Early View

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**Self-help cognitive behavioural therapy for anxiety in pulmonary hypertension: pilot
randomised controlled trial**

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Abstract

Objective: People with pulmonary hypertension (PH) are at an increased risk of experiencing anxiety disorders. This study developed and tested the acceptability, feasibility and preliminary effectiveness of a cognitive behavioural self-help intervention for anxiety in adults with PH using a pilot randomised control trial design.

Methods: Individuals with PH recruited from Pulmonary Hypertension Associations were randomised to either receive a newly developed self-help intervention (n=37) or a wait-list condition (n=40). Acceptability was explored using mixed-methods questionnaires. A 2x3 repeated-measures analysis of variance was used to explore anxiety (GAD-7), depression (PHQ-9), health-related quality of life (emPHAsis-10), dyspnoea (D-12), self-mastery (Self-Mastery Scale) and mood-related cognitions and behaviours (CBP-Q) at baseline, post-intervention and one-month follow up. A mediation analysis was performed to examine potential mechanisms of change.

Results: Dropout was low at 15.6%. All participants would recommend the intervention to another person with PH and felt it had helped with their anxiety. Participants in the intervention group reported a reduction in anxiety, depression and cognitive and behavioural processes linked with mood disorders, compared to the control group. Change in unhelpful cognitions and behaviours mediated the relationship between intervention condition, and change in anxiety and depression.

Discussion: The intervention was found to be acceptable, feasible and safe. Anxiety at baseline was higher than previously observed, which could be associated with the additional burden caused by COVID-19 as the intervention was trialled during the pandemic. Findings add to the growing evidence supporting the use of psychological therapies, including cognitive behavioural therapy, in this clinical group.

Take home message

- Mood disorders are highly prevalent in people with pulmonary hypertension (PH), although there is limited research examining effective treatments. We developed and piloted a self-help intervention targeting anxiety specifically in PH.

Key words

CBT; Self-Management; Pulmonary Arterial Hypertension; Mental Health;

Psychological therapy

Introduction

Anxiety and depression disorders are highly prevalent in people with pulmonary hypertension (PH) with rates of up to 50% [1, 2]. Such figures may also be underestimated as mental health conditions may be underrecognised in this group [2-4], and a recent UK survey found 85% of people with PH (n=824) reported greater symptoms of anxiety associated with the current COVID-19 pandemic [5]. Indeed, anxiety is a pertinent issue in PH due to its prevalence, the physiological effects on cardiovascular functioning which could exacerbate difficulties [6] and overlapping symptoms. Anxiety in PH is also associated with reduced HRQoL and depression [7, 8].

There is limited research examining treatments for anxiety in PH. In terms of psychological interventions, a systematic review found only three trials, all of which used Cognitive Behavioural Therapy (CBT) or strategies commonly associated with CBT, such as psychoeducation and relaxation techniques [1]. CBT informed interventions were found to be associated with significant improvements in anxiety, depression and HRQoL in PH [1]. In the UK, treatment guidelines recommend the use of CBT for anxiety and panic disorder in the general population, delivered within a stepped care model - first providing CBT self-management interventions [9]. There is growing evidence suggesting self-help interventions offer an accessible and effective form of treatment, including condition-specific interventions, which have the benefit of addressing specific challenges within clinical groups [10].

In collaboration with Pulmonary Hypertension Association UK, we developed a CBT informed self-help intervention for anxiety in adults with PH. In the current study, we conducted a pilot randomised controlled trial investigating the intervention against a wait-list control condition. The aim was to explore the feasibility and acceptability of the intervention, and preliminary effectiveness to help inform a sample size calculation for a

definitive trial. Furthermore, potential active mechanisms of change associated with the trial were explored using a mediation analysis.

Methods

Design

Participants were block randomised 1:1 to the two groups using an online randomiser (<https://www.random.org>). Participants were asked to complete a series of measures at baseline, post-intervention and one-month post-intervention. Ethical approval was obtained from the Department of Psychology Ethics Committee at the University of Sheffield (034442). The study was registered at clinicaltrials.gov: NCT04450862. Consort guidelines were used.

Eligibility Criteria

Participants were required to be aged 18 years or over; have a diagnosis of PH; provide informed consent; complete a series of self-report questionnaires independently in English; and self-report difficulties with anxiety. Participants could be living in the UK or outside of the UK (see results for the list of countries where participants were living). Individuals were not eligible if they experienced thoughts of self-harm or suicide.

Procedure

A convenience sampling method was used; participants were recruited via advertisements from Pulmonary Hypertension Associations. Participants were recruited between August- April 2021 Before being randomised, participants were first asked to complete a consent form, demographic and health-related questionnaires.

Our goal was to eventually make the intervention available to all members of PHA UK. Although this is a UK based charity, they are contacted by people outside of the UK with whom it is more practical to correspond electronically. Therefore, in accordance with how the intervention may be used in practice, participants in the UK were sent a paper version of the intervention, while non-UK participants received an electronic version.

Two weeks after, participants were contacted by telephone (UK participants) or email (non-UK participants), to assess adherence and acceptability, using a semi-structured questionnaire. Phone calls lasted on average 12.36 minutes (SD=12.25).

Participants randomised to the wait-list condition were informed that they would be contacted again in four-weeks.

After four weeks, participants were asked to complete the health-related questionnaires, which were repeated one-month later. Participants in the intervention group were then asked to complete a semi-structured acceptability questionnaire. Participants in the control group were informed if the intervention was acceptable, they would receive a copy in the future. Questionnaires were hosted by Qualtrics (<https://www.qualtrics.com>).

Intervention

The intervention consisted of a four-week programme corresponding to four individual booklets (Figure 1 & Table 1). A multi-disciplinary stakeholders' group was established comprising of adults living with PH and caregivers (n=4), clinical psychologists (n=2), PH nurse specialists (n=2), professionals from PHA UK (n=3), a cognitive behavioural therapist and graphic designer. The lead researcher developed the intervention guided by clinical experience and the evidence-base. For example, the intervention was guided by CBT-related theories on generalised anxiety disorder (GAD) [11], panic disorder [12] and health anxiety [13], which have all previously been documented as prevalent in this group [1, 14].

The lead researcher kept in regular contact with members of the stakeholders' group through a series of meetings acting on suggestions until the intervention was finalised.

Table 1. Outline of the intervention (also see Supplement 1)

Booklet	Contents	Exercises
1	Psychoeducation on CBT, anxiety and anxiety in PH.	Develop a mini-CBT formulation and practice slow breathing exercises.
2	Psychoeducation on maintenance factors for anxiety relevant to PH.	Develop a broader CBT formulation focusing on short and long-term difficulties, and practice mindfulness exercises.
3	Psychoeducation on cognitive biases and normalisation of difficulties through case vignettes.	Practice thought monitoring and thought challenging exercises.
4	Psychoeducation on graded exposure taking into consideration impact of fatigue. Information on other factors affecting anxiety and relapse prevention	Develop an anxiety hierarchy, create a plan to manage setbacks and progressive muscle relaxation exercise.

CBT = Cognitive Behavioural Therapy; PH = Pulmonary Hypertension

Measures

Participants provided demographics and information concerning their diagnosis of PH, PH functional class, duration of PH and whether they had received treatment for anxiety in the last twelve-months.

Anxiety, as the primary outcome measure, was measured using the GAD-7 questionnaire [15]. A score of 0–4 indicates “*minimal difficulties*”, 5–9 “*mild*”, 10–14 “*moderate*” and 15–21 “*severe*” anxiety. The clinical cut-off score is ≥ 8 with a reliable change of ≥ 4 [16]. Depression was measured using the PHQ-9 [17]. A score of 0–4 indicates “*minimal difficulties*”, 5–9 “*mild*”, 10–14 “*moderate*”, 15–19 “*moderately severe*” and ≥ 20 “*severe*” depression. The clinical cut-off score is ≥ 10 , with a reliable change of ≥ 6 [16]. HRQoL was examined using the emPHAsis-10 [18]. Scores range from 0–50, with a higher

score indicative of lower HRQoL. Breathing difficulties were explored using the Dyspnoea 12 (D12) [19]. Total scores range from 0-36 with a higher score indicating greater difficulty. Given the relationship between coping and anxiety in PH [8], coping was measured using the Self-Mastery Scale [20]. Total scores range from 7-49 with a higher score suggesting a greater perceived capacity to cope. To measure unhelpful cognitions and behaviours that may underlie anxiety, the Cognitive and Behavioural Processes Questionnaire (CBP-Q) was used. Total scores range from 0-120. Higher scores are associated with more unhelpful responses, which have been found to positively relate to anxiety and depression [21].

Two mixed-methods questionnaires were developed to assess adherence and acceptability part-way through and at the end of the study. Participants were asked about their experiences of anxiety in PH, the different aspects of the intervention and their engagement in the research trial.

Sample Size

In accordance with Whitehead et al's., [22] sample size guidelines for a pilot trial, a sample size of 36 in each arm was required using 90% power and 5% significance level. This was based a standardised effect size of 0.53 (Hedges' g), and average dropout rate of 16.5% reported in a meta-analysis of 54 self-help interventions for anxiety [23].

Data Analysis

A series of independent samples t -tests and Pearson's chi-square tests were performed to investigate group differences between CBT vs. control; UK vs. non-UK participants, and completers vs. non-completers.

A series of two-way mixed analysis of variance (ANOVA) were ran for each health-related measure examining the interaction, with condition representing the between subjects factor and time the within subjects factor. Where the interaction was significant, a series of independent samples *t*-tests were performed to compare differences at each time point between the two groups. A series of repeated measures ANOVAs were then used to investigate effects of time for each condition for all outcome measures corrected using Bonferroni. The alpha level (0.05) was accepted for all other tests. G*Power-3 was used to perform a post-hoc sample size calculation. Partial Eta Squared was converted to Cohen's *f* ($f=0.1$ small, 0.25 medium and 0.4 large) [24].

A series of mediated regression analyses were performed to investigate whether the effects of condition on significant changes in health-outcomes were mediated by change in CBP-Q scores. Change in scores was calculated by subtracting scores at baseline from one-month follow-up. A Sobel test was performed and Cohen's benchmarks were used to interpret effect sizes ($r^2=0.02$ small, 0.15 medium and 0.35 large) [24].

SPSS25 was used for all statistical analysis with Hayes' process macro extension for the mediation analyses [25].

Qualitative data collected from the adherence and acceptability questionnaires were analysed using quantitative content analysis [26]. A coding scheme was developed by the authors corresponding to the questions asked, which was used to code the qualitative data before quantifying participants' responses.

Results

Recruitment

Overall, n=138 individuals accessed the link to read the participant information sheet, of whom n=77 (55.8%) completed the baseline measures (Figure 2).

Randomisation

No significant differences in demographic or health-related outcomes were observed between participants in the self-help or control group (Table 2).

Table 2. Baseline data for participants randomised to the CBT self-help or control group. Means (M) and (standard deviations) unless otherwise stated. Results are from independent samples *t*-tests and Pearson's chi-square tests.

Characteristics	CBT self-help (n)	Control (n)	<i>p</i> value
Number of participants	37	n=40	0.73
Demographics			
Age (years)	M=48.2 (14.03)	M=47.5 (13.1)	0.83
Gender			0.4
Male	1	3	
Female	35	37	
Other	1	0	
Ethnicity			0.57
White	25	10	
Not reported or not clear	12	12	
Asian	4	5	
Hispanic	0	1	
Black	0	1	
Latina	1	1	
Education (years)	M=15.3 (4.7)	M=15.6 (3.8)	0.77
Employment			0.73
Employed	14	13	
Not employed	8	14	
Retired	9	9	
Student	2	1	
Other	4	3	
PH factors			
PH Type			0.83
Idiopathic PH	19	16	
Chronic thromboembolic PH	7	7	
Connective tissue disease	2	4	
Congenital PH	2	2	
Familial PH	0	1	
Other	3	6	
Not sure	4	4	
PH Class			0.27
I	6	2	
II	7	10	
III	12	12	
IV	2	0	
Not sure	10	13	
Years since diagnosis	M=9.48 (10.02)	M=7.84 (7.6)	0.42
Psychological factors			
Prescribed medication for anxiety	9 (24.3%)	13 (32.5%)	0.43
Received therapy for anxiety	7 (18.9%)	9 (22.5%)	0.7
Anxiety	M=10.43 (5.11)	M=11.75 (5.78)	0.29
Depression	M=12.38 (5.12)	M=12.7 (6)	0.8
HRQoL	M=38.49 (10.57)	M=41.28 (11.39)	0.27

Dyspnoea	M=14.38 (9.91)	M=16.55 (8.96)	0.32
Self-mastery	M=25.95 (7.08)	M=24.33 (7.75)	0.34
CBP-Q	M=69.59 (13.43)	M=68.55 (19.67)	0.79

n = Number of participants; M = Mean; PH = Pulmonary Hypertension; HRQoL = Health-Related Quality of Life; CBP-Q = Cognitive and Behavioural Processes Questionnaire

UK vs. non-UK Participants

Non-UK participants lived in Canada (n=17), India (n=7), United States (n=5), the Netherlands (n=1) and Australia (n=1). No significant differences were observed between the two groups with the exception of ethnicity, as a greater number of UK participants self-identified as white. Given the ethnic composition of countries from where individuals were recruited (e.g., UK vs. India), this may be expected (Supplement 2).

Completers vs. Non-Completers

A dropout rate of 15.6% was observed (self-help=18.9%, control=12.5%). Non-completers were significantly younger, reported lower levels of anxiety and scored higher on self-mastery ($p<0.05$). There was a greater ratio of men who completed the study; however, males overall were greatly underrepresented accounting for 5% of the sample (Supplement 3).

Acceptability

Overall, 32/37 (86.5%) of participants in the intervention condition completed the Check-in Questionnaire at two-weeks. The majority of participants were working on booklet one (n=7) or two (n=17). The majority (>70%, n=23/32) understood most of the content, felt it could help and did not find the intervention difficult or distressing (Supplement 4).

In total, 25/32 (78.1%) of participants completed the Final Acceptability Questionnaire. Ten participants rated the intervention as “*Excellent*”, fourteen “*Good*”, and

one “*Fair*”. Nearly half reported the intervention as “*Moderately*” helpful. All participants would recommend the intervention to another person with PH. Most (>68%, n=17/25) found the different booklets “*Extremely*” – “*Very Helpful*”. At the end of the study, >80% (n=20/25) felt more in control of their anxiety, supported during the intervention, valued that it was specific to PH and felt others would benefit from the resource (Supplement 4).

Analysis of qualitative feedback revealed participants wanted to take part due to difficulties with their anxiety, which for some was linked to COVID-19 (n=10). The majority reported the benefits of learning more about their anxiety (n=17) and developing strategies to manage (n=22). Participants recognised the importance of continuing to practice the strategies and valued the ability to refer back to the booklets at a later date. The largest group (n=9) felt there was “*nothing*” they did not like about the intervention. For others, it seemed external sources of motivation could have helped them to engage better (Supplement 5).

Effectiveness

Reliable change

Both groups scored above the clinical cut-off on the GAD-7 and PHQ-9 measures at baseline, scoring within the “*moderate*” range. While anxiety and depression symptomatology reduced below the clinical level and to the “*mild*” range following the self-help intervention and remained at this level at follow-up, those in the control group continued to report “*moderate*” and clinical levels throughout.

In the self-help group, 64.9% and 67.6% of participants scored above the clinical level on anxiety and depression, respectively, which by the end of treatment, reduced to 25% and 32.1%. Overall, 71.4% and 39.3% of participants reported a reliable change in anxiety and depression, respectively, at the end of the study, compared to baseline or post-intervention scores. In the control group, 44.1% of individuals reported a reliable change in

anxiety at one-month follow up, although nearly two-thirds remained above the clinical cut-off. Similarly, over half of the participants in the control group experienced clinical levels of depression at the end of the study (Supplement 6).

Statistical change

A series of two-way mixed ANOVAs revealed statistically significant interactions between group and time for anxiety, depression and CBP-Q (Table 3).

Table 3. Means and (Standard deviations) in the CBT (n=27) and control (n=30) group for health-outcomes and results of 2x3 mixed ANOVAs, Partial Eta Squared and Cohens' *f*

Measure	Group	Baseline	Post	One-month	Two-way mixed ANOVA (group x time)			
					F	<i>p</i>	η^2	Cohens' <i>f</i>
Anxiety	CBT	11 (5.03)	7.56 (3.93)*	5.89 (4.4)	F(2,110)= 4.23	0.017	0.07	0.28
	Control	11.9 (5.29)	11.03 (5.78)*	10.23 (5.62)				
Depression	CBT	12.41 (5.47)	8.11 (4.21)	7.48 (5.11)*	F(1.76,97.71) =3.26 HF	0.048	0.06	0.24
	Control	13.5 (5.4)	11.3 (5.47)	11.7 (7.1)*				
HRQoL	CBT	37.7 (11.16)	35.59 (10.69)	34.37 (10.14)	F(2,110)= 0.1	0.9	0.002	0.04
	Control	42.07 (11.27)	41.67 (11.06)	39.5 (11.81)				
Dyspnoea	CBT	14.93 (10.6)	14.85 (10.07)	12.63 (8.54)	F(1.85, 101.67) =0.77 HF	0.46	0.01	0.12
	Control	16.77 (8.74)	15.77 (8.92)	15.3 (9.29)				
Self-Mastery	CBT	25.22 (6.7)	26.41 (5.66)	27.63 (4.79)	^F(2,110) =0.24	0.79	0.004	0.06
	Control	24.2 (7.59)	25.63 (5.3)	25.67 (4.73)				
CBP-Q	CBT	69.56 (14.3)	57.59 (15.93)	50.26 (18.8)	^F(1.67,91.74) =8.55 HF	0.001	0.13	0.39
	Control	67.77 (17.88)	63.87 (16.47)	64.17 (22.28)				

CBT = Cognitive Behavioural Therapy; η^2 = Partial Eta Squared; HF = Huynh-Feldt; ^ = Test of Equality of Covariance significant; *f* = ANOVA effect size; ANOVA= Analysis of Variance; CBP-Q = Cognitive Behavioural Processes Questionnaire; *= Levene's Test of Equality not met ($p>0.05$) for mixed ANOVA

While no statistical difference in anxiety was observed at baseline between the two groups ($t(55)=-0.66, p=0.51$), there was a statistical difference at the post-intervention ($t(51.31)=-2.68, p=0.01$) and follow-up stage ($t(55)=-3.33, p=0.002$), with those in the CBT group reporting lower scores. Similarly, for depression, differences at baseline was non-significant ($t(55)=-0.76, p=0.45$), but was at post-intervention ($t(55)=-2.45, p=0.018$) and follow-up ($t(52.56)=-2.55, p=0.01$) in the same direction. Finally, for the CBP-Q, differences were non-significant at baseline ($t(55)=0.41, p=0.68$) and post-intervention ($t(55)=-1.46, p=0.15$), however, at the one one-month stage there was a significant difference ($t(55)=-2.53, p=0.014$), with the controls reporting a greater number of unhelpful thoughts and behaviours.

A series of repeated measures ANOVAs revealed a statistically significant effect of time on anxiety, depression and CBP-Q in the CBT group, indicating reductions across time. A significant effect was also observed for depression in the control group; however, post-hoc tests revealed this reduction was only temporary as there was no significant difference between baseline and one-month follow-up (Table 4).

Table 4. Mean differences in the CBT (n=27) and control (n=30) group for health-outcomes and results of repeated measures ANOVAs and pairwise comparisons corrected using Bonferroni

Measure	Group	Repeated measures ANOVA				Pairwise comparison		
		F	<i>p</i>	η^2	<i>Cohens' f</i>	Baseline vs Post	Baseline vs. One-month	Post vs. One-month
Anxiety	CBT	F(2,52)=18.54	<0.001	0.42	0.84	-3.44 (<i>p</i> =0.003)	-5.11 (<i>p</i> <0.001)	-1.67 (<i>p</i> =0.045)
	Control	F(2,58)=1.84	0.17	0.06	0.25	-0.87 (<i>p</i> =0.92)	-1.67 (<i>p</i> =0.29)	-0.8 (<i>p</i> =1)
Depression	CBT	F(1.63,42.25)=13.02 HF	<0.001	0.33	0.71	-4.3 (<i>p</i> =0.002)	-4.93 (<i>p</i> =0.002)	-0.63 (<i>p</i> =1)
	Control	F(2,58)= 5.43	0.007	0.16	0.43	-2.2 (<i>p</i> =0.004)	-1.8 (<i>p</i> =0.11)	0.4 (<i>p</i> =1)
HRQoL	CBT	F(2,52)=2.54	0.09	0.09	0.31	-1.11 (<i>p</i> =1)	-3.33 (<i>p</i> =0.2)	-2.22 (<i>p</i> =0.34)
	Control	F(2,58)=2.85	0.07	0.09	0.31	-0.4 (<i>p</i> =1)	-2.57 (<i>p</i> =0.15)	-2.17 (<i>p</i> =0.16)
Dyspnoea	CBT	F(2,52)=2.45	0.1	0.09	0.31	-0.07 (<i>p</i> =1)	-2.3 (<i>p</i> =0.34)	-2.22 (<i>p</i> =0.13)
	Control	F(2,58)=1.64	0.2	0.05	0.24	-1 (<i>p</i> =0.67)	-1.47 (<i>p</i> =0.39)	-0.47 (<i>p</i> =1)
Self-mastery	CBT	F(2,52)=1.25	0.3	0.05	0.22	1.19 (<i>p</i> =1)	2.41 (<i>p</i> =0.54)	1.22 (<i>p</i> =1)
	Control	F(2,58)=0.3	0.29	0.04	0.21	1.43 (<i>p</i> =0.79)	1.47 (<i>p</i> =0.38)	0.03 (<i>p</i> =1)
CBP-Q	CBT	F(1.63,42.3)= 18.38 HF	<0.001	0.41	0.83	-11.9 (<i>p</i> =0.001)	-19.3 (<i>p</i> <0.001)	-7.33 (<i>p</i> =0.03)
	Control	F(2,58)=2.04	0.14	0.07	0.27	-3.9 (<i>p</i> =0.15)	-3.6 (<i>p</i> =0.52)	0.3 (<i>p</i> =1)

CBT = Cognitive Behavioural Therapy; η^2 = Partial Eta Squared; HF = Huynh-Feldt; ANOVA= Analysis of Variance; CBP-Q = Cognitive Behavioural Processes Questionnaire

Sample Size Analysis

A sample size analysis was calculated using an observed effect size of $f=0.28$ for the primary outcome measure. Accepting 0.05 as the alpha level, a level of statistical power of 80%, two groups (self-help and wait-list) and three measurements (baseline, post-intervention and one-month) and sphericity not violated, a final sample size of $n=68$ is required. Taking into consideration a dropout rate of 15.6%, 80 participants are required for a definitive trial.

Mechanisms of Change

Intervention group was significantly related to change in anxiety scores ($b=3.62(\text{standard error}(\text{se}))=1.35$), $t(59)=2.69$, $p=0.009$) and change in CBP-Q at one-month ($b=14.86(\text{se}=4.4)$, $t(59)=3.37$, $p=0.001$). Change in CBP-Q scores significantly predicted change in anxiety ($b=0.13(\text{se}=0.04)$, $t(58)=3.52$, $p<0.001$). Controlling for change in CBP-Q as a mediator, condition was not a significant predictor of change in anxiety ($b=1.72(\text{se}=1.35)$, $t(58)=1.27$, $p=0.21$). Overall, change in CBP-Q was a significant mediator of change in anxiety (effect=1.91(se=0.82), 95% CI=0.56 – 3.67), which was confirmed by a Sobel test ($Z=2.34(\text{se}=0.82)$, $p=0.02$).

For depression, intervention group significantly predicted change in depression scores ($b=3.31(\text{se}=1.38)$, $t(59)=2.4$, $p=0.02$) and change in CBP-Q ($b=14.86(\text{se}=4.4)$, $t(59)=3.37$, $p=0.001$). Change in CBP-Q was a significant predictor of change in depression ($b=0.19(\text{se}=0.03)$, $t(58)=5.87$, $p<0.001$). When change in CBP-Q was controlled, condition was not a significant predictor of change in depression ($b=0.46(\text{se}=1.2)$, $t(58)=0.38$, $p=0.7$). As in anxiety, change in CBP-Q was a significant mediator of change in depression (effect=2.84(se=1.23), 95% CI=0.78 – 5.6), which was further assessed using a Sobel test ($Z=2.98(\text{se}=0.95)$, $p=0.003$).

Discussion

Findings suggest that the intervention was feasible, acceptable and safe, as no adverse events were reported. The majority of participants reported the intervention as “*Excellent*” or “*Good*” and would recommend it to someone with PH. An overall attrition rate of 15.6% was observed, which is comparable to rates reported by a meta-analysis of 54 self-help interventions for anxiety (16.5%) [23], as well as a meta-analysis of 45 studies investigating face-to-face psychological therapies for adults with GAD (16.99%) [27].

Participants who completed the study were significantly older, scored higher on self-mastery and reported greater anxiety. This finding is consistent with research suggesting younger adults are more likely to drop out of psychological therapies [28]. It is plausible that people who were less affected by anxiety had lower motivation to engage. However, findings suggest that the intervention was helpful in reducing symptoms of anxiety in those with mild to severe symptoms, and therefore supporting patients with all levels of anxiety to engage with psychological care may be beneficial. Similarly, while it has been suggested that anxiety is greater in those recently diagnosed with PH [1], as participants had been living with PH for an average of seven-nine years, interventions targeting anxiety may be helpful to people across the clinical group. However, our current ability to explore anxiety in people with PH is likely to be impaired by the lack of standardised screening assessments. Research investigating the suitability, reliability and adaptation of non-PH and generic health-related questionnaires in this population is needed.

At baseline, 70.1% (n=54/77) and 64.9% (n=50/77) of participants scored above the clinical level on anxiety and depression, respectively, which is greater than previous studies [1]. Given that participants here had to be experiencing difficulties with anxiety, it is reasonable to suggest that rates are higher compared to the wider PH population.

Nevertheless, it could also reflect the impact of the COVID-19 pandemic [29], as highlighted

by ten participants who discussed the pandemic as a reason for their participation. While participants found the intervention was helpful in managing their anxiety during the pandemic, it is not known whether this was translated to COVID-19-related anxieties. Further research is required examining the psychosocial effects of the pandemic in people with PH and how best to support this group.

Participants in the CBT group reported a significant reduction in anxiety, depression and unhelpful cognitions and behaviours over time, compared to participants in the control group. As such, the results further justify a large-scale and definitive RCT of the current intervention. Within-group differences revealed a large and significant reduction in the CBT group in symptoms of anxiety and depression, with participants scoring below the clinical level at the post-intervention stage with gains maintained, and significantly improved upon for anxiety, at follow-up. Taken together, the findings support the evidence concerning the benefits of psychological therapy, specifically CBT, in this population. Participants in the control group also experienced an improvement in depression at the post-intervention stage. While it is not unusual for control participants to report therapeutic gains [30]; the majority of participants remained above the clinical level and gains were only temporary.

Results from the mediation analyses indicate that the intervention had an indirect effect on improvement in mood outcomes, through changing cognitions and behaviours. Further research is required to examine the relationship between the cognitive and behavioural strategies that individuals use to cope, and health-related outcomes in PH. Identifying factors associated with positive health outcomes can help to recognise important therapeutic targets, and guide the development of services [3, 8].

Due to the modest sample size, differences in outcomes between participants receiving paper vs. electronic versions of the intervention were not examined. Therefore, it is not clear what impact (if any) this had on observed effect sizes, especially given the difference in effectiveness between the two approaches observed [23].

Notwithstanding the female preponderance reported in PH [31], men were considerably underrepresented in the current sample, which is consistent with previous evidence suggesting females with PH are more likely to take part in research [32]. Further investigation is required to explore such gender differences (including the likelihood of males with PH engaging with psychological therapies) and develop strategies to help overcome barriers for men.

Finally, it could be a limitation that a wait-list control group was used as this could have inflated study effects than if an active treatment was used. However, this approach was utilised as it reflects the current support typically available to patients as information on anxiety in PH is limited.

Conclusion

We developed a self-help intervention for anxiety in PH, which was found to be acceptable, feasible and safe. Attrition was comparable to other psychological therapies, including face-to-face treatments. While only preliminary, participants in the self-help condition reported a significant and reliable reduction in symptoms of anxiety and depression, which were fully mediated by a change in cognitions and behaviours related to the intervention. Results add to the growing evidence supporting psychological therapies in people with PH. Self-help intervention offer an effective and accessible form of therapy that can be used as a stand-alone intervention or to supplement other treatments.

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Figure 1. Extract from booklet one showing the four booklets and providing participants with instructions

Figure 2. *CONSORT flow diagram*

Supplement 1

Outline of the intervention

Booklet	Content	Exercises
1 'Anxiety and PH'	<ul style="list-style-type: none">• Introduction to CBT model• Introduction to the intervention• Normalise difficulties with anxiety through case studies• Psychoeducation on psychosocial impact of PH, anxiety (panic, fear, stress and worry) and anxiety in PH• Highlight the importance of coping strategies	<ul style="list-style-type: none">• Explore and identify common symptoms of anxiety• Complete the four-areas model – develop a mini-formulation of difficulties• Slow breathing exercise
2 'What keeps anxiety going'	<ul style="list-style-type: none">• Psychoeducation on anxiety – focusing on avoidance, reassurance seeking, safety behaviours, symptoms and unhelpful thoughts about health – all specific to experiences of PH	<ul style="list-style-type: none">• Explore and recognise maintenance cycles• Develop a here and now formulation of difficulties recognising triggers, reactions, unhelpful coping strategies and short- and long-term consequences• Two mindfulness exercises – focusing on bodily symptoms and everyday events
3 'The power of thoughts'	<ul style="list-style-type: none">• Psychoeducation on cognitive biases grounded in the four-factor model: predicting the future, mind reading, over-generalisation, all-or-nothing thinking and self-criticism• Challenging unhelpful thoughts – evidence for and against• Normalise difficulties with anxiety through case studies• Develop techniques to help manage worry	<ul style="list-style-type: none">• Recognise unhelpful thinking biases• Thought challenging• Worry tree• Worry delay• Thought monitoring exercise
4	<ul style="list-style-type: none">• Psychoeducation on graded exposure to manage anxiety; pacing, and boom and bust cycle to manage pain and fatigue	<ul style="list-style-type: none">• Development of anxiety hierarchy• Development of pacing hierarchy

‘The power
of your
actions’

- Psychoeducation on other factors affecting anxiety: routine, diet, sleep, social support, keeping physically active and confidence/assurance
- Relapse prevention reviewing what participants have learnt, skills they have developed, and identifying and managing setbacks
- Signpost to other services for mental health and PH-related problems

- Create a plan to cope with setbacks
 - Progressive muscle relaxation exercise
-

CBT = Cognitive Behavioural Therapy; PH = Pulmonary Hypertension

Supplement 2

Baseline data for participants living in the UK or outside of the UK (international). Means and (standard deviations) unless otherwise stated

Characteristics	UK	International	<i>p</i> value
Number of participants	n=46	n=31	0.09
Condition			0.38
CBT Self-help	n=24	n=13	0.07
Control	n=22	n=18	0.53
Demographics			
Age (years)	47.91 (12.59)	47.67 (14.91)	0.94
Gender			
Male	n=2	n=2	0.43
Female	n=44	n=28	
Other	n=0	n=1	
Ethnicity			0.004
White	n=29	n=16	
Not reported or not clear	n=15	n=4	
Asian	n=1	n=8	
Hispanic	n=0	n=1	
Black	n=1	n=0	
Latina	n=0	n=2	
Education (years)	14.54 (5.07)	16.42 (2.75)	0.07
Employment			0.07
Employed	n=16	n=11	
Not employed	n=14	n=8	
Retired	n=12	n=6	
Student	n=3	n=0	
Other	n=1	n=6	
PH factors			
Type			0.08
Idiopathic PH	n=17	n=18	
Chronic thromboembolic PH	n=12	n=2	
Connective tissue disease	n=5	n=1	
Congenital PH	n=1	n=3	
Familial PH	n=0	n=1	
Other	n=5	n=4	
Not sure	n=6	n=2	
Class			0.38
I	n=4	n=4	
II	n=8	n=9	
III	n=19	n=8	
IV	n=2	n=0	
Not sure	n=13	n=10	
Years since diagnosis	8.21 (8.55)	9.25 (9.33)	0.62
Psychological factors			
Prescribed medication for anxiety	n=12	n=10	0.56
Received therapy for anxiety	n=20	n=13	0.89
Anxiety	11.74 (5.23)	10.19 (5.78)	0.23

Depression	13.13 (5.32)	11.68 (5.88)	0.26
HRQoL	40.5 (11.15)	39.1 (10.95)	0.59
Dyspnoea	16.5 (9.79)	14.03 (8.81)	0.26
Self-mastery	24.52 (7.58)	25.97 (7.23)	0.4
CBP-Q	69.59 (16.51)	68.32 (17.4)	0.75

n = Number of participants; PH = Pulmonary Hypertension; HRQoL = Health-Related Quality of Life; CBP-Q = Cognitive and Behavioural Processes Questionnaire; UK = United Kingdom

Supplement 3

Baseline data for participants defined as completers vs. non-completers. Means and (standard deviations) unless otherwise stated

Characteristics	Completers	Non-completers	<i>p</i> value
Number of participants	n=65	n=12	<0.001
Condition			0.44
CBT Self-help	n=30	n=7	
Control	n=35	n=5	
Demographics			
Age (years)	49.27 (13.11)	40.08 (13.17)	0.03
Gender			0.046
Male	n=4	n=0	
Female	n=61	n=11	
Other	n=0	n=1	
Country			0.17
UK	n=41	n=5	
International	n=24	n=7	
Ethnicity			0.43
White	n=29	n=6	
Not reported or not clear	n=17	n=2	
Asian	n=6	n=3	
Hispanic	n=1	n=0	
Black	n=1	n=0	
Latina	n=1	n=1	
Education (years)	15.47 (4.23)	15.18 (4.26)	0.84
Employment			0.39
Employed	n=25	n=2	
Not employed	n=17	n=5	
Retired	n=16	n=2	
Student	n=2	n=1	
Other	n=5	n=2	
PH factors			
PH Type			0.4
Idiopathic PH	n=30	n=5	
Chronic thromboembolic PH	n=14	n=0	
Connective tissue disease	n=5	n=1	
Congenital PH	n=3	n=1	
Familial PH	n=1	n=0	
Other	n=7	n=2	
Not sure	n=5	n=3	
PH Class			0.86
I	n=7	n=1	
II	n=15	n=2	
III	n=23	n=4	
IV	n=2	n=0	
Not sure	n=18	n=5	
Years since diagnosis	7.93 (8.36)	12.42 (10.64)	0.11
Psychological factors			

Prescribed medication for anxiety	n=18	n=4	0.69
Received therapy for anxiety	n=12	n=2	0.24
Anxiety	11.75 (5.44)	7.67 (4.4)	0.02
Depression	2.95 (5.54)	10.33 (5.37)	0.13
HRQoL	40.63 (10.89)	36.17 (11.4)	0.2
Dyspnoea	15.92 (9.57)	13.25 (8.65)	0.37
Self-mastery	24.35 (7.4)	29.17 (6.38)	0.04
CBP-Q	69.95 (17.2)	63.9 (13.58)	0.27

n = Number of participants; PH = Pulmonary Hypertension; HRQoL = Health-Related Quality of Life; CBP-Q = Cognitive and Behavioural Processes Questionnaire; UK = United Kingdom

Supplement 4

Check-in Questionnaire (n=32); values represent n unless otherwise stated

Question	Great Deal – A Lot	Moderate Amount	None at All – A Little	Mean (SD)
How much have you looked at the intervention?	15 (46.9%)	13 (40.6%)	4 (12.5%)	3.6 (0.97)
How much of the intervention have you understood?	27 (84.4%)	3 (9.3%)	2 (6.3%)	4.35 (1.02)
How distressing is the intervention?	2 (6.3%)	5 (15.6%)	25 (78.1%)	1.55 (0.74)
How difficult is the intervention?	3 (9.4%)	6 (18.7%)	23 (71.9%)	1.97 (1.08)
How often have you been using the skills?	16 (50%)	10 (31.2%)	6 (18.8%)	3.29 (1.19)
How much is the intervention helping with your anxiety?	15 (46.9%)	10 (31.2%)	7 (21.9%)	3.23 (1.02)
How much can the intervention help with your anxiety?	30 (93.8%)	1 (3%)	1 (3%)	4.23 (0.68)
How much is the intervention helping in other areas of your life?	10 (31.3%)	14 (43.7%)	8 (25%)	2.97 (0.95)
How much can the intervention help in other areas of your life?	26 (81.3%)	6 (18.7%)	0 (0%)	4.06 (0.68)

SD = Standard Deviation

Final Acceptability Questionnaire post-intervention (n=25). Values represent n (%) unless otherwise stated

Question	Great Deal – A Lot	Moderate Amount	A Little	Mean (SD)
How much did the intervention help you with your anxiety?	9 (36%)	12 (48%)	4 (16%)	3.3 (0.84)
How much did the intervention help in other areas of your life?	10 (40%)	11 (44%)	4 (16%)	3.3 (0.85)
How likely are you to recommend the intervention?	21 (84%)	3 (12%)	1 (4%)	4.3 (0.84)

No participants reported “Not at All Helpful”

Results from the Final Acceptability Questionnaire continued.

Item	Extremely - Very Helpful	Somewhat Helpful	Slightly Unhelpful	Mean (SD)
Booklet 1 (Anxiety and PH)	18 (72%)	6 (24%)	1 (4%)	3.8 (0.75)
Booklet 2 & 3 (What keeps anxiety going and The power of thoughts)	23 (92%)	2 (8%)	0 (0%)	4.1 (0.49)
Booklet 4 (The power of your actions)	17 (68%)	8 (32%)	0 (0%)	3.9 (0.74)
Booklet 4 – (Other factors affecting anxiety)	18 (72%)	7 (28%)	0 (0%)	3.9 (0.7)
Homework tasks	19 (76%)	6 (24%)	0 (0%)	3.9 (0.65)
No participant reported “Not at All Helpful”				

Results from the final acceptability questionnaire continued.

Question	Strongly- Somewhat Agree	Neither Agree nor Disagree	Strongly – Somewhat Disagree	Mean (SD)
I feel more in control of my anxiety	22 (88%)	3 (12%)	0 (0%)	4.1 (0.6)
People diagnosed with PH would benefit from the intervention	25 (100%)	0 (0%)	0 (0%)	4.8 (0.37)
Family and friends would benefit from this intervention	23 (92%)	2 (8%)	0 (0%)	4.4 (0.65)
I valued the intervention was specific to PH	24 (96%)	0 (0%)	1 (4%)	4.7 (0.85)
I valued being contacted part-way through	24 (96%)	1 (4%)	0 (0%)	4.6 (0.58)
The level of support I received was sufficient`	24 (96%)	1 (4%)	0 (0%)	4.6 (0.58)
I was given enough time to work through the intervention	24 (96%)	1 (4%)	0 (0%)	4.3 (0.95)
Health outcomes were relevant to my difficulties	20 (80%)	3 (12%)	2 (8%)	4 (1)

PH = Pulmonary Hypertension

Supplement 5

Results from the quantitative content analysis investigating acceptability (n=32)

Categories	Responses	Examples	n
Motivation to take part	Affected by anxiety and/or mental health	<i>"I have struggled with anxiety for over 15 years but have had times where it has been worse since my PH diagnosis"</i> (P31)	15
	Learn how to better understand and manage anxiety	<i>"To learn how to help manage my own anxiety and stress & I was so amazed that the help was specified towards PH."</i> (P67)	14
	COVID, lockdown or shielding made anxiety worse	<i>"Due to the COVID-19 situation my mental health was getting worse."</i> (P43)	10
	To help other people through participating in a trial	<i>"I wanted to help other sufferers"</i> (P22)	4
	Like to participate in trials for PH	<i>"I want to take part in any research that can help pulmonary hypertension patients"</i> (P20)	2
	Better understand PH	<i>"To better understand pulmonary hypertension as I was only diagnosed this year... it was very scary getting told that and that I have blood clots in lungs"</i> (P28)	1
Result of taking part	Developed coping strategies to manage and feel more in control of anxiety	<i>"Thank you for helping to give tools to help people dealing with PH and many other challenges!!!!!"</i> (P67)	22
	Improved how I understand anxiety	<i>"It also made me aware of things that I do when feeling anxious that I wasn't even aware of."</i> (P17)	17
	Normalised my experiences	<i>"...the true-life scenarios were very helpful as you do tend to feel that sometimes it must be only you that feels like this"</i> (P33)	10
	Made me confront my difficulties	<i>"It helped me look at my anxiety in a new way and helped me to confront it."</i> (P20)	5
	Induced my anxiety in the short-term	<i>"The women in the study makes me quite worried but I feel it is useful, but not a criticism"</i> (P2)	3
	Helped in other areas, such as self esteem	<i>"Improved my self-esteem and motivation."</i> (P18)	2

How will you continue to use what you learnt in the study?	Keep practicing and using helpful coping strategies	<i>"Day by day putting in the work"</i> (P62)	21
	Going back to the booklets to recap	<i>"I plan to keep going over the materials, and I hope to form habits that will help with anxiety and stress."</i> (P67)	16
	Share the booklets and my anxiety difficulties with others	<i>"I plan to continue using the breathing exercises and plan on letting my partner read my responses in the booklets to better understand what worries me when it comes to my health."</i> (P20)	1
Barriers (what did you like the least?)	None	<i>"I found it all useful."</i> (P14)	9
	Struggled to stay motivated, keep focused or keep on track	<i>"Nothing really, the only thing maybe would be a reminder to ensure you know what week you're on. For example a reminder text"</i> (P52)	5
	Takes time to remember all the recommendations	<i>"It's difficult making them a habit when you have years of over thinking."</i> (P80)	4
	Questionnaires were not always relevant or forgetting to do them	<i>"I probably found the questionnaires quite difficult to answer as the choice of boxes to tick did not really relate..."</i> (P17)	4
	Being asked to face to my fears	<i>"Having to confront the situations that made me feel anxious and worried"</i> (P26)	3
	More information about the symptoms and more resources	<i>"Maybe more info could be good? That said I found what it contained specifically helpful"</i> (P74)	3
	Some of my difficulties were not represented	<i>"In particular, the booklets didn't help me to work out how to help my family"</i> (P24)	2
	Doing it by yourself	<i>"It's good that we do this on our own, but it would've been nice to interact with someone."</i> (P67)	2
	Case studies were anxiety provoking	<i>"The case studies with other people who have PH"</i> (P20)	1
	Font was very small – electronic version	<i>"I was only able to read the booklets online because the font in the printed version was too small and almost impossible to read."</i> (P77)	1
	Needed longer than one-week	<i>"I think a two-week interval between starting the other booklets would have been more helpful."</i> (P18)	1

Facilitators (What did you like the best?)	The information was user friendly and informative	<i>"It was straightforward and easy to work through."</i> (P14)	22
	Booklets helped promote self-evaluation	<i>"It made me think about my situation and how I manage my reactions."</i> (P22)	8
	You can work at your own pace	<i>"Letting you work through one stage at a time and not being bombarded with information."</i> (P38)	7
	It was specific to PH	<i>"I was very happy that the material was geared to people with pulmonary hypertension because it is rare to hear of information specific to PH and even more rare to have help with the issues we face."</i> (P67)	6
	The promotion of coping strategies	<i>"Gaining skills and techniques from the booklets. Better understanding why I react the way I do in some situations and being able to cope with it."</i> (P43)	5
	Homework tasks	<i>"I like the practical strategies that are suggested and I have found them useful."</i> (P17)	4
	Case studies	<i>"The testimonies, to know others feel the same and have similar experiences."</i> (P34)	4
	I feel less alone	<i>"Felt less alone and isolated when dealing with anxiety."</i> (P54)	3
	It was effective	<i>"It was simple and effective and support was given."</i> (P52)	1
	Support from the researcher	<i>"Having a call during the intervention was helpful to discuss matters."</i> (P52)	1
	I felt empowered to help myself	<i>"The self-help exercises. Found it quite empowering to feel that I was able to help myself cope and manage my anxiety."</i> (P18)	1
	Not having to wait for treatment	<i>"I think it is a very worthwhile study, as mental health services are so underfunded you can wait ages to access any support."</i> (P38)	1
	Relevant to everyday life	<i>"Interventions were realistic."</i> (P80)	1

P = Participant number; PH = Pulmonary Hypertension

Supplement 6

Anxiety and depression scores for participants in the CBT (n=37) and control group (n=40), values = n (%)

Group	Measure	Baseline	Post-intervention	One-month follow up
CBT	Anxiety	0-4 Minimal	4 (10.8%)	15 (53.5%)
		5-9 Mild	16 (43.2%)	6 (21.4%)
		10-14 Moderate	7 (18.9%)	6 (21.4%)
		15-21 Severe	10 (27%)	1 (3.6%)
		Above cut off (≥ 8)	24 (64.9%)	7 (25%)
		Reliable change (≥ 4)	-	20 (71.4%)
	Depression	0-4 Minimal	3 (8.1%)	11 (39.3%)
		5-9 Mild	9 (24.4%)	8 (28.6%)
		10-14 Moderate	8 (21.6%)	7 (25%)
		15-19 Moderately Severe	15 (40.5%)	1 (3.6%)
		20-27 Severe	2 (5.4%)	1 (3.6%)
		Above cut off (≥ 10)	25 (67.6%)	9 (32.1%)
		Reliable change (≥ 6)	-	13 (44.8%)
	Anxiety	0-4 Minimal	3 (7.5%)	4 (12.9%)
		5-9 Mild	13 (32.5%)	10 (32.3%)
		10-14 Moderate	11 (27.5%)	7 (22.6%)
		15-21 Severe	13 (32.5%)	10 (32.3%)
		Above cut off (≥ 8)	30 (75%)	18 (58.1%)
		Reliable change (≥ 4)	-	9 (29%)
Control	Depression	0-4 Minimal	3 (7.5%)	4 (12.9%)
		5-9 Mild	12 (30%)	9 (29%)
		10-14 Moderate	6 (15%)	8 (25.8%)
		15-19 Moderately Severe	15 (37.5%)	7 (20.5%)
		20-27 Severe	4 (10%)	1 (3.2%)
		Above cut off (≥ 10)	25 (62.5%)	18 (58.1%)
		Reliable change (≥ 6)	-	7 (22.6%)

CBT = Cognitive Behavioural Therapy