Singing for Adults with Respiratory Illness:
A Systematic Review and Evaluation of a Community Programme

by

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Abstract

Background: Respiratory disorders manifest themselves with chest symptoms including shortness of breath and decline in lung function. An innovative cost-effective therapy is needed and there is a growing body of evidence to suggest that singing in a group may have health and wellbeing benefits for these patients.

Method: The key objectives of this research were to review current knowledge and to evaluate the potential impact of group singing as an intervention for health and wellbeing. The research is divided into three components which aimed to:

- Review, synthesise and consolidate current research in the area of singing for COPD.
- Evaluate the impact of group singing on health and wellbeing for a group of COPD patients
- Evaluate the usability of specially designed resources for home practice for respiratory patients.

Results: A broad systematic review showed that availability of high quality evidence that singing for COPD improves physical health, dyspnoea or respiratory-specific quality of life is still very limited. This is due to the low number of high quality studies and small sample sizes. The study reported here found statistically significant positive changes in both total mean Warwick-Edinburgh Mental Wellbeing Scale (WEMWBS) and COPD Assessment Test (CAT) scores for twenty-one COPD participants of two community singing groups over a twelve-week period. These changes were maintained up to thirty-weeks for sixteen participants that continued. The research also successfully piloted the use of resources designed for self-management.

Conclusion: There are promising findings from this and other studies on the impact of singing for COPD on health and wellbeing. However, the outcomes of this research are consistent with those outlined in recent reviews and it is recommended that larger randomised controlled trials with longer durations are conducted.

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Chapter 1 - Introduction

Background

Respiratory Diseases

Hundreds of millions of people suffer each day from Chronic Respiratory Diseases (CRDs). These chronic diseases of the airways and other structures of the lung are among the leading causes of death worldwide according to the World Health Organization (WHO) **(WHO, 2017*a*)** and these make up five of the thirty most common causes of death with more than one billion people suffering from either acute or chronic respiratory conditions **(FIRS, 2017).**

The World Health Organisation (WHO) use the term 'Respiratory Illness' to encompass developmental and pathological conditions affecting the organs and tissues, that make gas exchange possible in higher organisms. This includes conditions of the upper respiratory tract, trachea, bronchi, bronchioles, alveoli, pleura and pleural cavity, and the nerves and muscles of breathing **(WHO, 1978)**. Respiratory illness in this context refers to the disorders outlined in a collective list of common chronic respiratory diseases identified by the WHO classification **(Bousquet et al., 2007)** which were detailed in the initial protocol (Appendix A, sub-appendix I) along with their related symptoms. Respiratory disorders generally manifest themselves with chest symptoms including shortness of breath and decline in lung function. These in turn lead to further symptoms such as weakening of muscles, pain and other related complications.

It is estimated that CRDs accounted for four million deaths (almost eleven percent of non-communicable disease deaths) in 2012 **(WHO, 2015)** and it is expected pulmonary diseases are likely to remain a major burden for decades to come. Prevention and treatment will need to be improved if their impact on longevity and quality of life of individuals and their economic burden on society are to be reduced.

Chronic obstructive pulmonary disease (COPD) is the term used for a subset of these illnesses, which generally includes two main conditions; emphysema and chronic bronchitis. World Health Organisation estimates **(WHO, 2017*b*)** that more than three million deaths were caused globally in 2015 by COPD alone, which represented approximately five percent of all deaths globally in that year.

COPD is characterised by poorly reversible airflow obstruction and an abnormal inflammatory response in the lungs, which represents the innate and adaptive immune responses to long-term exposure to noxious particles and gases **(MacNee, 2006)**. The main section of the GOLD 2013 diagnostic criteria states that a clinical diagnosis of COPD should be considered in any patient who has dyspnoea, chronic cough or sputum production, and a history of exposure to

risk factors for the disease. Spirometry is required to make the diagnosis in this clinical context; the presence of a post-bronchodilator FEV1/FVC <0.70 confirms the presence of persistent airflow limitation and thus of COPD **(Vestbo et al., 2013)**.

The concept of COPD as a syndrome with specific entities has evolved over time. Several definitions of COPD currently exist the first arising from working groups of the major respiratory societies i.e. the American Thoracic Society (ATS) and the European Respiratory Society (ERS)**.** The definitions have not always been particularly precise and may easily have included disease entities that would not usually be regarded as COPD (cystic fibrosis, bronchiectasis etc.) **(Vestbo, 2014)**. Significant national guidelines have subsequently adopted and modified these definitions, which can add further complexity to research in this area. Whether other concepts such as phenotypes will evolve and be included in future standards for diagnosis and management of COPD still remains to be seen **(Vestbo, 2014).**

The diseases which make up COPD are progressive and therefore long-term treatment options are currently limited and usually involve relieving the symptoms with medication (or surgery in a very small number of cases) **(NHS UK, 2016)**, but significant efforts now focus on pulmonary rehabilitation (PR) for these patients. This generally involves therapies to help strengthen the respiratory muscles and limbs through exercise (which in turn improves exercise tolerance). This aims to slow the progression of the disease and to help educate patients to prepare for the challenges of living with the disease and help support them in managing their related symptoms.

Dyspnoea

Dyspnoea, or shortness of breath, is perhaps the most common accompaniment of lung disease **(Burki and Lee, 2010)**. Patients with chronic respiratory disease are often limited in their activities by respiratory discomfort of this kind **(Meek et al., 1999)**. A reduction in functional status and quality of life of these patients, along with disability are frequent consequences. Diseases producing this symptom may leave patients with significant breathlessness despite maximal therapy and they are estimated to debilitate millions of people worldwide **(Herigstad et al., 2010)**.

The American Thoracic Society **(Meek et al, 1999)** describe respiration as being unique to all other vital functions, in that it is not only regulated by automatic centres located in the brainstem but also by voluntary signals initiated in the cortex. Individuals have some control over their breathing in that sensations arising from respiratory activity affect the rate and pattern of their breathing, as well as their functional status. Therefore, derangements or disturbances in the mechanics of this respiratory system and in overall oxygenation process may underlie the uncomfortable breathing sensations generally referred to as dyspnoea.

Since breathlessness is often perceived as a threat to survival, researchers **(Ryan et al., 2014)** propose that episodic breathlessness may also engage the human stress-response, as regulated by the hypothalamic–pituitary–adrenal (HPA) axis. It can be considered that chronic breathlessness can cause excessive stimulation of this axis, resulting in dysfunctional regulation, along with associated neuropsychological, metabolic and immunological outcomes.

Rehabilitation programs in patients with CRDs have been shown to relieve dyspnoea and reduce hospitalisations, but the understanding of mechanisms by which they succeed continues to be controversial **(Celli, 1995, Killian et al., 1992, Burki and Lee, 2010)**. The specific mechanism contributing to dyspnoea **(Meek et al., 1999)** and the symptoms of the dyspnoea **(Herigstad et al., 2010)** will both vary within an individual patient and from patient to patient, confounding the isolation of a definitive mechanism of action.

More than one process may contribute to the patients' functional ability and the patients' own emotional response to the illness could also exacerbate their response to the respiratory discomfort **(Meek et al, 1999)**. The symptoms of dyspnoea are not fully explained by the differences in disease severity in regard to lung function either. Patients suffering from dyspnoea often have very different pathologies, and it is likely that the psychological components of these conditions are also not identical **(Herigstad et al., 2010)**.

A Cochrane review of randomised control trials of Pulmonary Rehabilitation (PR) for COPD **(Lacasse et al., 2006)** found that while rehabilitation (i.e. exercise training) in patients with COPD showed clinically significant results for relief of dyspnoea and fatigue, and emotional function improvement, it did not indicate clinical significance for lung function improvement (Functional (FEC) and Maximal (MEC) Exercise Capacity). Lung function was not considered as a primary endpoint for the review and it focussed on health-related quality of life and exercise capacity. This would support the concept that dyspnoea and other related symptoms of the disease may not be directly related simply to the lung function alone.

In one documented case study **(Davies et al., 1987)** a patient experienced exertional dyspnoea after pulmonary surgery. Investigation excluded other key causes of dyspnoea and exercise studies demonstrated activity consistent with excess vagal nerve stimuli to the respiratory centre. Vagal nerve interruption via surgery markedly altered the patient's respiratory pattern and improved their functional status. While this finding is not relevant to respiratory patients specifically, it does highlight that excess vagal nerve fibre or 'afferent' activity caused the dyspnoea in this case and these mechanisms are would likely be interrelated to the complex underlying causes of this sensation in respiratory patients.

While the mechanism(s) and pathways of the sensations of dyspnoea remain unclear, it is speculated **(Burki and Lee, 2010)** that dyspnoea occurs when there is an increase of reflex information above usual levels from the peripheral sensors, provided by the pulmonary stretch and other receptors stimulated by the vagal nerve and chest wall mechanoreceptors. When

processed, these generate a neural output to the respiratory system, which suggests that when a patient's central neural output has unexpected results (i.e. via airflow or ventilation) either because of muscle paralysis or abnormal lung mechanics, a sensation of dyspnoea is experienced.

When an imbalance is perceived between the current or future ability to meet respiratory demand, but where airflow into and out of the lungs is still occurring, ''work-of-breathing'' dyspnoea develops. This is where various stimuli give rise to a perceived need for increased ventilation. Increased metabolic activity, production of CO_2, a lowered pH, increase in body temperature, perhaps anxiety or panic and increased sympathetic tone, among other things, signal the respiratory centre to increase respiration **(Hallenbeck, 2012)**.

A diagram summarising the short-term and longer-term factors impacting the respiratory systems for patients with COPD related illness and involved the sensation of dyspnoea is illustrated in figure 1 below.

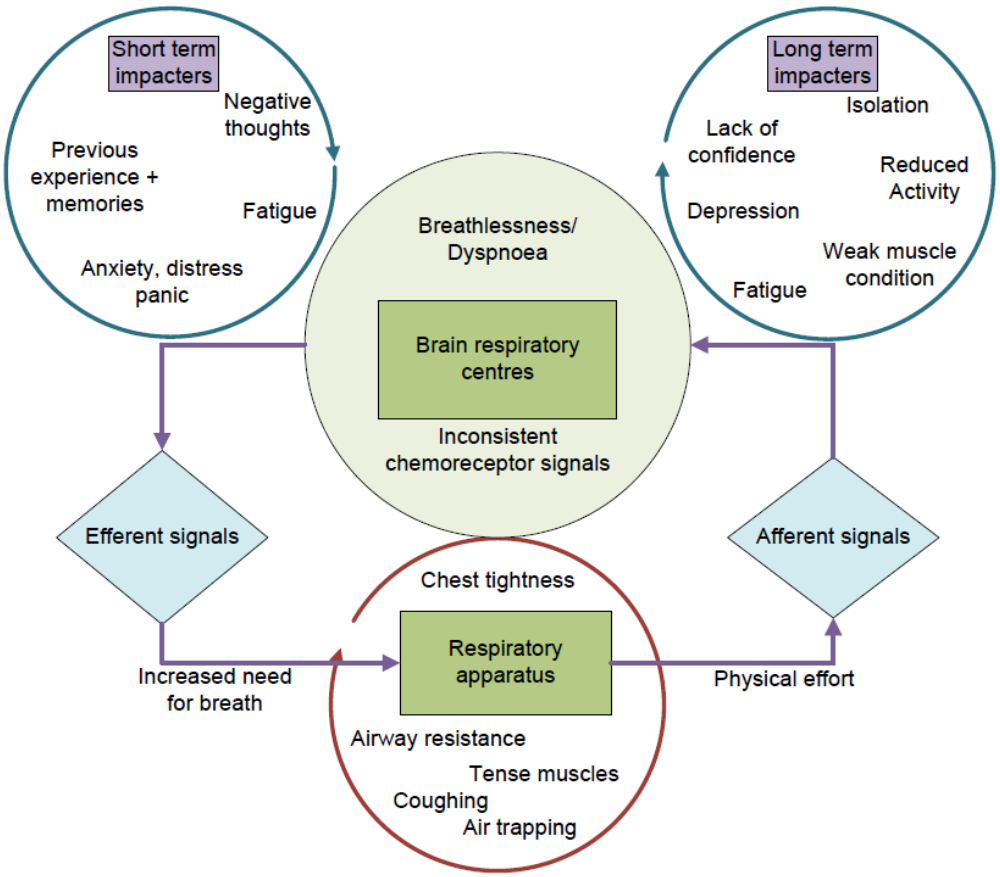

Figure 1 - Respiratory Stimuli Impacting Dyspnoea in COPD (created by author)

One study used functional MRI scanning and psychological and physiological assessments to indicate that breathlessness in COPD is associated with altered cognitive processing in the medial prefrontal cortex of the brain (i.e. differing brain activation patterns occurred in response to dyspnoea related 'word cues') between COPD patients and a control group **(Herigstad et al.,**

2013). In this study the control group displayed a similar activation pattern observed in previous functional MRI studies of breathlessness in healthy volunteers, while COPD patients displayed significantly greater activation in the medial prefrontal cortex (emotion control and memory consolidation). The behavioural data demonstrates that there may be a greater psycho-morbidity in the COPD patients **(Herigstad et al., 2013)**.

Imaging and behavioural findings indicate that in COPD, engagement of the medial prefrontal cortex may distort the processing of breathlessness sensations towards greater reliance or dependence on fear memories and expectations, contributing to a vicious cycle of fear and avoidance **(Herigstad et al., 2013)**. It has further been reported that distractive auditory stimuli decrease the level of exercise-induced dyspnea in patients with COPD by reducing their perceived unpleasantness of dyspnoea **(Von Leupoldt et al., 2007).**

The data obtained in a very recent study in animals suggest that star-shaped glial cells called astrocytes, characteristic in the brain and spinal cord, may also play a role in providing 'tonic excitation' of pre-Bötzinger complex (preBötC) circuits that generate the inspiratory rhythm **(Sheikhbahaei et al., 2018)**. The preBötC or pre-Bötzinger complex is a cluster of interneurons in the ventral respiratory centre of the brain, proven to be essential for the generation of respiratory rhythm and although this complex has not been definitively identified in humans, it is clearly identified and well-studied in animals **(Hallenbeck, 2012)**.

The role of preBötC astrocytes may be especially important in conditions where homeostatic adjustments of breathing are critical to support physiological and behavioural demands **(Sheikhbahaei et al., 2018)** and while this research is still in its infancy and the understanding of the physiologic mechanisms of dyspnoea and mechanisms of therapeutic action remains remarkably primitive **(Hallenbeck, 2012)**, it illustrates significant complexities. It is clear that there is still much to learn and understand about the engagement of the brain and respiratory systems and the mechanisms that might impact the links between exercise and breathlessness.

Inflammation and Depression

It is understood that untreated comorbid anxiety and depression in patients with COPD has devastating consequences and may increase healthcare utilisation. Depression and anxiety are challenging to identify and treat in this group because their symptoms often overlap with those of COPD. It is estimated that less than one-third of COPD patients with comorbid depression or anxiety symptoms are receiving appropriate treatment. The evidence for the efficacy of antidepressant drug therapy in patients with COPD with comorbid depression and anxiety is inconclusive **(Yohannes and Alexopoulos, 2014)**. There are however, promising findings regarding pulmonary rehabilitation, psychological therapy, and the collaborative care model in reducing depression and anxiety symptoms in patients with COPD, but these findings are currently limited by short-term follow-up periods **(Yohannes and Alexopoulos, 2014)**.

It is well established that a number of inflammatory cell types are involved in the pathophysiology of COPD including macrophages, neutrophils and T-cells (which are found in all tissues including the lungs). These cells release a variety of mediators in response to noxious particles and gases which orchestrate and perpetuate the inflammatory response in COPD **(Austin et al., 2016)**. In addition to an increase in the number of macrophages and neutrophils, these cells appear to have an impaired phagocytic function, resulting in impairment in clearance of apoptotic (dead or no longer needed) cells and potentially contributing to the chronic inflammatory state in the lungs of patients.

Those who develop COPD also have an enhanced or abnormal response to inhaling toxic agents. This amplified response may result in mucous hypersecretion as in chronic bronchitis patients, tissue destruction seen in emphysema, and disruption of normal repair and defence mechanisms causing small airway inflammation and fibrosis (bronchiolitis). These pathological changes result in increased resistance to airflow in the small conducting airways, increased compliance of the lungs (i.e. 'highly compliant' refers to lungs where elastic tissue is damaged by enzymes), air trapping, and progressive airflow obstruction **(MacNee, 2006)**.

Depressive symptoms are common in patients with COPD and both diseases are believed to be associated with inflammation. Those patients with severe COPD have a 2.5 times greater risk of developing depression than controls i.e. people without COPD **(van Manen et al., 2002).** Evidence suggests that pro-inflammatory cytokines and cortisol in particular, play a crucial role in the causes of COPD and depression. **(Du et al., 2014)** and depression is a disorder which remains easily undiagnosed due to under-presentation and because the symptoms are not very specific. It is important to consider this disorder in patients with COPD and research underscores the importance of reducing symptoms and improving physical functioning in patients with COPD rather than focusing on pulmonary function alone **(van Manen et al., 2002).**

A previous study **(Du et al., 2014)** investigated further the associations between depression, sputum cytokines and salivary cortisol in COPD patients. Diurnal rhythms of sputum biomarkers and salivary cortisol were measured in COPD patients with depression compared to those with depression alone, COPD alone and healthy controls. The combination of various sputum biomarkers and salivary cortisol VAR (relative diurnal variation) were found to perform best as a potential biomarker in the diagnosis of depression in COPD patients.

There is early ongoing laboratory research which indicates that the role of the immune system in depression could be due to T-cells protecting against stress and depression. These very early findings suggest that therapies that boost such T-cell responses could be used in some patients with depression. Decreased numbers of peripheral blood T-cells and reduced concentrations of anti-inflammatory cytokines in the blood have also been reported in depression. Thus, it appears that patients with depression may have impairments in neuroprotective and anti-inflammatory T-cell responses **(Miller and Raison, 2016)**. This research could also hold potential importance for COPD patients with depression.

There are ongoing efforts to understand and build on these known links between depression and inflammation, which could have relevance for COPD patients and their treatment. A recent study **(Cattaneo et al., 2016)** expanded on previous findings that depressed patients, resistant to conventional antidepressant therapies, appear to have higher concentrations of inflammatory biomarkers and that successful antidepressant treatment has been associated with a reduction in the levels of pro-inflammatory cytokines. This study looked in further detail at certain inflammatory biomarkers and found results that may support faster prediction of antidepressant response in particular patients. This could hold further implications for other research areas, such as COPD where inflammation and depression are key components.

Singing, Respiration and Science

Breathing, Stress and Wellbeing

It is accepted that breathing behaviour is an important area of study with implications for stress research **(Grossman, 1983)**. Specific breathing techniques are regularly recommended for relaxation, stress management, control of psycho-physiological states and to improve organ function (via improved circulation). Yogic breathing, defined as a manipulation of breath movement, has been shown to positively affect immune function, autonomic nervous system imbalances and psychological or stress-related disorders **(Zope and Zope, 2013)**. The slow-paced respiratory rate exercise has been found to have a strong tendency of improving or balancing the autonomic nervous system through enhanced activation of the parasympathetic system and thus can be practised for mental relaxation and reduction of stress of in daily life. This respiration and heart rate variability has been linked to physiological benefits, for example, lowered blood pressure **(Pramanik et al., 2009).** It is speculated that this may act similarly to mechanical hyperventilation and electronic unilateral vagal nerve stimulation (VNS), resulting in quieting of the frontal cerebral cortex **(Brown and Gerbarg, 2005)** the area of the brain involved in feelings of anxiety and stress.

There is building evidence globally that singing, which is highly dependent on controlled breathing holds similar implications. A report showed that 71% of singers in a university choral society agreed that singing was beneficial for their ‘mental wellbeing’ **(Clift and Hancox, 2001).** The results of a study conducted in Germany **(Kreutz et al., 2004)** supports the hypothesis that choir singing positively influences emotions as well as immune functions in humans, findings previously discussed by other researchers **(Unwin et al., 2002**, **Beck et al., 1999)**. The analysis of saliva samples taken at baseline and post group singing or listening indicated specific changes in the levels of secretory Immunoglobulin A (S-IgA), cortisol and emotional state **(Kreutz et al., 2004)** which collectively support the idea that choir-singing positively influences both emotional status and immune competence.

The results of a systematic review of the psychoneuroimmunological effects of music **(Fancourt et al., 2014)**, also demonstrates an apparent sensitivity of hormones to musical stimulation. Immunoglobulin A (S-IgA) has been revealed to be particularly responsive to music, increasing following exposure to a range of styles of music, as well as for both active involvement and simply listening to recorded music. Similarly, strong patterns can be noted with respect to cortisol, and response of epinephrine (adrenaline) and norepinephrine (non-adrenaline). The review indicated that more studies will be needed to confirm this pattern though, as previous studies have not managed to achieve statistical significance. A meta-analysis would have been useful in providing a balanced judgement across all studies. However, this was not possible, due to the variation in methods used by the studies selected.

There is also evidence from recent research in Cancer patients and their carers that greater improvements in mood, as a result of singing are associated with lower pro-inflammatory response, which appeared to be independent of stress levels. In this study, those with the lowest levels of mental wellbeing and highest levels of depression experienced the greatest short-term improvement in mood across the singing sessions and these larger mood changes were associated with lower levels of inflammation **(Fancourt et al., 2016)**.

It is unclear whether the increase in Immunoglobulin A holds any significance to physical functionality, but inflammation and depression are key factors in the pathophysiology of COPD, so this finding could hold important implications in further understanding the processes by which group singing might support the health and wellbeing of COPD patients.

The effects of stress and emotion are often measured using respiratory parameters **(Wientjes, 1992)**, with psychological distress generally leading to increases in respiration rate and a change over from diaphragmatic or 'deep' breathing to thoracic or 'shallow' breathing. In extreme cases, a lowering of partial pressure of carbon dioxide (pCO_2) values can be seen, with concomitant symptoms of hyperventilation **(Hibbert and Pilsbury, 1989)**. However, attention has focused more recently on relationships between respiration and vagal control over the heart. This direct cardiorespiratory interaction is reflected in a phenomenon known as respiratory sinus arrhythmia (RSA) **(de Geus et al., 1995)**.

Respiratory Sinus Arrhythmia and Heart Rate Variability

Respiratory sinus arrhythmia (RSA) is a cardiorespiratory phenomenon characterized in mammals by heart rate (HR) or R-R interval (RRI) fluctuations (i.e. cardiac beat-to-beat ECG intervals) that are in phase with inhalation and exhalation. RSA is defined as the coherence between respiration depth and heart rate. Typically, heart rate accelerates during inspiration and slows down during expiration, but the exact phase relationship, between respiratory and heart rate oscillations, is dependent upon the prevailing respiration rate **(Eckberg, 1983).** Respiratory sinus arrhythmia has been shown to reflect rhythmic waxing and waning of cardiac

vagal efferent effects upon the sinoatrial node (i.e. the carrying of impulses from the central nervous system to an effector) and thus on heart rate **(Eckberg, 2003)**.

Vagus Nerve Stimulation (VNS) (a technique providing targeted electrical therapy to the left cervical vagus nerve to control refractory epilepsy and treatment resistant depression) has also been seen to cause side effects involving the upper airway, lower respiratory tract, and upper gastrointestinal tract. These VNS devices can decrease airflow, oxygen saturation, and respiratory amplitude during sleep **(Ghanem, 2006)**. Electronic stimulation of left vagus nerve afferent fibres, which carry impulses to the central nervous system (at the current levels required to provide therapy in this patient population) has been seen to cause adverse events of vocal cord dysfunction, laryngeal spasm, cough, dyspnoea, nausea, and vomiting. Specific Vagus Nerve Stimulation (VNS) of this kind has been seen to cause an increase in respiratory rate, decrease in respiratory amplitude, decrease in tidal volume (volume of air inspired or expired in a single breath during regular breathing) and decrease in oxygen saturation during periods of device activation, **(Parhizgar et al., 2011).** This further supports the concept that vagal tone might have key implications for the sensations of dyspnoea and other related respiratory and vocal functions.

In summary, vagal tone (and specifically its influence on heart rate) represents a measure of the functional state of the parasympathetic nervous system, which is the division of the autonomic nervous system which acts to conserve energy. Vagal tone is most commonly measured by analysing the rhythmical variations in heart rate period or RSA, due to the respiratory cycle. Increases in vagal tone slow the heart and make heart rate more variable and although ‘vagal tone’ itself cannot be measured, changes in the heart rate due to RSA can be found by measuring periodic changes in the heart rate due to breathing and can be used to index autonomic vagal activity **(Thayer et al., 2011)**. This process is referred to as the heart rate variability (HRV).

Early research examined synchronization of respiration and HRV among choir members during singing **(Muller and Lindenberger, 2011)** and it was further investigated how singing, as a form of guided breathing, affects heart rate variability and respiratory sinus arrhythmia in healthy patients **(Vickoff et al., 2013)**. It was concluded that singing produces slow, regular and deep respiration which in turn triggers respiratory sinus arrhythmia. Respiratory sinus arrhythmia is associated with vagal influence and self-reported well-being, so researchers suggest that singing can be viewed as initiating the work of a vagal pump, sending relaxing waves through the choir. The singing itself guides respiration, resulting in compliances of frequencies and phases of respiration cycles and heart rate variability cycles between singers. This could hold implications for patients with a respiratory illness and may help researchers in understanding which aspects of group singing in particular, may have been effective in those trials **(Clift et al., 2013)** where encouraging results for COPD patients have been noted.

The influence of the inspiratory muscle weakness in COPD patients on cardiac autonomic control is as yet unknown. One study **(Reis et al., 2010)** evaluated the influence of respiratory muscle strength on autonomic control and found COPD patients demonstrated impaired cardiac autonomic modulation at rest and during RSA-M when compared with healthy subjects ($p < 0.05$). Patients with COPD presented impaired sympathetic-vagal balance at rest. In addition, cardiac autonomic control of heart rate was associated with inspiratory muscle weakness in COPD. It is speculated based on this evidence that future research applications of respiratory muscle training may bring to light a potentially valuable target for rehabilitation.

Group Singing for Respiratory Illness

Researchers have studied the effects of emotion induction on total respiratory resistance in asthma patients and the relationship with cardiac vagal activity and facial muscle activity **(Ritz et al., 2001)**. Significant increases of respiratory resistance in asthmatic patients were observed following 'depressing stimulation' (i.e. presentation of coloured pictures depicting scenes of hospital patients, scenes of catastrophe, soldiers in action, dead animals). Resistance increases were positively correlated with respiratory sinus arrhythmia and researchers concluded that the respiratory resistance increases in asthmatics following observation of these 'depressing stimulations' are dependent on vagal activity. Greater facial muscle activation during emotional stimulation is seen to reduce vagal activation too.

Evidence further suggests that singing has the potential to improve health outcomes in patients with other chronic respiratory diseases. A feasibility study on the health benefits of a community singing programme for older people with COPD **(Clift et al., 2013),** strongly support this. The measures of lung function and health-related quality of life significantly improved over a ten-month period of the singing programme **(Clift et al., 2013)** further supported by qualitative data collected from the same study **(Skingley et al., 2014).**

Group singing is an activity that may support lung function and enhance quality of life in people with certain respiratory disorders. The current literature and research though, has mixed and inconclusive findings, suggesting that several different but interrelated processes could potentially be active in differing degrees.

The key mechanism appears to be the active engagement of the muscles of the entire respiratory system and an increase in respiratory muscle strength, leading to increased lung volume and effective cough **(Kang et al., 2006, Wiens et al., 1999)**. Poor posture (hyperkyphosis) is often common in COPD patients and can restrict the expansion of the rib cage and movement of the diaphragm. Since singing requires the development of skills in controlling posture this may be transferable to activities in daily life in people with COPD **(Lord et al., 2010)**.

In addition, group singing includes a strong element of education by singing facilitator to help support patients with their breathing control. It is well accepted that teaching respiratory patients to breathe slowly and deeply during exercise, and avoid rapid upper thoracic patterns of breathing, should help to lessen dyspnoea and improve performance **(Macklem, 2010)**. Respiratory patients indicate that a crucial element in improvement to their breathing during group singing is the aspect of learning how to breathe correctly and to control their own breathing **(Skingley et al., 2018)**. It is also considered an important benefit that the action of breathing more slowly allows patients lungs more time to empty, thus reducing the acute effect of air trapping in the lungs **(Lord et al., 2010)** which impacts the mechanism of dyspnoea.

There are other mechanisms active that may potentially impact this population. One of these is mucus clearance. This could be due to the vibrations caused by the physical activities of singing, as the techniques used in singing itself follow similar scientific pathways that form the basis of successful chest physiotherapy, airway clearance, and airway clearance devices **(Goldenberg, 2018)**.

It was initially demonstrated in an animal model, that pressure oscillations at the chest wall cause the release of acetylcholine **(King et al., 1990)** stimulating cilia beating and helping to increase mucus flow. In singing, the frequency of the pressure oscillations will vary according to the pitch being sung, but any vibrations less than 350Hz can be felt at the chest wall **(Sundberg, 1992)**. These pressure oscillations during singing may also change the rheological properties of the mucus. The increasing shear rates from oscillating airflow in vitro were seen to reduce mucous viscoelasticity, making movement and thus clearance easier **(Tomkiewicz et al., 1994, Sturgess et al., 1970)**. Inspiration during singing would usually be around 5 l/s **(Sundberg, 1987)** and peak expiratory flow rates above 0.5 L/s can effectively shear and pull mucus within the airway **(Kim et al., 1987),** so although inspiratory airflow is not in the direction for expulsion of mucus the mucus would still be loosened such that it is easier to expel upon coughing.

It is common during singing sessions for COPD patients that the activity of singing induces a high prevalence of coughing and sputum expectoration **(Bonilha et al., 2009),** indicating this expected mobilisation of mucus. Studies of singing in cystic fibrosis patients in particular have observed participants coughing during singing though not during conversation **(Goldenberg, 2012),** which supports an understanding that singing not only improves respiratory muscle strength but as the physiology of singing resembles the principles behind airway clearance, it facilitates the mobilisation of mucus.

The other mechanisms that may have significance include a reduction of fear, anxiety and pain perception **(Kenny and Faunce, 2004)** possibly by increased breath control improving the autonomic nervous system through enhanced activation of the parasympathetic system, leading to higher vagal tone **(Pramanik et al., 2009, Kok and Fredrickson, 2010, Vickoff et al., 2013);** This in turn and in combination with the improvements in a patient's overall improved

consciousness of their own breathing due to the singing training, may reduce the occurrence or severity of dyspnoea episodes; Improvement of mood **(Unwin et al., 2002)** positively impacted by socialising and meeting people, thus combating isolation. Finally, immunoglobulin A (S-IgA) levels have been revealed to be particularly responsive to music **(Fancourt et al., 2014)** and there is evidence that greater improvements in mood as a result of singing were associated with lower pro-inflammatory response **(Fancourt et al., 2016)**. This leads to consideration of a potential link, still to be more rigorously examined, between regular singing and more sustainable improvements in mood and positive impact on depression and inflammation. This in turn would be a particularly important outcome for COPD patients specifically, where depression is more prevalent.

These multiple complex processes are thought likely to be interrelated, thus enhancing the overall potential benefit by form of a virtuous circle, combating the 'spiral of decline' **(Jones, 2009)** associated with the condition. It can be hypothesised that singing as an enjoyable pursuit, may help to prolong the continued engagement and retention of patients in regular activities which support their condition longer term. The variety of cost effective and self-sustaining options available for patients within this area needs to be explored further both in regard to longer treatment and follow-up periods, overall healthcare utilisation and particularly to understand further the mechanisms and processes employed.

Research Importance

The WHO European health report **(WHO, 2012)** indicates that the main specific causes of death from respiratory diseases are chronic obstructive pulmonary disease (COPD), pneumonia, influenza and asthma and that the interventions to prevent such respiratory diseases often reflect wider social determinants of health that require some inter-sectorial action to ensure an impact that lowers disease incidence and severity.

The research outlined here is in line with this analysis and also with existing UK Chronic Obstructive Pulmonary Disease (COPD) outcomes strategy discussions **(UK DH, 2012)** particularly objectives identified in the companion document, which outline directions for enhancing quality of life for these patients. It is also aligned more broadly with a general movement away from past models of health care which formally placed the patient in the role of a passive recipient. Many chronic disease patients now require more active involvement in managing their conditions, whereby responsibility for day-to-day disease management gradually shifts away from health care professionals to the individual **(Barlow et al., 2002)**.

Research suggests that specific theoretical work is required to link the nature, process and experiences of 'singing' with the nature, processes and experiences of wellbeing and health **(Clift et al., 2008)**. It considers that the key questions needing to be addressed are 'What?' and 'How?' What effects, if any, does active involvement in group singing have on wellbeing and

health? And how does singing have these effects – what mechanisms are at work? What mechanisms, for example, link the physicality of singing with the physical wellbeing of the body? What mechanisms link the psychological and social processes at work during singing with senses of emotional wellbeing and mental health?

There is indication from the research literature available that the potential mechanisms and interactive aspects of this intervention may support health outcomes in respiratory challenged patients more generally e.g. positively influencing both emotional status and immune competence **(Kreutz et al., 2004).** It can also be hypothesised that, depending on the nature of the underlying respiratory illness in question, differing aspects of the activity of singing may be more or less impactful to patients and their symptoms too i.e. The overall effect of singing may be variable dependant on the pathology of the disease, the area of the human respiratory system impaired or the respiratory apparatus impacted by the treatment. e.g. vocal training may reduce snoring or Obstructive Sleep Apnoea (OSA) by improving the pharyngeal muscle tone **(Ojay and Ernst, 2000)** or it may support cystic fibrosis by providing rigorous exercises for the whole respiratory system as well as a means for emotional expression, in turn enhancing quality of life **(Irons et al., 2010*a*).** This suggests that understanding the impact of singing on specific symptoms within a single disease group or population may prove an important step forward, in addition to further understanding the holistic effects on wellbeing in general.

The implications of group singing on respiratory sinus arrhythmia and other psycho-physiological functions of patients with a respiratory disorder and on related symptoms such as dyspnoea in particular, are still not fully understood. However, group singing has the potential to be an innovative, cost-effective, low risk therapy which helps participants to engage in physical and social activities and which can serve to support independence and improved quality of life. This planned research intends to provide additional knowledge in this area and enhance the impact of singing for patients with a respiratory illness.

Objectives

The aims of this research were to add to the growing body of evidence in the area of singing for wellbeing and health, particularly for patients with COPD, to help answer the following research questions.

1) What has been established so far by research regarding the effects of group singing for COPD patients?

2) What are the potential health and wellbeing benefits of regular participation in group singing for a small cohort of COPD patients?

3) What is the potential feasibility and usefulness of developing and implementing novel singing resources in a group of COPD patients, in addition to regular group singing?

The key objectives were to review current research systematically and evaluate the potential mechanisms, processes and effects of group singing as an intervention for pulmonary function and health status. The research was divided into three discrete components:

- Review, synthesise and consolidate current research in the area of group singing for COPD patients and use this information to support the design of an empirical research study

- Evaluate the impact of group singing on health and wellbeing for a cohort of COPD patients using patient reported outcomes measures (WEMWBS, CAT, ACQ-5) and provide recommendations to inform and support future study design and planning.

- Evaluate the feasibility and usability of specially designed resources for home singing practice for respiratory patients.

The initial planning for the systematic review was conducted during 2013 with an initial protocol completed in August 2013. Initial searches and data extraction started at that point. In December 2015 the Cochrane review title ‘Singing for Adults with COPD’ was registered and the Cochrane review protocol ‘Singing for Adults with COPD’ published in July 2016.

It was recognised at this point that the initial planned protocol closely aligned to the Cochrane methodology and as such this separate planned systematic review was largely replicating the planned Cochrane review. It was decided at that point that it would add more value to this research if the scope of the systematic review be broadened to incorporate the review of design and execution aspects of the available studies. This planned review would remain in close alignment with the Cochrane review but there would be no restriction on the types or quality of the studies to be included.

The first version of this broad systematic review was completed in December 2016 and this was used to inform the design of the Empirical study. Recruitment planning and taster sessions for planned COPD singing groups were initiated in spring and summer 2016 prior to the finalisation of the study design. This was to ensure the singing groups, resources and venues could be arranged during winter 2016 allowing for group sessions to be conducted in the following summer period. The protocol for the empirical study was completed in March 2017 and received Ethics approval in April 2017. The study was successfully conducted at two COPD singing groups during the summer 2017.

It was further decided during the ongoing study conduct in September 2017 that there would be value in continuing the study and collecting further data for these singing groups, so the protocol and study plans were revised to enable the collection of data for one additional visit timepoint. A

request was submitted and further ethics approval granted. The study then completed in November 2017 and all final data was analysed in winter 2017. The final thesis was completed and submitted in June 2018.

Chapter 2 – Background: Systematic Review of Current Literature

Systematic Review: Introduction

The Cochrane Collaboration has completed systematic reviews for singing in patients with certain specific respiratory disorders, for example 'Singing for Cystic Fibrosis' **(Irons et al., 2010*a*)** and 'Singing for Bronchiectasis' **(Irons et al., 2010*b*)**, however these were 'empty' reviews and did not lead to any conclusions. A good quality collective review of 'Singing for chronic respiratory disorders' or 'Singing for COPD' had not been published by Cochrane or other recognised organisation at the onset of this project in 2014. However a Cochrane review protocol was published for 'Singing for Adults with COPD' in 2016 **(McNamara et al., 2016)** and the full review and subsequent publication **(McNamara et al., 2017)** competed in parallel with this project. However, the total number of randomised controlled trials (RCTs) in this area, eligible for inclusion, was small and the total sample sizes used and follow-up periods employed did not provide enough robust information to provide significant conclusions.

Two other systematic reviews were published during this project which similarly agreed with the overall outcome of the Cochrane review 'Singing for Adults with COPD' **(McNamara et al., 2017)**. The first, **(Gick and Nicol, 2015)** considers studies of singing for both COPD and other respiratory conditions and indicates that improvements are not shown in respiratory health directly for most studies. It shows instead that improvements in quality of life or well-being have a positive, immediate impact and might further affect respiratory health over time. The other review **(Lewis et al., 2017)** provides a detailed and critical review of the existing literature on singing for lung health and concludes that further, larger-scale and more robust studies are needed to test the therapeutic effects of singing, but it does acknowledge that there is considerable qualitative data to support participation in singing groups as a safe and potentially valuable strategy for these patients.

In addition to the more rigorous Cochrane review and these further highly specific systematic reviews, a broader systematic review is still considered important. The aim is to research all study designs, methodology and outcomes published in the area of singing for adults with COPD to date, with the intent of consolidating existing evidence in one place, to support future research methodology and practice development. A further aim of this review is to explore whether there are any insights to be gained from currently published research that concern any additional aspects that group singing as an intervention may have for COPD symptoms specifically, which have not previously been considered.

The recently completed Cochrane review 'Singing for Adults with COPD' certainly holds more significance and impact in regard to the analysis of the previous data obtained, but since it

focuses exclusively on randomised controlled studies, there remains valuable qualitative information to be provided by an extended review of all ‘Singing for COPD’ related studies. A collective review of all studies including those that were not randomised controlled trials from any additional publications will provide a comprehensive review of current information. The researcher for this systematic review and project was also a contributing author for the ‘Singing for adults with COPD’ Cochrane review **(McNamara et al., 2017)** so these two reviews remain sympathetic and consistent.

This broader review aims to add context and compliment the Cochrane review. It focuses on the nature, methodologies and quality of all current work to date, including all studies in this area, i.e. mixed model, quantitative and qualitative. The intent of this is to supplement the outcome of a Cochrane review conducted in parallel and to help shape agenda for future research.

This extended review completed alongside the Cochrane review focuses on the following areas:

1. The quality, design and settings employed in execution of the studies, including the frequency and duration of the singing intervention.

2. The health and wellbeing measures used, data collected and the overall research problems identified by researchers.

3. The discussion and consideration of any general findings in this area with particular focus on any outcomes which may clarify further the aspects or mechanisms which group singing may employ for COPD patients

Systematic Review: Method

The design criteria outlined in the original protocol, ‘Singing for Chronic Obstructive Pulmonary Disease: A Systematic Review of Currently Published Literature’ as shown in Appendix A, was followed. The initial version of this protocol acted as the framework for a review of all work to date however, the scope for this protocol included research for singing for all patients with a respiratory disease and it was decided, following the initial database searches, to narrow this review criteria to focus more specifically on COPD related disease i.e. ‘singing for adults with COPD’ and to exclude studies on singing for other non-COPD respiratory illnesses. This also enabled the review to remain closely aligned with the Cochrane review protocol ‘Singing for Adults with COPD’ **(McNamara et al., 2016)**, developed in parallel, and to ensure direct comparability with the specific patient populations.

The scope for the broader systematic review was expanded both in its intent to review all aspects of study design and execution and also in regard to inclusion of all study types and designs. It aimed to include all papers to date for singing and COPD, without exclusion. All

studies that contained pertinent content for an expanded review of the methodology, implementation, practice implications and findings for group singing and COPD were incorporated. The initial searches had been deliberately conducted so as not to specify or exclude any particular study type, as the intent was for the researcher to procure all studies available for singing in COPD in their entirety and manually filter those with the required study type. Hence the original searches remained valid as they were designed to capture all randomised controlled trials, quasi-experimental designs, qualitative, quantitative and mixed model designs, recorded in any language containing the intervention and population terms as outlined in the original protocol.

Systematic Review: Results

Database Searches

The initial protocol was developed with the aim to focus on singing in all Chronic Respiratory Diseases (CRD). Hence the planned search strategy for this systematic review included all papers relating to Chronic Respiratory Diseases (CRD) as identified per WHO ICD10 by the Global Alliance against Chronic Respiratory Diseases **(Bousquet et al., 2007)** where singing was included as the intervention. The search criteria were very broad and thus the searches included studies which had less strict design criteria and a wide range of respiratory illnesses.

The initial searches based on this early strategy were completed in August 2014 and all papers were procured and the characteristics of each study were collated and databased as per protocol over the following year. This produced 2055 titles of which there were 165 duplicates leaving a total of 1963 unique studies. The titles were reviewed for relevance and all those meeting the specific criteria of singing for respiratory illness extracted and reviewed.

There were found to be a number of relevant studies including some that were excluded from previous Cochrane reviews. Studies were found to be highly variable over a number of parameters both within the design, respiratory illness type and in the quality of execution and reporting.

The results from the initial searches and identification of relevant papers led to the protocol being revised to narrow the focus to those studies involving COPD alone, and also to review studies with a more robust design methodology i.e. randomised design, feasibility and pilot studies in line with more distinct criteria. It was decided that studies with a high risk of bias or with confounding factors in the population (e.g. patients and carers, population bias, stratification by gender etc.) should be excluded.

The original key database searches (completed in 2014) plus additional manual searches and review of references within all relevant studies produced a total of twenty-six papers that met all

the original protocol inclusion criteria i.e. singing as an intervention for respiratory illness. A total of four titles were registered studies only without a full paper (which were all excluded) and one title was an abstract with no other information available. The remaining twenty-four papers detail relevant studies where diseases of interest could be more simply categorised into six groups by their pathology: Asthma; Sleep Apnoea/Snoring; Lung Cancer; Cystic Fibrosis; Emphysema; COPD/CRD.

These database and manual searches were performed using broad criteria from the original protocol and then this full list of studies and papers manually pared further to include only those where patients were specifically noted as having COPD or a related illness (e.g. Emphysema). This produced a total of sixteen papers from eleven unique studies. Updated searches were run in fourth quarter of 2015 and first quarter of 2016 but produced no further studies of relevance.

The Cochrane review title 'Singing for Adults with COPD' was registered December 2015 and the protocol published in July 2016. Database and manual searches were completed for this protocol in August - September 2016 and while Cochrane's methodology for effectiveness reviews requires that only randomised controlled trials (RCT) are included, the searches were much wider and acted as further important quality check for studies to be included in this review.

These searches in 2016 surfaced one additional more recently published study **(Canga et al., 2015)** along with a clinical trials registration abstract **(Canga et al., 2014)** for the same study, which met the criteria for this systematic review. These searches also identified additional conference abstracts for the study by **Bonilha et al. (2009) (i.e. Martinez et al., 2008),** for the **Lord et al. (2010) study (i.e. Lord et al., 2010*b***) and an abstract and an erratum paper for the **Lord et al. (2012) study (i.e. Lord et al., 2012*a*, Lord et al., 2012*e*)**. These studies had already been included in searches for this review and selected, but these five additional papers were included for completeness.

One further study was noted to be registered in the ANZCTR database of clinical trials i.e. **(McNaughton, 2014)** '*Sing Your Lungs Out: Effect of participation in weekly community singing group for 1 year, on lung function, anxiety and depression and health care use in patients with COPD who have completed pulmonary rehabilitation: a quantitative and qualitative study'* but this study data was not available at the time of the initial review.

In March 2018 further manual and database searches were conducted to ensure all updated information was captured. The study mentioned previously **(McNaughton, 2014)** was traced and two papers had since been published documenting this study. One paper was published in 2016 **(McNaughton et al., 2016)** and then a further updated paper was published in 2017 **(McNaughton et al., 2017)** both for the same study. These were both subsequently assessed and included along with the additional six papers surfaced in 2016.

Seven further published papers from six unique studies became available within this time and were subsequently added **(BLF, 2017, Clift et al., 2017, Liu et al., 2016, Jamaly et al., 2017, Skingley et al., 2018, Thomas et al., 2015, Trivedi, 2017)**. The outcome is that a total of sixteen additional eligible papers were identified between mid-2016 and spring 2018 and sourced from nine unique studies. These were subsequently added to the systematic review and detailed in figure 2 below:

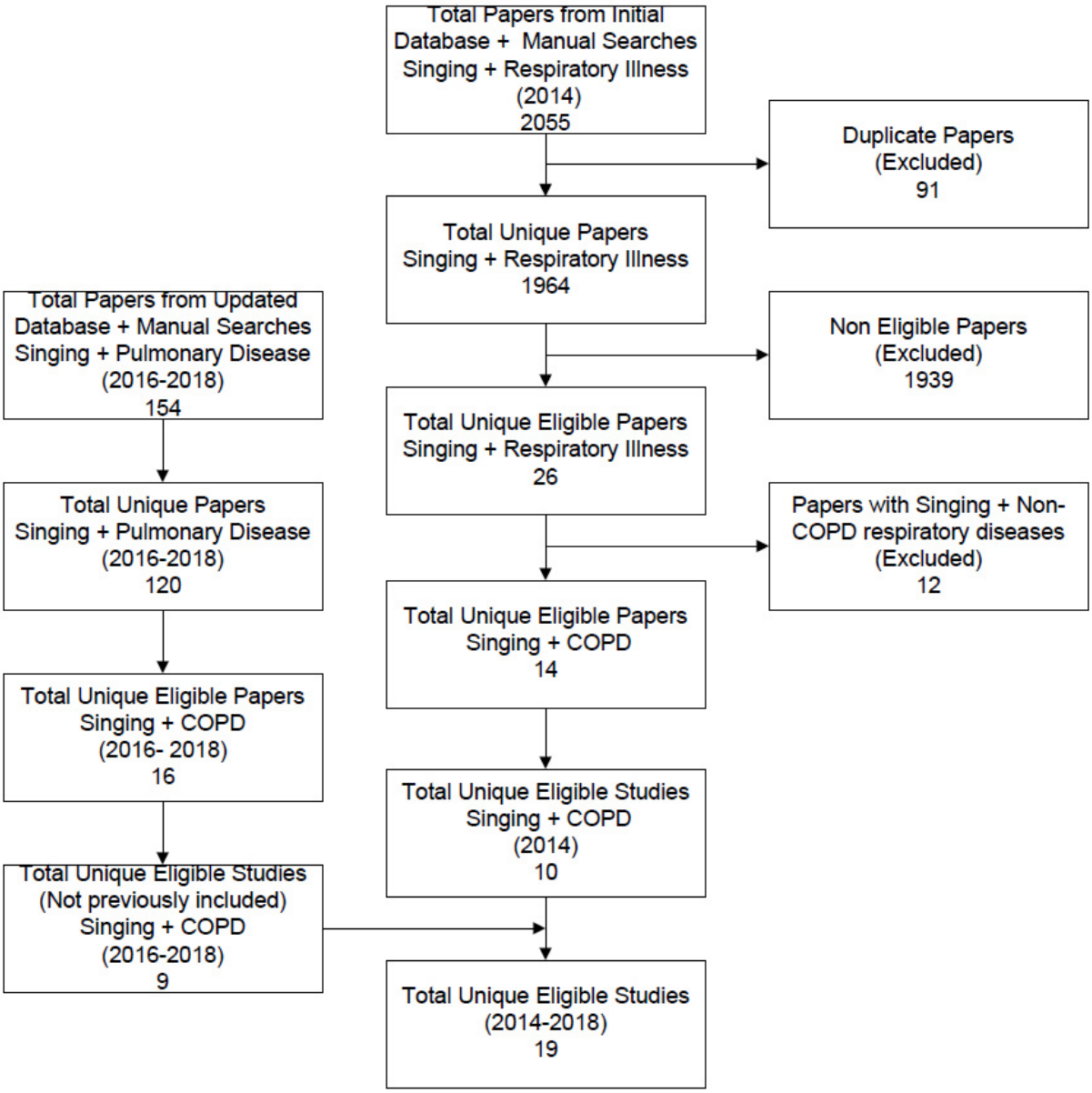

Figure 2 – Systematic Review - Flow of Papers and Studies through the Review

Study Selection

There were thirty key papers identified from the initial and subsequent searches and full reviews as detailed in Table 1 below, which reported on nineteen unique studies in total. All the papers by **Clift et al. (2013), Morrison et al. (2013) and Skingley et al. (2014)** present different

aspects of a single study, as do **Clift et al. (2017)** and **Skingley et al. (2018)**. **Lord et al. (2010*a*) and Lord et al. (2010*b*)** papers both present abstracts of the study as detailed by **Lord et al. (2010).** The paper by **Lord et al. (2012*a*)** describes an abstract of the study presented in full in **Lord et al. (2012),** and **Lord et al. (2012*e*)** also provides an erratum to this same study. **McNaughton et al. (2017)** is a later update (including quantitative findings) from the same study detailed by **McNaughton et al. (2016)** which detailed the initial qualitative findings. **Martinez (2008)** is a conference abstract of the study of **Bonilha et al. (2009)**. The details described in these papers, are grouped and collectively presented under a single study within subsequent tables and text, for simplicity i.e. **Clift et al. (2013), Lord et al. (2010), Lord et al. (2012), McNaughton et al. (2017)** and **Bonilha et al. (2009)**. If data is referenced uniquely only in one of the papers within the group, then that specific paper may be referenced. These papers are noted with an additional unique alphanumeric identifier relating it to the main study paper in the table, as appropriate.

Three papers **(Miyahara et al., 2001, Jamaly et al., 2017 and Liu et al., 2016)** were conference abstracts for relevant studies. The full study paper for each was not available during this systematic review despite attempts to follow up to obtain information, however key information available in the abstract was still utilised for the purpose of the extended review. The information in these studies appears to contain potentially important outcome data. This review will be updated should information become available at a later date.

The majority of the studies selected were conducted in Europe (UK, France, Germany and Portugal) with four studies in North America (US, Canada), four in Asia Pacific (New Zealand, India, China and Japan) and one in Brazil. No further publications were available for studies conducted in any other languages or further countries worldwide from the global databases that were searched or from the further manual searches.

The studies that have been published to date were, for the most part, conducted in higher income countries, with a bias overall to educated participants, where this information was recorded.

Demographic Information

In the majority of studies selected the demographic measurements for patients, as noted in Table 1 below, was limited to ‘Age’ and ‘Gender’. Fifteen of the unique studies captured age data in various formats while thirteen captured gender. The majority presented the average age rather than providing more detailed age data or any further analysis by age group.

The age or mean age of patients was presented for fifteen of the unique studies. The studies which captured this data, included participants aged between 48-91 years old. There is clearly a large differential within this age range both within the total sample and within the samples of the

individual studies. Since COPD has mostly been seen to affect people more often beyond retirement age **(BTS/BLF, 2002)** this sample is consistent with the target population.

Gender information was only collected for thirteen of the unique studies. Study samples generally included a higher proportion of females than males. Race and ethnicity data was rarely mentioned in any of the reviewed studies and only one study **(McNaughton et al., 2017)** made clear record of this.

Weight was captured in four studies **(Lichtenschopf et al., 2014, Lord et al., 2010, Lord et al., 2012, Pacheco et al., 2014)** and height and clinical history by one of these **(Pacheco et al., 2014),** though again only the average weight or average height was presented, but no analysis was carried out on this data, so little can be deduced from this.

Several studies captured smoking history **(Grasch, 2015, McNaughton et al., 2017, Thomas et al., 2015)**, or smoking and alcohol history **(Lichtenschopf et al., 2014)** but the total amount of data examined in these studies was limited and was not analysed in regard to current smoking and alcohol usage so again, nothing can be clearly deduced from this information.

Unique identifier (First Author/ Date Published) (Country)	**Publication title**	**Condition**	**Age range**	**Gender**	**N = Total patients evaluable**
Bonilha 2009** (Brazil)	Effects of singing classes on pulmonary function and quality of life of COPD patients	COPD	69.8 ±7.4 73.6 ±7.5	Male/ Female	43
BLF 2017 (UK)	British Lung Foundation Singing Project Report 2015-2017	CRD	Not Known	Male/ Female	80
Canga 2014* *(Canga 2015a*) (US)	Music therapy in the treatment of chronic pulmonary disease (CT.gov registration entry - NCT02146235)	COPD	Not Known	Not Known	Not Known
Canga 2015* (US)	AIR: Advances in Respiration - Music therapy in the treatment of chronic pulmonary disease	COPD	70.1 (48-88)	Male/ Female	68

Clift 2013 (UK)	An evaluation of community singing for people with Chronic Obstructive Pulmonary Disease (COPD)	COPD	69.5 (SD 7.64)	Male/ Female 1:2	126
Clift 2017 (UK)	Findings from the Lambeth and Southwark Singing and COPD Project	COPD	68.64 (SD 9.01)	Male/ Female 8:23	42
Engen 2005 (US)	The Singer's Breath: Implications for Treatment of Persons with Emphysema	COPD	M av. 81 F av. 72	Male/ Female	7
Goodridge 2013* (Canada)	Therapeutic singing as an adjunct for pulmonary rehabilitation participants with COPD: Outcomes of a feasibility study	Advanced COPD	Average age ~70	Male/ Female	14
Grasch 2013 (US)	Daily Singing Practice as a Means of Improving Pulmonary Function and Quality of Life in Emphysema Patients	COPD	69.44 +/- 9.43 (47-84)	Male/ Female (9:16)	25
Herer 2013 (France)	Outcomes of a pulmonary rehabilitation program including singing training	37 COPD + 8 CRDs	60 +/-10	Male/ Female	45
Jamaly 2017	The effect of singing therapy compared to standard physiotherapeutic lung sport in COPD	COPD	63 ± 6	Not Known	22
Lichtenschopf 2014 (Germany)	Singing as a new therapy in treating COPD patients: A pre-study. *[German] (Singen Als erweiterung des therapeutischen spektrums bei der behandlung der COPD)*	COPD	Not Known	Not Known	119

Liu 2016 (China)	Effect of Singing Therapy on Anxiety and Depression in Patients with Chronic Obstructive Pulmonary Disease at Stable Stage in Community	COPD	Not Known	Not Known	56
Lord 2010** (UK)	Singing teaching as a therapy for chronic respiratory disease - a randomised controlled trial and qualitative evaluation	CRD	67.3 (8.1)	Not Known	28
Lord 2010*a* (UK)**	Effect Of Singing Lessons In Patients With COPD A Randomised Controlled Trial [Abstract]	COPD	67.3 (8.1)	Not Known	28
Lord 2010*b* (UK) **	Singing for breathing™ effects of singing lessons in patients with COPD – A randomised control trial [Abstract]	COPD	Not Known	Not Known	28
Lord 2012** (UK)	Singing classes for COPD: A randomised controlled trial	COPD	68.3 (9.7)	Not Known	24
Lord 2012*a* (UK)**	Effects Of "Singing for breathing"™ In Patients With COPD - A Randomized Control Trial [Abstract]	COPD	NK	NK	24
Lord 2012*e* (2014) (UK)**	Erratum to: Singing classes for COPD: A randomized controlled trial <correction of trial number to ISRCTN39714922>	COPD	Not Known	Not Known	Not Known
Martinez 2008 *(Bonilha 2009a)* (Brazil)**	Effects of singing on pulmonary function and quality of life of COPD patients [Abstract]	COPD	M: 69.8 ±7.4 F: 73.6 ±7.5	Male/ Female	Not Known

McNaughton 2014 *(McNaughton 2017a)* (New Zealand)	Sing Your Lungs Out: Effect of participation in weekly community singing group for 1 year, on lung function, anxiety and depression and health care use in patients with COPD who have completed pulmonary rehabilitation: a quantitative and qualitative study' WHO registration entry - ACTRN12615000736549)	COPD	Not Known	Not Known	Not Known
McNaughton 2016 *(McNaughton 2017b)* (New Zealand)	Sing Your Lungs Out: a qualitative study of a community singing group for people with COPD	COPD	51–91	Male/ Female 1:1.3	21
McNaughton 2017 (New Zealand)	Sing Your Lungs Out: a community singing group for chronic obstructive pulmonary disease: a 1-year pilot study	COPD	68.8 (9.8) (51- 91)	Male/ Female 8:13	21
Miyahara 2001 (Japan)**	Benefits of singing training in chronic obstructive pulmonary disease patients	COPD	Not Known	Not Known	20
Morrison 2013 *(Clift 2013a)* (UK)	A UK feasibility study on the value of singing for people with Chronic Obstructive Pulmonary Disease	COPD	69.5 (SD 7.64)	Male/ Female 1:2	106
Pacheco 2014 (Portugal)*	Singing in chronic obstructive pulmonary disease patients: A pilot study in Portugal	COPD	2M= 68 1F=42 1F=50	Male/ Female 1:1	6/8

Thomas 2015 (UK)	'I really live for coming here'. The effect of a long-term singing group on control of breathlessness, social empowerment and psychological wellbeing of patients with respiratory disease: a qualitative study	CRD	72.6 (50–92) (Mean)	Male/ Female 1:3	16
Trivedi 2017 (India)	Effect of Singing along with Pulmonary Rehabilitation on Quality of Life and Dyspnea in Patient with Chronic Obstructive Pulmonary Disease	COPD	54 (Mean) (All <65)	Male/ Female 15:7	56
Skingley 2014 *(Clift 2013b)* (UK)	"Singing for Breathing": Participants' perceptions of a group singing programme for people with COPD	COPD	69.5 (SD 7.64)	Male/ Female 1:2	97
Skingley 2018 *(Clift 2017a)* (UK)	Community singing groups for people with chronic obstructive pulmonary disease: participant perspectives	COPD	Not Known	Male/ Female	37

Table 1 – Systematic Review – Study Demography Details

An *a* or *b* noted after the year of study denotes that the paper is a study registration title, a subsequent abstract title or an additional paper related to the original study and an *e* following the year of study denotes an erratum paper.

NB Those studies included in the Cochrane review are noted with two asterisks (**) and those discussed but excluded from the Cochrane review are noted with one asterisk (*).

Study populations and eligibility

The study populations i.e. sample size, study setting, eligibility and enrolment criteria are identified and presented in summary in Table 2 below. The majority of the information within this table is provided verbatim from the studies and papers.

Unique identifier (Country)	Participant eligibility	Participant population and enrolment details	Study Setting	Randomisation Method/ Control	Total Patient No.
Bonilha 2009 (Brazil)	- COPD diagnosis according to 'GOLD' criteria - Former smokers - Stable clinical conditions for at least two months - Excluded: Patients with severe comorbidities, still smoking, or using oxygen therapy	Patients invited to participate in the study during regular consultations at the University Hospital, or after answering a radio advertisement	Hospital (2008)	Randomisation – method not specified Control – Craft classes	30 n=15 singing group
BLF 2017 (UK)	- Participants experiencing a range of diagnosed lung conditions - There is a variety of diagnoses although the majority had COPD	A total of 32 leaders were trained, 29 groups were set-up with 26 self-sustaining singing groups delivering classes to over 300 people with a lung condition across England, Scotland and Wales. 70/80 enrolled patients evaluated	Community Groups (2016-17)	Pilot study Mixed method, No randomisation or control	70

Canga 2015 (US)	- COPD diagnosis according to the 'GOLD' criteria - Attending pulmonary rehabilitation sessions - Able to attend music therapy sessions once a week for 6 weekly sessions - Medically stable simultaneously allowing them to participate in PR	Patients newly enrolled in PR sessions randomly allocated to treatment or control group. 30 excluded as required number of sessions were not completed (due to time commitment or personal/ medical concerns)	Hospital (2008 - 2013)	Pilot study pulmonary rehabilitation plus musical instrument playing and singing - No randomisation or control	60
Clift 2013 (UK)	- COPD diagnosis defined by the FEV1% following GOLD (2010) and NICE (2010) guidance. - COPD pts with 15% mild, 45% mod% severe and 10% very severe. - 126 patients recruited via all routes, 5 did not attend for baseline assessments. 15/121 (12.3%) did not meet formal COPD inclusion criteria	COPD patients from COPD practices. 106 Pts Mean age 69.5 (SD 7.64) 1:3 (M:F) 75.1% Retired 13.5% due to COPD. 69.5% previous smokers; 11.4% current smokers 99% white, 51.4% continued in education, with > 1/3 holding a degree. 106 enrolled, 35 discontinued (33%) 71 followed up 216 comments from 97	Community Groups (2011-12) Six groups in a community setting with family, carers + supporters	Pilot study Mixed method six groups - No randomisation or control	106

Clift 2017 (UK)	- Participants with breathing difficulties - 31/42 met GOLD criterion and diagnosis of COPD (i.e. FEV1\FVC <0.7) at baseline - Six had mild severity, fifteen moderate, nine severe and one very severe. - 18 were recruited in Sep. and 13 in Jan.	44/ 60 CRD participants 10 males and 34 females recruited with breathing difficulties. 31 COPD patients 8 males and 34 females. Two cohorts from four groups comparable on all baseline measures, combined for the purpose of the evaluation	Community Groups (2015-16)	Pilot study Mixed method – No randomisation or control	44
Engen 2005 (US)	- Physician's diagnosis of emphysema or COPD. - Patient agreement to participate in 10/12 group lessons. - Screened for smoking and respiratory related hospitalisation. - Tested for FEV >35% <75% of predicted values	Emphysema or COPD outpatients in Gerontology clinic and pulmonary rehabilitation clinic	Classroom (2002)	Assigned to small group based on patient schedule preference (day/time) - No randomisation or control	7

Goodridge 2013 (Canada)	- Advanced COPD attending a PR program targeted to address symptoms - Participation in program for ≥ 3 months - Clinically stable - Able to complete ICD and speak/read English Exclusions - concomitant medical conditions significantly limiting exercise tolerance	COPD patients attending a PR program offered through the local health region in Western Canada	Private room at the PR facility following regularly scheduled PR session.	Participants consecutively assigned to the therapeutic singing intervention up to a capacity of 14. Remaining participants continue normal standard of care	14
Grasch 2013 (US)	- COPD or emphysema without any other major concomitant respiratory illnesses	COPD and Emphysema patients participating in pulmonary rehabilitation program	Hospital PR program.	PR study with singing alone daily as add on activity - No randomisation or control	13
Herer 2013 (France)	- COPD (n=37) or other chronic respiratory disorders (n=8)	COPD and CRD	Hospital	Open study - No randomisation or control	45
Jamaly 2017 (Germany)	- COPD FEV1 1.65 ± 0.65 - exacerbation-free	COPD patients Height 172 ± 10 cm, Weight 84 ± 24 kg	Hospital	Single centre Open, prospective, randomized, pilot study	22

Lichtenschopf 2014 (Germany/ Austria)	- COPD (n=92) or other chronic respiratory disorders (n=27)	COPD and CRD Patients requiring oxygen or recorded with global insufficiency were excluded	Rehab. Centre	Pilot study design - No randomisation or control	119
Liu 2016 (China)	- COPD patients	COPD patients	Hospital	Randomisation according to a digit table	56
Lord 2010 (UK)	- COPD diagnosis according to the 'GOLD' criteria at the Royal Brompton Hospital	COPD patients. 183 people approached initially 150 declined (82%) Total n=33 randomised, 8 discontinued (i.e. 24% attrition). n=24 completed - attending at least 8/12 sessions	Hospital	Randomisation - singing or control - block randomization with consecutive sealed envelopes Control - normal standard of care	28 n=15 singing group n=13 control group
Lord 2012 (UK)	- COPD diagnosis according to the 'GOLD' criteria attending respiratory clinics at the Royal Brompton and Harefield NHS Foundation Trust	Invitation to patients attending respiratory clinics at Royal Brompton and Harefield NHS Foundation Trust. Control - group film discussion	Hospital clinic	Randomisation - baseline blocks of 4 via consecutive sequentially numbered sealed envelopes	24 n=13 singing group n=11 control group

McNaughton 2017 (New Zealand)	- COPD or other chronic lung disease diagnosis - Attended 8-week, hospital-based PR programme. - Attending weekly maintenance community PR exercise class	Patients selected by a PR nurse from group attending weekly maintenance community PR exercise class. COPD patients with a wide range of disease severity and comorbidities	Urban community	Mixed methods feasibility study Qualitative description from interview transcripts and a focus group	21
Miyahara 2001 (Japan)	- COPD moderate to severe - FEV1 40+/-15%	COPD patients	Hospital	Randomisation - singing vs. control standard of care	Not Known
Pacheco 2014 (Portugal)	- Stable clinical condition for at least 6 weeks before admission to the study - COPD according to 'GOLD' criteria	COPD Patients attending a maintenance P programme (90 min, 2x week) at Physical Medicine and Rehabilitation Dept. in Hospital	Hospital Clinic	Pilot study - No randomisation or control	6
Thomas 2015 (UK)	- Singing group led by a music therapist and open to all patients with respiratory disease	Total n=16 COPD (11), Asthma (2), Bronchiectasis (2) and Fibrosis (1) All non-smokers (ex-smokers 12); 75% had previously attended PR. 10 lived alone, 8 had a history of mental health comorbidity	Hospital	Pilot study - No randomisation or control	16

Trivedi 2017 (India)	- Patient with recognized COPD according to 'GOLD' criteria - Grade A to C - Age < 65 years - Singing is their hobby. - Patient with other concurrent respiratory disease or associated medical condition, malignant disease or did not consider singing a hobby were excluded	COPD Patients	Hospital Outpatient	Randomised to pulmonary rehabilitation or pulmonary rehabilitation with Singing	56 n=30 singing group

Table 2 – Systematic Review – Study Patient Population Summary

The study papers selected cover the period from 2001 to 2017, and the majority i.e. seventeen of the nineteen studies were conducted within the past ten years.

A total of seven studies specifically mention GOLD staging criteria for COPD, **(Bonilha et al., 2009, Canga et al., 2015, Clift et al., 2013, Clift et al., 2017, Lord et al., 2010, Lord et al., 2012, Pacheco et al., 2014**) but use of the terminology COPD diagnosis or 'stable COPD' was often used more often, without any specific criteria being mentioned. There is little evidence to indicate if the criterion was checked individually for each patient within the studies either, which is a significant gap in the quality of the data.

Studies were frequently recruited using patients from pulmonary rehabilitation (PR) groups or recruited through pulmonary clinics **(Canga et al., 2015, McNaughton et al., 2017)** so COPD diagnosis may have been assumed. However, pulmonary rehabilitation is often offered in some countries to patients that are not necessarily diagnosed with COPD i.e. PR can be offered to patients considered functionally disabled by breathlessness (MRC score $\geq$ 3), patients with MRC score $\geq$ 2 who are symptomatic and disabled by their condition, patients with other chronic progressive lung conditions (e.g. bronchiectasis, interstitial lung disease, chronic asthma and chest wall disease and also patients pre and post thoracic surgery including lung transplant) as

well as patients with a confirmed diagnosis of COPD **(NHS UK, 2011)**. Hence, the expectation of meeting GOLD criteria should not just be assumed. It is apparent that even studies which mention GOLD criteria as being part of their inclusion may not have tested patients for this criterion specifically prior to enrolment.

Study Interventions

The study interventions i.e. the details of the singing intervention for each study were identified and are presented in summary in Table 3 below.

Unique identifier	**Duration of study and singing intervention details**	**Control interventions**
Bonilha 2009	24 weeks - one hour per week Classes for singing practice	Craft classes
BLF 2017	12 weeks (3 months) – one hour per week Singing for Lung Health sessions led by a local singing leader who has taken part in the BLF singing programme. Intervention includes physical warm-ups, breathing exercises and vocal exercises such as rhythm and pitch games. Songs were selected as suitable for people with a lung condition, as well as being fun. Leaders were trained to focus on developing breathing techniques to support participants in reducing breathlessness and preserve optimal lung function, to develop awareness of postural and breathing patterns, extend outbreaths through sung phrases, to build physical and vocal stamina and have fun. Emphasis is on the importance of fostering a positive social environment for participants and allowing them to express themselves creatively; whilst improving self-confidence, quality of life and counteracting feelings of stress and anxiety	None
Canga 2015	6 weeks - 45 minutes per week Music therapy session including live music visualizations, wind instrument playing with clinical improvisation and singing	None

Clift 2013	36 weeks (9 months) - one hour per week Exercises and singing with 30 minutes of socialising. Singing in classes with no prescribed practice at home. Singing session commenced with 20 minutes of relaxation, posture, breathing and vocal exercises followed by 40 minutes singing - familiar and new songs taught by ear. Two combined group workshops and two performance events	None
Clift 2017	38-40 weeks (10 months) - one hour per week Exercises and singing with 30 minutes of socialising/administration. Singing in classes with no prescribed practice at home. Singing sessions included relaxation, posture, breathing and vocal exercises followed by the singing of songs	None
Engen 2005	6 weeks - 45 minutes twice per week Twelve instructional sessions with nine weeks study time intervention sessions. Two weeks no-contact period prior to a final follow-up visit	None
Goodridge 2013	8 weeks - one hour per week Pulmonary rehabilitation program involving eight weeks of classroom instruction as well as structured exercise classes prior to singing	Normal care
Grasch 2013	12 weeks - 10 minutes per day (70 min per week) Singing pamphlet guide subjects through a series of breathing and vocal warm-ups. Upon completion, they follow with singing a song of their choice. Subjects were instructed to practice singing five minutes each morning and evening	None
Herer 2013	3 weeks - one hour per week Respiratory education through singing by two professional musicians (instrumentalists and choirmaster). Twenty minutes included breathing exercises consisting in particular of short inspirations deep and prolonged expirations. A dozen minutes included a vocal warm-up especially by vocalizations. Final part of the session was singing popular songs with musical accompaniment	None

Jamaly 2017	4 weeks - 8 course units of 1.5 hours Therapeutic Singing (TS) - a therapy with breathing exercises and techniques	Physiotherapeutic lung exercise
Liu 2016	24 weeks – once per week Singing therapy	Health Education + Mental Nursing
Lichtenschopf 2014	2 weeks - one hour twice per week Total of four sessions for about one hour with singing group meeting four times (1.5 hours each time). Include COPD assessed by GOLD II and III and who completed inpatient rehabilitation for three to four weeks, plus oral interview	None
Lord 2010	6 weeks – one hour twice per week All subjects received a thirty-minute standard session on breathing control and techniques to manage breathlessness, delivered by one of two senior respiratory physiotherapists involved in the study. ‘Pursed-lip’ breathing and nose breathing were also discussed in relation to managing episodes of shortness of breath. Subjects received a booklet and advised to practice the techniques at home	Normal standard of care
Lord 2012	8 weeks - one hour twice per week All subjects received a thirty-minute standard session on breathing control and techniques to manage breathlessness, delivered by one of three senior respiratory physiotherapists involved in the study. ‘Pursed-lip’ breathing and nose breathing were also discussed in relation to managing episodes of shortness of breath. Subjects received a booklet and advised to practice the techniques at home	Film discussion group – once per week
McNaughton 2017	6–9 months – one hour per week Singing group started with a 5 minute warm-up session, the group led by a musician, chose the singing option from a mix of genres with attention to the group’s voice range and capacity for phrase lengths. Musician also discussed breathing for singing techniques as the year progressed and as the group gained confidence. No music reading ability required. CD of songs provided to allow practice at home	None

Miyahara 2001	8 weeks – 5 times per week (duration of sessions unavailable) Japanese 'Shingin' singing programme (requiring slow deep breaths in between singing). Individualized intensity of training based on patient achieving a dyspnoea-score of 3-5 (on Borg scale 0-10)	Normal standard of care
Pacheco 2014	10 weeks - one hour per week Classes were coordinated by a singing teacher and a physiotherapist including relaxation exercises of neck and upper and lower limb muscles, conducted by a physiotherapist (15 min), vocalization exercises, led by a singing teacher, as a preparation for singing (10 min) and singing training for popular Portuguese songs, conducted by a singing teacher (35 min)	None
Thomas 2015	88 weeks - one hour per week Singing group led by a music therapist and open to all patients with respiratory disease. Duration of singing group attendance 15.3 ± 6.5 months	None
Trivedi 2017	4 weeks - one hour twice per week One class of singing included pre-singing session and singing practice. Pre-singing session included local relaxation of respiratory muscles (5 minutes), singing related exercises (slow and fast inspiration and expiration practice, vocalization exercises by loud and rhythmic pronunciation of vowels for 5-10 minutes) and demonstration of song and explanation of singing technique (5 minutes). Singing practice was done for 45 minutes with one intermittent rest of 5 minutes. Singing teacher was instructed to choose Indian classical Songs which have positive and inspirational lyrics. (Singing in addition to 45 minutes of PR four days a week for four weeks)	Pulmonary Rehabilitation provided to both groups (i.e. experimental and control) with dose tailored by patient according to AACVPR guidelines

Table 3 – Systematic Review - Study Intervention Summary

Duration, Frequency and Design

There were two key approaches taken in regard to study method which were either controlled studies or single group observational studies. There were eight randomised or quasi studies

with a control group **(Bonilha et al., 2009, Goodridge et al., 2013, Jamaly et al., 2017, Liu et al., 2016, Lord et al., 2010, Lord et al., 2012, Miyahara et al., 2001, Trivedi, 2017)** and eleven feasibility, pilot or observational studies **(BLF, 2017, Canga et al., 2015, Clift et al., 2013, Clift et al., 2017, Engen, 2005, Grasch, 2013, Herer, 2013, Lichtenschopf et al., 2014, McNaughton et al., 2017, Pacheco et al., 2014, Thomas et al., 2015)**.

The frequency of the singing interventions, for studies where this information was explicitly presented, was most commonly delivered in one hourly sessions within a group setting either once per week **(Bonilha et al., 2009, BLF, 2017, Clift et al., 2013, Clift et al., 2017, Goodridge et al., 2013, Herer, 2013, Liu et al., 2016, McNaughton et al., 2017, Pacheco et al., 2014, Thomas et al., 2015)** or twice per week **(Lichtenschopf et al., 2014, Lord et al., 2010, Lord et al., 2012, Trivedi, 2017)**. Ten of the studies included patients having one session per week and four studies presented data for patients attending two sessions per week.

Five other studies appeared to have a more novel or unique approach. In the first of these **(Grasch, 2013)**, patients completed a five-minute series of breathing and vocal warm-ups followed with singing a song morning and evening. Another **(Miyahara et al., 2001)** required patients to attend hourly group singing sessions five times per week and one **(Jamaly et al., 2017)** was set up as eight course units of therapeutic singing (breathing exercises and techniques) each one and half hours long and delivered over four weeks.

One study paper **(Canga et al., 2015)** described weekly music therapy sessions lasting forty-five minutes but these included live music visualizations, wind instrument playing and clinical improvisation, in addition to the target intervention of singing. This was conducted over a six-week duration hence there are serious confounders limiting inclusion of results within this review more fully. It was decided that this study could be included for completeness in regard providing valuable information regarding the design and measures used. There is relatively little statistical data reported from the study, so it is considered that the impact of including this would be of negligible impact to the key messages or conclusions.

The singing sessions for one of the studies **(Engen, 2005)** were documented to be twice per week but were conducted for forty-five minutes instead of one hour and results of the ANOVAs for breath management (extent of counting) and breath support (intensity of speech) for this study were significant ($p < 0.038$ & $p < 0.000$ respectively). Descriptive analyses of this study also showed a clear shift in breathing mode from clavicular to diaphragmatic breathing that was maintained two weeks after the treatment period. The overall length of study period i.e. period for which the singing or control interventions were conducted for participants, ranged from two weeks to thirty-six weeks.

There were eight randomised or quasi studies with a control group **(Bonilha et al., 2009, Goodridge et al., 2013, Jamaly et al., 2017, Liu et al., 2016, Lord et al., 2010, Lord et al., 2012, Miyahara et al., 2001, Trivedi, 2017)**, but the type of control activity was quite varied

between the studies identified, ranging from normal standard of care or treatment as usual **(Goodridge et al., 2013, Lord et al., 2010, Miyahara et al., 2001)**, physiotherapy lung exercises **(Jamaly et al., 2017),** Health Education/Mental Nursing **(Liu et al., 2016)** film discussion groups **(Lord et al., 2012)** to craft classes **(Bonilha et al., 2009)**. One study **(Trivedi, 2017)** included PR as a control but the singing intervention also included PR in addition to the singing intervention.

Several studies also enrolled patients directly from a pulmonary rehabilitation (PR) group (e.g. **Goodridge et al., 2013, Canga et al., 2015, McNaughton et al., 2017, Pacheco et al., 2014** etc.) and this does potentially manage some confounding factors and allows patients to start from a relatively similar educational perspective or baseline in regard to their respiratory understanding, though it may also lead to its own confounders depending on whether the PR continues throughout the study or ceases prior to the start. This was certainly the case for two studies **(Canga et al., 2015, Trivedi, 2017)** where the PR continues. This is not specifically mentioned for other studies, so it could not be confirmed either way.

The intervention for one of these studies **(Canga et al., 2015)** did not purely involve singing and PR either, it included additional aspects i.e. exercises with musical instruments in parallel with singing intervention and PR, which are significant confounding factors regarding outcome data in respect to expressing the outcomes within this systematic review.

One other limitation that should be noted is that none of the studies appeared to have recorded previous, recent or parallel singing activities of participants either alone or in groups, which could potentially be another confounding factor in the results of the studies included.

There are a number of studies which have confounding factors, limitations or concerns in regard unclear information. However, for this broader expanded systematic review, the study design, conduct and methodology for all these studies are still valuable. All related studies are included and discussed so that an understanding of the overall demographic of the work conducted to date and the various study designs, measures and implementations can be examined. The actual outcome data and conclusions would have significant limitations though and so these components are not explored further for this reason. The results of the more robustly designed studies are examined collectively and published in the Cochrane review ‘Singing for Adults with COPD’ **(McNamara et al., 2017)** to which the present author contributed.

Power calculation and sample size rationale

The information regarding power calculations and rationale for the size of the samples were limited. One study **(Bonilha et al., 2009)** had no prospective power calculation but reference was made to a previous study which showed that muscle training with 32 patients with COPD led to significant changes in respiratory pressures. This was used as justification on final number of 15 patients to be included in each arm of the trial.

One study **(Clift et al., 2013)** provided an explanation that if a minimum of fifty estimated participants were followed up at the end of the study it would provide appropriate information on feasibility and potential effects. A conservative approach was estimated for retention of 50% of subjects such that the aim would be to recruit one hundred participants to the study to accommodate this retention rate. The follow-up study **(Clift et al., 2017)** aimed to recruit this number of patients and further referenced power calculations in its recommendations, suggesting that a sufficiently powered trial may well have required finding over two hundred people with COPD at the outset to take into account estimated attrition. The authors indicated that future multi-centre randomised studies sufficiently powered and capable of assessing the effects of the intervention might require recruitment of five hundred to a thousand participants to achieve such enrolment numbers for seasonal variations and background pollution levels to be taken into account.

A further study **(Grasch, 2013)** referenced the significant change from pre-study to post-study on the quality of life assessment (SGRQ) from a previous study **(Bonilha et al., 2009)**, with effect size of 1.02, as being the key factor in calculating the sample size. It was calculated that n=30 would give a statistical power in excess of 0.90 for this proposed pilot study to find a statistically significant difference, with alpha=0.05, on the SGRQ. The sample size dropped to 25 patients and this was still felt to be adequate power to detect an effect size of $\geq$0.59.

However, the paper presenting the study referenced **(Bonilha et al., 2009)** expresses concerns regarding limitations of this study, including small statistical power for some of the comparisons, mainly due to excessive variation in the results. The authors comment that enrolment of a greater number of subjects is needed to assess whether significant changes occur in key parameters.

One other study **(Canga et al., 2015)** noted in their discussion that they would recommend future studies consider effectiveness of a lengthened duration of intervention period and suggested that a larger sample would increase statistical power. However initially there was no detail provided as to how the original sample size was calculated.

It was not clear from any of the other studies if there had been a clear discussion or decision on how study samples had been calculated. This rationale has not been provided for the majority of studies nor has an indication of whether this was considered.

Study Measures

The studies selected in this review included measurements of over seventy different parameters, ranging from demography and physical outcomes to depression and anxiety. Many also included qualitative data from participant feedback and interviews. While this does illustrate

well the wide diversity of options and data that can be collected when researching this area, unfortunately the data captured were largely inconsistent throughout the designs and research groups, so comparisons of results between studies or robust meta-analysis could not be executed particularly effectively. However, there is still interesting content for examination and collective discussion.

The measures used by each study are extracted and analysed in a tabular format as presented in Table 4 below and then further examined.

Measures	General theme	Total	Bonilha 2009	BLF 2017	Canga 2015	Clift 2013	Clift 2017	Engen 2005	Goodridge 2013	Grasch 2013	Herer 2013	Jamaly 2017	Lichtenschopf 2014	Liu 2016	Lord 2010	Lord 2012	McNaughton 2017	Miyahara 2001	Pacheco 2014	Thomas 2015	Trivedi 2017
Demographics: Gender	Demography	13	x		x	x	x	x	X	x	x		x				x		x	x	x
Demographics: Age	Demography	15	x		x	x	x	x	X	x	x	x			x	x	x		x	x	x
Demographics: Height	Demography	3				x						x							x		
Demographics: Weight	Demography	6				x						x	x		x	x			x		
Demographics: Body Mass Index (BMI) derived	Demography	1					x														
Demographics: Clinical history	Demography	2					x												x		
Demographics: Ethnicity	Demography	1															x				
Demographics: Smoking history	Demography	5		x						x			x				x			x	
Demographics: Alcohol	Demography	1											x								
Demographics: Adherence to singing	Demography	1								x											
Demographics: Medication changes during the study.	Demography	1								x											
Demographics: Social Measures (Income, Housing, Education)	Demography	1					x														
Demographics: Marital Status	Demography	1							X												
FVC	Spirometry	5	x			x	x			x									x		
FEV1	Spirometry	10	x			x	x	x	X	x	x	x					x		x		
FEV1/FVC	Spirometry	4	x				x			x									x		
FVC%.	Spirometry	4	x			x	x												x		
IC	Spirometry	1	x																		
ERV	Spirometry	1	x																		
PImax (cmH2O) (%)	Spirometry	1	x																		
PEmax (cmH2O) (%)	Spirometry	1	x																		

Maximal respiratory (inspiratory and expiratory) pressure measurements mouth level (PImax, PEmax)	Spirometry	4	x				x				x								x		
FEV1%	Spirometry	7				x	x		X	x					x	x	x				
FEV%	Spirometry	2	x			x															
TLC	Spirometry	3								x		x							x		
RV	Spirometry	3								x		x							x		
VO2max	Spirometry	1									x										
Breath management (extent of counting or single breath counting)	Breathing	3						x							x	x					
Breath hold time	Breathing	2													x	x					
Breath support (intensity of speech)	Breathing	1						x													
Breathing mode from clavicular to diaphragmatic breathing	Breathing	1						x													
O2 Saturation% (SpO2%)	Physical	1					x														
O2 Saturation recovery (s)	Physical	3							X						x	x					
Heart Rate recovery (s)	Physical	2													x	x					
Subjective recovery (s)	Physical	2													x	x					
Blood Pressure (Systolic /Diastolic)	Physical	1					x														
Arterial blood gases breathing room air;	Physical	1	x																		
Heart rate post walk	Physical	5					x		X						x	x			x		
Distance walked	Activity	1						x													
Six-Minute Walk Test (6MWT) min	Activity	5					x		X		x						x		x		
Incremental Shuttle Walk Test (ISWT) min	Activity	2													x	x					
Incremental shuttle Walk Test (ISWT) with recovery rates for heart rate	Activity	1														x					
Shuttle walk distance	Activity	3						x							x	x					

Steps per day	Activity	1														x					
Sedentary time (minutes per day)	Activity	1														x					
Physical activity duration (PAD) (minutes per day)	Activity	1														x					
Activity entry expenditure (AEE) (Kj per day)	Activity	1														x					
Routine exercise	Activity	1								x											
Physical activity was measured using SenseWear armbands pre/post intervention. Subjects were given a SenseWear Pro (SenseWear, Body Media, Pittsburgh, USA) activity monitor for a week prior to commencing sessions	Activity	1														x					
Exercise recorded (all activity)	Activity	7						x	X	x	x				x	x			x		
MRC breathlessness/ Dyspnoea scale	Dyspnoea	7		x		x	x		X		x								x		x
BDI evaluations of dyspnoea (Basal Dyspnoea Index)	Dyspnoea	2	x		x																
Borg dyspnoea score	Dyspnoea	6	x				x	x							x	x			x		
University of Cincinnati Dyspnoea Questionnaire (UCDQ)	Dyspnoea	1									x										
Chronic Respiratory Disease Questionnaire (CRQ)	Respiratory	3			x											x		x			
COPD Assessment Test Score (CAT)	Respiratory	4		x								x				x			x		
St George's Respiratory Questionnaire QoL (SGRQ)	Respiratory	8	x			x	x		X	x		x			x				x		

Brief Symptom Inventory-x8 Global Severity Index	Respiratory	1								x											
London Chest Activity of Daily Living Scale (LCADL)	Respiratory	1																	x		
Visual Simplified Respiratory Questionnaire (VSRQ)	Respiratory	1									x										
Clinical COPD Questionnaire (CCQ)	Respiratory	1															x				
Respiratory illness occurred during study.	Respiratory	1								x											
EQ-5D EuroQoL Test;	Health	4		x		x	x												x		
Short Form Health Survey (SF-36) or York SF-12	Health	3				x									x	x					
HAD Anxiety Score	Health	6											x	x	x	x		x		x	
HAD Depression Score	Health	5											x	x	x	x				x	
Beck Depression Inventory	Health	1			x																
DUKE physical health subscale	Health	1						x													
General Anxiety Disorder-7 (GAD-7)	Health	1		x																	
Measures of functional outcomes	Health	1						x													
Self-reported global health rating (10-point scale)	Health	1							x												
Brief Illness Perceptions Questionnaire (BIPQ)	Health	1							x												
Brief Symptom Inventory-18 Global Severity Index	Health	1								x											
A post intervention written evaluation survey (acceptability and enjoyment)	Survey	1							x												

Questionnaire - CareFusion, Kent, UK	Survey	1												x							
Specially prepared questionnaire	Survey	2						x				x									
Service Use (Healthcare Utilisation)	Survey	2					x													x	
Hospital admission days for acute exacerbation of COPD (AECOPD)	Survey	1														x					
Patient Activation Measure (PAM)	Survey	1		x																	
Principal themes expressed from participant feedback	Written text	3				x	x													x	
Interviews, Journals and Participant Feedback	Written text	10		x		x	x	x	x					x	x	x		x	x		
Evaluation of adverse event occurrence while attending the singing programme	Written text	1							x												

Table 4 – Systematic Review – Study Measures Summary

A full explanation or description of the types of measures used, along with general findings from the various different measures and tools employed within the studies are grouped into categories for summarisation as follows:

Physiological Measures

Lung Function Measures

Lung function or pulmonary health measures are most commonly performed in these studies using validated spirometer readings. This measures the amount and speed with which air can move in and out of the lungs. The following different spirometry (or pulmonary) measurements were captured in more than one of the studies identified:

Forced Vital Capacity (FVC)

This is the amount of air exhaled with force relative to population normal values, after inhaling as deeply as possible. Five studies **(Bonilha et al., 2009, Clift et al., 2013, Clift et al., 2017, Grasch, 2013, Pacheco et al., 2014)** collected readings of this measure and a small, though statistically significant reduction in FVC were shown only in one study **(Clift et al., 2013)**. No significant effects were seen in any of the other studies.

Percentage Forced Vital Capacity (FVC%)

This calculation is the % air exhaled with force after inhaling as deeply as possible. Four studies **(Bonilha et al., 2009, Clift et al., 2013 Clift et al., 2017, Pacheco et al., 2014)** collected and presented data for this endpoint, though improvements in FVC% were only shown by the same study **(Clift et al., 2013)** that showed improvement in FVC.

Forced Expiratory Volume (FEV)

This is the amount of air exhaled with force in one out-breath. The amount of air exhaled in one out-breath may be measured at one second (FEV1), two second (FEV2), or three second (FEV3) intervals. (FEV1 divided by FVC can also be determined). Ten studies **(Bonilha et al., 2009, Clift et al., 2013, Clift et al., 2017, Engen, 2005, Goodridge et al., 2013, Grasch, 2013, Herer, 2013, Jamaly et al., 2017, McNaughton et al., 2017, Pacheco et al., 2014)** described this measure. In some cases, this value was only noted as being captured at baseline to support inclusion criteria or the analysis of other measures and no further data was reported post intervention **(e.g. McNaughton et al., 2017)**. Only one study **(Clift et al., 2013)** appeared to show some improvements in this measure post intervention and another **(Pacheco et al., 2014)** observed that the FEV1 improved from 1.15 L (45%) to 1.21 L (47%) for one of four patients though this was not significant.

Percentage Forced Expiratory Volume (FEV%, FEV1%)

This is a calculated value of the % air exhaled with force relative to population normal values at intervals as above. Eight studies **(Clift et al., 2013**, **Clift et al., 2017, Goodridge et al., 2013, Grasch, 2013, Jamaly et al., 2017, Lord et al., 2010, Lord et al., 2012, and McNaughton et al., 2017)** presented FEV1%, and two studies **(Bonilha et al., 2009, Clift et al., 2013)** presented FEV% results. Interestingly there appears to be only one study **(Clift et al., 2013)** which showed improvement for FEV1% but there were no other significant findings from any of the other studies.

Calculated Ratio FEV1/FVC

This ratio (also called Tiffeneau-Pinelli index) represents the proportion of FVC that a patient is able to expire in the first second of forced expiration. Four studies calculated and referred to this ratio **(Bonilha et al., 2009, Clift et al., 2017, Grasch, 2013, Pacheco et al., 2014)** but there were no findings referenced except by one study **(Clift et al., 2017)** where FEV1 /FVC shows a significant improvement, but the research team note that this likely reflects the decrease in FVC within this sample.

Maximal Inspiratory Pressure (MIP or PImax)

Maximal Inspiratory Pressure (MIP or PImax) reflects the strength of the diaphragm and other inspiratory muscles **(Polkey et al., 1995)** (An alternative test of inspiratory muscle strength is maximal sniff nasal inspiratory pressure (SNIP) but this was not seen in any of the studies selected). Maximal inspiratory pressures at the mouth level (MIP or MIP%) were measured for four studies **(Bonilha et al., 2009, Clift et al., 2017, Herer, 2013, Pacheco et al., 2014)**. In one very small study **(Herer, 2013)** there was seen to be a principal significant variation observed for PImax between baseline and the end of the intervention (11W variation, +14.7%, p = 0.001), though this study included singing as part of a pulmonary rehabilitation programme confounding the overall finding. No significant changes were noted for any of the other studies where this measure was presented.

Maximal Expiratory Pressure (MEP or PEmax)

This reflects the strength of abdominal and other expiratory muscles. Maximal expiratory pressures at the mouth level (MEP or MEP%) were measured for four studies **(Bonilha et al., 2009, Herer, 2013, Pacheco et al., 2014)**. This measure provided the most important functional finding for one study **(Pacheco et al., 2014)** where the positive influence of singing exhibited an increase of 3 cm H^2O in mean PEmax at the end of the intervention, while the control group showed a decrease of 11.3 cm H_2O. One other study **(Bonilha et al., 2009)** noted that while the control group showed a clear decline in maximal expiratory pressure, the singing group showed a small improvement, and the difference was statistically significant. It also noted that the improvement of PEmax associated with singing could also contribute to better coughing.

General Spirometry Measures

A further ten to fifteen additional lung function or inspiratory measures were mentioned, recorded or calculated throughout all the selected studies. It is anticipated that some were similar or the same despite differing terminology e.g. The generic term spirometry was noted in place of explicit detailed spirometry measure readings being captured or presented i.e. FEV etc. which would usually be expected as part of a spirometry test.

There were two studies **(Bonilha et al., 2009, Pacheco et al., 2014)** in particular which included almost the full range of spirometry readings plus several other different function tests or calculations in addition, including: Expiratory Reserve Volume (ERV), which is the maximal volume of air that can be exhaled from the end-expiratory position; Tidal Volume (TV), the volume of air moved into or out of the lungs during quiet breathing; Inspiratory capacity (IC) which is the sum of ERV and TC **(Bonilha et al., 2009**); Total lung capacity (TLC) the volume in the lungs at maximal inflation, Residual volume (RV), the volume of air remaining in the lungs after a maximal exhalation and finally Total lung capacity (TLC) which is the volume in the lungs at maximal inflation and the sum of VC and RV) **(Pacheco et al., 2014)**. One additional study **(Jamaly et al., 2017)** recorded these two values and showed that there was a significant ($p = 0.002$) hyperinflation decrease in a therapeutic singing experimental group i.e. RV%/TLC 58 ± 12.3% to 51 ± 14.5%. However, the quality of this study is low due to the lack of published data currently available.

There seems to be no significant findings for the majority of these other lung function tests although it was noted in one randomised controlled trial that a session of singing was associated with distinct effects on ERV and IC, detected 2 minutes after its interruption. The control group showed an increase of ERV and a decrease of IC but the singing group had opposite outcomes **(Bonilha et al., 2009)**. The measurements that were utilised most commonly i.e. > 2 studies, were FVC (4 studies), FEV1 (7 studies) and FEV1/FVC (3 studies). There was only one study **(Clift et al., 2013)** which specifically showed significant improvement from singing alone in FEV1%, FVC and FVC% results.

Physical Measures (Non-Lung function)

There were more than twenty different types of physical and activity measures recorded, across all included studies. These were related to heart rate, exercise, activity etc. The measures that were utilised most commonly were the Six-Minute Walk Test (6MWT), incremental shuttle walk test (ISWT) and these are outlined in more detail below.

Six-Minute Walk Test (6MWT)

The 6MWT is used as a measure of functional status in patients with a wide variety of diseases, but it has been particularly helpful for determining prognosis and response to treatment in patients with pulmonary hypertension, congestive heart failure, and COPD. It also correlates with maximum oxygen uptake in patients with pulmonary hypertension, congestive heart failure, and COPD **(Wise and Brown, 2005)**.

This measure was utilised by five studies **(Clift et al., 2017, Goodridge et al., 2013, Herer, 2013, Pacheco et al., 2014, McNaughton et al., 2017)** without any valuable findings from two of these **(Goodridge et al., 2013, Pacheco et al., 2014)** but significant variations observed in one very small study **(Herer, 2013)** where there was a positive variation of +13.8% which was of

significance p = 0.006 and another **(McNaughton et al., 2017)** showed strong evidence for an increase in 6MWT after 4 months, with a mean increase of 28 (95%CI 5 to 52) m, p = 0.019, increasing further to 65 (95% CI 35 to 99) m, p < 0.001 at 1 year. However, this outcome was confounded by the fact that the intervention included participant engagement in physical exercise.

One further study **(Clift et al., 2017)** showed a reduction from 312.7 to 296.6 metres in the distance covered in the Six-Minute Walk Test (6MWT) by participants with COPD, but the change was not statistically significant, and the evaluations were plagued with unusually high temperatures on the days of post-test assessments, impacting exercise evaluation.

Incremental Shuttle Walking Test (ISWT)

The Incremental shuttle walk test (ISWT) is derived from field tests of maximum exercise capacity. The shuttle walk test (SWT) has been less extensively studied in COPD than the 6MWT but has been demonstrated to correlate with Maximal Oxygen Uptake (VO_2Max) and in particular patients with COPD have a strong correlation of SWT with VO_2Max **(Wise and Brown, 2005)**.

This test was employed by just two studies **(Lord et al., 2010, Lord et al., 2012)** with time to recovery of oxygen saturation, Borg Dyspnoea score and heart rate following the walk but there appeared to be no discernible change over time in walking. One of these studies **(Lord et al., 2010)** recorded no differences between the groups for single breath counting, incremental shuttle walking test (ISWT) score or recovery time following ISWT.

General Exercise Measurements

A total of seven of the studies **(Engen, 2005, Goodridge et al., 2013, Grasch, 2013, Herer, 2013, Lord et al., 2010, Lord et al., 2012, Pacheco et al., 2014)** included at least one exercise measure of some kind (including either one of the afore mentioned walk tests, accumulative steps per day, distance walked, routine exercise etc.). One study in particular **(Lord et al., 2012)** noted many different and detailed physical measures in addition due to the use of 'Activity Monitors' discussed further in the next section. There seems to be no clear message however from any of the studies individually or collectively in regard to exercise in response to the intervention.

Activity Monitors

Physical activity was measured using 'SenseWear©' armbands by just one study **(Lord et al., 2012)** which is a novel approach in this area of research. This study had a strong focus on patient activity, and in addition to other more standard exercise measures there was a detailed analysis of a variety of different monitored readings i.e. Steps per day, Sedentary time (min per

day), Physical Activity Duration (PAD) min per day and Activity Energy Expenditure (AEE) Kj per day with measures taken prior to and post intervention.

Subjects were given a SenseWear© Pro (Sense-Wear, Body Media, Pittsburgh, USA) activity monitor to wear for one week prior to commencing their sessions and given written instructions on its usage and cleaning **(Lord et al., 2012)**. It would be interesting to explore further the experiences of using such tools in this population as there is no indication of patient feedback on using this tool, the reliability or quality of the data or any measures of compliance.

This study **(Lord et al., 2012)** also utilised two measures to assess control of breathing, which were in routine use in the physiotherapy department for the assessment of hyperventilation. All members of the singing group reported being more aware of their breathing and how to control it more effectively but the difference in perceived improvements between the two groups was not accompanied by differences in measures of breathing control, functional exercise capacity or daily physical activity.

Oxygen uptake (VO_2), Maximal Oxygen Uptake (VO_2Max), Oxygen Saturation (SaO_2) Saturation of Peripheral Oxygen (SpO2) and Saturation recovery (s)

Pulse oximetry is a technology used for patients with COPD and other conditions to measure the oxygen level in the blood and the heart rate and can rapidly detect changes in blood oxygen level. Six of the studies **(Bonilha et al., 2009, Clift et al., 2017, Herer, 2013, Goodridge et al., 2013, Lord et al., 2010, Lord et al., 2012)** utilised various different oxygen readings and these are grouped together for simplicity.

Maximal oxygen uptake (VO_2Max) is the measurement of the maximum amount of oxygen that an individual can utilize during maximal exercise, measured such that it accounts for their weight i.e. ml/kg/min and oxygen saturation (SaO_2) measures the percentage of hemoglobin binding sites in the bloodstream occupied by oxygen. Saturation of Peripheral Oxygen (SpO_2) is an estimation of the oxygen saturation level and it is an indirect measurement of the oxygen content of blood using oximetry. SaO_2 is a direct measurement of the oxygen content of the blood by arterial blood gas sampling. In patients with COPD, the maximal oxygen uptake (VO_2) measured at peak exercise and the six-minute walk distance (6MWD) has also been associated with survival rates, 6MWD is as good predictor of mortality as the peak VO_2 in patients with COPD **(Cote et al., 2007)**.

Three of the studies **(Goodridge et al., 2013, Lord et al., 2010, Lord et al., 2012)** recorded time to recovery of oxygen saturation following ISWT but recovery time for oxygen saturation did not improve significantly for any of these. Another study **(Clift et al., 2017)** undertook oxygen saturation assessments only as a precautionary measure to ensure that values were in the normal range and did not use these as an outcome measure.

Similar to the results noted for 6MWT, the same small study **(Herer, 2013)** observed principal significant variations with VO_2Max to with a variance of +8.3% (p = 0.01) and one study **(Bonilha et al., 2009)** captured arterial blood gases while breathing room air and showed a significantly higher arterial oxygen saturation (SaO_2) during the act of singing.

Heart Rate (HR) and HR Variability (HRV)

Two studies **(Lord et al., 2010, Lord et al., 2012)** recorded Heart Rate (HR) and HR recovery, which is defined as the difference in heart rate between peak exercise and 1 minute later in seconds as well as subjective recovery post exercise, however neither of these appeared to improve. There were also three studies **(Clift et al., 2017, Goodridge et al., 2013, Pacheco et al., 2014)** which recorded Heart Rate following walk, but there were no significant findings. None of the studies selected captured HRV.

Patient Reported Outcome Measures

Dyspnoea Measures

The main goal of rehabilitation in chronic respiratory disease is to improve dyspnoea. Hence, quantifying dyspnoea is very important and a large number of scales are available to classify and characterise this symptom specifically. There are clinical scales (e.g. MRC or BDI/TDI) in which information is obtained directly from the patients via questioning and there are also psychophysical scales (e.g. Borg scale, VAS). These assess symptom intensity in response to a specific stimulus e.g. exercise **(Crisafulli and Clini, 2010)**.

Twelve of the studies included at least one form of dyspnoea questionnaire. Seven studies **(BLF, 2017, Clift et al., 2013, Clift et al., 2017, Pacheco et al., 2014, Goodridge et al., 2013, Herer, 2013**, **Trivedi, 2017)** utilised the 'MRC breathlessness/ Dyspnoea scale' or a modified version of the MRC. Six studies **(Bonilha et al., 2009, Clift et al., 2017, Engen, 2005, Pacheco et al., 2014 Lord et al., 2010, Lord et al., 2012)** utilised the 'Borg dyspnoea score' or a modified version of this scale. Two further studies **(Bonilha et al., 2009, Canga et al., 2015)** utilised 'Evaluations of dyspnoea (basal dyspnoea index (BDI))' and one study utilised the University of Cincinnati Dyspnoea Questionnaire (UCDQ) **(Herer, 2013)**. The details of these findings are further outlined below.

MRC Breathlessness/Dyspnoea Scale

The MRC breathlessness scale comprises five statements that describe almost the entire range of respiratory disability. The MRC breathlessness scale does not quantify breathlessness itself as with the Borg scale or visual analogue scales, but it quantifies the exercise limitation associated with breathlessness **(Stenton, 2008)**.

In has been indicated that the MRC score correlates well with the results of other breathlessness scales and lung function measurements **(Mahler and Wells, 1988)**. It also aligns closely with direct measures of disability such as walking distance **(Meek et al., 1999)**. The main disadvantage of the MRC measure over other more complex scales is its relative insensitivity to change **(Stenton, 2008)**. It has been in use for more than 50 years and has the important benefit of not being subject to copyright, making it easily accessible for clinical research work and thus more widely used.

Seven of the studies utilised this scale. Two observed significant variations from the initial value. One **(Herer, 2013)** observed 2.3 +/- 0.6 with % variation -21.7% (significance $p < 0.01$ and the other **(Trivedi, 2017)** observed a more significant change ($p = 0.001$) in the control group than in the experimental group ($p = 0.47$) suggesting an improvement in dyspnoea due to the singing intervention, but there were no significant changes found on the MRC scale by the other five feasibility studies **(BLF, 2017, Pacheco et al., 2014, Goodridge et al., 2013, Clift et al., 2013, Clift et al., 2017)**.

Borg Dyspnoea Scale

It was demonstrated that the Modified Borg Dyspnoea scale correlated well with other clinical parameters often used in the emergency department **(Kendrick et al., 2000)** and was found to be effective in assessing and monitoring treatments in non-acute and the acute environments. It has been a practical and effective way to provide documentation and continuity of care information and by incorporating the principles of self-rating, the patient is able to contribute valuable information to the medical and nursing treatment plan.

Six studies **(Bonilha et al., 2009, Clift et al., 2017, Engen, 2005, Pacheco et al., 2014, Lord et al., 2010, Lord et al., 2012)** utilised this scale. One study **(Bonilha et al., 2009)** showed a significant ($p = 0.02$) effect using this scale for dyspnoea i.e. transitory elevations on the Borg scale. Another study **(Grasch, 2013)** showed significant change at follow-up from the mean at baseline according to the Borg Fatigue rating (7.60 vs 8.88), however this study has a confounding factor that pulmonary rehabilitation continued throughout which could impact the effect of the intervention of interest i.e. singing. Finally, the distance walked in one study **(Clift et al., 2017)** was shown to have a small but non-significant reduction using the Borg breathlessness scale and participants reported no change in breathlessness following the 6MWT. This is a positive finding though as it showed no deterioration in exercise potential over the period of the evaluation.

Baseline/Basal Dyspnoea Index (BDI)

The baseline dyspnoea index (BDI) consists of five specific grades for each of three categories: functional impairment, magnitude of task and magnitude of effort. To grade dyspnoea with the

BDI, an observer interviews the patient and asks open-ended questions concerning the patient's symptoms which should take around three minutes. They then focus on specific criteria for the severity of breathlessness in each category as outlined in the BDI. The degree of impairment is based on patient responses and graded related to dyspnoea for all three components. Baseline focal score is obtained by adding the three ratings for functional impairment, magnitude of task and magnitude of effort **(Mahler and Wells, 1988)**.

This tool was implemented by one study **(Bonilha et al., 2009)** but noted no significant changes in BDI however commented that due to limitations in the study design it cannot be excluded the possibility that the enrolment of a greater number of subjects would provide evidence of significant changes in these parameters.

University of Cincinnati Dyspnoea Questionnaire (UCDQ)

The University of Cincinnati Dyspnoea Questionnaire (UCDQ) was developed to measure the impact of dyspnoea during physical activity, speech activity and simultaneous speech and physical activity and provides valid and reliable information about the effect of dyspnoea on speech and daily activities **(Hodge et al., 2003)**. This was also only utilised by one study **(Herer, 2013)** and again no significant changes were recorded.

Dyspnoea within other Heath Questionnaires

Several studies utilised tools which contain a component to measure aspects of dyspnoea as below:

Chronic Respiratory Disease Questionnaire (CRQ)

Chronic Respiratory Disease Questionnaire (CRQ) is an interviewer-administered questionnaire containing a dyspnoea element, utilised by three studies **(Canga et al., 2015**, **Miyahara et al., 2001**, **Lord et al., 2011)** but with no significant results or findings in regard to singing itself.

One study **(Canga et al., 2015)** utilised this tool along with a visual analogue scale (VAS), consisting of a succession of lung drawings representing the progressive severity of dyspnoea with a numerical rating scale. The analysis of the results of two independent tools shows a significant improvement of perceived dyspnoea on the control group, observed over session and time. The most significant change was observed after the 5th week of treatment. The author speculates this might relate to the effect of musical agency on perceived dyspnoea during exercise. However, the design of the study utilises several confounding elements in addition to singing within the intervention, so it is impossible to draw a firm conclusion about singing itself from this study.

Physiological Respiratory Measures

Researchers have found that physiological, functional and psychosocial consequences of COPD are only 'poorly to moderately' related to each other. Comprehensive assessment of the effects of COPD requires an extensive amount of instruments that not only capture the disease specific effects, but also the overall burden of the disease on patients everyday functioning and emotional wellbeing **(Engström et al., 2001)**.

There are a number of patient reported outcomes tools available specifically for respiratory illness, those measures most commonly providing key findings within these studies are:

St George's Respiratory Questionnaire (SGRQ - QoL)

The SGRQ is a self-completed multiple-choice questionnaire involving the patients' perceived wellbeing in regard to comfort with their lung function during daily activities and how the disease affects their life. Three component scores are calculated: Symptoms, Activity, and Impacts from which a total score is derived **(Grasch, 2013)**.

Three studies provided some positive findings utilising this tool. One study **(Clift et al., 2013)** showed a change of 3.3 points in the direction of health improvement. A second study showed a significant improvement of 41 ± 20 to 31 ± 18 points ($p = 0.016$) in the experimental singing intervention group compared with a change of 44 ± 12 to 37 ± 16 points ($p = 0.037$) in the control group where participants were assigned to a course of physiotherapy lung exercises **(Jamaly et al., 2017)**.

One study **(Clift et al., 2017)** showed a 0.2 increase in a negative direction on the total score of this scale but the change for the symptom scale showed a positive improvement of 7.8 points. However, there are no available guidelines on the interpretation of sub-scales within the SGRQ, as an assessment of the 'minimum clinical important difference' (MICD) in scores for the SGRQ can only be made for the total score.

One study **(Bonilha et al., 2009)** showed significant improvements of 'Quality of Life' scale within group comparisons. The mean change in 'Quality of Life' score of the control group for this study was slightly higher than the improvement of the singing group though, but the difference did not reach statistical significance ($p = 0.06$).

Three further studies **(Goodridge et al., 2013, Grasch, 2013, Lord et al., 2010, Pacheco et al., 2014)**, employing the SGRQ, did not find improvements in health-related quality of life, exercise capacity, or perceptions of illness for participants in the singing program compared to those receiving usual care.

COPD Assessment Test (CAT)

The COPD Assessment Test (CAT) is a questionnaire designed to measure the impact of COPD on a person's life, and how this changes over time. It was developed in response to a need for a validated short, simple instrument to quantify chronic obstructive pulmonary disease (COPD) impact in routine practice to aid health status assessment and communication between patient and physician**.** It is a short, simple questionnaire for assessing and monitoring COPD. It has good measurement properties, is sensitive to differences in state and provides a valid, reliable and standardised measure of COPD health status **(Jones et al., 2009)**. The CAT tool contains questions which are similar but not as detailed or specific as those contained in the SGRQ.

This measure was employed by four studies **(BLF, 2017, Lord et al., 2012, Jamaly et al., 2017, Pacheco et al., 2014)**. Two studies showed impact of COPD on a patients' life as measured by the CAT improved significantly during the singing intervention. One study **(Jamaly et al., 2017)** recorded significant improvement ($p = 0.029$) in the singing intervention group (17 ± 9 to 12 ± 8 points) though a significant change ($p = 0.161$) was not seen in the control group (physiotherapy lung exercises). Another study **(BLF, 2017)** recorded significant mean improvement of 1.1 ($p = 0.02$) post singing intervention. No significant findings were recorded with the CAT by the other two studies **(Lord et al., 2012, Pacheco et al., 2014)**.

Visual Simplified Respiratory Questionnaire (VSRQ)

The Visual Simplified Respiratory Questionnaire (VSRQ) is designed to assess health-related quality of life (HRQoL) in patients with COPD. It contains eight items: dyspnoea, anxiety, depressed mood, sleep, energy, daily activities, social activities and sexual life. Psychometric properties of this tool were assessed during a clinical trial of COPD patients, including the determination of structure, internal consistency reliability and concurrent validity with the St George's Respiratory Questionnaire (SGRQ), test-retest reliability, clinical validity and responsiveness to change over two weeks **(Perez et al., 2009)**.

This was utilised by two studies **(Bonilha et al., 2009, Herer, 2013)** but only one study **(Herer, 2013)** showed principal significant variations (+50.0% $p < 0.01$ i.e. 87.5%).

Breath Management and Control

Patients control of breathing was assessed by two studies **(Lord et al., 2010, Lord et al., 2012)** using two measures. These were the breath hold test, where subjects held their breath from maximum inspiration and also single breath counting, where subjects were instructed to breathe in and then count out loud in time with a metronome running at 60 beats per minute (bpm). Three attempts at each manoeuvre were made and the mean values recorded. However, there was no improvement shown in either study.

One study **(Engen, 2005)** also captured data for breath management including single breath counting as above but with the metronome set at 92 beats/ minute (bpm) falling behind by more than one beat stopped the count and the measure taken was the mean of 3 trials. Breath support was also captured and defined as the power behind the breath, measured as intensity of speech.

Persons with emphysema are known to have a breathy vocal quality and so a measure of vocal support (intensity of speech) in decibels, was measured in this study too **(Engen, 2005)**. A RadioShack® digital sound level meter was used to record the maximum decibel level during speaking (as if to an audience). A simple visual check was also taken at each measurement period to determine whether or not the patient could learn to breathe diaphragmatically.

Physiological and Health Measures

Short Form Health Survey SF-36 (SF-36) and York SF-12 (SF-12)

The SF-36 is a generic health status measure which has gained popularity as a measure of outcome in a wide variety of patient groups and social surveys. The SF-12 is an even shorter measure, which reduces respondent burden and in studies has been seen to accurately reproduce the two summary component scores which can be derived from both surveys. Physical Component Summary Score (PCS) and Mental Health Component Summary Score (MCS) calculated from the SF-36 or a sub-set of 12 items (the 'SF-12') were virtually identical and indicated the same magnitude of ill-health and degree of change overtime **(Jenkinson et al., 1997)**.

One study utilised the shorter version (SF-12) **(Clift et al., 2013)** and two studies **(Lord et al., 2010, Lord et al., 2012)** utilised the full version (SF-36). No changes were seen on SF-12 components **(Clift et al., 2013)**, but two studies showed a significant difference in response for the physical component summary (PCS) score of the SF-36, favouring the singing group i.e. one study **(Lord et al., 2010)** showed improvement of +7.5(14.6) vs. -3.8(8.4) ($p = 0.02$) and another study **(Lord et al., 2012)** showed improvement of +12.9 (19.0) vs -0.25(11.9) ($p = 0.02$).

One study **(Clift et al., 2013)** referred to the fact that a previous study **(Coulton et al., 2015)** showed SF12 mental health improvements for older people participating in singing over twelve weeks, as rationale for using this score, as well as noting the previous changes on SF36 reported by two earlier studies **(Lord et al., 2010, Lord et al., 2012)**. Evidence shows that the SF12 and SF36 scores are strongly correlated **(Jenkinson et al., 1997)**.

There is no further information available to explain which aspects of the PCS score specifically were potentially impacted by the singing intervention in the studies that showed improvement

(Lord et al., 2010, Lord et al., 2012). It may be interesting to research these characteristics further in this population following these results.

EuroQoL Test (EQ-5D)

The EuroQOL five dimensions questionnaire (EQ-5D) is one of the most commonly used generic questionnaires to measure health related quality of life (HRQoL). The conceptual basis of the EQ-5D includes the medical definition, as well as the fundamental importance of independent physical, emotional and social functioning. The concept of health in EQ-5D also encompasses both positive aspects (well-being) and negative aspects (illness). The EQ-5D is deemed to be short, easy to use and flexible and has been used successfully in several different settings (scientific trials, health policies, pharmacoeconomics, clinics, etc.). It consists of a questionnaire and a visual analogue scale (EQ-VAS). The EQ-VAS is a self-rated health status using a VAS to record patient perceptions of their own current overall health and to monitor changes with time. The self-assessment questionnaire is self-reported description of the subject's current health in 5 dimensions i.e., mobility, self-care, usual activities, pain/discomfort and anxiety/depression **(Gusi et al., 2010)**.

Four studies **(BLF, 2017, Pacheco et al., 2014, Clift et al., 2013, Clift et al., 2017)** utilised this health questionnaire though no significant changes were found in any of these studies.

Hospital Anxiety and Depression Scale (HADS)

The Hospital Anxiety and Depression Scale, or HADS, was designed to provide a simple yet reliable tool for use in medical practice. The term 'hospital' in its title suggests that it is only valid in such a setting, but many studies conducted throughout the world have confirmed that it is valid when used in community settings and primary care medical practice **(Snaith, 2003)**.

Five studies **(Lord et al., 2010, Lord et al., 2012, Liu et al., 2016, McNaughton et al., 2017, Trivedi, 2017)** utilised both the HAD anxiety and HAD depression scales. One study **(Liu et al., 2016)** recorded a significant improvement ($p < 0.001$) in the experimental group following a twenty-four-week singing intervention which was significantly different ($p < 0.01$) between experimental group and control group (control group were provided with health education and mental nursing though no further details could be identified). Another study **(Trivedi, 2017)** showed a significant change in total HAD score in both experimental and control groups (both also employing PR) but the change in HAD score following intervention for the experimental group was more significant ($p = 0.001$) than that seen in the control group ($p = 0.023$) suggesting an improvement in anxiety and depression due to the singing intervention.

One further study **(McNaughton et al., 2017)** showed a difference in the mean total HAD score between baseline and 12 months but this was not clinically significant ($p = 0.37$) but a significant ($p = 0.038$) mean reduction in HADS anxiety score of 0.9 units was observed at 1 year though

this did not exceed the minimal clinically important difference (MCID) of 1.32. This replicated a significant (p = 0.03) improvement in HAD anxiety score -1.1(2.7) vs. +0.8(1.7) noted by one of the other earlier studies **(Lord et al., 2010)**.

The other remaining study **(Lord et al., 2012)** that utilised both the HAD anxiety and HAD depression scales and one further study **(Pacheco et al., 2014)** that utilised the HAD anxiety scale alone, showed no further significant results recorded using these tools.

General Anxiety Disorder-7 (GAD-7)

The General Anxiety Disorder-7 GAD-7 is a self-reported anxiety questionnaire that is often used in mental health assessment. It is a valid and efficient tool for screening for general anxiety disorder and assessing its severity in clinical practice and research **(Spitzer et al., 2006)**.

This measure was employed for just one study **(BLF, 2017)** but a significant improvement (where p = 0.04) in levels of anxiety in participants was shown after three months.

General Outcome Measures

There were a number of other validated measures which were only utilised within one study, most of which did not produce significant study findings. These might however provide useful information to support selection of tools for future study design.

Duke Health Profile

The Duke Health Profile is a physical health subscale which addresses quality of life and general well-being by asking about self-perception, relationships, breathlessness, overall health, and socialisation. Scores are scaled in two broad categories of "function" and "dysfunction." One study **(Engen, 2005)** employed this scale and four of the six functional scales were scored: physical health score, mental health score, social health score, and self-esteem score. Additionally, three or the five dysfunctional scales were considered: anxiety score, depression score, and anxiety depression (DUKE-AD) score. Two of the seven subscales analysed, mental health and social health, showed an upward shift in quartile scores though this was not noted as significant.

Brief Illness Perceptions Questionnaire (BIPQ)

The Brief Illness Perceptions Questionnaire (BIPQ) is a nine-item scale designed to rapidly assess the cognitive and emotional representations of illness **(Broadbent et al., 2006)**. It was utilised by one study **(Goodridge et al., 2008)** but there were no significant differences for this scale within or between the groups at baseline or following the programme.

London Chest Activity of Daily Living (LCADL)

The London Chest Activity of Daily Living (LCADL) scale is a tool aimed at assessing the level of dyspnoea during ADL. The tool is divided into self-care; household activities; physical activity; and leisure activities and is considered an inexpensive and user-friendly instrument. It can be a feasible clinical tool for assessment and monitoring of dyspnoea-related functional impairment in chronic lung disease patients **(Muller et al., 2013)**, as well as for pre- and post-intervention assessment as utilised by one other study **(Pacheco et al. 2014)**. This provided no significant outcome, but the study had a very small sample size which means the study was under-powered to detect change.

Brief Symptom Inventory−18 (BSI-18)

The Brief Symptom Inventory−18 (BSI-18) is an 18-item self-report screening tool designed to assess current (past seven days) psychological distress in “a broad spectrum of adult medical patients 18 or older and adult individuals in the community who are not currently assigned patient status” **(Meachen, 2008)**. It measures depression, anxiety, and somatic symptoms, with a subscale score in each of these three categories. The General Severity Index is a composite score derived from this tool, which assesses the overall distress of the patient **(Grasch, 2013)**. It is the second abbreviated version of the 90-item Symptom Checklist based on the Hopkins Symptom Checklist **(Meachen, 2008)**. It was successfully utilised by one study **(Grasch, 2013)** but without any significant changes.

Patient Activation Measure (PAM)

The Patient Activation Measure (PAM) measures patient engagement with their self-management by examining their knowledge, skills and confidence that are essential in their own self-care. It is a valid, highly reliable, unidimensional scale that reflects a developmental model of activation. Activation is regarded as having four stages: (1) believing the patient role is important, (2) having the confidence and knowledge necessary to take action, (3) actually taking action to maintain and improve one's health, and (4) staying the course even under stress. The measure has good psychometric properties indicating that it can be used at the individual patient level to tailor intervention and assess changes **(Hibbard et al., 2004)**. This was employed in only one study **(BLF, 2017)** but with no significant results.

There were some remaining unique tools used e.g. Questionnaires CareFusion, Kent, UK **(Lord et al., 2010)** but no further significant results were produced from these.

Qualitative data - Interviews, Journals and Participant Feedback Surveys

There were a number of interview, journal or participant feedback methods utilised to capture comments, experience and other outcomes as provided by participants in verbatim text.

Interesting qualitative information is captured extensively within some of these studies **(e.g. Clift et al., 2013, Clift et al., 2017, McNaughton et al., 2016)** and common themes could be extracted even though the efforts and methods employed are variable throughout the group of studies identified.

A total of nine mixed model studies **(BLF, 2017, Clift et al., 2013, Clift et al., 2017, Engen 2005, Goodridge et al., 2013, Lord et al., 2010, Lord et al., 2012, Pacheco et al., 2014, McNaughton et al., 2017)** utilised interview, journal or written feedback survey techniques in addition to other standardised outcome measures. Only one wholly qualitative study **(Thomas et al., 2015)** was located and included. The written comments provided from individuals during interview for three of these studies **(Clift et al., 2013, Clift et al., 2017, McNaughton et al. 2017)** were captured and presented with some rigor and the common themes from these were further examined using content and thematic analysis **(Skingley et al., 2014, Skingley et al., 2018, McNaughton et al. 2016)** to provide in-depth summaries and important considerations.

The findings from one study **(Skingley et al., 2014)** suggest that singing was perceived as both acceptable and beneficial to the participants, not only for breathing but also in relation to general physical, psychological and social well-being for example "*I love coming to COPD Singing Research Project and I always feel much better physically and emotionally afterwards*", *"It certainly appears to have helped with my general breathing",* and *"I believe that the project is teaching me how to understand my breathing and how to control it. This is very useful; it stops me hyperventilating when my breathing is under pressure i.e. climbing a steep hill".* A further similarly designed study **(Skingley et al., 2018)** provided rich and practical information regarding how singing provided meaningful impact to participants, supplementing the quantitative data in this area and strengthening the potential for generalisability. The majority of participants in this later study agreed that the singing programme led to improvements in respiratory symptoms and in mental wellbeing and led to social benefits, often acting as support and avenues to seek advice. These qualitative data results were significantly more positive than those results expressed by the quantitative data for the same studies.

One mixed model feasibility study conducted in New Zealand **(McNaughton et al., 2016)** analysed transcripts from individual interviews and a focus group meeting, regarding their experience after six to nine months of group singing. Individual interviews each lasted thirty to forty minutes and focussed on three topics of interest: participants' experiences of participation in the singing group, any perceived effects or consequence as a result of being in the group and finally whether the singing group was similar to or different from other community support groups or singing groups they had been involved in. Overall, the results indicated that the singing group was a highly positive experience and participants reported perceived health benefits from participation in the group, and no adverse events. These health benefits included improvements in breathing, sputum clearance and exercise tolerance, as well as a general sense of improved wellbeing. The key themes from the interviews and focus groups provided an explanation for the possible social mechanisms which contributed to the high attendance rates

and self-reported health improvements. Four key themes were identified including *'being in the 'right space', 'developing a sense of connection with others', 'experiencing shared purpose and growth'* and *'participation in meaningful physical activity'*. The findings for the qualitative componant of the study again further supported the positive outcome of the quantitative results.

A large singing for lung health programme **(BLF, 2017)** reported positive results from qualitative scoping work completed at three months post start, as part of a larger mixed-model study. The themes of the feedback provided by patients in the community singing groups were all very positive. These themes included *'Increased understanding of breathing techniques to reduce breathlessness and maintain optimal lung function', 'Increased awareness of the benefits of singing for lung health and self-management', 'Increased capacity for creative self-expression leading to increased self-confidence and quality of life whilst counteracting feelings of stress and anxiety', Increased accessibility to services which are open and approachable', 'Reduced social isolation - initial research has demonstrated how groups are removing such barriers, helping to bring people back into their local communities'*. This positive qualitative data further supported the outcomes from the quantitative data from this project.

A further two studies utilised structured interviews lasting for thirty minutes, both with the same template **(Lord et al., 2010, Lord et al., 2012)**. Subjects were asked to discuss their perception of any physical and emotional benefits or harm they had experienced from attending the sessions. A selection of comments indicated a much more positive conclusion than the measurement data for example "*It has made my life easier; I would have liked this when I was first diagnosed*", "*I increased my out-breath from 4 to 14 counts*", "*I started breathing much better, from the stomach*", "*The exercises, thinking about breathing and relaxing when I have (breathing) problems, this has been very useful*", "*I always felt better afterwards physically*". There were other positive physical effects described which indicated positive impact on lifestyle and functional ability i.e. "*I have better posture now*", "*Walking better, I go out more when it's not cold*", "*Now things are less of a chore, housework is no longer a struggle*" **(Lord et al., 2010)**. The overall message from the qualitative componant of these studies was again more encouraging than the positive quantitative outcome measures assessed.

The only solely qualitative study included in the systematic review **(Thomas et al., 2015)** identified four themes from the qualitative analysis of the semi-structured interviews employed at the end of an almost two-year study. The themes identified were 'Control of Symptoms', 'Community and Friendship', 'Psychological Benefits', and 'Mastery of Illness'. The singing group was reported to improve breathlessness symptoms, enable access to further sources of support and formed new friendships. The study concluded that dominant effects were improving mood, providing a sense of mastery over breathing to better cope with breathlessness and tackling social isolation. The results of this study were positive, but partially confounded in regard to the systematic review criteria, because only eleven of the sixteen participants included in the study were diagnosed with a COPD related illness.

One study **(Engen, 2005)** reports that throughout the duration of the treatment, subjects were asked to keep a journal. Information on practice times, musical preference, frustrations, health comments and so forth. These were not analysed as independent measures but were used to monitor participants' reactions. Journal sheets were provided to all participants, but not equally used i.e. Study results show male participants only made entries when reminded during sessions, while the frequency and quality of entries by women varied widely.

One small feasibility study **(Goodridge et al., 2013)** included free response journal entries and added a satisfaction survey which was mailed to participants post participation. This post-intervention programme survey was used to evaluate acceptability and enjoyment of the singing programme and additionally served to identify factors that affected sustainability of the intervention. This data was provided from written feedback and although neutral findings were established from the quantitative measures recorded in the study, the qualitative data from the written feedback from participants was uniformly positive about the experience of participating in the singing intervention. Twelve participants indicated that they felt singing helped their breathing during the session itself and would continue singing after the programme and ten also believed that the programme helped their breathing overall.

The final study **(Pacheco et al., 2014)** held only limited qualitative information. It analysed the answers to a specifically designed questionnaire which indicated that all patients reported improvement in lung function control, reduced anxiety and more self-esteem after the singing lessons. In addition, all patients reported a feeling of well-being and recorded the experience as very pleasant. This data was very limited as the sample size included four participants, however it did improve the overall outcome of the study which was initially negative based on the quantitative data alone.

Several studies that indicated that feedback had been solicited or that interviews had been completed did not provide all the details about the questions asked or how the questions or interviews were implemented **(Engen, 2005, Lord et al., 2010, Lord et al., 2012, Thomas et al., 2015)**, while other studies noted full details of the scripts or indicated that templates or qualitative questionnaires were used or specified that researchers were trained to ensure consistency of interview technique **(BLF, 2017, Clift et al., 2013, Clift et al., 2017, McNaughton et al., 2016, Pacheco et al., 2014)**.

Adverse Outcomes

Adverse events were not formally captured in any of the selected studies. The proportion of patients experiencing adverse events due to the intervention appear to be very low though, as are the total number of hospitalised days, symptomatic days and proportions of participants who had respiratory exacerbations and/or hospitalisations. These are not presented in detail in any of the studies reviewed.

There is some anecdotal information provided about problems experienced from observations by the authors and interviews for example, one study **(Bonilha et al., 2009)** mentions that vocal exercises and singing popular songs were well tolerated by the patients and they did not complain of severe dyspnoea, chest pain, regurgitation or dizziness in the course of the activity. However, a high prevalence of coughing and sputum expectoration was observed during the resting intervals. Another study **(McNaughton et al., 2017)** asked patients about adverse events and noted that a participant had a lung transplant during the year of the study but continued to sing with the group indicating it was well tolerated.

Treatment Adherence

Adherence to the classes was not presented for the majority of studies. There were several studies that built adherence into the design by ensuring that a patient could only be considered for collection of post treatment results if they had attended a number of singing sessions or classes e.g. Subjects were required to have attended at least three sequential classes before the final assessment **(Bonilha et al., 2009)** or patients were excluded from the analysis if they did not complete the required number of sessions **(Canga et al., 2015)**.

Patient Retention

The retention of patients over all the studies is generally very good and feedback from patients was positive. Any detailed explanations provided by each of the authors is summarised in Table 5 as below (where the information was provided).

Unique identifier (Author/Year)	**Study Recruitment and Retention Details**
Bonilha 2009	Seventy-eight patients were evaluated. Thirty-five were excluded due to not meeting inclusion criteria or refusal to participate. Out of forty-three patients who agreed to participate in the investigation, twenty-three were allocated to the Singing Group and twenty to the Control Group. A total of thirteen patients discontinued the study, but at a proportion not significantly different between groups (Singing group: n=8; Control group: n=5). Reasons for abandoning the study in the singing group included non-medical causes (3) and non-pulmonary medical conditions (4). Only one patient left the singing group due to repeated episodes of acute COPD exacerbation and difficulties in regularly attending classes. Reasons for dropping out of the Control Group were non-medical causes (4) and a non-pulmonary medical condition (1). Fifteen patients concluded the entire protocol in both arms of the interventions. The final gender composition was similar in both groups, with a predominance of males. There was 30% attrition over 24 weeks.

BLF 2017	At baseline 167 questionnaires were collected and at three months this reduced to 80 with a maximum of 70 paired samples. The drop-in follow-up was due to some participants not completing the course as well as a smaller number of groups folding before completion though the reasons are not specified. The majority of singers were women (116/167) and most were ex-smokers. Approximately 50% of patients had participated in an exercise class or attended PR. The British Lung Foundation (BLF) maintained regular phone and email contact with the leaders in order to monitor participant attendance and support ongoing recruitment and sustainability of their groups. The BLF and local leaders have also been promoting SLH through targeted press and social media posts. E.g. featuring in two reputable medical journals, The Lancet Respiratory Journal and the Practice Nursing Journal. Locally each singing leader was provided with a press release to send out, which helped to generate interest locally. One of the participants wrote a blog sharing personal stories of involvement and benefits. The BLF also created a video to highlight the positive benefits and during World COPD Day many groups helped to highlight the day by recording a video and sharing it on YouTube.
Canga 2015	Ninety-eight patients were randomly allocated to the treatment and control group. Thirty patients out of the ninety-eight enrolled were excluded from analysis due to non-completion of required number of sessions either due to anticipated time commitment or personal/medical concerns. Therefore, a total of sixty-eight patients were included in the final analysis
Clift 2013	Thirty-four participants left the study overall and did not complete the final questionnaire (32.1% attrition). A further six participants were unable to attend for the final spirometry assessments due to other commitments and illness (37.7% attrition). The rate of loss to the study is less than the conservative estimate of 50% per design. Comparisons were made between those dropping out and those remaining in the study and no significant differences emerged on any measure taken at baseline, so attrition did not introduce bias into the study. In addition, only a small number of withdrawals (2.8%) were due specifically to COPD related health issues, and only a small proportion of missed attendances during the programme were due to COPD related health issues (1.5%).
Clift 2017	Participants referred themselves into the study on the basis of information received through their surgeries or various other routes. Everyone, describing themselves as affected by breathing problems, was accepted. Groups ran on a weekly basis with good regular attendance. Study did not recruit to the level originally hoped. In retrospect, the degree of time and effort needed to communicate effectively with health professionals and those affected by COPD and to encourage participation was

	underestimated. At the outset a well thought out strategy was adopted. Contact was made with the only BLF Breathe Easy group locally, enlisting their support. Suitable venues were found and taster sessions run. Times/dates decided so that they could be advertised early. Attractive recruitment posters and flyers were used and website created. A high-profile launch event at the Royal Festival Hall conducted and high quality promotional created. Lead GP's + Practice Managers at 10-12 health centres within 30 minutes walking distance of the advertised venues were contacted. Posters/flyers sent to surgeries and a request made to surgeries to send out information to patients on their COPD registers. The request was made a second time as there was low up-take + eventually only six practices undertook to write to patients. Recruitment still fell short so further efforts were made to enhance publicity through advertising in local newspapers, street level leafleting, particularly with the recruitment efforts. Baseline assessments took place in September 2015, and three groups commenced. At the end of the recruitment process, sixty patients were enrolled out of the one hundred targeted.
Engen 2005	Twelve patients participated but only seven completed the requirements for inclusion. One patient withdrew due to a family emergency, two had initial breath measures too low for inclusion and two had measurements too high for inclusion. The four participants with measures outside inclusion limits had signed informed consent and completed the programme as they wished to participate.
Goodridge 2013	Twenty-one participants from the original twenty-two identified actually met the eligibility criteria. The first fourteen participants were assigned to the singing intervention. The remaining seven were assigned to the control group receiving usual care. Two of those assigned to the control group declined to participate as they were interested in the singing intervention, leaving five in the control group and a total of nineteen patients overall.
Grasch 2013	Thirty-one subjects diagnosed with emphysema consented to participate. Twenty-five patients completed the study, nine males and sixteen females
Herer 2013	All the patients were invited to these sessions a near-maximum attendance; However, an asthmatic patient, having a string dysfunction antecedent voice, had to prematurely terminate these sessions.
Lichtenschopf 2014	Ninety-two COPD patients and twenty-seven patients were included with other underlying disease and all completed the study though no further details were included.
Liu 2016	Total of fifty-six COPD patients were enrolled and divided into intervention group and control group.
Lord 2010	Thirty-six patients were selected with twenty allocated to the singing group and sixteen to the control group. Two subjects withdrew from the singing

	group once the workshop had begun and three did not attend the final assessment once completing the workshops. In the control group, one withdrew and two did not attend the final assessment. Data are therefore presented for the fifteen singers and thirteen controls that completed the study. Six of these attended twelve sessions, four attended eleven, two came to ten and three came to nine. Attrition rate was 22% over six weeks.
Lord 2012	One hundred and eighty-three patients were approached to participate. One hundred and fifty declined to take part. Eighteen patients were allocated to the singing group and fifteen to the film group. Five subjects from the singing group withdrew once the sessions had begun. In the film group, one subject died after randomization but before attending any sessions and three subjects withdrew from the study once the sessions had begun. Data were presented for thirteen subjects in the singing group and eleven in the film group that completed the study. Median number of sessions attended was 14.5 for the singing group and 7 in the film group. Attrition rate was 19% over eight weeks.
McNaughton 2017	Twenty-eight patients were enrolled in the singing group: Five patients withdrew within one month of enrolment (disinterest or joining another therapeutic trial). After six to nine months of singing, twelve participants contributed to interviews about their experiences and after analysing data from the individual interviews nine of the remaining eleven group member participated in a focus group. Two could not attend due to other commitments.
Miyahara 2001	Twenty patients were enrolled in the study and ten patients were randomly assigned to an experimental group. No further details were available regarding drop out or attrition rates
Pacheco 2014	Eight patients were selected to participate but two were subsequently excluded. Two patients were dropped from the study analysis because they did not attend at least seven sessions.
Thomas 2015	Sixteen patients (four male and twelve female) with mean age 72.6 (50–92) years, were interviewed. Diagnoses included COPD (11/16), asthma (2/16), bronchiectasis (2/16) and fibrosis (1/16). All were non-smokers (ex-smokers 12/16); 12/16 (75%) had previously attended pulmonary rehabilitation. 10/16 lived alone and 8/16 had a history of mental health comorbidity requiring treatment. No further details were available regarding drop out or attrition rates.
Trivedi 2017	Ninety patients were screened patients and based on inclusion and exclusion criteria fifty-six patients were recruited in the study. All fifty-six patients were divided into two groups (n=28) and two patients dropped from the experimental group of the study due to non-medical reasons.

Table 5 – Systematic Review - Study Retention and Withdrawal Summary

One of the studies **(Skingley et al., 2014)** reports that reference to the social aspects of the singing groups were universally positive in nature. This exceeded the number of comments related to breathing with the comments broadly pertaining to friendship and company generally or support gained from meeting those similarly experiencing COPD. This might indicate that there would be a positive impact on retention rate for singing groups and also a further up-take of this intervention in comparison to other traditional alternative activities such as exercise classes and pulmonary rehabilitation. This view is supported by the low attrition rate of 32.1% noted in this same study **(Clift et al., 2013)**.

Health Care Utilisation

Health care utilisation data acquisition was non-existent in all but four studies **(Clift et al., 2013, Clift et al., 2017, Liu et al., 2016, Thomas et al., 2015)**. There was one study **(Clift et al., 2017)** that attempted to carefully address this issue by monitoring health and social care service usage over the study duration to assess whether any changes took place which could provide evidence on potential cost savings associated with participation in group singing. A section entitled 'Treatments and services to help you with your breathing' was included which raised questions on oxygen and inhaler use, admission to hospital and the health and social services used (including visits to a hospital accident and emergency department, appointments with a GP, appointments with a practice nurse and home visits). There did not appear to be any evidence of significant changes between baseline and follow-up and the research concluded that there appeared to be no evidence of either a clear increase or decrease over the period of the project.

One study **(Liu et al., 2017)** confirmed that data for self-reported primary care (GP) visits were fewer in the year following commencement of singing but this was not a significant change and another **(Thomas et al., 2015)** noted there was no difference in hospital admissions in the year after starting singing compared to the year before.

Study Quality

Randomised controlled trials are the most rigorous way of determining whether a cause-effect relation exists between treatment and outcome and for assessing the cost effectiveness of a treatment **(Sibbald and Roland, 1998)**. This is largely due to more effective elimination of bias by ensuring random allocation to intervention groups and ensuring groups are treated identically except for the treatment. It also allows the analysis to be focused on estimating the size of the difference in predefined outcomes between intervention groups.

There are four randomised controlled studies (RCTs) within the selected group that compare singing alone specifically against an active control **(Bonilha et al., 2009**, **Jamaly et al., 2017, Lord et al., 2010, Lord et al., 2012)**. These trials are therefore more impactful within this area

of research and the results from the earlier of these trials are more well-known within the research community **(Bonilha et al., 2009**, **Lord et al., 2010, Lord et al., 2012)** and as such have generally been cited more often to date.

Randomised controlled trials are powerful tools, but their use is limited by ethical and practical concern **(Sibbald and Roland, 1998)**, this is particularly true when the intervention itself is dependent on the patient's active involvement, understanding and participation.

Studies including singing or similar activities are certainly impacted by practical concerns due to the nature of the intervention. Ethical aspects could be a consideration too, even though singing is largely considered as an activity complimentary to standard care rather than a replacement. One of the key ethical concerns would be if singing were offered where pulmonary rehabilitation is not offered as part of standard of care or offered as an alternative. Pulmonary rehabilitation is now widely used as part of the mechanism for COPD management and meta-analysis as part of a Cochrane review strongly supports respiratory rehabilitation as part of the spectrum of management for patients with COPD **(Lacasse et al., 2009)**.

Observational studies such as cohort studies or case studies have often been more widely executed successfully within this research genre. One study **(Lord et al., 2010)** reported that in addition to a randomised study, an open workshop was also completed. However, the selective bias of this study is questionable since certain measures were included or mentioned at baseline but not presented post treatment.

The quality of each study was measured using the extended Cochrane Collaboration tool for assessing risk of bias **(Higgins and Green, 2011)** Cochrane Handbook for Systematic Reviews of Interventions Version 5.1.0 [updated March 2011]. In this case the questions within the risk of bias tool were collated in a tabular format showing the questions and responses of the tool and documented in a study risk of bias summary table (This is illustrated in Appendix B*).

The risk of bias generally is very high with the rating of all papers considered to be weak or moderate at best. This is largely due to the nature of the intervention i.e. finding a suitable blinding method is challenging, such that very few studies actually included robust randomisation methods and blinding of participants is not possible. There were only four studies **(Bonilha et al., 2009**, **Jamaly et al., 2017, Lord et al., 2010, Lord et al., 2012)** which mentioned randomisation and included some reference to the assessors or investigators being blinded to the intervention, although as mentioned, blinding of patients is challenging in this area even in the more robustly designed randomised trials. This increases the potential for treatment bias and the challenges faced by researchers are great.

Systematic Review: Discussion

Study Demographics

Demographic information was variable in quality across the studies selected and there was little subgroup analysis regarding demographics, for example to understand how efficacy may be dependent on gender or age factors. Nine of the selected studies captured gender information and these were generally seen to contain higher proportions of females than males. This could likely be due to singing groups and choirs in general often seeing more females having an interest in taking part. Singing studies in general speak to the challenges choir directors have in recruiting male participants. Research shows that this may be because of the perception of singing as being feminine gendered **(Warzecha, 2013)** and other more complex gender related issues regarding physiological changes and challenges particularly during male adolescence **(Freer, 2007, Freer, 2012)**.

One of the major steps forward in the field of COPD was a global consensus on the definition, classification and management of the disease through the “GOLD” guidelines. This was not only was a landmark with regard to providing reliable and up-to-date knowledge about this disease, but it also enabled worldwide harmonisation of epidemiological, clinical and experimental research studies in patients with COPD. The latter is essential for the comparison of results and therefore, for further advances in the field **(Sterk, 2004)**.

Eight studies specifically mention that GOLD staging criteria were used as inclusionary diagnosis qualification for COPD, but more often other terminology such as COPD diagnosis or ‘stable COPD’ was used in the studies identified. In studies where it is mentioned, there is little evidence presented to indicate if the criterion were fully adhered to or checked individually for each patient **(e.g. Lord et al., 2012)** or whether there was an assumption per specific rationale made by the researchers that patients adhered to this definition. The studies selected for this review were often recruited using patients from pulmonary rehabilitation groups, for example, or recruited through pulmonary clinics, so it is possible that GOLD criteria could have been utilised to diagnose the patients within these groups, even when this was not explicitly mentioned as part of the study protocol. It would seem an important step to ensure that agreed diagnostic criteria for patients are adopted in future studies in this area, so that data across studies within this field can be compared more consistently.

It was estimated in 2012 that more than 90% of COPD deaths occurred worldwide in low and middle-income countries **(WHO, 2017*b*)**, with COPD being more prevalent in these countries than in higher income countries. However, the studies identified within this review indicate that research in this area to date for the most part has been conducted in high income countries, with a bias overall to white, educated participants as specifically highlighted by one study **(Clift et al., 2013)**. That said, in 2012 it was accepted that this disease now affects men and women

almost equally, due in part to increased tobacco use among women in higher-income countries **(WHO, 2017*b*)**. This suggests a gap in the research base and a need for more research globally, particularly across a wider scope of income and educational groups and regions.

The burden of chronic diseases, such as COPD, poses a special challenge in low-income countries, where health-care resources are traditionally designed to respond episodically to acute disease, especially infectious diseases, and are thus not well adapted to treating chronic diseases **(Lopez-Campos et al., 2016)**. Respiratory diseases in general receive little attention and funding in comparison with other major causes of global morbidity and mortality. In particular, COPD has been a major public health problem and researchers consider it will remain a challenge for clinicians within the 21st century **(Lopez-Campos et al, 2016)**.

The additional issue here is that even if financial barriers to access were removed, access to quality health care could still be compromised by other barriers, including patients' beliefs, difficult access to care centres, insufficient transportation, language barriers, and inadequate cultural competency of health-care providers **(Finn and Cohen, 2015)**. It would therefore seem potentially important that the relatively low cost and ease of access of singing as an intervention could be a key driver in expanding research to low or middle-income countries and extending promotion, education and exposure globally.

Study Design

Duration, Frequency and Design

The rationale for study duration and frequency, intensity or duration of the intervention is not provided in detail for the majority of studies, which is a significant gap. Those that do provide this information show that their designs vary quite widely, and the quality and consistency of the intervention is rarely assessed for consistency or quality.

There were only a few studies which included a control or randomised patients and only two studies compared singing to an alternative therapy or activity such as film discussion groups **(Lord et al., 2012)** and craft classes **(Bonilha et al., 2009)**. It is challenging to find an activity that is similar enough to singing that could be effectively utilised as a control. Types of classes or activities such as those mentioned would be a control for the social and logistical aspects of the singing class. The only activity close to singing for COPD patients is Pulmonary Rehabilitation.

Pulmonary Rehabilitation (PR)

It has been established that rehabilitation relieves dyspnoea and fatigue, improves emotional function and enhances patients' sense of control over their condition. The improvements of

pulmonary rehabilitation are moderately large and clinically significant, so rehabilitation now forms an important component of the management of COPD **(Lacasse et al., 2009)**.

Several studies enrolled patients directly from pulmonary rehabilitation training groups **(e.g. McNaughton et al., 2017)** as part of their design which is good practice in that it helps manage some confounding factors, allowing patients to start from a relatively similar educational perspective in regard to their respiratory understanding. Several studies included pulmonary rehabilitation as part of the design **(Canga et al., 2015, Grasch, 2013, Trivedi, 2017)** and this could have a potentially confounding effect on any quantitative or qualitative outcomes.

There could certainly be ethical issues in utilising pulmonary rehabilitation specifically as a control for singing for COPD however, due to the proven benefits of PR. It could potentially be used as a control for studies where patients enrolled having previously rejected PR but again this could certainly impact outcomes or bring about its own bias.

Pulmonary rehabilitation is now accepted within health systems as an essential strategy in the ongoing management of people with COPD and results of a Cochrane review meta-analysis strongly support including at least four weeks of exercise training, as part of the spectrum of treatment for patients with COPD **(McCarthy et al, 2015)**. However, at present there is often poor access to pulmonary rehabilitation internationally or a low uptake in certain regions. Measurable benefits of pulmonary rehabilitation also diminish over time and thus increasing uptake of PR and sustaining the benefits of this activity are very important **(McNaughton et al., 2017)**. Interventions to prolong the beneficial effects of pulmonary rehabilitation have been studied but most are not practical or financially feasible, and none have unequivocally been shown to be effective **(Spruit et al., 2013)**.

Singing as an intervention could clearly support or enhance this type of treatment since it is both enjoyable and has a positive impact on health and well-being of COPD patients. It can act both as an activity to potentially improve retention to PR groups or prolong adherence to PR activity or more importantly, in its own right as an alternative to PR. It would seem important to establish further the patient retention capabilities for singing groups both in their own right and compared to retention levels seen in PR groups.

Study Measures

It has been proposed that “disease modification” in regard to COPD be defined as any change in a metric, related to the disease, that is maintained over time and that any treatment given over time changes one or more of these metrics can be considered disease modification **(Mahler and Criner, 2007)**. This concept goes beyond the traditional approach of using lung function alone (in particular FEV1) for this purpose. COPD measures are therefore now much more extensively used to look at a variety of health aspects in relation to the condition. In the studies selected lung function tests showed varied and inconclusive results overall, so the

inclusion of additional non-lung function measures and outcomes is key to building a picture of how the physiological and psychological mechanisms of action of singing might impact these patients' health and wellbeing.

A large number and variety of measures were used throughout the studies reviewed and there was relatively little consistency of tools used between studies. There were also a number of studies which highlighted significant positive results, but from tools that are not particularly common or were only used for those studies in isolation e.g. a significant proportion of patients (87.5%) were seen to have an increased score of greater than four points for QoL using the VSRQ **(Herer, 2013)**, but this tool was only used by one study.

In addition to this issue and also the fact that the risk of bias was calculated to be high and many studies were lacking robust quality, it was deemed not to be feasible to perform a collective data analysis of any of the quantitative measures, as had been outlined for consideration in the original review protocol i.e. statistically combining the data from these studies could not deliver meaningful results. The Cochrane review protocol was designed to select the most robust randomised controlled studies and present any meaningful data analysis from these **(McNamara et al., 2016)**.

Journal data would have been valuable in this area but unfortunately in the studies where this was used it seemed to be inconsistently administered or spuriously tracked or adhered to e.g. missing journal records etc. **(Engen, 2005)**. Information collected from journals, especially in regard to treatment adherence and other ongoing patient behaviour data could provide useful insight within this research. It would be helpful for further emphasis to be placed on these aspects for future studies, particularly where home tools or self-management resources are utilised to extend or expand the treatment exposure. It would also be interesting to explore the specific components of all those tools where singing was shown to have positive results e.g. FVC, FVC%, FEV1, PCS - SF-36 etc. and to further understand whether there is any correlation with patient feedback from interviews and journals. This could lend quantitative support to the overall positive effects, which in turn could provide insight as to the quality or accuracy of the journal information.

Interview techniques and open text questionnaires provided the most positive of all results and gave wider insight and information regarding the physiological and psychological mechanisms of action involved in singing. The respondents reported improvements in mental wellbeing, attributing much of this to the singing group and many social benefits were recorded. These extended beyond singing sessions, with fellow participants often acting as support and advice channels **(Clift et al., 2017, Skingley et al., 2018)**. One study found that major effects were in mood improvements, providing a sense of control over breathing to manage dyspnoea more effectively and for tackling social isolation **(Thomas et al., 2015)**.

These interview techniques provide a wide source of data, though it can be challenging to review or analyse this collectively, due to the varying methodology and formats used. Interviews and participant feedback has provided important supportive data within this area and qualitative research has much to offer with methods that enrich knowledge. Analysing qualitative data is not a simple or quick task **(Mays and Pope, 2000)** and with quantitative data currently becoming cheaper and more accessible to capture and analyse, this is often the most cost-effective option and more often favoured globally to inform data-driven policy making **(Etsy et al., 2007)**. However, it would be recommended that further work to extract and analyse all the available qualitative data collected for singing for COPD to date would be worthwhile to help inform future research.

COPD progression is traditionally assessed using spirometry, but it has been noted **(Mahler and Criner, 2007)** that lung function is poorly related to other clinical outcomes i.e. dyspnoea, exercise performance and exacerbations and these patient-centred metrics are in fact, more important to the patients themselves than lung function measures or spirometry readings. This is clearly a very important factor regarding the methods of action of singing on COPD, since it has the potential to impact the patient in several ways, the magnitude of which could be variable between patients too. Individuals may place differing importance on the varying symptoms or outcomes of their disease.

Several researchers have studied the predictive accuracy of composite indices that include non-pulmonary factors to determine whether they can predict outcome better than lung function measurements such as FEV1. The use of some tools which are common to recent COPD research have not been employed by any of the singing for COPD studies available for this review. One tool in particular, which may have provided insight into the more holistic effects of singing on the patient is the BODE index. This specific index is constructed from four factors found to predict the risk of death in COPD patients. It is so called because it is constructed from body-mass index (B), the degree of airflow obstruction as measured by FEV1 (O), dyspnoea (D) and exercise capacity (E), measured by the six-minute–walk test **(Celli et al., 2004)**.

The BODE index was prospectively evaluated and patients with higher scores were shown to be at greater risk. This showed that the index was able to discriminate the probability of survival among patients more strongly than the conventional standard of FEV1 assessment alone. In recent studies, this index has also been shown to be an accurate predictor of COPD hospitalization and mortality as well as a useful index to track the effect of disease modification. It is a simple multidimensional grading system, better than the FEV1 at predicting the risk of death from respiratory causes among patients with COPD **(Celli et al., 2004).** This index was created in 1997 and validated by 2004 **(Celli et al., 2004)** although only one of the studies included in this review **(Herer, 2013)**, conducted after 2004 mentions this index or considers specifically whether it might be relevant. More importantly there in not a great deal of detail within the studies generally that explains the specific rationale as to why interviews or in fact any particular tools or measures were selected.

There appears to be no clear positive outcomes for any of the exercise measures for the majority of studies, although there is anecdotal information within comments and interview feedback that there is a positive effect on the symptoms patients are experiencing. There is such a large variety of different tests and measures being used both across and also within these studies though, that it makes it challenging to arrive at general conclusions from the quantitative data. This further enhances the importance of looking at the correlation of data from several different measures or outcomes within patient profiles or in relation to ongoing regular visit data or journal information.

One aspect worthy of further investigation might be to study the varying effect of frequency, consistency and duration of singing sessions e.g. exploring whether increased frequency of short singing sessions versus less frequent but longer or more intense sessions have greater value for adults with COPD. In addition, it would be of value to know whether group sessions need to be continued regularly or if bursts of sessions periodically might deliver similar results. These issues are not discussed in any of the papers in detail but exploring them could provide insight regarding the optimum duration and frequency of singing sessions, the quality of instruction and training given and the importance of repetition and continued practice at home.

Technology Advancements

The studies identified were all quite simple in design in regard to delivery of intervention and collection of data there is relatively little use of technology. There is an untapped potential to utilise technology in various ways, particularly in the area of data capture/collection but also as tools to aid in the delivery of the intervention, retain patients in group sessions or to encourage or remind continued practice either for self-management or to enhance engagement with a regular singing group. However, it is clear that this often requires investment of significant time and cost resources which is often not available in this area of research.

The use of ‘Sensewear©’ armbands for example, **(Lord et al., 2011, Lord et al., 2012)** was a very novel approach but there was no strong positive indication shown within the data. This outcome would seem to have impacted any further expansion of this technique as there appears to be little development of this technology or use of any other more sophisticated tools to capture health data in this area of research since these studies were conducted. It would also have been interesting if experiences of using such tools in this population had been explored further. There appears to be no indication of patient feedback on using this tool, the reliability or quality of the data or any measures of compliance etc.

There is some opportunity to investigate relatively low-cost options which would easily capture user data on an ongoing basis, allowing for real time analysis and reporting. Technology used for collecting diary info, health data etc. is now more widely available and diary data is not necessarily limited to collection using paper journals or specialised and expensive data capture tools e.g. e-diary solutions etc. used for large scale clinical trials.

The usage of smart phones and other similar personal devices that contain sophisticated software applications for sharing, storing and collecting healthcare data is still increasing. The reliance on patient-generated data has also been demonstrated using mobile devices to tailor diagnostic and treatment decisions as well as educational messages to support desired patient behaviours **(Roski et al., 2014)**.

There has been a significant acceleration in data collection, particularly over the last fifteen years, which has been driven by advances in technology, data science and the usage of 'big data' and data mining i.e. wider usage and accessibility to the internet, social media, smart phone technology etc. which means that more opportunities are becoming available **(Wu et al., 2014)**.

However, for health organizations to rely on this type of data, the enabling IT infrastructure has to be available. Installing such infrastructure and its components is becoming less expensive. Nonetheless, the installation still requires a significant investment of time, training and money, and it can involve lost productivity during the transition process. Organizations need to develop processes and policies that accommodate new protocols, time requirements, risk factors, and mandates for managing data too, especially in the area of privacy and security **(Roski et al., 2014)**, so this does all need to be taken into consideration.

It is recognised that novel technology maybe less widely used within the regions and populations where COPD is often seen to be more prevalent i.e. low and middle-income regions, people beyond retirement age, however further investigation into this would be important both to improve the available data and inform further research within this population.

There was a brief identification of this gap mentioned by one study paper **(Bonilha et al., 2009)** where it notes a specific deficiency of the research was that changes on respiratory pattern during the act of singing were not investigated, and the pulmonary volumes were not measured by body plethysmography. Thora3Di© is a technology which allows the measurement of abdominal and chest wall movement, using structured light plethysmography. Measurements are assessed using a grid of light beamed onto a patient's chest. Measurements can be seen real-time and instant feedback on an individual's breathing pattern can be given. Apart from traditional pulmonary measurements, this method has the ability to show traditional lung function data in a 3D format.

There is much value in exploring further options for technology and tools particularly investigating whether there might be more effective ways of establishing potential physiological mechanisms of the body during singing, particularly in the COPD population. This information could enhance the current understanding of the mechanisms of singing and the reasons why COPD patients indicate feeling positive effects in qualitative data even if the quantitative data is less robust in supporting this outcome. In turn this information has the potential to assist

development of singing training and other resources for use by singing for COPD or other health groups, with an aim to improving the overall effectiveness of the treatment.

Heart Rate Variability

Heart Rate Variability (HRV), as referenced in Chapter 1 is becoming a more commonly used measure for athletes, sport scientists, and many more of the general public simply interested in improving or monitoring their own health via personal technology. It is more widely accepted as an indicator of health with many simple personal aids with low cost phone apps and devices becoming easily available to measure improvements in HRV. There is much emerging data and research on tracking of behaviours related to negative affect and stress, based on sensors and using simple smartphone technology to calculate HRV **(Muaremi et al., 2013)**.

Heart rate variability (HRV) is a measure that is reliably used to assess the functioning of the Autonomic Nervous System (ANS) of a patient or healthy person. HRV spectral analysis is a non-invasive tool for the quantification of spontaneous HRV in normal humans **(Volterrani et al., 1994)**. Interest in HRV, as a measurement of autonomic function, lies in its clinical importance. A reduced HRV is a powerful and independent predictor of an adverse prognosis in patients with heart disease and in the general population **(Nunan et al., 2010)**.

There is increasing evidence indicating that COPD is more complex than originally thought and is not related to airflow obstruction alone. It has been recognized that COPD is a systemic disease which has been shown to negatively affect both the cardiovascular and the autonomic nerve system **(Gestel et al., 2011)**. In addition to being associated with a strength and resistance decrease, COPD is also now closely associated with the dysfunction of the ANS **(Ricci-Vitor et al., 2013)**. Preliminary study results suggest that in patients with COPD, there is an imbalance in ANS activity. This is apparently driven at rest by an increase in vagal activity and through the lack of responsiveness to sympathetic stimulation. This altered balance could also contribute to the airways obstruction in COPD **(Volterrani et al., 1994)**.

The autonomic nervous system (ANS) regulates multiple physiological processes, so dysfunctions of the ANS are recognized by the symptoms that result from failure of both sympathetic and parasympathetic components. The complexity of the physiologic basis by which autonomic dysfunction occurs in patients with COPD is considerable and the knowledge in this field is still in early stages. The insight into ‘sympathovagal’ imbalance as a pathological phenomenon in COPD may be important in understanding the pathophysiology of COPD and may have potential clinical importance for improving risk stratification and treatment of patients with COPD **(Gestel et al., 2011)**.

Singing is known to produce slow, regular and deep respiration which in turn triggers respiratory sinus arrhythmia **(Vickoff et al., 2013)** this is associated with vagal influence of HRV cycles

between singers and thus could hold significant implications for patients with a respiratory illness. It has been shown that when singing structure is regular, HRV profiles between healthy singers tends to conform in terms of frequency and phase **(Muller and Lindenberger, 2011 and Vickoff et al., 2013)**. There has been no published investigation or data to date to understand the impact of singing on this phenomenon within patients with COPD diagnosis or that are otherwise respiratorily compromised. It is an important next step to understand whether there is any impact on these same measures by a singing intervention, where the respiratory state of patients is impacted. This could be particularly important given that group singing has already been seen to impact many other aspects of these patient's health outcomes and may give insight into the various potential physiological and psychological mechanisms of action of singing on the respiratory system.

Airway Clearance

Mucus hypersecretion and chronic productive cough are the features of chronic bronchitis and chronic obstructive pulmonary disease (COPD). The overproduction and hypersecretion by goblet cells and decreased elimination of mucus are the primary mechanisms responsible for excessive mucus. Mucus accumulation in COPD patients affects several important outcomes such as lung function, health-related quality of life, COPD exacerbations, hospitalizations, and mortality **(Ramos et al., 2014)**.

Chronic cough and sputum production are common features of COPD and have a significant impact on exacerbation frequency and quality of life. Techniques to assist with removal of sputum from the airway do not have a well-defined role in COPD management though. Clinical trials of Airway Clearance Techniques (ACTs) in COPD have shown mixed results with little evidence of long-term benefit. However, many studies have failed to account for the heterogeneity of COPD lung disease. Analysis of short-term studies suggests that there may be a cohort of patients who would benefit from prescription of a sputum clearance regimen **(Holland and Button, 2006)**, though current evidence based on a systematic review in airway clearance reports that uncertainty still exists about the clinical effectiveness of airway clearance techniques for patients with stable COPD **(Wang, 2015)**.

Anecdotal evidence from interviews and observations of some of the studies within this review shows that there is an increased level of airway clearance for some COPD patients during the singing sessions and it could be hypothesised that the act of singing may potentially have similar properties to techniques used in chest physiotherapy for airway clearance e.g. chest percussion, vibration, cough stimulation. As outlined in detail previously in Chapter 1, the techniques used in singing itself follow similar scientific pathways that form the basis of successful chest physiotherapy, airway clearance, and airway clearance devices **(Goldenberg, 2018)**. Daily observations of children have also showed that during breathing exercises, playing and laughing and during FEV1 manoeuvres, mucus moved better than most of the time during

postural drainage etc. **(Schöni, 1989)**. The effect of group singing versus singing alone, would likely involve additional activities such as talking, moving and laughing with others in the group, which could enhance this mucus clearance and as such COPD symptoms.

In one study **(Bonilha et al., 2009)** the investigators observed that the patients coughed and eliminated a substantial amount of sputum just after ending the practice of vocal exercises or songs. This finding was not registered by employing a formal research protocol though. The author does highlight that 'this indicates that singing may also exhibit bronchial hygiene properties' and that the 'performance of prolonged and robust expirations has the potential to facilitate the mobilization of respiratory secretions towards the upper airways, eliciting the cough reflex'. The participants reported in another study **(McNaughton et al., 2017)** also perceived health benefits including sputum clearance from participation in the group. It is also suggested by the author that the "improvement of PEmax associated with singing could also contribute to better coughing". Singing did exhibit an increase in mean PEmax for one study **(Pacheco et al., 2014)** and their control group showed a decrease, so this certainly indicates further analysis using this measure would be interesting.

Dyspnoea Management

There was no clear positive improvement in dyspnoea symptoms shown by any of the quantitative data in the studies selected with the exception of two studies **(Trivedi, 2017, Jamaly et al., 2017)**. The first **(Trivedi, 2017)** showed significant improvement in MRC Breathlessness/Dyspnoea Scale for the experimental group over the control, however the study continued pulmonary rehabilitation in both groups for the duration, which might have a confounding effect in the construct of this review. The other study **(Jamaly et al., 2017)** showed a significant improvement for the singing intervention compared with control group where participants were assigned to a course of physiotherapy lung exercises, however further details on the quality and conduct of this study are still pending. It is clear that with such a high variation of dyspnoea scales at play and different elements of dyspnoea symptoms being captured and assessed, analysing this data collectively is very challenging.

Studies which used multiple tools to capture similar information may contain useful information to look at the consistency of responses in this regard and it could prove useful to obtain raw data for these studies to look at this specifically or build this in to future design. Comparisons of these tools could hold valuable information about the suitability of the outcome measures used within this population and potentially aid future study design. It may be helpful to obtain a consensus of the most robust tool for future studies, so that dyspnoea could be reviewed further.

Dyspnoea is one of the most important symptoms for COPD patients and has the potential to be positively impacted by singing intervention, so finding a way to improve data capture and

analysis of this data seems to be important but to date does not appear to have received the attention it might deserve.

These studies contain key information about the use and suitability of dyspnoea tools employed in this therapeutic area. It could therefore be useful to compare the outcomes of these questionnaires within this small batch of studies and potentially analyse this in further detail. This could aid and advise which tools might be the most appropriate or sensitive to utilise for future research of the impact of dyspnoea in singing for respiratory disorders.

Inflammation and Depression

Inflammation and depression are key factors in the pathophysiology of COPD and there are ongoing efforts to understand the links between the two and how these could have relevance for COPD treatment. There is an emerging recognition that inflammation may cause depression in certain subgroups of individuals. Epidemiological studies on large community samples (as well as smaller samples of medically ill individuals) have in addition demonstrated that increased inflammation serves as a risk factor for the future development of depression **(Miller and Raison, 2016).**

Singing studies in general support the hypothesis that choir singing positively influences emotions as well as immune functions in humans **(Kreutz et al., 2004)** and research so far points to the pivotal role of stress pathways in linking music to an immune response **(Fancourt et al., 2014)**. There is evidence that greater improvements in mood as a result of singing are associated with lower pro-inflammatory response **(Fancourt et al., 2016)**. In light of the importance of emerging data and the implications for singing and COPD research, it is surprising that there has been relatively little data collected for these factors within the singing for COPD studies.

Four studies **(BLF, 2017, Pacheco et al., 2014 Clift et al., 2013, Clift et al., 2017)** utilised EUROQL questionnaire which captured some depression data, although no significant findings were presented in regard to the singing intervention.

One of these **(Pacheco et al., 2014)** also utilised the HAD anxiety scale and five others **(Lord et al., 2010, Lord et al., 2012, Liu et al., 2016 McNaughton et al., 2017, Trivedi, 2017)** utilised both the HAD anxiety and HAD depression. A significant improvement was recorded by one of these studies **(Liu et al., 2016)** following a twenty-four-week singing intervention vs. control however the quality of this study is uncertain due to lack of further publication. Another study **(Trivedi, 2017)** showed a suggested significant improvement in anxiety and depression due to the singing intervention however, PR was employed throughout for both arms of the study, so this result may have been compromised. One further study **(McNaughton et al., 2017)** showed a mean reduction in HADS anxiety score observed at one year though this did

not exceed the minimal clinically important difference. This also replicated a similar result seen over a shorter period as noted by one of the other earlier controlled studies **(Lord et al., 2010)**. No other significant results were recorded using these tools in any of the other studies.

There is some investigation into depression in the studies selected but this is only via outcome questionnaires and there has been no investigation into biomarkers for depression or inflammation in any of the singing studies for COPD patients specifically. This would seem to be a considerable gap within the research available.

Health Care Utilisation, Service Use and Cost-effectiveness Considerations

One important study **(Lopez-Campos et al., 2016)** notes that the predominant health-care cost item for COPD patients is hospitalizations for exacerbations and it suggests high priority should be given to interventions aimed at delaying the progression of disease, preventing exacerbations and reducing the risk of comorbidities. This is in order to alleviate the clinical and economic burden of COPD in Western and Asian countries as studies on health-care use and economic cost to date in Asian countries, confirmed that these findings are now universal.

Health care utilisation does not appear to be captured consistently in any studies which would indicate that the current tools in use are possibly not ideal or this area is more challenging to assess. Only four studies made mention of this aspect **(Clift et al., 2013, Clift et al., 2017, Liu et al., 2016, Thomas et al., 2015)**. One small qualitative study **(Thomas et al., 2015)** mentioned that there was no difference in hospital admissions in the year after starting singing compared to the year before. A larger study **(Clift et al., 2017)** also reported a stable picture of service use though which is encouraging and a positive finding in itself. There were no other significant findings. It would be important to explore this for a longer period, across a greater number of COPD patients with differing severity of disease and particularly in randomised trial setting. It may then be possible to examine in more detail whether a singing intervention for this degenerative disease, where service use is likely to increase over time, could be seen to maintain levels of health care utilisation at a steady state. It is clearly important that this area should continue to be pursued in future research.

It would also seem important to explore the cost-effectiveness of providing singing interventions to this population, as the provision of opportunities to meet and sing together have been shown to provide cost-effective and acceptable options to maintaining and enhancing health in other populations **(Coulton et al., 2015)**, and this would provide useful data to support decisions on future policy in this area.

Systematic Review - Conclusion

Singing is a well-tolerated intervention that has a number of positive outcomes in support of patients with COPD. Singing was shown to have positive outcomes for physical activity (PCS - SF-36) **(Bonilha et al., 2009)** and improvements in lung function **(Clift et al., 2013, Pacheco et al., 2014)** which is possibly the most important clinical finding from the studies.

The overwhelming message from the collective outcomes of the systematic review and studies included is a positive one. The majority of the studies, i.e. eighteen of the nineteen studies included, reported one or more positive finding, with varying degrees of significance and none of the studies concluded any negative findings. Several of these studies **(Clift et al., 2017, Engen, 2005, Grasch, 2013, Herer, 2013, Jamaly et al., 2017)** presented both positive and neutral (no change) findings and one study **(Goodridge et al., 2013)** reported only neutral findings. However, a neutral finding in this population, with a degenerative disease, may also be considered positive in the sense of a lack of negative change in the measure i.e. an indicator of 'no change' in an outcome measure might suggest lack of deterioration. The qualitative data from this study was positive, providing a slightly positive message overall.

Only two studies **(Pacheco et al., 2014, Clift et al., 2017)** reported any negative findings. The first was due to a very small sample i.e. a single patient on one data point. The other clearly stated that some of the results were likely to have been significantly impacted due to extenuating temperature changes on the days of the assessments.

Principal significant variations with maximal oxygen uptake (VO_2) and higher arterial oxygen saturation (SaO_2) respectively **(Herer, 2013, Bonilha et al., 2009)** have also been observed, during the act of singing. These results are provided from a very small sample, but this could be interesting for further research. Patients with COPD often show slow, progressive deteriorations in arterial blood gases during the night, particularly during rapid eye movement (REM) sleep. This is mainly due to hypoventilation, while a deterioration of ventilation/perfusion mismatch plays a minor role. Current indications on treatment in this area are based on limited knowledge **(Marrone et al., 2006)** hence any investigation which could improve understanding in this area could also be useful.

There have been attempts to address the limitations of previous research **(Clift et al., 2013)** by establishing much larger groups, meeting weekly over a longer period of time. In addition to teaching good posture, breathing techniques and engaging in singing, having additional social time with others in the group and working towards combined performance events may have contributed to the more positive outcomes seen. Hence the quality, consistency and persistence of the singing intervention and delivery method may well be essential components of any effective singing intervention and such issues need to be a significant focus for future research.

The lack of standardisation and agreement of key outcome measures is apparent, and it seems important that an agreement is reached and shared among the key opinion leaders and researchers in this field so that a more consolidated and consistent approach is taken worldwide moving forward. Initial headway has been made in this area and a consensus group convened in the UK in 2016 to address a variety of issues pertaining to Singing for Lung Health **(Lewis et al., 2016)**. The group agreed that the patient-reported outcome measure, the COPD assessment test (CAT) score, is a strong candidate measure for Singing for Lung Health, both as feedback to patients and to ensure that the group is functioning effectively **(Lewis et al., 2016)**. However, further consensus needs to extend globally and encompassing other health and wellbeing measures.

This would apply to the collection tools in use, methodology for study design (e.g. inclusion and exclusions criteria) and the optimum target intervention adherence (i.e. frequency and duration of sessions). This would allow more efficient collaboration between researchers in different regions and allow more opportunity to pool and share data across multiple studies. There is also little information available regarding the health care utilisation and cost-effectiveness of singing interventions compared with other treatment options, which would be helpful to understand further and consider for future research for this intervention to aid in policy making.

The anecdotal comments and interview output provided by the qualitative data give much more positive reflections on singing interventions, not always reflected in quantitative outcome measures **(Goodridge et al., 2013)**. It was even commented in the conclusion of one study that given the subjective benefits in physical sensation described by patients the lack of change in exercise capacity is interesting i.e. comments such as “housework is no longer a struggle” **(Lord et al., 2010)**. It is crucial to continue to pursue the relevant physical data to build on these findings as anecdotal information, while encouraging, cannot be relied on to support long term solutions or policy change. It is recommended that future work might include a comprehensive systematic review of all obtainable interview output for ‘singing for adults with COPD’ and related patient comments, as this data would be much more extensive than the current quantitative data and could provide further support and direction for future work.

It is recommended that research in this area should extend to incorporate other important personal and health data i.e. further exploration of biomarkers, particularly those that provide information on the immune system, links between inflammation and depression biomarkers and the possibility of improved HRV in patients. This is vital in order to build on the previous important research in this area **(Vickoff et al., 2013, Kreutz et al., 2004, Fancourt et al., 2014)** and improve knowledge and understanding of both the physiological and psychological mechanisms of action involved in singing. The research to date indicates further investigation as to whether singing could impact these biomarkers in COPD patients would be an important step in understanding how singing positively impacts symptoms in this population specifically.

It is clearly established that the primary cause of COPD is tobacco smoke (either through tobacco use or second-hand smoke) and in 2012 it was accepted that this disease now affects men and women almost equally, due in part to increased tobacco use among women in higher-income countries **(WHO, 2017*b*)**. It is relevant to acknowledge that even with generalised, confirmed, continuous reductions in COPD standardised mortality rates, plus some recent successful anti-smoking reductions in a number of Western countries, the overarching ageing of an ever-expanding world population and other factors such as high rates of smoking in Asia specifically, this will continue to make COPD an even larger problem for years to come **(Lopez-Campos et al., 2016)**. Further global studies, particularly in both higher income and less affluent regions is therefore becoming even more desirable.

The overwhelming conclusion from the studies within this broad systematic review are in close alignment with the recent Cochrane review **(McNamara et al., 2017)** i.e. That the availability of high quality evidence that singing for COPD improves physical health, dyspnoea or respiratory-specific quality of life is still limited. This is largely due to the low number of studies and the small sample size of each study. The existence of research examining the longer-term effect of singing for people with COPD is limited and robust information from randomised controlled trials over extended periods is non-existent.

It is essential that researchers build on this existing knowledge through larger, longer, multi-centre global randomised controlled trials. These should certainly extend to provide more consistent data across diverse regions and countries with low, middle and high incomes and target more diverse COPD populations. This is where the gap in the current research lies. Key data to support these areas is fundamental, both to help determine the impact of group singing on health-related quality of life and dyspnoea, and as an established evidence base to support health improvements and policy making in the future.

Chapter 3 – Methodology

Research Study: Introduction

In order to address two of the three key research questions ‘What are the potential health and wellbeing benefits of regular participation in group singing for a small cohort of COPD patients?’ and ‘What is the potential feasibility and usefulness of developing and implementing novel singing resources for a group of COPD patients, in addition to regular group singing?’ a two-part empirical study was designed and executed. This study was of a single group quasi-experimental design.

The intent of this empirical research was to evaluate the impact of weekly group singing sessions for people with COPD including newly developed supplementary resources for promotion of self-management in this population. To meet this aim and to provide suitable input and learnings for future planning of a larger randomised controlled trial, a study was designed with two interrelated but distinct components.

The first component of the study (Part I) concerns review of the overall impact of weekly group singing sessions on health and wellbeing measures, for people with COPD, who are also using a newly developed supplementary resources. It employs a quasi-experimental pre-test post-test design which aims to answer the question ‘What has been established so far by research regarding the effects of group singing for COPD patients?’. It is also intended that this study will be a feasibility study to help to further understanding of the viability of conducting future research in this area of Kent and to potentially inform the methodology of this and future larger research study designs.

The second component of this research study (Part II) was concerned with piloting the use of newly prepared self-management resources for COPD patients and aimed to answer the question ‘What is the potential feasibility and usefulness of developing and implementing novel singing resources for a group of COPD patients, in addition to regular group singing?’

The empirical study was designed in parallel with the systematic review and the finalised design of the study followed the initial completion stages of the systematic review in late 2016. This was to ensure any important and valuable information from the conclusions of the systematic review could be used to inform the research study design. The following components were specifically informed by the systematic review.

The frequency and duration of individual sessions, as seen in the majority of those studies with positive outcomes, to be one hour of singing and exercise once every week. This was also deemed to be the simplest format and in line with the regular arrangements for community groups, so this was adopted for this study. The pattern and structure of the singing and exercise

sessions was also planned to closely follow those laid out in previous studies **(Clift et al., 2013, Clift et al., 2017).**

Participants were aged between 49-91 years old for the fifteen studies where this was presented, so since this was a wide range it was decided that no stipulations were made on age for inclusion for the design of this study, except that participants should be adults over 18 years old and date of birth would be captured as a data point.

The studies included in the systematic review were conducted across four continents i.e. North America, Latin America, Europe and Asia Pacific with the majority in Europe in temperate climates. It was considered feasible that a study in South East England would allow achievable recruitment. It was also considered most advantageous to enrol and conduct the singing groups in spring and summer seasons, similar to previously conducted studies **(Clift et al., 2013, Clift et al., 2017)** and the taster sessions and patient recruitment activities had been previously organised with this schedule in mind.

It was decided to keep the data collection simple to administer and as unobtrusive as possible to participants, both to enhance good compliance and to ensure the key focus in the time commitment for participants remained on singing and exercise activities as the priority.

It was deemed important to capture at least one health related questionnaire specific to COPD. The COPD Assessment Test (CAT) is a questionnaire designed to measure the impact of COPD on a person's life, and how this changes over time and has good measurement properties. The CAT had been used previously by two other studies included in the review. One study **(Jamaly et al., 2017)** recorded significant improvement ($p = 0.029$) in the singing intervention group and another study **(BLF, 2017)** recorded significant mean improvement of 1.1 ($p = 0.02$) post singing intervention on this measure. Since it is sensitive to differences in state and provides a valid, reliable and standardised measure of COPD health status **(Jones et al., 2009)** this measure was selected.

It was also decided that is could be important in this group of participants to measure the adequacy of asthma control in addition to the CAT and as such the ACQ was selected for this purpose. However, this was not informed specifically by the systematic review. The selection of the other wellbeing outcome measure similarly was not informed by the outcome of the systematic review directly. However, the review did reveal the importance of including a simple wellbeing outcome and the Warwick Edinburgh Mental Wellbeing Scale (WEMWBS) was selected as it had been validated for use in the UK with those aged 16 and above and commissioned by NHS Health Scotland **(Stewart-Brown and Janmohamed, 2008)** and could be implemented simply without extensive cost.

Since the qualitative outcomes measures from the studies discussed in the systematic review provided data that was shown to enhance the quantitative data for those mixed model studies

where this was collected, it was also decided to include an open text questionnaire to give participants the opportunity to make comments and provide further feedback in an unstructured way.

It would have been ideal to implement a more robust study design here, as recommended by the outcome of the systematic review however, this was not feasible at this time due to the time and resource availability so a quasi-experimental pre-test post-test design was selected.

Research evidence in healthcare can be classified according to levels of quality known as the hierarchy of evidence. There are various versions of the evidence pyramid most of which focus on showing weaker study designs (basic science and case series) at the bottom or level 5 followed by case–control and cohort studies in the middle, then randomised controlled trials (RCTs) and systematic reviews and meta-analysis at the top **(Murad et al., 2016)**. The details of the levels of this hierarchy are shown in a tabular format in table 6 below.

Level	Research Design
Level 1	Systematic Reviews or Collective Randomized Controlled Trials (High Quality)
Level 2	One Randomized Controlled Trial (High Quality) or Collective Randomized Controlled Trials (High-Low Quality)
	Prospective Controlled Trial
	Cohort
Level 3	Case Control Study
Level 4	Pre-Post Test Study
	Post Test Study
	Case Series
Level 5	Observational Study
	Clinical Consensus
	Case Reports

Table 6 - Levels of the Hierarchy of Evidence (created by author)

The research study components here are relatively small but good quality in regard to conduct. However, since the core study has a quasi-experimental pre-test post-test study design and the secondary study has a post-test design these would align with level 4 in this hierarchy.

One of the strengths of a quasi-experimental pre-test post-test design are such that it allows a study to be designed, implemented and conducted simply and conveniently, both for

researchers and participants alike, which was a key consideration for the study schedule and patient population. It allowed design to be conducted and granted ethics approval in parallel with the planning and recruitment for the singing groups, so that both could start concurrently in the spring. This allowed the advantage of conducting the study during the more favourable seasonal conditions for COPD patient recruitment and maximised the impact of the previously conducted taster sessions on enrolment into the study.

The additional strengths of a pre-test post-test design, relevant to the decision to use it in this case, were that it is relatively low cost compared to other types of study design, it can work well with surveys such as the patient reported outcome questionnaires, as selected for this study, and it can still provide a reasonable evaluation or assessment of a post intervention change. The main weaknesses are that this design is not rigorous at eliminating bias or other explanations, as is the case with a robust randomised controlled and blinded study designs and it can usually only show short term changes.

While the study design meets most of the requirements in terms of execution it clearly reduces the overall confidence in the reliability of the results that could be achieved particularly with a randomised controlled study or a design which allows some possibility of blinding the interventions, since there is strong potential for bias.

The outcome of the literature review highlights more generally that the quality, consistency, frequency, length and duration of the singing intervention and the delivery method are essential components which need to hold a more significant focus for future research and while this study could not be designed to meet these important focus points, it does aim to address the aspect of a longer term follow up period and to provide important feasibility information to planning for future larger randomised trials, which are essential in this research area.

One potential way to improve the aspects of the group singing intervention, while exploring use of low-cost, broad-reaching and more innovative approaches, might be to understand the feasibility of tools which encourage good quality home-practice or self-management, that can be used to encourage, compliment and support regular and prolonged attendance of group singing sessions.

Feasibility of Self-Management Resources

Overall, there is a growing body of evidence to show that, when compared to no intervention (i.e. standard care) self-management approaches can provide benefits for participants particularly in terms of knowledge, performance of self-management behaviours, self-efficacy and aspects of health status **(Barlow et al., 2002)**.

The findings from one study that looked at utilising a DVD specifically to improve compliance with home exercise programs **(Kingston et al., 2014)** demonstrated the multidimensional nature of compliance and showed an improved understanding of exercises when utilising home resources such as DVDs. This suggests self-help guides could be utilised as part of a programme that facilitates a patient-therapist relationship. It is considered important to trial additional resources and hypothesised that varying and/or combining the medium of delivery of this intervention may enhance the experience of group singing, provide a more versatile offering and encourage retention and continued adherence to singing exercises in the longer term.

A previous study **(McAuley et al., 2013)** provided cautiously optimistic evidence that the use of DVD-delivered programs designed for a target population can be both effective and utilized on a broad scale. This study demonstrated the feasibility, acceptability, and efficacy of delivering regularly scheduled activities focusing on older adults via the medium of DVD. This study highlighted and recognised that this type of novel intervention is also capable of reaching participants from outlying geographical areas who typically would not be able to attend centre-based activity trials conducted at university or medical centres.

The novel 'Singing for Better Breathing' (SfBB) resources created for use in this study have been developed such that they may also have other potential uses in the future to provide support to those COPD patients who might benefit from the activity of singing but are unable to attend group sessions regularly, outside their home environment due to extent of their disease or other limiting factors. It might also be speculated that it could be helpful to improve consistency of good practice between group sessions and encourage longer term continuation of singing.

COPD self-management tools

The current comparable research evidence in the area of self-management specifically in the COPD patient population is largely in regard to Pulmonary Rehabilitation (PR) and the results of several recent studies of PR home use indicate that home training may be a feasible option for COPD patients, in addition to hospital-based PR or instead of hospital-based PR where resources for this option are limited. While preliminary studies have limitations, initial reports suggest that home-based PR is safe and may improve clinical outcomes **(Holland et al., 2017, Guell et al., 2008, Puente-Maestu et al., 2000)**.

An early study **(Strijbos et al., 1996)** compared hospital outpatient and home supervised PR in groups of COPD patients with moderate to severe airflow limitation. Equal improvements were detected in exercise capacity and in Borg dyspnoea for both groups, however, whereas after the group-led sessions values tended to return to baseline outcome, after home training a further ongoing significant improvement in exercise capacity was observed, while Borg dyspnoea scores remained significantly improved over 18 months. Improvements in cycle workload and

dyspnoea score were significantly better maintained in the home supervised group as compared with hospital outpatient group. A significant improvement in wellbeing was maintained over 18 months in both rehabilitation groups. Therefore, while beneficial effects were achieved in both researchers recommended initiation of home supervision as improvements were maintained longer and are even further strengthened in this setting.

One study demonstrated that the improvement in exercise tolerance achieved by COPD patients with an unsupervised home pulmonary rehabilitation program is similar to the gains of patients in an intensive hospital-based program **(Guell et al., 2008)**. A further study showed that both hospital and home rehabilitation strategies were associated with statistically and clinically significant improvements and that the home intervention was not inferior to the outpatient intervention at 3 months and 1 year, using the primary end-point of improvement in dyspnoea **(Maltais et al., 2008)** further supporting the understanding home rehabilitation is a useful, equivalent alternative to outpatient rehabilitation in patients with COPD.

A more recent randomised controlled equivalence study in the COPD population explored the use of home-based PR and found that a home-based PR model, delivered with minimal resources, produced short term clinical outcomes that were equivalent to centre based PR. Although neither model was effective in maintaining gains at 12 months, it was concluded that home-based PR could be considered for people with COPD who cannot access centre-based PR **(Holland et al., 2017)**.

Finally, a meta-analysis of quality of life outcomes in COPD patients revealed that self-management interventions can significantly improve COPD patients' health outcomes and recommended that due to the multifaceted nature of self-management programmes the process of self-management become integrated into patients' usual care, providing patients with ongoing feedback, bolstering their support, in addition to improving their overall health and well-being **(Cannon et al., 2016)**.

While the activities and details of many of these PR activities are not directly comparable to singing self-practice, it could be speculated that singing home-practice tools and self-management techniques might be a feasible option for COPD patient population, particularly those unable to attend group singing sessions at all or unable to continue group sessions, adding to the arsenal of tools and options available to support patients with this debilitating illness.

The aim of this next aspect of the research is to further explore these themes by evaluating the feasibility of specific tools that might support the improvement of quality, consistency, frequency and length of a singing intervention. It is speculated that these tools may support by encouraging good practice and persistence between group sessions, in addition to good quality group training weekly with a singing leader.

The research aims to evaluate the use and usability of a supplementary element of self-management (in the form of DVD and booklet resources designed specifically for the COPD population) in addition to weekly group singing sessions, by evaluating data and feedback from a group of patients with respiratory illness over specified durations.

Study Objectives and Endpoints

Objectives

The primary objectives of the research were to review and evaluate the impact of weekly group singing on COPD patients' wellbeing and COPD impact using patient reported outcome measures (The Warwick-Edinburgh Mental Well-Being Scale (WEMWBS), The COPD Assessment Test (CAT) and The Asthma Control Questionnaire (ACQ-5)) and to assess feasibility of specifically created resources "Sidney De Haan (SDHR) 'Singing for Better Breathing' Resource©" (SfBB), used in conjunction within the target population using a novel questionnaire designed for this purpose. The hypothesis being, if participants attend singing for COPD groups regularly, there will be improvements noted in their health and wellbeing as reported within these outcome measures.

The secondary objectives were to assess recruitment uptake, retention and acceptability and adherence to the SfBB resources with the objective of assessing feasibility for this type of intervention in this population. Compliance data of group singing and use of resources will be collected through both group session attendance records and participant feedback questionnaires.

Primary Endpoints

- Mean total score difference from baseline of WEMWBS, CAT and ACQ-5 at Follow-Up Visit V_2, Week 12 $\pm$14 days

Secondary Endpoints

- Mean total score difference from baseline of WEMWBS, CAT and ACQ-5 at Mid-Term Visit V_1 Week 6 $\pm$14 days
- Mean total score difference from baseline of WEMWBS, CAT and ACQ-5 at Final Follow-Up Visit V_3 Week 30 $\pm$21 days
- Mean total score SFBBQ for usage and usability of SDHR SfBB resources at Follow-Up Visit V_2, Week 12 $\pm$14 days
- Summary of all written feedback, as captured in patient evaluation forms.
- Overall compliance and drop-out rate for singing groups

Research Study: Method and Design

The study was conducted following a formal protocol (See Appendix C)

Ethics and Ethics Committee Review

To ensure this research complies with the major ethical issues i.e. Informed consent, risk vs benefit to the participant, respect for anonymity, confidentiality and privacy, it is the researcher's responsibility to have prospective approval of the study design including any amendments, informed consent documents, patient information sheets and other relevant documents, from a local ethics committee. In this case the Faculty Research Ethics Committee of Canterbury Christ Church University. All correspondence with the ethics committee is retained by the researcher and the formal approval letter is noted in Appendix C, sub-appendix V.

The logistics of the 'Singing for COPD' group sessions were planned discretely from the research component, with full risk assessments carried out. The intent was that these groups with self-referred participants would be maintained and progress independently of the research itself. Therefore, the ethical considerations of the research component were specifically related to the data collection aspects i.e. capture of documented informed consent prior to the use of participants' data and respect for their anonymity, confidentiality and privacy.

One amendment was considered to continue to collect data for an additional period and this was approved by the ethics committee prospectively. The only circumstance in which an amendment to the study would be considered to be initiated prior to ethics committee approval is where a change is necessary to eliminate apparent immediate hazards to the participants. In that unlikely event, the research team would have contacted the ethics committee in writing immediately after the implementation. However, this was not required during the conduct of this study.

Participant Information and Consent

All potential participants were provided with an information sheet explaining the study (see Appendix C, sub-appendix II) and asked to sign an informed consent form (Appendix C, sub-appendix III). Personal details other than those necessary for demographic information were not requested and only data to be analysed and reported as part of the researched was collected. All data is treated as confidential and no comments from any participant in the study will be attributed to them personally in any publication.

When study data is analysed or compiled for transfer or publication the participant names, addresses, and other identifiable data is replaced by a numerical code in order to de-identify study participants. The researcher maintained a confidential list of participants who participated

in the study linking their numerical code to the participant's actual identity. In case of data transfer, the researcher will maintain high standards of confidentiality and protection of participant personal data consistent with applicable privacy laws.

The informed consent documents were in compliance with Faculty Research Ethics Committee of Canterbury Christ Church University including applicable privacy laws. The informed consent documents used during the informed consent process were reviewed by the researcher, submitted and approved by the ethics committee before use and available for inspection. The research has been carried out to ensure that each study participant is fully informed about the nature and objectives of the study and any other information associated with participation.

Ethical approval of the Informed Consent Document (ICD) and Information sheet was sought and obtained from the Faculty Research Ethics Committee of Canterbury Christ Church University. It was expected that participants were living independently and not affected by serious physical or mental frailty. However, in circumstances where participants might potentially be considered vulnerable in view of their age or any related health condition, care was taken to present information about the project with due attention to all participants needs and capabilities. It was considered that this research project did not carry any risks of harm to individuals who were invited to participate. All care was taken to ensure informed consent, risk vs. benefit to the participant, respect for anonymity, confidentiality and privacy were upheld at all times.

The researcher, or a person designated by the researcher (e.g. singing leader), obtained written informed consent from each participant, before any study-specific activity was performed. The researcher then retained the original of each participant's signed consent document and a copy of this was also provided to the participant for their information and records.

All information gathered is stored securely in the offices of the Sidney De Haan Research Centre for Arts and Health, Canterbury Christ Church University. Members of the Sidney De Haan Research Centre are the only persons having access to the primary research material. All data will be anonymised prior to analysis.

Participant Selection and Recruitment

Initial feasibility was established from patients and carers attending pulmonary rehabilitation (PR) groups conducted in the Medway area during summer months (April-July) 2016. Taster sessions were arranged for these patients at the end of their pulmonary rehabilitation sessions. Originally there were approximately eighty people that expressed an interest in participating in attending a Singing for Better Breathing (SfBB) group to be arranged in the Medway area of Kent, UK. The potential participants also expressed preference of location for attending the sessions.

They were each contacted again to confirm interest and establish the most effective locations that they could attend regularly bearing in mind the limitations of their respiratory condition. The information acquired allowed the set-up of two sites for assessments and weekly singing sessions. Total recruitment was confirmed in early spring 2017 with fifty-four potential self-referred participants, including both those that confirmed the had a respiratory diagnosis and those that considered themselves undiagnosed attendees, all identified as strong reliable candidates to join one of the two groups. Participants were self-referred based on information provided to them in confidence by their Health Care Provider (HCP) or pulmonary rehabilitation team. A decision was made to collect and analyse findings for the whole group together, for the purposes of retention and to obtain the widest possible feedback on the self-management resource tools, while focusing on those diagnosed with COPD for the analysis and reporting of health and wellbeing outcomes.

The locations and times of the weekly sessions were decided using criteria of availability at four centres identified to have appropriate facilities along with the availability of two trained singing leaders, associated with the SDHR centre in the local Kent area in South East England (UK). The sessions were selected to be conducted on a Wednesday morning at 10-11 AM (local time) at Centre A, a community centre in Gillingham, Kent (UK) and on Thursday afternoons at 1-2 PM (local time) at Centre B, a community centre in Chatham, Kent (UK).

Leaflets were also used as a recruitment resource to outreach other participants local to the selected community centres shortlisted for conducting the sessions and other attendees of local British Lung Foundation (BLF) ‘Breath Easy’ groups in the Medway area. While there was some interest, the recruitment of all eligible was from the original established NHS PR groups. This therefore ensured that all of the enrolled participants had been previously diagnosed with a respiratory illness and had attended a course of pulmonary rehabilitation (PR) prior to enrolment in the study or were attending as carers of the same.

Inclusion and Exclusion Criteria

This study was only able to fulfil its objectives if appropriate participants were enrolled. The singing groups included self-referred participants from pulmonary rehabilitation groups in the area and so the majority were diagnosed with COPD or another CRD but also included several undiagnosed participants and carers due to the needs and dynamics of the groups. The following eligibility criteria below were designed to select only self-referred participants for whom the protocol intervention is considered appropriate by their health care provider (HCP) or pulmonary rehabilitation team.

The initial enrolment time was very short due to the availability of the venues, so all candidates attending the initial two sessions were included in the evaluation, including carers that did not

have a lung condition diagnosis, but only participants meeting all of the following criteria were deemed eligible for the study evaluations:

Inclusion Criteria

- Over 18 years of age.
- Shows evidence of a personally signed and dated informed consent document, indicating that the participant has been informed of all pertinent aspects of the study and provided with an information sheet.
- Previously diagnosed with a COPD related lung condition by their Physician or Health Care Provider (HCP)
- Willing to commit to participating in the project over the course of thirteen weeks (health permitting)
- Enrolled into a singing for COPD group conducted by the Sidney De Haan Research Centre
- Physically mobile and able to travel to sessions independently or with support

Exclusion Criteria

Participants with any of the following characteristics/conditions were not included in the study though no participants meeting these criteria were identified for exclusion:

- Research site staff members directly involved in the conduct of the study and their family members, site staff members otherwise supervised by the researcher, or participants who are employees of Sidney De Haan Research Centre or Canterbury Christ Church University, including their family members, directly involved in the conduct of the study
- Participants who are physically unable to attend group singing evaluation sessions
- Participants with severe dementia or other cognitive or communication problems, which in the opinion of the researcher would render consent problematic
- Participants with ongoing or planned participation in pulmonary rehabilitation, scheduled to take place within project timescale.

Study Intervention

The intervention combined a thirteen-week programme of regular weekly group singing sessions with guided home singing practice between sessions.

Singing groups were led by two skilled and experienced singing leaders and the scheduled exercise and singing largely followed the guidance within the specially created resources provided to the participants to ensure a broadly consistent approach.

Singing sessions were weekly during academic term time, from May 2017 to July 2017 with a break during the summer starting again from September 2017 to November 2017. Sessions were held in two community halls booked specifically for the purpose. They were private and afforded acceptable levels of comfort (heating, light, ambient sound, refreshment making facilities, etc.). One had close integral car parking and the other had roadside parking with access to public transport and both had flat access to the hall on the ground floor.

Sessions were a total of one hour with thirty minutes available either side of the session for setting up, socialising, ‘meeting and greeting’ and clearing away after singing. The sixty-minute singing session commenced with twenty minutes of relaxation, posture, breathing and vocal exercises followed by forty minutes of singing. Participants also steered the musical direction of song choices for their group according to their interests, so that the programme was enjoyable, stimulating but consistent across both groups. Songs were taught by ear and were sung mainly without accompaniment.

The feasibility study was designed with the aim of enrolling and consenting thirty patients across the spectrum of COPD severity to attend one of two research ‘singing for COPD’ groups in Medway in Kent. This was to primarily explore the impact of group singing and to obtain feedback on two specially created resources. The rationale was to collect initial thoughts and comments on booklet and DVD resources produced specifically for home use created for potential use with future singing for COPD groups and projects.

The intervention aimed to employ skilled singing facilitators and a planned programme of group singing and singing-related exercises run weekly over a period of thirteen weeks ensured standardisation of delivery throughout the trial.

Participants were also provided with the SfBB resources in addition, at their baseline visit and asked to trial these at home between group sessions. Participants were involved in baseline data assessment before the start of the singing groups and after the first seven weeks of intervention. Follow-up data collection then took place thirteen weeks after the intervention period of the trial to provide the key assessment data. Group singing was then halted for several weeks over the summer period and initiated again for a further 12 weeks, at which point a final

follow-up assessment was completed to allow for additional longer-term follow-up data to be analysed. The intent was then that the sessions might continue depending on interest, funding and availability of venue, singing leader etc.

The structured singing during group sessions closely followed the guidance provided in the resource booklet (A summary of the songs provided for singing practise are noted in Appendix C, sub-appendix IV) and hard copies of the booklet containing full instructions and an accompanying DVD were provided to all participants. Guidance was given for participants to follow the resource instructions at home outside the group sessions and 'self-administer' the intervention according to the guidance. The intent was to provide limited information verbally about the resources, so as to assess the quality of the instruction and content. This evaluation was planned to occur at the Follow-Up Visit V_2, twelve weeks post-baseline.

Allocation to Intervention

A single, open non-randomised unblinded design was employed so participants were recruited from willing volunteers into newly established self-referred community singing groups with approximately thirty to forty participants recruited across the groups. Participants were assigned a number sequentially by the principal investigator and the participants DOB and postcode was used as a primary key for database analysis and quality control the case report forms collected at each visit point.

The investigators knowledge of treatment intervention did not influence the decision to enrol a particular participant or affect the order in which participants are enrolled as the study is open.

Study Procedures

Study procedures carried out at each visit are outlined below and specific questionnaire evaluations are noted in further detail in the following section.

Baseline Study Visit - V_0 Week 0 ($\pm$7 days)

- Information Sheet and Informed Consent
- Demography Questionnaire
- Warwick-Edinburgh Mental Well-Being Scale (WEMBMS)
- The COPD Assessment Test (CAT)
- Asthma Control Questionnaire (ACQ-5)
- Initial General Feedback Questionnaire
- Distribution and explanation of SDHC SfBB Booklet/DVD resource

Mid-Term Study Visit V_1 - Week 6 (±14 days)

- Warwick-Edinburgh Mental Well-Being Scale (WEMBMS)
- The COPD Assessment Test (CAT)
- Asthma Control Questionnaire (ACQ-5)

Follow-Up Study Visit V_2 - Week 12 (±14 days)

- Warwick-Edinburgh Mental Well-Being Scale (WEMBMS)
- The COPD Assessment Test (CAT)
- Asthma Control Questionnaire (ACQ-5)
- SfBB self-help resource feedback questionnaire
- Final General Feedback Questionnaire

Final Follow-Up Study Visit V_3 - Week 30 (±14 days)

- Warwick-Edinburgh Mental Well-Being Scale (WEMBMS)
- The COPD Assessment Test (CAT)
- Asthma Control Questionnaire (ACQ-5)

A flow diagram of the study visits is noted in figure 3 below.

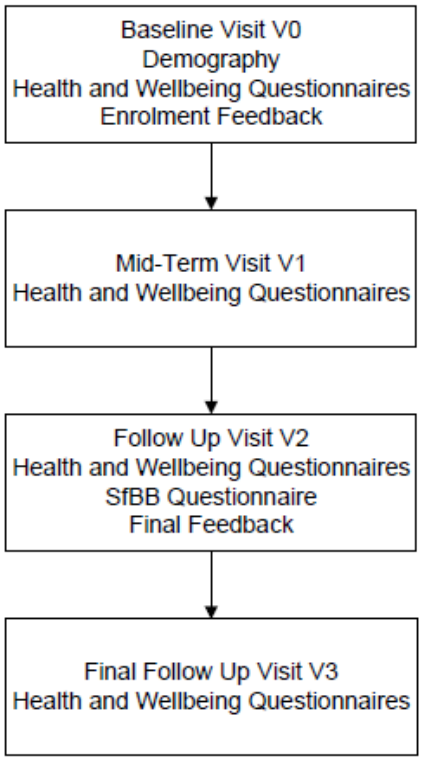

Figure 3 - Research Study Design: Flow Diagram for Study Evaluations

Measures and Data Collection

The selection of research forms and measures was guided by a variety of rationale and a description of each is noted in more detail below. The study was conducted within a community setting and the aim was to ensure measures and data could be collected in the most simple and unobtrusive way. The measures and means used were required to be acceptable to participants and conducted within the confines of a community setting and the time available i.e. Lack of access to specialist or diagnostic equipment (e.g. weight scales, spirometers) or trained personnel etc.

Questionnaires

Demography Questionnaire

The demography questionnaire was a short form created for collection of key demography data as supplied by the participant i.e. Gender, Date of Birth, Respiratory diagnosis and Length of time affected by lung problems? It also included questions regarding smoking status and previous group singing experience.

It also included a request for the participants' postcode, which was specifically to be used in association with their data of birth to provide a unique anonymised identifier (primary key) for matched analysis of samples within the dataset. These two critical data points (date of birth and postcode) were further collected at each subsequent visit and retained with the participants research form.

Warwick-Edinburgh Mental Well-Being Scale (WEMWBS)

The Warwick Edinburgh Mental Wellbeing Scale (WEMWBS) is a validated questionnaire assessing positive mental wellbeing and aims to measure mental well-being and not the determinants of mental well-being. It comprises a fourteen-item scale that relates to an individual's state of mental well-being (thoughts and feelings) within the previous two weeks. It is a scale of mental well-being covering subjective well-being and psychological functioning, in which all items are worded positively and address aspects of positive mental health. The scale is scored by summing responses to each item answered on a 1 to 5 Likert scale where the minimum scale score is 14 and the maximum is 70. WEMWBS has been validated for use in the UK with those aged 16 and above **(Stewart-Brown and Janmohamed, 2008)**. The Warwick-Edinburgh Mental Well-being Scale was funded by the Scottish Government National Programme for Improving Mental Health and Well-being, commissioned by NHS Health Scotland, developed by the University of Warwick and the University of Edinburgh, and is jointly owned by NHS Health Scotland, the University of Warwick and the University of Edinburgh.

The COPD Assessment Test (CAT)

The COPD Assessment Test (CAT) provides clinicians and patients with a simple and reliable measure of overall COPD-related health status for the assessment and long-term follow-up of individual patients. It consists of eight items, each formatted as a numerical response scale from zero to six, making the tool easy to administer and easy for patients to complete. The items cover a wide range of disease severity, with the intention that the greatest discriminant power would be in the mild to moderate range **(Jones et al., 2009)**.

Asthma Control Questionnaire (ACQ-5)

The Asthma Control Questionnaire (ACQ-5) is an asthma control measurement tool consisting of five questions on symptom control; each of the questions is scored on a scale of 0–6 where 0 represents excellent asthma control and 6 represents extremely poor control **(O'Byrne et al., 2010)**. The ACQ-5 can include responses to seven questions, five relating to symptoms, one relating to rescue symptom use and one to capture FEV1. The shortened, five-question 'symptom only' questionnaire is considered just as valid and is adequate for the purposes of this study.

The quantitative variables for all feedback from each of the above questionnaires will be expressed as mean values with standard deviation (SD). A comparison of the mean scores of each particular question within the questionnaires, as well as the mean overall scores for each questionnaire will be completed using an appropriate two-way statistical test against the null hypothesis. A *p*-value less than 0.05 will be considered as statistically significant. Statistical analysis will be performed using the Statistical Package for the Social Sciences (SPSS) version 24 for Windows.

SfBB Questionnaire (SfBB-Q)

A simple SfBB questionnaire was developed with the sole purpose of gaining quantitative feedback on the SFBB resources from participants in the study. The design is similar to the format of the CAT questionnaire but consists of eight items, each formatted as a numerical response scale from zero to six, making the tool simple to administer and easy for participants to complete. The items covered various questions regarding the usability of the tools, frequency of use and whether the participants would use these or similar tools in the future.

The eight items were as follows:

1. I used the booklet and/or DVD regularly
2. I found the instructions in the booklet easy to follow
3. I found the instructions on the DVD easy to follow
4. Using the booklet and DVD made me feel more confident at the group sessions

5. It was easy to fit in regular practice at home
6. I will continue to use the booklet and DVD to practice
7. I would recommend the booklet and DVD to others
8. I would be likely to use this or other singing resources online or via an electronic device, mobile phone ‘app’ or similar

Data Collection and Statistical Analysis Methods

Sample Size Determination

Sample size is determined based on sample size calculated for two prior outpatient pulmonary rehabilitation studies **(Bakarat et al 2008, Mendes de Oliveira et al 2010)** where total patients recruited into the pulmonary rehabilitation home resources cohort was 19-23. The aim for this study was to enrol and consent at least n=30 participants over two groups with conservative anticipated drop-out rate of 30-50% allowing for 15-21 participants or over, completing the full evaluation.

It would have been advantageous to recruit a larger number of patients if possible to increase the power which is low but since this is a pilot study and resources are limited i.e. opportunities for wide reaching recruitment options, availability of skilled singing leaders and venues, the aim of thirty participants is deemed reasonable given that this will still provide important feasibility and proof of concept input for planning of a sufficiently powered larger trial in the near future.

Data Capture and Electronic Data Records

The term Case Report Form (CRF) is understood to refer to either a paper form or an electronic data record or both. A Case Report Form is required and completed for each included participant. The completed original CRFs are the sole property of the Sidney De Haan Research Centre and should not be made available in any form to third parties, except for authorised representatives of the Sidney De Haan Research Centre or appropriate, without written permission from the researcher.

The researcher has ultimate responsibility for the collection and reporting of all data entered on the questionnaires and any other data collection forms, ensuring that they are accurate, authentic/original, attributable, complete, consistent, legible, timely (contemporaneous), enduring and available when required. The participant records must be signed by the researcher to attest that the data contained therein is true. Any corrections to entries made in the source documents are dated, initialled and explained (if necessary) and do not obscure the original entry.

Data Handling Conventions are utilised, documented and applied to missing or inconsistently recorded data for the purposes of final statistical analysis.

Data Handling and Record Keeping

Data for this feasibility study was collected as raw source documentation. This consisted of paper questionnaires (See protocol Appendix C, sub-appendix I) which were provided and self-administered by patients and the data then manually entered into a database by the researcher. This data was entered in real time and cleaned on an ongoing basis for processing, analysis and delivery as a final study results report. The data handling conventions were documented prospectively as recorded in this report.

It is advised that use of estimations to fill in missing values for the WEMWBS should only be done in situations where at least a certain proportion of items are answered. If less than this proportion has been answered the respondent's score should be set to missing **(Stewart-Brown and Janmohamed, 2008)**. This assumption was followed for all three scale measures (WEMBMS, CAT and ACQ-5) and if more than one question within any questionnaire or questionnaire domain was missing, the score was set to missing (i.e. 999) within the dataset. If only one question or statement response was missing or left unanswered then the missing response was imputed with the questionnaire or domain-specific mean based on the remaining answered question scores.

Statistical Analysis

It was planned to use the paired t-test to compare means of two related observations i.e. a comparison of given values both before and after an intervention and recorded in pairs per participant. The paired t-test was used due to the small sample sizes to compare mean differences. The difference between the paired values is assumed to be normally distributed and the null hypothesis, that the expectation is zero, is tested by paired t test.

The test is used to compare the means for all three health and wellbeing evaluations over each possible interval i.e. between V_0 and V_1, V_1 and V_2, V_2 and V_3, V_1 and V_3, V_0 and V_2 and V_0 and V_3 for both the total score and individual question scores. Those with values of significance i.e. $p < 0.05$ are reported.

The p-value produced from the paired t-test indicates the likelihood of the observed difference under the null hypothesis. The substantive significance i.e. what the findings indicate about population effects themselves will be assessed using a comparison of the mean difference expressed as meaningful for each of the individual health and wellbeing scores as noted from previous validations of each one.

Record Retention

To enable evaluations and/or audits from regulatory authorities all records will be kept records, including the identity of all participating participants (sufficient information to link records) and all original signed informed consent documents, copies of all CRFs, safety reporting forms, source documents, and detailed records of treatment disposition, and adequate documentation of relevant correspondence (e.g., letters, meeting minutes, telephone calls reports). The records should be retained by the research team according to International Conference on Harmonisation (ICH) or according to local regulations, whichever is longer.

All information gathered will be stored securely in the offices of the Sidney De Haan Research Centre for Arts and Health, Canterbury Christ Church University. Members of the research team are the only people who will have access to the primary research material. All data will be anonymous prior to analysis.

If the research team becomes unable for any reason to continue to retain study records for the required period (e.g., retirement, relocation) the study records must be transferred to a designee, such as another institution, or to an independent third party. Records must be kept for a minimum of 15 years after completion or discontinuation of the study or for longer if required by applicable local regulations.

Publication of Study Results

Publication of study results will be at the discretion and agreement of Canterbury Christ Church University, the Sidney De Haan Research Centre and the researcher.

Publications relating to the study will comply with Canterbury Christ Church University requirements and recognised ethical standards concerning publications and authorship, including Section II - “Ethical Considerations in the Conduct and Reporting of Research” of the Uniform Requirements for Manuscripts.

Chapter 4 – Results and Discussion – Part I: Core Study

Research Study: Results and Data Analysis Part I

Study Enrolment and Demography

Total recruitment was confirmed as fifty-four potential self-referred participants, including those that confirmed a Chronic Respiratory Disease (CRD) diagnosis (COPD and non-COPD) and those that considered themselves undiagnosed attendees. All were identified as strong, reliable candidates, available for the requested time frame and willing to join one of two singing groups.

Thirty-eight of these participants attended an initial singing group session at one of the two selected community centres in the Medway area of Kent in the UK i.e. Centre A, a community centre in the centre of Gillingham, Kent (UK) and Centre B, a community centre in the surrounding area of Chatham, Kent (UK). A total of thirty-seven participants, were assessed for inclusion and provided their informed consent for enrolment in the study across the two newly formed singing groups.

In total, twenty-eight of the total thirty-seven participants that completed assessments at Baseline Visit V_0 were followed up and completed Mid-term Visit V_1 and Follow-Up Visit V_2. Twenty of these original participants also continued attending sessions up to thirty weeks post Baseline and were followed up at the Final Follow-Up Visit V_3. The demographic details of all participants are given in table 7 below.

Diagnosis	Total	Consented	Male	Female	Mean Age	V_0	V_1	V_2	V_3
CRD - COPD	26	26	11	15	67.65 $\pm$8.7	26	23	21	17
CRD - Non-COPD	7	7	0	7	63.1 $\pm$7.1	7	4	4	1
Undiagnosed	5	4	1	3	62.5 $\pm$7.7	4	3	3	2
Total	38	37	12	25	66.24 $\pm$8.4	37	28	28	20

Table 7 – Research Study Part I: All Participants – Demography, Enrolment and Assessment Visits Summary

Research Study: Sample for Health and Wellbeing Evaluation

Participants were only considered for inclusion in the health and well-being outcome analysis if they strictly met all protocol inclusion criteria, including adherence to inclusion criteria #3 having '*Previously been diagnosed with a COPD related lung condition by their Physician or Health Care Provider (HCP)*'.

Participants included in the health and wellbeing assessments were also required to complete at least Baseline Visit V_0 and Follow-Up Visit V_2 to be included in the Follow-Up Visit V_2 evaluation group. Similarly, participants were required to complete at least Baseline Visit V_0 and Mid-term Visit V_1 for inclusion in the Mid-term Visit V_1 evaluation group and at least Baseline Visit V_0, Follow-Up Visit V_2 and Final Follow-Up Visit V_3 for inclusion in the Final Follow-Up Visit V_3 evaluation group.

Twenty-six participants indicated that they had previously been diagnosed with a COPD related lung condition by their HCP and were confirmed as meeting all inclusion criteria for the study protocol (Appendix C). These participants were considered as the 'per protocol' (PP) sample group and only participants from this group were assessed to evaluate the primary endpoint of the study and all health and wellbeing endpoints.

A flow diagram to illustrate the number of participants at each assessment up to Follow-Up Visit V2 is shown in figure 4 below.

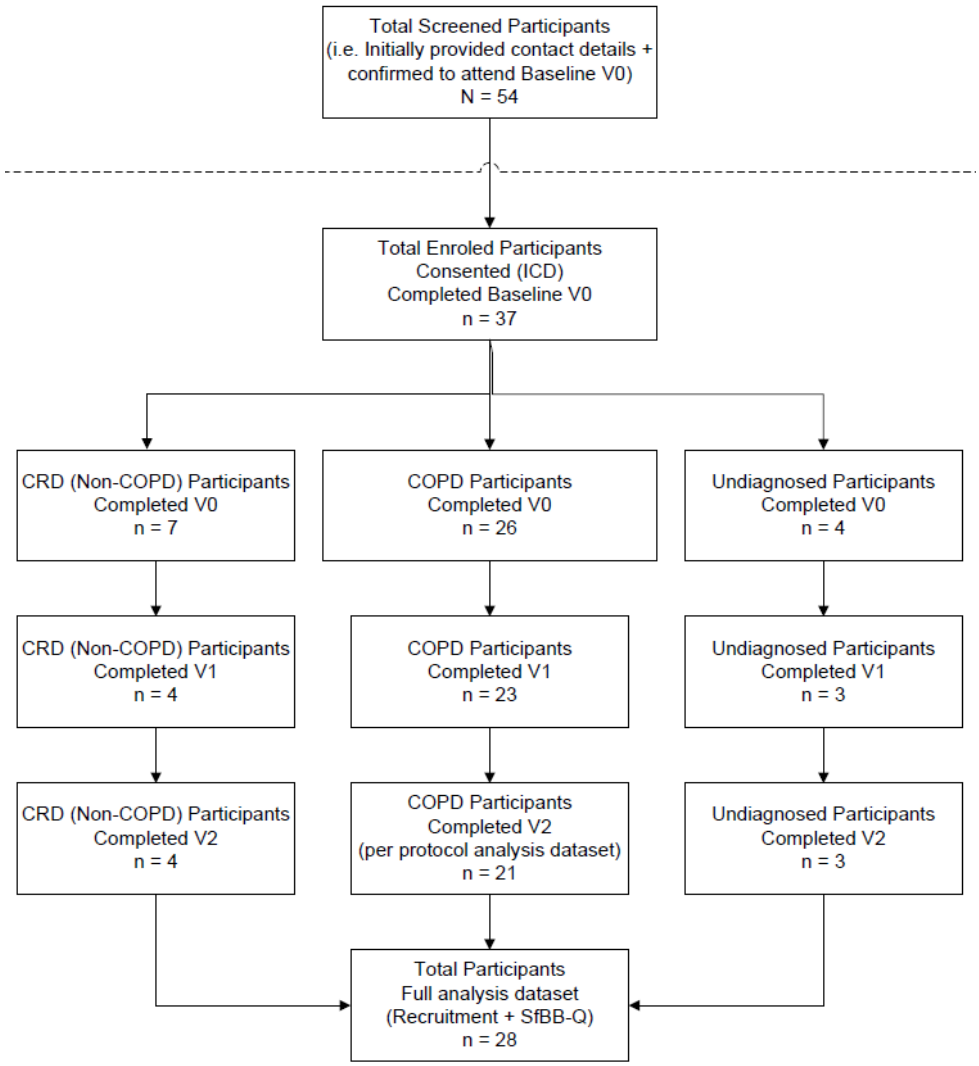

Figure 4 - Research Study Results Part I: Demography - Flow Diagram of Participants from Baseline Visit V_0 to Follow-Up Visit V_2

Twenty-one of these COPD participants continued with regular weekly participation up to Follow-Up Visit V_2 and provided evaluations at Baseline Study Visit V_0 (Week 0 $\pm$7 days), Mid-Term Study Visit V_1 (Week 6 $\pm$14 days) and Follow-up Study Visit V_2 (Week 12 $\pm$14 days) assessment points, having attended at least 75% (n=13) of all group sessions (i.e. ten or more of the total of thirteen possible group sessions) within this time frame. This group comprised thirteen females and eight males with a mean age of 66.62 $\pm$9.3 years. Five of these participants (four females and one male) were enrolled at the Centre A and sixteen of these participants (nine females and seven males) were enrolled at Centre B. A summary of the enrolment numbers and visits completed is shown in table 8 below.

Diagnosis Group	Total	Consented	Evaluable	Male	Female	Mean Age	V_0	V_1	V_2	V_3
COPD	26	26	21	8	13	66.62 $\pm$9.3	26	23	21	16

Table 8 – Research Study Part I: COPD Participant Enrolment and Completion of Visits

The data collected from these participants were assessed to evaluate the primary endpoint of the study i.e. the 'Mean total score difference from baseline of WEMWBS, CAT and ACQ-5 at Follow-Up Visit V_2, Week 12 +14 days and an additional secondary endpoint concerned with health and wellbeing i.e. 'Mean total score difference from baseline of WEMWBS, CAT and ACQ-5 at Mid-Term Visit V_1 Week 6 +14 days. A flow diagram to illustrate the number of participants at each assessment is shown in figure 5 below.

All participants were then given the option to return, following a six-week hiatus during the summer months, to continue singing in a single group comprising all participants from the original two separate groups. These were combined for logistical purposes at a single location, Centre B, led by the singing leader from the original Centre B group. The group sessions continued to be conducted weekly from September to November 2017. Sixteen of the original twenty-one COPD (PP) participants evaluated at Follow-Up Visit V_2 and were further evaluated at Final Follow-Up Visit V_3 after returning to join the combined singing group.

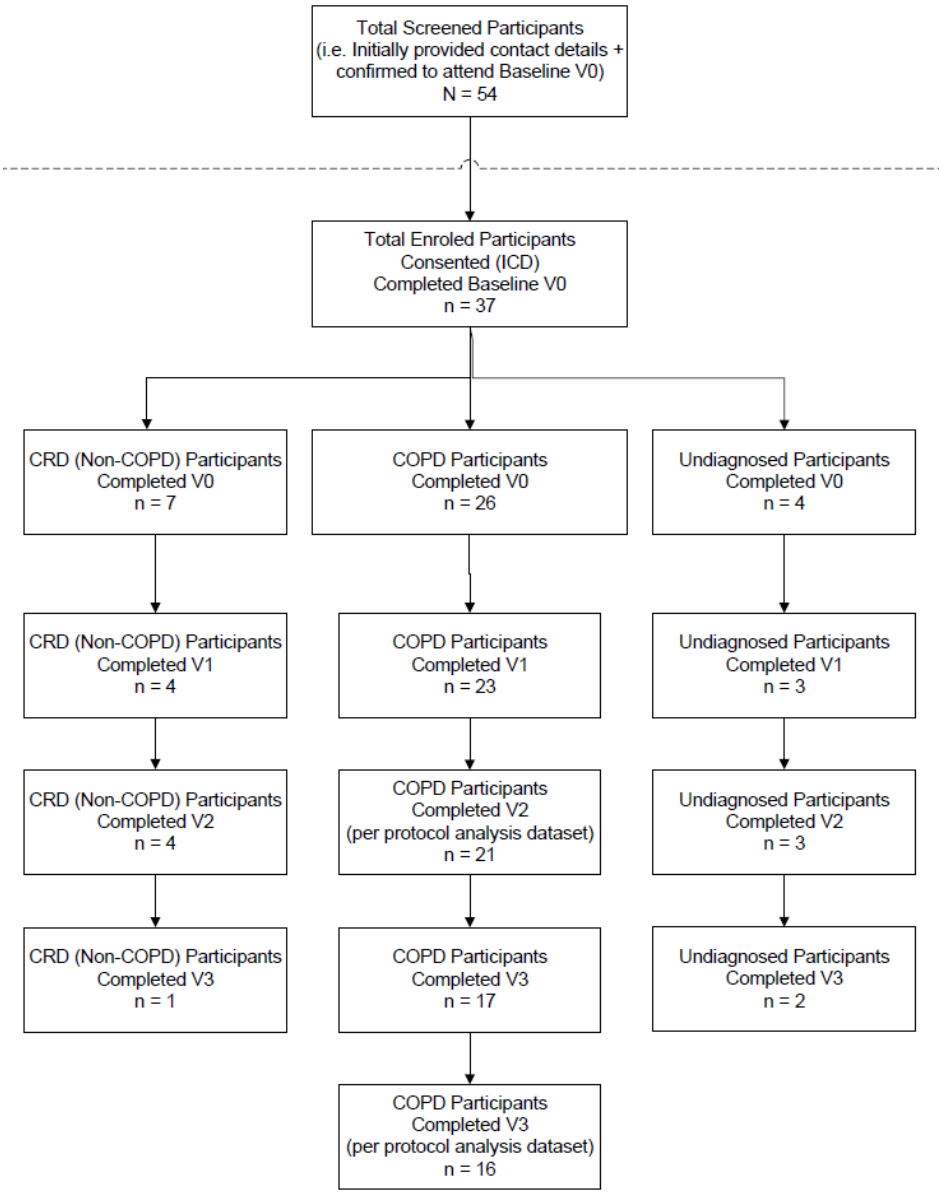

Figure 5 - Research Study Results Part I: Flow Diagram up to Follow-Up Visit V_3

Health and wellbeing data collected from the sixteen per protocol participants was further assessed at Final Follow-Up Visit V_3 Week 30 from Baseline Visit V_0 to evaluate a further secondary endpoint which was also concerned with health and wellbeing i.e. 'Mean total score difference from baseline of WEMWBS, CAT and ACQ-5 at Final Follow-Up Visit V_3 Week 30 +21 days'.

NB One additional participant was evaluated at this visit, but their data was excluded from the 'per protocol' analysis set as they did not fully comply with the criteria. Details are noted in the next section.

Study Enrolment and Recruitment

Participants were self-referred and contacted via outreach to pulmonary rehabilitation and other self-help breathing groups in the Medway area in Kent (UK) during the year prior to the study. Initial feasibility was established from respiratory patients and carers attending pulmonary rehabilitation (PR) groups conducted in the Medway area during summer months (April-July) 2016. Taster sessions were arranged for these patients at the end of their PR sessions and approximately eighty potential candidates from these sessions expressed an interest in participating.

All candidates were further contacted and a total of fifty-four participants agreed to attend on one of the two initial dates at the selected two centres and of these, thirty-eight participants attended an initial session. There was an overall drop-out rate of 26% for the whole population (n=37) by Follow-Up Visit V_2 compared to a drop-out rate of just 19% for participants with a COPD diagnosis alone (n=26). Similarly, there was an overall drop-out rate of 47% for the whole population (n=37) by Final Follow-Up Visit V_3 compared to just 38% for participants with a COPD diagnosis (n=26).

Patients were asked to estimate the number of years since they were first diagnosed with their respiratory illness and out of a total of thirty-three diagnosed participants that provided this information, there were fifteen respiratory patients diagnosed >10 years ago of which twelve were diagnosed specifically with COPD. Eighteen respiratory patients indicated they had received a diagnosis within the last 10 years of which fourteen of these were diagnosed with a COPD-related condition.

Only one participant indicated that they were also involved in another singing group, however this participant (#2021) dropped out of the initial assessment due to ill health. They returned for the Final Follow-up V_3 assessment but had not completed >2 of the original group sessions between baseline visit V_0 and Follow-Up Visit V_2, so they were deemed to be ineligible for inclusion in the per protocol (PP) sample. The data for this participant were therefore excluded from all health and wellbeing analyses.

Attendance rates overall were very high. All twenty-eight participants that completed a Follow-Up Visit V_2 had an attendance rate of >60% up to that visit. A total of 66% of all these participants (n=28) had an attendance rate >85% (n=13) which further increased to 81% for those twenty-six participants that were diagnosed with a COPD-related respiratory illness.

Health and Wellbeing Outcomes

The primary endpoint of this research study is the evaluation of the impact of the group singing intervention on health and wellbeing scores total score for WEMWBS, CAT and ACQ from

baseline evaluation (Baseline Visit V_0) to the twelve-week follow-up evaluation (Follow-Up Visit V_2). Secondary endpoints included evaluation of these same scores at additional six-week mid-term (V_1) and thirty-week follow-up (Final Follow-Up Visit V_3) time points.

The null hypothesis in each case is that 'no change occurred over time on these measures' such that a beneficial impact of the singing intervention over time is assumed as the alternative hypothesis. If this were the case, then improvements would be recorded. A two-tailed criterion was employed to test the null hypothesis to allow for the possibility of a decrease in mean score indicating a decline in participants' health or wellbeing which is likely in degenerative conditions such as COPD.

Health and Wellbeing Outcomes – Total Scores

Paired t-tests were conducted on the total scores for the available sample of patients at every visit on for each one of the health and wellbeing outcome scales to explore any potentially significant differences between the full range of visits including Baseline to Mid-term, Baseline to Follow-up, Baseline to Final Follow-up and also Mid-term to Follow-up and Follow-up to Final Follow-up i.e. V_0 to V_1, V_0 to V_2, V_0 to V_3, V_1 to V_2 and V_2 to V_3. The full results of all analysis combinations are listed in the appendices (Appendix D) and the most significant results are summarised as follows.

Paired Samples Test - Change from Baseline Visit V_0								
	Paired Differences							
				95% Confidence Interval of the Difference				
	Mean	Std. Deviation	Std. Error Mean	Lower	Upper	t	df	Sig. (2-tailed)
$WEMV_0$ - $WEMV_1$	-2.836	7.316	1.525	-6.000	0.327	-1.859	22	0.076
$WEMV_0$ - $WEMV_2$	-6.095	10.720	2.339	-10.975	-1.216	-2.606	20	0.017
$WEMV_0$ - $WEMV_3$	-8.380	9.362	2.340	-13.368	-3.392	-3.581	15	0.003
$CATV_0$ - $CATV_1$	2.156	6.451	1.375	-0.704	5.016	1.567	21	0.132
$CATV_0$ - $CATV_2$	4.755	7.828	1.708	1.192	8.319	2.784	20	0.011
$CATV_0$ - $CATV_3$	5.036	7.001	1.750	1.305	8.766	2.877	15	0.012
$ACQV_0$ - $ACQV_1$	1.600	3.979	0.890	-0.262	3.462	1.798	19	0.088
$ACQV_0$ - $ACQV_2$	2.000	4.449	0.995	-0.082	4.082	2.011	19	0.059
$ACQV_0$ - $ACQV_3$	2.000	4.648	1.162	-0.477	4.477	1.721	15	0.106

Table 8 – Research Study Part I: Summary of T-Test Analysis Results for All Health and Wellbeing Questionnaires – Change in Total Score from Baseline Visit V_0

There was a wide range of results for total score across all three health and wellbeing questionnaires, particularly at V_0 where CAT scores ranged from 4 to 38, WEMWBS ranged from 27 to 70 and ACQ ranged from 2 to 21. There are considerable variations in the reported health outcomes levels for this group of patients, which would be expected due to the different stages of disease and length of time of diagnosis which was reported as anything from 2 years to 71 years.

The most important findings were those pertaining to the primary endpoint of the study, which are reflected in the analysis of the WEMWBS and CAT scores between Baseline Visit V_0 and Follow-Up Visit V_2 at Week 12 occurring between May to July 2017 along with the results of the subsequent Final Follow-Up Visit V_3 scores at week 30.

There were statistically significant changes recorded in both total mean WEMWBS and total mean CAT scores from Baseline Visit V_0 to Follow-Up Visit V_2 i.e. $p < 0.05$. There was an increase in the mean WEMWBS scores between Baseline Visit V_0 (M=49.14, SD=12.397) and Follow-Up Visit V_2 at Week 12 (M=55.23, SD=10.150); $t(20)=2.606$, $p = 0.017$ and decrease in the mean CAT score between Baseline Visit V_0 (M=25.03, SD=7.774) and Follow-Up Visit V_2 at Week 12 (M=20.27, SD=10.659); $t(20)=2.784$, $p = 0.011$ for the twenty one 'per protocol' COPD participants across the two singing groups combined.

Participants were also given the option to continue in a single singing group composed of the original two separate groups. These were combined at a single location (Centre B) with the singing leader from the original Centre B group. These group sessions were conducted weekly from September to November 2017 and included a smaller subgroup of sixteen per protocol patients (five participants less than the original total) returning to join the combined singing group, following a six-week hiatus during the summer.

The mean WEMWBS score for those participants that were assessed at the Final Follow-Up Visit V_3 at Week 30 support a finding that the effect from Follow-Up Visit V_2 at Week 12 was maintained for this outcome measure up to Final Follow-Up Visit V_3 i.e. there was a significant difference in the mean WEMWBS score between V_0 (M=48.43, SD=12.886) and V_3 (M=56.81, SD=8.719); $t(15)=3.581$, $p = 0.003$. In fact, this suggested a further slight overall increase in mean WEMWBS score of ~2.285 between Baseline Visit V_0 to Follow-Up Visit V_2 and Baseline Visit V_0 to Final Follow-Up Visit V_3.

The mean CAT score for those participants that were assessed at the Final Follow-Up Visit V_3 at Week 30 also showed a similar finding in that the effect from Follow-Up Visit V_2 at Week 12 was maintained for this outcome measure up to Final Follow-Up Visit V_3 i.e. CAT score between Baseline Visit V_0 (M=24.16, SD=7.282) and Final Follow-Up Visit V_3 (M=19.13, SD=9.563); $t(15)=2.877$, $p = 0.012$. Similar to the results for the WEMWBS scores, this also revealed a further slight overall improvement in mean CAT score of ~0.281 between Baseline Visit V_0 to Follow-Up Visit V_2 and Baseline Visit V_0 to Final Follow-Up Visit V_3.

There was an improvement in total mean ACQ-5 score between Baseline Visit V_0 and Follow-Up Visit V_2 (Week 12) but this did not achieve statistical significance (p = 0.059). The results for the change in total mean ACQ-5 score showed no significant results for overall score at any stage. A demographic analysis was conducted to understand if there were any changes in mean total score difference between Baseline Visit V_0 and the Follow-Up Visit V_2 with regard to either age or gender.

A Pearson correlation coefficient analysis of the correlation between age and the three scale scores was undertaken, but no significant strong correlations were seen for any of the three Health and Wellbeing scales. There was a weak non significant negative correlation detected between participant age and change in WEMWBS score (r = -0.499 p = 0.21) and weak positive correlation detected between age and change in the CAT score (r = 0.250 p = 0.274) and the ACQ score (r = 0.063 p = 0.274) but with such a small number of participants this was negligible.

Independent t-tests were conducted to compare for the total change in scores for the available sample of patients on each of the health and wellbeing outcome scales against participants' gender, to explore any potentially significant differences (see Appendix D). There was no significant difference in the change in WEMWBS scores for males (M=2.88, SD=7.72) and females (M=8.08, SD=12.07); t (19) = -1.085, p = 0.292, in the change in CAT scores for males (M=-5.66, SD=11.86) and females (M=-4.20, SD=4.38); t (8) = -0.335, p = 0.746 or in the change in ACQ scores for males (M=-3.14, SD=6.176) and females (M=-1.38, SD=3.33); t (18)= -0.836, p = 0.414 between the Baseline Visit V_0 and the Follow-Up Visit V_2.

The results suggest there is no real effect of age or gender on the difference of any of the health and wellbeing scales between Baseline Visit V_0 and the Follow-Up Visit V_2.

Weekly Progress – Singing Leader Feedback

General ad hoc feedback and observations from the two trained singing group leaders and the researcher indicated that participants were already starting to interact quite quickly and mix with each other, exchanging numbers and socialising very early on in the first sessions. They generally indicated they were having fun and felt relaxed, and that as time went on friendships formed and additional social bonds expanded e.g. participants started arriving earlier than the session start just to meet for a coffee and social catch-up prior to singing, bringing additional refreshment items, sharing home grown produce, exchanging contact details to connect outside the group sessions.

The ongoing ad hoc feedback from participants indicated they were becoming more confident in the groups, were finding singing easier and were noticing the difference to their breathing as

early as the fourth weekly session. An increasingly common theme of discussion within both groups from the second or third sessions onwards was regarding how the groups could be continued and sustained beyond the research project. This became more important with time as the groups became more cohesive.

General anecdotal comments and feedback regarding how the group was progressing, socialising and interacting were also collected weekly in a diary format by one of the singing leaders to share with the researcher, including a selection of general unsolicited participant comments. These details provide support to the observations above and they help provide further understanding of progress, participant acceptability to attending the groups and other tacit information not collected as part of the study research e.g. challenges faced by participants regarding venue logistics regarding participants with respiratory conditions. They also provide supportive information to the observations noted anecdotally above and to the quantitative results.

(A high-level summary of these comments made to the singing leader at the Centre A group, recorded verbatim and ad hoc, are noted in Appendix E).

General Participant Feedback: Group Session

Baseline Visit V_0 – Enrolment Feedback Comments

Participants were asked to provide open text feedback comments at Baseline Visit V_0 (Appendix C, sub-appendix I). The questions were related to how the participant had heard about the sessions, specifically whether it was from their GP and they were asked to advise what they hoped to gain from these sessions i.e. Questions asked were '*How did you hear about this project?'*, '*Did your GP or other health care professional recommend you attend?*', '*What do you hope to gain from joining the group?*'.
The responses were all very similar in nature as the group of participants that were included were recruited from the same Pulmonary Rehabilitation groups and 'British Lung Foundation' - Breath-easy meeting groups where the initial taster sessions were conducted.

Nineteen of the twenty-four COPD patients that provided responses, had been advised about the course from either their General Practitioner or through a pulmonary rehabilitation or COPD exercise group (i.e. nurse, HCP) and the remainder had heard from leaders or friends at their local BLF 'Breatheasy' group. Only two refrained from responding.

In response to the question 'What do you hope to gain from joining the group?' responses were broadly similar and as expected i.e. participants hoped to gain some improvement to their breathing, wellbeing, mood and confidence. To learn how to manage their condition, control their breathing and generally feel better.

Follow-Up Visit V_2 – Participant Feedback Comments

Participants were asked to provide open text feedback comments at Follow-Up Visit V_2 (Questions detailed in Appendix C, sub-appendix I).

A total of eighteen participants of the possible twenty-eight that completed Follow-Up Visit V_2 (as summarised in figure 6 below), completed the final page of the questionnaire and provided free text feedback comments.

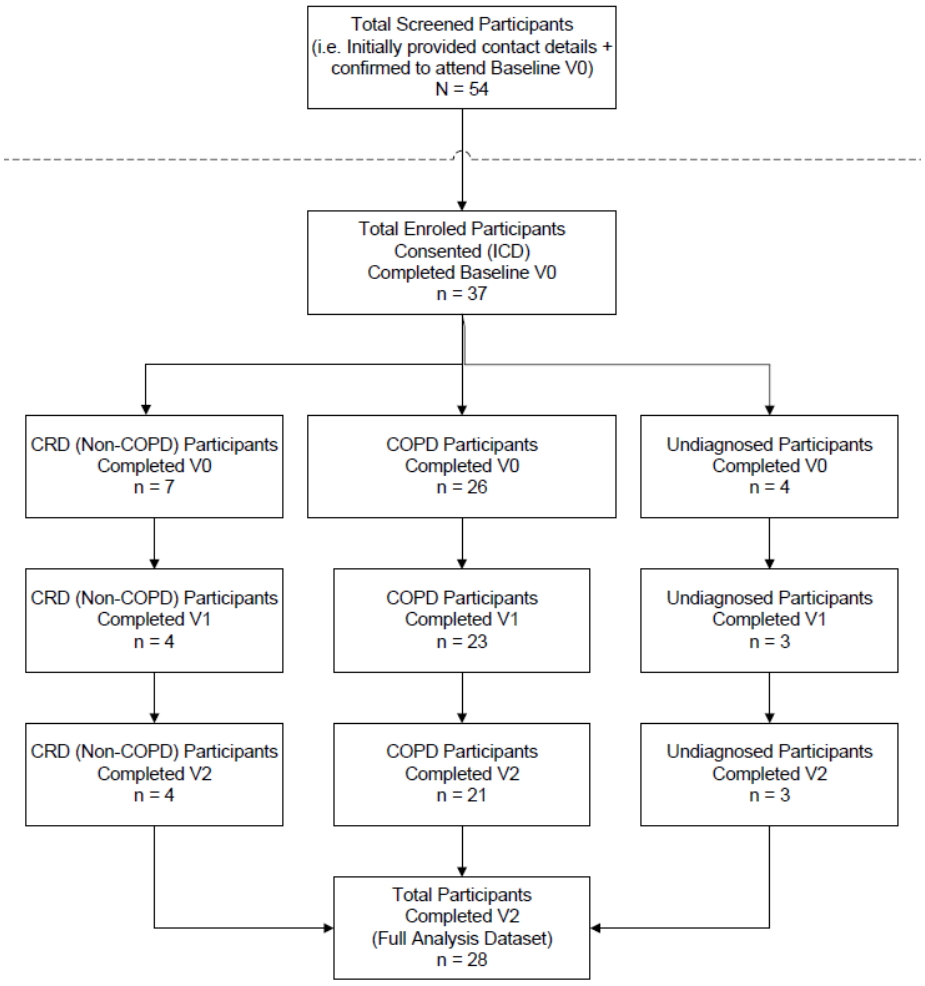

Figure 6 - Research Study Results Part I: Flow of Participants up to Follow-Up Visit V_2

These comments were not extensive and since the number was very small a detailed analysis was not possible however, there were some key themes noted and the comments were reviewed manually. Details regarding either physical health or mental health outcomes were identified and collated as below.

A total of eleven patients made a comment that specifically related to physical activities e.g. breathing apparatus, lungs, exercise or respiratory condition:

“[The leader] has really helped us a lot in exercises and singing, I can now let breathe out twice as long as I was able to before we started”

“My lung condition is progressive but singing has helped in the short term”

“I find singing does help a lot with my breathing”

“My diaphragm feels looser after singing”

“We soon got refreshed on how to sing well, this in turn helped improve my breathing when I got in a panic situation i.e. moving fast to the toilet in public and fearing I would not make it.”

“It has helped my breathing and taught me a level of control previously lacking”

“The singing has aided my lung condition I would say there is a slight improvement however on days I don't sing so much there is a decline in my lung condition”

“It has been a great experience and a noticeable improvement in breathing”

“It has helped me to learn to breathe from my diaphragm”

“I felt some improvement in my breathing which I think would improve if the group exercise continued”

“The singing group was very useful for my lung condition”

“Helped to improve my breathing by correct lung exercises”

“A noticeable improvement in breathing”

The greatest number of comments were made and recorded in relation to participant mental health and wellbeing, i.e. the enjoyment of the activities and exercises, improvements in confidence, social connectivity, having fun with others etc. Over 77% of participants (n=18) that provided free text feedback mentioned one or more comments of this nature.

NB. The written comments noted below reflect many more similar verbal comments made to the singing leaders and researcher at the start and end of each of the sessions.

"It gave me an interest"

"It is very much a spiritual help as well as physical"

"It has been absolutely wonderful - physically and mentally"

"I feel happy when we are singing"

"There are times when I have only got out of bed/house because of singing"

"The sound of all our voices is spiritually uplifting"

"I have more confidence now"

"I have widened out my singing to the wider community so I will be doing charity singing"

"Singing is now part of my day to day life again after a long absence"

"I feel better since starting singing class"

"Really enjoyable and a brilliant way to meet new friends"

"Overall feeling much better"

"I enjoyed coming"

"I have really enjoyed the group"

"Helped with mixing in a group"

"It has been a great experience"

There were in addition to physical and mental wellbeing comments eight other comments of note which specifically referred to a responders' sadness at the cessation of the groups or desirability for continuation of the groups. The general comments made to the researchers and singing leaders, during socialising following the singing sessions, continued with this same theme.

"Will miss our weekly get together a lot"

"I felt some improvement in my breathing which I think would improve if the group exercise continued"

"Would like to request a repeat course"

"Am sad that it is ending"

"I hope it can continue in some form"

"The group is very good but needs to continue for it to have a lasting benefit and I would be willing to pay a modest sum to enable this"

"It would be brilliant to continue the singing with the group"

"I will sadly miss the group and hope there will be some way of continuing"

It should also be mentioned that the discussion regarding continuation of the groups and feasibility of raising funds to continue was continually on the agenda of the participants. The comments above reflect the strong desirability of long-term continuation of the groups and maintenance of support to achieve this end.

Two participants also indicated in their comments some reservations about the sound of their singing voice which they had 'overcome' due to the regular group singing.

"I have never sung at school I always mimed when singing, in church I mimed. Now I sing all the time because of this project"

"I cannot sing, I have joined in and no-one complained"

Finally, one participant provided a comment which was non-positive in regard to the group sessions. This patient was in fact a protocol violator and was diagnosed with a CRD other than COPD (i.e. Bronchiectasis) so would not have been eligible to be included in the health and wellbeing feedback assessments, however their general feedback seemed important to report.

"Although I enjoyed the singing sessions it was difficult to commit to weekly sessions. There was also a feeling that some of the group wished to perform and make the sessions more public. I would be extremely uncomfortable with this and consequently have decided not to continue.

In general, the participant feedback was limited, though the information collected still provides valuable insight.

Participant Attendance

One of the five secondary endpoints of the study was to assess the overall compliance and drop-out rate for singing groups. The compliance was collected in the form of registers, maintained weekly by the singing leaders of each of the groups. These were submitted regularly for data entry by the researcher. The primary aim of this was for compliance purposes i.e. to ensure that participants included in the study analysis had attended at least 75% of the available group sessions (n=13) during the study period i.e. to be considered regular attendees. It was also useful information in understanding if there were any pattern to the retention and whether this could be utilised for future study planning.

There was a very low drop-out rate of <20% observed for singing group participants with a COPD diagnosis prior to the Mid-Term Visit V_1 Week 7 (n=26) with retention remaining steady and no further drop out for these participants through to the Follow-Up Visit V_2 Week 14. A further 19% of participants (n=26) were seen to drop out between Follow-Up Visit V_2 and Final Follow-Up Visit V_3 which also included the hiatus period during the summer when the groups ceased.

The overall attendance at the weekly sessions was well maintained by all included participants over time. There was an initial high drop-out around the first two weeks but after this the attendance remained steady. The main drop-out reasons, where a reason was provided verbally to singing leaders, indicated that participants found the timing or location (e.g. parking facilities) of the sessions inconvenient and there was no indication that this was related to participants being dissatisfied however, the majority of the participants that dropped out were 'lost to follow-up'.

Research Study: Discussion Part I

Health and Wellbeing

In order to assess the endpoints of health and wellbeing quantitatively, the difference in mean total score from baseline, for each of the three validated health and wellbeing scales was the most important focus of this research. A two-tailed criterion was employed to test the null hypothesis i.e. 'no change occurred over time on these measures' such that a beneficial impact of the singing intervention over time is assumed as the alternate hypothesis.

Questionnaire Data – Total Score

The key outcome of this research indicated that group singing positively affected the health and wellbeing of this group of participants diagnosed with COPD. The mean total score for mental wellbeing as measured by the Warwick-Edinburgh Mental Wellbeing Scale (WEMWBS) and the mean total score for impact of a patient's COPD and experience of breathing difficulties and how this changes over time, as measured by the COPD Assessment Test (CAT) showed significant improvement at Follow-Up Visit V_2 (12 weeks $\pm$14 days) post Baseline Visit V_0 i.e. There was an increase in the mean WEMWBS score between Baseline Visit V_0 (M=49.14, SD=12.397) and Follow-Up Visit V_2 at Week 12 (M=55.23, SD=10.150); t(20)=2.606, p = 0.017 and a decrease noted in the mean CAT score between Baseline Visit V_0 (M=25.03, SD=7.774) and Follow-Up Visit V_2 at Week 12 (M=20.27, SD=10.659); t(20)=2.784, p = 0.011 for the 'per protocol' COPD participants across the two separate singing groups.

This improvement was maintained and in fact showed further improvement at a level of significance at Final Follow-Up Visit V_3 (30 weeks $\pm$21 days post Baseline Visit V_0) i.e. There was an increase in the mean WEMWBS score between V_0 (M=48.43, SD=12.886) and V_3 (M=56.81, SD=8.719); t(15)=3.581, p = 0.003 and decrease in mean CAT score between Baseline Visit V_0 (M=24.16, SD=7.282) and Final Follow-Up Visit V_3 (M=19.13, SD=9.563); t(15)=2.877, p = 0.012 for these two same scales, despite a short cessation in group singing sessions post Follow-Up Visit V_2.

The mean improvement in WEMWBS and CAT scores between Baseline Visit V_0 and Follow-Up Visit V_2 met required significance i.e. $p \leq 0.05$ and this required significance was maintained at Final Follow-Up Visit V_3. The values for difference in mean total ACQ score at the same intervals also showed improvement which was maintained at Final Follow-Up Visit V_3, however the values for the change in mean total score for this scale fell just short of the required significance level (i.e. p = 0.059) for the same periods.

The most reliable estimate of the minimum important difference (MID) for the CAT questionnaire, particularly in response to intervention studies, has been assessed to be a change of two or more points **(Kon et al., 2014)**. The results obtained from this scale in the current study showed a change of five points which was maintained after thirty weeks post baseline. This finding is clearly very encouraging in regard to the impact of regular singing on COPD symptoms, though caution has to be exercised regarding the small sample size and low power and also the low quality of the study due to design constraints.

There is statistical significance in the mean difference, but this only shows the measurement precision. The substantive change here is relatively low due to the small sample size, which indicates the effect within the population is low.

It further supports finding from two previous singing for COPD studies **(BLF, 2017, Jamaly et al., 2017)** where this measure was employed. These are referenced in detail in the earlier systematic review. One study **(Jamaly et al., 2017)** recorded significant improvement of five points (17 ± 9 to 12 ± 8 points) in a singing intervention group not seen in the control group (physiotherapy lung exercises) after four weeks of more frequent singing sessions (average of twice per week), but the full details of this study cannot be assessed clearly for quality. The second **(BLF, 2017)** used this measure to show a significant (p-value = 0.02) mean improvement of 1.9 points following twelve weeks of singing intervention in COPD patients, which approached but did not exceed the MID value.

Similarly, for the WEMWBS scale it has been suggested **(Jaeschke et al., 1989)** that an improvement of 0.5 on each item in the 5-point Likert scale represents an improvement (one which is deemed important to an individual) implying that an improvement of seven points across the fourteen items could be deemed important. However, more recent research suggests that a meaningful change in WEMWBS score is seen between 1.0 and 2.77 SEM which is generally between three and eight points **(Maheswaran et al., 2012)** so again, the results of this study and improvement in six points, is within this range and are encouraging in regard to participant wellbeing.

No other similar studies selected for the systematic review used the WEMWBS measure specifically, so this study is the first study in this area of research to use this scale. Hence, as a result it is not possible to compare the scores obtained with previous findings. However, these findings are in alignment with similar studies within this field that found positive results utilising similar wellbeing and quality of life scales such as the Short Form Survey (SF-36) **(Bonilha et al., 2009, Lord et al., 2010)**. However, this study shows more positive outcomes than similar studies using scales such as the EuroQoL (EQ-5D) **(BLF, 2017, Clift et al., 2013, Clift et al., 2017, Pacheco et al., 2014)** where no significant outcomes were found.

These outcome values show that the intervention impact on participants' symptoms due to their COPD was slightly more significant than the impact on wellbeing outcomes, which would support the general understanding that respiratory changes are one of the most obvious and perceptible changes noted by patients **(Kang et al., 2017)** attributed to their group singing intervention.

Participant Feedback

Participant feedback was solicited at Baseline Visit V_0 and Follow-Up Visit V_2 twelve weeks post baseline. The majority of participants chose to write comments to supplement quantitative information though due to time availability of venues and resources along with other priorities, the written feedback provided was not extensive and is utilised simply for illustration purposes i.e. to provide context to the quantitative feedback by the groups. The obvious key themes were

manually extracted and support the feasibility, acceptability and effectiveness of group singing in this population.

This aspect of the research was not a primary endpoint for the study and as such it was appropriate that the focus was placed on ensuring adequate time for the collection of quantitative responses. It was also deemed important that the aspect of social engagement within the groups (at the start and end of sessions) should be encouraged and this took precedence during data collection over requests to participants to provide extensive written comments. In future studies, it would be recommended to provide more structured interview questions similarly to other recent qualitative studies **(Skingley et al., 2018)** and to grant more time for this during study design and logistic arrangements.

The feedback provided by participants at Baseline Visit V_0 was collected to provide an overall picture of recruitment regarding the set-up of the singing groups and to provide support to future study design and logistic consideration. The comments provided were not extensive, but they showed that the majority (~80%) of the participants diagnosed with COPD (n=26) had been advised about the course from a healthcare provider and almost all those that responded mentioned that the aim was to gain some improvement to their breathing, wellbeing, mood and confidence and to learn how to manage their condition, control their breathing and generally feel better.

The feedback solicited and captured on the final page of the questionnaire at the Follow-Up Visit V_2 was aimed at addressing individuals' state of health and subjective accounts of any health benefits or considerations potentially due to the singing intervention. This data was analysed to address a secondary endpoint of the study i.e. Summary of all written feedback, as captured in patient evaluation forms or by singing leaders. These questionnaires were completed by eighteen participants, as recorded in free text feedback comments. Interestingly the participants identified various mechanisms whereby benefits were perceived from the singing intervention.

Eleven individuals recorded one or more positive comments that specifically related to their physical activities and abilities in particular e.g. their breathing apparatus, lungs, exercise or respiratory condition. All these participants made comments mentioning that singing was useful, aided or helped their health in some way or that they gained a perceived physical health improvement from attending the groups. The participant comments noted in relation to mental health and wellbeing i.e. the enjoyment of the activities and exercises, improvements in confidence, social connectivity, having fun with others etc. was slightly more significant with over 77% of participants (n=28) recording comments of this nature which supports recent studies that collected similar participant comments **(Skingley et al., 2014, Skingley et al., 2018)** that the majority of participants in the research study agreed that the singing programme led to improvements in respiratory symptoms between baseline and follow-up.

In addition, the responders raised issues of sustainability from early on and while they were particularly mentioned at the Follow-Up Visit V_2 the discussion around the continuation of the groups was a common theme in written feedback and personal comments made to the researcher and singing leaders. This even seemed to cause consternation for those members for whom the group singing had made a perceived impact on their lives. This discussion regarding continuation of the groups and feasibility of raising funds to continue was continually on the agenda of the participants and it should be an ethical consideration from the outset to ensure that discussions regarding this aspect are brokered with participants during early sessions to manage expectations.

Summary of Qualitative Data

All but one of the participant provided comments regarding the intervention, group interactions and impact on health and wellbeing, were positive in nature and strongly supported the positive findings from the quantitative data. There were comments regarding general enjoyment, feeling happy and feeling improvements in their health and wellbeing, including social connectivity and self-confidence, all of which provided strongly supportive evidence to the significant increase in mean score for individual CAT and WEMWBS statements at Follow-Up Visit V_2 in regard responses pertaining to feeling optimistic, good about oneself, confident, interested in new things, cheerful and having more energy to spare.

Unexpected Results

Two participants also expressed some reservations about the sound of their singing voice, which they considered had been ‘overcome’ following regular singing within the group, which might illustrate an underlying lack of confidence or even concern. This is also often voiced by potential participants during recruitment for singing groups. It is possible that this could be a hurdle for encouraging future recruitment as potential participants may be deterred from joining similar future groups, particularly in a population which may experience greater issues with self-confidence. It is important that this worry might be addressed in recruitment material and to extend taster sessions more widely to allay this potential fear.

There was one unexpected result in a comment noted from a non-COPD participant (i.e. a participant with a respiratory condition which was not COPD and thus was ineligible for inclusion in health and wellbeing analysis) who had not returned following the hiatus of the sessions in the summer. It raised personal concerns of performing in public and giving rationale that this issue along with difficulty in commitment to the sessions held the responder back from returning. The participant did not make this concern known during the sessions attended and it was obviously unclear to this participant that there would not be pressure to perform or make sessions more public.

If these concerns had been brought to the attention of the researcher and singing leaders then they would certainly have highlighted this clarification more broadly to both groups, as it may have been something other participants were also potentially concerned about, then any fears or worries could have been addressed.

These findings indicate the importance for organisers or leaders of future groups to continue to elicit ongoing feedback from their own participants wherever possible and to ensure they feel comfortable with expectations regarding changes to the group structure or public performances. In groups where patients are managing health conditions such as COPD which involve symptoms such as anxiety and lack of confidence finding avenues to convey and reiterate that there is no expectation is key. Participants may feel pressure to enter situations that might cause them stress or anxiety which may cause them to refrain from attending further sessions, rather than vocalising their concerns.

Similarly, it should be an important consideration for future recruitment and retention of singing for health groups that any resources or materials should indicate that public performances are optional and that potential attendees should not feel self-conscious about the sound they are making. It should be clear that while the aim of the group would be to achieve a quality sound to improve the feelings of achievement, cohesiveness and thus wellbeing, the level of the singing or performance should not discourage those with lower levels of self-confidence.

It is also important that contact with participants who drop-out of sessions is maintained wherever possible or that there is further outreach or contact beyond the group to reduce the potential numbers of ‘lost to follow-up’ participants and to gain more feedback of this kind. This has not been widely addressed in the previous studies and it is vital for providing learnings for communications with future groups and more effectively address potential concerns before they ‘take root’. It is also essential to provide guidance to research groups and singing leaders to ensure these concerns might be addressed more regularly in case there are underlying concerns or worries by individuals that may be hidden or not vocalised due to the enthusiasm of other group participants.

Enrolment, Compliance and Retention

It is notable that the majority of those recruited into these groups i.e. nineteen of the twenty-four COPD patients that provided responses, had been advised about the course from either their General Practitioner (GP) or through a pulmonary rehabilitation or COPD exercise group i.e. nurse, health care professional (HCP). Since COPD and respiratory disease is predominantly seen in older patients it should be recognised that these patients are often seen to respect their GP, health care provider or physician's advice and have regular contact with their family doctor, so healthcare workers and advisors can also play key and pivotal role in the initiation and

maintenance of these types of exercise behaviours among the population **(Schutzer and Graves, 2004)**.

There was a low drop-out rate of <20% of participants (n=37) observed for all COPD singing group participants prior to the Mid-Term Visit V_1 Week 7 with retention remaining steady with only a 0.05% drop out through to the Follow-Up Visit V_2 Week 14. This could correlate with the high number of positive comments regarding social interaction and cohesion between the participants.

There were a further 22% of participants (n=37) seen to drop out between Follow-Up Visit V_2 and Final Follow-Up Visit V_3 which also included the hiatus period during the summer when the groups ceased. It is possible that this may have impacted the retention rate over this period although there is no specific data to support the reason for reduced retention management over this timeframe.

Interestingly, during this same period the two separate singing groups that existed initially at Centre A and Centre B were amalgamated to one group, located at Centre B. It might be anticipated that the group of participants attending sessions at Centre A would be impacted by the change in some way, thus impacting retention of this subgroup. However, the five participants that dropped-out during this period were all from the original Centre B group and all the participants from the separate Centre A group that were given the option to continue at a different venue with a new singing leader were all retained up to the Final Follow-Up Visit V_3.

Since it has been seen that group cohesion is an effective predictor of both short and long-term adherence to exercise in older adults **(Estabrooks and Carron, 1999)** it might be hypothesised that group cohesion could be effective for improving adherence to continued exercise practise in this study where a similar effect has been observed. Singing can be seen as a form of exercise and the strong cohesive bonds made within these groups may in turn be responsible for the positive retention rates seen. This has been highlighted in the participant feedback and it is likely due to a variety of other factors brought about by the activity of group singing itself, as previous research indicates i.e. that singing increases the release of the hormone oxytocin which can induce a socio-biological bonding response **(Kreutz, 2014, Pearce et al., 2015)**.

It is also interesting that the overall drop-out rate for diagnosed COPD patients (n=26) was less than that seen for the full group (n=37) which included participants with other lung conditions and participants that were not previously diagnosed. This was 7% less between baseline and Final Visit V_2 and 9% less across the full study i.e. baseline to Final Follow-Up Visit V_3 which might indicate that the COPD patients may have felt more engagement or benefit from attending or alternatively they felt that the activities and exercises were more targeted to the improvement of health and wellbeing of their condition of COPD specifically.

The reasons for drop-out were not captured systematically and this is something that could be improved upon in future study planning. The information provided verbally to singing leaders indicated that participants found the timing or location (e.g. parking facilities) of the sessions inconvenient and there was no indication that this was related to participants being dissatisfied, however since the majority of the participants that dropped out were lost to follow-up and the data was not captured or solicited systematically this cannot be analysed. It would be recommended that future studies consider follow-up during study plan and capture this critical data formally to inform future planning.

Chapter 5 – Results and Discussion – Part II: Sub-Study

Research Study: Results and Data Analysis Part II

Study Enrolment and Demography

Research Study: Sample for Self-Management Resources Evaluation

A total of twenty-eight participants were considered eligible to utilise the prepared SfBB resources and provide evaluations on these. These same twenty-eight participants were also included in the analysis of the overall compliance and drop-out rate for singing groups.

There were initially twenty-six participants diagnosed with COPD and a further eleven other participants enrolled at Baseline Visit V_0. Five of these participants were undiagnosed with a respiratory condition, one of whom declined to give consent for evaluation and was excluded from the data collection. (However, they continued in the singing group to support another participant diagnosed with a respiratory illness). A further seven other participants, previously diagnosed by a Physician or Health Care Provider (HCP) with varying non-COPD Chronic Respiratory Diseases (CRDs) i.e. Asthma, Bronchiectasis, Aspergillosis, Bird Lung Disease and who were not previously diagnosed with a COPD related lung condition by their Physician or Health Care Provider (HCP) were also included.

Three of the undiagnosed participants and four of the participants diagnosed with a CRD (non-COPD) continued with the singing group sessions and attended the Follow-Up Visit V_2 evaluations where the SFBB-Q was administered. While these seven participants did not meet the full inclusion criteria of the protocol and deviated from inclusion #3 ‘i.e. has ‘previously been diagnosed with a COPD related lung condition by their Physician or Health Care Provider (HCP)’, they did fully meet all other protocol criteria.

These participants were excluded from health and wellbeing outcome analysis as their diagnosis deviated from the strict protocol inclusion criteria regarding COPD. However, since these participants were assessed individually as being involved in self-managing respiratory conditions in some way, the researcher deemed that their feedback for evaluation of the self-management resources pilot would be appropriate and valuable. These participants provided informed consent and demographics at Baseline Visit V_0 and met all other inclusion criteria of the study. They were included in all singing sessions which enhanced the overall attendee numbers and supported the retention and engagement of the singing groups. A flow diagram to illustrate the number of participants at each assessment is shown in figure 7 below.

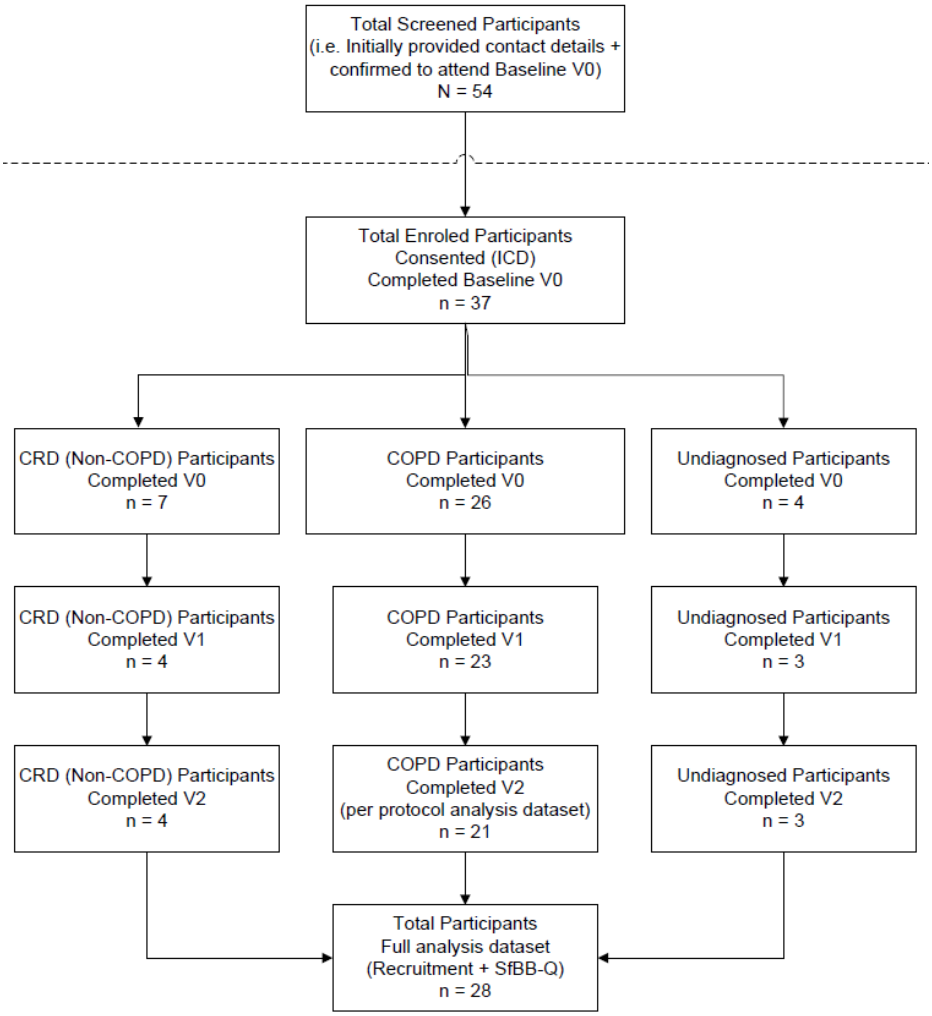

Figure 7 - Research Study Results Part II: Flow of Participants for SfBB Resources Evaluation at Follow-Up Visit V2

A total of thirty-seven participants completed baseline evaluations for the study and were supplied with SfBB resources (Booklet and DVD) at their Baseline Visit V_0, for home use throughout the study. Twenty-eight of these thirty-eight participants attended evaluations at both baseline (Baseline Visit V_0) and post-baseline follow-up (Follow-Up Visit V_2) to provide feedback on the SFBB resources. The demographic details of all participants are given in table 11 below.

Demography of those Participants Included in V_2 Evaluation	Male	Female	Mean Age	V_2	No. of Participants with SfBB-Q evaluable for total score
CRD - COPD (PP)	13	8	66.62 $\pm$9.3	21	19*
CRD - Non-COPD)	0	4	63 $\pm$9.8	4	4
Undiagnosed	0	3	59.33 $\pm$5.5	3	3
Total	13	15	65.32 $\pm$9.1	28	26

Table 11 – Research Study Results Part II: Summary of Participants with Evaluation at V_2

NB The reduced number of evaluable questionnaires (*) for the total score, is due to two participants with questionnaires having >1 missing response value to an individual question.

Participants were considered for inclusion in the SfBB Resource evaluation group if they met all protocol inclusion criteria and had completed at least Baseline Visit V_0 and Follow-Up Visit V_2 evaluations. In addition, those participants who had showed a deviation from inclusion criteria #2 'Patients previously diagnosed with a COPD related lung condition by their Physician or Health Care Provider (HCP)', were also considered to be evaluable. This included three undiagnosed participants and four participants, previously diagnosed by an HCP with similar Chronic Respiratory Diseases (CRDs) i.e. Asthma, Bronchiectasis, Aspergillosis, Bird Lung Disease but not suffering specifically from a COPD related condition.

Research Study: Sample for SfBB Resource Evaluation

One of the key secondary endpoints for this research aimed to assess and review the feasibility of specifically designed and created resources for home singing. These were created to support and encourage home singing for COPD patients specifically. The tools have been designed to assist the consistency and persistence of participant singing outside of the singing sessions. These resources consisted of a booklet with songs and instructions, along with a DVD of songs with music and lyrics which could be watched and followed for practice between sessions to encourage participants to continue their singing exercise beyond just the group sessions

A bespoke questionnaire, created for the purposes of capturing relevant feedback, was utilised for this study. The aim of the next section is to present the results for these evaluations. An evaluation of the usage and limitations of the questionnaire itself will be presented in subsequent sections.

SfBB Resource Evaluation Definition

It was decided that although they deviated from this single protocol inclusion criteria, their feedback of the resources would be considered valuable and these participants could be included in the SfBB-Q analysis to allow for the maximum evaluation of the SfBB resources.

There were twenty-eight evaluable questionnaires completed overall. Feedback from twenty-eight participants was available to evaluate SfBB questions #5 to #8, twenty-seven for questions #1 to #2 and twenty-six for questions #3 to #4. Two participants left >1 question unanswered but provided feedback that they experienced some technical issues and they were unable to utilise the SfBB DVD resources and as such were not able to give full feedback on the tools.

SfBB Resource Evaluation

There was a total of twenty-eight participants from the group that provided feedback on the resources and of these twenty-six that recorded a response to seven or more of the eight individual questions i.e. did not leave more than one question blank or marked as not done.

SfBB Questionnaire (SfBB-Q) Total Scores

A secondary endpoint for the study research included the mean total SFBB Questionnaire score for usage and usability of SDHR SfBB resources as recorded at V_2 (Week 13).

The total score is provided by adding the scores from the eight questions to derive a total score (out of a maximum of 48) with 0 being the most positive and 48 being the most negative. A score <24 is deemed to indicate an overall positive score and a score >24 deemed negative, with 24 being neutral.

Over 88% of the participants that completed the questionnaire (n=28) indicated a positive score <24 with only three female participants, (aged 64 -75) recording a negative score >24.

There were two COPD participants whose feedback data were excluded from the overall summary score data analysis as they left >1 question unanswered. The overall summary score total was available for twenty-six participants out of a total of twenty-eight. This was quite misleading and problematic to analyse due to the variety of the questions and challenges faced by participants that were not able to access the DVD resources.

Two of these participants showed scoring and feedback which appeared inconsistent, indicating that they might have had some confusion in completing the questionnaire or instructions were not clear.

Participant #2010 recorded a negative score (>3) for Q1 '*I used the booklet and/or DVD regularly'* and for Q2/Q3 **'I found the instructions in the booklet/on the DVD easy to follow'* but a positive score for Q5 '*It was easy to fit in regular practice at home'* (2) and Q4 '*Using the booklet and DVD made me feel more confident at the group sessions*' which appeared inconsistent in comparison with other scores. The data for these participants appeared spurious for this reason but was retained since there was no clear indication that this was completely inaccurate.

Participant #2018 recorded a score of six for all the responses to the questionnaire and in the verbatim comment section noted the verbatim text:

'I didn't get to play the DVD as I haven't got the resource to play it on, so sorry I can't comment'

So again, this could be deemed spurious but since this participant provided a negative response for all questions, it was included in order to ensure the approach taken with the data was 'most conservative' possible and did not introduce any positive bias.

The demographic data analysis was conducted to understand if there were any differences in total score related to the age of the participants, but no strong correlations were seen. A Pearson correlation coefficient analysis was computed to assess the relationship between age and the total SfBB score and a very weak positive correlation was detected between these two variables $r = 0.401$ $p = 0.035$, which was seen to be significant and thus may warrant further analysis in future studies.

Independent t-tests were conducted to compare the total SfBB-Q score with gender for the available sample of patients, to explore any potentially significant differences (see Appendix D).. There was no significant difference in the score for males (M=10.56, SD=4.667) and females (M=17.47, SD=9.851); $t(26) = -1.989$, $p = 0.057$ between the Baseline Visit V_0 and the Follow-Up Visit V_2.

The overall analysis of the total score was indicative of positive reception to the resources but was not particularly revealing, due to the variability of the questions and responses. Thus further analysis of individual scores was deemed to be more pertinent in examining this feedback.

SfBB Questionnaire (SfBB-Q) Individual Question Scores

The data analysis from each of the questions was deemed more revealing than the total score in helping to understand the usage and usability of the tool, so the analysis is further broken down into the feedback provided for the individual questions.

The questionnaire has been designed so that a score of <3 shows a positive indication where the participant is completely (0), mostly (1) or slightly (2) in agreement with the original positive statement on the left and a score of >3 is similarly a negative indication where the participant was in agreement completely (6), mostly (5) or slightly (4) with the negative answer on the right. A score of 3 indicates neither agreement nor disagreement with either statement. A summary of the totals for each of the individual question scores are summarised in Table 13 below.

Category *(Pt = Participant)*	V_2 SfBB Q1 I used the booklet and/or DVD regularly	V_2 SfBB Q2 I found the instructions in the booklet easy to follow	V_2 SfBB Q3 I found the instructions on the DVD easy to follow	V_2 SfBB Q4 Using the booklet and DVD made me feel more confident at the group sessions	V_2 SfBB Q5 It was easy to fit in regular practice at home	V_2 SfBB Q6 I will continue to use the booklet and DVD to practice	V_2 SfBB Q7 I would recommend the booklet and DVD to others	V_2 SfBB Q8 I would be likely to use this or other singing resource online or via an electronic device, mobile phone 'app' or similar
Total Pts n completing question	27	27	26	26	28	28	28	28
Total No. Pts with SfBB-Q Score <3 (positive)	8	19	21	18	14	18	22	19
Total No. Pts with SfBB-Q Score =3 (neutral)	13	5	1	5	4	6	4	4
Total No. Pts with SfBB-Q Score >3 (negative)	6	3	4	3	10	4	2	5
Total % of Pts with Positive SfBB-Q Score	30	70	81	69	50	64	79	68

Table 13 – Research Study Part II: SFBB-Q - Individual Question Score Summary

Since there was a small sample the scores for the whole group of participants answering that question are grouped as positive, neutral or negative for analysis purposes and presented as a percentage of the total number of participants answering each individual question. This is to help identify any significant patterns within those groups.

Q1 I used the booklet and/or DVD regularly

Only 30% of participants (n=27) that completed this question indicated a score of <3 (where 0 indicates strong agreement with the statement 'I used the booklet and/or DVD regularly' and 2 indicates slight agreement) but a further 48% indicated a neutral response of 3 neither agreeing with the statement or agreeing with the negative alternative statement. Only 22% (n=27) recorded a value of >3 indicating 'I did not use the booklet of DVD regularly'. These two participants along with one additional participant (who did not answer this question) indicated by written comment on the form or by verbal comments to the facilitator, that they were not able to use the DVD in their PC or did not have a PC or DVD player facilities.

Examples of comments noted or made to the researcher or singing leaders were:

> "The *DVD wouldn't work"*
> *"The DVD didn't play on my PC"*
> *"I don't have a DVD player"*
> *"I didn't get to play the DVD as I haven't got the resource to play it on"*

This indicates that the majority of participants included in the review had used the booklet and/or DVD at least once during the first 12 weeks of the study and almost a third of participants used the resources more than once. It would also appear that those that did not use it were likely impacted by the issues with the DVD access, even though they were encouraged to still use the booklet alone. Several of the participants made ad hoc comments to the investigator at the mid-way and final visits that they were now singing regularly outside the sessions. However, whether it was using the resources as a guide was not always apparent.

Q2 I found the instructions in the booklet easy to follow

Over 70% of participants that completed this question (n=27) indicated a score of <3 (where 0 indicates strong agreement with the statement 'I found the instructions in the booklet easy to follow' and 2 indicates slight agreement). Only one of these participants recorded a value of 5 which was the highest score selected, indicating I did not find the instructions in the booklet easy to follow'.

Q3 I found the instructions on the DVD easy to follow

Over 80% of participants that completed this question (n=26) indicated a score of <3 (where 0 indicates strong agreement with the statement 'I found the instructions on the DVD easy to follow' and 2 indicates slight agreement). However, over 69% indicated a score of <2 though (i.e. strongly or mostly in agreement with the positive response compared to 65% for the booklet

instructions. It is possible that the delivery method of verbal instructions on the DVD, similarly to the singing teacher in the sessions, was more readily received. The simplicity of audio/visual use impacted the score and may be worth further investigation.

Q4 Using the booklet and DVD made me feel more confident at the group sessions

Over 69% of participants that completed this question (n=26) a score of <3 (where 0 indicates strong agreement with the statement 'Using the booklet and DVD made me feel more confident at the group sessions' and two of these twenty-six indicates slight agreement). Only three of these twenty-six participants recorded a value of >3 indicating agreement with the statement 'Using the booklet and DVD did not made me feel more confident at the group sessions'.

Q5 It was easy to fit in regular practice at home

Only 50% of participants that completed this question (n=28) indicated a score of <3 (where 0 indicates strong agreement with the statement ''It was easy to fit in regular practice at home' and 2 indicates slight agreement). Only ten of these participants recorded a value of >3 indicating agreement with the statement 'It was not easy to fit in regular practice at home'.

Q6 I will continue to use the booklet and DVD to practice

Over 64% of participants that completed this question (n=28) indicated a score of <3 (where 0 indicates strong agreement with the statement 'I will continue to use the booklet and DVD to practice' and 2 indicates slight agreement). Only four of these participants recorded a value of >3 indicating agreement with the statement 'I will not continue to use the booklet and DVD to practice'.

Q7 I would recommend the booklet and DVD to others

Over 78% of participants that completed this question (n=28) indicated a score of <3 (where 0 indicates strong agreement with the statement 'I would recommend the booklet and DVD to others' and 2 indicates slight agreement). Only two of these participants recorded a value of >3 indicating agreement with the statement 'I would not recommend the booklet and DVD to others'

Q8 I would be likely to use this or other singing resources online or via an electronic device, mobile phone 'app' or similar

Over 67% of participants that completed this question (n=28) indicated a score of <3 (where 0 indicates strong agreement with the statement 'I would be likely to use this or other singing resources online or via an electronic device, mobile phone 'app' or similar' and 2 indicates slight

agreement). Only two of these participants recorded a value of >3 indicating agreement with the statement 'I would not recommend the booklet and DVD to others'.

SfBB Qualitative Data – Participant Written Feedback

The qualitative information provided by participants regarding the SfBB resources was extremely limited. While it was encouraged that participants mention their views and comments regarding the resources specifically in the written feedback questionnaire, there were only four comments provided. Two of these were related to accessing the DVD i.e. '*DVD wouldn't work'* and *'I didn't get to play the DVD as I haven't got the resource to play it on, so sorry I can't comment'* which supported the evidence obtained in the quantitative data.

The other two comments both gave a positive message i.e. *'The DVD and booklet were very helpful'* and *''I sang out loud with the DVD and heard my own voice singing for the first time in years and felt good about it'* which also supported the quantitative data from the questionnaire.

It was noted that participants did seem eager to provide verbal feedback about the resources during the sessions, so on reflection it might be advantageous in future to include discussion group methodology to capture verbal feedback from participants regarding their use and views of the of the resources or to incorporate questions via interview techniques. The collection of more coherently verbal comments could provide rich feedback to improve the development and usage of future resources.

Research Study: Discussion Part II

SfBB Questionnaire – SfBB Resources Evaluation

Total SfBB-Q scores were generally very positive with over 88% of the participants that completed this questionnaire and had an evaluable total score (n=26), indicating an overall positive score <24 with only three female participants, aged between 64 and 75, indicating a negative score >24.

The questions or statements that indicated the lowest overall positive scores were two related to regularity of use and finding time to practice i.e. question SFBB-Q1 'I used the booklet and/or DVD regularly' where only 30% of the participants that completed this question indicated a positive score and question SFBB-Q5 'It was easy to fit in regular practice at home' where only half the participants that completed this question indicated a positive score.

This indicates that one focus for improving the implementation and success of the resources might be to find simple ways to encourage regularity of use and improve consistency. This

might perhaps be by way of reminders by singing leaders at the group sessions or suggestions that the group might practice a song in readiness for the next group session etc. It might be by encouraging individuals in the group to find time to regularly complete activities between sessions e.g. employing good practices to remember or build activities into their daily or weekly routine or to even to take this further by exploring development of new tools to remind and enhance the experience e.g. simple reminder tools via a mobile app to increase more frequent use, singing exercises delivered via online application, website, mobile app or similar for easy practice 'on the go'.

Materials of this kind should be designed with different kinds of learning styles in mind **(Franzoni et al., 2008)** *as* people sometimes find it easier to learn and practice in different ways and at different speeds, so a variety of teaching resources, tools and methods might be best employed for maximum efficacy across different groups of people. Group methods have however been rated significantly higher than online tools in relation to content, interaction, participation, faculty preparation, and communication **(Ryan et al., 1999)**, so implications call for efforts in accessibility and creativity in these tools to ensure long term engagement.

In particular, research based on methods where information is processed through one of two usually independent channels where one channel processes verbal information such as text or audio and other processes visual information like diagrams, images, animations, etc., has concluded that a combination of electronic media used to expose learners, helps to improve their learning results **(Beacham et al., 2002)**, so further exploration of methods and tools could definitely be advantageous in this area. When singing instructions or tools for practicing these activities or exercises for health and wellbeing purposes are developed in future, it is important that these factors are considered. There is a need to offer a wider variety of options in addition to just verbal instruction, which is commonly used for community singing groups. This research supported this concept even though the media itself had somewhat mixed reviews. DVDs did not work well as a medium for some participants, but others gave praise to this way of presenting guidance. The verbal participant comments to the singing leaders also indicated that some participants felt encouraged to seek different types of media themselves e.g. online resources.

SfBB-Q statements SFBB-Q2 and SFBB-Q3 refer to the instructions of the booklet and the DVD i.e. the ease of use of the resources. The outcome of these questions would initially suggest that the participants found the DVD easier to follow than the booklet. The data was further explored, and this phenomenon was seen in very few participants i.e. only four participants. Three females and one male noted a difference in their individual question score between the instructions in the booklet versus the instructions on the DVD, so this would merit further exploration before any real conclusions could be made.

If these findings are an accurate reflection of the participants' views, then it may be possible that the delivery method of verbal instructions on the DVD or the simplicity of audio/visual

appearance impacted the score and may be worth further investigation i.e. those that used both the DVD and booklet may have found the instructions easier to see or read when visualised on a screen in front of them depending on the resolution and size of their monitor screen, system or equipment. There are many unknown factors here though, since this was external to the control of the research i.e. while the font size in the booklet was known and identical for all patients the participant experience of viewing the text or hearing the audio via the DVD was variable and unknown to the researcher. The instructions on the DVD were identical to those of the booklet, so the slightly less positive score for this statement may have been affected by the accessibility challenges some participants faced with use of the technology.

At least three participants were not able to utilise the DVD for technical reasons and provided feedback on this. It is possible others may not have been able to use the DVD but may not have noted this on the form and simply responded to the items on the scale. One participant (Participant ID #2018) recorded a value of six (the most negative response) for almost every statement except SfBB-Q2 "*I did not find the instructions on the DVD easy to follow* which was left blank. This impacted the overall average total score for the statements. However, since it appeared from the text that they were not able to use the DVD at all (i.e. Participant ID #2018 comment recorded "*I didn't get to play the DVD as I haven't got the resource to play it on"*) it is difficult to ascertain clearly if these responses of a negative score regarding the instructions on the DVD and booklet or all other statements were actually valid or if the participants were simply expressing negative scores because they were deterred from using the resources because they could not access the DVD at all.

One participant regularly mentioned verbally during the evaluation sessions that he didn't have his glasses/spectacles or told the researcher that he could not read or write though when advised that the researcher could support with questionnaire and answers, had replied that he was 'just joking' and continued to complete the questionnaires. This is just anecdotal information from one patient and not recorded in the results, however, this does raise the further question of whether lack of confidence may play a factor in asking for help to complete study questionnaires or use the resources, particularly within groups where the underlying disease has symptoms which could affect self-confidence. It might even be speculated this could be a possible reason why instructions on the audio-visual material received differing responses to some statements than paper versions for some participants.

The final three questions (i.e. SfBB-Q6 *"I will continue to use the booklet and DVD to practice",* SfBB-Q7 *"I would recommend the booklet and DVD to others"* and SfBB-Q8 *"I would be likely to use this or other singing resources online or via an electronic device, mobile phone 'app' or similar"*) are collectively attempting to elicit specific feedback from the participants to indicate and support the value of further exploration and investment of this type of media and technology in this population or if there would be little uptake or usage. The overall responses to questions SfBB-Q6, SfBB-Q7 and SfBB-Q8 were all positive with >64% of the participants completing these questions (n=28) indicating a score of <3, where 0 indicates strong agreement with the

most positive response. The results in this small group would suggest that the future investigation and development of self-help materials either these or similar resources would be valuable and that this engages participants and encourages practice between group singing sessions.

The overall outcome of the key evaluations showed improvement in all scores and improvement in three of the health and wellbeing questionnaires, two of these meeting the required significance levels. This could be for a number of reasons. One possible difference in this study to the others as identified in the systematic review of all other previously published studies in this area is that novel tools specifically designed to accompany the sessions were utilised. Since this is a pilot study with no randomisation or control group, it is only possible to speculate rather than draw clear conclusions. It would certainly be a strong suggestion that further investigations and trials be conducted to explore whether these tools or similar resources might be a factor in the engagement of the participants and the positive outcome.

It was a clear finding that using a variety of media to present the audio or visual recordings works well but still needs to be explored further to ensure the most targeted approach for specific populations. However, the overall results of the study do support similar previous research in this area where specially created novel self-management resources have been provided in addition to group singing **(Lord et al., 2010)** to provide a positive outcome.

SfBB Assessment Questionnaire – SfBB-Q Design Evaluation

The novel SfBB questionnaire was developed specifically for use in this study to collect and capture relevant quantitative feedback on the novel SfBB resources created by the Sidney De Haan Research Centre (Appendix C, sub-appendix I). The questionnaire design is similar to the format of the CAT questionnaire but consists of eight items, each formatted as a numerical response scale from zero to six.

The eight items were as follows:

1. I used the booklet and/or DVD regularly
2. I found the instructions in the booklet easy to follow
3. I found the instructions on the DVD easy to follow
4. Using the booklet and DVD made me feel more confident at the group sessions
5. It was easy to fit in regular practice at home
6. I will continue to use the booklet and DVD to practice
7. I would recommend the booklet and DVD to others
8. I would be likely to use this or other singing resources online or via an electronic device, mobile phone ‘app’ or similar

The participants were all familiar with the CAT questionnaire and since the SfBB-Q was created to be similar in design it was generally well accepted by participants and allowed for some useful evaluation of the SfBB tools. The questionnaire was administered easily with little instruction.

Several participants posed verbal queries to their neighbouring participants or the researcher during the administration of the questionnaire which were all pertaining to a similar theme regarding the 'direction of the scale' e.g. 'Is a score of zero the best and six worst?', "Is a zero good?" indicating that this was the key area needing clarification for improved understanding and ease of administration.

In two cases participants completed the scale in error and then corrected some responses. In just one of these cases (Participant ID #2010) some early responses then appeared completely contrary to later statements which lead to a supposition that participants completing the questionnaire may have been confused with the direction of the scale. There was only one in total, out of a possible twenty-eight that completed the questionnaire, where this showed up in the results.

Though the aim of the design was to make the questionnaire similar to the CAT to aid the users that were familiar with this questionnaire, it was a clear limitation of the design process that it was not noticed that the CAT responses are slightly different from those of the SfBB-Q. i.e. while the CAT scoring is from zero to six, where zero is the most positive and six is most negative the SfBB questionnaire includes some questions that relate to a frequency variable i.e. 'frequency of use' where participants inherently might feel that a 'more frequent' response should be recorded as a higher numerical value on the scale. Therefore, it might be advantageous to consider reversing the scale for the SfBB-Q to allow the higher value of six to indicate the most positive score or value of maximum frequency. One other option, if it is to be used again, might be to leave the scale is it stands but to add a further comment in the questionnaire instruction to make it more easily understandable when administered.

It was also noted that three participants recorded a value of six for questions regarding the ease of use of a specific item e.g. *'I did not find the instructions on the DVD easy to follow'* indicating that they did not find the instructions easy to follow. However, the same participant had also indicated by written comment on their evaluation form that they were not able to use the DVD, hence this could outline a further limitation of the questionnaire i.e. In future participants should be instructed to refrain from answering questions that are not relevant or to include an additional 'Not applicable' option.

To measure the internal consistency or correlation between different items on the SfBB scale, the Cronbach's alpha value was calculated as $\alpha = 0.813$, where internal consistency (α) ranges between 1 and 0. A value $0.8 \leq \alpha < 0.9$ is deemed 'Good' and a value $0.7 \leq \alpha < 0.8$ is 'Acceptable' with a value of $\alpha < 0.5$ being 'Unacceptable' **(George and Mallery, 2003)** indicating

there was ‘good’ or adequate internal consistency for this questionnaire within this population overall.

It was also noted from this analysis that Question 5 on the scale i.e. ‘It was easy to fit in regular practice at home’ was the only question on the scale to show negative correlation in the Inter-Item Correlation Matrix and as such showed a Corrected Item-Total Correlation score of 0.214. Since this value is between 0.2 and 0.7 it would still indicate validity, but is of note as it was the only question to have a ‘revised Cronbach’s alpha score if the item were deleted’ higher than the overall original Cronbach’s alpha i.e. α = 0.839 for the scale. This might indicate this question was not as reliable as the other questions. It might therefore be a consideration to remove or revise this question if using the questionnaire in future.

Question 1 on the scale i.e. ‘I used the booklet and/or DVD regularly’ showed a Corrected Item-Total Correlation score of 0.378 which was slightly lower that the other remaining questions too. However it was still well within the range of 0.2 to 0.7 and the revised Cronbach’s alpha score ‘if the item were deleted’ was still lower than the overall Cronbach’s alpha i.e. α = 0.810. All other questions recorded high Corrected Item-Total Correlation scores > 0.5 and revised Cronbach’s alpha score ‘if the item were deleted’ less than the original Cronbach’s alpha score, indicating that the questionnaire was reliable and removal of any question in this case, except question 5, would result in a lower Cronbach's alpha and thus lower reliability.

The initial intent of the SfBB-Q was to calculate an overall mean score per patient, summarising their feedback, which could then be comparable between participants and across the study. However, since the values from each of the questions can be quite variable and inconsistent due to the limitation of the SfBB-Q, the analysis of each individual response may hold more useful information on the usability of the SfBB guide and future use and uptake of such similar self-management resources.

These resources or alternatively, investment in further development of similar materials could help improve the quality, consistency and the persistence of the singing intervention and delivery method for future research. This has been outlined as an essential component from this and previous research **(Clift et al., 2013)** and this needs to be considered with more focus for future research.

The SfBB-Q form provided useful feedback and was readily accepted and completed by participants. The qualitative data was very minimal but it did support the findings from the SfBB-Q. The key learning from this pilot administration is that the SFBB-Q questionnaire would need further adaptation and improvement if it is to be used in future analysis. This would ensure it is more easily understood by participants, can be self-implemented by participants without additional instruction or advice and can provide better quality data. Qualitative feedback in the form of verbal group discussions or via participant interview techniques with questions dedicated to providing insight into how the resources were utilised would also be strongly

recommended for future studies, This could extract valuable tacit information from participants to help improve the rollout and uptake of such resources in future.

Chapter 6 – Discussion and Conclusions

The overall aims of this research were to answer the following questions:

1) What has been established so far by research regarding the effects of group singing for COPD patients?

2) What are the potential health and wellbeing benefits of regular participation in group singing for a small cohort of COPD patients?

3) What is the potential feasibility and usefulness of developing and implementing novel singing resources in a group of COPD patients, in addition to regular group singing?

The first question 'What has been established so far by research regarding the effects of group singing for COPD patients?' was addressed by execution of a broad systematic review of all available published research on singing for COPD to date, to explore all the evidence and supporting information available in this area and to use this knowledge where possible to inform future study design.

The key messages from this systematic review and the data for those studies included were generally very positive, with the majority of the studies reporting one or more positive finding and none of the studies concluding any negative findings. However, the overall conclusion of the review is in close alignment with the recent Cochrane review **(McNamara et al., 2017)**, such that, the availability of high quality evidence that singing for COPD improves physical health, dyspnoea or respiratory-specific quality of life is still limited, largely due to the low number of studies and their small sample sizes. This theme also aligns with arts and health studies more broadly, as discussed in a report for arts, health and wellbeing which found that evaluation still presents challenges and there is a need to address difficulties such as the use of randomisation, control and adequate sample sizes in this area of research **(RSPH, 2013)**.

A core study (Part I) was designed using information from the outcome of this systematic review to address the question 'What are the potential health and wellbeing benefits of regular participation in group singing for a small cohort of COPD patients?' and while it was not feasible to design a study at this time which met the recommendations outlined for large global randomised trials, the durations, population, measures and other aspects were informed by the outcome of the systematic review and the study did meet its aim in addressing this question. It also achieved its primary endpoint to assess the mean total difference scores from baseline on three health and wellbeing questionnaires (WEMWBS, CAT and ACQ-5) at twelve weeks post baseline (i.e. Follow-Up Visit V_2, Week 12 $\pm$14 days). The mean improvement in WEMWBS and CAT scores between Baseline Visit V_0 and Follow-Up Visit V_2 met required significance i.e. $p \leq 0.05$ and this required significance was maintained at Final Follow-Up Visit V_3 for those

participants that continued. The results obtained from this scale showed a change of five points which was maintained after thirty weeks post baseline. This finding is clearly very encouraging in regard to the impact of regular singing on COPD symptoms, though caution must be exercised regarding the small sample size and low power and also the low quality of the study due to the pre-test post-test design.

It also employed secondary endpoints which included assessment of the mean score differences of these questionnaires at six weeks post baseline (i.e. Mid-Term Study Visit V_1 (Week 6 $\pm$14 days) and again at thirty weeks post baseline (i.e. Final Follow-Up Visit V_3 at Week 30 $\pm$21 days) along with a review of written feedback, compliance and retention data also collected at baseline and twelve weeks. This core study aimed to address the question of whether participants engaged in group singing for COPD, including the use of new resources, express an improvement in any of these measures at the various time points. The clear indications from this study show there are some significant improvements.

The research further included a secondary endpoint which aimed to answer the third research question: ‘What is the potential feasibility and usefulness of developing and implementing novel singing resources in a group of COPD patients, in addition to regular group singing?’ It addressed this using a secondary sub-study (Part II) which piloted newly created self-management resource tools, developed specifically for singing for adults with COPD and respiratory illness. The intent of this sub-study was to provide deeper understanding of the usability and feasibility of these specific resources and also the potential use more generally of these types of tools for this population to provide an insight into any key themes for consideration in future use. In addition, this provided opportunities to examine the research instruments i.e. potential questionnaires and techniques involved in reviewing such tools for group singing in this population.

It was found that the use of novel singing resources in a group of COPD patients, in addition to regular group singing is a feasible enterprise, as implementing a variety of media to present audio or visual recordings was found to work well in this small population. The majority (78%) of participants indicated they would recommend the resources that had been provided and 67% agreed that they would use these resources of other types of media to practice in the future. These results were encouraging but this was a small population and the study had a high risk of bias due to lack of randomised control or blinding, so clearly there is still much to be explored to ensure effective design and delivery of these tools to maximise uptake and usability for COPD patients to help encourage continued singing practice.

Methodology, Questionnaires and Measures

A European Respiratory Society (ERS) task force report was published with the aim of establishing clear diagnostic criteria and standardized methods to examine COPD and it

strongly recommended measuring as many different characteristics of COPD patients as possible (e.g., respiratory symptoms, exacerbation frequency, comorbidity assessment, body mass index (BMI), biological markers, chest radiography, and risk factors assessment) in addition to spirometry, to provide a better understanding of the disease **(Bakke et al., 2014)**.

A recent study reviewing the diagnosis of COPD in epidemiological studies concluded that single spirometry or clinical respiratory symptoms alone are not enough for accurate COPD diagnosis. It further suggests that a comprehensive approach, including clinical assessment and follow-up spirometry should be taken into consideration for the diagnosis and management of COPD as well as for any screening program or prevalence study conducted in the future **(Andreeva et al., 2017)**.

The strategy of recording as many different characteristics of COPD patients as possible is advantageous and certainly more easily achievable for large epidemiological studies. However, in 'Singing for COPD' research, where budgets may be limited, and sample size of the available trials is small, it would be more advantageous to utilise consistent outcome measures wherever possible across trials so that results could be more widely compared. A key finding from the wider systematic review, as documented in Chapter 2 is that there is such a wide variety of questionnaires and measures used across the body of work all with varying success and small sample sizes, that it makes it very challenging to compare or pool data for analysis to draw conclusions. This is supported by a recent Cochrane review **(McNamara et al., 2017)**. There are also important new technological advances in lung function that may be more pertinent, viable for fast, accurate data collection in future studies e.g. the application of Structured Light Plethysmography (SLP), a non-invasive diagnostic method that uses structured light to perform pulmonary function testing **(de Boer et al., 2010)** already holds significant potential in this area.

The research forms and measures were selected for this research within the confines of the community setting i.e. without access to specialist or diagnostic equipment (e.g. weighing scales, spirometers) or trained personnel etc. Several alternative measures were explored during the early planning phase, for feasibility, including measures used by previous studies e.g. St. George's Respiratory Questionnaire (SGRQ), Spirometry measures, BODE Index, HRV recordings and plethysmography but due to the additional data collection requirements i.e. requests for detailed health measures, requirements for specialist equipment etc. and due to the additional informed consent and ethical hurdles, this was not considered to be feasible within this group. If future larger studies are conducted in a healthcare or hospital setting however, it would be recommended that a full review of wider measures is considered.

The overall view on the selected measures was that these were effective, well received and permitted a significant amount of quality data to be collected in a simple and unobtrusive way, within the time available and logistical constraints. The participants were provided pens, time and space to complete their forms and were encouraged to ask for guidance from the

researcher regarding completion, but questions were limited, and all participants completed feedback wholeheartedly and without any objection.

One other more minor consideration was the cost of the usage of the questionnaire. The resources available to conduct such trials in this area of study are limited and the most cost-effective options were selected to capture pertinent data and support the overall aims of this research. It is anticipated that future studies in this field, as in arts and health research more generally, would likely be conducted by small self-funding groups with minimal budget such as this study. Therefore, low cost solutions were selected with the expectation that other researchers would be encouraged to use the same inexpensive and easily accessible questionnaires within their studies in future, so more effective consolidation of data from multiple trials and improved comparisons of research results might be made.

Research Limitations

The research limitations noted were similar or the same as other singing for COPD trials conducted in this area due largely to the impossibility of blinding participants to the nature of the intervention and leaving the results exposed to an unmitigated risk of bias i.e. risk of 'expectation of benefit' and risk of potential desire by the participant to please or impact the results. The lack of a control group here further exposes the study to the risk of assessor bias. A credible randomised control with larger enrolment numbers would have eliminated much of the risk of bias, though this was not possible within the confines of the resources available and would be recommended in future. It is recommended that future studies employ some form of randomised control and that the assessors are blinded to the intervention.

In addition the questionnaires and outcome measures were not completed by the participant in isolation e.g. in a quiet space without distraction, so there was a potential for participants to be influenced by singing leaders, assessor and other participants, which may have introduced additional bias. It is recommended that participants are assessed in isolation where possible for future research, to help mitigate this potential for bias.

The total outcome measures were not extensive and the data collection for these patients was deliberately limited to make effective use of the resources i.e. the time availability at the venues, the singing leaders' availability, to ensure all required engagement and administration of the measures was respectful of participants' time and energy reserves. The deliberate selection of these simple measures was made to allow data collection to be as non-invasive on participants' time and to be as efficient as possible. Those previous randomised control studies **(Bonilha et al., 2009, Lord et al., 2010, Lord et al., 2012)** and the larger scale community studies **(Clift et al., 2013, Clift et al., 2017)** conducted in this area had the advantage of being completed either in a hospital setting or with larger clinical support teams available which allowed for additional physical and spirometry readings to be taken etc. However, despite the limitations of this study

it was conducted with rigour and the data collected was robust even though the study would be considered low quality due to the lack of randomisation, blinding or control.

The attrition rate of the original participants that consented (n=37) was approximately 50% over all up to the final assessment reflecting what has been seen previously in larger studies with a similar population **(Clift et al., 2017)** and as noted there, a sufficiently powered future trial would need to take this attrition into consideration. It is again a limitation of this study that larger population could not be recruited due to resource constraints i.e. suitable locations, availability of singing leaders etc.

This research strongly supports previous findings of similar studies in this area **(Bonilha et al., 2009, Lord et al., 2010, Lord et al., 2012, Clift et al., 2013, Clift et al., 2017)** that group singing does positively impact both health and wellbeing outcomes of these patients in a variety of ways. In addition, it further suggests that if sustained, this intervention has the potential to support maintenance of these positive effects over longer periods, even if there is a break in the group singing intervention.

Final Conclusions

The broad systematic review detailed here provides further evidence in alignment with the recent Cochrane review **(McNamara et al., 2017)** that the availability of high quality evidence to support singing for COPD improving physical health, dyspnoea or respiratory-specific quality of life is very limited due to the low number of high quality studies. Existence of research examining the longer-term effect of singing for people with COPD with large samples sizes is limited and randomised controlled data with large samples over extended periods is non-existent.

It can be concluded that although lung function is poorly related to other clinical outcomes **(Mahler and Criner, 2007)** and it is essential to pursue the use of measures and outcomes beyond lung function or spirometry alone, the variety of different measures currently being used is too overwhelming and there is a lack of standardisation in this area. This issue applies to study methodology and design too, including intervention type, frequency and duration. Global or at least regional standards in this area would allow more efficient collaboration and potential for shared data and pooled analysis. It is important that health care utilisation, service use and cost effectiveness of the intervention is also considered and further explored over the longer term to increase the impact of future trials.

The empirical study research here makes a useful contribution to the growing literature on the potential value of regular group singing for people with COPD. The study was designed to build upon the earlier studies conducted in South East England **(Clift et al., 2013, Clift et al., 2017)** and to assist in understanding further, the feasibility of potentially conducting a larger

randomised trial in the Medway area. In this context it successfully met its objective in addition to the primary aim which was to show group singing had a significant positive effect on mental wellbeing as measured by the Warwick-Edinburgh Mental Wellbeing Scale (WEMWBS) and a significant positive effect on the impact of a patient's COPD and how this changes over time, as measured by the COPD Assessment Test (CAT). It also concluded that despite a short cessation in group singing sessions this effect was maintained at thirty weeks post baseline.

This research is low in quality with regard to risk of bias, sample size and low power, largely due to the lack of a randomised controlled arm and the pre-test post-test design, which negatively impacts the overall confidence in the results. Nevertheless, it does indicate that significant improvements could occur within this population in only a very short timeframe. It suggests that just a one-hour session continued weekly for approximately twelve weeks could provide significant benefits to wellbeing and impact to COPD symptoms. The data further supports these effects being maintained up to thirty weeks with continued weekly sessions, even with a short cessation in group singing attendance.

This research highlights the importance of continued efforts to encourage this offering in this population particularly following initial pulmonary rehabilitation sessions to ensure prolongation of the effects. As previously discussed, researchers should continue to strive to use more reliable and valid quantitative measures in this area of research and ensure that a framework is used to promote use of measures that are validated for the specific population. It would be ideal if the leaders within this research community could agree consensus regarding the key measures used so that future study data is collected more consistently so that study data across multiple studies could potentially be pooled.

Consistent with recent research **(Skingley et al., 2018)**, the qualitative data provided by participants was in general agreement that the singing groups contributed to their improved state of wellbeing and the majority of participants in this research study agreed that the singing programme led to improvements in respiratory symptoms and in their wellbeing, confidence and social connections between baseline and follow-up visits. The COPD participants noted improvements in mental wellbeing, attributing much of this to the singing group. Social benefits were also in evidence, extending to the groups meeting outside of the singing sessions and often acting as external support networks. The data collected within this study supplements both the current qualitative and quantitative data in this area and adds to the growing body of evidence that singing is a safe, enjoyable pastime that helps health and wellbeing for adults with a respiratory illness.

This pre-test post-test study was designed to review the impact of group singing on adults with COPD within the Medway area and to pilot newly devised tools for the specific purpose of home support within the COPD population. This study shows significant positive outcomes and contributes to the expanding body of evidence on the potential value of group singing for adults with respiratory illnesses more broadly and particularly adults with COPD. This research

supports and builds on existing evidence indicating that group singing may offer many potential health and well-being benefits for adults with COPD related respiratory conditions **(Clift et al., 2017, Bonilha et al., 2009, Lord et al., 2010)**.

Recommendations for Future Research

The collective recommendations from the systematic review and the research studies are that, it is essential that researchers build on existing knowledge and that, longer, larger, multi-centre global randomised controlled trials are conducted with increased standardisation of measures between studies. These should certainly extend to provide more consistent data across diverse regions and countries with low, middle and high incomes and target more diverse COPD populations.

These findings are consistent with arts and health literature more broadly and in particular with a review of art, healing, and public health **(Stuckey and Nobel, 2010)** which collectively reviewed four primary therapies (music engagement, visual arts therapy, movement-based creative expression and expressive writing) suggesting researchers should make better attempts to establish meaningful control groups, to quantify interventions and outcome variables at higher levels of standardisation and precision allowing more cross-study comparison. It also suggested similar to the outcomes of this research, that future research should expand study populations to allow exploration of the effects of interventions in groups, and should plan for longer term follow-ups to assess the sustainability of outcomes over time.

It is strongly recommended based on that the group singing intervention for future studies includes at least one hour of regular singing and exercise each week and that participants are encouraged to practice in between sessions. Careful consideration and planning should be undertaken regarding logistical matters. It is recommended that selected venues singing and assessments have adequate access (ground floor level facilities where possible) and are central for public transport and with good parking facilities.

It is also suggested that at least thirty minutes is allowed at the start and the end of singing sessions to allow for setting up the space in readiness for the participants and clearing everything away afterwards, so that patients with respiratory needs do not feel ‘rushed’ and stressful situations are avoided. It would also be ideal where possible for questionnaires and outcome measures to be completed by participants in isolation and further recommended that assessors and data collection teams are blinded to the intervention.

The intervention period of the empirical study was conducted from spring to autumn in South East England, with the majority of assessments performed specifically in the spring and autumn, avoiding excessive temperature and environmental changes potentially experienced in summer and winter months. The temperatures were considered mostly temperate during the intervention

period, which allowed researcher and singing leaders to accommodate participants with COPD symptoms during these periods, since the condition is often impacted by temperature, allergens and other pollutants usually associated with more extreme seasonal climates both positively and negatively. Research suggests a distinct and independent influence of season on exacerbation outcomes and mortality **(Rabe et al., 2013)** and participants' conditions i.e. dyspnoea combined with disease progression can cause inability to travel, especially in winter months **(BLF, 2017)**. The arrangement of sessions around the seasons in this study appeared to positively support recruitment and retention of participants and avoided seasonal changes that could potentially have impacted assessment variables as seen previously **(Clift et al., 2017)**. It is recommended that future research planning strongly take this into account.

The recruitment and retention of participants into research trials has become more sophisticated in recent years. Researchers are more widely pursuing extensive feedback from the target population early on during feasibility planning to ensure input for study design, timings and logistics for particular populations is provided. It would be useful if this data could be more critically discussed in the planning of the research and delivery of group singing sessions within these studies too. This could potentially help to reduce attrition rates. It was not feasible to employ more sophisticated measures in the case of this study due to the time available and nature of the study but for larger complex randomised studies it is recommended.

It is recommended, that future work might involve a further comprehensive mapping of collective qualitative interview output for 'singing for adults with COPD' too, since there could be an extensive trove of this information from previous studies which has not yet been fully assessed holistically and this might direct future work. It is also recommended that sufficient time is made available during study planning to collect additional feedback comments and to ensure there is time or space available beyond the singing groups to allow time for socialising. This was considered valuable by the participants of this research and is something that could also potentially help with retention of participants.

The data collected for this research was limited to outcome measures to assess impact of COPD on participants' life and quality of life, but other symptoms and measures would be very desirable for future research i.e. Pulmonary function tests, other more detailed dyspnoea scales, changes in breathing pattern during and after singing, physical measures, hormone changes and effect on frequency and amount of expectoration of sputum. This might expand the knowledge base and also help understand more fully the physiological and psychological aspects and mechanisms of action that group singing may have on COPD symptoms.

The predominant health-care cost item for COPD patients is hospitalizations for exacerbations and high priority should be given to interventions aimed at delaying the progression of disease, preventing exacerbations and reducing the risk of comorbidities **(Lopez-Campos et al., 2016)**. It has been shown that the provision of opportunities to meet and sing together provide cost-effective and acceptable options for maintenance and enhancement of health in other

populations **(Coulton et al., 2015)**. However, much more robust data in these areas are clearly vital for the COPD population. A particular need is the exploration of service use and cost of COPD group singing over a longer period, across greater numbers of COPD patients with differing severity of disease and particularly in a randomised trial setting.

In alignment with previous recommendations and ideals **(BLF, 2017)** it is important that the consideration of sustainability of groups beyond the research period remains high on the agenda for any forthcoming projects. The singing group intervention in this study was so well received that there were feelings of concern and possibly even anxiety raised by participants as the study drew to a close, as to how the groups might continue. These issues were raised and discussed on many occasions and were a theme within the verbal and written comments provided by the majority of participants. It is therefore a recommendation that considerations of sustainability are addressed with participants from the outset wherever possible.

The findings of this research, including both the systematic review and the empirical research are consistent with the fundamental outcomes outlined in other recent reviews **(Lewis et al., 2016, McNamara et al., 2017)** and studies **(Bonilha et al., 2009, Lord et al., 2010, Lord et al., 2012, Clift et al., 2013, Clift et al., 2018)** in this area, in that there are critical limitations in the current body of evidence and it is vital that larger randomised controlled trials with longer durations and robust methodology be conducted to build on the existing evidence.

Ideally these trials should extend across more diverse regions globally and should provide more consistent data collected more frequently with a wider outreach to include a broad range of societies, incomes and educational levels. There should also be a focus on long-term follow-up data, regularity and consistency of group singing and consistency of the types of quantitative data collected to support core policy making in the future.

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Appendices

Appendix A – Initial Systematic Review Protocol

Singing for Chronic Obstructive Pulmonary Disease

A Systematic Review of Currently Published Literature [1]

Version – Final 1.2

Author: Charlotte Epsley

Protocol Summary

Abstract [(2)]

Respiratory disorders manifest themselves with chest symptoms including shortness of breath and a decline of lung function and a therapy is needed to provide treatment and support for these patients. There is a growing body of evidence to suggest that therapies that include breath control exercises, such as singing, may have health benefits for respiratory function and psychological well-being.

The objective of this review is to search the current body of evidence and evaluate the evidence available for singing as an intervention for health and wellbeing for patients diagnosed with a respiratory illness.

Terms: Singing, Respiratory Illness, Respiratory disorder, Chronic Respiratory disease

Table of Contents

Introduction

Rationale [3]

Respiratory diseases are among the leading cause of death worldwide. Lung infections (mostly pneumonia and tuberculosis), lung cancer and chronic obstructive pulmonary disease (COPD) together accounted for 9.5 million deaths worldwide during 2008, one-sixth of the global total. The World Health Organization (WHO) estimates that the same four diseases accounted for one-tenth of the 'disability adjusted life-years' (DALYs) lost worldwide in the same year.

Respiratory diseases are likely to remain a major burden on European societies for decades to come. The prevention and treatment of lung diseases will need to be improved, if their impact on longevity and quality of life (QoL) of individuals and their economic burden on society are to be reduced in Europe and worldwide **(WHO 2012)**.

Respiratory Disorders manifest themselves with chest symptoms including shortness of breath and a decline of lung function. Singing is an activity that may support lung function and enhance quality of life in people with certain respiratory disorders.

Bonilha et al. (2009) concluded, based on a small, randomised study of COPD patients, that singing classes are a well-tolerated activity for selected patients and that the regular practice of singing may improve QoL, and preserve the maximal expiratory pressure of these patients. The feasibility study on the health benefits of a community singing programme for older people with COPD by **Clift et al. (2013)** further supports this, showing that measures of lung function and health-related quality of life significantly improved over a 10 months period of the singing programme.

This research is in line with existing UK Department of Health (DH) Chronic Obstructive Pulmonary Disease (COPD) outcomes strategy discussions, particularly objectives identified in the UK Department of Health companion document **(DH May 2012)**, which outlines directions for enhancing quality of life for these patients.

The evidence suggests that singing has the potential of improving health outcomes in patients with respiratory illness. However, although individual systematic reviews of research literature have been completed for singing in patients with certain specific respiratory disorders for example 'Singing for Cystic Fibrosis' and 'Singing for Bronchiectasis' **(Irons et al., 2010*a*, Irons et al., 2010*b*)**, a systematic review of singing for chronic respiratory disorders collectively and in particular COPD, has not yet been carried out.

This systematic review aims to research current knowledge in the area of singing for respiratory illness specifically those collectively known as COPD, with the intent to consolidate existing

evidence in one place and support future research methodology and practice development. This could potentially impact patient welfare positively and reduce health care utilisation for patients with impaired respiration.

Condition

Respiratory illness encompasses developmental, pathological conditions affecting the organs and tissues that make gas exchange possible in higher organisms, and includes conditions of the upper respiratory tract, trachea, bronchi, bronchioles, alveoli, pleura and pleural cavity, and the nerves and muscles of breathing **(WHO, 1978)**.

The respiratory tract is the site of an exceptionally large range of disorders for three main reasons:
1) It is exposed to the environment and therefore may be affected by inhaled organisms, dusts or gases
2) It possesses a large network of capillaries through which the entire output of the heart has to pass, which means that diseases that affect the small blood vessels are likely to affect the lung
3) It may be the site of “sensitivity” or allergic phenomena that may profoundly affect function **(Britannica, 2018)**

Respiratory disease in the context of this review refers to one of those disorders included in the list of common chronic respiratory diseases as identified by WHO classification 2007 and is noted along with the related symptoms in Appendix I of this document.

Intervention

The intervention of singing and the suggested method of action are summarised very comprehensively within Cochrane review protocol ‘Singing for Adults with COPD’ **(McNamara et al., 2016)** as detailed verbatim below:

Intervention - description **(McNamara et al., 2016)**

> 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 et al., 2010*b*).** 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 et al., 1987, Leanderson and Sundberg, 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 et al., 2011)**. Trained singers have greater breathing efficiency and greater use of their lung capacity than non-trained singers **(Gould and Okamura 1973, Salomoni et al., 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 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 et al., 2010*a*)**.

Intervention - suggested method of action **(McNamara et al., 2016)**

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 et al., 2013)**. 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 **(Clift et al, 2013, Skingley et al, 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 et al., 1996)**, alleviate dyspnoea and improve quality of life **(Gosselink, 2003, Gosselink, 2004)**, and improve functional exercise capacity **(Holland et al., 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 et al., 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 et al., 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 et al, 2010)**.

Objectives (4)

The key objective of the review is to systematically search the existing published body of research on singing for respiratory illnesses, and to formally present findings to enable comparison, assessment and summarisation of current knowledge.

Primary Endpoint

Assessment of singing for pulmonary function in patients with a respiratory illness.

Secondary Endpoint

Assessment of singing for quality of life and wellbeing in patients with a respiratory illness.

Methods

Protocol (5)

The conduct of this study protocol is strongly guided by the framework outlined in the Cochrane Collaboration Handbook, 2011 **(Higgins and Green, 2011)** and based on the reporting principles of PRISMA statement guidelines (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) **(Prisma, 2015)**. The heading numbers in parentheses throughout this protocol, refer to the PRISMA checklist headings detailed in Appendix V.

This protocol is not currently registered.

Eligibility Criteria (6)

Criteria for considering studies for inclusion in this review are as detailed below:

Study types

Randomised controlled trials or quasi experimental designs and recorded in any language and meeting relevant eligibility criteria below.

Patient Inclusion Criteria

Studies where the patients included have one or more diagnosed respiratory disorder (defined clinically or radiologically) as identified under the subheading of 'Chronic obstructive lung disease' in the list of chronic respiratory disorders outlined by the World Health Organisation (WHO) in Appendix I.

Intervention Inclusion Criteria

Studies where diagnosed patients of any age have participated in programs that include singing activities, carried out in a group (choir) or one-to-one setting with a singing teacher or instructor for a minimum of two half-hour sessions.

Outcome Measures [13]

Primary Outcomes

1. Mean difference in respiratory function from baseline that reflects respiratory muscle function (maximal inspiratory and expiratory flow, cough peak flow)
2. Mean difference in other lung function indices from baseline (Spirometry or other lung volumes)
3. Mean difference in other subjective scores (Likert scale, Visual analogue scale (VAS), Level of interference of Cough, Cough diary, etc.)
4. Mean difference in quality of life (QoL) from baseline

Secondary Outcomes

1. Proportions experiencing adverse effects of the intervention
2. Total number of hospitalised days or symptomatic days
3. Proportions of participants who had respiratory exacerbations and/or hospitalisations
4. Treatment adherence + retention in the study
5. Psychological assessments measuring well-being, self-efficacy, depression and anxiety
6. Physical function assessments

Information Sources (7)

Searches will target the key bibliographic databases noted in Appendix II to identify relevant studies.

Searches of other resources will also be carried out for completeness i.e.
1. Searches of other databases and search engines, including grey literature
2. References cited in relevant publications or included at recent conference proceedings
3. Communication with the authors of trials included in the review.

Search (8)

Specific search criteria will be applied to the key bibliographic databases detailed in Appendix II. This is broken down into three key components to identify all unique research studies where singing is an intervention in this specific patient population. The search criteria will be developed by combining searches for the following terms:

‘Intervention’ term

The term ‘Singing’ is the intervention of interest and following initial preliminary investigation during pilot searches, singing is a 'Medical Subject Heading' (MeSH) term and delivers by far the highest specificity of literature in most key bibliographic databases. Those databases, which do not recognise MeSH terms, will be searched to retrieve studies where ‘singing’ is recorded as a key term.

‘Population’ term

The term Respiratory illness is also a MeSH term and following initial preliminary investigation during pilot searches delivered by far the highest specificity of literature too. Databases, which do not recognise MeSH terms, will be searched for ‘respiratory illness’, ‘respiratory disease’ or ‘respiratory disorder’ as a key term (depending on the individual database definitions). Key additional respiratory terms selected from those identified by WHO as detailed in Appendix I will potentially be used as an alternative in those databases which are require more specificity or which do not recognise generic ‘respiratory’ terms. All searches are to be documented for reproducibility.

Reverse searches and quality checks have been completed on MeSH terms to ensure relevant illness/disorder terms and singing terms are included prior to use.

The results of these two searches are merged to provide a list of unique literature in preparation for manual review for those studies that potentially meet the inclusion criteria.

Study Selection [9]

The abstracts of those studies retrieved by the search process, along with the title and descriptors, are to be reviewed independently by at least two reviewers. The reviewers will identify all studies in which singing has been observed as an intervention for patients with a diagnosed form of a respiratory illness. Only studies clearly unrelated to the area of interest are to be excluded at this stage. A final list will then be agreed, and full details retrieved for all available included studies.

Data Collection Process [10]

The full details for all studies selected by the initial screening process and appearing to meet eligibility criteria will be independently reviewed comprehensively by at least two reviewers. The key data will be extracted and collated for comparison in a specifically developed data collection tool using **Microsoft XLS™**.

Studies identified are to be further screened at this stage to exclude studies where one or more of the measures identified in Appendix III have not been assessed at baseline and $\geq$1 other time-point or visit.

If any disagreement occurs regarding inclusion of a study either at this stage or at the search stage, this is to be resolved by third party adjudication.

Data Items [11]

Data points identified in Appendix IV will be considered for each eligible study, extracted and captured in the data collection tool. If data are missing, the field will be noted as NA (Not applicable) or UNK (unknown) or an explanatory comment will be recorded.

If important key data impacting an endpoint appear to be missing or unclear, an attempt will be made to contact the study author(s) to acquire additional information or clarification.

Reviewers will compare and discuss outcomes to deliver a final agreed version of the database.

Risk of bias in individual included studies [12]

Studies are to be evaluated on:
(i) Methodological quality - extent to which design and conduct are likely to have prevented systematic errors
(ii) Precision - measurement of likelihood of random errors (usually confidence interval width around result)

(iii) External validity - extent to which results are applicable to patients with a respiratory disorder and other populations

The risk of bias will be assessed for all studies, following criteria outlined in the *Cochrane Handbook for Systematic Reviews of Interventions.* **(Higgins and Green, 2011).** The Cochrane Collaboration's tool for assessing risk of bias shown in Appendix VI will be utilised to support reviewers in critically assessing bias for various domains within the review.

To assess the study level risk of bias the quality of the studies included will be assessed with reference to the following domains and graded with either a 'low', 'high' or 'unclear' risk of bias:

(i) Selection/Randomisation bias
(ii) Performance bias
(iii) Detection/Blinding bias
(iv) Attrition/Incomplete data bias
(v) Selective reporting bias
(vi) Other (e.g. design-specific)

Those assessing risk of bias will not be blinded to the names of the study authors, institutions, journal or results of a study.

Quality Appraisal

The findings will be critically assessed, identifying common themes with a particular focus on the study design, intervention details and measures implemented for existing research. It is thought that these components in particular could be potentially important in supporting the direction and design of future research in this area.

Synthesis of Results and Data Analysis [14]

Analysis of the impact of the intervention on the quantitative outcome measures will be the focus of this review. The nature of the intervention in studies included may have some 'mixed method' designs or will have elements of qualitative data reported, i.e. within patient interviews or comments. This information could potentially enhance the relevance of the review and help with interpretation of the quantitative data results. While the qualitative data will not be included in the review itself, a parallel synthesis approach will be adopted to ensure pertinent qualitative concepts or themes are synthesised for inclusion in the discussion.

Quantitative Data

A review will be conducted for all available meta-data extracted for primary and secondary endpoints, to assess if this is comparable. If the same efficacy scales of interest have been used (e.g. Lung function measures) and meta-data are comparable, then the data will be pooled and the mean difference (MDs) and 95% confidence interval (CI) change from baseline will be calculated. These results will be compared with established published standards to suggest if any clinical significance may be determined.

Qualitative Data

Qualitative evidence will be assessed and coded for any distinct and comparable themes or concepts. These will be grouped and discussed separately and if deemed relevant, incorporated or commented upon in the overall review discussion.

The differences in study populations, inclusion/exclusion criteria, interventions, design and outcome assessment will be compared and discussed for all studies selected. Details of studies excluded from analysis will be recorded with explanation.

Risk of bias across included studies (15)

Risk of bias affecting the cumulative evidence acquired i.e. publication bias or selective reporting bias will be considered and discussed if sufficient publications are included. If the combination of data and meta-analysis (with at least 10 studies) is possible, publication bias will also be assessed using methodology according to the recommendation of the Cochrane Collaboration Handbook **(Higgins and Green, 2011)**.

Investigation of heterogeneity (16)

If there are a sufficient number of studies meeting the inclusion criteria then the clinical, methodological and statistical heterogeneity between included studies will be investigated using guidance from the Cochrane Collaboration Handbook **(Higgins and Green, 2011)**.

Heterogeneity between the study results will be examined, described and tested to see if it reached statistical significance. The I^2 statistic will be used to measure heterogeneity among the trials in each analysis if sufficient data are available. Interpretation of statistical heterogeneity will be according to the recommendation of the Cochrane Collaboration's tool **(Higgins and Green, 2011)**.

(i) 0% > I^2 ≤ 40% indicates low heterogeneity
(ii) 30% > I^2 ≤ 50% indicates moderate heterogeneity
(iii) 50% > I^2 ≤ 90% indicates substantial heterogeneity

Where substantial heterogeneity (I^2 > 50%) is identified, it will be explored using a pre-specified subgroup analyses where possible.

Potential subgroup analysis (16)

The following subgroup analyses may be considered, where sufficient studies can be included, dependant on assessment of results:

1. Children (aged 18 years or less) and adults (>18 years)
2. Participant type (Respiratory Disorder as primary disease vs Respiratory Disorder as co-existent disease)
3. Type and length of singing intervention (e.g. short term: <1 month; medium term: 1-6 months; long term >6 months; type of training)
4. Stable state vs exacerbation phases of Respiratory Disorder
5. Severity of Respiratory impairment (based on FEV1: >80% classified as mild, 50-79% classified as moderate, 30-49% classified as severe, <30% classified as very severe)
6. Types of singing material used for the intervention

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Appendices

Appendix I - Term selection

(i) Respiratory Disorders

(WHO Table 2 Common chronic respiratory diseases (ICD-10)) (Bousquet et al 2007)

Asthma
Bronchiectasis
Chronic obstructive lung disease
(Incl. chronic obstructive pulmonary disease (COPD), bronchitis and Emphysema)
Chronic rhinosinusitis
Hypersensitivity pneumonitis
Lung cancer and neoplasms of respiratory and intrathoracic organs
Lung fibrosis
Chronic pleural diseases
Pneumoconiosis
Pulmonary eosinophilia
Pulmonary heart disease and diseases of pulmonary circulation
(Incl. pulmonary embolism, pulmonary hypertension and corpulmonale)
Rhinitis
Sarcoidosis
Sleep apnoea syndrome

(ii) Respiratory Symptoms

(WHO Table 3 Symptoms and signs involving the respiratory system)

Haemorrhage from respiratory passages
-Epistaxis
-Haemoptysis
Cough
Abnormalities of breathing
-Dyspnoea
-Stridor
-Wheezing
-Hyperventilation
-Sneezing
Pain in the throat and chest
Other symptoms and signs involving the circulatory and respiratory systems
-Asphyxia
-Pleurisy
-Respiratory arrest (cardiorespiratory failure)
-Abnormal sputum

Appendix II - Selected Research Databases

Trials will be identified from the following key bibliographic database sources:

BNI (British Nursing Index)
CINAHL (1982 to present) Cumulative Index to Nursing and Allied Health Literature
Clinical Trials Register (www.clinicaltrials.gov)
International Clinical Trials Registry Platform (WHO)
The Cochrane Airways Group Specialised Trials Register
The Cochrane Central Register of Controlled Trials (CENTRAL)
EMBASE (1980 to present)
MEDLINE (1966 to present)
OLDMEDLINE (1950 to 1965)
PsycINFO (1872 to present)
Web of Science
ZETOC (Grey literature database)
Music & Medicine
Arts & Health
Singing for Snorers web
ATS - American Thoracic Society
IBSS - International Bibliography of the Social Sciences
PubMED US & EU

Appendix III - Outcome Measures

Studies selected should contain ≥1 of the following measures:

MRC breathlessness scale
Chest x-ray
Chest computed tomography (CT) scan
Arterial blood gas test
Borg dyspnea scale
6-minute walk test (6MWD)
Inspiratory muscle strength
Exercise capacity
Lung function recordings:
- *Maximum voluntary ventilation*
- *Residual lung volume (RV)*
- *Total lung capacity (TLC)*
- *Forced expiratory volume in one second (FEV1)*
- *Forced vital capacity (FVC)*
- *Mean forced expiratory flow during the middle half of FVC*
- *FEV1 / FVC*
- *RV/TLC*
- *Peak expiratory pressure*
- *Peak inspiratory pressure*
- *Spirometry*

Biomarkers:
- *Salivary Immoglobulin A (SIg A)*
- *Cortisol,*
- *Oxytocin*
- *TNF-alpha*
- *Prolactin*

Heart rate
Heart rate variability
Blood pressure
Electromyographic tension
Peripheral skin temperature
Skin conductance
Spielberger's trait anxiety inventory (STAI)
Shortness of Breath Questionnaire (SOBQ)
EQ-5 Questionnaire
St George's questionnaires (SGRQ)
York SF-12
UWIST mood adjective checklist
Positive and Negative Affect Scale (PANAS)
Oxford Happiness Inventory (OHI)
Qualitative patient feedback via patient interviews
Patient statements/comments verbatim
General Hospital Questionnaire-28 (GHQ-28)
Hospital Anxiety and Depression Scale (HADS)
Geriatric Depression Scale – Short Form

Appendix IV - Data Extraction Fields

The key fields below will be captured for each study in a tabular/spreadsheet format for analysis. This may be reformatted in one or more tables for reporting purposes and delivery of results.

Study setting
Year of study
Patient details (e.g. country, health status, age, gender, total population sample and eligibility criteria)
Numbers of patients
Study methods (e.g. Design, schedule of activities, method of allocation concealment used)
Duration of intervention
Interventions (e.g. methods, delivery type (individual/group), session number/duration)
Outcome measures recorded (Type, administration frequency, Patient reported outcomes (PRO))
Randomisation method
Key results and findings (e.g. Mean PRO values, respiratory symptoms, acute exacerbations)
Study conclusions

The following fields will also be captured if available and pertinent

Source of funding
Inclusion criteria
Exclusion criteria
Other symptoms
Blinding (masking) procedure(s)
Care provider/Outcome assessors
Previous singing training
Co-interventions
Numbers of patients not followed up
Reasons for withdrawals from study protocol
Details on adverse events of therapy
Patient comments or interview text
Other measurement results
Mean Difference from baseline for any measure
Intervention details (vocal exercises, song type, pitch and dynamic range, delivery format, control methods and prescribed singing practice time)

Appendix V - PRISMA checklist

Numbers in blue superscript parentheses refer to the PRISMA 2009 checklist headings 1-16 below – for illustration purposes only:

PRISMA 2009 Checklist

Section/topic	#	Checklist item	Reported on page #
TITLE			
Title	1	Identify the report as a systematic review, meta-analysis, or both.	
ABSTRACT			
Structured summary	2	Provide a structured summary including, as applicable: background; objectives; data sources; study eligibility criteria, participants, and interventions; study appraisal and synthesis methods; results; limitations; conclusions and implications of key findings; systematic review registration number.	
INTRODUCTION			
Rationale	3	Describe the rationale for the review in the context of what is already known.	
Objectives	4	Provide an explicit statement of questions being addressed with reference to participants, interventions, comparisons, outcomes, and study design (PICOS).	
METHODS			
Protocol and registration	5	Indicate if a review protocol exists, if and where it can be accessed (e.g., Web address), and, if available, provide registration information including registration number.	
Eligibility criteria	6	Specify study characteristics (e.g., PICOS, length of follow-up) and report characteristics (e.g., years considered, language, publication status) used as criteria for eligibility, giving rationale.	
Information sources	7	Describe all information sources (e.g., databases with dates of coverage, contact with study authors to identify additional studies) in the search and date last searched.	
Search	8	Present full electronic search strategy for at least one database, including any limits used, such that it could be repeated.	
Study selection	9	State the process for selecting studies (i.e., screening, eligibility, included in systematic review, and, if applicable, included in the meta-analysis).	
Data collection process	10	Describe method of data extraction from reports (e.g., piloted forms, independently, in duplicate) and any processes for obtaining and confirming data from investigators.	
Data items	11	List and define all variables for which data were sought (e.g., PICOS, funding sources) and any assumptions and simplifications made.	
Risk of bias in individual studies	12	Describe methods used for assessing risk of bias of individual studies (including specification of whether this was done at the study or outcome level), and how this information is to be used in any data synthesis.	
Summary measures	13	State the principal summary measures (e.g., risk ratio, difference in means).	
Synthesis of results	14	Describe the methods of handling data and combining results of studies, if done, including measures of consistency (e.g., I^2) for each meta-analysis.	
Risk of bias across studies	15	Specify any assessment of risk of bias that may affect the cumulative evidence (e.g., publication bias, selective reporting within studies).	
Additional analyses	16	Describe methods of additional analyses (e.g., sensitivity or subgroup analyses, meta-regression), if done, indicating which were pre-specified.	
RESULTS			
Study selection	17	Give numbers of studies screened, assessed for eligibility, and included in the review, with reasons for exclusions at each stage, ideally with a flow diagram.	
Study characteristics	18	For each study, present characteristics for which data were extracted (e.g., study size, PICOS, follow-up period) and provide the citations.	
Risk of bias within studies	19	Present data on risk of bias of each study and, if available, any outcome level assessment (see item 12).	
Results of individual studies	20	For all outcomes considered (benefits or harms), present, for each study: (a) simple summary data for each intervention group (b) effect estimates and confidence intervals, ideally with a forest plot.	
Synthesis of results	21	Present results of each meta-analysis done, including confidence intervals and measures of consistency.	
Risk of bias across studies	22	Present results of any assessment of risk of bias across studies (see Item 15).	
Additional analysis	23	Give results of additional analyses, if done (e.g., sensitivity or subgroup analyses, meta-regression [see Item 16]).	
DISCUSSION			
Summary of evidence	24	Summarize the main findings including the strength of evidence for each main outcome; consider their relevance to key groups (e.g., healthcare providers, users, and policy makers).	
Limitations	25	Discuss limitations at study and outcome level (e.g., risk of bias), and at review-level (e.g., incomplete retrieval of identified research, reporting bias).	
Conclusions	26	Provide a general interpretation of the results in the context of other evidence, and implications for future research.	
FUNDING			
Funding	27	Describe sources of funding for the systematic review and other support (e.g., supply of data); role of funders for the systematic review.	

From: Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group (2009). Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement. PLoS Med 6(6): e1000097. doi:10.1371/journal.pmed1000097

For more information, visit: www.prisma-statement.org.

Appendix VI - Cochrane Collaboration Risk of Bias Table

Domain	Support for judgement	Review authors' judgement
Selection bias.		
Random sequence generation.	Describe the method used to generate the allocation sequence in sufficient detail to allow an assessment of whether it should produce comparable groups.	Selection bias (biased allocation to interventions) due to inadequate generation of a randomised sequence.
Allocation concealment.	Describe the method used to conceal the allocation sequence in sufficient detail to determine whether intervention allocations could have been foreseen in advance of, or during, enrolment.	Selection bias (biased allocation to interventions) due to inadequate concealment of allocations prior to assignment.
Performance bias.		
Blinding of participants and personnel *Assessments should be made for each main outcome (or class of outcomes).*	Describe all measures used, if any, to blind study participants and personnel from knowledge of which intervention a participant received. Provide any information relating to whether the intended blinding was effective.	Performance bias due to knowledge of the allocated interventions by participants and personnel during the study.
Detection bias.		
Blinding of outcome assessment *Assessments should be made for each main outcome (or class of outcomes).*	Describe all measures used, if any, to blind outcome assessors from knowledge of which intervention a participant received. Provide any information relating to whether the intended blinding was effective.	Detection bias due to knowledge of the allocated interventions by outcome assessors.
Attrition bias.		
Incomplete outcome data *Assessments should be made for each main outcome (or class of outcomes).*	Describe the completeness of outcome data for each main outcome, including attrition and exclusions from the analysis. State whether attrition and exclusions were reported, the numbers in each intervention group (compared with total randomized participants), reasons for attrition/exclusions where reported, and any re-inclusions in analyses performed by the review authors.	Attrition bias due to amount, nature or handling of incomplete outcome data.
Reporting bias.		
Selective reporting.	State how the possibility of selective outcome reporting was examined by the review authors, and what was found.	Reporting bias due to selective outcome reporting.
Other bias.		
Other sources of bias.	State any important concerns about bias not addressed in the other domains in the tool. If particular questions/entries were pre-specified in the review's protocol, responses should be provided for each question/entry.	Bias due to problems not covered elsewhere in the table.

(Higgins and Green, 2011)

Appendix B – Systematic Review Risk of Bias **(illustration purposes only)**

Systematic Review – Risk of Bias Table for All Included Studies

	QUALITY ASSESSMENT TOOL FOR QUANTITATIVE STUDIES (as formatted)	Bonilha 2008	BLF 2017	Canga 2015	Clift 2013/	Clift 2017	Engen 2005	Goodridge 2013	Grasch 2013	Herer 2013	Liu 2016	Lichtensc hopf 2014	Jamaly 2017	Lord 2010	Lord 2012	McNaughton	Miyara	Pacheco 2014	Thomas 2015	Trivedi 2017
A	**A) SELECTION BIAS**																			
Q1	**(Q1) Are the individuals selected to participate in the study likely to be representative of the target population?**	GOLD		GOLD			Physicians diagnosis	Medical diagnosis of COPD	Physicians diagnosis	GOLD COPD + CRD		GOLD		GOLD	GOLD					Global initiative for COPD criteria
	1 Very likely	1		1	1		1	1	1	1		1		1	1	1	1	1		1
	2 Somewhat likely		2								2		2						2	
	3 Not likely																			
	4 Can't tell																			
Q2	**(Q2) What percentage of selected individuals agreed to participate?**	55.13%		68/98 =	121/126 = 96%		12 partic	19/22 = 9	31					36/92	24/32	23/28 = 82%	6/8 = 75%	16/16		56/90 = 62%
	1 80 - 100% agreement				1	1		1	1							1			1	
	2 60 – 79% agreement			2													2			2
	3 less than 60% agreement	3												3	3					
	4 Not applicable																			
	5 Can't tell		5				5			5	5		5				5			
A	RATE THIS SECTION																			
	1 STRONG				1	1		1	1							1				
	2 MODERATE	2		2			2					2		2	2		2	2	2	2
	3 WEAK		3							3	3		3							
	See dictionary																			
B	**B) STUDY DESIGN**																			
Q1	**Indicate the study design**	RCT	Feasibility/ Pilot	RCT	Feasibility/ Pilot	Feasibility/ Pilot	Cohort study	Two cohorts 14 Tx v. 5 SoC	quasi-experimental pre–po	open, non-randomized	RCT	Feasibility/ Pilot	RCT	RCT	RCT	Mixed method	RCT	Feasibility/ Pilot		
	1 Randomized controlled trial	1		1							1			1	1		1			1
	2 Controlled clinical trial				2							2						2		
	3 Cohort analytic (two group pre + post)					3		3	3	3						3				
	4 Case-control																		4	
	5 Cohort (one group pre + post (before and after))		5				5													
	6 Interrupted time series																			
	7 Other specify																			
	8 Can't tell																			
	Was the study described as randomized? If NO, go to Component C.																			
	No Yes	Yes		Yes	No	No	No	No	No	No	Yes	No	Yes	Yes	Yes	No	Yes	No	No	Yes
	If Yes, was the method of randomization described? (See dictionary)																			
	No Yes	No		Yes							Yes		No	Yes	No		No			No
	If Yes, was the method appropriate? (See dictionary)																			
	No Yes	N/A		Yes							Yes		NA	Yes	NA		NA			NA
B	RATE THIS SECTION																			
	1 STRONG	1		1							1			1	1					
	2 MODERATE				2	2	2	2	2	2		2	2			2	2	2		2
	3 WEAK																		3	
C	**C) CONFOUNDERS**																			
Q1	**(Q1) Were there important differences between groups prior to the intervention?**											COPD + CRD							CRD not just COPD	
	1 Yes											1				1				1
	2 No	2	2	2	2	2	2	2	2					2	2		2			
	3 Can't tell									3	3		3				3		3	
	Information captured and important difference observed regarding:																			
	i Tx/ Medication Standard of Care, Pt diagnosed by Physician	NK	NK	NK	NK	No	No	No	NK	NK	NK	NK	NK	NK	NK	NK	NK	NK	NK	NK
	ii Previous singing/external singing or other confounding activity (e.g. musical instrument lessons)	NK	NK	Yes	NK	NK	NK	NK	NK	NK	NK	NK	NK	Yes	NK	NK	NK	NK	NK	NK
	iii Demographics - Age, Location	No	No	No	No	No	No	No	NK	No	No	NK	No	No	NK	Yes	NK	No	NK	No
	iv Respiratory health status/underlying disease	No	No	No	No	No	No	No	NK	Yes	No	Yes	No	No	No	No	NK	No	NK	No
	v Smoking history	No	No	No	No	No	No	NK	NK	NK	NK	NK	NK	NK	NK	NK	NK	NK	NK	NK
	vi Pulmonary Rehabiliation	No	NK	No	NK	No	NK	NK	Nk	NK	NK	NK	Yes	NK	NK	No	NK	No	NK	Yes
Q2	**(Q2) If yes, indicate the percentage of relevant confounders that were controlled (either in the design (e.g. stratification, matching) or analysis)?**	RCT		RCT					experimental pre/pos		RCT		RCT	RCT	RCT		RCT			RCT
	1 80 – 100% (most)	1		1							1		1	1	1		1			1
	2 60 – 79% (some)																			
	3 Less than 60% (few or none)							3												
	4 Can't Tell		4		4	4	4		4	4		4				4		4	4	
C	RATE THIS SECTION																			
	1 STRONG	1		1										1	1					1
	2 MODERATE				2	2					2		2							
	3 WEAK		3				3	3	3	3		3				3	3	3	3	
D	**D) BLINDING**																			
Q1	**(Q1) Was (were) the outcome assessor(s) aware of the intervention or exposure status of participants?**																			
	1 Yes	1	1	1	1	1	1	1				1	1	1	1	1		1	1	1
	2 No																			
	3 Can't tell								3	3	3						3			
Q2	**(Q2) Were the study participants aware of the research question?**																			
	1 Yes	1	1		1	1					1	1	1	1	1	1		1	1	1
	2 No																			
	3 Can't tell			3			3	3	3	3							3			
D	RATE THIS SECTION																			
	1 STRONG																			
	2 MODERATE																			
	3 WEAK	3	3	3	3	3	3	3	3	3	3	3	3	3	3	3	3	3	3	3
	See dictionary																			
E	**E) DATA COLLECTION METHODS**																			
Q1	**(Q1) Were data collection tools shown to be valid?**	Standard validate	Standard validate	Standard validate	Standard validate	Standard validate	Standard validate	Standard validated Tools	Standard validate	Standard validate	Standard validate	Questions only - no questionna	Standard validated Tools used	Standard valida	Standard validate		Standard validate	Standard validated Tools used	Standard validated Tools used	Standard validated Tools used
	1 Yes	1		1	1	1	1	1	1	1	1		1	1	1		1	1	1	1
	2 No																			
	3 Can't tell											3				3				
Q2	**(Q2) Were data collection tools shown to be reliable?**																			
	1 Yes	1	1	1	1	1	1	1	1	1	1		1	1	1		1	1	1	1
	2 No																			
	3 Can't tell											3				3				
E	RATE THIS SECTION																			
	1 STRONG	1	1	1	1	1	1	1	1	1	1		1	1	1		1	1	1	1
	2 MODERATE															2				
	3 WEAK											3				3				
F	**F) WITHDRAWALS AND DROP-OUTS**																			
Q1	**(Q1) Were withdrawals and drop-outs reported in terms of numbers and/or reasons per group?**																			
	1 Yes	1		1	1	1	1	1	1	1				1	1	1		1		
	2 No		2									2					2		2	
	3 Can't tell										3		3							3
	4 Not Applicable (i.e. one time surveys or interviews)																			
Q2	**(Q2) Indicate the percentage of participants completing the study. (If the percentage differs by groups, record the lowest).**	43/78 = 55%		68/98 = 69%	66%	44/60 = 73%	7/12 = 58%	19/19	25/31 = 80%	45/48	56	114/119		13/16	23/183: Sing 13/18 =72%	21/28 = 75%	singing + control n=10	6/8		54/56
	1 80 -100%							1	1	1	1			1	1		1	1	1	1
	2 60 - 79%			2	2	2										2	2			
	3 less than 60%	3					3													
	4 Can't tell		4									4								
	5 Not Applicable (i.e. Retrospective case-control)																			
F	RATE THIS SECTION																			
	1 STRONG							1	1	1	1		1				1			
	2 MODERATE	2		2	2	2								2	2	2		2	2	3
	3 WEAK		3				3					3								
	4 Not Applicable																			
G	**G) INTERVENTION INTEGRITY**																			
Q1	**(Q1) What percentage of participants received the allocated intervention or exposure of interest?**	8/23 = 34%		68/98 = 69%			~7/7	14/14	25/31 = 80%	45/48		118	22 =	15/16	Sing 13/18	21/28 = 75%	10/10 = 100%	~06/08		54/56
	1 80 -100%						1	1	1	1	1	1	1	1			1	1	1	1
	2 60 - 79%			2		2										2	1			
	3 less than 60%	3																		
	4 Can't tell		4																	
Q2	**(Q2) Was the consistency of the intervention measured?**																			
	1 Yes					1	1													
	2 No	2	2	2								2		2	2	2				
	3 Can't tell						3	3	3	3	3		3				3	3	3	3
Q3	**(Q3) Is it likely that subjects received an unintended intervention (contamination or co-intervention) that may influence the results?**																			
	1 Yes			1																
	2 No																			
	3 Can't tell	3	3		3	3	3	3	3	3	3	3	3	3	3	3	3	3	3	3
H	**H) ANALYSES**																			
Q1	**(Q1) Indicate the unit of allocation (circle one)**																			
	community organization/institution practice/office individual	institution practice	community organization	institution practice	community organization	community organization	institution practice	institution practice	institution practice	institution practice	institution practice	institution practice	institution practice	institution practi ce	institution practice	institution practice	institution practice	institution practice	institution practice	institution practice
Q2	**(Q2) Indicate the unit of analysis (circle one)**																			
	community organization/institution practice/office individual	institution practice	community organization	institution practice	community organization	community organization	institution practice	institution practice	institution practice	institution practice	institution practice	institution practice	institution practice	institution practi ce	institution practice	institution practice	institution practice	institution practice	institution practice	institution practice
Q3	**(Q3) Are the statistical methods appropriate for the study design?**																			
	1 Yes	1	1	1	1	1	1	1	1	1	1		1	1	1	1		1	1	1
	2 No																			
	3 Can't tell											3					3			
Q4	**(Q4) Is the analysis performed by intervention allocation status (i.e. intention to treat) rather than the actual intervention received?**																			
	1 Yes																			
	2 No	2	2	2	2	2	2	2	2				2	2	2	2	2	2	2	2
	3 Can't tell									3	3	3								
	GLOBAL RATING																			
	COMPONENT RATINGS																			
	Please transcribe the information from the grey boxes on pages 1-4 onto this page. See dictionary on how to rate this section.																			
A	SELECTION BIAS																			
	1 STRONG	0	0	0	1	1	0	1	1	0	0	0	0	0	0	1	0	0	0	0
	2 MODERATE	2	0	2	0	0	2	0	0	0	0	2	0	2	2	0	2	2	2	2
	3 WEAK	0	3	0	0	0	0	0	0	3	3	0	3	0	0	0	0	0	0	0
B	STUDY DESIGN																			
	1 STRONG	1	0	1	0	0	0	0	0	0	1	0	0	1	1	0	0	0	0	0
	2 MODERATE	0	0	0	2	2	2	2	2	2	0	2	2	0	0	2	2	2	2	2
	3 WEAK	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0
C	CONFOUNDERS																			
	1 STRONG	1	0	1	0	0	0	0	0	0	0	0	0	1	1	0	0	0	0	1
	2 MODERATE	0	0	0	2	2	0	0	0	0	2	0	2	0	0	0	0	0	0	0
	3 WEAK	0	3	0	0	0	3	3	3	3	0	3	0	0	0	0	3	3	3	0
D	BLINDING																			
	1 STRONG	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0
	2 MODERATE	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0
	3 WEAK	3	3	3	3	3	3	3	3	3	3	3	3	3	3	3	3	3	3	3
E	DATA COLLECTION METHOD																			
	1 STRONG	1	1	1	1	1	1	1	1	1	1	0	1	1	1	0	0	1	1	1
	2 MODERATE	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	2	0	0	0
	3 WEAK	0	0	0	0	0	0	0	0	0	0	3	0	0	0	3	0	0	0	0
F	WITHDRAWALS AND DROPOUTS																			
	1 STRONG	0	0	0	0	0	0	1	1	1	1	0	1	0	0	0	1	0	0	0
	2 MODERATE	2	0	2	2	2	0	0	0	0	0	0	0	2	2	2	0	2	2	3
	3 WEAK	0	3	0	0	0	3	0	0	0	0	3	0	0	0	0	0	0	0	0
	4 Not Applicable	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0
	GLOBAL RATING FOR THIS PAPER (circle one):																			
	1 1 STRONG (no WEAK ratings)																			
	2 2 MODERATE (one WEAK rating)	2		2	2	2								2	2					2
	3 3 WEAK (two or more WEAK ratings)		3				3	3	3	3	3	3	3			3	3	3	3	

Appendix C – Research Study Protocol

Singing for Chronic Obstructive Pulmonary Disease (COPD)

An Evaluation of a Community Programme including Feasibility of a Self-help Booklet and DVD Guide: "Sidney De Haan Research Centre - 'Singing for Better Breathing' Resource©"

Version – Final V1
Version date – 07-Apr-17
Author: Charlotte Epsley

Protocol Summary

Abstract

Respiratory disorders manifest themselves with chest symptoms including shortness of breath and a decline of lung function and a therapy is needed to provide treatment and support for these patients. There is a growing body of evidence to suggest that therapies that include breath control exercises, such as singing, may have health benefits for respiratory function and psychological well-being.

In addition to group singing, supplementary activities which encourage regular and ongoing singing practice, participation in group singing and observing the correct techniques have potential to further support the objectives of singing for respiratory health. This protocol outlines the planned evaluation of a community-based singing group for adults with COPD and usability of carefully prepared home practice guides provided to participants to use between singing sessions.

Terms: Singing, COPD, Chronic Respiratory Disease, self-study resources

Table of Contents

Introduction

Respiratory diseases are among the leading causes of death worldwide and are likely to remain a major burden for decades to come. It is estimated that sixty-five million people have moderate to severe COPD worldwide and more than three million deaths occurred (5% of all deaths globally) in 2015 **(WHO, 2016)**. The prevention and treatment of lung diseases will need to be improved, if their impact on longevity and the quality of life (QoL) of individuals and their economic burden on society are to be reduced in Europe and worldwide **(WHO, 2012)**.

Respiratory Disorders manifest themselves with chest symptoms including shortness of breath and a decline of lung function. Chronic obstructive pulmonary disease (COPD) is one such term for a group of these airway conditions which are characterised by progressive, irreversible limitation of airflow, and a major goal of its treatment is to ensure that the patient's health is optimised **(Jones et al., 2009)**.

Dyspnoea, or shortness of breath, is perhaps the most common accompaniment of lung disease **(Burki and Lee, 2010)**. Patients with chronic respiratory disease, including COPD, are often limited in their activities by respiratory discomfort of this kind. Airflow limitation in patients with COPD leads to dynamic hyperinflation, particularly during exercise and several important consequences of dynamic hyperinflation serve to worsen dyspnoea **(Meek et al., 1999)**. A reduction in functional status and quality of life of these patients, along with disability are frequent consequences. These diseases may leave patients presenting with this significant breathlessness symptom despite maximal therapy and are estimated to debilitate millions of people worldwide **(Herigstad et al., 2010)**. Group singing is an activity that may support lung function and enhance quality of life in people with certain respiratory disorders and it has the potential to play an important role as an adjunct therapy to other established and proven offerings.

A small, randomised study of COPD patients **(Bonilha et al., 2009)** concluded that singing classes are a well-tolerated activity for selected patients and that the regular practice of singing may improve QoL and preserve maximal expiratory pressure of these patients. A feasibility study of the health benefits of a community singing programme for older people with COPD **(Clift et al., 2013)** further supports this, indicating that measures of lung function and health-related quality of life significantly improved over a ten months period of the singing programme. In addition, recent results published by the British Lung Foundation **(BLF, 2017)** indicate that taught exercises, specifically focused on improving lung conditions, compared to singing for wellbeing in general, demonstrate improvement in the impact living with a lung condition has on a patients' daily life.

This planned research is in line with these finding and with existing UK Department of Health (DH) Chronic Obstructive Pulmonary Disease (COPD) outcomes strategy discussions,

particularly objectives identified in the **UK DH (2012)** companion document, which outlines directions for enhancing quality of life for these patients and supporting outcome strategies to improve respiratory health and well-being of all communities.

It is still recommended that further research is necessary **(Bonilha et al., 2009, Lord et al., 2012**, **Clift et al., 2013)** to gain more robust results and further understand the outcome of group singing for this indication and population. It is also considered that additional singing practice outside group sessions may help improve the overall frequency, compliance and retention of patients to prolong their engagement, and maximise exposure and therapeutic effect.

To that end 'Singing for Better Breathing' (SfBB) resources, including a supportive guidance booklet and DVD have been developed by the research team at the Sidney De Haan Research Centre to support simple practice activities between sessions. It is the objective of this study to explore the feasibility of using such tools in this population and gain feedback and experience of this media in a real life setting with COPD patients.

There has been some limited research in self-help or home learning for comparable disciplines e.g. Pulmonary Retention for COPD patients **(Mendes de Oliveira et al., 2010),** exercise for older adults **(McAuley et al., 2013)** and for DVD exercise programmes more broadly **(Rasmussen et al., 2013, Kingston et al., 2014)** and some singing for COPD studies have even included a home singing practice element **(Lord et al., 2010, Lord et al., 2012)**. However, this is not explored in any detail and there is very little evidence of research in home delivery methods or programmes for singing guidance targeted specifically at COPD patients.

Mechanism of Action

The key mechanism of action of singing in these patients appears to be the active engagement of the muscles of the entire respiratory system and an increase in respiratory muscle strength, leading to increased lung volume and effective cough **(Kang et al., 2006, Wiens et al., 1999)**. Poor posture (hyperkyphosis) is often common in COPD patients and can restrict the expansion of the rib cage and movement of the diaphragm. Since singing requires the development of skills in controlling posture this may be transferable to activities in daily life in people with COPD **(Lord et al., 2010)**.

The other mechanisms that may have significance include a reduction of fear, anxiety and pain perception **(Kenny and Faunce, 2004)** possibly by increased breath control improving the autonomic nervous system through enhanced activation of the parasympathetic system, leading to higher vagal tone **(Pramanik et al., 2009, Kok and Fredrickson, 2010, Vickoff et al., 2016).** This in turn and in combination with the improvements in a patients overall improved consciousness of their own breathing due to the singing training, may reduce the occurrence or

severity of dyspnoea episodes; Improvement of mood **(Unwin et al., 2002)** positively impacted by socialising and meeting people, thus combating isolation; Finally, although there is little evidence in this population specifically, Immunoglobulin A (S-IgA) levels have been revealed to be particularly responsive to music **(Fancourt et al., 2014)** and greater improvements in mood, as a result of singing have been associated with lower pro-inflammatory response **(Fancourt et al., 2016)** leading to a potential hypothesis that singing could lead to a positive impact on depression and inflammation which would be a particularly important effect for COPD patients. These mechanisms are all thought likely to be interrelated too, thus enhancing the overall benefit by form of a virtuous circle potentially combating the 'spiral of decline' **(Jones, 2009)** associated with the condition.

Background and Rationale

Condition

Respiratory illness encompasses developmental, pathological conditions affecting the organs and tissues that make gas exchange possible in higher organisms, and includes conditions of the upper respiratory tract, trachea, bronchi, bronchioles, alveoli, pleura and pleural cavity, and the nerves and muscles of breathing **(WHO, 1978)**.

COPD is characterised by poorly reversible airflow obstruction and an abnormal inflammatory response in the lungs, which represents the innate and adaptive immune responses to long term exposure to noxious particles and gases **(MacNee, 2006)**.

Intervention

The intervention combines a thirteen-week programme of regular weekly group singing sessions with guided home singing practice between sessions. The findings of a study looking at utilising a DVD to improve compliance with home exercise programs **(Kingston et al., 2014)** demonstrated the multidimensional nature of compliance and showed improved understanding of exercises when utilising home resources such as DVDs. This suggests self-help guides such could be utilised as part of a programme that facilitates the patient-therapist relationship. It is considered important to trial additional resources both to enhance the experience of group singing, vary the medium of delivery of this intervention, provide a more versatile offering and to encourage retention and continued adherence to singing exercises longer term.
A previous study **(McAuley et al., 2013)** provided cautiously optimistic evidence that the use of DVD-delivered programs designed for a target population can be both effective and utilized on a broad scale. This study demonstrated the feasibility, acceptability, and efficacy of delivering regularly scheduled activities focusing on older adults via the medium of DVD. This study highlighted and recognised that this type of novel intervention is also capable of reaching participants from outlying geographical areas who typically would not be able to attend centre-

based activity trials conducted at university or medical centres. The SfBB resources have been developed such that they may also have other potential uses in the future to provide support to those COPD patients who might benefit from the activity of singing but are unable to attend group sessions outside their home environment due to extent of their disease or other limitations.

Although centre-based interventions for exercise programs are generally shown to have success in improving functional performance in older adults, such approaches can be costly, have limited reach, can be challenging to implement and sustain, and are often not generalizable **(Glasgow et al., 1999)**. Low-cost, broad-reaching, and innovative approaches to delivering physical activity programs are therefore needed.

Study Objectives and Endpoints

Objectives

The primary objectives of the research is to review and evaluate the impact of weekly group singing on COPD patients and to assess specifically created resources “Sidney De Haan (SDHR) ‘Singing for Better Breathing’ Resource©” (SfBB) used in conjunction within the target population, and obtain patient feedback on the usefulness, usability and format of this tool to support or enhance the effectiveness of group singing for patients with COPD

The secondary objectives will be to assess recruitment uptake, retention and acceptability and adherence to the SfBB resources with the objective of assessing feasibility for this type of intervention in this population. Compliance data of group singing and use of resources will be collected through group attendance records and participant feedback questionnaires.

Primary Endpoint

- Participant feedback of SDHR SfBB resources via specially designed SFBB Questionnaire
- Impact of weekly group singing on participants using WEMWBS, CAT and ACQ-5 data

Secondary Endpoints

- SDH SfBB suggestions for improvement per written feedback from patient evaluation form
- Suggestions for improvements as per feedback from patient questionnaire
- Compliance with SDH SfBB resource guidance schedule
- Overall compliance and drop-out rate

- Patient reported outcomes from COPD patients that have agreed to use the tools in real time, including evaluation solicited from patients written comments regarding the usability of the resources.

Study Design

Participant Selection

This study can fulfil its objectives only if appropriate participants are enrolled. The following eligibility criteria are designed to select only self-referred participants for whom the protocol intervention is considered appropriate by their health care provider (HCP) or pulmonary rehabilitation team.

Inclusion Criteria

Participants must meet all of the following inclusion criteria to be eligible for enrolment in the study:

Over 18 years of age.

Shows evidence of a personally signed and dated informed consent document, indicating that the participant has been informed of all pertinent aspects of the study and provided with an information sheet.

Previously diagnosed with COPD or lung condition by their Physician or other Health Care Provider (HCP)

Willing to commit to participating in the initial project over the course of 13 weeks (health permitting)

Enrolled into a singing for COPD group run by the Sidney De Haan Research Centre

Physically mobile and able to travel to sessions independently or with support

NB. Participant eligibility should be reviewed and documented by an appropriate member of the research team before participants are included in the study.

Exclusion Criteria

Participants with any of the following characteristics/conditions will not be included in the study:

Research team site staff members directly involved in the conduct of the study and their family members, site staff members otherwise supervised by the research team, or participants who are employees of Sidney De Haan Research Centre or Canterbury Christ Church University, including their family members, directly involved in the conduct of the study

Participants who are physically unable to attend group singing evaluation sessions

Participants with severe dementia or other cognitive or communication problems, which in the opinion of the research team would render consent problematic

Participants with ongoing or planned participation in pulmonary rehabilitation, scheduled to take place within project timescale.

Study Intervention

Singing groups will run weekly over a period of thirteen weeks. Skilled singing facilitators and a planned programme of singing and singing-related exercises will ensure standardisation of delivery throughout the trial. Participants will be involved in baseline data assessment before the start of the singing groups and after the first seven weeks of intervention. Follow-up data collection will take place thirteen weeks after the intervention period of the trial.

The structured singing during group sessions will closely follow the guidance provided in the resource booklet (Appendix I) and copies of the booklet instructions and an accompanying DVD will be provided to all participants. Guidance will be given for participants to follow the resource instructions at home outside the group sessions and ‘self-administer’ the intervention according to the guidance.

Allocation to Intervention

A single, non-randomised un-blinded design will be employed. Participants will be recruited from willing volunteers in newly established self-referred community groups with approximately thirty to forty participants recruited across the groups. Participants will be assigned a number sequentially by research team members.

The research team knowledge of treatment should not influence the decision to enrol a particular participant or affect the order in which participants are enrolled.

Study Procedures

Baseline Visit V_1 - Week 1 (±7 days)

- Information Sheet and Informed Consent Document Sign-off
- Demography Questionnaire
- Warwick-Edinburgh Mental Well-Being Scale (WEMWBS)
- COPD Assessment Test (CAT)
- Asthma Control Questionnaire (ACQ-5)
- Initial General Feedback Questionnaire
- Handout SDHC SfBB Booklet/DVD resources

Study Period Visit V_2 - Week 7 (±7 days)

- Warwick-Edinburgh Mental Well-Being Scale (WEMWBS)
- The COPD Assessment Test (CAT)
- Asthma Control Questionnaire (ACQ-5)

Follow Up Visit V_3 - Week 13 (±7 days)

- Warwick-Edinburgh Mental Well-Being Scale (WEMWBS)
- COPD Assessment Test (CAT)
- Asthma Control Questionnaire (ACQ-5)
- SfBB self-help resource feedback questionnaire (SfBBQ)
- Final General Feedback Questionnaire

Assessments

- Demography Questionnaire
- Warwick-Edinburgh Mental Well-Being Scale
- COPD Assessment Test (CAT)
- Asthma Control Questionnaire
- General Feedback Questionnaire (pre-intervention)
- General Feedback Questionnaire (post-intervention)

Questionnaires

Warwick-Edinburgh Mental Well-Being Scale

The Warwick Edinburgh Mental Wellbeing Scale (WEMWBS) (Appendix I) is a validated questionnaire assessing positive mental wellbeing and aims to measure mental well-being itself and not the determinants of mental well-being. It comprises 14 item scale that relate to an individual's state of mental well-being (thoughts and feelings) in the previous two weeks. It is a scale of mental well-being covering subjective well-being and psychological functioning, in which all items are worded positively and address aspects of positive mental health. The scale is scored by summing responses to each item answered on a 1 to 5 Likert scale. The minimum scale score is 14 and the maximum is 70. WEMWBS has been validated for use in the UK with those aged 16 and above **(Stewart-Brown et al., 2008)**.

The COPD Assessment Test (CAT)

The COPD Assessment Test (CAT) (Appendix I) provides clinicians and patients with a simple and reliable measure of overall COPD-related health status for the assessment and long-term follow-up of individual patients. It consists of eight items, each formatted as a semantic six-point differential scale, making the tool easy to administer and easy for patients to complete. The items cover a wide range of disease severity, with the intention that the greatest discriminant power would be in the mild to moderate range **(Jones et al., 2009)**.

Asthma Control Questionnaire (ACQ-5)

The Asthma Control Questionnaire ACQ-5 (Appendix I) is an asthma control measurement tool consisting of five questions on symptom control; each of the questions is scored on a scale of 0–6 where 0 represents excellent asthma control and 6 represents extremely poor control **(O'Byrne et al., 2010)**. The ACQ can include responses to seven questions, five relating to symptoms, one relating to rescue symptom use and one to capture FEV1. The shortened, five-question 'symptom only' questionnaire is considered just as valid and is adequate for the purposes of this study.

Participant Withdrawal

'Withdrawal of Consent': Participants who request to discontinue will remain in the study and must continue to be followed for protocol specified follow-up procedures. The only exception to this is when a participant specifically withdraws consent for any further contact with him/her or persons previously authorized by participant to provide this information. Participants should

notify the research team of the decision to withdraw consent from future follow-up whenever possible. All attempts should be documented.

'Lost to Follow-Up': All reasonable efforts must be made to locate participants to determine and report their current status. Lost to follow-up is defined by the inability to reach the participant after a minimum of three documented efforts (phone calls, faxes, letter or emails). All attempts should be documented.

The study team will consult publicly available sources, such as public health registries and databases, in order to obtain updated contact information. If after all attempts, the participant remains lost to follow-up, then the last known contact date as determined by the study team should be reported and documented.

Participants may withdraw from the study at any time at their own request, or they may be withdrawn at any time at the discretion of the study team.

If a participant does not return for a scheduled visit, every effort should be made to contact the participant. All attempts to contact the participant and information received during contact attempts will be documented. In any circumstance, every effort should be made to document participant outcome, if possible. The study team should inquire about the reason for withdrawal and request the participant to return for a final visit, if applicable.

If the participant withdraws from the study, and also withdraws consent for disclosure of future information, no further evaluations should be performed, and no additional data should be collected. The researcher may retain and continue to use any data collected before such withdrawal of consent.

Data Analysis and Statistical Methods

The paired t-test will be used to compare means of two related observations i.e. a comparison of given values both before and after an intervention and recorded in pairs per participant. The paired t-test is selected due to the small sample sizes to compare mean differences. The difference between the paired values is assumed to be normally distributed and the null hypothesis, that the expectation is zero, is tested by paired t test.

The test is used to compare the means for all three health and wellbeing evaluations over each possible interval i.e. between V_0 and V_1, V_1 and V_2, V_2 and V_3, V_1 and V_3, V_0 and V_2 and V_0 and V_3 for both the total score and individual question scores. Those with values of significance i.e. $p < 0.05$ are reported.

The calculation of the statistical significance shows the improbability of findings drawn from assumptions about the null hypothesis. The substantive significance i.e. what the findings indicate about population effects themselves will be assessed using a comparison of the mean difference expressed as meaningful for each of the individual health and wellbeing scores as noted from previous validations of each one.

Sample Size Determination

Sample size is determined based on sample size calculated for two prior outpatient pulmonary rehabilitation studies **(Bakarat et al., 2008, Mendes de Oliveira et al., 2010)** where total patients recruited into the PR home resources cohort was 19-23. The aim for this study will be to enrol at least 30 participants over two groups with conservative anticipated drop-out rate of 30-50% allowing for 15-21 participants or over, completing the full evaluation.

Data Sources

Data sources will be questionnaires designed to solicit specific feedback directly from the participants regarding their health and wellbeing and the tools under study. This will include a novel questionnaire designed for participants to answer simple questions about use of the SfBB tools. Attendance data for the singing group sessions will also be collected to capture participant compliance to regular weekly singing.

Data Handling and Record Keeping

Case Report Forms - Electronic Data Record

The term Case Report Form (CRF) as used in this protocol should be understood to refer to either a paper form or an electronic data record or both, depending on the data collection method(s) used. A CRF is required and should be completed for each included participant. The completed original CRFs are the sole property of the Sidney De Haan Research Centre and should not be made available in any form to third parties, except for authorized representatives of the Sidney De Haan Research Centre or appropriate regulatory authorities, without written permission from the research team.

The study team has ultimate responsibility for the collection and reporting of all data entered on the CRFs and any other data collection forms and ensuring that they are accurate, authentic / original, attributable, complete, consistent, legible, timely (contemporaneous), enduring and available when required. The CRFs must be signed by a member of the research team to attest that the data contained on the CRFs is true. Any corrections to entries made in the CRFs or

source documents must be dated, initialled and explained (if necessary) and should not obscure the original entry. Data Handling Conventions may be utilised, documented and applied to missing or inconsistently recorded data for the purposes of final statistical analysis.

Record Retention

To enable evaluations and/or audits from regulatory authorities, the research team agrees to keep records, including the identity of all participating participants (sufficient information to link records, e.g., CRFs), all original signed informed consent documents, copies of all CRFs, safety reporting forms, source documents, and detailed records of treatment disposition, and adequate documentation of relevant correspondence (e.g., letters, meeting minutes, telephone calls reports). The records should be retained by the research team according to International Conference on Harmonisation (ICH) or according to local regulations, whichever is longer.

All information gathered will be stored securely in the offices of the Sidney De Haan Research Centre for Arts and Health, Canterbury Christ Church University. Members of the research team are the only people who will have access to the primary research material. All data will be anonymous prior to analysis.

If the research team becomes unable for any reason to continue to retain study records for the required period (e.g., retirement, relocation) the study records must be transferred to a designee, such as another institution, or to an independent third party. Records must be kept for a minimum of 15 years after completion or discontinuation of the study or for longer if required by applicable local regulations.

Ethics

Institutional Review Board (IRB)/Ethics Committee (EC)

It is the responsibility of the research team to have prospective approval of the study protocol, protocol amendments, informed consent documents, and other relevant documents, if applicable, from the local IRB/EC. All correspondence with the IRB/EC should be retained by the study team.

The only circumstance in which an amendment may be initiated prior to IRB/EC approval is where the change is necessary to eliminate apparent immediate hazards to the participants. In that event, the research team must notify the IRB/EC in writing immediately after the implementation.

Ethics Conduct of the Study

The study will be conducted in accordance with legal and regulatory requirements, as well as the general principles set forth in the Guidelines for GCP (ICH 1996), and the Declaration of Helsinki (World Medical Association 1996 & 2008). In addition, the study will be conducted in accordance with the protocol, the ICH guideline on GCP, and applicable local regulatory requirements and laws.

Participant Information and Consent

All potential participants will be provided with an information sheet explaining the study and asked to sign a consent form. Personal details other than those necessary for demographic information will not be requested. All data will be treated as confidential and no comments from any participant in the study will be attributed to them personally in any publication.

When study data is compiled for transfer outside the investigating team, participant names, addresses, and other identifiable data will be replaced by a numerical code in order to de-identify study participants. The study team will maintain a confidential list of participants who participated in the study linking their numerical code to the participant's actual identity. In case of data transfer, the research team will maintain high standards of confidentiality and protection of participant personal data consistent with applicable privacy laws.

The informed consent documents must be in compliance with ICH GCP, local regulatory requirements, and legal requirements, including applicable privacy laws. The informed consent documents used during the informed consent process must be reviewed by the research team, approved by the IRB/EC before use and available for inspection. The research team must ensure that each study participant is fully informed about the nature and objectives of the study and possible risks associated with participation.

Ethical approval will be sought from the Faculty Research Ethics Committee of Canterbury Christ Church University. The study will be conducted in accordance with GCP/MRC GRP guidelines and the Declaration of Helsinki. It is expected that participants will generally be living independently and not affected by serious physical or mental frailty. However, participants may be considered vulnerable in view of their age, therefore care will be taken to present information about the project with due attention to their needs and capabilities. It is not believed that the research project outlined carries any risks of harm to individuals who will be invited to participate.

All information gathered will be stored securely in the offices of the Sidney De Haan Research Centre for Arts and Health, Canterbury Christ Church University. Members of the research team

are the only people who will have access to the primary research material. All data will be anonymous prior to analysis.

The research team, or a person designated by the research team, will obtain written informed consent from each participant, before any study-specific activity is performed, unless a waiver of informed consent has been granted by an IRB/EC. The research team will retain the original of each participant's signed consent document.

Participant Recruitment

Participants will be recruited from a pool of participants attending newly formed or established singing groups. These participants will have been self-referred on the basis of information provided by their Health Care Provider (HCP) or pulmonary rehabilitation team. Participants will have been invited to attend ‘taster’ sessions in advance of visit 1 to encourage interest and recruitment.

Definition of End of Study

End of Study (EOS) is defined as last participant’s last visit.

Premature termination of this study may occur because of a regulatory authority decision, change in opinion of the IRB/IEC or at the discretion of the Sidney De Haan Research Centre, Canterbury Christ Church University.

If a study is prematurely terminated or discontinued the research team will be notified. All study materials will be collected and CRFs completed to the greatest extent possible.

Publication of Study Results

Publication of study results will be at the discretion and agreement of Canterbury Christ Church University, Sidney De Haan Research Centre research team and the Primary Investigator.

Publications relating to the study will comply with Canterbury Christ Church University requirements and recognized ethical standards concerning publications and authorship, including Section II - “Ethical Considerations in the Conduct and Reporting of Research” of the Uniform Requirements for Manuscripts Submitted to Biomedical Journals, http://www.icmje.org/index.html#authorship established by the International Committee of Medical Journal Editors.

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Appendices

Appendix I – Participant Response Questionnaires

‘Singing for Better Breathing’ Singing and COPD Project Evaluation Questionnaire

Dear Singing Group Participant,

Canterbury Christ Church University is running Singing for Better Breathing groups in Kent to gather feedback from participants to assess whether they have gained any benefits from participation and to assess whether they have found new tools useful.

This is the purpose of this questionnaire, which we will ask you to complete at the beginning and end of the course. **It is entirely your choice whether to complete it.**

The questionnaire is **confidential and anonymous**, but on the first page we ask for your date of birth and also your post code. This is so that your questionnaires before and after the course can be matched up.

Please read the statements or questions carefully and tick the answer that is best for you. The questionnaire ends with some further questions about you and there is a space to write any comments you would like to make about the project.

Thank you for your help.

Charlotte Epsley
Primary Investigator
Sidney De Haan Research Centre for Arts and Health
Canterbury Christ Church University

Personal details

Please circle your answers to the following questions or write in the details

Sex/Gender: _____

Date of birth: (*DD-MMM-YY*) _____

Do you have: Asthma COPD Another lung problem

If another please describe _____

How long have you been affected by lung problems? _____ years

Do you smoke? Yes Have given up Never

As an adult, have you been part of a singing group? Yes No

Please give your post code: _____

Personal details

Date of birth: (*DD-MMM-YY*) _____

Please give your post code: _____

The Warwick-Edinburgh Mental Well-Being Scale

Below are some statements about your thoughts and feelings. Please tick the box that BEST describes your experience of each over the last TWO WEEKS.

STATEMENTS	None of the time	Rarely	Some of the time	Often	All of the time
I've been feeling optimistic about the future					
I've been feeling useful					
I've been feeling relaxed					
I've been feeling interested in other people					
I've had energy to spare					
I've been dealing with problems well					
I've been thinking clearly					
I've been feeling good about myself					
I've been feeling close to other people					
I've been feeling confident					
I've been able to make up my own mind about things					
I've been feeling loved					
I've been interested in new things					
I've been feeling cheerful					

The COPD Assessment Test (CAT)

This questionnaire is designed to measure the impact COPD (Chronic Obstructive Pulmonary Disease) has on wellbeing and daily life. It can also identify difficulties experienced due to other lung conditions like asthma. For each item below, circle the number that best describes you CURRENTLY. Be sure to only select one response for each question.

I never cough	0 1 2 3 4 5 6	I cough all the time
I have no phelgm (mucus) in my chest at all	0 1 2 3 4 5 6	My chest is completely full of phelgm (mucus)
My chest does not feel tight at all	0 1 2 3 4 5 6	My chest feels very tight
When I walk up a hill or one flight of stairs I am not breathless	0 1 2 3 4 5 6	When I walk up a hill or one flight of stairs I am very breathless
I am not limited doing any activities at home	0 1 2 3 4 5 6	I am very limited doing activities at home
I am confident leaving my home despite my lung condition	0 1 2 3 4 5 6	I am not at all confident leaving my home because of my lung condition
I sleep soundly	0 1 2 3 4 5 6	I don’t sleep soundly because of my lung condition
I have lots of energy	0 1 2 3 4 5 6	I have no energy at all

Asthma Control Questionnaire

This questionnaire is designed to measure the impact Asthma has on wellbeing and daily life. It can also identify difficulties experienced due to other lung conditions like COPD. For each item below, circle the number of the response that best describes how you have been during the PAST WEEK.

1. On average, during the past week, how often were you woken by your asthma (or other conditions) during the night?

0 Never
1 Hardly ever
2 A few minutes
3 Several times
4 Many times
5 A great many times
6 Unable to sleep because of asthma

2. On average, during the past week, how bad were your asthma symptoms (or other conditions) when you woke up in the morning?

0 No symptoms
1 Very mild symptoms
2 Mild symptoms
3 Moderate symptoms
4 Quite severe symptoms
5 Severe symptoms
6 Very severe symptoms

3. In general, during the past week, how limited were you in your activities because of your asthma (or other conditions)?

0 Not limited at all
1 Very slightly limited
2 Slightly limited
3 Moderately limited
4 Very limited
5 Extremely limited
6 Totally limited

4. In general, during the past week, how much shortness of breath did you experience because of your asthma (or other conditions)?

0 None
1 A very little
2 A little
3 A moderate amount
4 Quite a lot
5 A great deal
6 A very great deal

5. In general, during the past week, how much of the time did you wheeze?

0 Not at all
1 Hardly any of the time
2 A little of the time
3 A moderate amount of the time
4 A lot of the time
5 Most of the time
6 All the time

We are very keen to have feedback from you

How did you hear about this project?
Did your GP or other health care professional recommend you attend?
What do you hope to gain from joining the group?

Please continue overleaf if you need more space

Thank you for completing this questionnaire.
Please check that you have answered all the items.

SfBB Booklet and DVD Resource© - Final Overall Feedback

<u>To be completed only once and after the last session</u>

Circle the number that best describes how you feel about the booklet and DVD overall - complete this section at the end of the programme and only select one response for each question.

I used the booklet and/or DVD regularly	0 1 2 3 4 5 6	I did not use the booklet and/or DVD regularly
I found the instructions in the booklet easy to follow	0 1 2 3 4 5 6	I did not find the instructions in the booklet easy to follow
I found the instructions on the DVD easy to follow	0 1 2 3 4 5 6	I did not find the instructions on the DVD easy to follow
Using the booklet and/or DVD made me feel more confident at the group sessions	0 1 2 3 4 5 6	Using the booklet and/or DVD did not make me feel more confident at the group sessions
It was easy to fit in regular singing practice at home	0 1 2 3 4 5 6	It was difficult to fit in regular singing practice at home
I will continue to use the booklet and DVD to practice	0 1 2 3 4 5 6	I will not continue to use the booklet and DVD to practice
I would recommend the booklet and DVD to others	0 1 2 3 4 5 6	I would not recommend the booklet and DVD to others
I would be likely to use this or other singing resources online or via an electronic device, mobile phone ‘app’ or similar	0 1 2 3 4 5 6	I would not be likely to use this or other singing resources online or via an electronic device, mobile phone ‘app’ or similar

We are very keen to have any other feedback from you about the impact of group singing on you and your lung condition and about the SfBB Booklet and DVD resource

Please continue overleaf if you need more space

Thank you for completing this questionnaire.
Please check that you have answered all the items.

Appendix II – Participant Information Sheet

Participant Information Sheet

Study title: 'Singing for Better Breathing' - Singing and COPD Project

The above study is being conducted by Charlotte Epsley with the Sidney De Haan Research Centre for Arts and Health at Canterbury Christ Church University (CCCU)

Before you decide to take part in the study it is important for you to understand why the research is being done and what it will involve. Please take time to read the following information carefully before making a decision to take part in the study.

Background

A number of recent research studies have shown that regular group singing can be beneficial for people with lung conditions that make breathing difficult, lead to problems of breathlessness and limit physical activity. This study aims to set up and evaluate a number of weekly singing groups for residents in Kent to further research this idea. The aim is to test whether regular community singing is helpful for people with respiratory illness (chronic obstructive pulmonary disease/ COPD) and test whether some tools (a booklet and a DVD) are helpful for practising singing outside the group meetings.

What will you be required to do?

You will have the opportunity to attend a singing group meeting every week in a venue in your neighbourhood for thirteen weeks. We would like you to attend every week if this is possible for you, but understand that your health and other commitments may mean you have to miss occasional sessions. To help us assess the benefits of this activity, you will be asked to complete some questionnaires about this and about health and wellbeing. You have the right to leave blank any questions you would rather not answer.

The data collection exercises will take place at the beginning, during and after the research project. Towards the end of the project we may also invite you to take part in a recorded interview with a member of the research team.

During the study we may also make a documentary record using photography and filming. You will be asked at every stage for your permission to be involved in any photography or filming that happens during the project and the team will make sure that you are not photographed or filmed if you choose not to be involved. You can take part in the project but still refuse to be photographed or filmed without giving any reason for your decision.

To participate in this research, you must

- Be aged 18 or over
- Be able to attend a local venue where a singing group is taking place weekly for 13 weeks
- Be willing to practise at home using a booklet and/or DVD
- Be willing to complete questionnaires
- Be able to complete questionnaires in English (help will be provided)

Confidentiality

All data and personal information will be stored securely within CCCU premises in accordance with the Data Protection Act 1998 and the University's own data protection requirements. Data can only be accessed by members of the research team. After completion of the study, all data will be made anonymous (i.e. all personal information associated with the data will be removed).

Dissemination of results

We will be writing up the results of the study in a report. There will also be a summary of the results which will be available for people who have taken part. We may also publish the results in health journals. A film documentary of the project may also be produced which would be available through the internet. You will be asked for your permission to be involved at every stage of filming and you will have the opportunity to see the film before it is made public to ensure that you are happy to be included.

Deciding whether to participate

If you have any questions or concerns about the nature, procedures or requirements for participation do not hesitate to contact us. If you decide to participate that doesn't mean that you have to continue with the project. You can decide to stop coming to the singing groups and being part of the research project at any time without having to give a reason.

Any questions?

If you have any questions or concerns about the nature, procedures or requirements for participation do not hesitate to contact:

Charlotte Epsley
Sidney De Haan Research Centre for Arts and Health,
Canterbury Christ Church University, 65-69 Tontine Street, Folkestone, Kent CT20 1JR

Email: ce162@canterbury.ac.uk
Tel: 01303 220870

Thank you very much for taking the time to consider being involved in this study

Appendix III – Participant Informed Consent

Canterbury Christ Church University

CONSENT FORM

Title of Project: 'Singing for Better Breathing' – Singing and COPD Project – Initial consenting for participation in the evaluation

Name of Researcher: Charlotte Epsley (Primary Investigator)

Supervisor – Prof. Stephen Clift

Contact details:

Address:	Sidney De Haan Research Centre for Arts and Health, Canterbury Christ Church University, 69 Tontine Street, Folkestone, Kent, CT20 1JR
Tel:	01303 220870
Email:	ce162.canterbury.ac.uk

Please initial box

1.	I confirm that I have read and understand the information sheet for the above study and have had the opportunity to ask questions.	☐
2.	I understand that my participation is voluntary and that I am free to withdraw at any time, without giving any reason.	☐
3.	I understand that any personal information that I provide to the researchers will be kept strictly confidential	☐
4.	I agree to take part in the above study.	☐

_____	_____	_____
Name of Participant	Date	Signature
_____	_____	_____
Name of Person taking consent (if different from researcher)	Date	Signature
_____	_____	_____
Researcher	Date	Signature

Copies: 1 for participant, 1 for researcher

Participant consent forms Version 1 07Apr17

CONSENT FORM

Title of Project: 'Singing for Better Breathing' – Singing and COPD Project – Further consenting for interview, photographing and filming

Name of Researcher: Charlotte Epsley (Primary Investigator)

Supervisor – Prof. Stephen Clift

Contact details:

Address:	Sidney De Haan Research Centre for Arts and Health, Canterbury Christ Church University, 69 Tontine Street, Folkestone, Kent, CT20 1JR
Tel:	01303 220870
Email:	ce162.canterbury.ac.uk

Please initial box

1.	I confirm that I have previously given my consent to participate in this study and understand that my participation is voluntary and that any personal information I provide will be kept confidential	☐
2.	I agree to photography and filming during group singing sessions and combined performance events	☐
3.	I understand that I will have the opportunity to view any images before they are used in reports and presentations on the project and for purposes of disseminating findings from the project	☐
4.	I agree to audio recording of a short interview during group singing session	☐

_____	_____	_____
Name of Participant	Date	Signature
_____	_____	_____
Name of Person taking consent (if different from researcher)	Date	Signature
_____	_____	_____
Researcher	Date	Signature

Copies: 1 for participant, 1 for researcher

Participant consent forms Version 1 07Apr17

Appendix IV – Sidney De Haan ‘Singing for Better Breathing’ Resource© Summary of Songs

SfBB Main Film - with introduction and warm-up exercises.

DVD and Booklet containing recorded sing-a-long films and lyrics to the following:

Song 1 - Daisy Bell
Song 2 - Moon River
Song 3 - Kookaburra
Song 4 - I'd Like to Teach the World to Sing
Song 5 - My Bonnie
Song 6 - Consider Yourself
Song 7 - Train Is A-Comin'
Song 8 - Happy Talk
Song 9 - Some Enchanted Evening
Song 10 - Charlie Is My Darling
Song 11 - The Wild Mountain Thyme
Song 12 - What A Wonderful World

Appendix V – Ethics Committee Approval Letter

Canterbury Christ Church University

7th April 2017 Ref:16/H&W/CL181

Charlotte Epsley
c/o Sidney De Haan Research Centre for Arts and Health
Faculty of Health & Wellbeing

Dear Charlotte

Confirmation of ethics compliance for your study *"Singing for Chronic Obstructive Pulmonary Disease (COPD) – An evaluation of feasibility for self-help booklet & DVD guide:"*

I have received your Ethics Review Checklist and appropriate supporting documentation for proportionate review of the above project. Your application complies fully with the requirements for proportionate ethical review as set out in this University's Research Ethics and Governance Procedures.

In confirming compliance for your study, I must remind you that it is your responsibility to follow, as appropriate, the policies and procedures set out in the *Research Governance Framework* (http://www.canterbury.ac.uk/research-and-consultancy/governance-and-ethics/governance-and-ethics.aspx) and any relevant academic or professional guidelines. This includes providing, if appropriate, information sheets and consent forms, and ensuring confidentiality in the storage and use of data. Any significant change in the question, design or conduct of the study over its course should be notified via email to red.resgov@canterbury.ac.uk and may require a new application for ethics approval. It is a condition of compliance that you must inform me once your research has been completed.

Wishing you every success with your research.

Yours sincerely

Carol Clewlow

cc: Professor Stephen Clift

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Professor Rama Thirunamachandran, Vice Chancellor and Principal

Appendix D – Research Study - Analysis of Results

Analysis – T-Test Tables using SPSS

CAT – CAT total score

WEM – WEMBMS total score

ACQ – ACQ-5 total score

V_0 = Baseline Visit V_0

V_1 = Mid-Term Visit V_1

V_2 = Follow-Up Visit V_2

V_3 = Final Follow-up V_3

Health and Wellbeing Outcomes – Total Score Analysis

Analysis for per protocol dataset i.e. COPD = 1 and Attendance > 2 sessions

Paired Samples Statistics				
	Mean	N	Std. Deviation	Std. Error Mean
$WEMV_0$	48.30	23	12.150	2.533
$WEMV_1$	51.14	23	11.459	2.389
$WEMV_0$	49.14	21	12.397	2.705
$WEMV_2$	55.23	21	10.150	2.215
$WEMV_0$	48.43	16	12.886	3.222
$WEMV_3$	56.81	16	8.719	2.180
$CATV_0$	24.57	22	7.698	1.641
$CATV_1$	22.42	22	9.532	2.032
$CATV_0$	25.03	21	7.774	1.696
$CATV_2$	20.27	21	10.659	2.326
$CATV_0$	24.16	16	7.282	1.820
$CATV_3$	19.13	16	9.563	2.391
$ACQV_0$	11.80	20	5.064	1.132
$ACQV_1$	10.20	20	6.254	1.398
$ACQV_0$	12.05	20	4.989	1.116
$ACQV_2$	10.05	20	6.022	1.346
$ACQV_0$	11.81	16	4.490	1.123
$ACQV_3$	9.81	16	5.833	1.458

Paired Samples Correlations				
	N	Correlation	Sig.	
$WEMV_0$ - $WEMV_1$	23	0.810	0.000	
$WEMV_0$ - $WEMV_2$	21	0.563	0.008	
$WEMV_0$ - $WEMV_3$	16	0.687	0.003	
$CATV_0$ - $CATV_1$	22	0.739	0.000	
$CATV_0$ - $CATV_2$	21	0.680	0.001	
$CATV_0$ - $CATV_3$	16	0.685	0.003	
$ACQV_0$ - $ACQV_1$	20	0.772	0.000	
$ACQV_0$ - $ACQV_2$	20	0.688	0.001	
$ACQV_0$ - $ACQV_3$	16	0.622	0.010	

	Paired Differences					t	df	Sig. (2-tailed)
	Mean	Std. Deviation	Std. Error Mean	95% Confidence Interval of the Difference				
				Lower	Upper			
$WEMV_0$ - $WEMV_1$	-2.836	7.316	1.525	-6.000	0.327	-1.859	22	0.076
$WEMV_0$ - $WEMV_2$	-6.095	10.720	2.339	-10.975	-1.216	-2.606	20	0.017
$WEMV_0$ - $WEMV_3$	-8.380	9.362	2.340	-13.368	-3.392	-3.581	15	0.003
$CATV_0$ - $CATV_1$	2.156	6.451	1.375	-0.704	5.016	1.567	21	0.132
$CATV_0$ - $CATV_2$	4.755	7.828	1.708	1.192	8.319	2.784	20	0.011
$CATV_0$ - $CATV_3$	5.036	7.001	1.750	1.305	8.766	2.877	15	0.012
$ACQV_0$ - $ACQV_1$	1.600	3.979	0.890	-0.262	3.462	1.798	19	0.088
$ACQV_0$ - $ACQV_2$	2.000	4.449	0.995	-0.082	4.082	2.011	19	0.059
$ACQV_0$ - $ACQV_3$	2.000	4.648	1.162	-0.477	4.477	1.721	15	0.106

Health and Wellbeing Outcomes – Total Score Analysis

Analysis for per protocol dataset i.e. COPD = 1 and Attendance > 2 sessions

CAT – CAT total score

WEM – WEMBMS total score

ACQ – ACQ-5 total score

V_0 = Baseline Visit V_0

V_1 = Mid-Term Visit V_1

V_2 = Follow-Up Visit V_2

V_3 = Final Follow-up V_3

Paired Samples Test: COPD =1									
		Paired Differences					t	df	Sig. (2-tailed)
		Mean	Std. Deviation	Std. Error Mean	95% Confidence Interval of the Difference				
					Lower	Upper			
Total WEM Score	WEM V_0-V_1	-2.836	7.316	1.525	-6.000	0.327	-1.859	22	0.076
Total WEM Score	WEM V_0-V_2	-6.095	10.720	2.339	-10.975	-1.216	-2.606	20	0.017
Total WEM Score	WEM V_1-V_2	-3.608	7.456	1.627	-7.002	-0.214	-2.218	20	0.038
Total CAT Score	$CATV_0$ - $CATV_1$	2.156	6.451	1.375	-0.704	5.016	1.567	21	0.132
Total CAT Score	$CATV_0$ - $CATV_2$	4.755	7.828	1.708	1.192	8.319	2.784	20	0.011
Total CAT Score	$CATV_1$ - $CATV_2$	2.422	7.869	1.760	-1.261	6.104	1.376	19	0.185
Total ACQ Score	ACQ V_0-V_1	1.600	3.979	0.890	-0.262	3.462	1.798	19	0.088
Total ACQ Score	ACQ V_0-V_2	2.000	4.449	0.995	-0.082	4.082	2.011	19	0.059
Total ACQ Score	ACQ V_1-V_2	-0.050	4.322	0.966	-2.073	1.973	-0.052	19	0.959

Group Statistics

Sex/Gender *COPD = 1		N	Mean	Std. Deviation	Std. Error Mean
Change WEM FU V_2 score from Baseline V_0*	Male	8	2.88	7.717	2.728
	Female	13	8.08	12.068	3.347
Change CAT FU V_2 score from Baseline V_0*	Male	8	-5.66	11.863	4.194
	Female	13	-4.20	4.376	1.214
Change ACQ FU V_2 score from Baseline V_0*	Male	7	-3.14	6.176	2.334
	Female	13	-1.38	3.330	0.924
SfBB total score(All Pts)	Male	9	10.56	4.667	1.556
	Female	19	17.47	9.851	2.260

Independent Samples Test

* COPD = 1	Levene's Test for Equality of Variances		t-test for Equality of Means						
Gender	F	Sig.	t	df	Sig. (2-tailed)	Mean Difference	Std. Error Difference	95% Confidence Interval of the Difference	
								Lower	Upper
Change WEM FU V_2 score from Baseline V_0*	2.292	0.146	-1.085	19	0.292	-5.202	4.796	-15.240	4.836
			-1.205	18.923	0.243	-5.202	4.318	-14.242	3.839
Change CAT FU V_2 score from Baseline V_0*	6.354	0.021	-0.407	19	0.688	-1.464	3.593	-8.984	6.057
			-0.335	8.188	0.746	-1.464	4.366	-11.492	8.565
Change ACQ FU V_2 score from Baseline V_0*	3.609	0.074	-0.836	18	0.414	-1.758	2.102	-6.175	2.658
			-0.700	7.929	0.504	-1.758	2.510	-7.556	4.040
SfBB total score (All Pts)	2.985	0.096	-1.989	26	0.057	-6.918	3.478	-14.068	0.231
			-2.522	25.977	0.018	-6.918	2.744	-12.558	-1.278

Appendix E – Weekly Progress Information
Singing Leader Feedback and Comments

A high-level summary of singing leader comments along with a selection of unsolicited participant comments made to the singing leader at the Centre A group (recorded verbatim and ad hoc) between Baseline Visit V_0 and Follow-Up Visit V_3 are described below for illustration purposes. No further analysis has been conducted on this data it is presented here to provide supportive information.

Session 1 Comments

> *"Participants were already starting to interact quite quickly after an introduction about the study and 45 minutes of exercises and singing."*

Session 2 Comments

> *"Participants were smiling and starting to mix with each other, exchanging numbers and socialising."*

Session 3 Comments

> *"Participants indicated they were having fun and more relaxed. Starting to discuss what will happen when the study ends and whether they can continue."*

Session 4 Comments

> *"Participants indicated they were having fun some finding it easier and noticing the difference to their breathing. Feeling confident enough to say that song was too high for them to sing."*

Session 5 Comments

> *"Participants indicated they felt relaxed and comfortable. They sang songs in two parts and rounds. Some mentioned that they are practising in the week and finding songs they like at home and looking online e.g. Youtube was mentioned."*

> Participant commented *"Life has been bad with my heath for the past year but singing gives them something to look forward to", "Thoroughly enjoyed it"* and *"I downloaded all the songs and have been singing along non-stop"*

Session 6 Comments

"Participants starting to indicate which songs they preferred to sing and asked if they could join in and sing with other groups."

Session 7 Comments

"Visit from Medway healthcare team member and [study] researchers. [Group] completed assessments at the beginning of the session. Participants indicated songs they preferred to sing and asked if they could join in and sing with other groups."

Participant [that] missed the last two sessions indicated they had not felt able to come out but "Realised that the singing was doing me good - I'm glad that I came back."

Session 8 Comments

"Weather was warm so warm up was reduced so as not to stress the participants or over exert them reminding them to drink and switched on fan."

"Slightly increased attendee number possibly due to the better weather – led to more enthusiasm i.e. humorous chit-chat and anecdotes. Suggested to make the singing more operatic/dramatic to increase movement and make things more fun."

Session 9 Comments

"Attention given to good breathing, then introduced clapping to help with movement."
"Community centre staff looked in and made positive comments which lifted the mood of the group further. One new attendee joined in for just one session. The door to the centre is being left open so that others in the community centre can listen to the singing in the foyer."

"Participant indicated they couldn't play the DVD but was practising his own songs between sessions.
Participant indicated they had returned to Church "just for singing (I'm not religious just go for the singing)"
One participant indicated she had a bad experience on the way and was deciding whether to stay – indicated that she was very glad she stayed as she felt better for it."

Session 10 Comments

“Attention given to good posture and finding space and practising a ‘diving into a pool’ position to stretch the core and also working on sitting posture. Discussed attention to vowels and putting energy into the singing including singing with arms outstretched.”

“Group seemed confident from the first song today, enthusiastic and they commented they had lost inhibitions with each other. Chit-chat and socialising before the session - participants are arriving early to meet in the café and stay afterwards to socialise.”

“Participants commented “It was a good day”, “It went exceptionally well”, “It was good for us to meet and work together” and “It was lovely to come and sit and relax and sing – wonderful.””

Session 11 Comments

“Good session extra songs sung. Comments [were] raised again about meeting and singing with other groups. Laughter and collaboration and all stayed to have refreshments together and chat at the end.”

Participants commented “*It’s so enjoyable*.” and “*I’m looking forward to it each week.”*

Session 12 Comments

“Participants swapped contact details and discussed the differences between small and large sized singing groups.”

Participants commented *“we might be a small group but we have done really well.” and “It’s really social and we had got to know each other well.”*

Session 13 Comments – post-study period

“Group are planning to attend the session of another group tomorrow both to sing and discuss sustainability of the groups. All but one participant will be attending."

Participants commented “*I’ve never really sung before I used to always mime” and “This singing has given me a new lease of life and given me confidence”*

Appendix F – SfBB Questionnaire Output Summary

COPD = 1 (COPD Diagnosis)

COPD = 2 (CRD Diagnosis other than COPD)

COPD = 3 (Undiagnosed)

ID #	Gender	Year of Birth	COPD	Q1	Q2	Q3	Q4	Q5	Q6	Q7	Q8	SfBB total
1001	Female	1948	1	ND	5	ND	ND	5	2	1	3	NA
1002	Female	1948	1	6	1	ND	ND	0	0	0	0	NA
1003	Female	1941	2	2	3	4	6	1	3	5	5	29
1004	Male	1939	1	4	1	1	1	1	1	1	1	11
1006	Female	1957	1	2	2	1	3	1	1	1	1	12
1010	Female	1964	1	0	0	0	0	0	0	0	0	0
1011	Female	1965	2	3	1	1	3	4	1	3	1	17
1012	Male	1962	3	0	1	1	1	0	1	1	2	7
2001	Female	1940	1	3	1	1	2	4	3	1	5	20
2002	Female	1940	1	3	3	3	2	2	5	2	3	23
2003	Male	1946	1	3	0	0	3	5	1	0	5	17
2004	Male	1935	1	3	2	2	1	0	0	0	0	8
2007	Female	1967	1	2	0	0	0	0	0	0	0	2
2008	Female	1955	2	3	1	1	5	3	5	1	1	20
2009	Female	1955	1	3	4	4	0	3	3	3	3	23
2010	Female	1952	1	5	4	4	2	2	3	3	2	25
2011	Male	1954	1	1	1	1	1	4	2	1	1	12
2013	Male	1954	1	3	3	2	2	2	2	2	2	18
2014	Male	1957	1	3	0	0	0	1	0	0	0	4
2015	Male	1947	1	3	0	0	0	4	0	0	0	7
2016	Female	1960	1	2	1	1	1	2	1	1	2	11
2017	Male	1943	1	3	0	0	2	4	2	0	0	11
2018	Female	1945	1	6	ND	6	6	6	6	6	6	48
2019	Female	1953	2	5	1	1	1	3	3	1	3	18
2020	Female	1959	1	2	0	0	2	2	2	0	0	8
2022	Female	1957	3	3	0	0	0	5	1	0	5	14
2023	Female	1937	1	3	3	1	3	4	4	3	2	23
2025	Female	1952	3	4	3	2	3	3	3	2	2	22