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[Intervention Protocol]

Singing for children and adults with cystic fibrosis

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ABSTRACT

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

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

BACKGROUND

Description of the condition

Cystic Fibrosis (CF) is a common life-threatening genetically inherited 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 vulnerable and susceptible for 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, taking pancreatic enzymes and other medications, as well as hospital admissions. Such an intensive treatment regime may cause negative psychosocial impact on children and adolescents (Glasscoe 2008), but particularly in 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 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 assists with increased lung volumes and strengthened 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 2009; 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 (under 60 years of 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

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 a review on the efficacy of singing as an adjunctive intervention for people with CF would help guide clinical practice.

OBJECTIVES

To assess the effects of singing as an adjunctive therapy for children and adults with CF on outcomes including the 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 include diaphragmatic breathing, which are 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 does not involve the activation of the respiratory muscles will be considered. Studies using non face-to-face delivery format, such as DVD or CD will not be considered as these formats do not address some important issues such as how to use the voice in healthy way while singing, and to obtain immediate feedback on singing practice, the posture and the breathing technique. Further, individual or group singing sessions can also be adjusted to the levels of singers, which is hard to be achieved through pre-made mediums, 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^{TM} (Pediatric Quality of Life Inventory^{TM})
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 scale, visual analogue scale, level of interference of cough etc.)
2. Spirometry
   a. forced expiratory volume in one second (FEV_1)
   b. forced vital capacity (FVC)
   c. forced mid-expiratory flow rate (FEF 25-75%)
3. Number of participants experiencing adverse effects of the intervention (see Appendix 1 for details)
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 will be no language or publication restrictions.

Electronic searches

Relevant trials will be identified from the Group's Cystic Fibrosis Trials Register.

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.

We also plan to search the following sources:

- Allied and Complementary Database AMED (1985 to present)
- National Research Register (NRR) Archive (2000 to 2007)
- www.clinicaltrials.gov
- PsycINFO (1872 to present)
- CINAHL
- Dissertation Abstracts International (late 1960 to present)
- Music therapy research database (www.musictherapyworld.de)

Searching other resources

We also plan to search other relevant publications, including handsearching of music or singing or music therapy journals. Further, we will contact experts (singing or music therapy related researchers and singing facilitators or teachers) in this area and have written communication with the authors of trials included in the review as necessary.

Data collection and analysis

Selection of studies

One author (JYI) will run the searches and make records of search results. Two authors (JYI, AC) will independently select studies for inclusion in the review based on the inclusion criteria stated above. If there is any disagreement in this process, they will consult with the third author (DK) and resolve the issue by discussion. Studies excluded for the review will be listed in the 'Characteristics of Excluded Studies' and the reason for exclusion will be given.

Data extraction and management

Two authors (JYI, DK) will independently extract data from the eligible studies on to a standard data extraction form, and one author (JYI) will enter the data into RevMan for analysis (RevMan 2008). DK will check the entered data.

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 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. Data will be extracted 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). 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 as described in Chapter 8 of the Cochrane Handbook for Systematic Reviews of Interventions (Higgins 2008).

Generation of the allocation sequence

Each study will be graded 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 is 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 is entirely transparent before allocation.

Allocation concealment

We will assess whether allocation was adequately concealed, to prevent neither the participants nor the investigators could foresee assignment, for example, when using central allocation, opaque sealed envelopes, etc.

Blinding (or masking)

Due to nature of the intervention, it is impossible to blind participants. We will assess each study as to whether the outcome assessors were blinded 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.

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.

Measures of treatment effect

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 and 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.

For the dichotomous outcome variables of each individual study, we will calculate 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 will use the Cochrane statistical package RevMan 5.0 (RevMan 2008). Numbers needed to treat (NNT) will be 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 will calculate the mean difference and 95% CIs using RevMan 5 (RevMan 2008). If studies report outcomes using different measurement scales, the standardised mean difference will be 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 will be included.

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 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 will categorise heterogeneity 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 combination of data and meta-analysis (with at least 10 studies) 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, ideally by comparing the trial protocol with the final published paper, but alternatively by comparing the 'Methods' and 'Results' sections of the published trial.

Data synthesis

We will calculate the summary OR and mean differences with their 95% CIs using a fixed-effect model. We will use a random-effects model whenever there are concerns about statistical heterogeneity.

Subgroup analysis and investigation of heterogeneity

If there are sufficient studies included in the review and it is appropriate, we plan the following a priori sub-group analyses:

1. Children (under 18 years old) versus adults
2. Severity of exacerbation (based on FEV_1 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 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

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APPENDICES

Appendix 1. Possible adverse events

possible adverse events	lay description
h aemoptysis	coughing up blood
dyspnoea	difficulty in breathing

CONTRIBUTIONS OF AUTHORS

Protocol stage: draft the protocol	JY Irons, A Chang, D Kenny
Review stage: select which trials to include (2 + 1 arbiter)	JY Irons, A Chang + D Kenny
Review stage: extract data from trials (2 people)	JY Irons, D Kenny
Review stage: enter data into RevMan	JY Irons
Review stage: carry out the analysis	JY Irons, A Chang
Review stage: interpret the analysis	JY Irons, D Kenny, A Chang
Review stage: draft the final review	JY Irons, D Kenny, A Chang
Update stage: 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