

The effect of telerehabilitation on quality of life, anxiety, and depression in children with cystic fibrosis and caregivers: A single-blind randomized trial

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Abstract

Background: Postural and aerobic exercises are essential in rehabilitation in cystic fibrosis (CF). The aim of this study is to examine the effect of telerehabilitation on the quality of life, depression, and anxiety levels of children with CF and their caregivers' mood and anxiety levels.

Materials and Methods: Patients between the ages of 6–13 with CF were randomized into two groups. Study group received an exercise program three times a week via Zoom for 12 weeks. Cystic Fibrosis Revised Questionnaire (CFQ-R), Anxiety and Depression Scale in Children-Revised (RCADS) were applied to the patients and State-Trait Anxiety Scale (STAI) and Beck Depression Inventory (BDI) were applied to the caregivers in the beginning and at the end of the program. Patients' FEV1 levels and 6-min walk tests were also measured.

Results: Twenty-eight patient-caregiver dyads, 14 dyads in each group, completed the study. The initial mean RCADS-Major depressive disorder score of the patients in the exercise group was 6.21 ± 3.11 , and this value decreased to 3.92 ± 3.79 at the end of the study and was significantly better ($p < 0.02$). A similar significant change was observed when the RCADS-generalized anxiety disorder score decreased from the initial mean level of 6.28 ± 2.81 to 3.42 ± 2.65 ($p < 0.01$). There were significant changes in improvement in the body image in telerehabilitation group. Similar significant changes were not observed in the control group. Caregivers' anxiety and depression levels did not change significantly.

Conclusion: A short-term telerehabilitation program improved patients' anxiety and depression levels, body image, and functional status. However, caregiver anxiety and depression levels did not change significantly.

KEYWORDS

anxiety, cystic fibrosis, quality of life, rehabilitation, telehealth

1 | INTRODUCTION

Cystic fibrosis (CF) is an autosomal recessive genetic disease that occurs because of the damage to the chlorine channel called Cystic Fibrosis Transmembrane Conductance Regulator (CFTR) synthesized from the mutated CF gene.¹ Although CFTR protein is found in many epithelial cell membranes, its effects primarily manifest in the lung and gastrointestinal systems. Disruption of chlorine transport through the damaged or absent CFTR channel increases Na⁺ absorption and water absorption of epithelial cells. As a result, the amount of secretions decreases while the mucus becomes denser and thicker. Thick mucus clogs the airways and disrupts the mucus clearance system, paving the way for respiratory tract infections.² Due to the obstruction of the pancreatic duct, malabsorption and damage to the pancreatic tissue are also observed.³ The upper respiratory tract, reproductive system, and biliary tracts may similarly be involved.⁴

CF is a disease that shortens life expectancy and inversely affects the quality of life. Because it is a chronic, fatal disease that affects many systems, it may impact mental health in children and families. Many studies have shown that children with CF and their families have higher levels of anxiety and depression compared to the general population.⁵ Previous research found that CF patients with depression were three times more likely to be hospitalized for pulmonary exacerbation and health care cost was five times higher than in patients who did not experience depression.⁶

Studies have shown that there is a strong correlation between anxiety and depression in individuals with CF and their health outcomes. Anxiety and depression symptoms were associated with worse quality of life, lower lung function, decreased physical function, and exacerbation of chest symptoms.⁷ Parental anxiety and depression may also increase the risk of depression and anxiety in the child and may negatively affect treatment adherence.⁸ Identifying and treating parental mental health problems may increase the child's adherence to treatment.

The lack of physical activity in children with CF may be related to psychosocial effects. The participation of healthy children in physical activities is related to their parents' views of their physical competence. Parents of children with CF often express their concerns about their children's participation in various sports activities. Still, when exercise is used as a way of treatment, the patient, family, the caregiver's perspective on exercise is more favorable than other treatment modalities.⁹ As exercise is seen to be safe and beneficial in CF, patient-appropriate exercise programs have become one of the focal points of CF physiotherapy. Still, physicians need to consider the risk of cross-infection when encouraging patients to exercise.¹⁰ It may be difficult for the patient to comply with a long-term physiotherapy program in the home environment. Failure to comply with the physiotherapy program results in more frequent hospital visits and an increased risk of potential infection. To prevent this situation and improve the patient's access to health services, telehealth services via videoconferencing can be used. Various studies continue to suggest that telemedicine is beneficial for home rehabilitation in chronic diseases.¹¹

The primary aim of this study is to examine the effect of telerehabilitation on the quality of life, anxiety, and depression levels of children with CF. Secondary purposes were to examine the change in levels of anxiety and depression in caregivers of CF patients undergoing telerehabilitation and compare them to caregivers of patients who did not receive any exercise regimen.

2 | METHODS

Among a total of 78 patient-caregiver dyads that were being followed in the pediatric pulmonology clinic, 30 of them were suitable for this study and accepted to participate. The study was conducted in our Physical Medicine and Rehabilitation and Pediatric Pulmonology departments. All patients and caregivers have given written and verbal informed consent. The study was approved by the local ethics committee and registered in ClinicalTrials.gov with the number of NCT04923438.

The inclusion criteria were being between the ages of 6–13 and having a genetically confirmed diagnosis of CF. The exclusion criteria were experiencing serious dyspnea with exercise, presence of an acute exacerbation, a recent history of musculoskeletal injury or surgery, the existence of previously diagnosed psychiatric problems, and the lack of access to the internet or required internet software. Patients previously diagnosed with mental health disorders including depression were excluded from this study considering the fact that they might be receiving other treatments that might interfere with the scores that are used in this study. On this basis, 4 patients were removed from the initial pool of 78 patients according to their medical records. The patients who were unable to perform at least two technically acceptable maneuvers were considered to be unsuccessful, and they were excluded from the study.

Fifteen of the patients were randomly assigned to the exercise group and 15 to the control group with the help of a computer-generated list. Zoom application was used for telerehabilitation study with children with CF three times a week for 12 weeks. The exercise program to be applied by each researcher to the patients they are responsible for is based on high-intensity interval training and postural strengthening and was prepared by a specialist physiotherapist and can be found as a Supporting Information file. A combination of high-intensity interval training in the form of a letter game and postural exercises was selected for telerehabilitation sessions. These exercises were selected so that they do not require any additional equipment; they are easy to implement in an online setting and easy to understand for a child. The parents only supervised the programs but did not actively participate. Five of the researchers were involved with the exercise sessions. Each researcher was able to provide the same exercise training to their patients by receiving the exact same training from the physiotherapist who planned the exercises before the beginning of the sessions. The researchers applying the programs were not physiotherapists themselves. All researchers had written instructions of the exercises before this meeting to familiarize

themselves with the concept and the content of the exercises. In this initial meeting, they also discussed coping methods and modifications when exercises cannot be performed for any reason. All researchers that applied the program were able to reach each other and the responsible physiotherapist on demand. The physiotherapist also participated in the first session of each patient to make sure that the exercises were done properly. The researchers that applied the exercise program were present during the whole session of the telerehabilitation and supervised and participated with the patient in person. A researcher blinded to the allocation of the patients evaluated the patients before and after treatment. All patients were encouraged to go on with their selected airway clearance technique, but the technique and its application were not monitored for this study.

For data collection, the questionnaires were applied to patients before and after video telerehabilitation were Cystic Fibrosis Revised Questionnaire (CFQ-R)¹² and Anxiety and Depression Scale in Children-Revised (RCADS).¹³ CFQ-R parent-report versions were used in retrieving quality of life data, since all children were between the ages of 6–13. The questionnaires that were applied to the caregivers were State-Trait Anxiety Inventory (STAI) and Beck Depression Inventory (BDI).^{14–16} Pulmonary function tests and the distance in the 6-min walking test of the patients before and after the study were also recorded.^{17,18} The details of the evaluation methods are given as an appendix.

2.1 | Statistical analyses

Sampling size calculations were done by using G power. There is no study in the literature that previously measured the effect of internet-based exercises performed by CF patients on anxiety. Therefore, a pilot study was performed with the five patients and five controls which revealed an effect size of 0.6 according to the change in RCADS. Therefore 26 patients were needed for the study to be at

90% power and an error margin of alpha 0.05. Considering the potential dropout rate of 10% in each group, 30 patients were recruited to the study. The distribution of the data was analyzed with Shapiro–Wilks's test, and it was found to be normal. Basic descriptive analyses were used for the calculation of frequencies, means, and standard deviations. For baseline differences between groups, independent samples *t*-test were used. Two-way mixed ANOVA was used for within-group and between-group analyses. A *p* value below 0.05 was considered significant since there were only a baseline and after-treatment measurement. All statistical analyses were done by SPSS version 20.0.

3 | RESULTS

Two of the 30 patients who participated in the study were excluded from the study because they could not continue the sessions during the study due to scheduling conflicts and one decided not to participate in this study further, withdrawing their consent. The final analysis was completed with 14 patients in the telerehabilitation group and 14 patients in the control group (Figure 1). At the beginning of the study, there were no significant differences between the telerehabilitation group and the control group in terms of age-sex, FEV1 levels, age and gender of the parents, and baseline criteria of all participants (Table 1).

When the changes in anxiety and quality of life of the patients were analyzed, the major depressive disorder subscale showed significant changes in the telerehabilitation group decreasing from 6.33 ± 4.36 to 3.92 ± 3.79 ($p = 0.02$) as shown in within-group analyses. A similar improvement was seen in the generalized anxiety disorder score, decreasing from 6.28 ± 2.81 to 3.42 ± 2.65 ($p = 0.001$). The major depressive disorder subscale of the control group was significantly higher than the telerehabilitation group at the end of the treatment with a score of 7.78 ± 3.04 ($p = 0.006$). Similarly, telerehabilitation group had significantly lower generalized anxiety scores at the end of

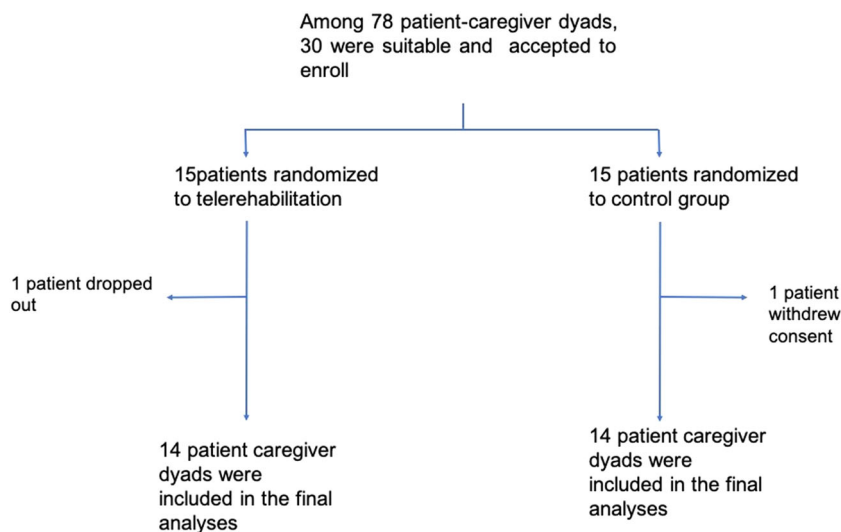


FIGURE 1 Patient flowchart [Color figure can be viewed at wileyonlinelibrary.com]

TABLE 1 Baseline group characteristics

	Telerehabilitation (n = 14)	Control (n = 14)	p
Mean age of the patient (years)	9.8 ± 2.14	10.0 ± 1.64	0.84
Number of female patients	n = 11 (78%)	n = 9 (64%)	0.70
Mean age of the caregiver (years)	37.14 ± 5.11	36.86 ± 4.94	0.88
Number of female caregivers	n = 13 (92%)	n = 14 (100%)	1.0
Baseline 6MWT (m)	431.78 ± 170.92	416.23 ± 115.30	0.77
Baseline FEV1 (% predicted)	90.02 ± 14.20	86.46 ± 10.07	0.41
Baseline Beck Depression Inventory Score	9.35 ± 7.37	12.06 ± 7.74	0.34
Baseline STAI state	45.00 ± 8.02	46.86 ± 5.33	0.46
Baseline STAI trait	30.00 ± 8.02	31.86 ± 5.33	0.46
Baseline RCADS-social phobia	5.14 ± 5.14	6.06 ± 3.84	0.58
Baseline RCADS-panic disorder	2.71 ± 2.78	3.80 ± 2.93	0.31
Baseline RCADS-separation anxiety disorder	8.13 ± 4.53	5.35 ± 3.43	0.08
Baseline RCADS generalized anxiety disorder	6.28 ± 2.81	4.33 ± 3.01	0.08
Baseline RCADS-obsessive-compulsive disorder	3.64 ± 2.92	4.80 ± 2.11	0.23
Baseline RCADS major depressive disorder	6.33 ± 4.36	6.21 ± 3.11	0.13
Baseline CFQ-R physical functioning	63.33 ± 29.59	78.17 ± 20.83	0.14
Baseline CFQ-R emotional functioning	74.47 ± 16.37	82.74 ± 9.92	0.14
Baseline CFQ-R social functioning	49.94 ± 18.14	56.12 ± 20.02	0.48
Baseline CFQ-R body image	89.63 ± 12.92	78.57 ± 25.21	0.34
Baseline CFQ-R eating problems	61.23 ± 25.09	65.33 ± 33.72	0.74
Baseline CFQ-R treatment burden	74.07 ± 17.64	69.04 ± 20.52	0.77
Baseline CFQ-R respiratory	83.92 ± 16.81	81.66 ± 16.12	0.82
Baseline CFQ-R digestive	68.89 ± 23.45	71.43 ± 17.81	0.82

Note: Data are given as mean ± standard deviation (percentage) unless stated otherwise.

Abbreviations: 6MWT, 6-min walking test; BDI, Beck Depression Inventory; CFQ-R, Cystic Fibrosis Questionnaire-Revised; FEV1, forced expiratory volume in the first second; RCADS, Anxiety and Depression Scale in Children-Revised; STAI, State-Trait Anxiety Inventory.

the treatment, with the mean score of control group being 6.64 ± 2.73 ($p = 0.004$). There were no significant changes in quality-of-life scores in both groups except for the body image scores in telerehabilitation group, which showed an improvement from 78.52 ± 25.21 to 86.50 ± 18.58 ($p = 0.04$). But between-group analyses showed no significant difference between groups in body image scores at the end of the treatment (Tables 2 and 3, Figure 2). There were no significant improvements in caregivers' depression and anxiety scores (Table 4).

While the mean 6-min walking test in the telerehabilitation group was 431.78 ± 170 m at baseline; in the control group, this distance was 416.23 ± 115.30 m. At the end of the study, this distance increased to 483.07 ± 107.04 in the telerehabilitation group, while it

was 405.66 ± 97.54 in the control group, and there was a significant difference between the two groups ($p = 0.05$; Figure 3). When the within-group changes were analyzed, it was seen that this increase was also significant compared to the baseline value of the telerehabilitation group ($p = 0.02$). Considering the estimated FEV1% values, the mean value of the telerehabilitation group at the beginning was 90.02 ± 14.20 , while it was 92.00 ± 12.28 at the end of the study, and no significant change was observed ($p > 0.05$). While the initial value was 86.46 ± 10.07 in the control group, it was 85.20 ± 8.45 at the end of the study, and no significant change was observed ($p > 0.05$).

No unwanted incidences occurred during the telerehabilitation sessions.

TABLE 2 Change between Cystic Fibrosis Questionnaire-Revised (CFQ-R) scores before and after treatment

	Group	Baseline	After treatment	Intra-group mean difference (95% CI of the difference); <i>p</i>
CFQ-R physical functioning	Telerehabilitation	78.17 ± 20.83	76.98 ± 19.47	1.37 (-7.57 to -10.31); <i>p</i> = 0.74
	Control	63.33 ± 29.59	67.45 ± 25.95	-2.77 (-11.39 to -5.84); <i>p</i> = 0.49
	Intra-group mean difference (95% CI of the difference); <i>p</i>	-14.83 (-34.47 to 4.79); <i>p</i> = 0.13	5.02 (-9.52 to 19.58); <i>p</i> = 0.28	
CFQ-R emotional functioning	Telerehabilitation	82.74 ± 9.92	81.54 ± 12.94	1.26 (-4.78 to -7.31); <i>p</i> = 0.65
	Control	74.47 ± 16.37	76.19 ± 14.56	-1.75 (-7.93 to 4.42); <i>p</i> = 0.55
	Intra-group mean difference (95% CI of the difference); <i>p</i>	-8.26 (-18.67 to 2.14); <i>p</i> = 0.11	5.20 (-16.06 to 5.34); <i>p</i> = 0.31	
CFQ-R social functioning	Telerehabilitation	56.12 ± 20.02	57.71 ± 20.25	-1.43 (-10.01 to 7.13); <i>p</i> = 0.72
	Control	49.94 ± 18.14	55.44 ± 19.11	-5.3 (-14.70 to -4.04); <i>p</i> = 0.24
	Intra-group mean difference (95% CI of the difference); <i>p</i>	6.17 (-20.71 to 8.37); <i>p</i> = 0.39	-5.35 (-16.06 to 5.34); <i>p</i> = 0.31	
CFQ-R body image	Telerehabilitation	78.52 ± 25.21	86.50 ± 18.58	-7.92 (-15.63 to 0.20); <i>p</i> = 0.04*
	Control	89.63 ± 12.92	87.30 ± 16.22	1.58 (-4.50 to -7.67); <i>p</i> = 0.58
	Intra-group mean difference (95% CI of the difference); <i>p</i>	11.05 (-4.04 to 26.16); <i>p</i> = 0.14	0.79 (-12.75 to 14.34); <i>p</i> = 0.90	
CFQ-R eating problems	Telerehabilitation	65.33 ± 33.72	65.42 ± 29.84	-2.18 (-16.83 -12.46); <i>p</i> = 0.75
	Control	61.23 ± 25.09	58.58 ± 28.15	3.05 (-7.23 to 13.35); <i>p</i> = 0.53
	Intra-group mean difference (95% CI of the difference); <i>p</i>	-4.29 (-26.83 to 18.24); <i>p</i> = 0.69	-6.83 (-29.37 to -15.70); <i>p</i> = 0.53	
CFQ-R treatment burden	Telerehabilitation	69.04 ± 20.52	65.87 ± 25.58	3.31 (-5.51 to 12.13); <i>p</i> = 0.42
	Control	74.07 ± 17.64	71.42 ± 21.22	3.97 (-2.49 to 10.43); <i>p</i> = 0.21
	Intra-group mean difference (95% CI of the difference); <i>p</i>	5.02 (-9.52 to 19.58); <i>p</i> = 0.48	5.55 (-12.70 to 23.81); <i>p</i> = 0.53	
CFQ-R respiratory symptoms	Telerehabilitation	83.92 ± 16.81	85.71 ± 11.97	1.66 (3.73-9.74); <i>p</i> = 0.66
	Control	81.66 ± 16.12	79.76 ± 15.91	3.87 (-2.39 to 11.43); <i>p</i> = 0.23
	Intra-group mean difference (95% CI of the difference); <i>p</i>	6.11 (-14.80 to 10.29); <i>p</i> = 0.71	-5.95 (-16.89 to 4.99); <i>p</i> = 0.27	
CFQ-R digestive symptoms	Telerehabilitation	71.43 ± 17.81	76.18 ± 20.37	-2.18 (-21.44 to 17.07); <i>p</i> = 0.81
	Control	68.89 ± 23.45	59.52 ± 29.75	7.14 (-1.05 to 15.33); <i>p</i> = 0.08
	Intra-group mean difference (95% CI of the difference); <i>p</i>	-2.54 (-18.50 to 13.42); <i>p</i> = 0.74	-16.66 (-36.47 to 3.15); <i>p</i> = 0.10	

TABLE 3 Intragroup and intergroup comparison of Anxiety and Depression Scale in Children-Revised (RCADS) before and after treatment in patients

Group	Baseline	After treatment	Intra-group mean difference (95% CI of the difference); <i>p</i>
RCADS-social phobia			
Telerehabilitation	5.14 ± 5.14	5.64 ± 5.28	-0.5 (-2.44 to -1.44); <i>p</i> = 0.58
Control	6.06 ± 3.84	5.07 ± 4.58	1.07 (-0.74 to 2.88); <i>p</i> = 0.22
Intra-group mean difference (95% CI of the difference); <i>p</i>	0.92 (-2.51 to 4.36); <i>p</i> = 0.58	-0.57 (-4.41 to 3.27); <i>p</i> = 0.76	
RCADS-panic disorder			
Telerehabilitation	2.71 ± 2.78	1.50 ± 1.69	1.21 (-0.51 to -2.94); <i>p</i> = 0.15
Control	3.80 ± 2.93	2.78 ± 3.26	1.07 (-0.81 to 2.95); <i>p</i> = 0.24
Intra-group mean difference (95% CI of the difference); <i>p</i>	1.08 (-1.09 to 3.26); <i>p</i> = 0.31	1.28 (-0.73 to 3.30); <i>p</i> = 0.20	
RCADS-major depressive disorder			
Telerehabilitation	6.33 ± 4.36	3.92 ± 3.79	2.28 (0.43-4.13); <i>p</i> = 0.02*
Control	6.21 ± 3.11	7.78 ± 3.04	-1.42 (-3.16 to 0.30); <i>p</i> = 0.10
Intra-group mean difference (95% CI of the difference); <i>p</i>	0.11 (-2.79 to 3.03); <i>p</i> = 0.93	3.85 (1.18-6.52) <i>p</i> = 0.006*	
RCADS-separation anxiety disorder			
Telerehabilitation	5.35 ± 3.43	5.50 ± 3.77	0.14 (-1.89 to -1.60); <i>p</i> = 0.86
Control	8.13 ± 4.53	5.50 ± 3.88	3.07 (0.54-5.59); <i>p</i> = 0.07
Intra-group mean difference (95% CI of the difference); <i>p</i>	2.77 (-0.30 to -5.85); <i>p</i> = 0.08	0.15 (-2.97 to 2.97); <i>p</i> = 0.95	
RCADS-generalized anxiety disorder			
Telerehabilitation	6.28 ± 2.81	3.42 ± 2.65	2.85 (1.39-4.32); <i>p</i> = 0.001*
Control	4.33 ± 3.01	6.64 ± 2.73	-2.21 (-4.11 to 0.31); <i>p</i> = 0.02*
Intra-group mean difference (95% CI of the difference); <i>p</i>	-1.95 (-4.17 to 0.27); <i>p</i> = 0.08	3.21 (1.12 to -5.30); <i>p</i> = 0.004*	
RCADS-obsessive compulsive disorder			
Telerehabilitation	3.64 ± 2.92	2.64 ± 2.81	1.00 (-0.86 to 2.86); <i>p</i> = 0.26
Control	4.80 ± 2.11	4.78 ± 3.21	0.21 (-1.88 to -2.31); <i>p</i> = 0.82
Intra-group mean difference (95% CI of the difference); <i>p</i>	1.15 (-0.77 to 3.19); <i>p</i> = 0.23	2.14 (0.2-4.49); <i>p</i> = 0.07	

*indicates statistical significance *p* < 0.05.

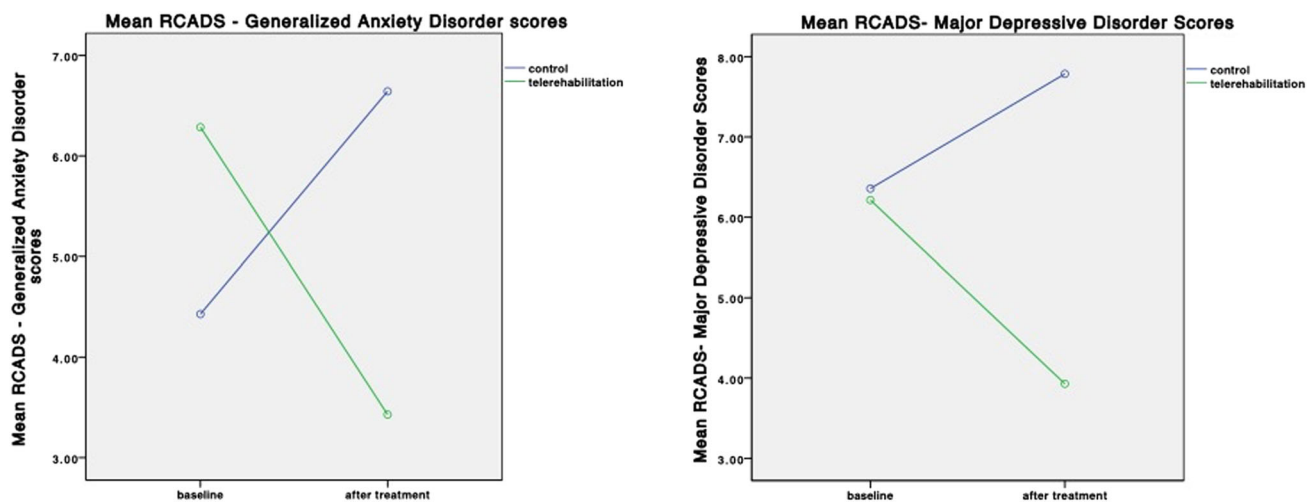


FIGURE 2 Changes in Anxiety and Depression Scale in Children-Revised major depressive disorder and generalized anxiety disorder domains before and after treatment [Color figure can be viewed at wileyonlinelibrary.com]

TABLE 4 Comparison of Beck Depression Inventory and State-Trait Anxiety Inventory (STAI) scores before and after treatment in caregivers

	Group	Baseline	After treatment	Intra-group mean difference (95% CI of the difference); <i>p</i>
Beck Depression Inventory	Telerehabilitation	9.35 ± 7.37	8.76 ± 6.02	0.57 (-1.83 to 2.97); <i>p</i> = 0.61
	Control	12.06 ± 7.74	9.78 5.72	2.42 (-1.26 to 6.12); <i>p</i> = 0.18
	Intra-group mean difference (95% CI of the difference); <i>p</i>	2.70 (-3.06 to 8.47); <i>p</i> = 0.34	1.00 (-3.56 to 5.56); <i>p</i> = 0.65	
STAI state	Telerehabilitation	45.00 ± 8.02	44.64 ± 7.38	0.35 (-2.51 to 3.22); <i>p</i> = 0.79
	Control	46.86 ± 5.33	44.42 ± 5.00	2.21 (-1.36 to 5.78); <i>p</i> = 0.20
	Intra-group mean difference (95% CI of the difference); <i>p</i>	1.86 (-3.29 to 7.02); <i>p</i> = 0.46	0.21 (-3.29 to 7.02); <i>p</i> = 0.92	
STAI trait	Telerehabilitation	30.00 ± 8.02	29.64 ± 7.38	0.35 (-2.51 to 3.22); <i>p</i> = 0.79
	Control	31.86 ± 5.33	29.42 ± 5.00	2.21 (-1.36 to 5.78); <i>p</i> = 0.20
	Intra-group mean difference (95% CI of the difference); <i>p</i>	1.86 (-3.29 to 7.02); <i>p</i> = 0.46	0.21 (-3.29 to 7.02); <i>p</i> = 0.92	

4 | DISCUSSION

This study has shown that a telerehabilitation approach that includes postural and high-intensity exercises can improve anxiety, depression, and functional performance in patients with CF. However, it does not seem to improve quality of life parameters, pulmonary function, and caregivers' anxiety and depression levels. It must be kept in mind that patients previously diagnosed with mental health disorders, such as depression, were not recruited for this study. While this was done to understand the effect of exercise and remove confounding treatments that might improve depression and anxiety scores. But this in turn would have excluded patients that had higher scores at the baseline, which might have altered the ability to detect change within our sample. This approach might have also excluded caregivers with higher anxiety and depression scores from the study

and resulted in nonsignificant changes within our study group. Normal values of RCADS scores were found to be 7.78 ± 6.24 in major depressive disorder and 6.01 ± 4.18 for the generalized anxiety disorder in the Turkish population.¹⁹ Our patients' RCADS scores were near these showing that patients with psychiatric diagnoses were eliminated successfully, but patients with higher scores are also left out. Therefore, the improvement in these domains in the telerehabilitation group might not be a clinical change but define the change in the mood and generalized anxiety levels of the patients. However, it is also impossible to determine if this change is clinically significant, since minimal clinically important difference scores were not calculated in Turkish population and developers of RCADS warn against using the English values in other populations.²⁰ This would also explain why telerehabilitation failed to show any improvement in other domains of the RCADS, since they are also near normal and

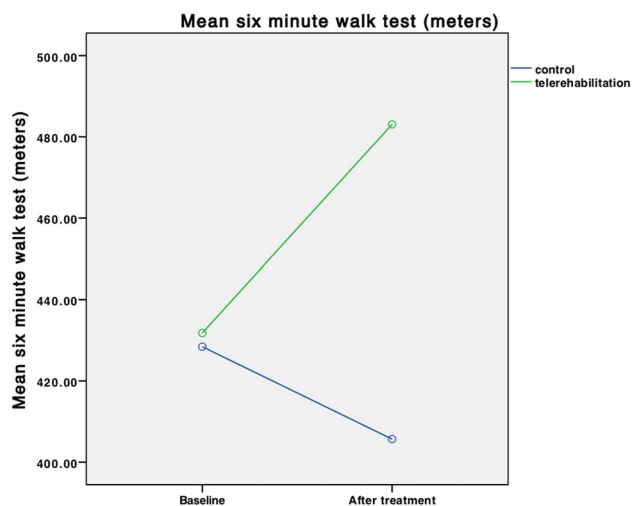


FIGURE 3 Changes in mean 6-min walking distances before and after treatment [Color figure can be viewed at wileyonlinelibrary.com]

shows the lack of pathologies such as obsessive-compulsive disorder. It must also be stated that the RCADS-GAD scores increased during the study period in the control group, showing the probable adverse effects of the pandemic period in this group.

The risk of cross-contamination with pathogens such as *Pseudomonas aeruginosa* has always been a major obstruction in the way of group therapies, including supervised aerobic exercises in patients with CF. For this purpose, telehealth has always been suggested as a viable alternative in these patients.²¹ Considering the positive effects of exercise therapies in patients with CF, this had been a major setback in the implementation of such therapies. The ongoing pandemic has shifted the priority in all healthcare, making telehealth applications, including telerehabilitation, a cornerstone.²² That is why telerehabilitation studies are of high importance in patients with CF. As of Fall 2021, the pandemic is raging on, and it seems that we will rely on these approaches for a while longer, maybe indefinitely, for high-risk patients such as patients with CF. Our study documented that telerehabilitation can be helpful in improving function and anxiety levels in these patients, and no unwanted events occurred, showing that these programs are safe and effective. However, telerehabilitation depends highly on the patients' ability to reach suitable equipment such as a computer and internet access and may not be applicable for all patients.

This study implemented high-intensity interval training but while named high intensity, it was not possible for us to monitor heart rate in a remote setting, so the exercise program was designed by the physiotherapist on the basis of their previous experience with on-site monitoring of children with CF. The design of our study documented that after a careful design by a competent physiotherapist, proper documentation of the exercises, which we provided with this manuscript, and proper training of the researchers, these exercises can successfully be applied and therefore can be used in all centers that have their own physiotherapy team that has internet access and enthusiastic patients. Also since the researchers applying the exercise

program were not physiotherapists themselves, a similar program can be taught to parents, medical students or nurses, nurse assistants and can be employed successfully with a lower cost. Still, not being able to monitor the heart rate is a major setback, and programs that can monitor patients remotely would be healthier. Previous studies have documented that high-intensity training programs are safe and effective in improving peak oxygen uptake in adults with CF, while they did not show any functional improvement.^{23,24} However, there are no previous studies that investigated its effects on pulmonary function, quality of life, and psychological aspects in children between the ages of 6–13, making our study important on this aspect. The limitations mentioned above are also why we did not want to rely on high-intensity training alone and added the postural exercise component to our programs. While we could not monitor the heart rate, we could see if the patient did the postural exercises and did them properly during telerehabilitation sessions, which is an advantage compared to home-based exercises alone. Also, a better posture has been shown to improve lung volume parameters in children with CF.²⁵ A previous study showed that postural exercises could help improve quality of life and functional parameters when added to a pulmonary rehabilitation program in this age group.²⁶ Our study did not implement a full-on pulmonary rehabilitation program, rather just implemented an exercise therapy for these patients. We did not monitor the airway clearing techniques, and that is a potential confounder. However, both postural and aerobic exercises are suggested to all patients with CF regardless of the type of airway cleaning technique used in their daily lives. However, a future study that could also control these aspects may be required to see if a certain regimen containing a type of airway clearance with an exercise is better than another.

Numerous studies have documented the positive effects of exercise on anxiety and depression in other populations.²⁷ But the data about the patients with CF are limited. Some studies also indicate that the ongoing pandemic has increased the anxiety levels in adults patients with CF.²⁸ Our study showed that a telerehabilitation approach significantly improved generalized anxiety and major depressive disorder domains in children with CF. This shows that short-term programs can help in improving a patient's psychological state. An improved psychological state can be helpful for patients to implement long-term changes in their treatment regimen, such as increasing their adherence to therapies, which is essential in patients with CF. Similarly, especially in the pediatric patient group, the status of caregivers' psychological condition is crucial for proper treatment implementation.²⁹ Previous studies documented the increased caregiver burden and the adverse effects of impaired mental health state on reaching health services in this patient population.^{30,31} Therefore, to decrease the burden and improve caregiver mental state must always be considered when a new treatment regimen is implemented. While our study failed to show any improvement on caregivers' level of anxiety and depression, other approaches such as direct tele-counseling sessions might be more helpful in this regard.

There were no changes in quality-of-life subscales except the fact that there was a significant increase in the body image subscale

in telerehabilitation group. However, the final score of the telerehabilitation group was not significantly better than the control group. Previous studies have shown that exercise positively influences body image both in children in general and in children with CF.^{32,33} Similar to RCADS, due to lack of previous data, we cannot state that this change is clinically significant. However, due to the lack of a significant difference in between-group analyses, we cannot attribute this change confidently to the telerehabilitation procedure.

There are serious limitations of this study. While the number of patients might be adequate for statistical purposes, only a small population could be recruited for this single-center study. A similar design with a multicentric approach could give different results while showing telerehabilitation can be used safely and effectively everywhere. Another serious limitation is the lack of heart rate monitoring during exercise, as mentioned above. Monitorization would improve the effectiveness and safety of the exercises, but it could not be implemented in this study due to funding issues. Finally, we could not exclude the negative effects of the pandemic on patients' and caregivers' anxiety levels. Especially the lack of change in caregivers' might be related to the ongoing situation, and without the end of the pandemic, there is no way of knowing its influence.

5 | CONCLUSION

A telerehabilitation approach that includes postural and aerobic exercises can help patients with CF to improve their functional status, depression, and anxiety levels and might positively influence body image. There were no changes in caregivers' levels of anxiety and depression. This single-centered and small-scale study can help to establish similar treatment modalities in other CF centers. However, larger-scale studies are needed for more robust results and generalizability for the entire CF population.

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CONFLICT OF INTERESTS

The authors declare that there is no conflict of interests.

AUTHOR CONTRIBUTION

Özge Kenis-Coskun: Conceptualization (lead); Data curation (lead); Formal analysis (equal); Investigation (equal); Methodology project administration (equal); Writing – original draft (equal); Writing – review & editing (equal). **Ahsen N. Aksoy:** Data curation (equal); Investigation (equal); Methodology (equal); Project administration (supporting); writing – original draft (supporting); writing – review & editing (supporting). **Eda N. Kumaş:** Methodology (equal); Project administration (supporting); Writing – original draft (supporting). **Aybike Yılmaz:** Data curation (equal); Investigation (equal); Methodology (equal); Project administration (supporting); Writing – original draft (supporting); Writing – review & editing (equal).

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DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

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SUPPORTING INFORMATION

Additional supporting information may be found in the online version of the article at the publisher's website.

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