RESEARCH ARTICLE

SingStrong—A singing and breathing retraining intervention for respiratory and other common symptoms of long COVID: A pilot study

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Background: Management of Long COVID (LC) is hugely challenging for clinicians. This pilot study evaluated a breathing retraining and singing programme (SingStrong for LC) to address common LC symptoms. The study hypothesized that this intervention would improve symptoms impacting disordered breathing and participant wellbeing.

Methods: The 10-week, bi-weekly online programme was comprised of a 45-min class of mindfulness, breathing retraining, vocal exercises, and singing. Sessions were recorded for non-attenders and conducted by a trained vocal coach experienced in respiratory cohorts. Persons with a confirmed COVID-19 diagnosis and persisting symptoms were invited to participate. Demographic and COVID-19 data were collected, and the DePaul Symptom Questionnaire Short Form (DSQ- SF) and COVID-19 Yorkshire Rehab Screen questionnaires were administered. Post-intervention focus groups were also conducted.

Results: Of 27 (F = 23(85%)) participants recruited, data from 21 who completed at least 10 (50%) classes were analysed. Participants showed significant pre–post-intervention improvements in all breathlessness symptoms (at rest: $P < 0.001$; dressing: $P = 0.01$; stairs: $P < 0.001$), fatigue ($P = 0.03$), usual activities ($P = 0.04$), pain/disability ($P = 0.03$), voice quality ($P = 0.01$), and communication/cognition ($P = 0.04$). Pre–post number of instances meeting DSQ-SF criteria for myalgic encephalomyelitis (ME) and chronic fatigue syndrome (CFS) decreased by a net of nine cases (14.3%). No association between COVID-19 hospitalisation status and diagnosis of ME/CFS was identified. Qualitative feedback from eight participants was overwhelmingly positive with all reporting improvements in breathing and general well-being.

Conclusion: The SingStrong programme shows promise as a viable treatment option for LC sufferers. Future studies are required to further investigate the efficacy of this intervention.

Key Words: *long COVID; singing rehabilitation; breathing retraining; dis-ordered breathing; lung disease*

INTRODUCTION

At the time of writing, there have been approximately 243 million confirmed cases of the Severe Acute Respiratory Syndrome Coronavirus-2 (SARS-CoV-2) or COVID-19 virus reported globally, claiming almost five million lives (https://COVID19.who.int/ (accessed 27 October 2021)). To date, 48.9% of the world's population has received at least one vaccination shot, but this figure falls to just 3.1% in low-income countries (https://ourworldindata.org/COVID-vaccinations (accessed 27 October 2021)). The trajectory of the virus remains uncertain as new COVID-19 variants threaten to undermine the efficacy of vaccines and prolong national lockdowns and recessions [1]. Scientists continue to learn about the pathophysiology and phenotypes of the virus in efforts to address the infectivity and transmissibility of this complex, multifaceted disease. However, as treatment efficacy has improved for those with acute disease [2], the clinical burden posed by so-called Long COVID (LC) grows more concerning [3].

The definition of LC, known also as "post-acute COVID-19 syndrome" or "post-acute sequelae of SARS-CoV-2 infection", has yet to be conclusively agreed. However, the term has previously been described as referring to persons with COVID-19 who experience symptoms for >28 days after initial diagnosis [3]. The persistent impact of symptoms and the onset of new problems post resolution of initial infection is a concerning trend in a significant proportion of COVID-19 patients [4]. Reported prevalence of LC fluctuates, but rates of 76% and 87% of hospitalised Chinese patients at 6 months [5] and an Italian cohort 60 days post COVID-19 onset [6], respectively, have been recorded. The duration of the condition is similarly unpredictable, with reports of symptoms persisting from a few weeks to many months [7].

The likelihood of developing LC is difficult to predict, with variability in age, sex, and acute disease severity all reported in affected patients [3]. A recent retrospective study of over 4000 patients found that

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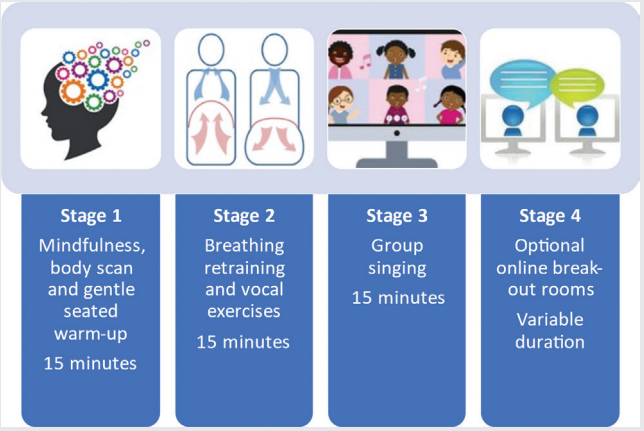
FIGURE 1
Stages of the SingStrong intervention.



older age, female sex (except females over 70 years), those who had required hospital assessment and a pre-existing asthma diagnosis were all associated with LC symptoms persisting for longer than 28 days [8]. In contrast, other studies have found no relationship between initial disease severity and developing LC [9]. Unsurprisingly, as dozens of LC phenotypes have been identified to date, detecting and predicting those at risk remains extremely challenging [10].

The spectrum of LC presentations is also bewildering with over 50 symptoms identified in a large systematic review on the subject [7]. Of these, the most common symptoms were fatigue, headache, attention disorder, hair loss, and dyspnoea. Common pulmonary symptoms included persistent cough, chest discomfort, reduced pulmonary diffusing capacity, sleep apnoea, and pulmonary fibrosis. Cardiac issues such as arrhythmias and myocarditis were also commonplace as were a range of neuropsychological issues including dementia, depression, and anxiety. Unsurprisingly then, the management of LC is challenging, requiring individualised assessment of the needs and symptoms of each patient. Available guidelines provide high level advice recommending extensive screening and examination, administration of medication for any acute and/or chronic conditions, education and ongoing follow-up [11]. However, effective management is further complicated by an inconsistent relationship between clinical findings and subjective patient symptoms [12].

In some patients with respiratory issues in particular, the degree of lung damage and gaseous exchange impairment observed does not always account for the marked levels of dyspnea, fatigue, and weakness reported [13]. Disordered breathing is postulated to precipitate weakness and fatigue due to ineffective ventilation and subsequent suboptimal gaseous exchange caused by poor recruitment of the diaphragm and intercostal respiratory muscles and exacerbated by apical breathing associated with anxiety [14]. It has further been proposed that the psychosocial trauma of LC may contribute to and exacerbate these symptoms, thus requiring a more holistic management strategy [9]. Inspiratory muscle training has been found to be effective for mechanically ventilated COVID-19 patients, improving pulmonary function, dyspnoea, and quality of life [15]. Disordered breathing in other respiratory cohorts such as those with Chronic Obstructive Pulmonary Disease (COPD) has benefitted similarly from breathing retraining using singing exercises and vocal techniques [16]. Singing interventions in lung pathologies aim to improve the efficiency of respiration through optimal recruitment of the primary muscles of respiration, while simultaneously enhancing quality of life and ameliorating psychological distress [17]. A systematic review exploring the benefits of singing for lung health more broadly found such interventions have the potential to improve bio-psychological health-related quality of life, without incurring significant side effects [18].

This current study conducted with a cohort of patients with LC in the Republic of Ireland hypothesised that a program of breathing retraining and singing techniques (SingStrong) would improve respiratory and other symptoms including fatigue in this group.

METHODS

This pilot study used a one-group pre-test–post-test research design with a mixed-methods approach to collect and evaluate data. Ethical Approval for this project was provided by the ethics committee of the relevant faculty in the local University (2019_04_06_EHS).

Participants

The following inclusion and exclusion criteria applied:

Inclusion:

- Adults aged 18 or older who had a confirmed clinical diagnosis of COVID-19.
- People still experiencing respiratory symptoms and/or increased fatigue at least 28 days post diagnosis.
- Good standard of spoken and written English.
- Access to appropriate technology and internet to access online classes.

Exclusion:

- Persons in the acute stages of COVID-19 (<28 days).
- Those with symptoms of LC that had resolved/largely resolved.
- Those who were actively engaged in singing on a regular basis or engaged in singing as a therapeutic intervention.
- Cognitive deficits that would impair the ability to participate in the intervention or answer questionnaires appropriately.

There were no limitations on whether participants required hospitalisation or supportive care such as intubation or non-invasive ventilation during their COVID-19 illness. Similarly, participants were not excluded if they were actively undertaking medical treatment for their LC elsewhere (apart from singing interventions). Participants were recruited through an information campaign shared with and by the clinical networks of the principal investigator (PI), promotion on various social media outlets including Twitter, and targeting of LC groups on Facebook and Instagram. Owing to the method of recruitment, it was impossible to accurately calculate recruitment rate. Prior to commencement of the intervention, potential participants were invited to attend an online information session with the PI. All participants provided signed informed consent.

Intervention

The SingStrong LC intervention was a 10-week, bi-weekly series of breathing retraining and singing classes, which was delivered live and online using Zoom™ between April and June of 2021. Classes were 45 min in

duration and included an initial mindfulness and body-scanning relaxation exercise, physical warm-up session, vocal and breathing exercises, and singing (Figure 1). The classes were designed by the main author (RC) who is an experienced respiratory physiotherapist and delivered by a trained vocal coach and choir leader (CM) who previously led SingStrong projects in other respiratory cohorts [16]. CM had also previously upskilled to facilitate singing classes for people with respiratory conditions. The structure of the classes was split in three; 15 min for mindfulness, head to toe body scanning, and gentle, seated, physical warm-ups. This was followed by 15 min of vocal and breathing exercises, followed by a final 15 min of singing. This breakdown reflected a growing body of evidence about the fatigue and limited physical reserves of patients with LC [19]. Songs were chosen in collaboration with participants, with consideration given to participant breathing capacity. Online breakout rooms after the classes were also offered to the participants to support a sense of community within the project but were not mandatory, monitored, or recorded. Classes were video recorded and made available to participants unable to attend in real time to facilitate compliance, and to encourage practice between classes. Participants were reminded to advise the PI if they caught up with classes using recordings to ensure accuracy in attendance figures.

Data collection

Online questionnaires were used pre- and post-intervention to collect data. Pre-intervention data included demographic data (age, sex, employment status, occupation, ethnicity), health-related data (health complaints prior to COVID-19 diagnosis, weight, self-reported overall health status pre-COVID-19 diagnosis), and details of acute COVID-19 experience (date of diagnosis, details of hospitalisation if applicable, treatments administered). Additionally, the COVID-19 Yorkshire Rehab Screen (C19-YRS) [20] was used. This is a 19-item questionnaire that collects data on multiple biopsychosocial aspects of health that the participant rates on a scale from 0 (worst) to 10 (best) for both pre-COVID and currently. Symptoms are dichotomised for analysis into domains of disease severity and impact/disability, with an additional Post Traumatic Stress Disorder (PTSD) screen included in the C19-YRS tool. Finally, the DePaul Symptom Questionnaire Short Form (DSQ-SF) was administered [21]. This is a widely used instrument that assesses typical symptoms of myalgic encephalomyelitis (ME) and chronic fatigue syndrome (CFS), commonly present in patients with LC [22]. The DSQ-SF asks about the severity and frequency of 14 common symptoms such as fatigue, pain, and brain fog, which the respondent rates from 0 (best) to 4 (worst). The DSQ-SF algorithm has excellent accuracy (>85%) in correctly identifying individuals diagnosed with ME and CFS by established methodologies including the Fukoda criteria, the Canadian ME/CFS criteria, and the Institute of Medicine (IOM) criteria [21].

The questionnaire was piloted and required approximately 20 min to complete. Only the C19-YRS and DSQ-SF were re-administered post-intervention. Focus groups were also administered on completion of the intervention with willing participants who had attended at least three of the 20 classes. The focus groups were administered by a researcher previously unconnected with the project. A semi-structured interview approach was adopted focusing on the experience of the intervention and impact (if any) on the health of participants. Focus groups were conducted via Microsoft Teams™ and were audio recorded for verbatim transcription. Themes were identified following multiple re-reading of transcripts. Line by line coding of the transcripts was conducted to identify themes. The data analysis was conducted manually since the quantity of text was amenable to manual inspection.

Data were anonymized and stored in the online cloud system of the host University. The system is General Data Protection Regulation compliant and only the authors had access to the relevant data files. The encryption key to decode data was stored on the secure laptop of the lead author.

Statistical analysis

Descriptive statistics are reported using mean and standard deviation (SD) for normally distributed variables, median (interquartile range (IQR)) for non-normal variables, and percentages for categorical variables. The distributions of all numeric variables were assessed for skewedness using formal tests (Kolmogorov–Smirnov and Shapiro–Wilk [23]) and through visual inspection of histograms. Chi-squared tests were used to investigate association between categorical variables. Paired sample *t*-tests were used to evaluate pre–post data, with Student's *t*-test (normal distribution) or Wilcoxan's *W* (non-normal distribution) used as appropriate. Statistical significance was set at $P \leq 0.05$. Familiarization with the focus group data was supported by verbatim transcription of the interviews. Iterative re-reading of scripts and reflexive note-taking was employed by the PI to track and record emerging impressions [24]. Line by line coding of written transcripts was conducted to identify themes. The data analysis was conducted manually due to the relatively small quantity of text.

RESULTS

Participants: Demographics

A total of 27 participants (female *n* = 23 (85%), mean (Standard Deviation (SD)) age: 48.4 (10.1) years) completed the baseline questionnaire. Of these, all except one were of white Irish ethnicity with the remaining participant being of "other" white ethnicity. Occupational status and occupation (when healthy) are illustrated in Table 1. Nine (33.3%) participants reported no medical history of note prior to their COVID-19 diagnosis. Of the remaining 18 participants, 17 (63.9%) reported between one and four pre-existing medical conditions with one participant reporting nine conditions. A total of 41 conditions in all were reported of which the most cited were asthma (*n* = 7 (17.1%)), hyperlipidaemia (*n* = 4 (9.7%)) and hypertension (*n* = 3 (7.3%)). Fifteen (55.5%) participants stated that they had a healthy body weight with the remainder (*n* = 12 (44.4%)) stating that they were overweight. It was not possible to collect objective anthropometric data to verify this self-reported response. Mean (SD) overall self-reported health status pre-COVID rated from 0 to 10 was 7.4 (5.5) as opposed to 3.7 (2.3) at the time of the baseline questionnaire.

COVID-19 experience

The median (IQR) time since COVID-19 diagnosis was 12 (4–13) months. Eleven (40.7%) participants were hospitalised, with a mean (SD) length of stay of 4.5 (2.8) weeks. Of these 11 participants, six (54.5%) received care in a standard non-critical care ward, two (18.1%)

TABLE 1

Current occupational status and occupation prior to COVID-19 diagnosis

Occupational status	*n*	%	Occupation (Pre-COVID)	*n*	%
Employed: 37 or more hrs p. week	8	29.7	Medical/health care professional	8	29.7
Employed: 1–36 h p. week	6	22.2	Education sector	7	25.9
Self-employed	3	11.1	Retired	3	11.1
Not employed and not looking for work	2	7.4	Business owner / Self-employed	2	7.4
Retired	2	7.4	Business professional	2	7.4
Disabled and unable to work	6	22.2	Milliner	1	3.7
			Restaurant/bar worker	1	3.7
			Technology/engineering	1	3.7
			Administrator	1	3.7
			Artist, creative entrepreneur	1	3.7
Total	**27**	**100**	**Total**	**27**	**100**

TABLE 2

DePaul Symptom Questionnaire Short Form: Participants with diagnostic indicators of myalgic encephalomyelitis and chronic fatigue syndrome

		Hospitalised						
Criteria		**Yes**	**No**	**Total**	**χ^2 value**	***P***	**OR**	**CI**
Fukoda	Yes	10	11	21	1.85	0.174	0.22	0.02–2.22
	No	1	5	6				
Canada	Yes	4	11	15	2.77	0.096	3.85	0.76–19.50
	No	7	5	12				
IOM	Yes	6	11	17	0.564	0.453	1.83	0.37–8.98
	No	5	5	10				

Note: IOM, International Association of Medicine; OR, Odds Ratio; CI, Confidence Interval.

TABLE 3

DePaul Questionnaire (Short Form): Meeting diagnostic criteria for chronic fatigue syndrome/myalgic encephalomyelitis pre- and post-SingStrong intervention

Participant	Pre: Fukoda	Post: Fukoda	Pre: Canada	Post: Canada	Pre: IOM	Post: IOM
1	Yes	Yes	Yes	No	Yes	No
2	Yes	Yes	No	No	Yes	Yes
3	Yes	Yes	Yes	Yes	Yes	Yes
4	Yes	Yes	Yes	Yes	Yes	Yes
5	Yes	Yes	Yes	Yes	Yes	Yes
6*	No	N/A	No	N/A	No	N/A
7	Yes	Yes	Yes	Yes	Yes	Yes
8	Yes	Yes	Yes	No	Yes	No
9*	Yes	N/A	Yes	N/A	Yes	N/A
10	No	Yes	No	Yes	No	Yes
11*	No	N/A	No	N/A	No	N/A
12	Yes	Yes	Yes	Yes	Yes	Yes
13	No	No	No	No	No	No
14	No	No	No	No	No	No
15	Yes	Yes	Yes	Yes	Yes	Yes
16	Yes	Yes	No	No	No	No
17	Yes	Yes	Yes	Yes	Yes	Yes
18*	Yes	N/A	No	N/A	Yes	N/A
19	Yes	No	No	No	No	No
20	Yes	Yes	Yes	No	Yes	No
21	Yes	Yes	Yes	No	Yes	No
22*	Yes	N/A	Yes	N/A	Yes	N/A
23	Yes	Yes	Yes	No	Yes	No
24	No	No	No	No	No	No
25*	Yes	N/A	Yes	N/A	Yes	N/A
26	Yes	No	No	No	No	Yes
27	Yes	No	No	No	No	No

Note: IOM, Institute of Medicine; N/A, Not Applicable. Green indicates change in diagnosis of ME/CFS from positive to negative; red indicates change in diagnosis of ME/CFS from negative to positive.

*Participants who were not included in post-intervention evaluation.

were admitted to an intensive care (ICU) or high dependency unit (HDU), and the remaining three patients (27.2%) were treated in both settings. Patients in ICU/HDU spent on average 5 weeks in these settings. Of these patients, one was ventilated, one required a tracheostomy, and three received non-invasive ventilation. For patients treated on non-critical care wards, supplemental oxygen was the main treatment administered to seven participants, followed by proning (n = 4), and chest physiotherapy (n = 2).

According to the DPQ-SF, 21 participants (77.8%) met the diagnostic criteria for ME/CFS per the Fukoda criteria, 15 (55.5%) met the Canadian ME/CFS criteria, and 17 (62.9%) met the IOM criteria for ME/CFS. There was no significant association between those patients hospitalised and those who met the criteria for ME/CFS under any of the algorithms evaluated by the DPQ-SF (Table 2).

Attendance at SingStrong classes

The mean (SD) attendance at the online classes was 13.3 (5.9) and ranged from 0 to 20 classes. Of the initial 27 participants, a mean (SD) class attendance of 18.3 (3.9) participants attended each of the twenty classes. Mean (SD) number of participants attending synchronously online was 12.9 (4.1) and catching up using the recording was 5.3 (1.9). There were five dropouts from the programme within 3 weeks of commencement. One reported that his symptoms had resolved but there was no contact from the remaining participants.

Post intervention DPQ-SF analysis

Only data from participants who had completed 10 or more classes (50%) were included in the analysis (n = 21, (F = 19 (91.5%)). All of these participants completed the post-intervention survey. Analysis of the DPQ-SF showed a reduction in the number of participants meeting the criteria set by three algorithms (Fukoda: n = 15 (71.4%), Canadian model: n = 8 (38.1%), IOM: n = 10 (47.6%)). Across the three algorithms and 21 participants, there were 13 instances of improvement (no longer meeting criteria for CFS/ME) and four instances of dis-improvement (newly meeting criteria for CFS/ME) (Table 3).

TABLE 4

Severity and impact/disability domains of the COVID-19 Yorkshire Rehab Screen—Disease severity domain

Variable	Pre-COVID mean (SD)/ median (IQR)	Intervention baseline mean (SD)/median (IQR)	Post-intervention mean (SD)/ median (IQR)	Statistic		*P*	Effect size	
Breathlessness at rest	0 (0–0)	2.5(2.2)	1(0–2)	Student's *t*	–4.5	**<0.001**	Cohen's *d*	–0.99
				Wilcoxon *W*	0	**<0.001**		
Breathlessness dressing	0 (0–0)	4.0 (2.4)	2 (1–3)	Student's *t*	2.89	**0.01**	Cohen's *d*	0.62
				Wilcoxon *W*	142.5	**0.01**		
Breathlessness stairs	0 (0–1)	6.7(2.5)	3 (3–5)	Student's *t*	5.11	**<0.001**	Cohen's *d*	1.12
				Wilcoxon *W*	166	**<0.001**		
Mobility	0 (0–4)	4.4(3.0)	4.1(3.1)	Student's *t*	0.44	0.67	Cohen's *d*	0.010
				Wilcoxon *W*	54.5	0.55		
Fatigue	0 (0–1)	8 (5.3–9)	5.5(3.0)	Student's *t*	2.41	**0.03**	Cohen's *d*	0.53
				Wilcoxon *W*	115	**0.02**		
Personal Care	0 (0–0)	3 (0–5)	2 (0–5)	Student's *t*	0.65	0.52	Cohen's *d*	0.14
				Wilcoxon *W*	47.5	0.53		
Usual activities	0 (0–0)	8 (5–9)	5.7 (3.2)	Student's *t*	**2.24**	**0.04**	Cohen's *d*	0.49
				Wilcoxon *W*	129.5	**0.05**		
Pain/disability	0 (0–0)	4.7 (2.8)	3.7(2.9)	Student's *t*	2.39	**0.03**	Cohen's *d*	0.52
				Wilcoxon *W*	112	**0.02**		
Anxiety	0 (0–3)	6 (2–7)	4.0 (2.5)	Student's *t*	1.84	0.08	Cohen's *d*	0.40
				Wilcoxon *W*	90	0.09		
Depression	0 (0–0)	2 (0–5)	2 (0–3)	Student's *t*	0.98	0.337	Cohen's *d*	0.21
				Wilcoxon *W*	42.5	0.417		

Note: Pre–post analysis of Severity variables performed for change between intervention baseline and post-intervention only. Pre Covid-19 data provided for information only. Bold text indicates statistically significant change. SD, standard deviation; IQR, inter quartile range. SD, standard deviation; IQR, inter quartile range.

TABLE 5

Severity and impact/disability domains of the COVID-19 Yorkshire Rehab Screen—Disease impact/disability domain

Variable	*N* = Yes @ Intervention baseline & post-intervention	Intervention baseline mean (SD)/median (IQR)	Post-intervention mean (SD)/ median (IQR)	Statistic		*P*	Effect size	
Airway complications	7 (33.3%) 9 (42.9%)	0 (0–5)	0 (0–4)	Student's *t*	–0.48	0.64	Cohen's d	–0.10
				Wilcoxon *W*	14	0.34		
Voice quality/issues	14 (66.6%) 11 (52.8%)	4 (0–6)	2 (0–4)	Student's *t*	2.79	**0.01**	Cohen's d	0.61
				Wilcoxon *W*	81	**0.01**		
Swallowing	3 (14.3%) 2 (9.5%)	0 (0–0)	0 (0–0)	Student's *t*	0.78	0.44	Cohen's d	0.18
				Wilcoxon *W*	3	1.00		
Nutrition/appetite	3 (14.3%) 2 (9.5%)	2 (0–4)	1 (0–4)	Student's *t*	–0.10	0.92	Cohen's d	–0.02
				Wilcoxon *W*	46.5	0.972		
Cognition/Communication	16 (76.2%) 13 (62.0%)	4.3 (3.3)	3 (0–5)	Student's *t*	2.19	**0.04**	Cohen's d	0.48
				Wilcoxon *W*	58	**0.03**		

Note: Pre–post analysis of Severity variables performed for change between intervention baseline and post-intervention only. Pre Covid-19 data provided for information only. Bold text indicates statistically significant change. SD, standard deviation; IQR, inter quartile range.

Analysis of C19-YRS

There were significant dis-improvements in all elements of the severity domain of the C19-YRS between the healthy baseline and intervention baseline of participants. These data were not collected for the impact/disability domain by the C19-YRS. Significant pre- to post-intervention improvements were observed in all breathlessness elements (at rest, dressing, stairs), fatigue, performance of usual activities and pain/disability (Severity domain; Table 4), and voice quality and cognition/communication (Impact/disability domain; Table 5).

At intervention baseline, three participants (four post-intervention) reported difficulty controlling their bowel since COVID-19 onset and two had bladder issues (three post-intervention). One participant had both bowel and bladder issues associated with COVID-19, which were unchanged post-intervention. Eighteen participants had COVID-associated new or worsened difficulty with concentration and 17 had new or worsened issues with short-term memory. These factors were unchanged post-intervention. There was a high level of PTSD reported, related to participants' COVID-19 illness and/or hospitalisation. Between 10 and 13 participants reported unpleasant dreams, memories, and active avoidance strategies to stop thinking about the experience at intervention baseline. These figures remained largely unchanged after the intervention. At no time point did any participant express self-harm ideation.

Focus group feedback

Two focus groups of four participants each, lasting approximately 50 min were conducted (female: n = 6). Participants were allocated to focus groups based on their availability. All participants had attended at least 10 classes (range 12–20). The main themes discussed were as follows:

Pre-conceptions, concerns, and motivations

Some participants had no previous knowledge of SingStrong or similar programmes. Others had heard of the programme on the media or through family members, or they had heard of similar singing interventions with other patient groups (e.g., Alzheimer's) or LC cohorts in other countries. Motivations included professional recommendations from clinicians managing their care; in response to symptoms noticed by their families; or simple desperation.

> Participant (P) 3: "I was singing ...and my daughter said your voice sounds so different. (It) prompted me like you know, obviously I'm not breathing properly. So, I thought that the program would really help that."

> P7: "I was getting pretty desperate so I was willing to try anything basically."

While some participants were excited about starting the programme, others cited concerns such as a lack of singing ability or a worry that it may dis-improve their symptoms;

> P3: "my voice was so bad at that stage, and I would get breathless quite a lot. I was a little bit nervous that it might actually make things worse."

Participants noted that any anxieties about the program were allayed by effective communication and the warm and friendly atmosphere created by the PI and SingStrong vocal coach.

Structure and delivery of classes

Participants were asked about the duration, composition and delivery of the 10-week programme. On the whole, most participants stated that they would have liked if the programme continued on for longer than 10 weeks, and also for the class duration to be longer than 45 min. They felt that while elements of relaxation and breathing techniques were valuable, more actual singing would have been welcome. However, there was a recognition that this might have been exhausting for some member.

> P1: "if we want to sing maybe we need to do all the stuff that Ciara (vocal coach) was doing with us and then a little bit more singing at the end and make it a longer class. But then you run into the problem of people getting too tired.... So it's going to be an awkward one because we're different."

Evening classes were more popular than daytime classes due to the pressure of work, and recordings were very popular to allow people to practice during the week or to catch up on missed classes. Also, as one participant noted:

> P2: "I definitely found it was good to have them there—the recordings, for when you missed a class. But I would never miss a class thinking, oh I'll do the recording instead."

Breakout rooms were poorly attended, and reasons given were a lack of time or fatigue after the class, or a fear of rooms becoming a forum for negativity.

> P5: "You're dealing with what you had gone through yourself and to be honest I didn't want to be taking on board anyone else's symptoms."

However, there was a frequently cited desire for a social forum, perhaps to be scheduled outside of class days, to allow participants to build on relationships the developed out of the classes.

Impact of intervention

Participants overwhelmingly reported a positive experience of the programme and improved biopsychosocial health upon its conclusion.

> P7: "It's had a really positive impact on my well-being... I get a lot more out of my day, I'm able to pace myself better. I'm able to keep my heart rate down, a huge change from being stuck in bed for 23 hours a day."

> P8: "My singing will never improve but my breathing has."

> P2: "It made me feel good. It was good for my mental health, to be there and to do this and to have a bit of fun.... I definitely learned lots in terms of how to better use my voice to continuously be aware of breathing in the correct way."

> P6: "I'm walking pretty much like I was before, but I find inclines hard still."

Suggestions for improvements

In addition to suggestions regarding additional social support, class timing, duration and programme length, as already discussed, there were a number of other suggestions regarding the programme. Practical considerations such as providing lyrics sheets for singing and videos of various breathing techniques were proposed as aids to memory, particularly as brain fog was a common problem in this cohort. An important point was also made regarding the heterogeneity of participants in the study:

> P7: "There were two distinct groups in in the programme ... there was one group that had very, very severe lung issues and were often struggling to keep up with breathing and singing exercises. And then, my side of things, which is that the singing was no problem but the some of the warm-ups and the relaxation techniques that ironically were very difficult."

DISCUSSION

This pilot study explored the impact of an online singing and breathing retraining intervention on the respiratory and other common symptoms of people with LC. The results showed the potential for such an intervention to positively impact on the biopsychosocial wellness of these patients, as well as establishing the feasibility of such a programme.

The prognosis for people with LC remains uncertain due to the novelty of the condition and the plethora of associated presentations and symptoms. The multi-organ nature of LC adds to the complexity of care required for these patients. Recent NICE guidelines (https://www.nice.org.uk/guidance/ng188 (accessed 27 October 2021)) support the importance of advice and education in managing breathlessness in this cohort, and non-pharmacological treatment strategies for chronic dyspnoea including breathing retraining exercises, pulmonary rehabilitation, and optimal body positioning are also recommended. Evidence for the treatment of other common LC symptoms including fatigue and cognitive impairments (so-called "brain-fog") is still emerging, but holistic management in a supportive setting is advocated [25]. The prescription of physical exercise however is controversial due to frequent issues with post-exercise malaise, and it is to be used with caution according to a recent NICE statement [26].

The SingStrong intervention adopted these general LC management principles to create a novel intervention for the holistic management of respiratory and other common LC symptoms. This pilot study indicates that benefits to mental and physical health may result, particularly in relation to breathlessness, fatigue, pain and function, voice quality, communication, and cognition. The study also saw an overall improvement in the number of people who meet a diagnosis of CFS/ME as measured by the DSQ-SF. It is plausible that these improvements do not reflect any measureable change in lung function or

morphology, but more likely a greater control and mastery of breathing. A high level of anxiety was noted at intervention baseline in these patients, which may have exacerbated physical symptoms of breathlessness, pain, and fatigue. Similar findings have previously been reported in patient cohorts with COPD [27]. Regaining mastery over effective breathing is likely to have positively impacted these and related factors such as endurance and activities of daily living. Targeting appropriate diaphragmatic breathing as well as using relaxation techniques such as body-scanning and mindfulness appears to have been a popular and effective strategy with this cohort. Adverse changes to voice quality in this population have been attributed to laryngological damage caused by persistent coughing, and also insufficient airflow due to pulmonary involvement [28]. Improvements in this facet of participant health was likely due to instruction in proper vocal technique in addition to enhanced breathing control.

The SingStrong intervention proved on the whole to be extremely popular with participants, but there are undoubtedly improvements to be made. Despite clear inclusion criteria, the cohort recruited for this study was quite heterogeneous, in terms of initial COVID-19 experience and severity of LC symptoms. While this may reflect clinical reality, it is advisable where possible to group patients by main LC issue, thus allowing for more targeted management. Levels of fatigue of participants is also an important consideration, as is encouraging participants to go at their own pace. The use of videos and other supports is a valuable asset to this population allowing patients to access resources when they feel physically able, and as a cue to those patients suffering the debilitating effects of brain fog. This practice may additionally empower patients to self-manage their recovery and symptoms as much as possible, which is a fundamental tenet in the management of any chronic condition [29]. Grouping patients of similar ability would also allow clinicians to tailor class length, by considering the capacity and fatigue limits of the group. Finally, opportunities should be sought to create non-clinical social interaction between members if so desired. This may allow for a patient-led community of support to further enhance well-being.

Limitations of this study include the aforementioned heterogeneity of the recruited cohort, and the relatively small sample size. There was no capacity or opportunity for objective clinical testing which may have further bolstered results. Additionally, there was no focus group representation from poor attenders despite invitation and, thus, a lack of valuable feedback that may have been garnered from those participants. Information was not collected on additional clinical supports available to participants during the trial period, thus it is difficult to know how much of the reported change was attributable solely to the SingStrong intervention. Participants also self-reported how often they watched the recorded session videos off-line and this may have been subject to bias and inaccuracy. The absence of a control group further limits robust interpretation of results and would be an appropriate strategy for future studies. However, design choice was a pragmatic decision based on available participants and complications presented by limited and piecemeal services available to the LC population at the time of writing.

CONCLUSIONS

The management of LC is complex due to the heterogeneity of presentations, multi-organ involvement, and the lack of robust evidence in the area. Existing guidelines propose holistic approaches that address the biopsychosocial needs of the patient. SingStrong for LC is an intervention focussed on addressing disordered breathing and associated issues through a combination of relaxation techniques, breathing retraining, and singing. Participants in this 10-week pilot program have experienced significant improvements in numerous LC issues including breathlessness, fatigue, pain, and communication and provided positive feedback on the programme itself. As evidence for robust management strategies for LC emerge, there is merit in holistic programmes such as SingStrong to enhance patient biopsychosocial well-being. Heterogeneity of the cohort in terms of LC symptoms should be considered by singing leaders and clinicians in structuring participant groups. Further high-quality randomised controlled trials are required to further evaluate the efficacy or otherwise of this program.

DISCLOSURES

Contributors

All authors contributed to the conception or design of the work, the acquisition, analysis, or interpretation of the data. All authors were involved in drafting and commenting on the paper and have approved the final version.

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Competing interests

All authors declare no conflict of interest.

Ethical approval

Ethical approval for this project was provided by the ethics committee of the relevant faculty in the local University (2019_04_06_EHS).

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