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[Intervention Protocol]

Singing for children and adults with bronchiectasis

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ABSTRACT

This is a protocol for a Cochrane Review (Intervention). The objectives are as follows:

To evaluate the effects of a singing program as an adjunctive therapy on the morbidity, respiratory muscles and pulmonary function of children and adults with bronchiectasis during

- (a) stable bronchiectasis and
- (b) an exacerbation of bronchiectasis

BACKGROUND

Description of the condition

(Please see [Appendix 1](#) for explanations of medical terminology used.)

Bronchiectasis, commonly termed an 'orphan disease' is increasingly recognized as a major cause of respiratory morbidity especially in developing countries ([Karadag 2005](#), [Karakoc 2001](#)) and in pockets of affluent countries ([Chang 2008](#)). The underlying aetiology of bronchiectasis varies from post recurrent respiratory infections to rare immune deficiencies. However, bronchiectasis is also a common pathway for a variety of diseases. Thus, the presence of bronchiectasis is also increasingly recognised in common (e.g. chronic obstructive pulmonary disease (COPD) ([O'Brien 2000](#)) and uncommon respiratory diseases (e.g. bronchiolitis obliterans and sarcoidosis) as well non primary respiratory (e.g. autoimmune) diseases. When bronchiectasis is present with another underlying disorder, it increases the morbidity and mortality of the underlying diseases ([Keistinen 1997](#)). For example, in diseases like COPD the presence of bronchiectasis has been reported in 29-50% ([O'Brien 2000](#)) and when present, increases the severity and frequency ([Gursel 2006](#)) of respiratory exacerbations. Thus, management of the symptoms and severity of bronchiectasis is important

The definition of bronchiectasis by Laenec was originally based on post mortem histopathology in 1819. Later, bronchograms, which were first described in 1951 became the gold standard and this has now largely been replaced by high-resolution computerized tomography (HRCT) scans of the chest. Currently bronchiectasis is defined by 'irreversible dilatation of peripheral airways', is usually diagnostically established radiologically by chest HRCT scans. The key features of bronchiectasis in HRCT scans are dilated bronchi in the periphery of the lung and bronchial wall thickening, and lacking of tapering. On a clinical level, particularly in children, this radiology-based definition is problematic. It is not known at what stage of the disease process HRCT signs of bronchiectasis occur, and CT definitions are from adult populations; moreover, chest HRCT scans performed in different states of health may yield different results ([Chang 2008b](#)).

The dominant symptoms and signs of bronchiectasis are productive or wet cough, dyspnoea on exertion and presence of other respiratory signs (clubbing, chest wall deformity, respiratory noises such as wheeze or crepitations on auscultation) ([Chang 2008b](#)). In the long term, pulmonary decline may occur. Also, like patients with COPD, children and adults with bronchiectasis also suffer from recurrent acute exacerbations, some of whom require hospitalised treatment ([Chang 2008](#)). Effective management regimes for bronchiectasis aim to improve quality of life (QoL) and could reduce the frequency or severity of respiratory exacerbations, and/or the long term pulmonary decline ([Chang 2008](#)).

In people with respiratory disease, anecdotal evidence suggests that adjunctive therapies that include breathing maneuvers, such as singing, have significant health benefits for the disease process ([Stacy 2002](#)) as well as for the psychological well being ([Unwin 2002](#)). In Engen and colleagues' non-controlled small study in adults with COPD, a singing program improved functional outcomes but had no effect on physical health (FEV₁, distance walked) ([Engen 2005](#)). Other studies have described that due to

diaphragmatic breathing, trained singers exhibit more efficient pulmonary capacity than non-trained singers ([Formby 1987](#), [Collyer 2008](#)).

Description of the intervention

Singing requires well-controlled respiratory manoeuvres due to high/low pitch of notes, long/short musical phrases and various dynamics in 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 subglottic pressure for singing. Louder and higher sounds are associated with higher lung volumes. Respiratory muscles such as transversus abdominis, external and internal obliques, and the intercostals are also 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 ([Leanderson 1988](#); [Thomasson 1999](#), [Collyer 2008](#)).

Respiratory muscles play an important role in producing effective cough, which is essential for lung health in patients with bronchiectasis. For an effective cough, high subglottic 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 assists with increased lung volumes and strengthened respiratory muscle capacity ([Sapienza 2002](#)).

Singing can provide not only health benefits but also enjoyment. Studies found that when singing/music was part of a breathing exercise, participants with asthma demonstrated better treatment compliance due to enjoyment, which reinforces motivation ([Fukuda 2000](#), [Lipawen 2000](#), [Wiens 1999](#))

Additionally, a number of anecdotal reports attest to the benefits of singing in enhancing QoL of people with bronchiectasis. Studies investigating the effects of a singing/music program on the lung health and QoL of people with COPD, emphysema and asthma indicate that singing could also be an enjoyable, low-cost and low-risk intervention for people suffering from bronchiectasis, to support their lung health and enhance QoL ([Griggs-Drane 1999](#), [Engen 2005](#); [Wade 2002](#)).

How the intervention might work

Singing as an intervention for respiratory conditions involves diaphragmatic breathing, not only use of the vocal cords, but activation of the muscles of the entire respiratory system. Diaphragmatic breathing during singing activates respiratory muscles, which support efficient deep inhalation and slow exhalation ([Sundberg 1987](#)). This increases respiratory muscle strength, which increases lung volume and assists effective coughing ([Kang 2006](#)).

Most songs contain musical phrases of greater length than spoken utterances, notes of various pitch, and changing dynamics (soft/loud), which actively engage 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 the participants to master the diaphragmatic breathing technique. This can vary from patient to patient, depending on their

age, background, illness severity and the relationship between the singing facilitator and the patient. A study with emphysema patients (> 60 age) indicates 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

Although there is some evidence suggesting as a relatively low-cost adjunct therapy to more conventional regimes, with the potential of improving health outcomes, no systemic review of singing in bronchiectasis has been carried out.

OBJECTIVES

To evaluate the effects of a singing program as an adjunctive therapy on the morbidity, respiratory muscles and pulmonary function of children and adults with bronchiectasis during

- (a) stable bronchiectasis and
- (b) an exacerbation of bronchiectasis

METHODS

Criteria for considering studies for this review

Types of studies

All randomised controlled trials will be considered.

Types of participants

Children or adults with bronchiectasis (defined clinically or radiologically) not related to cystic fibrosis. Exclusion criteria: Participants with cystic fibrosis or with other diseases where bronchiectasis is not present. (see objectives and Note below)

Types of interventions

All types of singing program that include diaphragmatic breathing, which are carried out in a group (choir) or one-to-one setting with a singing teacher or instructor for minimum of two half-hour sessions. Studies comparing singing with a sham group that does not involve activation of the respiratory muscles will be considered. Studies using non face-to-face delivery format, such as using DVD or CD, will not be considered.

Types of outcome measures

Primary outcomes

1. Mean difference in quality of life
2. Mean difference in respiratory function that reflect respiratory muscle function (maximal inspiratory and expiratory flow, cough peak flow)

Secondary outcomes

1. Mean difference in other subjective scores (cough diary, Likert scale, visual analogue scale, level of interference of cough, cough diary, etc)
2. Mean difference in other lung function indices (spirometry, other lung volumes)
3. Proportions experiencing adverse effects of the intervention, (e.g. haemoptysis)
4. Total number of hospitalised days or symptomatic days

For studies in stable state, additional secondary outcomes are

5. Proportions of participants who had respiratory exacerbations and/or hospitalisations
6. Satisfaction with the intervention
7. Treatment adherence
8. Psychological assessments measuring self-efficacy, depression and anxiety

Search methods for identification of studies

Electronic searches

The following topic search strategy will be used to identify the relevant randomised controlled trials listed on the electronic databases:

("bronchiectasis" OR "suppurative lung disease" as (textword) or (MeSH)) AND ("singing" OR "songs" OR "sing" OR "music" or "voice training" or "diaphragm" OR "breathing exercise" OR "voice" OR "voice training" OR "vocalise" OR "choir" as (textword) or (MeSH))

Trials will be identified from the following sources:

1. The Cochrane Airways Group Specialised Trials Register (please see the [Airways Group Module](#) for details)
2. The Cochrane Central Register of Controlled Trials (CENTRAL)
3. MEDLINE (1966 to present). Topic search strategy combined with the RCT search filter as outlined in the Airways Group module.
4. OLDMEDLINE (1950 to 1965). Topic search strategy combined with the RCT search filter as outlined in the Airways Group module.
5. EMBASE (1980 to present). Topic search strategy combined with the RCT search filter as outlined in the Airways Group module.
6. CINAHL (1982 to present). Cumulative Index to Nursing and Allied Health Literature.
7. AMED (1985 to present). Allied and Complementary Database
8. NRR (2000 to 2007). National Research Register Archive
9. Clinical Trials Register (www.clinicaltrial.gov)
10. PsycINFO (1872 to present)
11. LILACS (Latin American and Caribbean Health Science Literature)
12. Dissertation Abstracts International (late 1960 to present)
13. INSIDE (www.bl.uk/online/inside). British Library Database for journals and conference proceedings

Searching other resources

14. References in relevant publications and conference proceedings
15. Music Therapy Research CD Rom (AMTA 1999), Music Therapy World Info CD Rom (Aldridge 2002)

16. Written communication with the authors of trials included in the review as necessary.

We do not apply any language restrictions.

Data collection and analysis

Selection of studies

Retrieval of studies: From the title, abstract, or descriptors, two reviewers (JS, AC) will independently review the literature search results to identify potentially relevant trials for full review. Searches of bibliographies and texts will be conducted to identify additional studies. From the full text using specific criteria, the same two reviewers will independently select trials for inclusion. Disagreement will be resolved by third party adjudication (DK).

Data extraction and management

Trials that satisfy the inclusion criteria will be reviewed and the following information 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 patients 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. Data will be extracted on the outcomes described previously. Further information will be requested from the authors where required.

Assessment of risk of bias in included studies

In order to assess the risk of bias, two review authors will independently assess the quality of the studies included in the review using the RevMan 'Risk of Bias' table.

Allocation concealment

Whilst blinding may not always be possible, concealment of the treatment group at the time participants are enrolled into the study will be assessed.

Generation of the allocation sequence

Each study will be graded for allocation concealment as follows:

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

Blinding (or masking)

Each study will be graded for blinding as follows:

1. blinding of outcome assessor to treatment allocation.

Follow up

Each study will be graded as to whether numbers of and reasons for dropouts and withdrawals in all intervention groups were described; or if it was specified that there were no dropouts or withdrawals.

Dealing with missing data

The authors will request further information from the primary investigators where required.

Assessment of heterogeneity

We will describe any heterogeneity between the study results and test this to see if it reached statistical significance using the chi-squared test. We will consider heterogeneity to be significant when the P value is less than 0.10 (Higgins 2008). We also plan to use the I² statistic, where heterogeneity is categorised such that a value of under 25% is considered low, around 50% is considered moderate and over 75% is considered a high degree of heterogeneity (Higgins 2003).

Assessment of reporting biases

If we can combine data and meta-analysis is possible, we will assess publication bias using a funnel plot. We will try and identify and report on any selective reporting in the included trials.

Data synthesis

For the dichotomous outcome variables of each individual study, odds ratio (OR) will be calculated using a modified intention-to-treat analysis. An initial qualitative comparison of all the individually analysed studies will examine whether pooling of results (meta-analysis) is reasonable. This will take into account differences in study populations, inclusion/exclusion criteria, interventions and outcome assessment.

The results from studies that meet the inclusion criteria and report any of the outcomes of interest will be included in the subsequent meta-analyses. The summary weighted odds ratio and 95% confidence interval (fixed effects model) will be calculated (Cochrane statistical package, RevMan version 5).

Numbers needed to treat (NNT) will be calculated from the pooled OR and its 95% CI applied to a specified baseline risk using an online calculator (Cates 2003). If studies report outcomes using different measurement scales, the standardised mean difference will be estimated. Any heterogeneity between the study results will be described and explored. The 95% confidence interval estimated using a random effects model will be included whenever there are concerns about statistical heterogeneity.

Subgroup analysis and investigation of heterogeneity

The following a priori sub-group analyses are planned:

1. Children (aged 18 years or less) and adults (>18 years)
2. Participant type (bronchiectasis as primary disease vs bronchiectasis as co-existent disease)
3. Type of singing intervention (e.g. short term: less than one month; medium term: 1-6 months; long term > 6 months; type of training)
4. Stable state vs exacerbation phases of bronchiectasis

5. Severity of bronchiectasis (based on FEV1: >80% classified as mild, 50-79% classified as moderate, 30-49% classified as severe, <30% classified as very severe)

Sensitivity analysis

Sensitivity analyses are also planned to assess the impact of the potentially important factors on the overall outcomes:

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"

ACKNOWLEDGEMENTS

We thank Toby Lasserson, Dr Chris Cates and Elizabeth Arnold from the Airways Group for their advice, supportive role and comments to the protocol.

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APPENDICES
Appendix 1. Term used

Term used	Lay Description
Aetiology	Causes of disease
Crepitations	Sounds heard in the chest using a stethoscope
Dyspnoea	Difficulty in breathing
FEV1	Measurement of expiratory flow using a spirometer - detects lung disease related to airway obstruction
Morbidity	illness or disease
Orphan disease	A rare disease in the developed world or one that is poorly researched
HRCT scans	High Resolution CT scan of the chest - a CT technique for evaluation of small airway disease
Haemoptysis	Coughing up blood

WHAT'S NEW

Date	Event	Description
11 May 2009	Amended	Contact author name amended

CONTRIBUTIONS OF AUTHORS

All authors wrote the protocol based on previous protocols. JS is a music therapist, DK has music and psychology expertise and AC has clinical and research expertise in respiratory medicine.

DECLARATIONS OF INTEREST

The authors are currently conducting a RCT on the efficacy of a singing program in children and adolescence with cystic fibrosis.

SOURCES OF SUPPORT
Internal sources

- Royal Children's Hospital Foundation, Australia.

Research support for AC

Singing for children and adults with bronchiectasis (Protocol)

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External sources

- NHMRC, Australia.
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- Queensland Smart State Funds, Australia.
Support for AC
- Australian Cochrane Airways Group Network Award, Australia.
Scholarship for JS

INDEX TERMS**Medical Subject Headings (MeSH)**

*Breathing Exercises; *Voice Training; Bronchiectasis [*therapy]; Music Therapy [*methods]; Randomized Controlled Trials as Topic

MeSH check words

Adult; Child; Humans