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Singing Therapy for Young People With Cystic Fibrosis: A Randomized Controlled Pilot Study

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Abstract

Singing is not merely a vocal expression but also a physical activity involving the lungs and respiratory muscles. The purpose of this study was to evaluate singing as an adjunct therapy for young people with cystic fibrosis (CF). In a randomized controlled study, 51 hospitalized children (mean age = 11.6 years, 35% male) participated in either 8 singing or 8 recreational sessions. Respiratory muscle strength using maximal inspiratory and expiratory pressure (MIP and MEP, respectively), spirometry, and quality of life (QoL) were assessed at baseline (T1), postintervention (T2), and follow-up (T3). The singing group demonstrated statistically significant increases in MIP and MEP at T2 and T3, while no statistically significant changes were evident in the control group. In the QoL measurements, both groups showed improvement in some domains. The results indicate that singing has the potential to improve the respiratory status and QoL of young people with CF.

Keywords

cystic fibrosis, quality of life, respiratory muscle strength, singing

Introduction

Cystic fibrosis (CF) is a genetically inherited life-threatening condition that affects major organs, in particular the lungs.[1] Cystic fibrosis requires a multifaceted daily treatment regimen that involves a range of airway clearance therapies to maintain lung health.[2] Previous studies have found that, due to the chronic nature of the illness and the relentless demands of treatments, many individuals with CF are likely to have a poor quality of life (QoL).[3,4] A recent systematic review reported a lack of assistance to cope with the lifelong challenges of CF.[5]

Growing evidence suggests that singing may enhance QoL.[6] In the field of respiratory research, a study by Lord et al found that participants with chronic obstructive pulmonary disease (COPD) had reduced anxiety and depression ($P = .03$) and increased general well-being following 12 biweekly group singing sessions.[7] However, to date no study has examined the benefits of singing in enhancing QoL in people with CF.[8]

Anecdotal evidence also suggests that singing acts as a form of exercise for the respiratory system.[9,10] Singing is a complex interaction between the vocal apparatus, respiratory system, and abdominal musculature.[11] It places great demands on the respiratory system due to the changes required in pitch (high/low), dynamics (loud/soft), and phrasing (short/sustained).[12] The singer must actively engage the respiratory muscles to expand the rib cage and abdominal wall, in order to achieve the lung volume and subglottal pressure necessary to produce high and loud tones. During singing, the singer is often required to sustain a phrase for an extended period, during which the respiratory system has to work efficiently to generate constant subglottal pressures.[12]

A recent randomized controlled study evaluated singing as a therapy for people with COPD. Bonilha et al conducted 24 weekly singing sessions and measured the effects of singing on participants' respiratory muscle strength using maximal expiratory pressure (MEP) and maximal inspiratory pressure (MIP). Participants demonstrated improved MEP ($P = .05$) following the intervention, while increased MIP was observed without statistical significance.[13]

For individuals with CF, as for patients with COPD, the respiratory muscles play an important role in maintaining lung

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health.[2] Respiratory muscle function also affects exercise capacity in people with CF,[14] and greater levels of exercise capacity are correlated with lower levels of mortality.[15] To improve respiratory muscle function in CF, conventional treatment uses inspiratory muscle training (IMT) devices. Some research has shown that an IMT program can improve respiratory muscle function in children;[16] however, a recent systematic review detected only weak evidence that conventional IMT devices were beneficial.[17] Using an IMT device can also be seen as another chore for children with CF, given the complexity of the daily treatment regimen. In contrast, the present study aimed to provide an enjoyable activity that would also have therapeutic benefits. We hypothesized that hospitalized young people with CF would experience improved respiratory muscle strength and enhanced QoL after participating in a singing intervention.

Research Design and Method

Participants

We included young people (7-17 years) who had an established diagnosis of CF, who were being treated as inpatients for a respiratory exacerbation at 2 large pediatric hospitals (X and Y) in Brisbane, Australia. All participants were receiving intravenous antibiotic treatment and chest physiotherapy as the standard hospital care. We excluded children who had undertaken individual singing lessons within the past 5 years, who had an intellectual disability or who, according to their treating physician, were too ill to participate in singing activities. The human ethics committees of the participating hospitals and University of Sydney approved the study.

Study Protocol

A dual-center, single-blinded, parallel randomized controlled trial was conducted. Hospitalized children with CF at the X and Y were approached within 2 days of their admission. Children and their parents/caregivers were provided a study information sheet and were later approached to obtain informed consent. Signed consent was also obtained from children aged >12 years. Participants were then allocated to the next sequential number on the appropriate randomization list. Allocation was concealed with an opaque sticker. Stratification by age ($\leq$13 and >13 years) was used. The list was computer-generated using block randomization, which was performed by a person external to the research team. Using this randomization list, participants were allocated to either (1) the treatment group that participated in 8 standard individual singing lessons or (2) the control group that participated in 8 individual recreational sessions. The control group participants chose activities that did not encourage any respiratory muscle exercise, such as playing computer games, watching movies, or playing percussive instruments. Children in both groups received equal amounts of the researcher's time and attention.

Tests were conducted at 3 points in time: at baseline (T1), immediate postintervention (T2, following 8 individual sessions and prior to discharge), and at follow-up (T3, 6-8 weeks post-discharge). All participants undertook the following: respiratory muscle strength measurements (using MIP and MEP as surrogates), spirometry, and a CF-specific tool, the Cystic Fibrosis Questionnaire–Revised (CFQ-R).

Respiratory Measurements

Both MIP and MEP are simple, noninvasive methods that measure the strength of the respiratory muscles while breathing in and out.[18] The MIP is measured during maximum inspiratory effort against an occluded airway, following maximum exhalation. The MEP is measured during forcible exhalation through a blocked mouth piece, after full inhalation.[18] These measurements were performed in accordance with pediatric guidelines[19] by experienced respiratory scientists who were blinded to participants' group assignment. A pressure manometer (P.K. Morgan Kent, UK) was used at X and a SensorMedics V6200 Autobox (Legacy, Yorba Linda, California) at Y.

Spirometry tests (forced expiratory volume in 1 second [FEV_1], forced vital capacity [FVC], and forced expiratory flow 25% to 75% [$FEF_{25\%-75\%}$]) measure how quickly and effectively the lungs can be emptied and filled and are commonly used to detect ventilatory defects in the airways.[20] Forced expiratory volume in 1 second is the volume of the air that can be forcibly breathed out in 1 second after full inhalation. Forced vital capacity is the volume of air that can be forcibly exhaled after full inhalation. Forced expiratory flow 25% to 75% ($FEF_{25\%-75\%}$) measures the forced expiratory flow from 25% to 75% of vital capacity and is considered as an index of small airway function.[20] These were performed in accordance with the American Thoracic Society/European Respiratory Society guidelines[20] by experienced respiratory scientists who were blinded to participants' group assignment. A SensorMedics Vmax spirometer (Palm Springs, California) was used at X and a SensorMedics Vmax20C (Legacy) spirometer at Y.

Quality of Life Measurements—CFQ-R

The CFQ-R is an established CF-specific QoL instrument that has good reliability and validity and that measures the QoL during the previous 2 weeks.[21] It can be used to evaluate new therapies and inform clinical practice.[21] In younger children (7-11 years), an interview format was used to obtain answers, while older children (12-17 years) self-completed the CFQ-R. The questionnaire covers 6 generic domains: physical functioning, role, vitality, emotional status, social functioning, and health perceptions. It also includes 6 CF-specific domains: body image, eating disturbances, treatment burden, respiratory symptoms, digestive symptoms, and weight. For adolescents (>13), the questionnaire has 50 items across all 12 domains, while only 8 domains with 35 items apply for younger children ($\leq$13). Response choices are ratings of frequency and difficulty on a 4-point scale (1 = *always* to 4 = *never* or 1 = *very true* to 4 = *very false*). Scores of each domain are standardized on a 0- to 100-point scale, with higher scores reflecting better QoL.

Singing Protocol

A specialized singing program was developed by a musician and a music therapist, based on the well-established vocal pedagogy for young people by Phillips.[22] The program was based on diaphragmatic breathing (the essential technique in singing)[23] to provide optimal respiratory muscle exercise. A 4-stage program was developed: posture, diaphragmatic breathing, vocal warm-up, and singing. First, participants were taught how to adopt good posture while singing, which is essential for diaphragmatic breathing.[22] Second, diaphragmatic breathing was practiced.[22] Third, vocal warm-up exercises were performed using this technique. Fourth, songs were chosen to sing that would provide an optimal respiratory workout. The most effective songs are those that consist of high notes and sustained phrases.[24] Unlike normal speech, these require deeper breathing and the active engagement of respiratory muscles.[24] To maximize enjoyment, the songs were selected from among each participant's nominated list of favorite songs.

All sessions were conducted by a qualified, experienced music therapist. The therapist demonstrated the correct application of all exercises, adjusted the program to each participant's learning pace, and created a nonjudgmental and encouraging environment for singing. Upon discharge, following the completion of the singing intervention, participants were given a singing diary. This diary outlined 6 simple singing tasks to be completed each day for the following 6 to 8 weeks. Participants were encouraged to complete the tasks daily and to log their singing in the diary.

Statistical Analysis

Descriptive statistics were computed to describe participant characteristics, and Fisher exact chi-square, and *t* test were used to compare the groups on demographic and lung function characteristics at baseline. Kolmogorov-Smirnov test was used to examine the normality of the data. Due to a lack of power, repeated measures *t* tests were performed for each group, to compare improvement of the major outcome variables (respiratory measurements) across the 3 time points. Wilcoxon signed rank tests were used to analyze the QoL data, as the data violated normality assumptions. All statistical analyses were performed in SPSS (version 20). Two-tailed $P \leq .05$ values were considered significant.

Results

Between April 2008 and July 2010, potential participants (n = 73) were approached. One child did not meet the inclusion criteria and 21 declined to participate. Among the 51 patients who agreed to participate, 26 were allocated to the singing group and 25 to the control group. Eleven were lost at postintervention (T2). A total of 40 (20 in each group) were included for the baseline and postintervention analysis. Ten were lost at follow-up (T3); therefore, a total of 30 (15 in each group) were included for the baseline and follow-up analysis (Figure 1 consort flow diagram). As Table 1 indicates, the baseline characteristics of the treatment and control groups were similar.

Respiratory Measurements

Repeated measures *t* tests were conducted to evaluate the effects of singing on respiratory muscle strength and spirometry. None of the variables showed significant differences in profiles between the treatment and control groups except for MEP ($P = .021$), and there were no significant differences between groups at T2 or T3. In the treatment group, there was a statistically significant increase in MIP at T2 ($P < .001$) and T3 ($P = .016$) but not in the control group (all P values > .075). For MEP, there was a statistically significant increase in the treatment group at T2 ($P = .034$) and at T3 ($P = .002$) but again not in the control group (all P values > .395). Results of paired samples *t* tests for MIP and MEP, with effect sizes (Cohen *d*) ranging from small to large are presented in Table 2. Between-group differences in changes of the means in respiratory muscle strength measurements (T2-T1 & T3-T1) are presented graphically in Figure 2. There were increases in the means of MIP and MEP in the control group. However, the changes were smaller than those in the singing group and no statistical significance was detected.

In relation to spirometry (FEV_1, FVC, and $FEF_{25\%-75\%}$), a statistically significant increase was observed only in $FEF_{25\%-75\%}$ at T2 ($P = .035$) in the treatment group, which was not maintained at T3 ($P = .896$). No statistical significance in FEV_1 (all P values > .19) or FVC (all P values > .425) was observed in the treatment group. No statistically significant changes were evident in spirometry data for the control group (FEV_1 all P values > .123; FVC all P values > .068; and $FEF_{25\%-75\%}$ all P values > .267; Table 2).

Quality of Life Measurements

Of the 6 generic domains, no statistical significance was observed in either group for the physical functioning, role, or health perception domains (all P values > .065 at both T2 and T3). In the emotional domain, the treatment group showed a statistically significant increase at T2 ($P = .031$), but this was not maintained at T3 ($P = .694$). No statistical significance was observed in the control group for this domain ($P > .899$). In both the social and vitality domains, the control group showed statistically significant increases at T2 ($P = .044$ and $P = .043$, respectively), but these were not maintained at T3 ($P = .807$ and $P = .176$, respectively). The treatment group showed no statistically significant increase in either the social or vitality domain (all P values > .053).

Of the 6 CF-specific domains, statistically significant improvements in respiratory symptoms were evident in both groups at T2 (treatment group $P = .001$; control group $P = .002$) and T3 (treatment group $P = .016$; control group $P = .013$). A between-group comparison of the mean differences for this domain is illustrated in Figure 3. Further, statistically significant decreases were revealed in

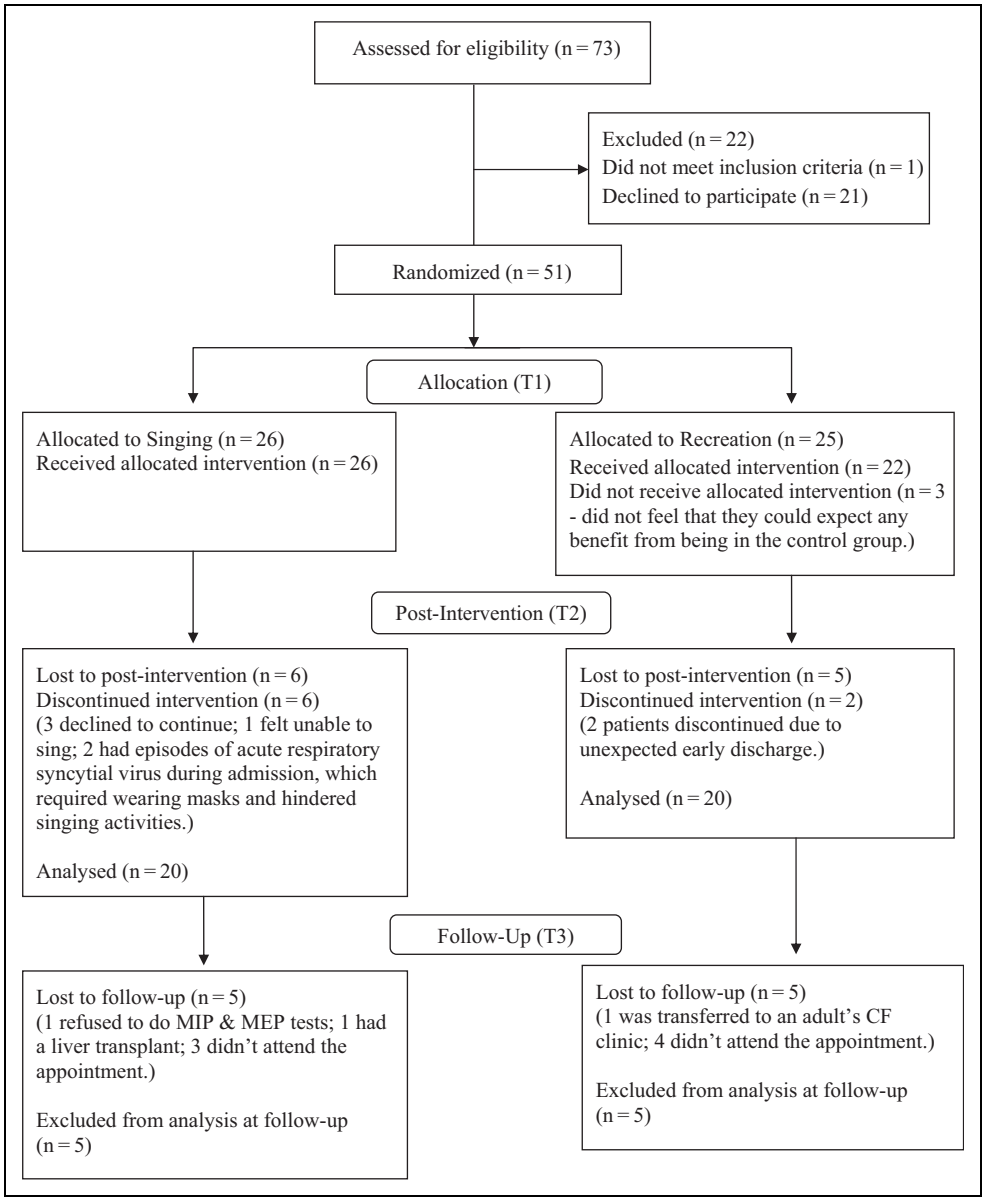

Figure 1. Consort flow diagram

treatment burden for both groups at T2 (treatment group $P = .014$; control group $P = .026$). However, the reduced treatment burden was only maintained in the treatment group at T3 ($P = .031$). A between-group comparison in the mean differences for this domain is illustrated in Figure 4. For the digestion domain, a statistically significant increase was evident only in the treatment group at T2 ($P = .024$) that was not sustained at T3 ($P = .833$), and there was no statistical significance in the control group at either T2 or T3 (all P values > .335). There were no significant changes in either group in the eating (all P values > .051), weight (all P values > .058), and body image domains (all P values > .122). A summary of the results and effect sizes (r) are presented in Table 3. The changes for those domains that are most relevant to the present study are discussed in the following section.

Singing Diary

Seventy-five percent of the participants returned their singing diaries at T3, and 80% had completed more than half of the daily singing activities.

Discussion

In this study, the first randomized controlled trial on singing as an adjunct intervention for hospitalized children with an acute exacerbation of CF, we have found that singing increases MIP, MEP, and some QoL domains.

Study results showed that participants in both groups demonstrated improved MIP and MEP at T2 and T3, indicated by the improved group means in Table 2. However, the changes were

Table 1. Participant Characteristics at Baseline (T1)

	Treatment group, N = 20 (%)	Control group, N = 20 (%)	*P* value
Gender			.337
Male	7 (35)	10 (50)	
Female	13 (65)	10 (50)	
Ethnicity			.231
Caucasian	17 (85)	20 (100)	
Non-Caucasian	3 (15)	0	
Age			1.000
7-13 years	14 (70)	14 (70)	
14-17 years	6 (30)	6 (30)	
Hospital site			1.000
X	16 (80)	15 (75)	
Y	4 (20)	5 (25)	
CF severity (based on FEV_1% at T1)			
Severe (30%-49%)	2 (10)	3 (15)	.828
Moderate (50%-79%)	9 (45)	7 (35)	
Mild (>79%)	9 (45)	10 (50)	
Previous music experience			.735
Participated in general school music	13 (65)	13 (65)	
Sang in a choir or played a wind instrument	4 (20)	2 (10)	
Played a string or other instrument	1 (5)	3 (15)	
None	2 (10)	2 (10)	
Physical activities			1.000
Regular participation in sports	14 (70)	15 (75)	
None	6 (30)	5 (25)	
BMI			
M (SD)[a]	18.66 (3.01)	17.49 (3.11)	.234
Lung function			
FEV_1 % predicted M (SD)[a]	.76 (.19)	.76 (.22)	.95
MIP M (SD)[a]	84.00 (16.97)	89.15 (35.6)	.563
MEP M (SD)[a]	98.6 (25.47)	108.95 (51.37)	.425

Abbreviations: CF, cystic fibrosis; FEV_1, forced expiratory volume in 1 second; BMI, body mass index; MIP, maximal inspiratory pressure; MEP, maximal expiratory pressure; SD, standard deviation.
[a]Mean (SD).

only statistically significant for the treatment group. The key component of the intervention was diaphragmatic breathing and, given that MIP measures the strength of the diaphragm, the increase observed in MIP in the singing group is an important finding. However, the amount of increase observed in MEP in the treatment group at T3, as shown in Figure 2, was unexpected. This result may be due to the program of singing undertaken by the treatment group during the postintervention period, through the use of the singing diary. Other studies have also shown similar improvements. A 12-week individualized singing program with 20 adults with impaired respiratory muscle strength from advanced multiple sclerosis found improved MEP in the treatment group, however due to a small sample size no statistical significance was observed.[25] In a second, uncontrolled study involving 20 patients with Parkinson disease, a 13-week group singing program showed statistically significant improvements in both MIP and MEP.[26] Our findings offer support to the hypothesis that singing promotes the efficient use of respiratory muscles and can be used as a type of respiratory muscle exercise.

Compared to the studies described earlier, the intervention period in our study was shorter (ie, 4 hours over approximately 2 weeks). It is possible that a longer intervention period (eg, 3, 6, or 12 months), accompanied by a relevant clinical outcome such as reduced exacerbations, may result in a more robust study and may show greater improvements in MIP and MEP and statistically significant between-group outcomes. On the other hand, our findings may indicate that a 4-hour singing program is the minimum needed for the CF population to maintain or improve respiratory muscle strength during an acute pulmonary exacerbation.

In relation to QoL, the respiratory, treatment burden, and emotional domains are most relevant to the present study. Participants in both the treatment and the control groups showed similar improvements. This is consistent with the findings of Bonilha et al.[13] Participants of both groups demonstrated significant improvement in the respiratory symptom domain at T2 and T3, with a medium-to-large effect size. This domain asks questions related to respiratory symptoms, such as coughing and breathing problems. As all children hospitalized for an acute respiratory exacerbation receive the standard medical treatment, it was not possible for this study design to assess the impact of singing independent of this treatment. It is interesting

Table 2. Respiratory Measurements[a]

	Treatment group (n = 20)						Control group (n = 20)					
	M	SD	Pair	*t*	*df*	*P* Value (effect size)	M	SD	Pair	*t*	*df*	*P* value (effect size)
MIP (cm H_2O) T1	84	16.97	T1-T2	5.53	19	<.000	89.15	35.59	T1-T2	1.88	19	.076
MIP (cm H_2O) T2	96.1	17.09				(*d* = .71)	95.25	37.93				
MIP (cm H_2O) T3[b]	98.27	21.52	T1-T3*	2.73	14	.016 (*d* = .64)	101.07	41.78	T1-T3[b]	1.51	14	.153
MEP (cm H_2O) T1	98.6	25.47	T1-T2	2.28	19	.034	108.95	51.37	T1-T2	.87	19	.396
MEP (cm H_2O) T2	107.6	28.09				(*d* = .33)	114.9	58.4				
MEP (cm H_2O) T3[b]	126.53	34.21	T1-T3*	3.85	14	.002 (*d* = .83)	116.67	50.48	T1-T3[b]	.08	14	.937
FEV_1 (l/s) T1	1.88	.62	T1-T2	1.14	19	.268	1.76	.93	T1-T2	1.51	19	.147
FEV_1 (l/s) T2	1.93	.58					1.85	.87				
FEV_1 (l/s) T3[b]	1.9	.53	T1-T3*	.40	14	.693	2.05	1.04	T1-T3[b]	1.64	14	.124
FVC (l) T1	2.41	.72	T1-T2	.31	19	.761	2.34	1.06	T1-T2	1.93	19	.069
FVC (l) T2	2.43	.66					2.45	1.03				
FVC (l) T3[b]	2.42	.60	T1-T3*	.66	14	.519	2.58	1.27	T1-T3[b]	1.05	14	.312
$FEF_{25\%-75\%}$ (l) T1	1.74	.79	T1-T2	2.27	19	.035	1.76	1.34	T1-T2	1.11	19	.279
$FEF_{25\%-75\%}$ (l) T2	1.9	.79				(*d* = .2)	1.95	1.29				
$FEF_{25\%-75\%}$ (l) T3[b]	1.79	.79	T1-T3*	.14	14	.896	2.27	1.42	T1-T3[b]	1.15	14	.268

Abbreviations: FEV_1, forced expiratory volume in 1 second; MIP, maximal inspiratory pressure; MEP, maximal expiratory pressure; SD, standard deviation; FVC, forced vital capacity; FEF, forced expiratory flow; (l/s), liters/second; (l), liters.
[a]Paired samples *t* test.
[b]At T3: treatment n = 15, control n = 15.

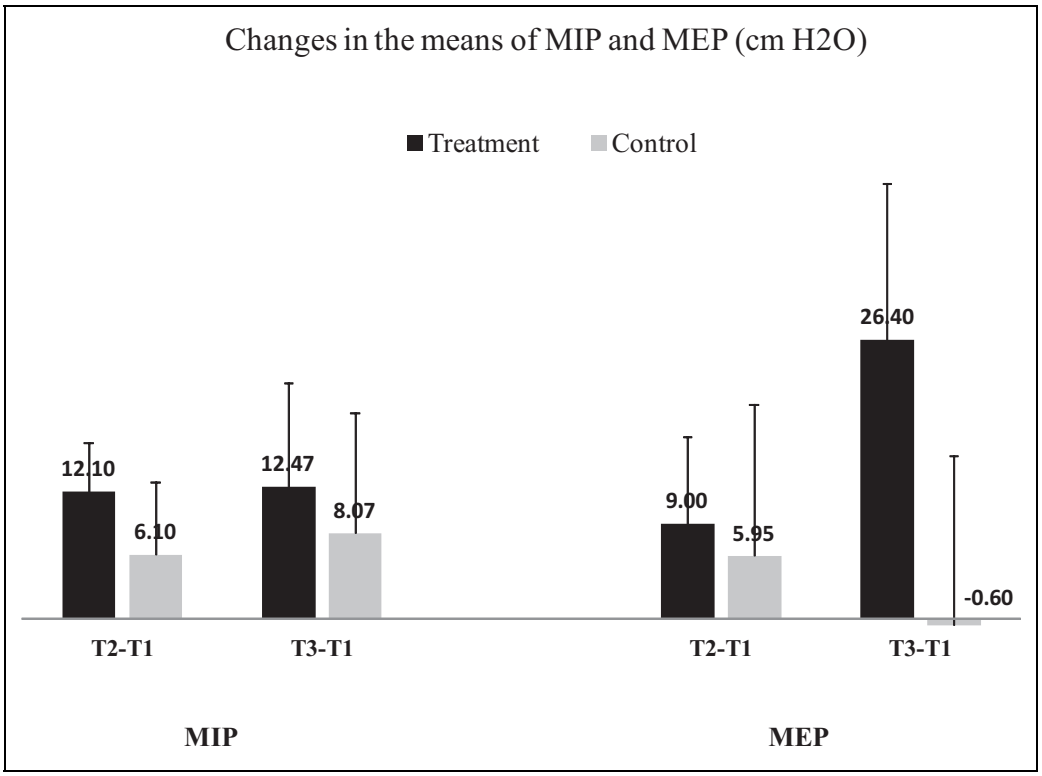

Figure 2. Maximal inspiratory and expiratory pressure (MIP and MEP): between-group comparison of changes in means

to note that the improvement in the control group seems to have been greater than in the treatment group (Figure 3). However, this observation is not consistent with the results of the objective respiratory measurements, as the control group showed no improvement in any of the respiratory measurements.

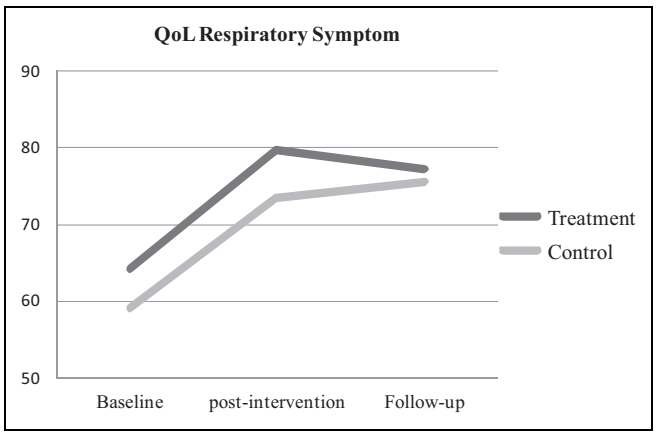

Figure 3. Quality of life (QoL) respiratory symptom: between-group mean comparison

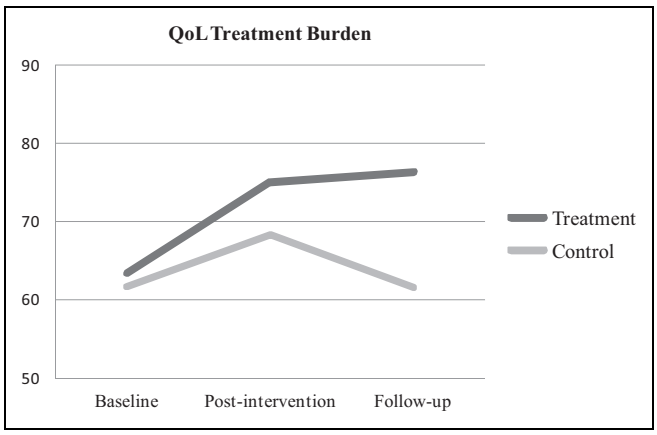

Figure 4. Quality of life (QoL) treatment burden: between-group mean comparison

Table 3. Quality of Life Measurements[a]

	Treatment group (n = 20)			Control group (n = 20)		
	Median (25th-75th percentiles)	Pair	*P* value (effect size)	Median (25th-75th percentiles)	Pair	*P* value (effect size)
Physical T1	80.50 (54.1-88.88)	T1-T2	.066	72.21 (56.94-88.80)	T1-T2	.320
Physical T2	83.30 (62.48-93.74)			83.30 (51.39-95.46)		
Physical T3[b]	83.30 (70.8-95.83)	T1-T3	.239	91.66 (75.00-100.00)	T1-T3	.083
Role T1[c]	58.33 (47.90-77.08)	T1-T2	.593	62.47 (54.13-87.48)	T1-T2	.752
Role T2[c]	58.30 (39.53-66.62)			66.63 (58.30-77.08)		
Role T3[d]	75.00 (58.33-75.00)	T1-T3	.109	83.30 (49.95-95.83)	T1-T3	1.000
Health perception T1[c]	61.05 (38.86-80.50)	T1-T2	.581	44.42 (30.53-80.48)	T1-T2	.078
Health perception T2[c]	61.05 (47.18-83.28)			77.70 (61.06-100.00)		
Health perception T3[d]	55.50 (33.31-83.29)	T1-T3	.317	88.80 (58.32-94.40)	T1-T3	.068
Emotion T1	81.65 (71.46-87.50)	T1-T2	.031 ($r = .34$)	84.97 (60.63-92.88)	T1-T2	.900
Emotion T2	83.30 (73.73-92.91)			80.00 (64.58-91.65)		
Emotion T3[b]	75.00 (66.60-91.60)	T1-T3	.694	83.30 (66.66-100.00)	T1-T3	.916
Social T1	71.42 (57.10-79.76)	T1-T2	.308	71.80 (61.90-85.70)	T1-T2	.044 ($r = .32$)
Social T2	71.81 (62.48-83.30)			66.60 (54.50-76.17)		
Social T3[b]	71.42 (66.6-85.71)	T1-T3	.054	72.20 (52.38-90.47)	T1-T3	.807
Vitality T1[c]	45.80 (33.32-54.17)	T1-T2	1.000	49.95 (33.32-64.55)	T1-T2	.043 ($r = .58$)
Vitality T2[c]	41.63 (33.30-66.62)			66.65 (45.83-91.60)		
Vitality T3[d]	50.00 (41.67-58.33)	T1-T3	.180	75.00 (49.95-85.40)	T1-T3	.176

(continued)

Table 3. (continued)

	Treatment group (n = 20)			Control group (n = 20)		
	Median (25th-75th percentiles)	Pair	*P* value (effect size)	Median (25th-75th percentiles)	Pair	*P* value (effect size)
Respiratory T1	66.60 (52.08-77.03)	T1-T2	.001 ($r = .53$)	58.30 (50.00-72.92)	T1-T2	.002 ($r = .48$)
Respiratory T2	80.54 (75.00-88.80)			79.15 (60.38-87.43)		
Respiratory T3[b]	83.30 (72.20-91.66)	T1-T3	.016 ($r = .44$)	83.30 (66.60-88.80)	T1-T3	.013 ($r = .45$)
Treatment burden T1	66.63 (55.50-77.77)	T1-T2	.014 ($r = .39$)	61.08 (47.21-77.70)	T1-T2	.026 ($r = .35$)
Treatment burden T2	77.74 (66.62-97.20)			66.60 (44.44-94.44)		
Treatment burden T3[b]	77.70 (55.50-100.00)	T1-T3	.031 ($r = .39$)	66.60 (44.40-77.77)	T1-T3	.207
Digestion T1	83.33 (66.62-100.00)	T1-T2	.024 ($r = .36$)	94.40 (66.60-100.00)	T1-T2	.523
Digestion T2	100.00 (88.88-100.00)			100.00 (66.62-100.00)		
Digestion T3[b]	100.00 (88.80-100.00)	T1-T3	.833	100.00 (66.66-100.00)	T1-T3	.336
Eating T1	100.00 (55.55-100.00)	T1-T2	.859	100.00 (66.62-100.00)	T1-T2	.306
Eating T2	88.88 (66.60-100.00)			83.29 (66.62-100.00)		
Eating T3[b]	88.88 (66.60-100.00)	T1-T3	.052	100.00 (88.80-100.00)	T1-T3	.832
Weight T1[c]	100.00 (0.00-100.00)	T1-T2	.317	33.30 (24.98-50.00)	T1-T2	.059
Weight T2[c]	100.00 (24.98-100.00)			66.60 (33.32-100.00)		
Weight T3[d]	100.00 (0.00-100.00)	T1-T3	1.000	66.60 (33.31-100.00)	T1-T3	.141
Body image T1	83.29 (66.62-100.00)	T1-T2	.123	77.77 (69.42-100.00)	T1-T2	.205
Body image T2	94.44 (66.66-100.00)			83.25 (66.66-97.22)		
Body image T3[b]	100.00 (66.60-100.00)	T1-T3	.136	88.80 (77.70-100.00)	T1-T3	.553

[a]Wilcoxon signed rank test.
[b]Treatment n = 15, control n = 15.
[c]Apply only for adolescents: treatment n = 6, control n = 6.
[d]Treatment n = 5, control n = 5.

Further, participants in both groups demonstrated reduced treatment burden at T2. As illustrated in Figure 4, the singing group continued to improve at T3, while the control group dropped to the baseline level. Both groups were receiving the standard hospital treatment, which may explain the improvement in both groups at T2. However, at T3, after the treatment group had continued their singing activities at home, only this group continued to have reduced treatment burden. This may indicate that diaphragmatic signing has an impact on perceived treatment burden over and above the effect of the standard hospital treatment or the attention from the researcher. Treatment burden is a significant problem in chronic diseases because of the number, frequency, and complexity of treatments that must be administered on a daily basis.[27] High treatment burden[27] and depressive symptoms associated with poor adherence[3] are reported in people with CF. Therefore, the finding that singing may have an effect in reducing perceived treatment burden is promising.

The enhanced emotional status in the treatment group is also important and is consistent with previous studies.[7] A qualitative study with choir singers revealed that singing improved emotional well-being and perceived physical status.[28] Young people with CF may experience a range of emotional challenges due to frequent hospitalization.[29] As our CFQ-R results indicate, the singing intervention had a positive impact on participants' emotional status. Singing

may provide a means for emotional expression, which may help individuals cope with stress and pain. Further research is required to understand young people's singing experience and its impact on coping and long-term clinical outcomes.

While pharmacological treatments may carry side effects, no adverse effects from the singing intervention were observed. Some participants reported increased coughing following singing, as singing may trigger more frequent coughing. Individuals with CF habitually cough and clear their throats, which can contribute to voice hoarseness over time.[30] Therefore, consideration should be given to voice care when patients with CF participate in singing.

Finally, there are limitations to the study that further research may help address. First, the sample size was not large enough to attain the necessary study power, which may have resulted in insignificant intergroup differences. Second, an overall dropout rate of 21% was higher than expected (10%) and may represent a bias. Third, as singing is likely to be better accepted by those who enjoy it and whose prior singing experiences were positive, there may also be a selection bias. Fourth, while the primary outcome was objective and blinded neither the participants nor the music therapist were blinded to the outcome measures. Given the nature of the intervention, blinding the participants was not feasible. Finally, MIP and MEP were chosen as surrogates of respiratory muscle strength because they are noninvasive and were feasible in the study context. However, there is a wide range in the normal values of both measures in children and adolescents,[31] and both lack data on clinically important changes for children with CF. Other tests (eg, diaphragmatic electromyography [EMG], nasal sniff pressures, and cough peak flows) also have similar limitations and were not considered feasible in our study context.

Conclusion

In children with CF who were hospitalized for an acute respiratory exacerbation, singing improved MIP and MEP and had a positive impact on QoL, although the difference between the singing and control groups was not statistically significant due to the small sample size. This study has provided pilot findings that suggest that a singing intervention is safe and effective and may be a valuable adjunct therapy to standard CF treatment. These findings warrant further research on the longer term effects of singing with a larger sample.

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Authors' Note

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Declaration of Conflicting Interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

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References

1. Chang AB, Sawyer SM. An approach to chronic cough and cystic fibrosis in children. In: Robertson D, South M, eds. *Practical Paediatrics*, 6th ed. Philadelphia, PA: Elsevier; 2006:506-514.
2. Robinson KA, McKoy N, Saldanha I, Odelola OA. Active cycle of breathing technique for cystic fibrosis. *Cochrane Database Syst Rev*. 2010;(11):CD007862.
3. Smith BA, Modi AC, Quittner AL, Wood BL. Depressive symptoms in children with cystic fibrosis and parents and its effects on adherence to airway clearance. *Pediatr Pulmonol*. 2010;45(8):756-763.
4. Ziaian T, Sawyer MG, Reynolds KE, et al. Treatment burden and health-related quality of life of children with diabetes, cystic fibrosis and asthma. *J Paediatr Child Health*. 2006;42(10):596-600.
5. Glasscoe CA, Quittner AL. Psychological interventions for people with cystic fibrosis and their families. *Cochrane Database Syst Rev*. 2008;(3):CD003148.
6. Gick ML. Singing, health and well-being: a health psychologist's review. *Psychomusicology*. 2011;21(1/2):176-207.
7. Lord VM, Cave P, Hume VJ, et al. Singing teaching as a therapy for chronic respiratory disease - a randomised controlled trial and qualitative evaluation. *BMC Pulm Med*. 2010;10:41.
8. Irons JY, Kenny DT, Chang AB. Singing for children and adults with cystic fibrosis. *Cochrane Database Syst Rev*. 2010;(5): CD008036.
9. Bolger EP, Judson MA. The therapeutic value of singing. *N Engl J Med*. 1984;311(26):1704.
10. Stacy R, Brittain K, Kerr S. Singing for health: an exploration of the issues. *Health Educ*. 2002;102(4):156.
11. Sundberg J. *The science of the singing voice*. Illinois, IL: Northern Illinois University Press; 1987.
12. Leanderson R, Sundberg J. Breathing for singing. *J Voice*. 1988; 2(1):2-12.
13. Bonilha AG, Onofre F, Vieira ML, Prado MY, Martinez JA. Effects of singing classes on pulmonary function and quality of life of COPD patients. *Int J Chron Obstruct Pulmon Dis*. 2009;4:1-8.

14. Enright S, Chatham K, Ionescu AA, Unnithan VB, Shale DJ. Inspiratory muscle training improves lung function and exercise capacity in adults with cystic fibrosis. *Chest*. 2004;126(2):405-411.
15. Nixon PA, Orenstein DM, Kelsey SF, Doershuk CF. The prognostic value of exercise testing in patients with cystic fibrosis. *N Engl J Med*. 1992;327(25):1785-1788.
16. Sawyer EH, Clanton TL. Improved pulmonary function and exercise tolerance with inspiratory muscle conditioning in children with cystic fibrosis. *Chest*. 1993;104(5):1490-1497.
17. Reid WD, El Geddes, O'Brien K, Brooks D, Crowe J. Effects of inspiratory muscle training in cystic fibrosis: a systematic review. *Clin Rehabil*. 2008;22(10-11):1003-1013.
18. Pierce RJ, Hillman D, Young IH, et al. Respiratory function tests and their application. *Respirology*. 2005;10(suppl 2):S1-S19.
19. Fauroux B, Aubertin G. Measurement of maximal pressures and the sniff manoeuvre in children. *Paediatr Respir Rev*. 2007; 8(1):90-93.
20. Miller MR, Hankinson J, Brusasco V, et al. Standardisation of spirometry. *Eur Respir J*. 2005;26(2):319-338.
21. Quittner AL, Buu A, Messer MA, Modi AC, Watrous M. Development and validation of The Cystic Fibrosis Questionnaire in the United States: a health-related quality-of-life measure for cystic fibrosis. *Chest*. 2005;128(4):2347-2354.
22. Phillips KH. *Teaching Kids to Sing*. Belmont, CA: Wadsworth Group/Thomson Learning; 1996.
23. Sonninen A, Laukkanen AM, Karma K, Hurme P. Evaluation of support in singing. *J Voice*. 2005;19(2):223-237.
24. Leanderson R, Sundberg J, von Euler C. Breathing muscle activity and subglottal pressure dynamics in singing and speech. *J Voice*. 1987;1(3):258-261.
25. Wiens ME, Reimer MA, Guyn HL. Music therapy as a treatment method for improving respiratory muscle strength in patients with advanced multiple sclerosis: a pilot study. *Rehabil Nurs*. 1999; 24(2):74-80.
26. Di Benedetto P, Cavazzon M, Mondolo F, Rugiu G, Peratoner A, Biasutti E. Voice and choral singing treatment: a new approach for speech and voice disorders in Parkinson's disease. *Eur J Phys Rehabil Med*. 2009;45(1):13-19.
27. Sawicki GS, Sellers DE, Robinson WM. High treatment burden in adults with cystic fibrosis: Challenges to disease self-management. *J Cyst Fibros*. 2009;8(2):91-96.
28. Clift S, Hancox G. The perceived benefits of singing: findings from preliminary surveys of a university college choral society. *J R Soc Promot Health*. 2001;121(4):248-256.
29. D'Auria JP, Christian BJ, Henderson ZG, Haynes B. The company they keep: the influence of peer relationships on adjustment to cystic fibrosis during adolescence. *J Pediatr Nurs*. 2000; 15(3):175-182.
30. Sataloff RT. Professional singers: the science and art of clinical care. *Am J Otolaryngol*. 1981;2(3):251-266.
31. Domenech-Clar R, Lopez-Andreu JA, Compte-Torrero L, et al. Maximal static respiratory pressures in children and adolescents. *Pediatr Pulmonol*. 2003;35(2):126-132.

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