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Self-management education for cystic fibrosis (Review)

Savage E, Beirne PV, Ni Chroinin M, Duff A, Fitzgerald T, Farrell D

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Self-management education for cystic fibrosis

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ABSTRACT

Background
Self-management education may help patients with cystic fibrosis and their families to choose, monitor and adjust treatment requirements for their illness, and also to manage the effects of illness on their lives. Although self-management education interventions have been developed for cystic fibrosis, no previous systematic review of the evidence of effectiveness of these interventions has been conducted.

Objectives
To assess the effects of self-management education interventions on improving health outcomes for patients with cystic fibrosis and their caregivers

Search strategy
We searched the Cochrane Cystic Fibrosis and Genetic Disorders Group Trials Register (date of the last search: 23 February 2011). We also searched databases through EBSCO (CINAHL; Psychological and Behavioural Sciences Collection; PsychInfo; SocINDEX) and Elsevier (EMBASE) and handsearched relevant journals and conference proceedings (date of the last searches: 30th March 2011).

Selection criteria
Randomised controlled trials, quasi-randomised controlled trials or controlled clinical trials comparing different types of self-management education for cystic fibrosis or comparing self-management education with standard care or no intervention.

Data collection and analysis
Two authors assessed trial eligibility and risk of bias. Three authors extracted data.

Main results
Four trials (involving a total of 269 participants) were included. The participants were children with cystic fibrosis and their parents or caregivers in three trials and adults with cystic fibrosis in one trial. The trials compared four different self-management education interventions versus standard treatment: (1) a training programme for managing cystic fibrosis in general; (2) education specific to...
aerosol and airway clearance treatments; (3) disease-specific nutrition education; and (4) general and disease-specific nutrition education. Training children to manage cystic fibrosis in general had no statistically significant effects on weight after six to eight weeks, mean difference -7.74 lb (95% confidence interval -35.18 to 19.70). General and disease-specific nutrition education for adults had no statistically significant effects on: pulmonary function (forced expiratory volume at one second), mean difference -5.00 % (95% confidence interval -18.10 to 8.10) at six months and mean difference -5.50 % (95% confidence interval -18.46 to 7.46) at 12 months; or weight, mean difference -0.70 kg (95% confidence interval -6.58 to 5.18) at six months and mean difference -0.70 kg (95% confidence interval -6.62 to 5.22) at 12 months; or dietary fat intake scores, mean difference 1.60 (85% confidence interval -2.90 to 6.10) at six months and mean difference 0.20 (95% confidence interval -4.08 to 4.48) at 12 months. There is some limited evidence to suggest that self-management education may improve knowledge in patients with cystic fibrosis but not in parents or caregivers. There is also some limited evidence to suggest that self-management education may result in positively changing a small number of behaviours in both patients and caregivers.

Authors’ conclusions

The available evidence from this review is of insufficient quantity and quality to draw any firm conclusions about the effects of self-management education for cystic fibrosis. Further trials are needed to investigate the effects of self-management education on a range of clinical and behavioural outcomes in children, adolescents and adults with cystic fibrosis and their caregivers.

**PLAIN LANGUAGE SUMMARY**

**Self-management education for cystic fibrosis**

We set out to review the effects of self-management education for cystic fibrosis on a range of health outcomes in individuals of all ages with cystic fibrosis and their caregivers. Our search for available evidence identified four trials, and all four compared a form of self-management education to standard treatment. The precise focus of self management differed between trials and included a training programme for managing cystic fibrosis, education on chest treatments, education on nutrition specific to cystic fibrosis, and education on general and disease-specific nutrition. Self-management education had no positive effects on lung function, weight, or intake of fatty food. There is some evidence to suggest that self-management education improves knowledge about cystic fibrosis and its management in patients with this condition and some self-management behaviours in patients and caregivers. However, due to the small number of trials in this review, and because of concerns about the quality of these trials, we are unable to reach any firm conclusions about the effects of self-management education for cystic fibrosis. We recommend that further trials are conducted to evaluate the effects of self-management education interventions.

**BACKGROUND**

**Description of the condition**

Cystic fibrosis (CF) is the most common life-limiting, autosomal recessively inherited disease in Caucasian populations with an estimated incidence of 1 per 3000 births per annum (Walters 2007). Most individuals are diagnosed in their first year of life and many countries now have newborn screening programmes. The disease manifests as pancreatic insufficiency, leading to malabsorption and failure to thrive and impaired mucociliary clearance, leading to recurrent chest infections and bronchiectasis. Advances in the treatment of this disease have resulted in a marked increase in survival rates over the past three decades and individuals can now be expected to live into their fourth decade (Dodge 2007). Nonetheless, CF remains a progressive disease involving a complex regimen of daily treatment including high fat, high calorie dietary intake, pancreatic enzyme replacement, vitamin supplementation, chest physiotherapy, nebulized medication, and antibiotic therapy in the event of respiratory infection. This daily regimen places considerable responsibility on patients and family members (especially parents of children and adolescents) to implement treatment requirements in an effort to optimise health and slow down disease progression.

**Description of the intervention**

The role that individuals with CF and family members play in the active management of their care is now seen as important for increasing the likelihood of positive health outcomes (Savage 2007; Sawicki 2007; Williams 2007). A number of self-management education interventions for patients with CF and their families, or both, have been developed since the 1990s (e.g. Bartholomew 1991; Bartholomew 1997; Downs 2006). Self-management can be described as helping patients and their families to choose, monitor and adjust treatment requirements for their illness, and also manage the effects of illness on their lives. The aim is to help them achieve the best possible health, and to fit treatment requirements into their everyday activities around a flexible management plan. The role of health care professionals is to support patients and families in this task (Newman 2004).

How the intervention might work

In order to make a difference, self-management education interventions should help patients and families to solve problems, set goals, and then plan changes in the ways they behave, so that they are motivated to manage their illness in the best possible way toward optimum health outcomes (Lorig 2003; Schreurs 2003). Traditionally, patient education programmes typically provided disease-specific knowledge aimed at increasing compliance with medical treatment and healthcare professional advice (Lorig 2002). In contrast, self-management education interventions should equip patients and families with knowledge, confidence, and skills to take responsibility for daily decisions concerning their health and to take effective control over managing the demands of chronic illness in ways that are flexible and relevant to their lives (Lorig 2002). Self-management education should work in ways that position patients and their families as ‘experts’ working in partnership with health care professionals (Department of Health 2001).

Why it is important to do this review

Although self-management education interventions for patients with CF or family members, or both, continue to be developed and advocated, there remains uncertainty over the effects of these interventions and to date no previous systematic review of the evidence has been conducted.

OBJECTIVES

To assess the effects of self-management education interventions on improving health outcomes for patients with CF and their caregivers.

METHODS

Criteria for considering studies for this review

Types of studies

Randomised controlled trials, quasi-randomised controlled trials and controlled clinical trials. Both published and unpublished studies were considered and no language restrictions were applied.

Types of participants

Individuals of all ages with a diagnosis of CF (diagnosed clinically and by sweat or genetic testing) or family members, or both.

Types of interventions

Self-management education programmes designed to help patients, of any age group, or family members, or both, to solve problems, set goals, and to plan how best to manage treatment requirements of CF in their daily lives. Education programmes were only included if a focus on self-management was explicitly specified in the aims of the programme or the content of the programme, or both. Programmes involving any structured educational or instructional approach were considered, e.g. web-based learning; computer-aided programme; video or audiotapes; written materials; one-to-one or group educational sessions. The interventions included, but were not limited to, self-management education designed to assist patients or their caregivers or both with dietary management including pancreatic enzyme replacement and vitamin supplementation, physiotherapy techniques and exercises; and medication management.

The following comparisons were considered:

1. a self-management education intervention versus another self-management educational intervention;
2. a self-management education intervention versus no intervention;
3. a self-management education intervention versus ‘standard treatment’.

Types of outcome measures

Primary outcomes

1. Pulmonary function (analysed as per cent predicted)
   i) forced expiratory volume at one second (FEV₁)
   ii) forced vital capacity (FVC)
   iii) residual volume/total lung capacity (RV/TLC)
   iv) forced expiratory flow 25–75% (FEF25–75%)
2. Indices of nutritional health or growth
Secondary outcomes

1. Self-management behaviour: any measure of the abilities of the patient (or family member, or both) to fit treatment requirements for CF into their everyday activities. For the purpose of this systematic review, we included measures of self-management skills (e.g. monitoring symptoms, monitoring calorie intake, regulating pancreatic enzymes according to fat content of food, performance of breathing techniques; goal setting and planning care; communicating about illness or aspects of care). We also included measures of independence, self-efficacy, coping, problem solving.

2. Adherence to CF treatment requirements: any measure of the patient’s or family member’s, or both, adherence including pill counts, self-report forms, diaries, electronic monitoring, prescription refill history.

3. Knowledge: any measure of the patient’s or family member’s, or both, knowledge of CF and its management.

4. Health-related quality of life: generic or disease-specific, or both: physical, psychological, social, cognitive, school functioning.

5. Utilisation of health services: e.g. number of acute hospitalisations, average length of hospital stay, clinic appointments (scheduled and unscheduled), number of visits to general practitioner, number of respiratory exacerbations requiring systemic antibiotics.

Search methods for identification of studies

Electronic searches

We identified relevant trials from the Group’s Cystic Fibrosis Trials Register using the terms: educat* OR program* OR ‘self care’ OR self-care OR selfcare OR self-management OR ‘self management’. The Cystic Fibrosis Trials Register is compiled from electronic searches of the Cochrane Central Register of Controlled Trials (Clinical Trials) (updated each new issue of The Cochrane Library), quarterly searches of MEDLINE, a search of EMBASE (to 1995 and the prospective handsearching of two journals - Pediatric Pulmonology and the Journal of Cystic Fibrosis. Unpublished work was identified by searching the book of abstracts of three major cystic fibrosis conferences: the International Cystic Fibrosis Conference; the European Cystic Fibrosis Conference and the North American Cystic Fibrosis Conference. For full details of all searching activities for the register, please see the relevant sections of the Cochrane Cystic Fibrosis and Genetic Disorders Group Module. Date of the last search of the Cochrane Cystic Fibrosis and Genetic Disorders Group Cystic Fibrosis Trials Register: 23 February 2011. We also undertook separate searches of the following databases: CINAHL with full text (EBSCO) (Appendix 1); Psychological and Behavioural Sciences Collection (EBSCO) (Appendix 2); PsychINFO (EBSCO) (Appendix 3); SocINDEX (EBSCO) (Appendix 4); Embase (Elsevier) (Appendix 5). No language restrictions were applied to separate searches of databases. Date of the last search of each of these databases: 30th March 2011.

Searching other resources

Reference lists of relevant trials identified were examined for additional citations. Specialists in the field and authors of the included trials were contacted to identify possible unpublished data.

Data collection and analysis

Selection of studies

To identify potentially eligible trials, two authors (ES, PB) independently screened the titles and abstracts of all reports gleaned through the search strategy. Where it was not possible to tell from the title and abstract whether a study was potentially eligible for inclusion, the authors retrieved full text copies of the studies. We applied no language restrictions to our search strategy. We planned to have any papers written in a foreign language translated prior to evaluating eligibility for inclusion if this could not be determined from the title and abstract (if available in the English language), or if an abstract was not available. We identified one non-English paper (French) (David 2008). One author (MNiC), who is fluent in this language, translated this paper. Two authors (ES, PB) independently read full text copies of all trials appearing to meet the inclusion criteria to determine their eligibility for inclusion in the review. We resolved any disagreements by discussion. If resolution was not possible, we planned to consult the other members of review team to adjudicate and reach consensus, however, this was not necessary.

Data extraction and management

For all trials that met the inclusion criteria, one author (ES) extracted data, two authors (PB, DF) independently cross-checked these. We resolved discrepancies by discussion. If needed, we planned to consult the other members of review team to resolve any disagreements. We used a standardised form adapted from the checklist of items in Table 7.3a in the Cochrane Handbook for...
Systematic Reviews of Interventions (Higgins 2011a), to extract data from each trial:

- general information (e.g. title, authors, citation and contact details);
- methods (trial design, randomisation process and other concerns about bias, study duration);
- participants (total number and flow of participants through trial, reasons for attrition, sample size estimations, settings, severity of illness, age and sex, details on co-morbidity);
- interventions (description of intervention including its content, mode of delivery, duration, setting, number of groups, treatment of controls);
- outcomes (primary and secondary outcomes relevant to this review, measures used, time points of data collection, intention to treat analysis);
- results (for each outcome - sample size, number of missing participants, summary data for each intervention group, estimate of effect, and subgroup analyses);
- miscellaneous (funding source, key conclusions by study authors, references to other relevant articles).

A third author (DF) cross-checked data on number of participants, mean scores and standard deviations (SD) entered into RevMan for each outcome against the data extraction forms and published records (RevMan 2011). We contacted trial authors for information either missing or unclear in published records. Where possible, we grouped outcome data into those measured at 1 to 6 months, 7 to 12 months, 13 to 18 months, 19 to 24 months and 6 monthly intervals after these time points if applicable.

Assessment of risk of bias in included studies

Two authors (ES, PB) assessed each of the included trials for risk of bias and disagreements were resolved through discussion without the need to consult other members of the review team. We assessed the risk of bias using the six specific domain-based evaluation criteria as described in the Cochrane Handbook for Systematic Reviews of Interventions 5.1 (Higgins 2011b). These were sequence generation, allocation concealment, blinding, incomplete outcome data, selective outcome reporting, and other sources of bias. For ‘other sources of bias’ we assessed efforts at minimizing cross-contamination bias (i.e. unofficial delivery of any aspect of the intervention to the ‘control’ group) including selecting intervention and control groups from different CF centres, asking participants in the ‘control’ group not to access or use self-management education material from sources such as the Internet, other CF families, CF organisations; asking participants in intervention group not to discuss the intervention with others until the end of the study; asking the control group what information about managing CF they have accessed during the course of the study. In evaluating the risk of bias for each of the six domains within each study included in the review, we made a judgement of ‘low risk’, ‘high risk’ or ‘unclear risk’ on the following basis:

1. ‘low risk’ of bias if the description of a domain indicated that it was adequately addressed;
2. ‘high risk’ of bias if the description of a domain indicated that it was not adequately addressed;
3. ‘unclear risk’ of bias if insufficient detail about a domain was reported.

Measures of treatment effect

To assess differences between groups, we recorded post-treatment mean difference (MD) values with 95% confidence intervals (CI) as our treatment effect measure for continuous variables. For dichotomous outcome data, we planned to assess treatment effects by calculating risk ratios (RR) with 95% confidence intervals (CIs). However, the trials included in this review only reported continuous outcome data.

Unit of analysis issues

For longitudinal measurements, we analysed data at each assessment time-point post treatment.

Dealing with missing data

To allow an intention-to-treat analysis, we planned to seek data on the number of participants with each outcome event, by allocated treated group, irrespective of adherence and whether or not the individual was later thought to be ineligible or otherwise excluded from treatment or follow-up. We contacted primary authors of trials to clarify data where necessary or to advise on data missing from published papers. We have listed the authors who replied to our requests for further information in the Acknowledgements section.

Assessment of heterogeneity

We planned to pool the results of studies only if they were judged to be sufficiently similar in terms of populations, interventions and outcomes. We planned to measure the inconsistency of trial results using $I^2$ statistic to determine if variation in outcomes across trials was due to heterogeneity rather than occurring by chance (Deeks 2011). The $I^2$ statistic quantifies heterogeneity in terms of overlapping percentage intervals: 0% to 40% (might not be important); 30% to 60% (may represent moderate heterogeneity); 50% to 90% (may represent substantial heterogeneity); and 75% to 100% (considerable heterogeneity) (Deeks 2011).

Assessment of reporting biases

We planned to assess funnel plot asymmetry for publication biases and other causes. However, this was not possible because tests for funnel plot asymmetry are not recommended unless there are at least 10 trials included in a meta-analysis (Sterne 2011).
Data synthesis

If we had identified studies as being clinically (e.g. similar age groups) or methodologically (e.g. similar interventions) homogeneous but statistically heterogeneous, we planned to conduct a random-effects meta-analysis. However, we did not conduct any meta-analysis in this review since studies were either clinically or methodologically diverse (or both). Conducting a meta-analysis on data from diverse studies runs the risk of obscuring genuine differences in effect (Deeks 2011). For future updates of this review, we will continue to plan for meta-analysis if appropriate. A narrative synthesis of the data is currently presented.

Subgroup analysis and investigation of heterogeneity

If we had identified substantial heterogeneity and included a sufficient number of trials, we planned to conduct subgroup analyses to investigate possible reasons for variations in results across trials. For subgroup analyses, we planned to make comparisons between subsets of participants, subsets of interventions, subsets of settings in which interventions were delivered, and subsets of personnel delivering interventions. We planned to stratify studies into:

1. participant age-group subsets (infants and toddlers up to two years, pre-school children aged 2 years to 5 years, primary school children aged 6 years to 12 years, adolescents aged 13 years to 17 years, adults aged 18 years and over);
2. intervention type (e.g. web-based learning, computer-aided programme, written materials, etc) and duration;
3. settings in which intervention was conducted (e.g. home, hospital, school);
4. personnel delivering intervention (e.g. dietitians, nurses, physicians, physiotherapists, CF advocacy or voluntary groups).

In future updates of this review, we will continue to adopt this plan for subgroup analysis and investigation of heterogeneity.

Sensitivity analysis

If appropriate, we planned to conduct sensitivity analysis to determine the influence on effect size of: published and unpublished trials; risk of bias as outlined above; length and size of studies. However, there were insufficient studies to perform this analysis.

Results of the search

A total of 123 records were identified through our search strategy as potentially relevant for inclusion. Of these, 35 citations reporting on 23 studies were identified by a search of the Cystic Fibrosis and Genetic Disorders Group’s CF Trials Register. An additional 87 records were identified from our search of individual databases. One additional record was identified in a newsletter published in Cystic Fibrosis Worldwide. Of the 123 records examined, a total of 11 records reporting on four trials were identified as meeting the inclusion criteria (Cottrell 1996; Downs 2006; Stapleton 2001; Watson 2008). Seven additional records (on six studies) were identified as potentially eligible for inclusion and are awaiting classification (Bergman 2007; Cannon 1999; Jessup 2008; Johnson 2001; Van der Gieesen 2006; Wainwright 2009). The remaining 103 records were excluded.

Included studies

The four included trials were published in peer-reviewed journals (Cottrell 1996; Downs 2006; Stapleton 2001; Watson 2008). Multiple records for three of the trials were identified: one was reported in three journal articles (Stapleton 2001); one was reported in four conference proceeding abstracts and one journal article (Watson 2008); and one was reported in an unpublished thesis and in a journal article (Cottrell 1996). For multiple records, data were extracted from the most recent publication and then from earlier publications as necessary.

Trial design

All four trials were of parallel design. Three trials were conducted in a single centre (Cottrell 1996; Stapleton 2001; Watson 2008). One trial was multicentre involving CF clinics of three public hospitals (Downs 2006). Two trials were undertaken in Australia (Downs 2006; Stapleton 2001); one in the USA (Cottrell 1996) and one in the UK (Watson 2008).

Participants

A total of 368 participants were recruited and randomised across the four trials: 139 children with CF; 155 caregivers of children; and 74 adults with CF. A total of 269 participants completed the trials: 104 children; 117 parents/caregivers (Cottrell 1996; Downs 2006; Stapleton 2001); and 48 adults (Watson 2008).

1. Children

Children with CF were included in three trials, aged 8 years to 18 years in one trial (Cottrell 1996), and 6 years to 11 years in two trials (Downs 2006; Stapleton 2001).

R E S U L T S

Description of studies

See: Characteristics of included studies; Characteristics of excluded studies; Characteristics of studies awaiting classification.
2. Caregivers/parents
Caregivers of children were included in three trials (Cottrell 1996; Downs 2006; Stapleton 2001). Caregivers were explicitly stated as parents in one trial (Cottrell 1996) and as the adult most responsible for managing children's nutrition in another trial (Stapleton 2001). It was not explicitly stated who the caregivers were in one trial (Downs 2006).

3. Adults
Adults with CF were aged 16 years to 43 years. None of the adults were waiting on a “heart/lung transplant list or were pregnant or lactating” at the time of taking part in the trial (Watson 2008).

Interventions

1. A self-management education intervention versus another self-management educational intervention
No included trial made this comparison.

2. A self-management education intervention versus no intervention
No included trial made this comparison.

3. A self-management education intervention versus 'standard treatment'
The four included trials made this comparison.

a. Self-management training programme versus standard treatment
One trial evaluated the effects of a self-management training programme in reducing the impact of CF on children and parents (Cottrell 1996). The programme was delivered in two six-hour group sessions in a hospital setting, facilitated by a registered nurse or psychologist. In the group sessions, knowledge of the nature of CF, principles of self-management, and strategies for managing CF-related problems were addressed. Skills training included problem solving and stress management.

b. Self-management education on aerosol and airway clearance treatments ('Airways') versus standard treatment
One trial evaluated the effects of an education programme ('Airways') on the self-management of aerosol and airway clearance treatments (Downs 2006). The 'Airways' programme was home based using written material containing child friendly information and behavioural exercises. Over a period of 10 weeks, children and their caregivers completed weekly exercises, each lasting approximately 20 minutes. The knowledge content of the programme drew on disciplines of medicine, physiotherapy, psychology and education. Self-management skills addressed in the programme were assessment, treatment implementation, decision making, and strategies to overcome barriers to treatment.

c. Nutrition self-management education versus standard treatment
Two trials made this comparison focusing on either disease-specific nutrition education (Stapleton 2001) or general and disease-specific nutrition education (Watson 2008).

i. Sub-comparison: Disease-specific nutrition education ('Go and Grow with CF') versus standard treatment
One trial evaluated the effects of a nutrition education programme ('Go and Grow with CF') on disease-specific nutrition knowledge and self-management skills (Stapleton 2001). The 'Go and Grow with CF' programme was home based using child-friendly written material on nutrition management. Over a period of 10 weeks, children and their caregivers completed weekly exercises, each lasting approximately 60 minutes. The programme included supplementary introductory and concluding workshops for separate groups of children and caregivers in a hospital setting, facilitated by dietitians. Knowledge content of the programme included disease-specific nutrition topics: enzymes; energy and fat; malabsorption; vitamins and minerals; growth; snacks; and salt. Self-management skills addressed in the programme were goal setting in small incremental steps to increase self efficacy, and self-monitoring adherence to daily goals.

ii. Sub-comparison: General and disease-specific nutrition education ('Eat Well with CF') versus standard treatment
One trial evaluated the effects of a nutrition education programme ('Eat Well with CF') on general and disease-specific knowledge and self-management skills training (Watson 2008). The 'Eat Well for CF' programme was home-based using written materials. Over a period of 10 weeks, participants completed weekly activities, each lasting approximately 30 minutes. Knowledge content of the programme included general and disease-specific nutrition topics: energy intake; digestion; pancreatic enzyme replacement; managing appetite; exercise; dietary fibre; reading food labels; body image. Self-management skills included goal setting in small incremental steps to establish new behaviours. The programme included supplementary group workshops (introductory weeks 5 and 10) in a hospital setting and weekly telephone calls, facilitated by dietitians. During the course of the trial, microbiological segregation was introduced following which workshops could no longer be held. Consequently, the trial was terminated.

Outcomes
Only outcomes of trials comparing 'Self-management education intervention versus standard treatment' are reported since no trials were identified for the remaining two comparisons groups considered in this review.

3. Self-management education intervention versus 'standard treatment'

a. Self-management training programme versus standard treatment

In the one trial that made this comparison, one primary and four secondary outcomes relevant to this review were assessed (Cottrell 1996). Only our pre-defined outcomes that were reported in the included trials are listed below.

Primary outcomes
1. Indices of nutritional health or growth
   Change in weight was measured in pounds using participants’ home scales (Cottrell 1996).
2. Adherence
   Medications and aerosol treatment taken by children as well as the number of chest physiotherapy sessions were assessed using a self-report diary. It was unclear whether children or parents completed the diary. Percentage ‘compliance’ was computed for each aspect of treatment by comparing reported ‘compliance’ with the schedule prescribed by physicians (Cottrell 1996).
3. Knowledge
   Knowledge was assessed using an established ‘CF Knowledge Survey’ consisting of multiple-choice questions for children, adolescents, and parents. The percentage of correct answers from each participant was recorded (Cottrell 1996).
4. Health-related quality of life
   Children's quality of life was measured using the ‘quality of well-being scale’ comprising three sub scales of functioning (mobility, physical activity, social activity) and twenty two problems/symptoms that could impair function. The total quality of life score ranged from zero (dead) to one (optimal functioning) (Cottrell 1996).

All outcomes were assessed at baseline and at six- to eight-week follow-up. Results for all outcomes were expressed as means and SDs (Cottrell 1996).

b. Self-management education on aerosol and airway clearance treatments ('Airways') versus standard treatment

In the one trial that made this comparison, three secondary outcomes relevant to this review were assessed (Downs 2006).

Secondary outcomes
1. Self-management behaviours
   Self-management behaviours of caregivers relating to aerosol and airway clearance treatments were assessed. Caregivers completed a newly developed one-week diary card constructed around three self-management sub scales: assessment; treatment; and communication. The unit of measure was a fractional score with one being the best possible score (Downs 2006).
   Self-management responsiveness of caregivers to airway clearance treatment during children's unwell days was recorded. The performance of longer and additional airway clearance treatment was considered to be responsive to the child's treatment needs. A mean responsiveness score for all unwell days was calculated (Downs 2006).
   Self-efficacy of caregivers to manage airway clearance treatments was assessed using an established 'self-efficacy scale' with five being the best possible score (Downs 2006).
2. Adherence
   Adherence to aerosol and airway clearance treatment was reported by caregivers in a one-week diary and was measured as a percentage of prescribed treatments taken by children (Downs 2006).
3. Knowledge
   Children’s knowledge on airway clearance treatment was assessed using a newly developed questionnaire with 23 being the best possible score (Downs 2006).

In this trial, adherence, self-management behaviours (assessment, treatment, communication) and self-efficacy were assessed at baseline, at immediate post test, and at 6- and 12-month follow-up. Self-management responsiveness was assessed at baseline and at post test. Knowledge was assessed at baseline, post test and at 12-month follow-up. Results for all outcomes were expressed as means and SDs (Downs 2006).

c. Nutrition self-management education versus standard treatment

i. sub-comparison: Disease-specific nutrition education ('Go and Grow with CF') versus standard treatment

In the one trial that made this sub-comparison, two secondary outcomes relevant to this review were assessed (Stapleton 2001).
Secondary outcomes

1. Self-management behaviours

Self-management skills of both children and caregivers were assessed using scenarios designed to yield open responses categorised as appropriate or inappropriate. For children, scenarios related to signs of malabsorption and communicating to caregivers about nutritional management; the highest possible scores being 23 for appropriate responses and five for inappropriate responses. For caregivers, scenarios related to malabsorption and assessment of what age they expected their children to manage their own pancreatic enzyme replacement therapy. The highest possible scores for caregivers were 61 for appropriate responses and 41 for inappropriate responses (Stapleton 2001).

3. Knowledge

Nutritional and enzyme knowledge was assessed using similar but separate newly developed questionnaires for children and caregivers. Each correct response was allocated a score of one. For children, the best possible score was 37. For caregivers, the best possible score was 42 (Stapleton 2001).

All outcomes in this trial were assessed at baseline, at immediate post-test, and at 12 month follow-up. Results for all outcomes were reported as mean score change and standard error (SE) values from baseline (Stapleton 2001). On request, the author provided unpublished data on mean differences and SEs for intervention effects.

ii. sub-comparison: General and disease-specific nutrition education ('Eat Well with CF') versus standard treatment

In the one trial that made this sub-comparison, two primary outcomes and three secondary outcomes relevant to this review were assessed (Watson 2008).

Primary outcomes

1. Pulmonary function

Watson assessed FEV1, analysed as per cent predicted (Watson 2008).

2. Indices of nutritional health or growth

Change in weight was assessed in kilograms using the same medical weighing scale for all participants (Watson 2008).

Dietary fat intake was assessed using a 17-item self-reported food frequency questionnaire, yielding a maximum score of sixty three points as the best possible score (Watson 2008).

Secondary outcomes

1. Self-management behaviour

Self-efficacy of adults to cope with a special diet was assessed using a newly developed measure, with 27 being the best possible score (Watson 2008).

3. Knowledge

Disease-specific and general nutrition knowledge were assessed using separate questionnaires adapted from previously established questionnaires designed for adults; the highest possible scores being 55 for disease-specific knowledge and 21 for general knowledge (Watson 2008).

4. Health-related quality of life

Quality of life was assessed using an established disease-specific measure for adults comprised of nine CF-specific domains (physical functioning, social functioning, treatment issues, chest symptoms, emotional responses, concerns for the future, interpersonal relationships, body image, career issues). The best possible health-related quality of life score that could be attained was 100 (Watson 2008).

All outcomes in this trial were assessed at baseline, and at 6- and 12-month follow-up. Results for all outcomes were expressed as means and SDs with the exception of quality of life, which were presented as differences in scores for each domain between intervention and control group (Watson 2008).

Excluded studies

Of the 123 records examined, 62 records were excluded following a review of title and abstracts because they were: review papers; reported on practice initiatives and were not studies; reported on instrument development; were clearly not education interventions; or did not include participants with CF. An additional 44 records (reporting on 23 studies) were excluded following review of abstracts and related full text publications because they were not RCTs, quasi-RCTs or CCTs or did not explicitly address self-management education in the aims or content of the programme. Details of the 23 excluded studies are presented in the Characteristics of excluded studies table.

Missing data

The principal authors of the four included trials were contacted for information missing from published records. Missing data in the four trials related mainly to criteria for assessing risk of bias. Three authors provided additional information (Stapleton 2001; Downs 2006; Watson 2008). Details of missing data are provided in the Characteristics of included studies table.

Studies awaiting classification

Six studies await classification, five of which were published as abstracts (Cannon 1999; Jessup 2008; Johnson 2001; Van der Gieesen 2006; Wainwright 2009). The remaining study was published in the Cystic Fibrosis Worldwide Newsletter targeting a lay and professional audience (Bergman 2007). All studies are awaiting classification because insufficient details on characteristics of the studies are available, and data on outcomes could not be extracted from publications in the format required for analysis. The principal authors of five studies have been contacted for further information (Bergman 2007; Jessup 2008; Johnson 2001; Van der Gieesen 2006; Wainwright 2009), two of whom have responded (Johnson 2001; Van der Gieesen 2006). Efforts to locate contact details on any of the authors concerning one study have failed (Cannon 1999). Information available to date on the six studies is...
Risk of bias in included studies

Based on the six domain-based evaluation criteria recommended in the Cochrane Handbook for Systematic Reviews of Interventions 5.1 (Higgins 2011b), none of the four included trials were judged as adequately meeting all criteria (Figure 1; Figure 2).

Figure 1. Risk of bias graph: review authors’ judgements about each risk of bias criterion presented as percentages across all included studies.
Figure 2. Risk of bias summary: review authors' judgements about each risk of bias criterion for each included study.

Please refer to the risk of bias tables for each individual trial located within the Characteristics of included studies table.

**Allocation**

Sequence generation was judged to be low risk in three trials (Downs 2006; Stapleton 2001; Watson 2008) and unclear in one trial (Cottrell 1996). Allocation concealment was judged to be unclear in two trials (Cottrell 1996; Stapleton 2001), high risk in one trial (Downs 2006), and low risk in one trial (Watson 2008).

**Blinding**

Details on blinding were unclear in the four trials (Cottrell 1996; Downs 2006; Stapleton 2001; Watson 2008). Blinding of participants is not possible for any of the interventions considered, however in two trials outcome assessors were blinded to at least some of the outcomes (Downs 2006; Stapleton 2001).

**Incomplete outcome data**

For incomplete outcome data, one trial was judged to be low risk (Watson 2008) and unclear in three trials (Cottrell 1996; Downs 2006; Stapleton 2001).
Selective reporting

One trial was judged to be high risk in terms of being 'free of selective reporting' (Stapleton 2001) and unclear in three trials (Cottrell 1996; Downs 2006; Watson 2008).

Other potential sources of bias

For the criterion on 'free from other bias', three trials were deemed to be low risk (Downs 2006; Stapleton 2001; Watson 2008), and high risk in one trial (Cottrell 1996).

Effects of interventions

Only the effects of 'a self-management education intervention versus standard treatment' are reported since no trials made the remaining two comparisons considered in this review. Only our pre-defined outcomes that have been reported on within the included trials are listed below. A summary of effects of interventions is presented in Table 1.

Table 1. Summary of the Effects of Interventions

<table>
<thead>
<tr>
<th>Outcome</th>
<th>Assessment time points: 6- to 8-weeks follow-up</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>PRIMARY OUTCOME (Children)</strong></td>
<td></td>
</tr>
<tr>
<td>Indices of nutritional health or growth</td>
<td></td>
</tr>
<tr>
<td>Change in weight</td>
<td>No statistically significant difference between groups in change in weight: MD -7.74 lb (95% CI -35.18 to 19.70) (Analysis 1.1).</td>
</tr>
<tr>
<td><strong>SECONDARY OUTCOMES (Children)</strong></td>
<td></td>
</tr>
<tr>
<td>Self-management behaviours</td>
<td></td>
</tr>
<tr>
<td>Number of digestive and pulmonary system behaviours</td>
<td>Statistically significantly greater number of digestive behaviours in the standard treatment group than training group, MD -5.30 (95% CI -9.29 to -1.31) (Analysis 1.2). No statistically significant difference between groups in the number of pulmonary system behaviours, MD -1.00 (95% CI -6.31 to 4.31) (Analysis 1.3). No statistically significant differences between groups for digestive system, pulmonary system, or both systems combined, MD -0.35 (95% CI -1.05 to 0.35); MD -0.28 (95% CI -0.90 to 0.34); and MD -0.18 (95% CI -0.81 to 0.45) respectively (Analysis 1.4; Analysis 1.5; Analysis 1.6).</td>
</tr>
<tr>
<td>Frequency of digestive and pulmonary system behaviours</td>
<td></td>
</tr>
<tr>
<td><strong>Adherence</strong></td>
<td></td>
</tr>
<tr>
<td>Percentages of medications, aerosol treatments, and chest physiotherapy taken.</td>
<td>No statistically significant differences between groups in the % of prescribed treatments taken by children: medications MD 2.00% (95% CI -16.31 to 20.31); aerosol treatments, MD 13.00% (95% CI -20.11 to 46.11), and chest physiotherapy, MD -8.00 % (95% CI -46.13 to 30.13) (Analysis 1.7; Analysis 1.8; Analysis 1.9).</td>
</tr>
<tr>
<td><strong>Knowledge</strong></td>
<td></td>
</tr>
<tr>
<td>Assessment time points: 6- to 8-weeks follow-up</td>
<td>Statistically significant greater knowledge scores about CF and its management in the training group than the standard treatment groups, MD 19.25% (95% CI 7.57 to 30.93) (Analysis 1.10).</td>
</tr>
<tr>
<td><strong>Health-related quality of life</strong></td>
<td></td>
</tr>
<tr>
<td>Quality of well-being</td>
<td>Assessment time points: 6- to 8-weeks follow-up</td>
</tr>
</tbody>
</table>
### Secondary Outcomes (Parents)

**Self-management behaviours**

- **Assessment time points**: 6- to 8-weeks follow-up
  - No statistically significant differences between groups for digestive system, pulmonary system, or both systems combined, MD -1.00 (95% CI -3.47 to 1.47), MD 0.40 (95% CI -2.73 to 3.53), and MD -0.60 (95% CI -5.20 to 4.00) respectively (Analysis 1.12; Analysis 1.13; Analysis 1.14).
  - No statistically significant differences between groups for digestive system, pulmonary system, or both systems combined, MD -0.09 (95% CI -0.65 to 0.47); MD 0.02 (95% CI -0.45 to 0.49); and MD 0.00 (95% CI -0.44 to 0.44) respectively (Analysis 1.15; Analysis 1.16; Analysis 1.17).

**Knowledge**

- **Assessment time points**: 6- to 8-weeks follow-up
  - No statistically significant difference between groups, MD 2.11% (95% CI -6.65 to 10.87) (Analysis 1.18).

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### Secondary Outcomes (Children)

**Adherence**

- **Assessment time points**: Post test, 6-months follow-up, 12-months follow-up
  - Statistically significant greater % of prescribed aerosol treatments taken by 'Airways' group at each time point (Analysis 2.1): Post test MD 29.70% (95% CI 14.29 to 45.11); 6-months follow-up MD 21.00% (95% CI 5.59 to 36.41); 12-months follow-up MD 17.50% (95% CI 5.50 to 29.50).
  - Statistically significant greater % of prescribed airway clearance treatments taken by 'Airways' group significantly greater at 6-months follow-up only (Analysis 2.2): Post test MD 19.00% (95% CI -0.62 to 38.62); 6 months follow-up MD 21.60% (CI 95% 7.04 to 36.16); 12-months follow-up MD 15.10% (95% CI -3.18 to 33.38).

**Knowledge**

- **Assessment time points**: Post test, 12-months follow-up
  - Significantly greater knowledge scores in the 'Airways' group at each time point (Analysis 2.3): Post test MD 3.80 (95% CI 2.29 to 5.31); 12-months follow-up MD 4.60 (95% CI 2.83 to 6.37).

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### Secondary Outcomes (Caregivers)

**Self-management behaviours**

- **Assessment time points**: Post test, 6 months follow-up, 12 months follow-up
  - Statistically significant greater assessment behaviour scores in the 'Airways' group post test only (Analysis 2.4): Post test MD 0.17 (95% CI 0.02 to 0.32); 6 months follow-up MD 0.15 (95% CI -0.01 to 0.31); 12 months follow-up MD 0.08 (95% CI -0.06 to 0.22).
  - Statistically significant greater treatment behaviour scores in the 'Airways' group at 6 month only (Analysis 2.5): Post test MD 0.07 (95% CI -0.01 to 0.15); 6 months follow-up