire in Adolescents and Adults; HRQoL, Health-related quality of life.

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care. Both the U.S. Food and Drug Administration and the European Medicines Agency have formally recognized their importance [1,2], and thus, PROs are increasingly used as primary or secondary outcomes in the drug approval process [3]. The most appropriate PRO depends on its purpose (natural course of disease, efficacy of treatment).

Health-related quality of life (HRQoL) measurements in PROs are very important in cystic fibrosis (CF) because they allow the inclusion of the patient's perspective in research and clinical practice [4–8]. A validated CF questionnaire is the Cystic Fibrosis Questionnaire-Revised (CFQ-R). The CFQ-R was developed to evaluate the effects of the disease and its treatments on daily functioning in several domains (e.g., respiratory symptoms, physical functioning, and vitality) and has been used as an end-point for clinical trials of new medications [3,9–11]. Specific CFQ-R scales have been shown to predict pulmonary exacerbations and correlate with lung function, body mass index (BMI), and mortality [12–15]. Recently, the score of some scales of the CFQ-R has been used for the assessment of candidates for lung transplantation [16].

Despite the relevance of HRQoL measurements to monitor the impact of CF, the CFQ-R questionnaire is not routinely used in clinical practice in Europe. Among the reasons for this could be a lack of experience in the use of the questionnaire, the fact that it is time-consuming to analyze the data, there could be missing items in the paper copy, and the delay in receiving results in real time. These difficulties cause this valuable tool to be restricted to use in epidemiological studies and trials, and without a direct impact on daily clinical care [4,17]. Accordingly, an electronic version would help, and we have developed an electronic application of this questionnaire (e-CFQ-R+14). This version provides an automatic result in all domains, reduces data entry errors (which are common in the paper version) and the time required to enter and calculate the data [18]. The results are saved and stored in a secure database. The goal of our study was to evaluate the reliability and validity of the e-CFQ-R compared with the paper version.

2. Materials and methods

2.1. Description of the paper questionnaire

The Cystic Fibrosis Questionnaire-Revised Teen/Adult (CFQ-R+14) is a disease-specific, health-related quality of life instrument for adolescents and adults (≥14 years of age) with CF. The CFQ-R measures daily functioning from the patients' perspective, and thus provides unique information to facilitate clinical interventions. More broadly, it provides systematic data on the frequency and severity of respiratory symptoms that are not measured using conventional clinical tests [17].

The CFQ-R+14 has 12 scales or domains that evaluate functioning over the previous 2 weeks in the following domains or scales: physical functioning, role, vitality, emotional functioning, social, body image, eating disturbances, treatment burden, health perceptions, weight, respiratory symptoms, and digestive symptoms [19–22]. The CFQ-R+14 includes 50 items across 12 domains. The first 6 domains measure general aspects of HRQoL: 8 items for physical functioning; 4 for role functioning; 4 for vitality; 3 for health perceptions, 5 for emotional functioning, and 6 for social functioning. The other 6 domains measure symptoms and effects of the disease: 3 items for body image, 3 for eating disturbances, 3 for treatment burden, 1 for weight, 7 for respiratory symptoms, and 3 for digestive symptoms. Each domain is standardized on a 0–100 scale; higher scores indicate better HRQoL [23]. In the paper copy, scores for each domain can be calculated if at least two-thirds of the questions are completed [9,23].

The CFQ-R has been translated and validated in various languages, such as Latin-American Spanish and for the Spanish population from Spain [9].

2.2. Construction of the electronic version

The e-CFQ-R+14 is the electronic version of the paper questionnaire. Patients can easily complete the questionnaire in its electronic format. The electronic application is linked to an online database that can be adapted for any electronic device, such as tablets, phones, or computers. Once the questionnaire has been completed, this application calculates the results, sends them by email to the caregiver, and saves the data in a centralized and protected database. As soon as the patient completes the survey, the result is available.

The e-CFQ-R+14 web version and cross-platform (Windows, Linux, Android, IOS) has been developed using PHP programming, and is linked to a MySql database to record the calculated scores. It has been hosted at Strato, an ISO 27001-certified hosting provider, and is accessible through www.ecfqr.com, a domain specifically registered for this purpose. We have included two versions of the CFQ-R+14 in this program: English and Spanish. To access the English version, access this web address: http://www.ecfqr.com/en/ and for the Spanish version: http://www.ecfqr.com/. To grant privacy and security, access is restricted with an ID number and password, which is unique and personal for authorized staff.

We registered the www.ecfqr.com domain on 2015-11-26T00:00:00Z, where this application is hosted. By the end of 2015 and the beginning of 2016, we had performed the design and working tests on this electronic version to confirm its proper functioning.

2.3. Study procedure

Sixty patients with CF ≥14 years of age were consecutively enrolled in two CF adult units in Spain. At the end of the study, 50 patients had completed both questionnaires and repeated the electronic version 15 days later. The distribution was 24 patients in Málaga (Regional University Hospital) and 26 in Valencia (University Hospital La Fe). We used two centers to reduce single-center bias and to have a more heterogeneous sample. Recruitment took place during April and June of 2016. All the patients were in stable condition and completed the questionnaire before any other assessments to avoid bias (e.g., weight or pulmonary function test). On day 1, half the patients completed the paper version first and the other half started with the electronic version, but all the patients answered both formats on day 1. To

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