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Coping styles associated with depression, health anxiety and health-related quality of life in pulmonary hypertension: Cross-sectional analysis

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Coping styles associated with depression, health anxiety and health-related quality of life in pulmonary hypertension: Cross-sectional analysis

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Figure 1 - Scree plot prior to final four factor analysis of coping in pulmonary hypertension

1

Abstract

Objectives: Pulmonary hypertension (PH) is a life-shortening disease associated with early mortality and high morbidity. With advancements in medical treatment, people are living longer with the disease and research is now needed to explore factors that help to enhance patient-reported outcomes. This study investigated the coping strategies of individuals with PH, and examined the relationship between coping, depression, health anxiety and health-related quality of life (HRQoL).

Design: A cross sectional survey design was used.

Participants: Participants (n=121) were recruited from membership of Pulmonary Hypertension Association (PHA) UK.

Outcome measures: Participants completed a series of questionnaires assessing depression (Patient Health Questionnaire-9), health anxiety (Short Health Anxiety Inventory), HRQoL (emPHasis-10) and coping (Brief COPE). An exploratory factor analysis was used to identify participants' coping profile. A series of correlational, linear and moderated multiple regression analyses were performed to examine the relationship between coping and health-related outcomes.

Results: Overall, 43% participants met criteria for potential clinical depression. Depression and health anxiety were strongly associated with HRQoL, explaining 37% and 30% of variance respectively (p<0.001). A principal component factor analysis identified a four-factor model of coping. Dimensions were named based on construct items: "cognitive coping" (7-items), "passive coping" (4-items), "external coping" (7-items) and "substance use coping" (2-items). Cognitive and external coping moderated the relationship between depression and HRQoL, with high use of these coping strategies reducing the impact of depression on HRQoL. External coping also moderated the effect of health anxiety on HRQoL.

Conclusions: The results uniquely highlight the importance of coping styles and psychological distress in predicting HRQoL in PH. Our findings indicate the importance to assess for psychological distress in this population and suggest the need to offer psychological interventions that take into account coping resources and strategies.

Strengths and limitations

- Understanding how patients cope with the demands of pulmonary hypertension (PH) can be used to help inform services and patient-reported outcomes.

- The results demonstrate high rates of distress in PH within a community sample, with 43% of people scoring above the clinical cut off for depression.

- Coping strategies were found to moderate the relationship between health anxiety and depression, and HRQoL in PH.

- Coping by seeking external support and cognitive-based strategies buffered the impact of psychological distress on HRQoL.

- Clinical factors, such as PH severity were not included as covariates and therefore, it is not clear how objective measures of the impact of PH influence coping.

Key words: Pulmonary arterial hypertension; adjustment; chronic; long-term; coping

Pulmonary hypertension (PH) is a progressive disease characterised by elevated pulmonary artery pressure, which ultimately leads to right heart failure and death [1]. PH is a global term used to describe five distinct groups of the disease: Group 1, pulmonary arterial hypertension (PAH); Group 2, PH due to left heart disease; Group 3, PH caused by lung disease; Group 4, chronic thromboembolic PH (CTEPH); and Group 5, PH due to miscellaneous causes [2]. Common symptoms of PH can include shortness of breath, fatigue, pain, weakness, dizziness, oedema and in some cases syncope [3].

Better disease awareness and a greater understanding of the aetiologies of PH have helped to advance diagnostic and therapeutic approaches, dramatically improving treatment outcomes, and resulting in lower mortality rates [4, 5]. As in other conditions in which people are living longer with a disease, research is now needed to examine the impact on daily life and identify factors associated with patient-outcomes such as quality of life, the evidence from which can be used to inform care and support [6].

The World Health Organisation (WHO) functional class can be used to help indicate the severity of patients' symptoms of PH. This index ranges from I, indicative of patients experiencing none to mild PH-related symptoms during physical activity, to IV suggesting patients will experience severe symptoms at rest [7]. However, the WHO classification system was not designed to reflect the psychosocial impact of living with PH. A qualitative synthesis of adults' lived accounts summarised the specific challenges posed by the disease, including the burden of living with disabling symptoms; intensive treatment regimens; impact on loved ones; stigma of PH and lack of understanding from others [8].

Coping has been viewed as cognitive, emotional and behavioural strategies employed in an attempt to manage distress perceived as threatening [9]. This response requires the individual to recognise and appraise the stressor, taking into consideration the expected outcome and their perceived ability to cope [10]. A large evidence base exists examining how

people cope with living with long-term health conditions [11-14]; however, no studies to date have quantitively investigated coping in people with PH. Understanding how patients cope with a disease can help to identify targets for intervention, and assist in identification of individuals most at risk of experiencing difficulties. This is particularly relevant in PH, as treatments aim to sustain health-related quality of life (HRQoL) and early intervention is viewed as essential in reducing morbidity [5].

The primary aim of this study was to explore the specific coping strategies used by people with PH. We used the Brief COPE measure [15], which explores 14 different coping strategies, including: self-distraction, denial, substance use, behavioural disengagement, venting, self-blame, active coping, use of emotional support, use of instrumental support, positive reframing, planning, acceptance, humour and religion. This tool is one of the most commonly used measures to assess coping in clinical populations [16]. While some studies have grouped coping strategies dichotomously (i.e. approach- and avoidance-focused coping strategies), there is no clear consensus on the underlying factors. In fact, numerous studies using this measure have applied factor analyses to examine the psychometric properties of higher-order factors, proposing 2-15 factor models of coping [17].

A secondary aim of the current study was to assess how coping strategies are related to depression, health anxiety and HRQoL, using a series of correlational and regression analyses. As observed elsewhere, the relationship between psychological factors and burden of a long-term health condition is likely to be influenced by the coping strategies an individual utilises [18]. Lower HRQoL [19], and depression and anxiety are commonly reported as highly prevalent in people with PH [19-22]. Although health anxiety has not specifically been examined in this condition, there is evidence to suggest individuals experience anxiety about their condition, which could further contribute to distress [8].

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Therefore, we hypothesised depression and health anxiety would be related to lower HRQoL, and this relationship would be moderated by specific coping strategies.

Methods

Participants

A cross-sectional design was employed, utilising a convenience sampling method. The article has been conducted in line with the STROBE checklist (supplement A) [23].

Bose et al., [24] examined the relationship between HRQoL and coping in individuals with chronic heart failure. Coping significantly predicted the physical (R^2=0.26, $p<0.001$) and mental (R^2=0.4, $p<0.001$) health composite of the QoL measure, equating to an observed effect size (f^2) of 0.35-0.67. A prior analysis using G*Power, with an effect size f^2 of 0.35, alpha level of 0.05, 80% power suggested a total sample size of at least 25 was required.

Participants were recruited over the span of two weeks from advertisements electronically sent to members of Pulmonary Hypertension Association (PHA) UK. To be eligible, participants must have been aged 18 years or over, living in the UK, diagnosed with PH, able to complete self-report questionnaires independently in English, and provide informed consent. Ethical assessment and approval were sought and obtained from the University of Sheffield (035318).

Procedure

Individuals interested in the study were instructed to access a website link directing them to the participant information sheet. If individuals still wished to take part after reading the information sheet, they were asked to complete a consent form and series of questionnaires (see below). All measures were hosted by Qualtrics.

Measures

Demographic questionnaire

This included name, age, gender, ethnicity, living status, employment status, level of education, PH diagnosis and functional class, and duration of PH.

Quality of life

The emPHasis-10 is a PH specific HRQoL measure [25]. Participants are asked to rate how ten problems related to life with PH have affected them "recently" on a unidimensional scale. Possible scores range from 0-50, with a higher score indicative of lower HRQoL. The measure has excellent internal reliability ($\alpha = 0.9$) [25]; Cronbach's alpha within the current study was $\alpha = 0.94$.

Depression

The Patient Health Questionnaire-9 (PHQ-9) [26] was used to measure symptoms of depression. This is a nine-item measure asking participants how often they have been bothered by a range of depression-related difficulties over the previous two weeks. Scores range from 0-27; a score of 0-4 indicating "no difficulties with depression", 5-9 "mild difficulties", 10-14 "moderate", 15-19 "moderately severe", and 20 or higher "severe depression". A cut off of ≥ 10 has been found to be a valid and reliable measure for major (clinical) depression [27]. This measure has previously been used in those with PH [28]. The PHQ-9 was found to have good internal consistency ($\alpha = 0.88$) in the current sample.

Health anxiety

The Short Health Anxiety Inventory (SHAI) [29] was used to measure participants' anxiety in response to how they appraised their health. This measure asks respondents to

endorse items assessing their thoughts, feelings and behaviours about health anxiety over the

past week on a scale ranging from 0–3 according to specific statements. While the SHAI is

typically an 18-item measure, it can be converted to 14-items when exploring health anxiety

in people with an existing medical disease. The 14-item measure has been found acceptable

in clinical samples, demonstrating high internal consistency (Cronbach's alpha 0.84) [30].

Internal consistency was excellent ($\alpha = 0.9$) in this sample.

Coping styles

The Brief COPE measure [15] was used to examine participants' coping. Individuals

were asked how often they engage in 28 behaviours when experiencing hardship in their life,

which are aggregated into 14 coping strategies (two items per each coping strategy). Possible

scores for each strategy ranged from 2-8, with a higher score indicating greater use. Cronbach

alpha's were all within the acceptable range or higher ($\alpha = 0.65 - 0.94$), with the exception of

denial ($\alpha = 0.58$).

Data analysis

HRQoL scores were normally distributed (Kolmogorov-Smirnov $p = 0.085$), therefore

parametric tests were used. Descriptive statistics were utilised to describe participants' socio-

demographic and clinical characteristics, and coping profile. No data was missing.

An exploratory factor analysis was performed using a principal components method.

A direct oblimin rotation was applied as there is a strong theoretical basis for the coping

strategies investigated here to be related [31]. The determinant of the correlation matrix was

1.379, suggesting this approach was suitable. Solutions were analysed using Kaiser-Meyer-

Olkin measure of sampling adequacy, Bartlett's test of sphericity, communalities, scree plot,

variance explained and the component matrix. Missing data were excluded using pairwise deletion.

A series of Pearson's correlation tests were performed to explore the association between HRQoL and self-reported depression, health anxiety and coping strategies. To test whether depression or health anxiety predicted HRQoL and explore whether different coping styles moderated this relationship, a series of moderated multiple regression analyses were conducted - all predictor variables were mean-centred. Significant interaction terms were decomposed using a series of simple slope analyses; transforming coping strategies into dichotomous variables to examine the relationship at high (1SD above the mean) and low (1SD below the mean) use of each coping strategy. It is not common practice to correct alpha levels in regression studies and so the alpha level of 0.05 was accepted [32]. All data analyses were performed using SPSS25.

Patient, Public Involvement

The conception and research design of the study was developed in collaboration with Pulmonary Hypertension Association UK.

Results

Socio-demographic and clinical characteristics

Demographic characteristics of the sample (n=121) are summarised in Table 1. On average, participants had PH for 8.8 years; however, a large variance was observed. Most participants had either idiopathic PH or chronic thromboembolic PH; however, 13.2% were not sure of the group of their diagnosis. The largest group of individuals did not know their functional class (39.7%); of those who did, class III and II were most reported.

Participant's mean HRQoL was 23.6 (SD = 13.5) and health anxiety 15.4 (SD = 7.3). On average, participants scored 8.9 (SD = 6.2) on the measure of depression. Findings

suggested less than one third experienced no symptoms of depression (29.8%), whereas

42.9% of the sample met criteria indicative of the possible presence of clinically significant

depression warranting the need for intervention (see Table 1).

Table 1 Socio-demographic and clinical factors (n=121), mean and (standard deviation) unless otherwise stated

Factors		Responses
Age		58.3 (14.7)
Gender		
	Female	73.6%
	Male	26.4%
Ethnicity		
	White	99.2%
	Asian	0.8%
Living status		
	Alone	18.2%
	With family	81%
	With friends	0.8%
Employment status		
	Employed	28.1%
	Retired	52.9%
	Student	1.7%
	Unemployed	17.4%
Education		
	High school	23%
	College or higher	77%
Years since PH diagnosis		8.5 (8.8)
Type of PH		
	Idiopathic PH	38%
	Connective tissue disease	6.6%
	Chronic thromboembolic PH	25.6%
	Congenital PH	8.3%
	Other	8.3%
	Not sure	13.2%
PH Class		
	Class I	6.6%
	Class II	21.5%
	Class III	28.9%
	Class IV	3.3%
	Not sure	39.7%
HRQoL		
		23.6 (13.5)
PHQ9		
	Overall	8.9 (6.2)
	None (0-4)	29.8%
	Mild (5-9)	27.3%
	Moderate (10-14)	24%
	Moderately severe (15-19)	12.4%
	Severe (20-27)	6.5%
SHAI		
		15.4 (7.3)

PH = Pulmonary Hypertension; HRQoL = Health Related Quality of life; PHQ9 = Patient Health Questionnaire-9; SHAI = Short Health Anxiety Inventory

Coping profiles

Participants' scores on each of the 14 strategies of coping are displayed in Table 2.

Overall, the most frequently used coping strategy was acceptance and self-distraction,

whereas substance abuse and behavioural disengagement were the least common.

Table 2 Participant's scores on Brief COPE items

Brief COPE coping strategy	Mean (standard deviation)
Acceptance	6.1 (1.8)
Self-distraction	5.2 (1.9)
Active coping	5 (1.8)
Use of emotional support	4.7 (1.8)
Planning	4.7 (1.9)
Positive refraining	4.5 (1.9)
Use of informational support	4 (1.7)
Self-blame	3.6 (1.7)
Humour	3.6 (1.8)
Venting	3.5 (1.6)
Religion	3.2 (1.9)
Denial	3 (1.3)
Behavioural disengagement	2.9 (1.2)
Substance use	2.6 (1.2)

Results from the exploratory factor analysis using a principal component extraction

method of the 28 Brief COPE items, indicated the Kaiser-Meyer-Olkin measure of sampling

adequacy was 0.745 (above the recommended value of > 0.6). Bartlett's test of sphericity was

significant ($\chi2$ (378) = 1719.24, $p < 0.001$). Communalities for the 28 items were > 0.3.

Therefore, findings support the use of a factor analysis to examine data collected using the

Brief COPE.

The initial eigenvalues and scree plot indicated eight factors (>1 eigenvalue)

accounted for 70.45% of the cumulative variance; the first four factors accounted for 25.1%,

10.3%, 9.2% and 7% (>1.97 eigenvalue), whereas the remaining four explained 5.3%, 5.1%,

4.5% and 3.9% (<1.49 eigenvalue). A four-factor solution was preferred because of the

levelling off of variance as indicated by eigenvalues and visual inspection of the scree plot

(Fig. 1), which demonstrated a discontinuity in the curve after the fourth factor. Moreover,

many of the items that loaded (>0.4) on factors five to eight, were also co-loaded (>0.3) on

the first four factors.

>insert figure 1 here<

The final four-factor model explained 51.62% of the cumulative variance (all

communalities remained at >0.3). After inspecting the pattern matrix, eight items were

removed because they co-loaded (>0.3) on more than one factor, which were related to active

(n=2), denial (n=2), humour (n=2), planning and venting - thus, no items relating to denial,

active coping or humour were included in the final model. All 28 items loaded on at least one

primary factor (>0.4).

The first factor included items relating to distraction (n=n2), positive reframing (n=2),

acceptance (n=2) and planning (n=1). Cronbach's alpha was good (α = 0.83). As these

strategies reflect participants' thinking processes, the factor was named "cognitive coping".

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The second factor consisted of items assessing behavioural disengagement (n=2) and self-

blame (n=2), which had an acceptable level of reliability (α=0.73). These strategies are

characteristic of avoidance and unhelpful approaches; as such, this factor was labelled

"passive coping". The third factor represented items assessing emotional support (n=2),

instrumental support (n=2), religion (n=2) and venting (n=1). Reliability was good (α=0.823).

Given all these strategies involve seeking support from others, it was termed "external

coping". The final factor included items addressing substance use (n=2). Alpha level was

excellent (α=0.938); this construct was subsequently named "substance use

coping". Composite scores were created for each strategy of coping by summing the items; a

higher score indicating greater use (Table 3).

Table 3 Factor loading from principle factor analysis with direct oblimin rotation for a four-factor solution model

Item		Factor loading				Communality
		Cognitive coping	Passive coping	External coping	Substance use coping	
1	I've been turning to work or other activities to take my mind off things	.55				.48
4	I've been using alcohol or other drugs to make myself feel better				.87	.78
5	I've been getting emotional support from others			-.73		.59
6	I've been giving up trying to deal with it		.67			.47
10	I've been getting help and advice from other people			-.66		.54
11	I've been using alcohol or other drugs to help me get through it				.9	.84
12	I've been trying to see it in a different light, to make it seem more positive	.67				.59
13	I've been criticising myself		.67			.52
15	I've been getting comfort and understanding from someone			-.77		.6
16	I've been giving up the attempt to cope		.77			.6
17	I've been looking for something good in what is happening	.67				.59
19	I've been doing something to think about it less, such as going to movies, watching TV, reading, daydreaming, sleeping or shopping	.56				.41
20	I've been accepting the reality of the fact it has happened	.65				.45
21	I've been expressing my negative feelings			-.6		.48
22	I've been trying to find comfort in my religion or spiritual beliefs			-.48		.33
23	I've been trying to get advice or help from other people about it			-.71		.59
24	I've been learning to live with it	.69				.46
25	I've been thinking hard about what steps to take	.6				.49
26	I've been blaming myself for things that happened		.65			.44
27	I've been praying or meditating			-.45		.32
Eigenvalue		7	2.89	2.58	1.97	
% of variance		25.6	10.3	9.2	7	
Mean		18.38	6.45	13.98	2.58	
Standard deviation		4.95	2.5	4.6	1.15	

Co-loaded items have been removed

Relationship between coping style, depression, health anxiety and HRQoL

A series of Pearson's correlational analyses demonstrated HRQoL was significantly related to health anxiety ($r = 0.55$, $p <0.001$) and depression ($r = 0.61$, $p <0.001$). In turn, health anxiety and depression were significantly correlated ($r =0.65$, $p <0.001$).

Regarding coping strategies, passive coping was significantly positively correlated with depression, health anxiety and lower HRQoL. Substance use was positively related to depression and passive coping. External coping was positively associated with health anxiety and cognitive coping. Cognitive coping was not significantly related to any clinical measure (Table 4).

Table 4. Pearson's correlation coefficients between coping and health measures

	HRQoL	Depression	Health anxiety	Cognitive coping	Passive coping	External coping
Depression	0.61***					
Health anxiety	0.55***	0.66***				
Cognitive coping	0.04	0.1	0.14			
Passive coping	0.42***	0.68***	0.52***	0.08		
External coping	0.02	0.1	0.19*	0.47***	0.12	
Substance use coping	0.11	0.28**	0.12	-0.008	0.22*	-0.06

HRQoL = Health Related Quality of life (higher scores suggesting lower HRQoL)
*<0.05
**<0.01
***<0.001

A linear regression analysis revealed depression significantly predicted HRQoL, accounting for 37% of the variance ($F(1,119) = 70$, $p < 0.001$). A series of moderated regression analyses were performed by adding in the interaction term between coping strategy (cognitive coping, passive coping, external coping and substance use coping) and depression. The relationship between depression and HRQoL ($\Delta R2 = 0.4$, $\Delta F(3,116) = 26.1$, standardised $\beta = 0.63$, $p < 0.001$) was found to be moderated by cognitive coping (standardized $\beta = -0.18$, $p = 0.017$) as indicated by significant interaction effects. The

relationship between the same variables ($\Delta R2 = 0.4$, $\Delta F(3,116) = 25.2$, standardised $\beta = 0.62$, p <0.001) was also moderated by external coping (standardized $\beta = -0.14$, p = 0.05). No significant interaction effects were observed in passive coping or substance use. The interactions were decomposed using simple slopes analyses, which demonstrated there was a buffering effect of both cognitive coping and external coping strategies, such that high levels of cognitive (standardized $\beta = 0.47$, p < 0.001) and external coping (standardized $\beta = 0.48$, p p < 0.001) weakened the relationship between depression and HRQoL. Conversely, low levels of coping strengthened the association (standardized β cognitive = 0.79, p < 0.001; standardized β external = 0.76, p < 0.001).

Health anxiety was also found to significantly predict HRQoL, explaining 30% of the variance ($F(1,119) = 51.1$, p < 0.001). The same analysis described above suggested the relationship between health anxiety and HRQoL ($\Delta R2 = 0.35$, $\Delta F(3,116) = 20.45$, standardized $\beta = 0.58$, p<0.001) was moderated by external coping (standardized $\beta = -0.17$, p = 0.026), as indicated by significant interaction effects. A simple slopes analysis revealed high use of external coping reduced the association between health anxiety and HRQoL (standardized $\beta = 0.42$, p<0.001), whereas low use increased the relationship (standardized β = 0.74, p<0.001).

Discussion

This is the first study to investigate the coping profile of people with PH. Using the Brief COPE tool, we identified a four-factor model of coping in PH and we have investigated how these four coping styles relate to depression, health anxiety and HRQoL. Our findings are consistent with other research demonstrating the high prevalence rates of depression and anxiety in those with PH [20]. However, given that the current sample was recruited from the community, as opposed to individuals under the care of services, it is alarming that 42.9% of

participants met the cut-off indicative of the possible presence of clinically significant

depression. Moreover, to our knowledge, this is the first study examining anxiety relating to

health specifically in PH. On the group level, participants' scores were greater than what has

been reported using the same measure in patients with a diagnosed anxiety disorder (M =

14.9); individuals attending gastroenterology (M = 11.4), general practitioner appointments

(M = 11.2), and MRI appointments (M = 10.6); students (M = 9.6) and non-patient controls

(M = 9.4) [29]. The high levels of depression and anxiety found in our community sample are

at a stark contrast to the fact that just over one-third of patients with PH who present with

mood difficulties receive treatment [33].

People with PH demonstrated employing a range of strategies to cope with life

adversities, with a greater tendency to use acceptance, self-distraction and active coping, as

opposed to substance abuse and behavioural disengagement. Results indicate this is likely to

be a helpful approach, as our factor analysis demonstrated that the latter two coping strategies

loaded on two of the four factors - passive coping and substance use coping - which were

related to depression, health anxiety and lower HRQoL. This suggests that although these

coping strategies are used less frequently in this group, it is important to identify individuals

who are at risk of engaging in such behaviours.

The third coping strategy identified in the factor analysis was termed external coping,

characterised by behaviours through which participants sought practical, emotional and

spiritual support from others. This coping strategy was found to be an important moderator,

mitigating (although not fully eliminating) the impact of both depression and health anxiety

on HRQoL. Receiving support from others is a common strategy of coping observed across

populations living with a long-term condition [34]. However, patients with PH have

previously described difficulties when seeking care. For instance, fear of stigmatisation and

lack of understanding from others regarding the nature of the disease has resulted in some

feeling the need to conceal their symptoms and avoid situations where they may have to

disclose their diagnosis [35, 36]. Given the importance of external networks, support-targeted

interventions are likely to be beneficial in helping to reduce the stigma and misunderstanding

surrounding the condition, thus better supporting people with PH to share their experiences

and receive care from others. Moreover, while carers' needs are all too often overlooked in

medical provision, evidence has demonstrated the social and emotional impact PH can also

have on carers [37]. Therefore, helping to increase carers' knowledge, confidence and

develop helpful strategies of coping is also likely to contribute to further improvements in

patients' HRQoL.

Interestingly, our correlation analysis suggested that greater use of external coping

was related to higher levels of health anxiety. While it should be noted that this relationship

was low ($r = 0.19$), it is nevertheless consistent with the tendency of people with health

anxiety to seek reassurance and support from others as a way of managing anxiety and

uncertainty about their symptoms. While this can be an effective strategy reducing feelings of

anxiety in the short-term, over-reliance on reassurance-seeking tends to serve an unhelpful

maintenance function in the long-term [38, 39]. However, this needs to be balanced with the

need for individuals with PH to be vigilant towards change in their PH symptoms and seek

medical care when necessary.

The final factor identified in the analysis represented cognitive coping strategies. This

consisted of planning, acceptance, self-distraction and positive reframing. One of the most

well-known models of coping developed by Lazarus and Folkman [9] distinguishes between

problem-focused and emotion-focused (also known as active and passive) coping, suggesting

problem-focused coping efforts to be an adaptive way of responding to stressors – although

this is debated [40]. Indeed, qualitative evidence has demonstrated patients with PH engage

in extensive planning to help manage the short and long-term effects of the disease [41].

Importantly, in the present study, cognitive coping was found to moderate the relationship between depression and HRQoL, serving a protective function for those who made greater use of this strategy. Helping patients with PH develop cognitive coping skills may therefore be a helpful target for intervention. While there is a dearth of evidence examining psychological interventions in PH, there is some evidence to support their use [20, 42]. A pilot randomised controlled trial of a self-help Cognitive Behavioural Therapy intervention for anxiety in PH found changes in unhelpful cognitions and behaviours mediated the relationship between intervention group (CBT vs. waitlist) and change in anxiety and depression [22]. The use of cognitive techniques to alleviate distress is at the core of CBT where positive reframing and distraction are often utilised, as well as third-wave CBT approaches, such as Acceptance and Commitment Therapy (ACT), which makes use of acceptance and mindfulness. Both types of treatment may be beneficial to patients with PH and the use of these approaches in this patient group merits future research.

Limitations

Given that participants self-reported the aetiology of their PH group and WHO function classification, with a considerable proportion reporting to be "not sure" of either, we did not add these as factors in the regression analyses. This approach was taken in order to avoid type one and two errors. Indeed, PH class has been shown to predict participants' HRQoL using the emPHasis10 measure [25].

A further limitation was the representativeness of our sample. While many of the demographic characteristics were typical of PH samples (e.g., a higher proportion of females and those aged 50 and above) [43], participants in this study were a self-selected group of relatively highly educated, English-speaking individuals proactively accessing support from a PH organisation online. As such, participants' involvement in the charity may be impacting

on their coping style, as reflected by their scores on seeking emotional and instrumental support.

The findings should be viewed in light of the knowledge that the data was collected in the weeks following the easing of shielding (this involved people in the UK with a pre-existing medical condition that made them more susceptible to COVID-19 (such as PH) spend twelve weeks taking additional infection control precautions). This may have conceivably contributed to increased levels of health-related worry and anxiety within our sample not only for oneself but also for loved ones, and therefore a greater need to turn to others for practical and emotional help and support.

Conclusions

This study has provided a unique insight into the coping styles of people with PH. Four distinct coping styles emerged, which were related to measures of depression, health anxiety and HRQoL. While coping by seeking external support as well as cognitive coping strategies had a buffering effect on the impact of health anxiety and depression on HRQoL, the tendency to use more passive ways of coping and coping by substance misuse were associated with poorer psychological outcomes. The results are consistent with previous research suggesting people with PH are at a greater risk of experiencing psychopathologies. Findings add to growing evidence base demonstrating the role of psychological factors in predicting quality of life in those with this disease. Understanding the coping styles of this sample has highlighted a range of potential therapeutic targets, supporting the use of psychological treatments aimed at cognitive and emotional processes. Given the observed importance of support systems, patients are also likely to benefit from interventions focused on the provision of information and support to caregivers, although further research is required in these areas.

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Authors' contribution

Dr Gregg H Rawlings was responsible for the conception of the study, data collection and analysis, and writing up the study for publication. He approved the final version for publication.

Prof Andrew R. Thompson was responsible for the conception of the study, provided feedback on data collection and analysis, and writing up the study for publication. He approved the final version for publication.

Dr Iain Armstrong was responsible for the conception of the study, provided feedback on data collection, and writing up the study for publication. He approved the final version for publication.

Dr Barbora Novakova was responsible for data analysis and writing up the study for publication. She approved the final version for publication.

Prof Nigel Beail was responsible for the conception of the study, provided feedback on data collection and analysis, and writing up the study for publication. He approved the final version for publication.

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Competing interest statement

The authors report no conflict of interest.

Ethics approval

Ethical permission was obtained from the University of Sheffield, UK (035318).

Consent to participate

All participants provided informed consent.

Consent for publication

The authors give consent for publication

Data availability statement

The data that supports the findings of this study are available from the corresponding author

upon reasonable request.

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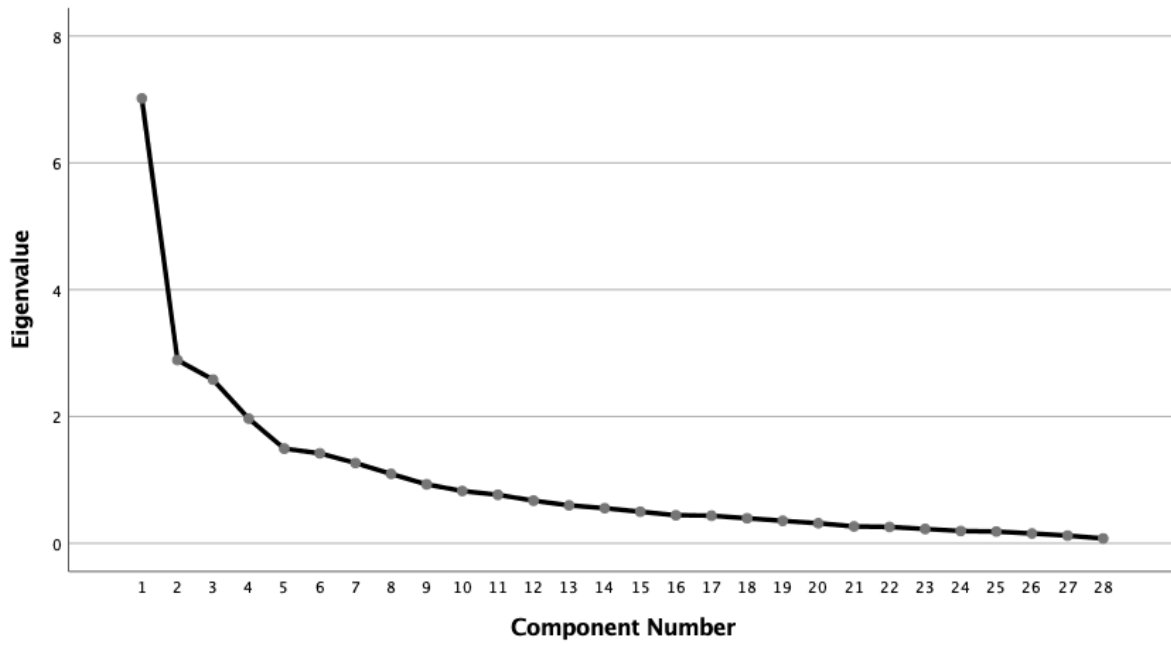patients' and carers' lives. *European Respiratory Review, 22*, 535-542.

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Figure 1. Scree plot prior to final four factor analysis of coping in pulmonary hypertension

Figure 1. Scree plot prior to final four factor analysis of coping in pulmonary hypertension

STROBE 2007 (v4) Statement—Checklist of items that should be included in reports of *cross-sectional studies*

Section/Topic	Item #	Recommendation	Reported on page #
Title and abstract	1	(*a*) Indicate the study's design with a commonly used term in the title or the abstract	1
		(*b*) Provide in the abstract an informative and balanced summary of what was done and what was found	2-3
Introduction			
Background/rationale	2	Explain the scientific background and rationale for the investigation being reported	4-6
Objectives	3	State specific objectives, including any prespecified hypotheses	5-6
Methods			
Study design	4	Present key elements of study design early in the paper	6
Setting	5	Describe the setting, locations, and relevant dates, including periods of recruitment, exposure, follow-up, and data collection	6
Participants	6	(*a*) Give the eligibility criteria, and the sources and methods of selection of participants	6
Variables	7	Clearly define all outcomes, exposures, predictors, potential confounders, and effect modifiers. Give diagnostic criteria, if applicable	7-8
Data sources/ measurement	8*	For each variable of interest, give sources of data and details of methods of assessment (measurement). Describe comparability of assessment methods if there is more than one group	7-8
Bias	9	Describe any efforts to address potential sources of bias	7-9
Study size	10	Explain how the study size was arrived at	6
Quantitative variables	11	Explain how quantitative variables were handled in the analyses. If applicable, describe which groupings were chosen and why	8-9
Statistical methods	12	(*a*) Describe all statistical methods, including those used to control for confounding	8-9
		(*b*) Describe any methods used to examine subgroups and interactions	8-9
		(*c*) Explain how missing data were addressed	8-9
		(*d*) If applicable, describe analytical methods taking account of sampling strategy	na
		(*e*) Describe any sensitivity analyses	8-9
Results			

Participants	13*	(a) Report numbers of individuals at each stage of study—eg numbers potentially eligible, examined for eligibility, confirmed eligible, included in the study, completing follow-up, and analysed	9-10
		(b) Give reasons for non-participation at each stage	Na
		(c) Consider use of a flow diagram	Na
Descriptive data	14*	(a) Give characteristics of study participants (eg demographic, clinical, social) and information on exposures and potential confounders	11
		(b) Indicate number of participants with missing data for each variable of interest	Na
Outcome data	15*	Report numbers of outcome events or summary measures	9-17
Main results	16	(*a*) Give unadjusted estimates and, if applicable, confounder-adjusted estimates and their precision (eg, 95% confidence interval). Make clear which confounders were adjusted for and why they were included	9-17
		(*b*) Report category boundaries when continuous variables were categorized	9-17
		(*c*) If relevant, consider translating estimates of relative risk into absolute risk for a meaningful time period	Na
Other analyses	17	Report other analyses done—eg analyses of subgroups and interactions, and sensitivity analyses	9-17
Discussion			
Key results	18	Summarise key results with reference to study objectives	17-18
Limitations	19	Discuss limitations of the study, taking into account sources of potential bias or imprecision. Discuss both direction and magnitude of any potential bias	20-21
Interpretation	20	Give a cautious overall interpretation of results considering objectives, limitations, multiplicity of analyses, results from similar studies, and other relevant evidence	17-21
Generalisability	21	Discuss the generalisability (external validity) of the study results	17-21
Other information			
Funding	22	Give the source of funding and the role of the funders for the present study and, if applicable, for the original study on which the present article is based	22

*Give information separately for cases and controls in case-control studies and, if applicable, for exposed and unexposed groups in cohort and cross-sectional studies.

Note: An Explanation and Elaboration article discusses each checklist item and gives methodological background and published examples of transparent reporting. The STROBE checklist is best used in conjunction with this article (freely available on the Web sites of PLoS Medicine at http://www.plosmedicine.org/, Annals of Internal Medicine at http://www.annals.org/, and Epidemiology at http://www.epidem.com/). Information on the STROBE Initiative is available at www.strobe-statement.org.

BMJ Open

Coping styles associated with depression, health anxiety and health-related quality of life in pulmonary hypertension: Cross-sectional analysis

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Coping styles associated with depression, health anxiety and health-related quality of life in pulmonary hypertension: Cross-sectional analysis

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Abstract

Objectives: Pulmonary hypertension (PH) is a life-shortening disease associated with early mortality and high morbidity. With advancements in medical treatment, people are living longer with the disease and research is now needed to explore variables that help to enhance patient-reported outcomes. This study investigated the coping strategies of individuals with PH, and examined the relationship between coping, depression, health anxiety and health-related quality of life (HRQoL).

Design: A cross sectional survey design was used.

Participants: Participants (n=121) were recruited from membership of Pulmonary Hypertension Association (PHA) UK.

Outcome measures: Participants completed a series of questionnaires assessing depression (Patient Health Questionnaire-9), health anxiety (Short Health Anxiety Inventory), HRQoL (emPHasis-10) and coping (Brief COPE). A principal component analysis was used to identify participants' coping profile. A series of correlational, linear and moderated multiple regression analyses were performed to examine the relationship between coping and health-related outcomes.

Results: Overall, 43% participants met criteria for potential clinical depression. Depression and health anxiety were strongly associated with HRQoL, explaining 37% and 30% of variance respectively (p<0.001). A principal component analysis identified a four-component model of coping. Dimensions were named based on construct items: "cognitive and affirmation coping" (7-items), "passive coping" (4-items), "external coping" (7-items) and "substance use coping" (2-items). Cognitive and affirmation and external coping moderated the relationship between depression and HRQoL, with high use of these coping strategies reducing the impact of depression on HRQoL. External coping also moderated the effect of health anxiety on HRQoL.

Conclusions: The results uniquely highlight the importance of coping styles and

psychological distress in predicting HRQoL in PH. Our findings indicate the importance to

assess for psychological distress in this population and suggest the need to offer

psychological interventions that take into account coping resources and strategies.

Strengths and limitations

- Understanding how patients cope with the demands of pulmonary hypertension (PH)

  can be used to help inform services and patient-reported outcomes.

- The results demonstrate high rates of distress in PH within a community sample, with

  43% of people scoring above the clinical cut off for depression.

- Coping strategies were found to moderate the relationship between health anxiety and

  depression, and HRQoL in PH.

- Coping by seeking external support and cognitive-based strategies buffered the

  impact of psychological distress on HRQoL.

- Clinical factors, such as PH severity were not included as covariates and therefore, it

  is not clear how objective measures of the impact of PH influence coping.

Key words: Pulmonary arterial hypertension; adjustment; chronic; long-term; coping

Pulmonary hypertension (PH) is a progressive disease characterised by elevated pulmonary artery pressure, which ultimately leads to right heart failure and death if untreated [1]. PH is a global term used to describe five distinct groups of the disease: Group 1, pulmonary arterial hypertension (PAH); Group 2, PH due to left heart disease; Group 3, PH caused by lung disease; Group 4, chronic thromboembolic PH (CTEPH); and Group 5, PH due to miscellaneous causes [2]. Common symptoms of PH can include shortness of breath, fatigue, pain, weakness, dizziness, oedema and in some cases syncope [3].

Better disease awareness and a greater understanding of the aetiologies of PH have helped to advance diagnostic and therapeutic approaches, dramatically improving treatment outcomes, and resulting in lower mortality rates [4, 5]. As in other conditions in which people are living longer with a disease, research is now needed to examine the impact on daily life and identify variables associated with patient-outcomes such as quality of life, the evidence from which can be used to inform care and support [6].

The World Health Organisation (WHO) functional class can be used to help indicate the severity of patients' symptoms of PH. This index ranges from I, indicative of patients experiencing none to mild PH-related symptoms during physical activity, to IV suggesting patients will experience severe symptoms at rest [7]. However, the WHO classification system was not designed to reflect the psychosocial impact of living with PH. A qualitative synthesis of adults' lived accounts summarised the specific challenges posed by the disease, including the burden of living with disabling symptoms; intensive treatment regimens; impact on loved ones; stigma of PH and lack of understanding from others [8].

Coping has been viewed as cognitive, emotional and behavioural strategies employed in an attempt to manage distress perceived as threatening [9]. This response requires the individual to recognise and appraise the stressor, taking into consideration the expected outcome and their perceived ability to cope [10]. A large evidence base exists examining how

people cope with living with long-term health conditions [11-14]; however, no studies to date have quantitively investigated coping in people with PH. Understanding how patients cope with a disease can help to identify targets for intervention, and assist in identification of individuals most at risk of experiencing difficulties [15]. This is particularly relevant in PH, as treatments aim to sustain health-related quality of life (HRQoL) and early intervention is viewed as essential in reducing morbidity [5].

The primary aim of this study was to explore the specific coping strategies used by people with PH. We used the Brief COPE measure [16], which explores 14 different coping strategies, including: self-distraction, denial, substance use, behavioural disengagement, venting, self-blame, active coping, use of emotional support, use of instrumental support, positive reframing, planning, acceptance, humour and religion. This tool is one of the most commonly used measures to assess coping in clinical populations [17]. While some studies have grouped coping strategies dichotomously (i.e. approach- and avoidance-focused coping strategies), there is no clear consensus on scales underlying composition across samples. Moreover, given that the questionnaire was not designed for this purpose, the developer suggests that researchers should group items based on datasets collected from specific samples as the pattern of relationships between items will differ between groups of individuals [18]. Indeed, the variation in subscales identified across clinical samples is likely to reflect the idiosyncratic challenges associated with specific life events. In fact, numerous studies using this measure have applied different statistical analyses to examine the psychometric properties of higher-order structures [19], including principal component analyses to combine items to identify components that define coping styles components [20, 21].

A secondary aim of the current study was to assess how coping strategies are related to depression, health anxiety and HRQoL, using a series of correlational and regression

analyses. As observed elsewhere, the relationship between psychological factors and burden of a long-term health condition is likely to be influenced by the coping strategies an individual utilises (Hundt et al., 2015). Lower HRQoL [22], and depression and anxiety are commonly reported as high in people with PH [22-24]. Although health anxiety has not specifically been examined in this condition, there is evidence to suggest individuals experience anxiety about their condition, which could further contribute to distress [8]. Therefore, we hypothesised depression and health anxiety would be related to lower HRQoL, and this relationship would be moderated by specific components of coping strategies.

Methods

Participants

A cross-sectional design was employed, utilising a convenience sampling method. The article has been conducted in line with the STROBE checklist (supplement A) [25].

Bose et al., [26] examined the relationship between HRQoL and coping in individuals with chronic heart failure. Coping significantly predicted the physical (R^2=0.26, p<0.001) and mental (R^2=0.4, p<0.001) health composite of the QoL measure, equating to an observed effect size (f^2) of 0.35-0.67. A prior analysis using G*Power, with an effect size f^2 of 0.35, alpha level of 0.05, 80% power suggested a total sample size of at least 25 was required.

Participants were recruited over the span of two weeks from advertisements electronically sent to members of Pulmonary Hypertension Association (PHA) UK. To be eligible, participants must have been aged 18 years or over, living in the UK, diagnosed with PH, able to complete self-report questionnaires independently in English, and provide informed consent. Ethical assessment and approval was sought and obtained from the University of Sheffield (035318).

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Procedure

Individuals interested in the study were instructed to access a website link directing them to the participant information sheet. If individuals still wished to take part after reading the information sheet, they were asked to complete a consent form and series of questionnaires (see below). All measures were hosted by Qualtrics.

Measures

Demographic questionnaire

This included name, age, gender, ethnicity, living status, employment status, level of education, PH diagnosis and functional class, and duration of PH.

Quality of life

The emPHasis-10 is a PH specific HRQoL measure [27]. Participants are asked to rate how ten problems related to life with PH have affected them "recently" on a unidimensional scale. Possible scores range from 0-50, with a higher score indicative of lower HRQoL. The measure has excellent internal reliability ($\alpha = 0.9$) [27]; Cronbach's alpha within the current study was $\alpha = 0.94$.

Depression

The Patient Health Questionnaire-9 (PHQ-9) [28] was used to measure symptoms of depression. This is a nine-item measure asking participants how often they have been bothered by a range of depression-related difficulties over the previous two weeks. Scores range from 0-27; a score of 0-4 indicating "no difficulties with depression", 5-9 "mild difficulties", 10-14 "moderate", 15-19 "moderately severe", and 20 or higher "severe depression". A cut off of ≥ 10 has been found to be a valid and reliable measure for major

(clinical) depression [29]. This measure has previously been used in those with PH [30]. The PHQ-9 was found to have good internal consistency ($\alpha = 0.88$) in the current sample.

Health anxiety

The Short Health Anxiety Inventory (SHAI) [31] was used to measure participants' anxiety in response to how they appraised their health. This measure asks respondents to endorse items assessing their thoughts, feelings and behaviours about health anxiety over the past week on a scale ranging from 0–3 according to specific statements. While the SHAI is typically an 18-item measure, it can be converted to 14-items when exploring health anxiety in people with an existing medical disease. The 14-item measure has been found acceptable in clinical samples, demonstrating high internal consistency (Cronbach's alpha 0.84) [32]. There is not a cut-off point that has been used consistently in the literature; however, one study performed a discriminant function analysis that suggested a score of 18 or higher identified people with significant health anxiety (hypochondriasis) [33]. Internal consistency was excellent ($\alpha = 0.9$) in this sample.

Coping styles

The Brief COPE measure [16] was used to examine participants' coping. Individuals were asked how often they engage in 28 behaviours when experiencing hardship in their life, which are aggregated into 14 coping strategies (two items per each coping strategy). Possible scores for each strategy ranged from 2-8, with a higher score indicating greater use. Cronbach alpha's were all within the acceptable range or higher ($\alpha = 0.65 – 0.94$), with the exception of denial ($\alpha = 0.58$).

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Data analysis

HRQoL scores were normally distributed (Kolmogorov-Smirnov $p = 0.085$), therefore

parametric tests were used. Descriptive statistics were utilised to describe participants' socio-

demographic and clinical characteristics, and coping profile. No data was missing.

A principal components analysis was used. A direct oblimin rotation was applied as

there is a strong theoretical basis for the coping strategies investigated here to be related [34].

The determinant of the correlation matrix was 1.379, suggesting this approach was suitable.

Solutions were analysed using Kaiser-Meyer-Olkin measure of sampling adequacy, Bartlett's

test of sphericity, communalities, scree plot, variance explained and the component matrix.

Missing data were excluded using pairwise deletion.

A series of Pearson's correlation tests were performed to explore the association

between HRQoL and self-reported depression, health anxiety and coping strategies. To test

whether depression or health anxiety predicted HRQoL and explore whether different coping

styles moderated this relationship, a series of moderated multiple regression analyses were

conducted - all predictor variables were mean-centred. Significant interaction terms were

decomposed using a series of simple slope analyses; transforming coping strategies into

dichotomous variables to examine the relationship at high (1SD above the mean) and low

(1SD below the mean) use of each coping strategy. Stepwise multiple regression analyses

were performed to identify whether coping components significantly predicted scores of

psychological distress (depression and health anxiety). It is not common practice to correct

alpha levels in regression studies and so the alpha level of 0.05 was accepted [35]. All data

analyses were performed using SPSS25.

Patient, Public Involvement

The conception and research design of the study was developed in collaboration with

Pulmonary Hypertension Association UK.

Results

Socio-demographic and clinical characteristics

Demographic characteristics of the sample (n=121) are summarised in Table 1. On

average, participants had PH for 8.8 years; however, a large variance was observed. Most

participants had either idiopathic PH or chronic thromboembolic PH; however, 13.2% were

not sure of the group of their diagnosis. The largest group of individuals did not know their

functional class (39.7%); of those who did, class III and II were most reported.

Participant's mean HRQoL was 23.6 (SD = 13.5) and health anxiety 15.4 (SD = 7.3).

On average, participants scored 8.9 (SD = 6.2) on the measure of depression. Findings

suggested less than one third experienced no symptoms of depression (29.8%), whereas

42.9% of the sample met criteria indicative of the possible presence of clinically significant

depression warranting the need for intervention (see Table 1).

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Table 1 Socio-demographic and clinical factors (n=121), mean and (standard deviation) unless otherwise stated

Factors		Responses
Age		58.3 (14.7)
Gender		
	Female	73.6% (n=89)
	Male	26.4% (n=32)
Ethnicity		
	White	99.2% (n=120)
	Asian	0.8% (n=1)
Living status		
	Alone	18.2% (n=22)
	With family	81% (n=98)
	With friends	0.8% (n=1)
Employment status		
	Employed	28.1% (n=34)
	Retired	52.9% (n=64)
	Student	1.7% (n=2)
	Unemployed	17.4% (n=21)
Education		
	High school	23% (n=28)
	College or higher	77% (n=93)
Years since PH diagnosis		8.5 (8.8)
Type of PH		
	Idiopathic PH	38% (n=46)
	Connective tissue disease	6.6% (n=8)
	Chronic thromboembolic PH	25.6% (n=31)
	Congenital PH	8.3% (n=10)
	Other	8.3% (n=10)
	Not sure	13.2% (n=16)
PH Class		
	Class I	6.6% (n=8)
	Class II	21.5% (n=26)
	Class III	28.9% (n=35)
	Class IV	3.3% (n=4)
	Not sure	39.7% (n=48)
HRQoL		23.6 (13.5)
PHQ9		
	Overall	8.9 (6.2)
	None (0-4)	29.8% (n=36)
	Mild (5-9)	27.3% (n=33)
	Moderate (10-14)	24% (n=29)
	Moderately severe (15-19)	12.4% (n=15)
	Severe (20-27)	6.5% (n=8)
SHAI		
		15.4 (7.3)
	Scoring 18 or above	34.7% (n=42)

PH = Pulmonary Hypertension; HRQoL = Health Related Quality of life; PHQ9 = Patient Health Questionnaire-9; SHAI = Short Health Anxiety Inventory; n = number of participants

Coping profiles

Participants' scores on each of the 14 strategies of coping are displayed in Table 2. Overall, the most frequently used coping strategy was acceptance and self-distraction, whereas substance abuse and behavioural disengagement were the least common.

Table 2 Participant's scores on Brief COPE items

Brief COPE coping strategy	Mean (standard deviation)
Acceptance	6.1 (1.8)
Self-distraction	5.2 (1.9)
Active coping	5 (1.8)
Use of emotional support	4.7 (1.8)
Planning	4.7 (1.9)
Positive refraining	4.5 (1.9)
Use of informational support	4 (1.7)
Self-blame	3.6 (1.7)
Humour	3.6 (1.8)
Venting	3.5 (1.6)
Religion	3.2 (1.9)
Denial	3 (1.3)
Behavioural disengagement	2.9 (1.2)
Substance use	2.6 (1.2)

Results from the principal component analysis of the 28 Brief COPE items, indicated the Kaiser-Meyer-Olkin measure of sampling adequacy was 0.75 (above the recommended value of > 0.6). Bartlett's test of sphericity was significant ($\chi2$ (378) = 1719.2, $p < 0.001$). Communalities for the 28 items were > 0.3. Therefore, findings support the use of the proposed method to examine data collected using the Brief COPE.

The initial eigenvalues and scree plot indicated eight components (>1 eigenvalue) accounted for 70.5% of the cumulative variance; the first four components accounted for 25.1%, 10.3%, 9.2% and 7% (>2 eigenvalue), whereas the remaining four explained 5.3%, 5.1%, 4.5% and 3.9% (<1.5 eigenvalue). A four-component solution was preferred because of the levelling off of variance as indicated by eigenvalues and visual inspection of the scree

plot (Fig. 1), which demonstrated a discontinuity in the curve after the fourth components. Moreover, many of the items that loaded (>0.4) on components five to eight, were also co-loaded (>0.3) on the first four components.

>insert figure 1 here<

The final four- components model explained 51.6% of the cumulative variance (all communalities remained at >0.3). After inspecting the pattern matrix, eight items were removed because they co-loaded (>0.3) on more than one component, which were related to active (n=2), denial (n=2), humour (n=2), planning and venting - thus, no items relating to denial, active coping or humour were included in the final model. All 28 items loaded on at least one primary component (>0.4).

The first component included items relating to distraction (n=2), positive reframing (n=2), acceptance (n=2) and planning (n=1). Cronbach's alpha was $\alpha = 0.8$. As these strategies reflect participants' thinking processes, the component was named "cognitive and affirmation coping". The second component consisted of items assessing behavioural

disengagement (n=2) and self-blame (n=2). Cronbach's alpha was α=0.7. These strategies are

characteristic of avoidance and unhelpful approaches; as such, this component was labelled

"passive coping". The third component represented items assessing emotional support (n=2),

instrumental support (n=2), religion (n=2) and venting (n=1). Cronbach's alpha was α=0.8.

Given all these strategies involve seeking support from others, it was termed "external

coping". The final component included items addressing substance use (n=2). Cronbach's

alpha was α=0.9; this construct was subsequently named "substance use coping". Composite

scores were created for each strategy of coping by summing the items; a higher score

indicating greater use (Table 3).

Table 3 Loading from the principal component s analysis with direct oblimin rotation for a four-component solution model

Item		Component loading				Communality
		Cognitive and affirmation coping	Passive coping	External coping	Substance use coping	
1	I've been turning to work or other activities to take my mind off things	.55				.48
4	I've been using alcohol or other drugs to make myself feel better				.87	.78
5	I've been getting emotional support from others			-.73		.59
6	I've been giving up trying to deal with it		.67			.47
10	I've been getting help and advice from other people			-.66		.54
11	I've been using alcohol or other drugs to help me get through it				.9	.84
12	I've been trying to see it in a different light, to make it seem more positive	.67				.59
13	I've been criticising myself		.67			.52
15	I've been getting comfort and understanding from someone			-.77		.6
16	I've been giving up the attempt to cope		.77			.6
17	I've been looking for something good in what is happening	.67				.59
19	I've been doing something to think about it less, such as going to movies, watching TV, reading, daydreaming, sleeping or shopping	.56				.41
20	I've been accepting the reality of the fact it has happened	.65				.45
21	I've been expressing my negative feelings			-.6		.48
22	I've been trying to find comfort in my religion or spiritual beliefs			-.48		.33
23	I've been trying to get advice or help from other people about it			-.71		.59
24	I've been learning to live with it	.69				.46
25	I've been thinking hard about what steps to take	.6				.49
26	I've been blaming myself for things that happened		.65			.44
27	I've been praying or meditating			-.45		.32
Eigenvalue		7	2.89	2.58	1.97	
% of variance		25.6	10.3	9.2	7	
Mean		18.38	6.45	13.98	2.58	
Standard deviation		4.95	2.5	4.6	1.15	

Co-loaded items have been removed

Relationship between coping style, depression, health anxiety and HRQoL

A series of Pearson's correlational analyses demonstrated HRQoL was significantly related to health anxiety ($r = 0.55$, $p < 0.001$) and depression ($r = 0.61$, $p < 0.001$). In turn, health anxiety and depression were significantly correlated ($r = 0.65$, $p < 0.001$).

Regarding coping strategies, passive coping was significantly positively correlated with depression, health anxiety and lower HRQoL. Substance use was positively related to depression and passive coping. External coping was positively associated with health anxiety and cognitive and affirmation coping. Cognitive and affirmation coping was not significantly related to any clinical measure (Table 4). A stepwise multiple regression analyses revealed passive coping significantly predicted both depression ($F(1,118)=103.79$,, p<0.001) accounting for 46% of the variation, and health anxiety ($F(1,118) = 43.53$, p<0.001) accounting for 26% – the other approaches were excluded by the analysis.

Table 4. Pearson's correlation coefficients between coping and health measures

	HRQoL	Depression	Health anxiety	Cognitive and affirmation coping	Passive coping	External coping
Depression	0.61***					
Health anxiety	0.55***	0.66***				
Cognitive and affirmation coping	0.04	0.1	0.14			
Passive coping	0.42***	0.68***	0.52***	0.08		
External coping	0.02	0.1	0.19*	0.47***	0.12	
Substance use coping	0.11	0.28**	0.12	-0.01	0.22*	-0.06

HRQoL = Health Related Quality of life (higher scores suggesting lower HRQoL)
*<0.05
**<0.01
***<0.001

A linear regression analysis revealed depression significantly predicted HRQoL, accounting for 37% of the variance (F(1,119) = 70, p < 0.001). A series of moderated regression analyses were performed by adding in the interaction term between coping

strategy (cognitive and affirmation coping, passive coping, external coping and substance use coping) and depression (Table 5). The relationship between depression and HRQoL ($\Delta R2 = 0.4$, $\Delta F(3,116) = 26.1$, standardised $\beta = 0.63$, $p < 0.001$) was found to be moderated by cognitive and affirmation coping (standardized $\beta = -0.18$, $p = 0.017$) as indicated by significant interaction effects. The relationship between the same variables ($\Delta R2 = 0.4$, $\Delta F(3,116) = 25.2$, standardised $\beta = 0.62$, $p < 0.001$) was also moderated by external coping (standardized $\beta = -0.14$, $p = 0.05$). No significant interaction effects were observed in passive coping or substance use. The interactions were decomposed using simple slopes analyses, which demonstrated there was a buffering effect of both cognitive and affirmation coping and external coping strategies, such that high levels of cognitive and affirmation (standardized $\beta = 0.47$, $p < 0.001$) and external coping (standardized $\beta = 0.48$, $p < 0.001$) weakened the relationship between depression and HRQoL. Conversely, low levels of coping strengthened the association (standardized β cognitive and affirmation = 0.79, $p < 0.001$; standardized β external = 0.76, $p < 0.001$).

Health anxiety was also found to significantly predict HRQoL, explaining 30% of the variance ($F(1,119) = 51.1$, $p < 0.001$). The same analysis described above suggested the relationship between health anxiety and HRQoL ($\Delta R2 = 0.35$, $\Delta F(3,116) = 20.45$, standardized $\beta = 0.58$, $p<0.001$) was moderated by external coping (standardized $\beta = -0.17$, $p = 0.03$), as indicated by significant interaction effects (Table 5). A simple slopes analysis revealed high use of external coping reduced the association between health anxiety and HRQoL (standardized $\beta = 0.42$, $p<0.001$), whereas low use increased the relationship (standardized $\beta = 0.74$, $p<0.001$).

Table 5 Moderated regression analyses without simple slopes analysis (HRQoL as dependent variable)

Models	R2	β	t
Depression	0.4	0.63	8.68***
Cognitive and affirmation coping		-0.04	-0.53
Depression x Cognitive and affirmation coping		-0.18	-2.42*
Depression	0.38	0.61	6.07***
Passive coping		0.5	0.41
Depression x Passive coping		-0.08	-0.85
Depression	0.4	0.62	8.53***
External coping		-0.06	-0.79
Depression x External coping		-0.14	-1.98*
Depression	0.38	0.63	8.11***
Substance use coping		-0.06	-0.66
Depression x Substance use coping		0.01	0.08
Health anxiety	0.32	0.56	7.23***
Cognitive and affirmation coping		-0.05	-0.65
Health anxiety x Cognitive and affirmation coping		-0.09	-1.17
Health anxiety	0.34	0.47	5.25***
Passive coping		0.21	2.1*
Health anxiety x Passive coping		-0.07	-0.76
Health anxiety	0.35	0.58	7.56***
External coping		-0.1	-1.27
Health anxiety x External coping		-0.17	-2.25*
Health anxiety	0.31	0.55	7.04***
Substance use coping		0.05	0.57
Health anxiety x Substance use coping		0.001	0.02

P<0.05, **p<0.01, p<0.001

Discussion

This is the first study to investigate the coping profile of people with PH. Using the

Brief COPE tool, we identified a four-component model of coping in PH and we have

investigated how these four coping styles relate to depression, health anxiety and HRQoL.

Our findings are consistent with other research demonstrating the high rates of depression

and anxiety in those with PH [23]. However, given that the current sample was recruited from

the community, as opposed to individuals under the care of services, it is alarming that 42.9%

of participants met the cut-off indicative of the possible presence of clinically significant

depression and 34.7% for health anxiety. Moreover, to our knowledge, this is the first study

examining anxiety relating to health specifically in PH. On the group level, participants' scores (M=15.4) were greater than what has been reported using the same measure in patients with a diagnosed anxiety disorder (M = 14.9); individuals attending gastroenterology (M = 11.4), general practitioner appointments (M = 11.2), and MRI appointments (M = 10.6); students (M = 9.6) and non-patient controls (M = 9.4) [31]. The high levels of depression and anxiety found in our community sample are at a stark contrast to the fact that just over one-third of patients with PH who present with mood difficulties receive treatment [36].

People with PH demonstrated employing a range of strategies to cope with life adversities, with a greater tendency to use acceptance, self-distraction and active coping, as opposed to substance abuse and behavioural disengagement. Results indicate this is likely to be a helpful approach, as our analysis demonstrated that the latter two coping strategies loaded on two of the four components - passive coping and substance use coping - which were related to depression, health anxiety and lower HRQoL. In fact, in a stepwise regression analysis, passive coping was the only significant predictor of depression and health anxiety suggesting that although these coping strategies are used less frequently in this group, it is important to identify individuals who are at risk of engaging in such behaviours. However, it should also be noted that due to the cross-sectional nature of this study, we cannot make any strong conclusions regarding the direction of the relationship; for example, whether passive coping is a causal factor or a symptom of depression or anxiety.

The third coping strategy identified in the analysis was termed external coping, characterised by behaviours through which participants sought practical, emotional and spiritual support from others. This coping strategy was found to be an important moderator, mitigating (although not fully eliminating) the impact of both depression and health anxiety on HRQoL. Receiving support from others is a common strategy of coping observed across populations living with a long-term condition [37]. However, patients with PH have

previously described difficulties when seeking care. For instance, fear of stigmatisation and lack of understanding from others regarding the nature of the disease has resulted in some feeling the need to conceal their symptoms and avoid situations where they may have to disclose their diagnosis [38, 39]. Given the importance of external networks, support-targeted interventions are likely to be beneficial in helping to reduce the stigma and misunderstanding surrounding the condition, thus better supporting people with PH to share their experiences and receive care from others. Moreover, while carers' needs are all too often overlooked in medical provision, evidence has demonstrated the social and emotional impact PH can also have on carers [40]. Therefore, helping to increase carers' knowledge, confidence and develop helpful strategies of coping is also likely to contribute to further improvements in patients' HRQoL.

Interestingly, our analysis suggested that greater use of external coping was related to higher levels of health anxiety. While it should be noted that the correlation was low ($r = 0.19$), it is nevertheless consistent with the tendency of people with health anxiety to seek reassurance and support from others as a way of managing anxiety and uncertainty about their symptoms. It is also important to note that this coping approach was the only component that moderated the relationship between HRQoL and both health anxiety and depression suggesting high use reduced the association. While this can be an effective strategy reducing feelings of anxiety in the short-term, over-reliance on reassurance-seeking tends to serve an unhelpful maintenance function in the long-term [41, 42]. However, this needs to be balanced with the need for individuals with PH to be vigilant towards change in their PH symptoms and seek medical care when necessary. It may therefore be important for future research to identify helpful ways for individuals with the disease to receive external support, which does not perpetuate their anxiety.

The final component identified in the analysis represented cognitive and affirmation coping strategies. This consisted of planning, acceptance, self-distraction and positive reframing. One of the most well-known models of coping developed by Lazarus and Folkman [9]distinguishes between problem-focused and emotion-focused (also known as active and passive) coping, suggesting problem-focused coping efforts to be an adaptive way of responding to stressors – although this is debated [43]. Indeed, qualitative evidence has demonstrated patients with PH engage in extensive planning to help manage the short and long-term effects of the disease [44].

Importantly, in the present study, cognitive and affirmation coping was found to moderate the relationship between depression and HRQoL, serving a protective function for those who made greater use of this strategy. Helping patients with PH develop cognitive coping skills may therefore be a helpful target for intervention. While there is a dearth of evidence examining psychological interventions in PH, there is some evidence to support their use [23, 45]. A pilot randomised controlled trial of a self-help Cognitive Behavioural Therapy intervention for anxiety in PH found changes in unhelpful cognitions and behaviours mediated the relationship between intervention group (CBT vs. waitlist) and change in anxiety and depression [46]. The use of cognitive techniques to alleviate distress is at the core of CBT where positive reframing and distraction are often utilised, as well as third-wave CBT approaches, such as Acceptance and Commitment Therapy (ACT), which makes use of acceptance and mindfulness. Both types of treatment may be beneficial to patients. Indeed, while there is growing evidence demonstrating a link between psychological factors and patient-report outcomes, further research is needed. Coping strategies identified in the present sample merits future research to validate proposed components for example using a confirmatory factor analysis. This could also be assessed in specific sub-groups of people with PH such as those with diagnosed anxiety or depression, or individuals from non-white

communities (120 of 121 self-reported themselves as white in the current study).

Longitudinal research would also clarify the relationship between coping styles and

challenges associated with PH overtime, for example, experiences of diagnosis, managing the

daily demands of the condition and palliative and end of life care.

Limitations

Given that participants self-reported the aetiology of their PH group and WHO

function classification, with a considerable proportion reporting to be "not sure" of either, we

did not add these as factors in the regression analyses. This approach was taken in order to

avoid type one and two errors. Indeed, PH class has been shown to predict participants'

HRQoL using the emPHasis10 measure [27].

A further limitation was the representativeness of our sample. While many of the

demographic characteristics were typical of PH samples (e.g., a higher proportion of females

and those aged 50 and above) [47], participants in this study were a self-selected group of

relatively highly educated, English-speaking individuals proactively accessing support from a

PH organisation online. As such, participants' involvement in the charity may be impacting

on their coping style, as reflected by their scores on seeking emotional and instrumental

support.

The findings should be viewed in light of the knowledge that the data was collected in

the weeks following the easing of shielding (this involved people in the UK with a pre-

existing medical condition that made them more susceptible to COVID-19 (such as PH)

spend twelve weeks taking additional infection control precautions). This may have

conceivably contributed to increased levels of health-related worry and anxiety within our

sample not only for oneself but also for loved ones, and therefore a greater need to turn to

others for practical and emotional help and support.

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Conclusions

This study has provided a unique insight into the coping styles of people with PH. Four distinct coping styles emerged, which were related to measures of depression, health anxiety and HRQoL. While coping by seeking external support as well as cognitive and affirmation coping strategies had a buffering effect on the impact of health anxiety and depression on HRQoL, the tendency to use more passive ways of coping and coping by substance misuse were associated with poorer psychological outcomes. The results are consistent with previous research suggesting people with PH are at a greater risk of experiencing psychopathologies. Findings add to growing evidence base demonstrating the role of psychological factors in predicting quality of life in those with this disease. Understanding the coping styles of this sample has highlighted a range of potential therapeutic targets, supporting the use of psychological treatments aimed at cognitive and emotional processes. Given the observed importance of support systems, patients are also likely to benefit from interventions focused on the provision of information and support to caregivers, although further research is required in these areas.

Figure 1. Scree point prior to final four factor analysis

Authors' contribution

Dr Gregg H Rawlings was responsible for the conception of the study, data collection and analysis, and writing up the study for publication. He approved the final version for publication.

Prof Andrew R. Thompson was responsible for the conception of the study, provided feedback on data collection and analysis, and writing up the study for publication. He approved the final version for publication.

Dr Iain Armstrong was responsible for the conception of the study, provided feedback on data collection, and writing up the study for publication. He approved the final version for publication.

Dr Barbora Novakova was responsible for data analysis and writing up the study for publication. She approved the final version for publication.

Prof Nigel Beail was responsible for the conception of the study, provided feedback on data collection and analysis, and writing up the study for publication. He approved the final version for publication.

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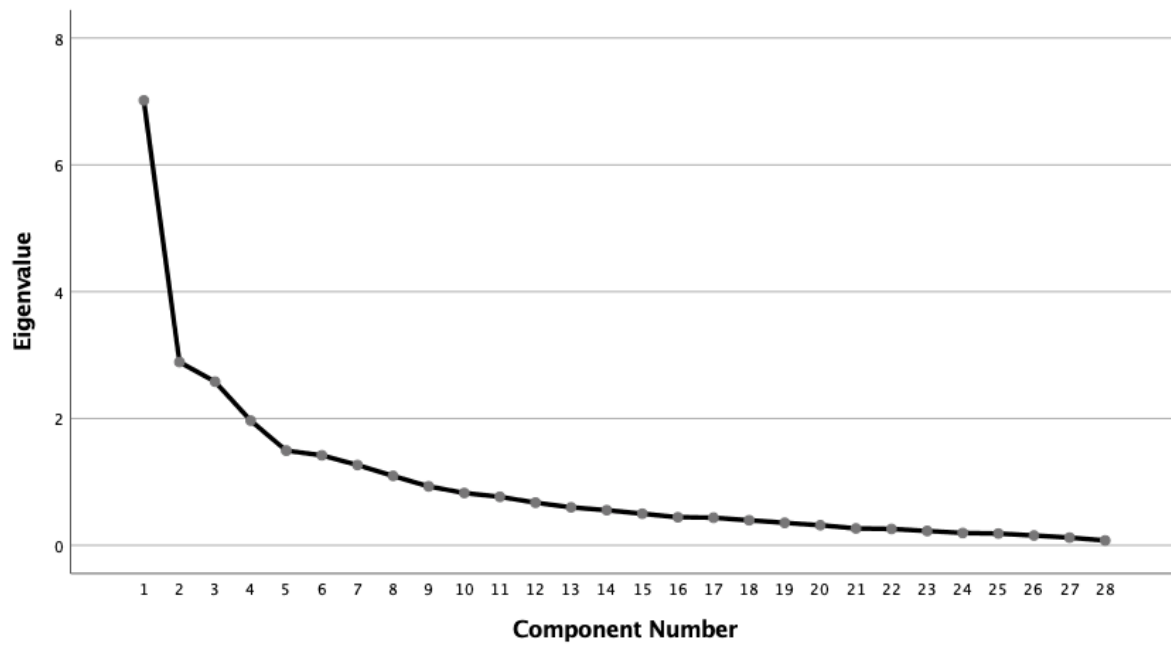43
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Figure 1. Scree plot prior to final four factor analysis of coping in pulmonary hypertension

STROBE 2007 (v4) Statement—Checklist of items that should be included in reports of *cross-sectional studies*

Section/Topic	Item #	Recommendation	Reported on page #
Title and abstract	1	(*a*) Indicate the study's design with a commonly used term in the title or the abstract	1
		(*b*) Provide in the abstract an informative and balanced summary of what was done and what was found	2-3
Introduction			
Background/rationale	2	Explain the scientific background and rationale for the investigation being reported	5-7
Objectives	3	State specific objectives, including any prespecified hypotheses	6-7
Methods			
Study design	4	Present key elements of study design early in the paper	7
Setting	5	Describe the setting, locations, and relevant dates, including periods of recruitment, exposure, follow-up, and data collection	7-8
Participants	6	(*a*) Give the eligibility criteria, and the sources and methods of selection of participants	7-8
Variables	7	Clearly define all outcomes, exposures, predictors, potential confounders, and effect modifiers. Give diagnostic criteria, if applicable	8-9
Data sources/ measurement	8*	For each variable of interest, give sources of data and details of methods of assessment (measurement). Describe comparability of assessment methods if there is more than one group	8-10
Bias	9	Describe any efforts to address potential sources of bias	7-10
Study size	10	Explain how the study size was arrived at	7
Quantitative variables	11	Explain how quantitative variables were handled in the analyses. If applicable, describe which groupings were chosen and why	10
Statistical methods	12	(*a*) Describe all statistical methods, including those used to control for confounding	10
		(*b*) Describe any methods used to examine subgroups and interactions	10
		(*c*) Explain how missing data were addressed	10
		(*d*) If applicable, describe analytical methods taking account of sampling strategy	10
		(*e*) Describe any sensitivity analyses	10
Results			

Participants	13*	(a) Report numbers of individuals at each stage of study—eg numbers potentially eligible, examined for eligibility, confirmed eligible, included in the study, completing follow-up, and analysed	11-12
		(b) Give reasons for non-participation at each stage	na
		(c) Consider use of a flow diagram	Na
Descriptive data	14*	(a) Give characteristics of study participants (eg demographic, clinical, social) and information on exposures and potential confounders	12
		(b) Indicate number of participants with missing data for each variable of interest	Na
Outcome data	15*	Report numbers of outcome events or summary measures	11-19
Main results	16	(*a*) Give unadjusted estimates and, if applicable, confounder-adjusted estimates and their precision (eg, 95% confidence interval). Make clear which confounders were adjusted for and why they were included	11-19
		(*b*) Report category boundaries when continuous variables were categorized	11-19
		(*c*) If relevant, consider translating estimates of relative risk into absolute risk for a meaningful time period	Na
Other analyses	17	Report other analyses done—eg analyses of subgroups and interactions, and sensitivity analyses	11-19
Discussion			
Key results	18	Summarise key results with reference to study objectives	19-24
Limitations	19	Discuss limitations of the study, taking into account sources of potential bias or imprecision. Discuss both direction and magnitude of any potential bias	23
Interpretation	20	Give a cautious overall interpretation of results considering objectives, limitations, multiplicity of analyses, results from similar studies, and other relevant evidence	19-24
Generalisability	21	Discuss the generalisability (external validity) of the study results	19-24
Other information			
Funding	22	Give the source of funding and the role of the funders for the present study and, if applicable, for the original study on which the present article is based	4

*Give information separately for cases and controls in case-control studies and, if applicable, for exposed and unexposed groups in cohort and cross-sectional studies.

Note: An Explanation and Elaboration article discusses each checklist item and gives methodological background and published examples of transparent reporting. The STROBE checklist is best used in conjunction with this article (freely available on the Web sites of PLoS Medicine at http://www.plosmedicine.org/, Annals of Internal Medicine at http://www.annals.org/, and Epidemiology at http://www.epidem.com/). Information on the STROBE Initiative is available at www.strobe-statement.org.