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Irons JY, Kenny DT, Chang AB

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[Intervention Review]

# Singing for children and adults with cystic fibrosis

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## ABSTRACT

### Background

Cystic fibrosis is a genetically inherited, life-threatening condition that affects major organs. The management of cystic fibrosis involves a multi-faceted daily treatment regimen that includes airway clearance physiotherapy, taking pancreatic enzymes and other medications. Previous studies identified that compliance with this intensive treatment especially among adolescents with cystic fibrosis is poor. Because of both the nature and consequences of the illness and the relentless demands of treatments, many individuals with cystic fibrosis are likely to have a poor quality of life. Anecdotal evidence suggests that singing may provide rigorous exercises for the whole respiratory system as well as a means for emotional expression, which may enhance quality of life.

### Objectives

To evaluate the effects of a singing intervention in addition to usual therapy on the quality of life, morbidity, respiratory muscle strength and pulmonary function of children and adults with cystic fibrosis.

### Search methods

We searched the Group's Cystic Fibrosis Trials Register, the Cochrane Central Register of Controlled Trials, major allied complementary data bases, and clinical trial registers. Hand searching for relevant conference proceedings and journals was also carried out.

Date of search of Trials Register: 02 September 2009.

Date of additional searches: 17 September 2009.

### Selection criteria

Randomised controlled trials in which singing (as an adjunctive intervention) is compared with either a sham intervention or no singing in people with cystic fibrosis.

### Data collection and analysis

No trials were found that met the selection criteria.

### Main results

No meta-analysis could be performed.

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**Authors' conclusions**

As no studies that met the criteria were found, this review is unable to support or refute the benefits of singing as a therapy for people with cystic fibrosis. Future randomised controlled trials are required to evaluate singing therapy for people with cystic fibrosis.

**PLAIN LANGUAGE SUMMARY****The effects of singing on lung function and quality of life for children and adults with cystic fibrosis**

Individuals with cystic fibrosis are at risk for respiratory infections due to too much mucus in their airways. Airway clearance is therefore an important part of cystic fibrosis management. Increasing anecdotal evidence suggests that singing may support lung function and enhance quality of life of people with cystic fibrosis. We searched for trials using the standard search module of the Cochrane Cystic Fibrosis and Genetic Disorders Group, as well as extensive searches in the relevant databases and publications. We did not find any randomised controlled trials that evaluated the benefits of singing in people with cystic fibrosis. The effects of singing in addition to standard therapy for people with cystic fibrosis remain unknown.

## BACKGROUND

### Description of the condition

Cystic fibrosis (CF) is a genetically inherited life-threatening condition that affects major organs, such as the lungs, pancreas, liver and kidneys. Individuals with CF produce abnormally excessive thick mucus in the airways, which makes them more susceptible to lung diseases. Repeated infections in the airways lead to irreversible lung damage (Phelan 1994).

The management of CF involves a multifaceted daily treatment regimen that usually includes airway clearance physiotherapy, physical exercise, taking pancreatic enzymes and other medications, as well as hospital admissions. Such an intensive treatment regime may have a negative psychosocial impact on children and adolescents (Glasscoe 2008), but particularly on adolescents (D'Auria 2000). Previous studies identified that individuals with CF are likely to have poor quality of life (QoL) (Quittner 2008).

### Description of the intervention

Several studies have reported that people such as trained singers, who learn diaphragmatic breathing, exhibit more efficient pulmonary capacity than non-trained singers (Collyer 2009; Formby 1987). In people with respiratory disease, anecdotal evidence suggests that adjunctive therapies that include breathing manoeuvres, such as singing, have significant health benefits for the disease process (Stacy 2002), as well as for psychological well being (Unwin 2002).

Singing is not merely a form of vocal expression, but also a complex physical activity. Singing requires well-controlled respiratory behaviour due to the greater range of pitch accessed during singing compared with speaking, greater length of musical phrases than spoken language, and the greater dynamic range used in singing songs. To meet these artistic as well as physical challenges, singers employ the diaphragmatic breathing method. The diaphragm as a primary inspiratory muscle generates the necessary subglottal pressure for singing. Louder and higher sounds are associated with higher lung volumes (Sundberg 1987). Respiratory muscles such as the transversus abdominis, external and internal obliques, and the intercostals must also be fully engaged to regulate air flows during singing. Thus, classically trained singers exhibit efficient breath-management and greater use of their lung capacity than non-singers (Collyer 2009; Leanderson 1988; Thomasson 1999).

Respiratory muscles also play an important role in producing effective cough, which is essential for lung health in people with CF. For an effective cough, high subglottal pressure and strong expiratory force are necessary (Kang 2006). For increasing one's ability to produce maximal expiratory pressure, it is necessary to employ diaphragmatic breathing because it helps to increase lung volumes and strengthen respiratory muscle capacity (Spapienza 2002; Wiens 1999).

Singing can also reduce fear, anxiety and pain perception (Kenny 2004) and improve mood (Unwin 2002). Further, singing can provide not only health benefits but also enjoyment. Studies found that when singing or music was part of a breathing exercise, participants with asthma demonstrated better treatment compliance due to enjoyment, which enhances motivation (Fukuda 2000; Lipawen 2000).

Additionally, a number of anecdotal reports attest to the benefits of singing in enhancing QoL of people with lung diseases (Stacy 2002). Studies investigating the effects of a singing or music program on the lung health and QoL of people with chronic obstructive pulmonary disorder (COPD), emphysema and asthma indicate that singing can be an enjoyable, low-cost and low-risk intervention, to support their lung health and enhance QoL (Engen 2005; Griggs-Drane 1999; Irons 2010; Wade 2002).

### How the intervention might work

Singing as an intervention for respiratory conditions involves, in addition to the vocal cords and laryngeal apparatus, the activation of the muscles of the entire respiratory system through diaphragmatic breathing (Sundberg 1987). Diaphragmatic breathing can increase respiratory muscle strength, which leads to increased lung volume and effective cough (Kang 2006; Wiens 1999).

Most songs contain musical phrases of greater length than spoken utterances, notes of various pitch, and changing dynamics (soft or loud), which actively engage and work the respiratory muscles.

A singing intervention can be carried out in a one-to-one or group setting in a non-judgmental and supportive environment. The program needs to be of sufficient length and intensity to allow participants to master the diaphragmatic breathing technique. This can vary from individual to individual, depending on their age, background, illness severity, past experience with singing and music generally and the relationship between the singing facilitator and the patient. A study of people with emphysema (over 60 years of age) indicated that at least two half-hour sessions are necessary for learning the diaphragmatic breathing method correctly (Engen 2005).

### Why it is important to do this review

A recent Cochrane systematic review identified very few research studies on non-medical interventions in the field of CF (Glasscoe 2008). Despite progress in medical treatment for CF, psychological therapies to improve emotional well-being have not been forthcoming (Elgudin 2004). To date, CF treatment has been dominated by rigorous medical treatments, which are, of course, essential. However, the strong emphasis on medical treatment for CF may reflect a disease-oriented view, in which only the physical symptoms are treated. A single medical treatment cannot provide the care and management required to improve all dimensions of QoL in people living with CF. A multidisciplinary treatment regimen can be effective in meeting the complex needs of individuals with CF. Research in health psychology supports this view and provides strong evidence that numerous factors influence both illness and well-being (Bernard 1994; Knight 1998). These should be taken into account when providing a health service whose aim is to optimise health outcomes, both medical and psycho-social.

Singing is a relatively inexpensive adjunct intervention that can potentially enhance QoL and de-medicalise at least some of the treatment regimens for people with CF. Thus, we have conducted a review on the efficacy of singing as an adjunctive intervention for people with CF to help guide clinical practice.

## OBJECTIVES

To assess the effects of singing as an adjunctive therapy for children and adults with CF on outcomes including QoL, morbidity, respiratory muscles and pulmonary function.

## METHODS

### Criteria for considering studies for this review

#### Types of studies

Randomised or quasi-randomised clinical trials.

#### Types of participants

People with CF, of any age, diagnosed on the basis of sweat testing or genotype analysis.

#### Types of interventions

All types of singing intervention that included diaphragmatic breathing, which were carried out in a group or one-to-one setting, facilitated by singing instructors or teachers, voice coaches or trainers, or music therapists, for a minimum of two half-hour sessions. Studies comparing singing with a sham group that did not involve the activation of the respiratory muscles were to be considered. Studies using non face-to-face delivery format, such as DVD or CD were not considered as these formats could not address some important issues such as how to use the voice in a healthy way while singing, and to obtain immediate feedback on singing practice, posture and breathing technique. Further, individual or group singing sessions can also be adjusted to the levels of singers, which is hard to achieve through media, such as DVD or CD.

#### Types of outcome measures

##### Primary outcomes

1. Quality of life measured by validated instruments, e.g. CFQ-R (Cystic Fibrosis Questionnaire-Revised), St. George's respiratory questionnaire, PedsQL™ (Pediatric Quality of Life Inventory™)
2. Respiratory muscle function
  - a. Maximal inspiratory flow
  - b. Maximal expiratory flow
  - c. Cough peak flow

##### Secondary outcomes

1. Other subjective scores (cough diary, Likert scales, visual analogue scales, subjective assessment of interference of cough etc.)
2. Spirometry
  - a. forced expiratory volume in one second (FEV<sub>1</sub>)
  - b. forced vital capacity (FVC)
  - c. forced mid-expiratory flow rate (FEF 25-75%)

3. Number of participants experiencing adverse effects of the intervention (e.g. coughing up blood and difficulty in breathing)
4. Respiratory exacerbations
  - a. total number of hospitalised days
  - b. total number of symptomatic days
  - c. number of participants with respiratory exacerbations
5. Satisfaction with the intervention (e.g. measured by a check-list, or post-intervention interviews)
6. Adherence to other CF treatments (e.g. measured by a diary, self-evaluation check-list)
7. Psychological assessments measuring self-efficacy, depression and anxiety

### Search methods for identification of studies

There were no language or publication restrictions.

#### Electronic searches

We searched the Group's Cystic Fibrosis Trials Register for relevant trials using the terms: 'cystic fibrosis' AND 'singing'.

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 is identified by searching the abstract books 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 [Cystic Fibrosis and Genetic Disorders Group Module](#).

Date of search: 02 September 2009

We also searched the following sources; please see the appendices for details of search terms and dates:

- Allied and Complementary Database AMED (1985 to present) ([Appendix 1](#));
- PsycINFO (1872 to present) ([Appendix 2](#));
- CINAHL (all years) ([Appendix 3](#));
- Dissertation Abstracts International (late 1960 to present) ([Appendix 4](#));
- [www.clinicaltrials.gov](http://www.clinicaltrials.gov) (all years) ([Appendix 5](#));
- [Music therapy research database](#) (all years) ([Appendix 6](#));
- National Research Register (NRR) Archive (2000 to 2007) ([Appendix 7](#)).

#### Searching other resources

We searched other relevant publications, including hand searching of music or singing or music therapy journals.

Journal	Date searched
Nordic Journal of Music Therapy (2003 - 2009 vol 18 (issue 2))	24/09/09
Australian Journal of Music Therapy (1996 - 2009 vol 20)	01/12/09

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The New Zealand Journal of Music Therapy (1987 - 2005)	02/12/09
The British Journal of Music therapy (1987 - 2009)	03/12/09
Musiktherapeutische Umschau (German Music Therapy Journal) (2000 - 2009)	03/12/09

Further, we have contacted experts (singing or music therapy related researchers and singing facilitators or teachers) in this area and would have had written communication with the authors of trials included in the review as necessary.

### Data collection and analysis

We did not find any eligible studies and were unable to carry out plans for analysis stated in the protocol. However, for future updates, if studies are included, we will follow the methods set out in the protocol.

### Selection of studies

Two authors (JYI, AC) independently assessed studies for inclusion in the review based on the inclusion criteria stated above. If there was any disagreement in this process, they would have consulted with the third author (DK) and resolved by discussion.

### Data extraction and management

Two authors (JYI, DK) would have independently extracted data from the eligible studies on to a standard data extraction form, and one author (JYI) would have entered the data into RevMan for analysis (RevMan 2008). DK would have checked the entered data.

Trials that satisfied the inclusion criteria would have been reviewed and the following information would have been recorded: study setting, year of study, source of funding, participants recruitment details (including number of eligible people), inclusion and exclusion criteria, other symptoms, randomisation and allocation concealment method, numbers of participants randomised, blinding (masking) of participants, care providers and outcome assessors, duration of intervention, previous singing training, co-interventions, numbers of participants not followed up, reasons for withdrawals from study protocol (clinical, side-effects, refusal and other), details on side-effects of therapy, and whether intention-to-treat analyses were possible. We would have extracted data on the outcomes described previously at two points: short term (at less than one month) and longer term (over one month and up to six months, over six months and up to one year and annually thereafter). We would have requested further information from the authors where required.

### Assessment of risk of bias in included studies

In order to assess the risk of bias, two review authors would have independently assessed the quality of the studies included in the review using the RevMan 'Risk of Bias' table as described in Chapter 8 of the Cochrane Handbook for Systematic Reviews of Interventions (Higgins 2008).

### Generation of the allocation sequence

We would have graded each study for the generation of allocation sequence as follows:

1. Low risk of bias, if methods of randomisation include using a random number table, computer-generated lists or similar methods;
2. Uncertain risk of bias, if the trial was described as randomised, but no description of the methods used to allocate participants to treatment group was described;
3. High risk of bias, if methods of randomisation include alternation, the use of case record numbers, dates of birth or day of the week, and any procedure that was entirely transparent before allocation.

### Allocation concealment

We would have assessed whether allocation was adequately concealed, to prevent both participants and investigators from foreseeing assignment.

1. Adequate (low risk of bias), if the allocation of participants involved a central independent unit, on-site locked computer, identically appearing numbered drug bottles or containers prepared by an independent pharmacist or investigator, or sealed opaque envelopes;
2. Unclear (unclear risk of bias), if the method used to conceal the allocation was not described;
3. Inadequate (high risk of bias), if the allocation sequence was known to the investigators who assigned participants, for example it was based on day of admission or case record number.

### Blinding (or masking)

Due to the nature of the intervention, it is impossible to blind participants. We would have assessed each study as to whether the outcome assessors were blinded to treatment allocation (low risk of bias); unclear whether they were blinded (unclear risk of bias); or not blinded to treatment (high risk of bias).

### Follow up

We would have graded each study as to whether numbers of and reasons for dropouts and withdrawals in all intervention groups were described; or whether it was specified that there were no dropouts or withdrawals.

1. Low risk of bias, if reasons for dropouts and withdrawals described;
2. Uncertain risk of bias, if insufficient or selective reporting of dropouts and withdrawals;
3. High risk of bias, if not reporting reasons for dropouts and withdrawals likely to be related to true outcome.

### Selective outcome reporting

We would have tried to identify and report on any selective reporting in the included trials, ideally by comparing the trial

protocol with the final published paper, but alternatively by comparing the 'Methods' and 'Results' sections of the published trial.

1. Low risk of bias, if all outcomes reported as being measured were reported;
2. Uncertain risk of bias, if it was unclear whether all measured outcomes were reported;
3. High risk of bias, if not all outcomes that were measured were reported.

#### **Other potential sources of bias**

We would have tried to identify any other sources of bias not reported elsewhere in the review and assessed their potential for putting the trial results at risk of bias.

#### **Measures of treatment effect**

An initial qualitative comparison of all the individually analysed studies would have examined whether pooling of results (meta-analysis) was reasonable. This would have taken into account differences in study populations, inclusion and exclusion criteria, interventions and outcome assessment. The results from studies that met the inclusion criteria and report any of the outcomes of interest would have been included in the subsequent meta-analyses.

For the dichotomous outcome variables of each individual study, we would have calculated the odds ratio (OR) and 95% confidence intervals (CIs) using a modified intention-to-treat analysis (modified if there are missing values due to drop outs). We would have used the Cochrane statistical package RevMan 5.0 (RevMan 2008). Numbers needed to treat (NNT) would have been calculated from the pooled OR and its 95% CI applied to a specified baseline risk (from the control group) using an online calculator (Cates 2003).

For continuous outcomes we would have calculated the mean difference (MD) and 95% CIs using RevMan 5 (RevMan 2008). If studies report outcomes using different measurement scales, the standardised mean difference would have been estimated.

#### **Unit of analysis issues**

Cross-over trials are not appropriate for this intervention and thus only the first arm of any cross-over trials would be included.

#### **Dealing with missing data**

The authors would have requested further information from the primary investigators where required.

#### **Assessment of heterogeneity**

We would have described any heterogeneity between the study results and tested this to see if it reached statistical significance using the chi-squared test. We would have considered heterogeneity to be significant when the P value was less than 0.10 (Higgins 2008). We also planned to use the  $I^2$  statistic, to quantify inconsistency of the results of the studies as described in Chapter 9 of the Cochrane Handbook for Systematic Reviews of Interventions (Higgins 2008). We would have categorised heterogeneity such that a value of under 25% was considered low, around 50% was

considered moderate and over 75% was considered a high degree of heterogeneity (Higgins 2003).

#### **Assessment of reporting biases**

If combination of data and meta-analysis (with at least 10 studies) was possible, we would have assessed publication bias using a funnel plot.

#### **Data synthesis**

We would have calculated the summary OR and mean differences with their 95% CIs using a fixed-effect model. We would have used a random-effects model whenever there were concerns about statistical heterogeneity, i.e. when there was at least moderate heterogeneity as defined above using the  $I^2$  statistic, where  $I^2$  is at least 50%.

We will only combine 'other subjective scores' (see Secondary outcome 2) if we deem it clinically appropriate.

#### **Subgroup analysis and investigation of heterogeneity**

If there were sufficient studies included in the review and it was appropriate, we planned the following a priori sub-group analyses:

1. Children (under 18 years old) versus adults;
2. Severity of exacerbation (based on FEV<sub>1</sub> where over 80% is classified as mild; 50 to 79% is classified as moderate; 30 to 49% is classified as severe; and less than 30% is classified as very severe);
3. Type of singing intervention (e.g. type of training, i.e. individual or group singing, length of follow up);
4. Intervention conducted during an acute exacerbation versus non-exacerbation state.

#### **Sensitivity analysis**

Sensitivity analyses were also planned to assess the impact of the potentially important factors on the overall outcomes, if there were sufficient studies (10):

1. Variation in the inclusion criteria;
2. Risk of bias in the included studies, (particularly whether allocation was well concealed);
3. Differences in outcome measures;
4. Analysis using random-effects model;
5. Analysis by "treatment received" or "intention-to-treat".

## **RESULTS**

### **Description of studies**

#### **Results of the search**

The electronic search yielded 194 studies. Initial screening of the abstracts of these studies was undertaken by JYI and AC. A total of 17 papers were reviewed for detailed assessment; of these, none fulfilled the inclusion criteria and two were excluded. For details, please see '[Characteristics of excluded studies](#)' below. We have identified one ongoing study (Irons 2009).

#### **Included studies**

No study met the inclusion criteria.



## Excluded studies

A music therapy study used pre-recorded music as an adjunct to regular chest physiotherapy for toddlers with CF (under 24 months old) and their caregivers (Grasso 2000). However, singing was not explored in this study. Another study investigated the effects of listening to Bach's *Magnificat* (a choir singing a religious work) on 40 adults with pneumonia or acute bronchitis (Roux 2007). Singing (as opposed to listening to music) was not part of the intervention in this study (Roux 2007).

## Risk of bias in included studies

No studies were included in the review.

## Effects of interventions

In the absence of any data, the effect of singing as a therapy for people with CF remains unknown.

## DISCUSSION

### Summary of main results

No studies could be included in this review. Two excluded studies suggest that music may be beneficial as an adjunctive management strategy for people with CF; however, these studies looked at the effects of listening to music rather than actively singing. Future research on the efficacy of a non-expensive yet potentially effective therapy on respiratory muscles and psychosocial well-being is warranted.

## AUTHORS' CONCLUSIONS

### Implications for practice

There is currently insufficient evidence to advocate singing as an adjunctive therapy in the management of people with CF.

### Implications for research

Previous studies have demonstrated that singing has the therapeutic potential to enhance the quality of life of people with chronic respiratory disease, such as emphysema, asthma, and COPD (Engen 2005; Griggs-Drane 1999; Irons 2010; Wade 2002). Singing, at least certain types of singing that use diaphragmatic breathing, can strengthen respiratory muscles. The design of future RCTs should include detailed information about the singing intervention (vocal exercises, song type, pitch and dynamic range) and delivery format (individual or group), length and frequency of sessions and prescribed singing practice time. Trials evaluating short-term (acute admission), medium-term (up to and including 12 months) and longer-term outcomes (over 12 months) are also needed. Additionally, culturally appropriate musical items should be included, taking care that different songs make similar demands on the respiratory and vocal systems. Dose response effects should be assessed.

## ACKNOWLEDGEMENTS

The review authors wish to thank the Cochrane Cystic Fibrosis and Genetic Disorders Review Group for their support.

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Wade L. A comparison of the effects of vocal exercises/singing versus music assisted relaxation on peak expiratory flow rates of children with asthma. *Music Therapy Perspectives* 2002;**20**:31-7.

**Wiens 1999**

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. *Rehabilitation Nursing* 1999;**24**(2):74-80.

## CHARACTERISTICS OF STUDIES

### Characteristics of excluded studies [ordered by study ID]

Study	Reason for exclusion
<a href="#">Grasso 2000</a>	Intervention is not singing, but using pre-recorded music during chest percussion with toddlers.
<a href="#">Roux 2007</a>	Intervention is not singing, but listening to music; people with CF were not included.

### Characteristics of ongoing studies [ordered by study ID]

**Irons 2009**

Trial name or title	The effects of singing on quality of life and lung function of children and adolescents with cystic fibrosis
Methods	RCT
Participants	young people between 7 and 17 years old
Interventions	Individual singing session during hospital admission
Outcomes	Quality of life (CFQ-R), Respiratory muscle function (maximal inspiratory flow and maximal expiratory flow), cough peak flow, spirometry

**Irons 2009** (Continued)

Starting date April 2008

Contact information [http://www.anzctr.org.au/trial\\_view.aspx?ID=83944](http://www.anzctr.org.au/trial_view.aspx?ID=83944)

Notes

RCT: randomised controlled trial

**APPENDICES****Appendix 1. AMED Search Strategy (searched on 17/09/2009)****AMED Search Strategy**

1. exp Cystic Fibrosis/
2. Cystic Fibrosis.tw
3. CF.tw
4. Mucoviscidosis.tw
5. 1 or 2 or 3 or 4
6. Sing or Singing or Singer\* or Song\*.tw
7. (Vocal\* or breath\*) adj2 exercis\*.tw
8. Choir\*.tw
9. Diaphragm\* adj2 breath\*.tw
10. 6 or 7 or 8 or 9
11. Exp Clinical trials/
12. Clinical trial.pt
13. Random\*.tw
14. Placebo.tw
15. Crossover or Cross-over.tw
16. 11 or 12 or 13 or 14 or 15
16. 5 and 10 and 16

## Key

tw: textword

Exp/: exploded AMED Thesaurus term

pt: publication type

**Appendix 2. PsychINFO Search Strategy (searched on 17/09/2009)**

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**PsycINFO Search Strategy**

---

1. Cystic Fibrosis/ [PsycINFO Thesaurus Descriptor]
  2. Cystic Fibrosis [keyword]
  3. CF [keyword]
  4. Mucoviscidosis [keyword]
  5. 1 or 2 or 3 or 4
  6. Sing or Singing or Singer\* or Song\* [keyword]
  7. (Vocal\* or breath\*) adj2 exercis\*[keyword]
  8. Choir\*[keyword]
  9. Diaphragm\* adj2 breath\* [keyword]
  10. 6 or 7 or 8 or 9
  11. Clinical trials [PsychINFO Thesaurus Descriptor]
  12. Random\* [keyword]
  13. Placebo.tw [keyword]
  14. Crossover or Cross-over [keyword]
  15. 11 or 12 or 13 or 14
  16. 5 and 10 and 15
- 

**Appendix 3. CINAHL Search Strategy (searched on 08/12/2009)**

---

**CINAHL Search Strategy**

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1. (MH "Cystic Fibrosis") [CINAHL Subject Heading]
  2. Cystic Fibrosis [all text]
  3. CF [all text]
  4. Mucoviscidosis [all text]
  5. 1 or 2 or 3 or 4
  6. Sing or Singing or Singers or Song\* [all text]
  7. (Vocal\* or breath\*) adj2 exercis\* [all text]
  8. Choir\* [all text]
  9. Diaphragm\* adj2 breath\* [all text]
  10. 6 or 7 or 8 or 9
  11. (MH "Clinical Trials+") [Exploded CINAHL Subject heading]
  12. Clinical trial [publication type]
-

---

(Continued)

13. Random\* [all text]
  14. Placebo [all text]
  15. Crossover or Cross-over [all text]
  16. 11 or 12 or 13 or 15
  17. 5 and 10 and 16
- 

#### **Appendix 4. ProQuest Search Strategy: Dissertation Abstracts International (late 1960 to present) (searched on 23/09/2009 and 24/09/2009)**

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ProQuest Search Strategy: Dissertation Abstracts International (late 1960 to present)

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1. [as key words in citation and abstract] Cystic fibrosis
  2. [as key words in citation and abstract] Cystic fibrosis AND Breathing
  3. [as key words in citation and abstract] Cystic fibrosis AND Voice
  4. [as key words in citation and abstract] Cystic fibrosis AND music
  5. [as key words in citation and abstract] Cystic fibrosis AND singing
  6. [as key words in citation and abstract] Music therapy AND breathing
  7. [as key words in citation and abstract] Singing AND breathing
  8. [as key words in citation and abstract] singing OR breathing exercise AND respiratory
  9. [as key words in citation and abstract] choir AND effect
- 

#### **Appendix 5. Clinicaltrials.gov Search Strategy (searched on 18/09/2009)**

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##### **Clinicaltrials.gov Search Strategy**

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Advanced Search

Condition: Cystic Fibrosis

Interventions: sing/singing/singer/song/diaphragmatic/vocal/choir

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#### **Appendix 6. Music therapy research database ([www.musictherapyworld.de](http://www.musictherapyworld.de)) Search Strategy (searched on 24/09/2009)**

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##### **Music therapy research database ([www.musictherapyworld.de](http://www.musictherapyworld.de)) Search Strategy**

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No electronic search was possible using search terms. Thus, all abstracts were reviewed.

1. Dissertations on Music Therapy (1994 - 2006)
- 

**Singing for children and adults with cystic fibrosis (Review)**

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(Continued)

2. Conference Reports Archive (1993 - 2001)

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## Appendix 7. National Research Register Archive Search Strategy (2000 - 2007)

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### National Research Register Archive Search Strategy

cystic fibrosis AND sing OR singing OR breathing OR choir OR vocal

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## CONTRIBUTIONS OF AUTHORS

<i>Protocol stage:</i> draft the protocol	JY Irons, A Chang, DT Kenny
<i>Review stage:</i> select which trials to include (2 + 1 arbiter)	JY Irons, A Chang + DT Kenny
<i>Review stage:</i> draft the final review	JY Irons, DT Kenny, A Chang
<i>Update stage:</i> update the review	JY Irons

## DECLARATIONS OF INTEREST

The authors are currently conducting a single-blinded randomised controlled trial with inpatients with CF.

## INDEX TERMS

### Medical Subject Headings (MeSH)

Cystic Fibrosis [\*therapy]; Music Therapy [\*methods]; Randomized Controlled Trials as Topic

### MeSH check words

Adolescent; Adult; Child; Humans