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Singing for adults with chronic obstructive pulmonary disease (COPD)

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Abstract

Background—Singing is a complex physical activity dependent on the use of the lungs for air supply to regulate airflow and create large lung volumes. In singing, exhalation is active and requires active diaphragm contraction and good posture. Chronic obstructive pulmonary disease (COPD) is a progressive, chronic lung disease characterised by airflow obstruction. Singing is an activity with potential to improve health outcomes in people with COPD.

Objectives—To determine the effect of singing on health-related quality of life and dyspnoea in people with COPD.

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CONTRIBUTIONS OF AUTHORS

RJM: protocol initiation, development and writing; additional literature search; retrieval of papers; study screening, quality appraisal and data extraction; author contact; data entry; data analysis; data interpretation; manuscript writing.

CE: protocol development and writing; additional literature search; retrieval of papers; study screening, quality appraisal and data extraction; data analysis; data interpretation; manuscript writing.

EC: protocol development and writing; data interpretation; manuscript writing; manuscript review.

ZJM: protocol development and writing; data interpretation; manuscript review.

DECLARATIONS OF INTEREST

RJM: none known.

CE: none known.

EC: none known.

ZJM: none known.

DIFFERENCES BETWEEN PROTOCOL AND REVIEW

We added clarification regarding the types of interventions: the criterion 'in the case of interventions combining one or more components of music therapy, for example instrumental and singing training, the singing must form the majority of the intervention' was added.

Summary of findings table: only the primary outcomes of health-related quality of life and dyspnoea were reported. Secondary outcomes of respiratory muscle strength and adverse events were not added due to only one study and no data, respectively, being available.

Subgroup and sensitivity analyses were not performed due to the small number of trials included.

Heterogeneity was considered significant if the P value was less than 0.10 (Higgins 2011).

Search methods—We identified trials from the Cochrane Airways Specialised Register, ClinicalTrials.gov, the World Health Organization trials portal and PEDro, from their inception to August 2017. We also reviewed reference lists of all primary studies and review articles for additional references.

Selection criteria—We included randomised controlled trials in people with stable COPD, in which structured supervised singing training of at least four sessions over four weeks' total duration was performed. The singing could be performed individually or as part of a group (choir) facilitated by a singing leader. Studies were included if they compared: 1) singing versus no intervention (usual care) or another control intervention; or 2) singing plus pulmonary rehabilitation versus pulmonary rehabilitation alone.

Data collection and analysis—Two review authors independently screened and selected trials for inclusion, extracted outcome data and assessed risk of bias. We contacted authors of trials for missing data. We calculated mean differences (MDs) using a random-effects model. We were only able to analyse data for the comparison of singing versus no intervention or a control group.

Main results—Three studies (a total of 112 participants) were included. All studies randomised participants to a singing group or a control group. The comparison groups included a film workshop, handcraft work, and no intervention. The frequency of the singing intervention in the studies ranged from 1 to 2 times a week over a 6 to 24 week period. The duration of each singing session was 60 minutes.

All studies included participants diagnosed with COPD with a mean age ranging from 67 to 72 years and a mean forced expiratory volume in one second (FEV₁) ranging from 37% to 64% of predicted values. The sample size of included studies was small (33 to 43 participants) and overall study quality was low to very low. Blinding of personnel and participants was not possible due to the physical nature of the intervention, and selection and reporting bias was present in two studies.

For the primary outcome of health-related quality of life, there was no statistically significant improvement in the St George's Respiratory Questionnaire total score (mean difference (MD) -0.82, 95% confidence interval (CI) -4.67 to 3.02, 2 studies, n = 58, low-quality evidence). However, there was a statistically significant improvement in the SF-36 Physical Component Summary (PCS) score favouring the singing group (MD 12.64, 95% CI 5.50 to 19.77, 2 studies, n = 52, low-quality evidence). Only one study reported results for the other primary outcome of dyspnoea, in which the mean improvement in Baseline Dyspnoea Index (BDI) score favouring the singing group was not statistically significant (MD 0.40, 95% CI -0.65 to 1.45, 1 study, n = 30, very low-quality evidence).

No studies examined any long-term outcomes and no adverse events or side effects were reported.

Authors' conclusions—There is low to very low-quality evidence that singing is safe for people with COPD and improves physical health (as measured by the SF-36 physical component score), but not dyspnoea or respiratory-specific quality of life. The evidence is limited due to the low number of studies and the small sample size of each study. No evidence exists examining the long-term effect of singing for people with COPD. The absence of studies examining singing performed in conjunction with pulmonary rehabilitation precludes the formulation of conclusions about the effects of singing in this context. More randomised controlled trials with larger sample sizes and long-term follow-up, and trials examining the effect of singing in addition to pulmonary

rehabilitation, are required to determine the effect of singing on health-related quality of life and dyspnoea in people with COPD.

INDEX TERMS: Medical Subject Headings (MeSH)

*Quality of Life; Dyspnea [therapy]; Pulmonary Disease; Chronic Obstructive [*therapy]; Singing [*physiology]; Time Factors

MeSH check words

Aged; Humans

PLAIN LANGUAGE SUMMARY**Singing for COPD**

Singing uses the lungs to provide airflow to produce musical words or sounds with the voice. Singing can require a lot of effort for muscle contraction and co-ordination. This may benefit people with chronic obstructive pulmonary disease (COPD) in a manner similar to that of breathing exercises. Singing is said to be beneficial for health but we need evidence for this before it can be recommended specifically to address health conditions. We planned to examine whether singing had any effect on quality of life or breathlessness in people with COPD. We included three studies with a total of 112 participants. Participants were randomly assigned to singing training or to a non-singing control group. The control groups were either a film workshop, handcraft work, or nothing at all. The singing was performed in groups, once to twice a week for one hour, for a minimum of six weeks. There was diversity in the results of the studies and we were unable to combine many results in 'meta-analyses'. A meta-analysis is a statistical analysis which combines the results of two or more separate studies to give a pooled result. Some studies showed improvements in some aspects of quality of life, while others showed no improvement. Breathlessness was only measured in one study and no improvement was found. The studies did not report whether any effects lasted for a long time after the singing training was completed. No studies reported any side effects from singing, so singing appears to be safe for people with COPD. The studies were of low quality due to the small number of participants and missing information about the methods and some of the outcomes. We were unable to find enough evidence to sufficiently determine the effect of singing in people with COPD. More studies are required and they should concentrate on enrolling larger numbers of people.

SUMMARY OF FINDINGS FOR THE MAIN COMPARISON [Explanation]

Singing compared with control for chronic obstructive pulmonary disease (COPD)						
Patient or population: people with stable COPD Settings: hospital and community Intervention: singing Comparison: control						
Outcomes	Illustrative comparative risks [*] (95% CI)		Relative effect (95% CI)	No of Participants (studies)	Quality of the evidence (GRADE)	Comments
	Assumed risk	Corresponding risk				
	Control	Singing				
Health-related quality of life (respiratory specific) St George's Respiratory Questionnaire (SGRQ; total score) Scale from 0–100 Lower value post intervention is favourable, indicating improvement in health-related quality of life Follow-up: end of intervention (range 6 to 24 weeks)	The mean change in SGRQ (total score) ranged across control groups from –5.0 to –0.4	The mean change in SGRQ (total score) in the intervention groups was 0.8 units higher (3.0 units lower to 4.7 units higher)		58 (2 studies)	⊕⊕○○ low ^{1,2}	The minimal important difference is 4 units lower
Health-related quality of life (generic) SF-36 (Physical Component Summary (PCS) score) Scale from 0–100 Higher value post intervention is favourable, indicating improvement in health-related quality of life Follow-up: end of intervention (range 6 to 8 weeks)	The mean change in SF-36 (PCS score) ranged across control groups from –3.8 to –2.5	The mean change in SF-36 (PCS score) in the intervention groups was 12.6 units higher (5.5 units higher to 19.8 units higher)		52 (2 studies)	⊕⊕○○ low ^{3,4}	The minimal important difference is 4 units higher
Health-related quality of life (generic) SF-36 (Mental Component Summary (MCS) score) Scale from 0–100 Higher value post intervention is favourable, indicating	The mean change in SF-36 (MCS score) ranged across control groups from –3.2 to 4.3	The mean change in SF-36 (MCS score) in the intervention groups was 5.4 units higher (3.9 units lower to 14.7 units higher)		52 (2 studies)	⊕⊕○○ low ^{2,4}	The minimal important difference is 4 units higher

Singing compared with control for chronic obstructive pulmonary disease (COPD)

Patient or population: people with stable COPD

Settings: hospital and community

Intervention: singing

Comparison: control

Outcomes	Illustrative comparative risks* (95% CI)		Relative effect (95% CI)	No of Participants (studies)	Quality of the evidence (GRADE)	Comments
	Assumed risk	Corresponding risk				
	Control	Singing				
improvement in health-related quality of life Follow-up: end of intervention (range 6–8 weeks)						
Dyspnoea Basal Dyspnea Index (BDI) (score) Scale from 0–12 Higher value post intervention is favourable, indicating improvement in dyspnoea Follow-up: end of intervention (24 weeks)	The mean change in BDI (score) was 0.3	The mean change in BDI (score) in the intervention groups was 0.4 units higher (0.7 units lower to 1.5 units higher)		30 (1 study)	⊕○○○ very low ^{5,6}	The minimal important difference is 1 unit higher

* The **corresponding risk** (and its 95% confidence interval) is based on the assumed risk in the comparison group and the relative effect of the intervention (and its 95% CI).

CI: Confidence interval

GRADE Working Group grades of evidence

High quality: Further research is very unlikely to change our confidence in the estimate of effect.

Moderate quality: Further research is likely to have an important impact on our confidence in the estimate of effect and may change the estimate.

Low quality: Further research is very likely to have an important impact on our confidence in the estimate of effect and is likely to change the estimate.

Very low quality: We are very uncertain about the estimate.

¹ One study showed limitations in design: selection and detection bias unknown (risk of bias –1)

² Meta-analysis was limited to few studies with small sample sizes and wide confidence intervals (imprecision –1)

³ Meta-analysis was limited to few studies with small sample sizes (imprecision –1)

⁴ One study showed reporting bias (risk of bias –1)

⁵ Study showed limitations in design: selection and detection bias unknown (risk of bias –1)

⁶ No meta-analysis as only one study (imprecision –2)

BACKGROUND

Description of the condition

Chronic obstructive pulmonary disease (COPD) is a progressive lung disease characterised by airflow limitation that is not fully reversible and is associated with an abnormal inflammatory response of the lungs to noxious particles or gases (GOLD 2017). The

prevalence of COPD has been reported as ranging from 0.2% to 37%, and varies widely across countries and populations (Rycroft 2012). Prevalence and incidence is greatest in men and those aged 75 years and over (Rycroft 2012). COPD is a major cause of morbidity and is the third most common cause of death globally (Lozano 2012). People with COPD have abnormal respiratory muscle function as a result of a mechanical disadvantage due to lung hyperinflation, diaphragmatic dysfunction and asynchronous breathing, and a reduction in respiratory muscle strength (Luce 1982). Symptoms of COPD include dyspnoea (breathlessness), cough and fatigue. Postures that enhance the function of the diaphragm may assist with dyspnoea relief (Luce 1982). Reduced functional capacity and physical inactivity are common features (Troosters 2010; Singer 2011), which significantly increase the risk for hospitalisation and mortality (Garcia-Aymerich 2006). Pulmonary rehabilitation, encompassing exercise training, education and behaviour change (Spruit 2013), is an important component in the management of COPD and is beneficial in relieving dyspnoea and fatigue, and improving health-related quality of life and exercise capacity (McCarthy 2015).

Description of the intervention

Singing is the production of musical words or sounds with the voice (Oxford 2016). Singing can be performed individually or in a group (choir), and can be arranged or improvised. Singing is a much more complex physical activity than speaking due to the greater length of phrases and greater range of pitch required (Irons 2010). Singing is dependent on the use of the lungs for air supply. During normal tidal breathing, the diaphragm contracts for inhalation, while exhalation occurs passively. During singing, air flow must be regulated and larger lung volumes are required, thus exhalation is active and aided by the abdominal, internal intercostal and pelvic muscles. Singing requires a high degree of muscle co-ordination by highly developed muscle reflexes. There are four stages of breathing with singing: inhalation; suspension; controlled exhalation (when phonation occurs); and recovery. A singer controls these stages consciously until they become conditioned reflexes (Mathis 2009).

Diaphragmatic breathing requires an increase in abdominal wall motion with a reduction in upper rib cage motion (Gosselink 2004), and is the method of breathing employed by singers, as the diaphragm can generate the greatest inspiratory muscle force to increase lung volumes and change subglottal pressures necessary for singing (Sundberg 1993). The subglottal air pressure requirements are much greater for singing tasks than for speaking tasks (Leanderson 1987; Leanderson 1988), as higher subglottal pressures are required for loudness and higher pitch (Sundberg 1993). Audible speech can be produced with subglottal pressures as low as 2 cmH₂O (centimetre of water pressure), with ordinary speech ranging from 7 cmH₂O to 10 cmH₂O; however, singing can vary from 5 cmH₂O to 40 cmH₂O for soft to loud tones (Proctor 1980). An increase in subglottal pressure is achieved by decreasing the volume of the rib cage using muscular forces, elasticity forces and gravity (Sundberg 1993).

Posture can greatly affect the quantity of air, the capacity of the lungs and the ability to move air in and out when singing. Good posture facilitates an efficient breathing pattern and

can influence the voice (Bunch 1995; Staes 2011). Trained singers have greater breathing efficiency and greater use of their lung capacity than non-trained singers (Gould 1973; Salomoni 2016).

Mastery of diaphragmatic breathing is vital for singing. Data from Engen 2005 suggests a minimum of four half-hour group singing sessions could be sufficient for people with emphysema to learn the diaphragmatic breathing technique correctly. Thus, singing needs to be performed for a sufficient duration, and most likely at a sufficient intensity in order to ensure an effective stimulus for learning this technique and for potentially having an effect on important health outcomes. The precise 'dosage' will likely vary for each person and may depend on their age, disease severity, and previous experience with singing (Irons 2010).

How the intervention might work

Singing is an activity that has the potential to improve health outcomes, such as relieving dyspnoea and enhancing quality of life, in people with COPD due to employment of diaphragmatic breathing, altered posture, and improved breathing co-ordination. Qualitative studies of singing and health report that singing can enhance mood, provide social support and friendship, help develop self esteem and self confidence, relieve stress, promote good posture and distract attention from personal worries (MacDonald 2012). Singing in people with COPD has the potential to demonstrate similar effects due to the enjoyable and low-risk nature of the activity (Engen 2005), and may have a positive impact on the distressing effects of COPD such as breathlessness, reduced quality of life and fatigue. The perceptions of people with COPD following a group singing programme support this (Morrison 2013; Skingley 2014).

Therapies that incorporate breathing manoeuvres, such as controlled breathing techniques including diaphragmatic breathing and active expiration (as performed during singing), have been shown to improve lung function (Esteve 1996), alleviate dyspnoea and improve quality of life (Gosselink 2003; Gosselink 2004), and improve functional exercise capacity (Holland 2012) in people with COPD. Singing requires great control to ensure a smooth and sustained exhalation. This exhalation is similar to that of pursed lip breathing and controlled breathing, which have been shown to reduce breathlessness in people with COPD (Gosselink 2003; Bianchi 2004).

Education on breathing and air support is fundamental in the process of learning to sing, and knowledge of the physical processes that make up the act of singing, and how those processes function (Mathis 2009), may improve breathing awareness and efficiency in people with COPD.

Poor posture (hyperkyphosis), which is common in people with COPD (Gaude 2014), can restrict the expansion of the rib cage and movement of the diaphragm. Singing requires the development of skills in controlling posture that may be transferable to activities in daily life for people with COPD (Lord 2010).

Why it is important to do this review

Singing may have the potential to improve health outcomes in people with COPD. Systematic reviews of research literature have been completed for singing in other chronic respiratory diseases such as bronchiectasis (Irons 2010), and cystic fibrosis (Irons 2016), and found an absence of randomised controlled trials (RCTs) to support or refute the benefits of singing. However, the authors of these reviews found studies that reported an improvement in quality of life in people with COPD, and a systematic review of singing for COPD has not yet been carried out. Furthermore, whilst pulmonary rehabilitation improves physical and psychosocial health outcomes in people with COPD (McCarthy 2015), the potential additional benefits of adding singing to pulmonary rehabilitation has not been examined.

OBJECTIVES

To determine the effect of singing on health-related quality of life and dyspnoea in people with COPD.

METHODS

Criteria for considering studies for this review

Types of studies—We included randomised controlled trials (RCTs) reported as full-text, published as abstract only, and unpublished data. We used data from studies published as abstract only where authors provided study data. Where the data were not available, we recorded the studies as awaiting classification.

Types of participants—We included studies that involved adults with COPD, diagnosed according to the investigators' definition, of any age or disease severity. The COPD was required to be stable (i.e. optimal and stable respiratory medications with no exacerbation or hospitalisation within the previous month). We included participants with COPD who used supplemental oxygen. Participants with and without a history of singing training could be included, and we recorded the singing training history wherever possible.

Types of interventions—We included studies examining structured, supervised singing training of at least four weeks' duration with a minimum of four sessions. Studies were included that compared:

1. singing versus no intervention (usual care) or another control intervention;
2. singing plus pulmonary rehabilitation versus pulmonary rehabilitation alone.

The singing could be performed individually or as part of a group (choir) facilitated by a singing leader, and inpatient and outpatient programmes were included. In the case of interventions combining one or more components of music therapy, for example instrumental and singing training, the singing needed to form the majority of the intervention. We recorded the precise nature of the singing facilitators' professional backgrounds, singing training and any pulmonary rehabilitation programme (frequency, duration, type, intensity), wherever possible.

Types of outcome measures

Primary outcomes

1. Health-related quality of life, measured using total scores from either generic or respiratory-specific quality-of-life questionnaires.
2. Dyspnoea, measured using a dyspnoea scale (e.g. Medical Research Council (MRC) dyspnoea scale (Bestall 1999)) or dyspnoea scores from a respiratory-specific quality-of-life questionnaire (e.g. dyspnoea domain of the Chronic Respiratory Disease Questionnaire (Guyatt 1987)), or both.

Secondary outcomes

1. Respiratory muscle strength measured from a pressure gauge (e.g. maximal inspiratory and expiratory mouth pressures or maximal sniff nasal inspiratory pressure).
2. Pulmonary function measured by spirometry or plethysmography (e.g. forced expiratory volume in one second (FEV₁) measured in litres or as per cent of predicted, forced vital capacity (FVC), FEV₁/FVC ratio, total lung capacity (TLC), residual capacity (RC), functional residual capacity (FRC)).
3. Psychological status measured from generic psychological questionnaires or scales (e.g. Hospital Anxiety and Depression Scale (Zigmond 1983)).
4. Functional exercise capacity measured from a functional exercise test.
5. Peak exercise capacity measured from a peak exercise test.
6. Endurance exercise capacity measured from an endurance exercise test.
7. Healthcare utilisation recorded as hospitalisation or length of hospital stay, or both.
8. Physical activity level from objective measurement tools (e.g. pedometers, accelerometers, multi-sensor devices).
9. Adverse events/side effects.

Reporting one or more of the outcomes listed here in the study was not an inclusion criterion for the review. We reviewed primary and secondary outcomes at baseline and immediately following the intervention period. If outcomes were also measured in the long term (e.g. six or 12 months after completion of intervention), we reviewed each of these time points in addition to immediately following the intervention period. The selected primary outcome measures are important to patients and clinicians, and all outcome measures were clinically relevant and could potentially be altered by a singing intervention.

Search methods for identification of studies

Electronic searches—We identified trials from the Cochrane Airways Trials Register, which is maintained by the Information Specialist for the group. The Register contains studies identified from several sources:

1. monthly searches of the Cochrane Central Register of Controlled Trials (CENTRAL), through the Cochrane Register of Studies Online (crso.cochrane.org);
2. weekly searches of MEDLINE Ovid SP 1946 to date;
3. weekly searches of Embase Ovid SP 1974 to date;
4. monthly searches of PsycINFO Ovid SP 1967 to date;
5. monthly searches of CINAHL EBSCO (Cumulative Index to Nursing and Allied Health Literature) 1937 to date;
6. monthly searches of AMED EBSCO (Allied and Complementary Medicine);
7. handsearches of the proceedings of major respiratory conferences.

Studies contained in the Trials Register are identified through search strategies based on the scope of Cochrane Airways. Details of these strategies, as well as a list of handsearched conference proceedings are in Appendix 1. See Appendix 2 for search terms used to identify studies for this review.

We also conducted a search of ClinicalTrials.gov (www.ClinicalTrials.gov) and the World Health Organization (WHO) trials portal (www.who.int/ictrp/en/) and PEDro (www.pedro.org.au/). We searched all databases from their inception to 1 August 2017, and we imposed no restriction on language of publication.

Searching other resources—We checked reference lists of all primary studies and review articles for additional references. We searched for errata or retractions from included studies published in full-text on PubMed (www.ncbi.nlm.nih.gov/pubmed).

Data collection and analysis

Selection of studies—Two review authors (RJM, CE) independently screened titles and abstracts of all the potential studies we identified as a result of the search and coded them as 'retrieve' (eligible or potentially eligible/unclear) or 'do not retrieve'. We retrieved the full-text study reports or publications and two review authors (RJM, CE) independently screened the full text and identified studies for inclusion. They also identified and recorded reasons for excluding ineligible studies. We resolved any disagreement through discussion or we consulted a third review author (ZJM). We identified and excluded duplicates and collated multiple reports of the same study so that each study, rather than each report, was the unit of interest in the review. We recorded the selection process in sufficient detail to complete a PRISMA flow diagram (Figure 1) and 'Characteristics of excluded studies' table (Moher 2009).

Data extraction and management—We used a data collection form for study characteristics and outcome data. One review author (RJM) extracted study characteristics from included studies. A second review author (CE) spot-checked study characteristics for accuracy against the trial report. We extracted the following study characteristics:

1. methods: study design, total duration of study, details of any 'run-in' period, number of study centres and location, study setting, withdrawals and date of study;
2. participants: number, mean age, age range, gender, severity of condition, diagnostic criteria, baseline lung function, smoking history, inclusion criteria and exclusion criteria;
3. interventions: intervention, comparison, concomitant medications and excluded medications;
4. outcomes: primary and secondary outcomes specified and collected, and time points reported;
5. notes: funding for trial and notable conflicts of interest of trial authors.

Two review authors (RJM, CE) independently extracted outcome data from included studies. One review author (RJM) transferred data into Review Manager 5 (RevMan 2014). We double-checked that data were entered correctly by comparing the data presented in the systematic review with the study reports.

Assessment of risk of bias in included studies—Two review authors (RJM, CE) independently assessed risk of bias for each study using the criteria outlined in the *Cochrane Handbook for Systematic Reviews of Interventions* (Higgins 2011). We assessed the risk of bias according to the following domains:

1. random sequence generation;
2. allocation concealment;
3. blinding of participants and personnel;
4. blinding of outcome assessment;
5. incomplete outcome data;
6. selective outcome reporting;
7. other bias.

We graded each potential source of bias as high, low or unclear and provided a quote from the study report together with a justification for our judgement in the 'Risk of bias' table. We summarised the 'Risk of bias' judgements across different studies for each of the domains listed.

When considering treatment effects, we took into account the risk of bias for the studies that contributed to that outcome.

Assessment of bias in conducting the systematic review—We conducted this review according to the published protocol (Differences between protocol and review).

Measures of treatment effect—We analysed continuous data as mean differences (MDs). We entered data presented as a scale with a consistent direction of effect. We

undertook meta-analyses only where this was meaningful, that is, if the treatments, participants and the underlying clinical question were similar enough for pooling to make sense.

We narratively described skewed data reported as medians and interquartile ranges.

Where multiple trial arms were reported in a single trial, we included only the relevant arms. If two comparisons were combined in the same meta-analysis, we halved the control group to avoid double counting.

Unit of analysis issues—We did not include cross-over trials. If the search identified cluster-randomised trials, the intention was to consult the *Cochrane Handbook for Systematic Reviews of Interventions* (Higgins 2011), however no cluster-randomised trials were identified.

Dealing with missing data—We contacted investigators in order to verify key study characteristics and obtain missing numerical outcome data where possible (e.g. when a study was identified as abstract only).

Assessment of heterogeneity—We used the I^2 statistic to measure heterogeneity among the studies in each analysis. Heterogeneity was considered significant if the P value was less than 0.10 (Higgins 2011).

Assessment of reporting biases—We were unable to pool more than 10 studies, however if more studies are included in future review updates we will create and examine a funnel plot to explore possible small-study and publication biases.

Data synthesis—We used a random-effects model using Review Manager 5 (RevMan 2014) and used change from baseline results to final scores.

Where the outcomes were reported using adjusted analyses (such as ANOVA or ANCOVA), we used the generic inverse variance method to combine the results with other studies; where adjusted analyses were not available, we preferred change from baseline results to final scores.

'Summary of findings' table—We created a 'Summary of findings' table using the following outcomes: health-related quality of life and dyspnoea. We used the five GRADE considerations (study limitations, consistency of effect, imprecision, indirectness and publication bias) to assess the quality of the body of evidence as it related to the studies that contributed data to the meta-analyses for the prespecified outcomes. We used methods and recommendations described in Section 8.5 and Chapter 12 of the *Cochrane Handbook for Systematic Reviews of Interventions* (Higgins 2011), and GRADEpro software (GRADEpro GDT). We justified all decisions to downgrade or upgrade the quality of studies using footnotes, and we have made comments to aid the reader's understanding of the review where necessary.

Subgroup analysis and investigation of heterogeneity—We planned to carry out the following subgroup analyses if sufficient studies were retrieved:

1. severity of lung disease - severe (FEV₁ % predicted less than 40%) versus not severe (FEV₁ % predicted 40% predicted or greater);
2. mode of singing intervention - individual versus group (choir);
3. participant's experience with singing training - no previous history with singing training versus prior history of singing training;
4. singing facilitator's professional background - formally trained music or singing professional versus health or lay professional.

We planned to use the following outcomes in subgroup analyses:

1. health-related quality of life;
2. dyspnoea.

We were unable to perform subgroup analyses due to the small number of studies. If more studies are included in future updates of this review, we will perform subgroup analyses using the formal test for subgroup interactions in Review Manager 5 (RevMan 2014).

Sensitivity analysis—We planned to carry out the following sensitivity analysis:

1. studies with a low risk of bias (to examine the effects of removing studies with a high risk of bias).

We were unable to conduct this sensitivity analysis because of the small number of studies. If more studies are included in future review updates, a sensitivity analysis will be performed to analyse the effects of studies with a low risk of bias for at least three of the following domains: random sequence generation, allocation concealment, blinding of outcome assessment, and incomplete outcome data.

RESULTS

Description of studies

Refer to the Characteristics of included studies, Characteristics of excluded studies and Characteristics of studies awaiting classification for the details of the studies included, excluded and awaiting classification.

Results of the search—Our search of the databases identified 50 citations. We identified three additional citations through handsearching. After removing duplicates, we reviewed 49 citation titles and abstracts, of which we excluded 38. We screened the full-text versions of eleven citations for eligibility, and excluded two because they did not meet the review inclusion criteria. Nine citations were appropriate for inclusion in the review, however one citation was published in abstract form only and our attempts to contact the authors were unsuccessful. This study remains as a study awaiting classification. The remaining eight citations represented three studies. We created a PRISMA flow diagram to depict the search

results (Figure 1). The review authors agreed on the inclusion of all citations, with a Cohen's kappa measurement of 1, indicating excellent agreement.

Included studies—We identified three studies (a total of 112 participants) which met the inclusion criteria for this review. They were represented by eight citations which were reviewed in full-text. The full details of these studies can be found in the Characteristics of included studies table.

The sample size of studies ranged from 33 to 43 participants. All studies included participants diagnosed with chronic obstructive pulmonary disease (COPD), with a mean age ranging from 67 to 72 years and a mean forced expiratory volume in one second (FEV₁) ranging from 37% to 64% of predicted values.

All studies randomised participants to a singing group or another control intervention. The frequency of the singing intervention in the studies ranged from 1 to 2 times a week over a 6 to 24 week period. The duration of the singing sessions was 60 minutes and they were conducted in groups led by a singing teacher. The singing sessions were structured in nature and included relaxation exercises, breathing exercises, vocalisation exercises, and singing. All studies began with relaxation exercises of the neck and upper limb muscles, or postural work and physical stretches. One study had participants perform singing-related breathing exercises consisting of fast, deep inspirations, followed by slow, full or interrupted expirations; performing fast and deep respiratory incursions, paying attention to the upper abdominal movements; and generating breathing movements against, or with the help of, pressures generated by a hand placed on the upper abdominal region (Bonilha 2009). All studies performed vocal exercises, for example, pronouncing vowels such as “le”, “la”, “mi”, “mu”, and singing the melody of a familiar song using such vowels instead of actually singing the lyrics (Bonilha 2009). In all the studies, participants sang songs for 20 to 30 minutes. The comparison groups included a film workshop (Lord 2012), handcraft work (Bonilha 2009), and no intervention (Lord 2010).

The primary outcome of health-related quality of life was measured in all three studies, using the St George's Respiratory Questionnaire total score (Bonilha 2009; Lord 2010), the SF-36 Physical Component Summary (PCS) and Mental Component Summary (MCS) scores (Lord 2010; Lord 2012), and the COPD Assessment Test (CAT) total score (Lord 2012). The primary outcome of dyspnoea was assessed in one study using the Basal Dyspnea Index (BDI) (Bonilha 2009).

The following secondary outcomes were measured by the included studies: respiratory muscle strength (Bonilha 2009); pulmonary function (Bonilha 2009); psychological status (Lord 2010; Lord 2012); peak exercise capacity (Lord 2010; Lord 2012); and physical activity level (Lord 2012).

The following secondary outcomes were not measured by the included studies: functional exercise capacity; endurance exercise capacity; and healthcare utilisation.

Excluded studies—We excluded two citations (representing one study) from this review due to the intervention not meeting the inclusion criteria.

Risk of bias in included studies

Our assessment of the risk of bias in the included studies is presented in Figure 2 and Figure 3.

Allocation—All three studies reported participant randomisation, however one study did not provide sufficient information to determine the sequence generation or allocation concealment (Bonilha 2009).

Blinding—Blinding of personnel and participants was not possible due to the physical nature of the intervention. Two studies reported blinding of the outcome assessor (Lord 2010; Lord 2012).

Incomplete outcome data—All three studies reported dropouts and loss to follow-up ranging from 22% to 30% (Bonilha 2009; Lord 2010; Lord 2012).

Selective reporting—Two studies reported all outcome measures as prespecified in the methods (Bonilha 2009; Lord 2010). One study did not report all outcomes at the post intervention time point as prespecified in the methods (Lord 2012).

Other potential sources of bias—All three studies appeared to be free of other sources of bias.

Effects of interventions

See: Summary of findings for the main comparison

See Summary of findings for the main comparison.

The Data and analyses table summarises the results of the meta-analyses for the comparison of singing to a control group. All three studies reported the results as change from baseline measures.

Primary outcomes

Health-related quality of life: Two studies reported results which could be pooled for meta-analysis for the St George's Respiratory Questionnaire (Bonilha 2009; Lord 2010). Results of the meta-analysis are shown in Figure 4. There was no statistically significant improvement in the St George's Respiratory Questionnaire total score (mean difference (MD) -0.82, 95% confidence interval (CI) -4.67 to 3.02, $n = 58$). We assessed the quality of the evidence as low according to GRADE criteria (Summary of findings for the main comparison).

Two studies reported results which could be pooled for meta-analysis for the SF-36 (Lord 2010; Lord 2012). Results of the meta-analysis are shown in Figure 5. There was a statistically significant improvement in the SF-36 Physical Component Summary (PCS) score favouring the singing group (MD 12.64, 95% CI 5.50 to 19.77, $n = 52$). There was no statistically significant improvement in the SF-36 Mental Component Summary (MCS) score, but the confidence interval is wide (MD 5.42, 95% CI -3.90 to 14.74, $n = 52$). We

assessed the quality of the evidence as low according to GRADE criteria (Summary of findings for the main comparison).

Dyspnoea: Only one study (Bonilha 2009) reported results for dyspnoea (Figure 6). The mean improvement in the Basal Dyspnoea Index (BDI) score favouring the singing group was not statistically significant (MD 0.40, 95% CI -0.65 to 1.45, n = 30). We assessed the quality of the evidence as very low according to GRADE criteria (Summary of findings for the main comparison).

Secondary outcomes

Respiratory muscle strength: One study reported measures of respiratory muscle strength (Bonilha 2009). There was an improvement in maximal inspiratory pressure (P_Imax, cmH₂O) favouring singing, but the confidence interval is too wide to exclude a possible reduction in inspiratory muscle pressure with the intervention (MD 4.00, 95% CI -8.49 to 16.49, n = 30).

There was a statistically significant improvement in maximal expiratory pressure (P_Emax, cmH₂O) favouring the singing group (MD 14.30, 95% CI 0.87 to 27.73, n = 30).

Pulmonary function: One study (Bonilha 2009) reported measures of pulmonary function in 30 participants, including forced expiratory volume in one second (FEV₁), forced vital capacity (FVC), FEV₁/FVC ratio, end respiratory volume (ERV), and inspiratory capacity (IC). There were no statistically significant differences between the singing group and control group for any of these measures (FEV₁ litres (L) MD -0.03, 95% CI -0.20 to 0.14; FVC (L) MD -0.04, 95% CI -0.33 to 0.25; FEV₁/FVC (%) MD 0.40, 95% CI -4.05 to 4.85; ERV (L) MD 0.17, 95% CI -0.06 to 0.40; IC (L) MD -0.16, 95% CI -0.37 to 0.05).

Psychological status: Two studies reported results which could be pooled for meta-analysis for the Hospital Anxiety and Depression Scale (HADS) (Lord 2010; Lord 2012). There was no statistically significant improvement in the HADS anxiety score (MD -1.09, 95% CI -3.02 to 0.83, n = 52) or HADS depression score (MD -0.87, 95% CI -2.16 to 0.42, n = 52).

Functional exercise capacity: No studies measured functional exercise capacity.

Peak exercise capacity: Two studies reported results which could be pooled for meta-analysis using the Incremental Shuttle Walk Test (ISWT) (Lord 2010; Lord 2012). There is uncertainty due to imprecision about whether singing has an impact on ISWT distance (metres) compared to control (MD -9.26, 95% CI -43.10 to 24.57, n = 52).

Endurance exercise capacity: No studies measured endurance exercise capacity.

Healthcare utilisation: No studies measured healthcare utilisation or hospitalisation.

Physical activity level: One study (Lord 2012) reported measures of physical activity for 24 participants, including steps (steps per day), sedentary time (minutes per day), physical

activity duration (minutes per day), and active energy expenditure (kJ per day). There were no statistically significant differences between the singing group and control group for sedentary time (minutes per day), but the confidence interval is wide (MD -8.60, 95% CI -88.33 to 71.13). There were statistically significant differences in the remaining measures of physical activity favouring the control group (steps (steps per day) MD -1774.00, 95% CI -2847.73 to -700.27; physical activity duration (minutes per day) MD -142.20, 95% CI -262.56 to -21.84; active energy expenditure (kJ per day) MD -373.00, 95% CI -625.28 to -120.72).

Adverse events and side effects: No adverse events or side effects were reported by any of the included studies, and participant withdrawal reasons (where provided) were unrelated to the singing intervention. The study by Bonilha 2009 reported that the vocal exercises and singing was well tolerated by the participants, with no complaints of severe dyspnoea, chest pain, regurgitation or dizziness (Bonilha 2009). In the study by Lord 2010, no participants reported any negative effects from the singing (Lord 2010). Lord 2012 did not report on this outcome (Lord 2012).

DISCUSSION

Summary of main results

We included three studies with a total of 112 participants in this review. The sample size of studies ranged from 33 to 43 participants. All studies included participants diagnosed with chronic obstructive pulmonary disease (COPD) with a mean age ranging from 67 to 72 years and mean forced expiratory volume in one second (FEV₁) ranging from 37% to 64% of predicted values. All studies randomised participants to a singing or a control group. The comparison groups included a film workshop (Lord 2012), handcraft work (Bonilha 2009), and no intervention (Lord 2010). The frequency of the singing intervention in the studies ranged from 1 to 2 times a week over a 6 to 24 week period. The duration of the singing sessions was 60 minutes.

Results for health-related quality of life were diverse. There was no significant change in the St George's Respiratory Questionnaire total score between groups, however a statistically significant improvement in the SF-36 Physical Component Summary (PCS) score favouring the singing group was found (MD 12.64, 95% CI 5.50 to 19.77, 2 studies, n = 52) which surpassed the minimal important difference of 4 units (Hays 2001). No change in dyspnoea was demonstrated.

Measures of pulmonary function and inspiratory muscle strength were only measured in one study and showed no significant differences between the singing group and control group. There was a statistically significant improvement in expiratory muscle pressure favouring the singing group, although this improvement was not clinically significant. No improvement in anxiety, depression, exercise capacity or physical activity level following singing were found. Healthcare utilisation was not measured by any studies and no adverse effects from singing were reported. There are no data to draw conclusions about the long-term effects of singing in people with COPD.

The main results show few statistically or clinically significant health outcomes. There were baseline imbalances between the studies, especially in lung function. This, along with the small number of participants, may have affected the precision around the mean differences. There is a clear need for larger trials with longer duration of follow-up to gain a better understanding of the effects of singing in people with COPD.

Overall completeness and applicability of evidence

Applicability of evidence includes consideration of whether people with COPD would be willing or motivated to participate in singing. Participants in the included studies were recruited from hospital respiratory clinics and no information was provided on previous experience of COPD management, such as pulmonary rehabilitation, or indeed of singing. A recent qualitative study found that people with COPD perceive that pulmonary rehabilitation does not fit their perception of health and that participation may be time-consuming and conflict with daily activities (Mathar 2017). It is not known whether singing might be considered similarly and what the impact of this might be on recruitment to a randomised controlled trial. Nonetheless, qualitative research studies have reported high satisfaction with singing by people with COPD, including self-reported improvements in both breathing and psychological outcomes (Goodridge 2013; Pacheco 2014; McNaughton 2016).

This review included people with stable COPD of moderate to severe disease severity, therefore the results cannot be extrapolated to people with unstable disease such as during or following an exacerbation, or to people with milder COPD. The studies also only reported the short-term effects of singing. Without long-term studies, the effect of singing over a longer period of time cannot be determined. Furthermore, no healthcare utilisation data was reported in any of the selected studies which is particularly important in relation to commissioning and potential for incorporation of findings into healthcare guidelines in the future.

Two studies randomised participants to an active comparison group which matched the time and attention of the singing group. However, the comparison group of one study provided no intervention. With an insufficient number of trials randomising participants to active and non-active comparison groups, we cannot determine whether any improvements in health outcomes were simply a result of participation in a group with support from a leader and fellow participants.

From the data in this review, we cannot determine the optimal delivery mode or dosage of singing required to achieve positive health outcomes in people with COPD. The singing in each study was delivered by a singing teacher, however all studies delivered the singing in groups, so the effect of individual singing lessons in people with COPD cannot be ascertained. The frequency and duration of the singing programs ranged from once to twice a week, and from 6 to 24 weeks in length. It is unclear what frequency and duration is sufficient to provide an effective stimulus for learning the technique of singing and to have an effect on our health outcomes of interest. In all studies participants were instructed to practice their singing at home, however compliance was not measured, therefore it is unknown whether more frequent singing than the supervised group sessions may have contributed to some of the outcomes.

There was general consistency in direction of effects in most outcomes. However, the outcome of anxiety showed an opposite effect. The results from Lord 2010 favoured singing, whilst Lord 2012 results favoured the control group. This inconsistency may be explained by the clinical heterogeneity of the two trials with baseline differences in lung function, anxiety, quality of life and exercise capacity, or it may simply be explained by the impact of chance on such small numbers of participants in each group. Whilst there was one clinically significant result for singing demonstrating an improvement in the SF-36 PCS score, the diversity in results for health-related quality of life may be explained by the methods employed by the studies. A primary outcome and sample size calculation was only reported by one of the included studies (Lord 2012). Therefore, we cannot determine whether two of the three included studies had an adequate sample size calculation or were adequately powered to determine a significant change for the outcome of health-related quality of life. There was a clinically significant change in physical activity favouring the control group in one study (Lord 2012) which is difficult to explain. No other health outcome was clinically significant, although the improvement in anxiety following singing showed a trend towards the minimal important difference (Puhan 2008). The small number of studies and small sample sizes are most likely the major reasons why the changes in outcomes in this review were so variable.

Quality of the evidence

The overall quality of the evidence for the studies included in this review was very low to low. The major methodological shortcoming was the small sample size of the studies. The quality of the evidence was also impacted by the inability to blind the population and personnel due to the physical nature of the intervention. An unknown randomisation process and lack of blinding of the outcome assessor compromised the quality of one study (Bonilha 2009), whilst a reporting bias was present in another study (Lord 2012).

Potential biases in the review process

We adhered to the standard Cochrane methodological procedures to minimise bias, including having two authors independently screen trials, extract trial data and perform the 'Risk of bias' assessment. Attempts were made to contact trial authors where missing information was identified and for a study published in abstract form only, however data were not provided.

Agreements and disagreements with other studies or reviews

This findings of this Cochrane review are consistent with the overall findings of a recent review of singing for lung health in people with COPD (Lewis 2016). Cohort and qualitative data were also examined as part of the narrative literature review which found that participants generally reported positive impacts of singing on their activities of daily living such as housework, their ability to manage their breathlessness, and improved well-being (Lewis 2016). These findings are in agreement with the positive effects shown in this review for one aspect of health-related quality of life, even though the results of the meta-analyses with this small population did not demonstrate clinical significance for all health-related quality of life measures.

AUTHORS' CONCLUSIONS

Implications for practice

There is limited low-quality evidence that singing is safe for people with moderate to severe COPD and improves physical health (as measured by the SF-36 physical component score). Whilst singing may be appealing and subjectively beneficial to some people with COPD, there is currently insufficient evidence to advocate singing as an effective intervention to achieve clinically significant health outcomes above and beyond no intervention or group-based recreational activities.

Implications for research

More randomised controlled trials are required to determine the effect of singing on health-related quality of life and dyspnoea in people with COPD; trials examining the effect of singing in addition to pulmonary rehabilitation are also needed. In particular, large studies with long-term follow-up are necessary. Future studies need to incorporate important methodological features such as adequate sample sizes, randomisation, allocation concealment and blinding of outcome assessors, as well as longer follow-up, to ensure high-quality evidence is available on the effectiveness of singing in people with COPD.

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SOURCES OF SUPPORT

Internal sources

- No sources of support supplied

External sources

- Cochrane Complementary Medicine Field bursary, USA.

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APPENDICES

Appendix 1. Sources and search methods for the Cochrane Airways Group Specialised Register (CAGR)

Electronic searches: core databases

Database	Frequency of search
CENTRAL (<i>the Cochrane Library</i>)	Monthly
MEDLINE (Ovid)	Weekly
Embase (Ovid)	Weekly
PsycINFO (Ovid)	Monthly
CINAHL (EBSCO)	Monthly
AMED (EBSCO)	Monthly

Handsearches: core respiratory conference abstracts

Conference	Years searched
American Academy of Allergy, Asthma and Immunology (AAAAI)	2001 onwards
American Thoracic Society (ATS)	2001 onwards
Asia Pacific Society of Respiratory (APSR)	2004 onwards
British Thoracic Society Winter Meeting (BTS)	2000 onwards
Chest Meeting	2003 onwards
European Respiratory Society (ERS)	1992, 1994, 2000 onwards
International Primary Care Respiratory Group Congress (IPCRG)	2002 onwards
Thoracic Society of Australia and New Zealand (TSANZ)	1999 onwards

MEDLINE search strategy used to identify trials for the CAGR

COPD search

1. Lung Diseases, Obstructive/
2. exp Pulmonary Disease, Chronic Obstructive/
3. emphysema\$.mp.
4. (chronic\$ adj3 bronchiti\$).mp.
5. (obstruct\$ adj3 (pulmonary or lung\$ or airway\$ or airflow\$ or bronch\$ or respirat\$)).mp.
6. COPD.mp.
7. COAD.mp.
8. COBD.mp.

9. AECB.mp.

10. or/1–9

Filter to identify RCTs

1. exp “clinical trial [publication type]”/

2. (randomized or randomised).ab,ti.

3. placebo.ab,ti.

4. dt.fs.

5. randomly.ab,ti.

6. trial.ab,ti.

7. groups.ab,ti.

8. or/1–7

9. Animals/

10. Humans/

11. 9 not (9 and 10)

12. 8 not 11

We will adapt the MEDLINE strategy and RCT filter to identify trials in other electronic databases.

Appendix 2. Search strategy to identify relevant trials from the CAGR

#1 MeSH DESCRIPTOR Pulmonary Disease, Chronic Obstructive Explode All

#2 MeSH DESCRIPTOR Bronchitis, Chronic

#3 (obstruct*) near3 (pulmonary or lung* or airway* or airflow* or bronch* or respirat*)

#4 COPD:MISC1

#5 (COPD OR COAD OR COBD OR AECOPD):TI,AB,KW

#6 #1 OR #2 OR #3 OR #4 OR #5

#7 (sing or singing or singer* or song*):ti,ab,kw

#8 (voice* or vocal*) NEAR (exercis* or train*)

#9 diaphragm* NEAR2 breath*

#10 MeSH DESCRIPTOR Music Therapy

#11 choir*:ti,ab,kw

#12 #7 or #8 or #9 or #10 or #11

#13 #6 AND #12

[in search line #4, MISC1 denotes the field in the record where the reference has been coded for condition, in this case, COPD]

CHARACTERISTICS OF STUDIES

Characteristics of included studies [ordered by study ID]

Bonilha 2009	
Methods	Study design: randomised controlled trial
Participants	<p>Participants: n = 43 Included: COPD (according to GOLD criteria); stable for 2 months; ex-smoker Excluded: severe comorbidities; oxygen therapy; smoker Baseline characteristics: Intervention group - singing (n = 15)</p> <ol style="list-style-type: none"> <i>Gender, male:</i> n = 12 <i>Age, years:</i> mean (SD) 70 (7) <i>FEV₁, L:</i> 1.11 (0.47) <i>FEV₁, % predicted:</i> 49 (21) <i>FEV₁/FVC, %:</i> 46 (18) <p>Control group - handcraft (n = 15)</p> <ol style="list-style-type: none"> <i>Gender, male:</i> n = 12 <i>Age, years:</i> 74 (8) <i>FEV₁, L:</i> 1.18 (0.47) <i>FEV₁, % predicted:</i> 53 (20) <i>FEV₁/FVC, %:</i> 43 (11)
Interventions	<p>Intervention characteristics: Intervention group - singing</p> <ol style="list-style-type: none"> <i>Duration (session):</i> 1 hour <i>Frequency:</i> 1/week <i>Length (programme):</i> 24 weeks <i>Professional/s:</i> physiotherapist + singing teacher <i>Location:</i> not reported <i>Session details:</i> (1) relaxation exercises of neck and upper limb muscles, conducted by a physiotherapist (5 minutes); (2) singing-related respiratory exercises conducted by a singing teacher (10 minutes) - these exercises are part of regular singing teaching, and consisted of: performing fast, deep inspirations, followed by slow, full or interrupted expirations; performing fast and deep respiratory incursions, paying attention to the upper abdominal movements; generating breathing movements against, or with the help, of pressures generated by a hand placed on the upper abdominal region; (3) vocalisation exercises, lead by the singing teacher, as a preparation for singing (15 minutes) - participants loudly pronounced vowels such as "le", "la", "mi", "mu", and also sang the melody of a familiar song using such vowels instead of actually singing the lyrics; (iv) singing training of Brazilian folk songs, conducted by the singer teacher (30 minutes) <i>Additional information:</i> participants were also instructed to practice the folk songs at home for half an hour on at least two more days during the week <p>Control group - handcraft</p> <ol style="list-style-type: none"> <i>Duration (session):</i> 55 minutes <i>Frequency:</i> 1/week <i>Length (programme):</i> 24 weeks

Bonilha 2009

	<p>4 Professional/s: physiotherapist + handcraft work teacher</p> <p>5 Session details: (I) relaxation exercises of neck and upper limb muscles, conducted by a physiotherapist (5 minutes); (ii) execution of handcraft artwork such as paper folding, drawing, and collages (50 minutes)</p> <p>6 Additional information: participants were also routinely instructed to include some incomplete artwork or beginning a new one at home</p>
Outcomes	<p>Health-related quality of life - <i>St George's Respiratory Questionnaire</i>, total score</p> <p>1 Outcome type: continuous outcome</p> <p>2 Range: 0–100</p> <p>3 Unit of measure: percent, %</p> <p>4 Direction: lower is better</p> <p>5 Data value: change from baseline</p> <p>Dyspnoea - <i>Basal Dyspnoea Index (BDI)</i>, score</p> <p>1 Outcome type: continuous outcome</p> <p>2 Range: 0–12</p> <p>3 Unit of measure: score</p> <p>4 Direction: higher is better</p> <p>5 Data value: change from baseline</p> <p>Respiratory muscle strength - <i>PI_{max}</i>, cmH₂O</p> <p>1 Outcome type: continuous outcome</p> <p>2 Unit of measure: cmH₂O</p> <p>3 Direction: higher is better</p> <p>4 Data value: change from baseline</p> <p>Respiratory muscle strength - <i>PE_{max}</i>, cmH₂O</p> <p>1 Outcome type: continuous outcome</p> <p>2 Unit of measure: cmH₂O</p> <p>3 Direction: higher is better</p> <p>4 Data value: change from baseline</p> <p>Lung function - <i>FVC</i>, L</p> <p>1 Outcome type: continuous outcome</p> <p>2 Unit of measure: litre, L</p> <p>3 Direction: higher is better</p> <p>4 Data value: change from baseline</p> <p>Lung function - <i>FEV₁</i>, L</p> <p>1 Outcome type: continuous outcome</p> <p>2 Unit of measure: litre, L</p> <p>3 Direction: higher is better</p> <p>4 Data value: change from baseline</p> <p>Lung function - <i>FEV₁/FVC</i>, %</p> <p>1 Outcome type: continuous outcome</p> <p>2 Unit of measure: percent, %</p> <p>3 Direction: higher is better</p> <p>4 Data value: change from baseline</p>

Bonilha 2009

	<i>Lung function - ERV, L</i> 1 Outcome type: continuous outcome 2 Unit of measure: L 3 Direction: higher is better 4 Data value: change from baseline <i>Lung function - IC, L</i> 1 Outcome type: continuous outcome 2 Unit of measure: litre, L 3 Direction: higher is better 4 Data value: change from baseline	
Identification	Country: Brazil Setting: hospital Authors name: Amanda Gimenes Bonilha Institution: University of Sao Paulo Email: jabmarti@fmrp.usp.br Address: Internal Medicine Department, Avenida Bandeirantes 3900, CEP: 14048-800, Ribeirao Preto, Sao Paulo, Brazil	
Notes	Authors were contacted for further information, with no response. Sponsorship source: Not stated	
Risk of bias		
Bias	Authors' judgement	Support for judgement
Random sequence generation (selection bias)	Unclear risk	Not specified
Allocation concealment (selection bias)	Unclear risk	Not specified
Blinding of participants and personnel (performance bias) All outcomes	High risk	Not specified, but due to the physical nature of the intervention it is unlikely the participants were able to be blinded
Blinding of outcome assessment (detection bias) All outcomes	Unclear risk	Not specified
Incomplete outcome data (attrition bias) All outcomes	High risk	Outcomes measured reported for all participants completing post intervention assessment High dropout rate from singing group (35%)
Selective reporting (reporting bias)	Low risk	All outcome measures listed in methods were reported
Other bias	Low risk	Study appears to be free of other sources of bias
Lord 2010		
Methods	Study design: randomised controlled trial	
Participants	Participants: n = 36 Included: COPD (diagnosed according to the GOLD guidelines and attending respiratory clinics) Baseline characteristics Intervention group - singing (n = 15)	

Bonilha 2009

	<ol style="list-style-type: none"> 1 <i>Gender</i>: not reported 2 <i>Age, years</i>: mean (SD) 67 (9) 3 <i>FEV₁, % predicted</i>: 37 (15) 4 <i>Long-term oxygen therapy, n (%)</i>: 4 (27) <p>Control group - no intervention (n = 13)</p> <ol style="list-style-type: none"> 1 <i>Gender</i>: not reported 2 <i>Age, years</i>: 68 (7) 3 <i>FEV₁, % predicted</i>: 38 (22) 4 <i>Long-term oxygen therapy, n (%)</i>: 4 (31)
Interventions	<p>Intervention characteristics: Intervention group - singing</p> <ol style="list-style-type: none"> 1 <i>Duration (session)</i>: 1 hour 2 <i>Frequency</i>: 2/week 3 <i>Length (program)</i>: 6 weeks 4 <i>Professional/s</i>: singing teacher 5 <i>Location</i>: hospital 6 <i>Session details</i>: 20 minutes - postural work and physical stretches; 10 minutes - breath observation and management/relaxation; 10 minutes - vocal exercises; 10–20 minutes - singing songs 7 <i>Additional information</i>: each participant was given homework and an accompanying CD of songs to practice at home Control group - no intervention
Outcomes	<p><i>Health-related quality of life - SF-36 Physical Component Summary (PCS), score</i></p> <ol style="list-style-type: none"> 1 Outcome type: continuous outcome 2 Range: 0–100 3 Unit of measure: score 4 Direction: higher is better 5 Data value: change from baseline <p><i>Quality of life - SF-36 Mental Component Summary (MCS), score</i></p> <ol style="list-style-type: none"> 1 Outcome type: continuous outcome 2 Range: 0–100 3 Unit of measure: score 4 Direction: higher is better 5 Data value: change from baseline <p><i>Psychological status - Hospital Anxiety and Depression Scale (HADS) - anxiety subscale, score</i></p> <ol style="list-style-type: none"> 1 Outcome type: continuous outcome 2 Range: 0–21 3 Unit of measure: score 4 Direction: lower is better 5 Data value: change from baseline <p><i>Psychological status - Hospital Anxiety and Depression Scale (HADS) - depression subscale, score</i></p> <ol style="list-style-type: none"> 1 Outcome type: continuous outcome 2 Range: 0–21 3 Unit of measure: score

Bonilha 2009

	<p>4 Direction: lower is better</p> <p>5 Data value: change from baseline</p> <p><i>Peak exercise capacity - Incremental Shuttle Walk Test (ISWT), m</i></p> <p>1 Outcome type: continuous outcome</p> <p>2 Unit of measure: metres, m</p> <p>3 Direction: higher is better</p> <p>4 Data value: change from baseline</p>
Identification	<p>Country: United Kingdom</p> <p>Setting: hospital</p> <p>Authors name: Victoria M Lord</p> <p>Institution: Royal Brompton & Harefield NHS Foundation Trust</p> <p>Email: n.hopkinson@imperial.ac.uk</p> <p>Address: Royal Brompton Hospital, Sydney Street, London SW3 6NP, UK</p>
Notes	<p>Prior to randomisation, all study participants received a 30-minute standard session on breathing control and techniques to manage breathlessness, delivered by a respiratory physiotherapist. Pursed lip breathing and nose breathing were also discussed in relation to managing episodes of shortness of breath. Each participant received a standard Royal Brompton Hospital "Help Yourself - physiotherapy for people with respiratory symptoms" and was advised to practice the techniques at home</p> <p>Sponsorship source: Royal Brompton and Harefield Arts (rb&hArts)</p>

Risk of bias

Bias	Authors' judgement	Support for judgement
Random sequence generation (selection bias)	Low risk	Block randomisation
Allocation concealment (selection bias)	Low risk	Consecutive sealed envelopes
Blinding of participants and personnel (performance bias) All outcomes	High risk	Not specified, but due to the physical nature of the intervention it is unlikely the participants were able to be blinded
Blinding of outcome assessment (detection bias) All outcomes	Low risk	Assessors were blinded to treatment allocation
Incomplete outcome data (attrition bias) All outcomes	High risk	Outcomes measured reported for all participants completing post intervention assessment High dropout rate from singing group (25%)
Selective reporting (reporting bias)	Low risk	All outcome measures listed in methods were reported
Other bias	Low risk	Study appears to be free of other sources of bias

Lord 2012

Methods	Study design: randomised controlled trial
Participants	<p>Participants: n = 33</p> <p>Included: COPD (diagnosed according to the GOLD guidelines and attending respiratory clinics)</p> <p>Baseline characteristics:</p> <p>Intervention group - singing (n = 18)</p> <p>1 Gender: not reported</p>

Bonilha 2009

	<p>2 <i>Age, years:</i> mean (SD) 69 (11)</p> <p>3 <i>FEV₁, % predicted:</i> 44 (14)</p> <p>Control group - film workshop (n = 15)</p> <p>1 <i>Gender:</i> not reported</p> <p>2 <i>Age, years:</i> 68 (9)</p> <p>3 <i>FEV₁, % predicted:</i> 64 (26)</p>
Interventions	<p>Intervention characteristics:</p> <p>Intervention group - singing</p> <p>1 <i>Duration (session):</i> 1 hour</p> <p>2 <i>Frequency:</i> 2/week</p> <p>3 <i>Length (program):</i> 8 weeks</p> <p>4 <i>Professional/s:</i> singing teacher</p> <p>5 <i>Location:</i> not reported</p> <p>6 <i>Session details:</i> 20 minutes - postural work and physical stretches; 10 minutes - breath observation and management/relaxation; 10 minutes - vocal exercises; 10–20 minutes - singing songs</p> <p>7 <i>Additional information:</i> participants were given a CD of physical warm-ups, breathing exercises and songs to practice at home daily</p> <p>Control group - film workshop</p> <p>1 <i>Duration (session):</i> duration of film (variable) + 1 hour</p> <p>2 <i>Frequency:</i> 1/week</p> <p>3 <i>Length (program):</i> 8 weeks</p> <p>4 <i>Professional/s:</i> film studies graduate</p> <p>5 <i>Session details:</i> watched a film in a group and discussed salient points in the workshop afterwards</p>
Outcomes	<p><i>Health-related quality of life - SF-36 Physical Component Summary (PCS), score</i></p> <p>1 Outcome type: continuous outcome</p> <p>2 Range: 0–100</p> <p>3 Unit of measure: score</p> <p>4 Direction: higher is better</p> <p>5 Data value: change from baseline</p> <p><i>Health-related quality of life - SF-36 Mental Component Summary (MCS), score</i></p> <p>1 Outcome type: continuous outcome</p> <p>2 Range: 0–100</p> <p>3 Unit of measure: score</p> <p>4 Direction: higher is better</p> <p>5 Data value: change from baseline</p> <p><i>Health-related quality of life - COPD Assessment Test (CAT), score</i></p> <p>1 Outcome type: continuous outcome</p> <p>2 Range: 0–40</p> <p>3 Unit of measure: score</p> <p>4 Direction: lower is better</p> <p>5 Data value: change from baseline</p>

Bonilha 2009

Author Manuscript	<i>Psychological status - Hospital Anxiety and Depression Scale (HADS) - anxiety subscale, score</i>	
	1	Outcome type: continuous outcome
	2	Range: 0–21
	3	Unit of measure: score
	4	Direction: lower is better
Author Manuscript	5	Data value: change from baseline
	<i>Psychological status - Hospital Anxiety and Depression Scale (HADS) - depression subscale, score</i>	
	1	Outcome type: continuous outcome
	2	Range: 0–21
	3	Unit of measure: score
Author Manuscript	4	Direction: lower is better
	5	Data value: change from baseline
	<i>Peak exercise capacity - Incremental Shuttle Walk Test (ISWT), m</i>	
	1	Outcome type: continuous outcome
	2	Unit of measure: metres, m
Author Manuscript	3	Direction: higher is better
	4	Data value: change from baseline
	<i>Physical activity - steps per day, n</i>	
	1	Outcome type: continuous outcome
	2	Unit of measure: number, n
Author Manuscript	3	Direction: higher is better
	4	Data value: change from baseline
	<i>Physical activity - sedentary time per day, min</i>	
	1	Outcome type: continuous outcome
	2	Unit of measure: min
Author Manuscript	3	Direction: lower is better
	4	Data value: change from baseline
	<i>Physical activity - physical activity duration per day, min</i>	
	1	Outcome type: continuous outcome
	2	Unit of measure: minute, min
Author Manuscript	3	Direction: higher is better
	4	Data value: change from baseline
	<i>Physical activity - active energy expenditure per day, kJ</i>	
	1	Outcome type: continuous outcome
	2	Unit of measure: kilojoule, kJ
Author Manuscript	3	Direction: higher is better
	4	Data value: change from baseline
	Identification	
	Country: United Kingdom	
	Setting: hospital	
	Authors name: Victoria M Lord	
	Institution: Royal Brompton & Harefield NHS Foundation Trust	
	Email: n.hopkinson@ic.ac.uk	
	Address: Royal Brompton Hospital, Sydney Street, London SW3 6NP, UK	

Bonilha 2009

Notes	Prior to randomisation, all study participants received a 30-minute standard session on breathing control and techniques to manage breathlessness, delivered by a respiratory physiotherapist. Pursed-lip breathing and nose breathing were also discussed in relation to managing episodes of shortness of breath. Each participant received a standard Royal Brompton Hospital "Help Yourself - physiotherapy for people with respiratory symptoms" and was advised to practice the techniques at home Sponsorship source: Royal Brompton and Harefield Arts (rb&hArts)
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Risk of bias

Bias	Authors' judgement	Support for judgement
Random sequence generation (selection bias)	Low risk	The sequence was developed by an author who was not involved with the day to day conduct of the trial
Allocation concealment (selection bias)	Low risk	Consecutive sequentially numbered sealed envelopes
Blinding of participants and personnel (performance bias) All outcomes	High risk	Not specified, but due to the physical nature of the intervention it is unlikely the participants were able to be blinded
Blinding of outcome assessment (detection bias) All outcomes	Low risk	Assessors were blinded to treatment allocation
Incomplete outcome data (attrition bias) All outcomes	High risk	Outcomes measured reported for all participants completing post intervention assessment High dropout rate from singing group (28%)
Selective reporting (reporting bias)	High risk	Spirometry and exacerbation rate were not reported post intervention
Other bias	Low risk	Study appears to be free of other sources of bias

cmH₂O: centimetre of water

COPD: chronic obstructive pulmonary disease

ERV: end respiratory volume

FEV₁: forced expiratory volume in one second

FVC: forced vital capacity

GOLD: Global Initiative for Chronic Obstructive Lung Disease

IC: inspiratory capacity

L: litre

min: minute

PI_{max}: maximal inspiratory pressure

PE_{max}: maximal expiratory pressure

Characteristics of excluded studies [ordered by study ID]

Study	Reason for exclusion
Canga 2015	Intervention did not meet study inclusion criteria of singing forming the majority of the intervention

Characteristics of studies awaiting assessment [ordered by study ID]

Miyahara 2001	
Methods	Study design: randomised controlled trial
Participants	Participants: n = 20 Included: COPD (moderate to severe) Baseline characteristics: <ol style="list-style-type: none"> 1 <i>Gender:</i> not reported 2 <i>Age, years:</i> not reported 3 <i>FEV₁, % predicted:</i> mean (SD) 40 (15)
Interventions	Intervention characteristics: Intervention group - Japanese traditional "Shigin" singing programme <ol style="list-style-type: none"> 1 <i>Duration (session):</i> not reported 2 <i>Frequency:</i> 5/week 3 <i>Length (program):</i> 8 weeks 4 <i>Professional/s:</i> not reported 5 <i>Session details:</i> the Japanese traditional "Shigin" singing program requires slow and deep breaths during singing 6 <i>Intensity:</i> dyspnoea score of 3–5 (on BORG scale 0–10) Control group - no training
Outcomes	<i>Pulmonary function</i> <i>Respiratory muscle strength</i> <i>Peak exercise capacity</i> <i>Health-related quality of life - Chronic Respiratory Disease Questionnaire, score</i>
Notes	Abstract only Authors were contacted for further information, with no response

COPD: chronic obstructive pulmonary disease

FEV₁: forced expiratory volume in one second

Characteristics of ongoing studies [ordered by study ID]

Kaasgaard 2017	
Trial name or title	The effects of singing training for patients with chronic obstructive pulmonary disease
Methods	Cluster-randomised controlled trial
Participants	People with COPD
Interventions	Singing group versus pulmonary rehabilitation group; 90 minute sessions twice weekly for 10 weeks
Outcomes	Not known
Starting date	Not known
Contact information	Mette Kaasgaard (mk@clin.au.dk)
Notes	Researcher was contacted for further information, with no response

COPD: chronic obstructive pulmonary disease

DATA AND ANALYSES

Comparison 1. Intervention - Singing vs Control

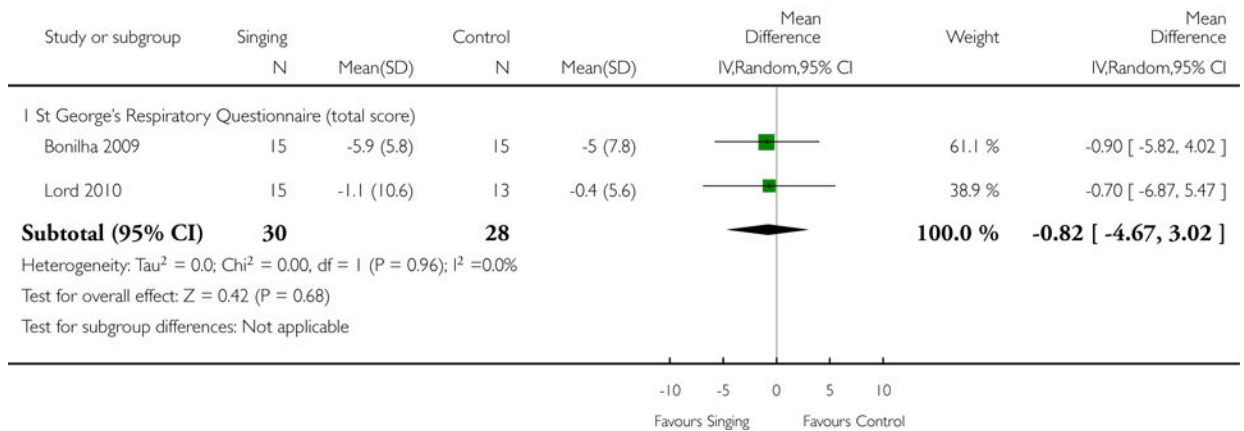
Outcome or subgroup title	No. of studies	No. of participants	Statistical method	Effect size
1 Health-related quality of life - respiratory specific (mean change)	2		Mean Difference (IV, Random, 95% CI)	Subtotals only
1.1 St George's Respiratory Questionnaire (total score)	2	58	Mean Difference (IV, Random, 95% CI)	-0.82 [-4.67, 3.02]
2 Health-related quality of life - generic (mean change)	2		Mean Difference (IV, Random, 95% CI)	Subtotals only
2.1 SF-36 PCS score	2	52	Mean Difference (IV, Random, 95% CI)	12.64 [5.50, 19.77]
2.2 SF-36 MCS score	2	52	Mean Difference (IV, Random, 95% CI)	5.42 [-3.90, 14.74]
3 Dyspnoea (mean change)	1		Mean Difference (IV, Random, 95% CI)	Totals not selected
3.1 BDI (score)	1		Mean Difference (IV, Random, 95% CI)	0.0 [0.0, 0.0]
4 Respiratory muscle strength (mean change)	1		Mean Difference (IV, Random, 95% CI)	Totals not selected
4.1 PImax (cmH ₂ O)	1		Mean Difference (IV, Random, 95% CI)	0.0 [0.0, 0.0]
4.2 PEmax (cmH ₂ O)	1		Mean Difference (IV, Random, 95% CI)	0.0 [0.0, 0.0]
5 Pulmonary function (mean change)	1		Mean Difference (IV, Random, 95% CI)	Totals not selected
5.1 FEV ₁ (L)	1		Mean Difference (IV, Random, 95% CI)	0.0 [0.0, 0.0]
5.2 FVC (L)	1		Mean Difference (IV, Random, 95% CI)	0.0 [0.0, 0.0]
5.3 FEV ₁ /FVC (%)	1		Mean Difference (IV, Random, 95% CI)	0.0 [0.0, 0.0]
5.4 ERV (L)	1		Mean Difference (IV, Random, 95% CI)	0.0 [0.0, 0.0]
5.5 IC (L)	1		Mean Difference (IV, Random, 95% CI)	0.0 [0.0, 0.0]
6 Psychological status (mean change)	2		Mean Difference (IV, Random, 95% CI)	Subtotals only
6.1 HADS - anxiety score	2	52	Mean Difference (IV, Random, 95% CI)	-1.09 [-3.02, 0.83]
6.2 HADS - depression score	2	52	Mean Difference (IV, Random, 95% CI)	-0.87 [-2.16, 0.42]
7 Peak exercise capacity (mean change)	2		Mean Difference (IV, Random, 95% CI)	Subtotals only
7.1 ISWT - metres	2	52	Mean Difference (IV, Random, 95% CI)	-9.26 [-43.10, 24.57]
8 Physical activity level (mean change)	1		Mean Difference (IV, Random, 95% CI)	Totals not selected
8.1 Sedentary time (minutes per day)	1		Mean Difference (IV, Random, 95% CI)	0.0 [0.0, 0.0]

Outcome or subgroup title	No. of studies	No. of participants	Statistical method	Effect size
8.2 Steps (steps per day)	1		Mean Difference (IV, Random, 95% CI)	0.0 [0.0, 0.0]
8.3 Physical activity duration (minutes per day)	1		Mean Difference (IV, Random, 95% CI)	0.0 [0.0, 0.0]
8.4 Active energy expenditure (kJ per day)	1		Mean Difference (IV, Random, 95% CI)	0.0 [0.0, 0.0]

Review: Singing for adults with chronic obstructive pulmonary disease (COPD)

Comparison: 1 Intervention - Singing vs Control

Outcome: 1 Health-related quality of life - respiratory specific (mean change)



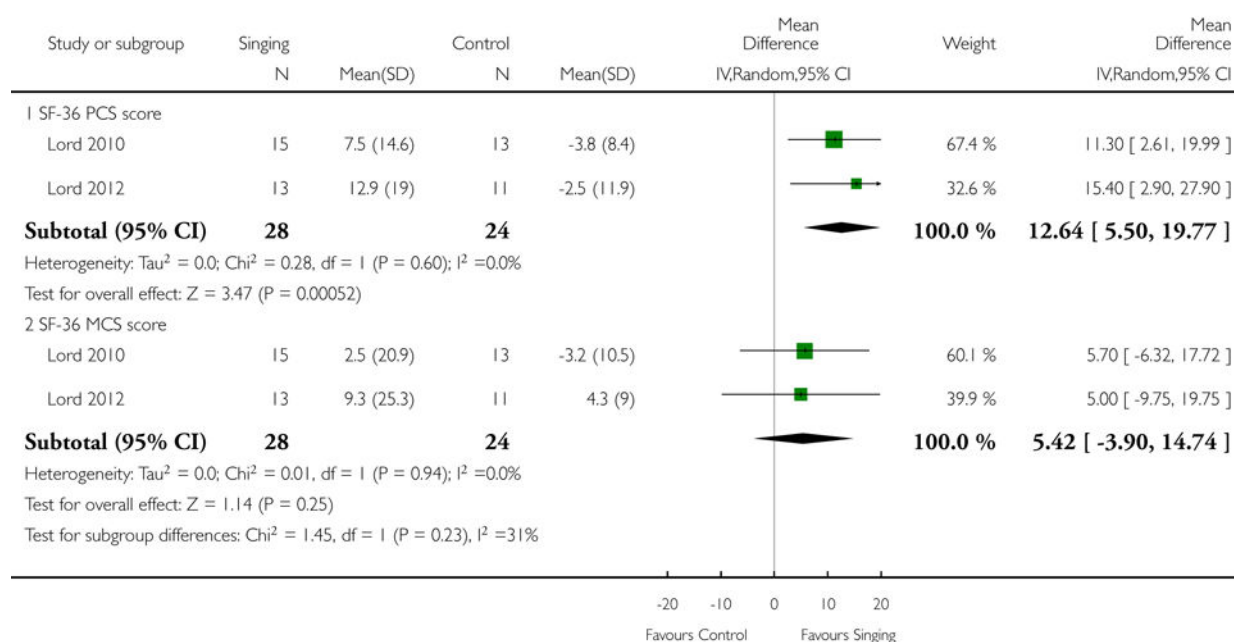
Analysis 1.1.

Comparison 1 Intervention - Singing vs Control, Outcome 1 Health-related quality of life - respiratory specific (mean change).

Review: Singing for adults with chronic obstructive pulmonary disease (COPD)

Comparison: 1 Intervention - Singing vs Control

Outcome: 2 Health-related quality of life - generic (mean change)



Analysis 1.2.

Comparison 1 Intervention - Singing vs Control, Outcome 2 Health-related quality of life - generic (mean change).

Review: Singing for adults with chronic obstructive pulmonary disease (COPD)

Comparison: 1 Intervention - Singing vs Control

Outcome: 3 Dyspnoea (mean change)



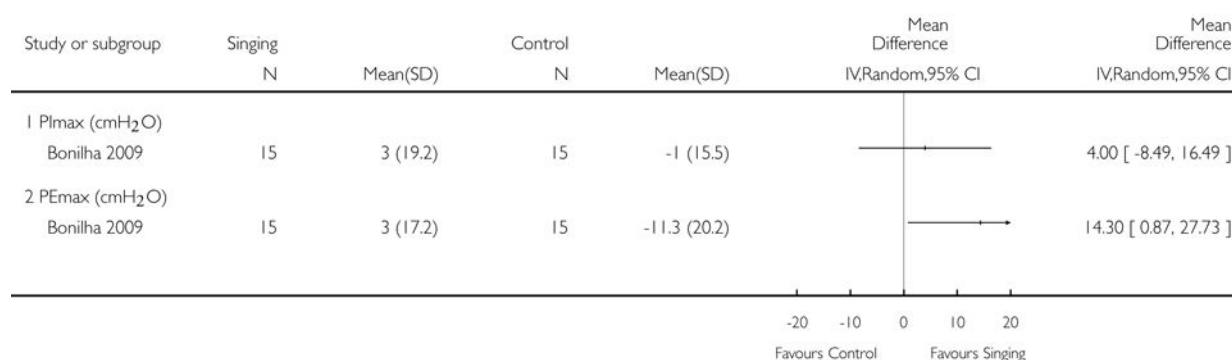
Analysis 1.3.

Comparison 1 Intervention - Singing vs Control, Outcome 3 Dyspnoea (mean change).

Review: Singing for adults with chronic obstructive pulmonary disease (COPD)

Comparison: 1 Intervention - Singing vs Control

Outcome: 4 Respiratory muscle strength (mean change)



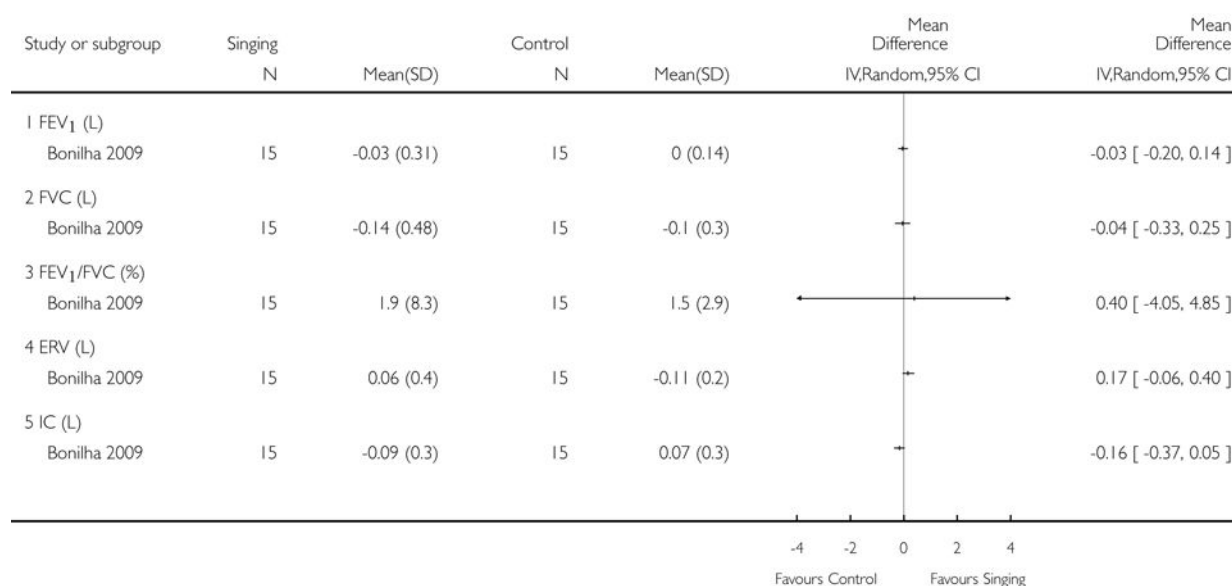
Analysis 1.4.

Comparison 1 Intervention - Singing vs Control, Outcome 4 Respiratory muscle strength (mean change).

Review: Singing for adults with chronic obstructive pulmonary disease (COPD)

Comparison: 1 Intervention - Singing vs Control

Outcome: 5 Pulmonary function (mean change)



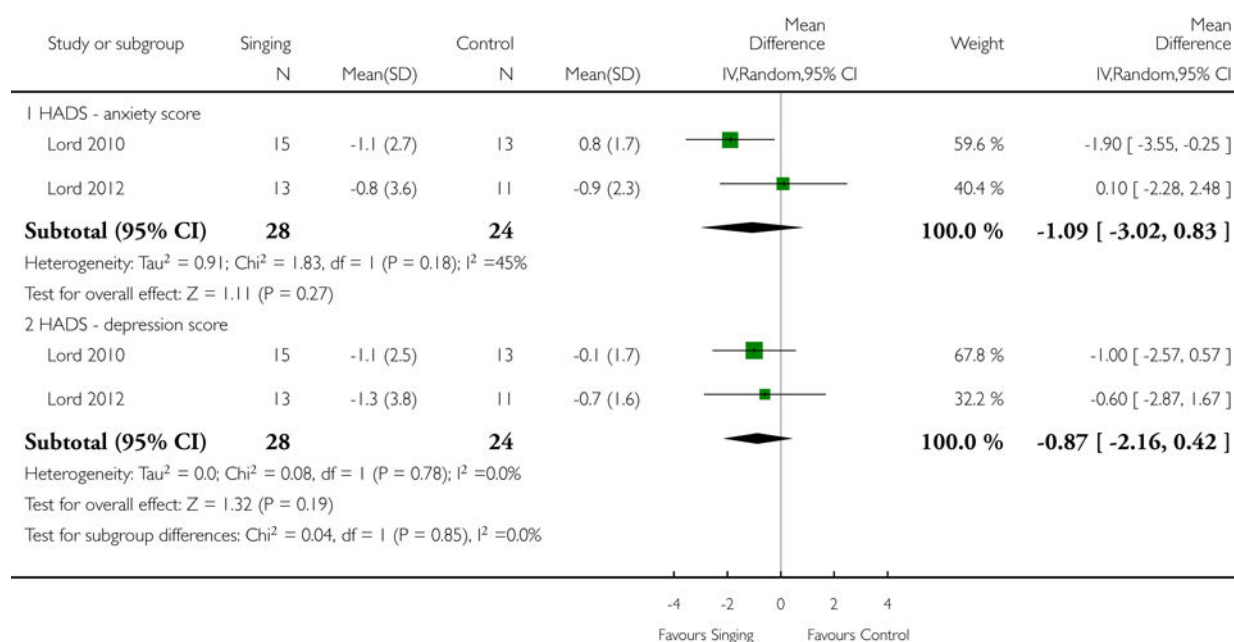
Analysis 1.5.

Comparison 1 Intervention - Singing vs Control, Outcome 5 Pulmonary function (mean change).

Review: Singing for adults with chronic obstructive pulmonary disease (COPD)

Comparison: 1 Intervention - Singing vs Control

Outcome: 6 Psychological status (mean change)



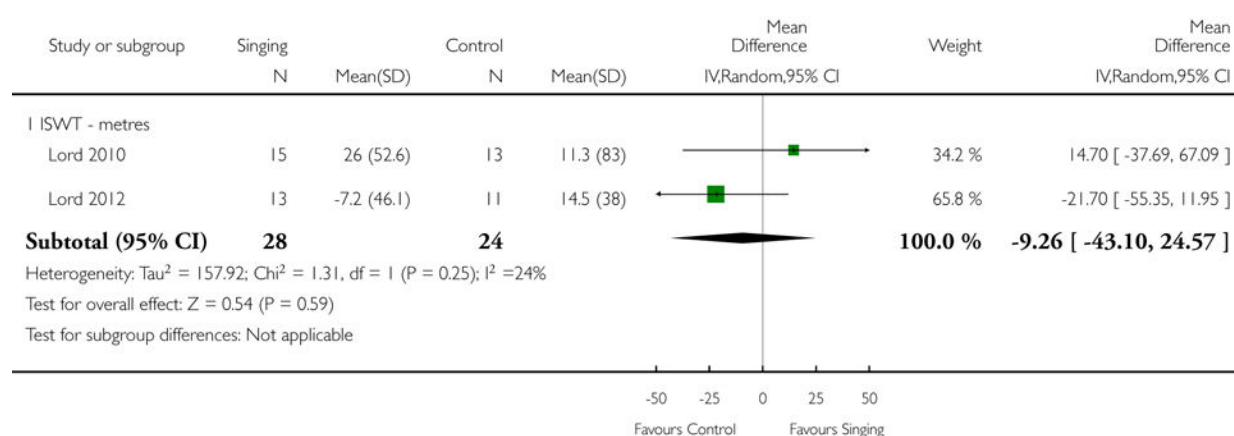
Analysis 1.6.

Comparison 1 Intervention - Singing vs Control, Outcome 6 Psychological status (mean change).

Review: Singing for adults with chronic obstructive pulmonary disease (COPD)

Comparison: 1 Intervention - Singing vs Control

Outcome: 7 Peak exercise capacity (mean change)



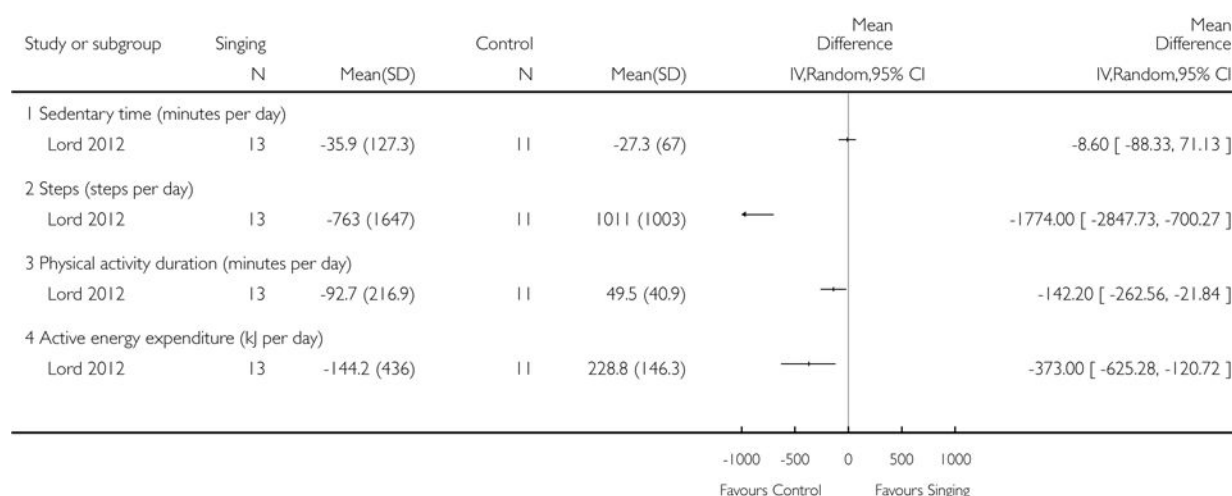
Analysis 1.7.

Comparison 1 Intervention - Singing vs Control, Outcome 7 Peak exercise capacity (mean change).

Review: Singing for adults with chronic obstructive pulmonary disease (COPD)

Comparison: 1 Intervention - Singing vs Control

Outcome: 8 Physical activity level (mean change)



Analysis 1.8.

Comparison 1 Intervention - Singing vs Control, Outcome 8 Physical activity level (mean change).

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* Indicates the major publication for the study

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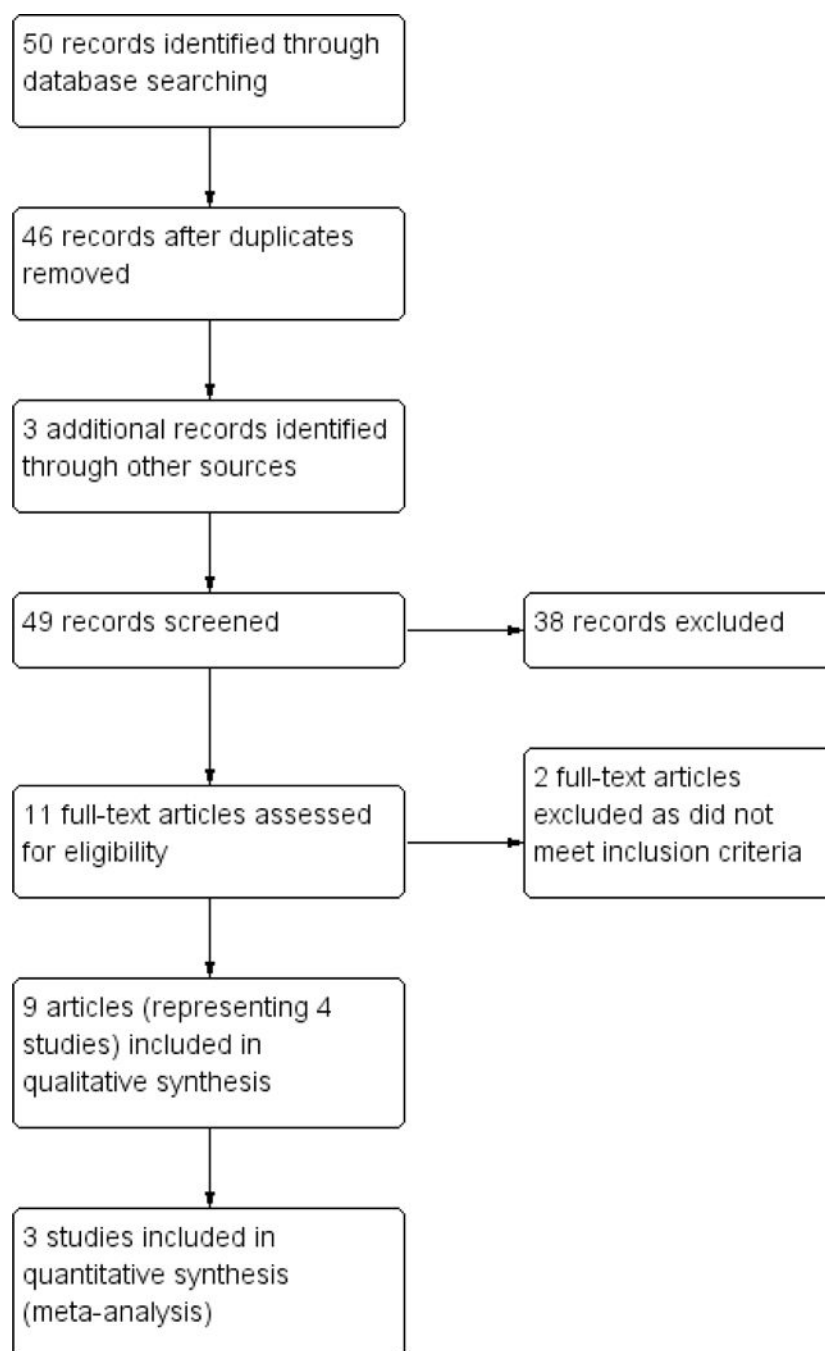


Figure 1.
Study flow diagram.

	Random sequence generation (selection bias)	Allocation concealment (selection bias)	Blinding of participants and personnel (performance bias)	Blinding of outcome assessment (detection bias)	Incomplete outcome data (attrition bias)	Selective reporting (reporting bias)	Other bias
Bonilha 2009	?	?	-	?	-	+	+
Lord 2010	+	+	-	+	-	+	+
Lord 2012	+	+	-	+	-	-	+

Figure 2.
Risk of bias summary: review authors' judgements about each risk of bias item for each included study.

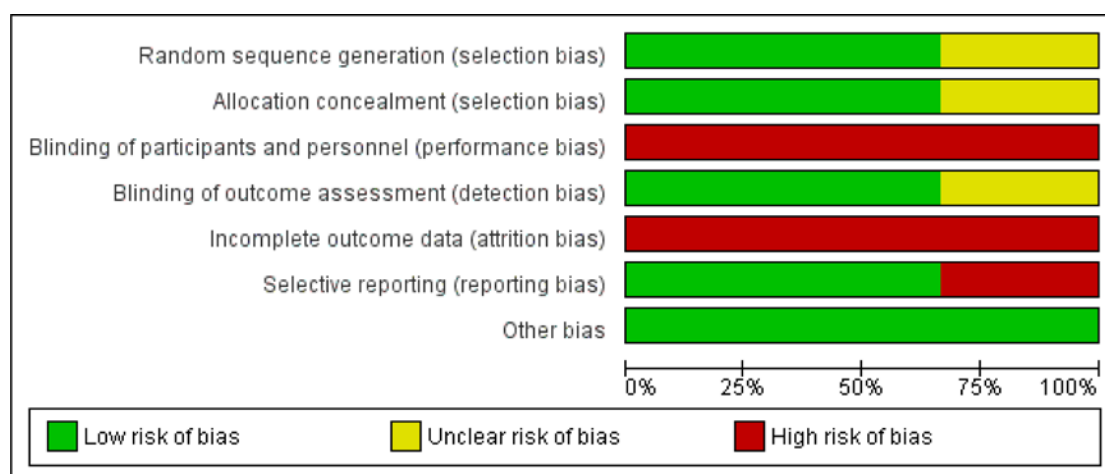


Figure 3.
Risk of bias graph: review authors' judgements about each risk of bias item presented as percentages across all included studies.

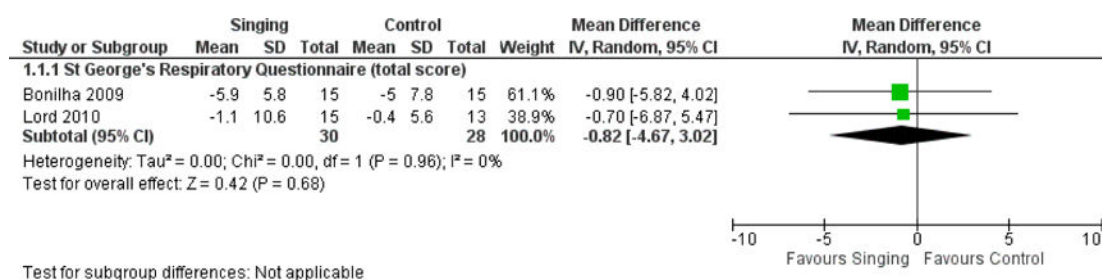
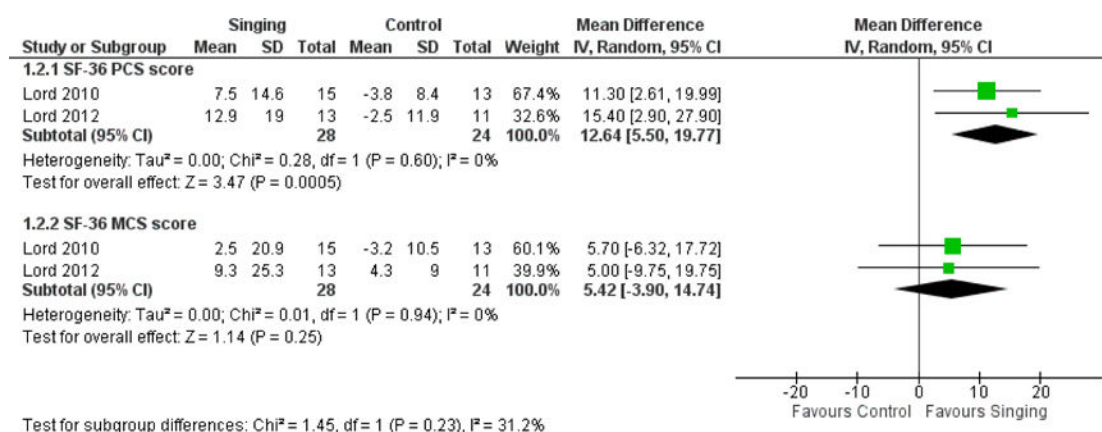


Figure 4.

Forest plot of comparison: 1 Intervention - Singing vs Control, outcome: 1.1 Health-related quality of life - respiratory specific (mean change).

**Figure 5.**

Forest plot of comparison: 1 Intervention - Singing vs Control, outcome: 1.2 Health-related quality of life - generic (mean change).

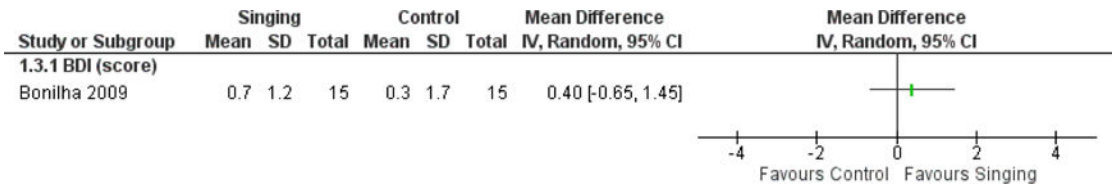


Figure 6.
Forest plot of comparison: 1 Intervention - Singing vs Control, outcome: 1.3 Dyspnoea (mean change).