



## AUDIT, RESEARCH AND GUIDELINE UPDATE

# Cochrane corner: psychological interventions for individuals with cystic fibrosis and their families

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## ABSTRACT

Psychological issues associated with cystic fibrosis may arise from the patients' lifelong disease- and treatment-related burden. This Cochrane Review aimed to determine psychosocial and physical outcomes of psychological interventions. Trial registries, databases and professional networks were used to identify relevant studies. Altogether, 16 studies involving 556 participants were included. They were heterogeneous in their methods, design, target groups, and outcomes. Overall, the current evidence for psychological interventions is insufficient. Preliminary evidence was available for interventions targeting specific aspects of the treatment regimen, such as behavioural nutrition interventions.

## INTRODUCTION

The growing population of individuals with cystic fibrosis (CF) face various psychological issues as a consequence of the disease burden and the complex treatment regimen. Research on psychological distress and adjustment to the disease in patients with CF indicates that most of the patients adjust well. Nevertheless, recent research highlights that symptoms of depression and anxiety are prevalent in particular subgroups of patients and their caregivers.<sup>1</sup> These findings have led to an ongoing discussion about how to identify and address potential comorbid mental health issues in clinical practice.<sup>2,3</sup>

The negative impact of psychological distress on adherence to treatment and disease outcomes are also outlined in the literature. Thus, psychological interventions for CF may aim to improve mental health and psychological function, but adherence to treatment and medical outcomes are additional challenges that need to be addressed.

In this review, we have updated a prior review to provide current evidence for psychological interventions for individuals with CF and their families.

## METHODS

Randomised controlled trials (RCTs) and quasi-randomised controlled studies evaluating psychological interventions were included provided they met the following inclusion criteria:

- ▶ Psychological interventions based on psychotherapeutic or psychosomatic techniques were applied.
- ▶ Interventions were facilitated by a psychologist or trained and supervised professional.
- ▶ Interventions were intended to improve psychological outcomes, adaptation to disease management or physiological outcomes.

- ▶ They were conducted in an individual, family or group setting.

Educational interventions to promote adherence were not included. All interventions were classified into the following types of interventions: cognitive-behavioural, cognitive, family systems or systemic, psychodynamic and other interventions. Well-being and psychological functioning, pulmonary function and weight and height or body mass index were predefined as primary outcomes. The search strategies were updated and the most recent search was run in December 2013. Three authors were involved in the screening of abstracts and checking full texts for eligibility. Data were extracted by two authors. All data were analysed using a random-effects model. Results were pooled when more than one study examined the same type of intervention, or when the intervention was comparable in their methods, setting, outcomes, participants and control condition.

More detailed information about the methods being used can be found in the original Cochrane Review.<sup>4</sup>

## RESULTS

### Included studies

The search revealed a total of 1705 titles and abstracts. Of those, 33 new studies were examined for eligibility, with eight of these studies included. Altogether, 16 studies with 556 participants were included in the current version of the review. Furthermore, 22 studies are awaiting classification and four studies were classified as ongoing. Participants were individuals with CF of all age groups (early childhood to adulthood), as well as their caregivers. Most of the studies (12) were conducted in the USA. A wide range of psychological interventions were evaluated and various designs and methods were used. Overall, the quality of studies was heterogeneous and just one study was judged to have low risk of bias across all criteria.

### Main effects of psychological interventions

The main findings are reported in this short version and details of those are demonstrated in [table 1](#). More detailed findings for each comparison of the studies in the review can be found in the original Cochrane Review.<sup>4</sup>

Five studies applying cognitive-behavioural interventions aimed to improve adherence and one study aimed to improve psychosocial adjustment. The single study addressing psychosocial adjustment used an intervention based on problem solving and developing social skills in a sample of children



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Table 1 Summary of findings

Psychological intervention/comparison	Outcomes	Illustrative comparative risks (95% CI)		Number of participants (studies)
		Assumed risk—control group(s)	Corresponding risk—intervention group(s)	
1) Cognitive behavioural 'Nutritional intervention plus behavioural management training compared with nutritional intervention alone' (children aged 4–12 years, group setting)	Secondary: <b>Total calories consumed per day</b> post-intervention	The mean ranged across control groups from 1316 to 2315 calories.	The mean in the intervention groups was <b>275.8 calories higher</b> (66.65–485.05 higher)	82 (3 studies)
	<b>Change in calorie intake</b> pre and post treatment	The mean ranged across control groups from 303.9 to 489 calories	The mean in the intervention groups was <b>364.06 calories higher</b> (191.99–536.13 higher)	82 (3 studies)
	<b>Percentage of estimated energy requirements (%EER)</b> post intervention	The mean in the control group was 127%	The mean in the intervention group was <b>21% higher</b> (7.76–34.24 higher)	67 (1 study)
	<b>Change in %EER</b> pre and post treatment	The mean of change in the control group was 27%	The mean of change in the intervention group was <b>21% higher</b> (9.22–32.78 higher)	67 (1 study)
2) Cognitive 'Decision aid for patients considering lung transplantation compared with usual care' (patients with advanced CF considering referral for lung transplantation, individual setting)	Secondary: <b>Participants' knowledge</b> four-item questionnaire (range 0–4)	The mean score of participants' knowledge in the control group was 1.974	The mean score of participants' knowledge in the intervention group was <b>0.98 higher</b> (0.66–1.31 higher)	149 (1 study)
	<b>Participants' knowledge—change in knowledge</b> week 3—baseline	The mean in the control group was 0.3	The mean in the intervention group was <b>0.94 higher</b> (0.53–1.35 higher)	149 (1 study)
	<b>Patient expectations</b> two-item questionnaire (range 0–2) 3-week follow-up	The mean in the control group was 0.58	The mean in the intervention group was <b>0.73 higher</b> (0.51–0.95 higher)	149 (1 study)
	<b>Patient expectations—change in expectation score</b> week 3—baseline	The mean in the control group was 0.05	The mean in the intervention group was <b>0.66 higher</b> (0.37–0.95 higher)	149 (1 study)
	<b>Decisional conflict—total score</b> range 0–100 (low decisional conflict to high decisional conflict) 3-week follow-up	The mean in the control group was 20.4	The mean in the intervention group was <b>8.8 lower</b> (13.7–3.9 lower)	149 (1 study)
3) Other interventions 'Biofeedback-assisted breathing re-training compared with biofeedback-assisted relaxation training' (individuals with CF; 10–41 years; individual setting)	Primary: <b>Pulmonary function—FEV<sub>1</sub>, expressed in litres</b>	The mean in the control group was 0.78	The mean in the intervention group was <b>0.54 higher</b> (0.15–0.93 higher)	24 (1 study)
	<b>Pulmonary function—forced expiratory flow 25%–75% expressed in litres per second</b>	The mean in the control group was 1.39	The mean in the intervention group was <b>0.67 higher</b> (0.1–1.24 higher)	26 (1 study)
4) Other interventions 'Massage therapy compared with bedtime reading control' (children and adolescents with CF aged 5–18 years; individual setting)	Primary: <b>Pulmonary function—peak expiratory flow rate</b> Follow-up day 30	The mean in the control group was 244	The mean in the intervention group was <b>53.9 higher</b> (43.27 lower to 151.07 higher)	20 (1 study)
	<b>Parent anxiety—State Trait Anxiety Inventory (STAI; range 0–80; higher scores=more anxiety)</b>	The mean score in the control group was 40	The mean in the intervention group was <b>9.1 lower</b> (17.84–0.36 lower)	20 (1 study)
	<b>Child anxiety—State Trait Anxiety Inventory for Children (STAIC; ranges from 0 to 80 with a higher score reflecting more anxiety)</b>	The mean child anxiety score in the control group was 32.9	The mean child anxiety score in the intervention group was <b>8.2 lower</b> (12.36–4.04 lower)	20 (1 study)

This table is based on a Cochrane Review published by Goldbeck *et al.*<sup>4</sup>

The basis for the assumed risk (eg, the median control group risk across studies) is provided in footnotes. The corresponding risk (and its 95% CI) is based on the assumed risk in the comparison group and the relative effect of the intervention (and its 95% CI).  
CF, cystic fibrosis.

with CF (8–12 years). The five studies applying cognitive-behavioural techniques used educational and cognitive-behavioural methods to improve the nutritional status of underweight children (4–12 years). Data of three of the five studies could be pooled for the variables 'total calories consumed per day', and data of two studies for 'change in weight' to evaluate the effects of behavioural management plus nutrition education versus nutrition education alone. Underweight children receiving the additional behavioural component consumed more calories

per day at post-intervention and showed significantly more increase in calorie intake compared with the group receiving nutrition education alone. No significant differences were found for the primary outcome 'change in weight'. Furthermore, one study of those five demonstrated significant differences between groups, with the combined intervention leading to higher achieved percentage of the individual estimated energy requirements (%EER) at post-intervention and greater increases in pre-post %EER.

Interventions in three studies used cognitive-only techniques. One of these studies aimed to improve adherence via motivational interviewing and one via a written self-disclosure intervention. The third study provided an evidence-based decision aid for patients being considered for lung transplantation. In this study, patients aged 18 years or older using this structured decision aid reported significant improvements of their knowledge of their options and their realistic expectations of risk of surgery at 3 weeks of follow-up. These improvements were significantly greater in the intervention group compared with the control group. Furthermore, patients receiving the cognitive intervention reported lower decisional conflict scores 3 weeks later. Group differences for change scores were not significant.

One family systems study was included, which evaluated an intervention providing support to parents and children within the CF-community. No study using a psychodynamic approach met the inclusion criteria.

Moreover, six studies providing other types of interventions, namely self-hypnosis, biofeedback, massage therapy, music therapy, telemedicine intervention to support patients awaiting transplantation and dance/movement therapy were included. In one study, biofeedback-assisted breathing re-training was compared with biofeedback-assisted relaxation training. Adolescent and adult patients using the biofeedback-assisted breathing re-training had significantly better lung function measured with FEV<sub>1</sub> expressed in litres and forced expiratory flow 25%–75% than the relaxation control group. One study evaluated massage therapy against bedtime reading for children and adolescents. Massage therapy provided by parental caregivers was superior to bedtime reading in reducing parents' anxiety and children's anxiety at 30 days of follow-up.

An overall effect of psychological interventions could not be obtained within this review due to the heterogeneous interventions and outcomes.

## DISCUSSION

Limited evidence of psychological interventions for children with CF and their caregivers is reported in this systematic review, though some of the included studies showed promising findings. There is substantial evidence supporting cognitive-behavioural interventions combined with education to improve calorie intake in children. Preliminary evidence was found for an intervention combining an educational session with a standardised decision-making aid, as well for interventions involving biofeedback, massage and music therapy.

While there is some evidence for interventions targeting specific aspects of the treatment regimen, the overall evidence on psychological interventions for individuals with CF and their families is insufficient. Important psychological outcomes, such as adherence to inhalation treatments, or comorbid symptoms of depression or anxiety, have not been targeted so far. The primary limitations of research were the lack of randomised controlled studies and the diversity of interventions used of the studies meeting inclusion criteria. Although a wide range of psychological methods, target groups, stages of the disease and outcomes have been examined, most findings are based on single studies with small samples which may limit the reliability of these results. Only cognitive-behavioural interventions have demonstrated an effect in larger samples across multiple studies.

Overall, the quality of study methods was poor. Authors' reporting of information about study protocols was particularly inadequate. Although the most recent studies paid more

rigorous attention to standards of intervention research and reporting, there is still room for improvement. CONSORT standards, which are available as well for non-pharmacological trials,<sup>5</sup> should be consulted.

For future investigations, we recommend to investigate studies addressing issues that have not yet been targeted, such as parents' adjustment to diagnosis, patients' adherence to treatment, transition to adult care, comorbid depression and anxiety, and palliative care. Replication of findings in larger multicentre studies is necessary to consolidate and extend the preliminary findings of studies in this review. New technological approaches, such as internet-based psychological interventions, should be considered to overcome the barriers for conducting large trials in an orphan disease.

With regard to clinical practices, implementation of interventions as clinical routine requires identification of individuals in need of psychological or psychotherapeutic support. The availability of professionals who are located in CF teams or who train members of the team to provide interventions must also be addressed. The results of the international epidemiological data on symptoms of anxiety and depression<sup>1</sup> initiated an open discussion about sense and necessity of routine screenings in CF care.<sup>2–3</sup> The International Guidelines Committee on Mental Health for Patients with CF and Their Caregivers is currently preparing recommendations on how to identify and treat patients and caregivers in need. Although specific intervention studies in patients with CF and comorbid mental disorders are missing, evidence-based treatments for anxiety and/or depression, such as exposure-based cognitive-behavioural therapy (CBT) for anxiety, or CBT aiming at activation and cognitive restructuring for depression should be considered for those patients and caregivers and are likely to improve their well-being and their adaptation to treatment.

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