Review Article

Control and Context Are Central for People With Advanced Illness Experiencing Breathlessness: A Systematic Review and Thematic Synthesis

Natasha Lovell, MBChB, Simon N. Etkind, MB, BChir, Sabrina Bajwah, PhD, Matthew Maddocks, PhD, and Irene J. Higginson, PhD
Cicely Saunders Institute of Palliative Care, Policy and Rehabilitation, King's College London, London, United Kingdom

Abstract

Context. Breathlessness is common and distressing in advanced illness. It is a challenge to assess, with few effective treatment options. To evaluate new treatments, appropriate outcome measures that reflect the concerns of people experiencing breathlessness are needed.

Objectives. The objective of this study was to systematically review and synthesize the main concerns of people with advanced illness experiencing breathlessness to guide comprehensive clinical assessment and inform future outcome measurement in clinical practice and research.

Methods. This is a systematic review following Preferred Reporting Items for Systematic Reviews and Meta-Analyses methodology. MEDLINE (1946–2017), PsycINFO (1806–2017), and EMBASE (1974–2017), as well as key journals, gray literature, reference lists, and citation searches, identified qualitative studies exploring the concerns of people living with breathlessness. Included studies were quality-assessed using the Critical Appraisal Skills Program checklist and analyzed using thematic synthesis.

Results. We included 38 studies with 672 participants. Concerns were identified across six domains of "total" breathlessness: physical, emotional, spiritual, social, control, and context (chronic and episodic breathlessness). Four of these have been previously identified in the concept of "total dyspnea." Control and context have been newly identified as important, particularly in their influence on coping and help-seeking behavior. The importance of social participation, impact on relationships, and loss of perceived role within social and spiritual domains also emerged as being significant to individuals.

Conclusion. People with advanced illness living with breathlessness have concerns in multiple domains, supporting a concept of "total breathlessness." This adapted model can help to guide comprehensive clinical assessment and inform future outcome measurement in clinical practice and research. J Pain Symptom Manage 2019;57:140–155.

Key Words
Breathlessness, dyspnea, experience, concern, advanced disease, palliative care

Background

Although there are a number of definitions, breathlessness is usually referred to as "a subjective experience of breathing discomfort that consists of qualitatively distinct sensations that vary in intensity" as defined by the American Thoracic Society.[1] Breathlessness that persists despite optimal treatment of the underlying pathophysiology and results in disability for the patient is often referred to as chronic breathlessness as agreed by recent Delphi consensus.[2]

Address correspondence to: Natasha Lovell, MBChB, Cicely Saunders Institute of Palliative Care, Policy and Rehabilitation, King's College London, Bessemer Road, London SE5 9PJ, United Kingdom. E-mail: natasha.lovell@kcl.ac.uk

Accepted for publication: September 26, 2018.

0885-3924/$ - see front matter
https://doi.org/10.1016/j.jpainsymman.2018.09.021

Chronic breathlessness may be accompanied by episodic breathlessness, defined as a severe worsening of breathlessness intensity or unpleasantness beyond usual fluctuations in the patient's perception.[3]

Chronic breathlessness is common and distressing, affecting almost all people living with chronic obstructive pulmonary disease (COPD) and interstitial lung disease (ILD), and most people with chronic heart failure and advanced cancer.[4–6] Current treatment options for chronic breathlessness are limited, and its management remains an important clinical challenge. While clinicians often recognize the significant impact for patients, time constraints, and inadequate resources and training, they feel ill-equipped in assessment and management.[7] Furthermore, to establish the clinical effectiveness of new treatments, appropriate outcome measures that capture the concerns and problems that matter to patients are vital. Although over 30 different outcome measures have been validated for breathlessness, consensus is lacking on which measure to use and when.[8,9]

An improved understanding of the main concerns for people experiencing breathlessness can provide clinicians with a framework for assessment and guide the choice of appropriate outcome measures in clinical practice and research. It may also identify potential targets for new treatments. The American Thoracic Society provides a helpful framework to guide outcome measure selection and proposes three domains of breathlessness: "sensory-perceptual experience"; incorporating what breathing feels like to the patient; "affective distress" that may include the unpleasantness experienced during breathing; and "symptom impact or burden" that might include functional ability or health-related quality of life.[1] Individual qualitative studies have explored what it is like for people to live with breathlessness, including the experience of care and impact of illness, and a recent systematic review considers the role of coping, help-seeking behavior, and clinician responsiveness.[10] In this review, Hutchinson describes the concept of breathing space and highlights the importance of clinician response in determining future coping and help-seeking behavior.[10]

However, there has been no attempt to systematically synthesize the concerns for people experiencing breathlessness with the aim of informing outcome measurement in clinical practice and research. Our systematic review aims to determine the main concerns for people with advanced illness experiencing breathlessness, to guide comprehensive clinical assessment, optimize clinical interactions, and inform future outcome measurement in clinical practice and research.

Methods

Study Design

We conducted a systematic literature review and thematic synthesis in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses guidelines.[11]

Information Sources and Search Strategy

We searched titles, abstracts, and keywords of articles indexed within three databases: PsycINFO (1806 to March Week 4 2017), MEDLINE (1946 to March Week 5 2017), and EMBASE (1974 to 2017 Week 14). Search terms were developed and piloted to ensure inclusivity and included a combination of the following terms: dypsn* OR short* of breath OR breathless* AND experience* OR concern* OR expect* OR prefer* AND palliate* OR chronic lung disease OR advanced disease. The full electronic search strategy is shown in Appendix 1. Key journals, gray literature, reference lists, and forward citation searches identified additional relevant articles.

Inclusion and Exclusion Criteria

Inclusion criteria were as follows: Primary qualitative or mixed-method studies, from any setting (hospital, community, or outpatient), which explored the concerns of adults experiencing breathlessness and living with advanced illness (including but not limited to COPD, ILD, chronic heart failure, and cancer), were eligible for inclusion.

Exclusion criteria were as follows: Studies presenting only quantitative data, published in a language other than English, and where patient concerns were described in relation to their illness experience, and not explicitly breathlessness, were excluded.

Procedures for Study Selection and Data Extraction

Articles were initially screened by title and abstract. All full-text articles were assessed against the eligibility criteria by one researcher (N. L.), with 25% reviewed by another researcher. Any disagreements about inclusion of articles were resolved by discussion within the author team. Data were extracted using a *pro forma* on study setting, participants, and qualitative approach. All included articles were assessed against the Critical Appraisal Skills Program (CASP) qualitative research checklist. The CASP checklist is a recognized tool developed and piloted by a group of experts and includes 10 brief questions relating to methodological rigor, credibility, and relevance. The CASP checklist is suited to systematic reviews of qualitative research and was considered an appropriate choice for this review.[12,13]

All subsections of text relating to breathlessness in the "results" or "findings" sections of included articles were extracted and imported verbatim into NVivo 11 qualitative data software (Version 11, 2015; QSR International Pty Ltd.). Extracted text included both direct quotations but also the authors' interpretation of findings.

Analysis

Data were collated and analyzed using thematic synthesis.[14] This involved three stages: 1) coding of text "line-by-line" to enable the translation of concepts from one study to another; 2) development of "descriptive themes"; and 3) the generation of "analytical themes." This process enabled the data to be considered in relation to the specific research question of this review and allowed for interpretation beyond what was been presented in the primary articles. During the course of the review, we also collated information on models of breathlessness and used this to structure the results.

Results

The search identified 5082 individual articles, of which 69 full-text articles were assessed against inclusion criteria and 38 separate articles were included

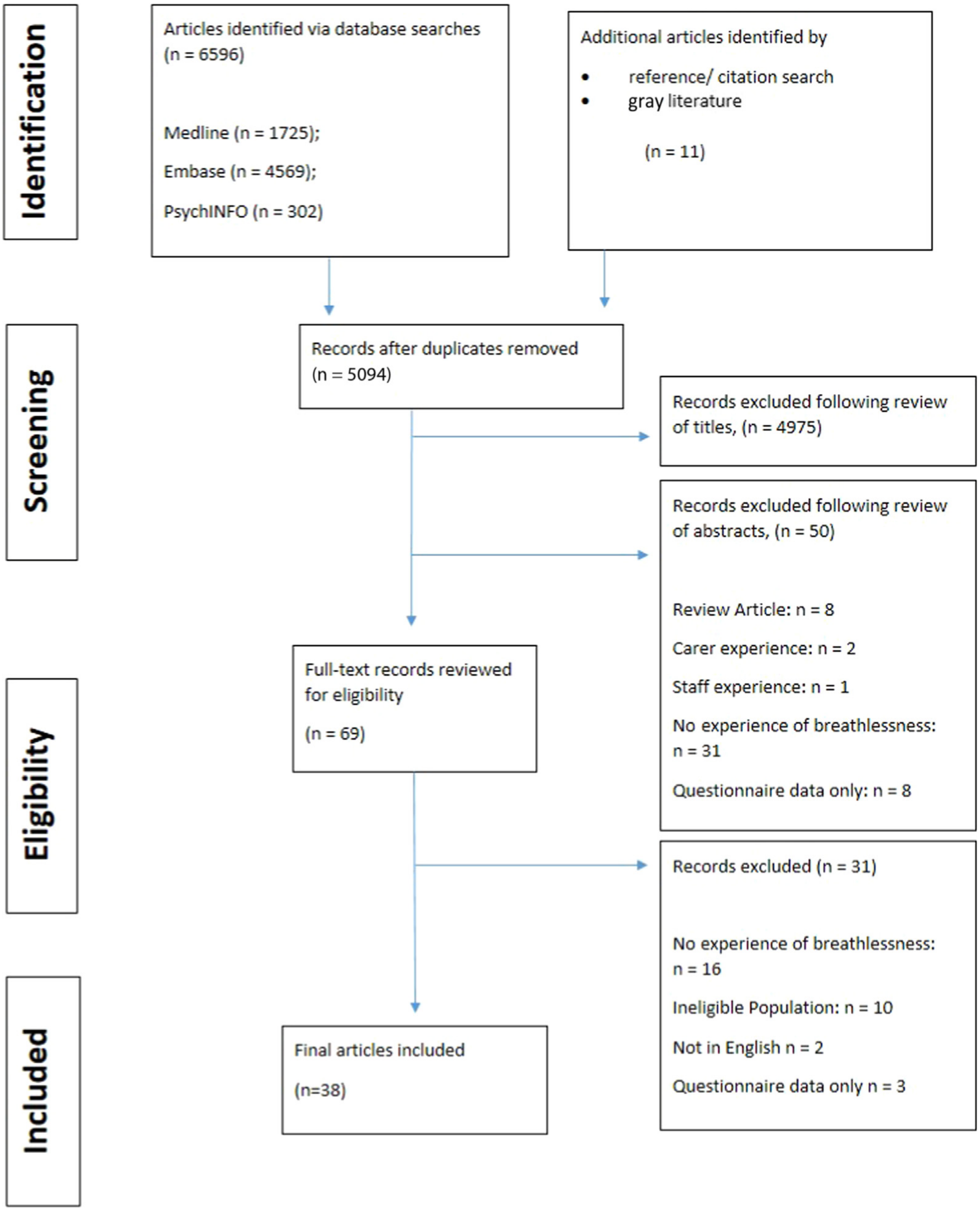

Fig. 1. PRISMA chart. PRISMA = Preferred Reporting Items for Systematic Reviews and Meta-Analyses.

(Fig. 1). A total of 672 participants were included across all studies. The majority of studies were conducted in the U.K. ($n = 24/38$) and in the outpatient or community setting ($n = 30/38$). Twenty-four studies included participants with a diagnosis of COPD; four with cancer; two with chronic heart failure; two with interstitial lung disease; and six studies focused on the symptom not the disease, including participants across different disease groups.

Articles were published between 1993 and 2017. The method of data collection was varied although a semistructured interview format was most common ($n = 28/38$), with the remaining studies following a narrative or unstructured approach. Approaches to data analysis included thematic analysis, hermeneutic analysis, grounded theory, framework analysis, and phenomenological analysis. See Table 1 for full characteristics of included studies. Thirty-two of the included articles met at least nine of the 10 CASP checklist criteria when assessed, and none scored less than seven (see Appendix 2).

Synthesis and Model

While conducting the synthesis, we considered how concerns mapped onto existing models of breathlessness, and chose "total dyspnea" as our conceptual framework.[31,53] This model forms the basis for the presentation of synthesis results (Fig. 2). It is based on the concept of "total pain" described by Dame Cicely Saunders, comprising four domains: 1) physical, including subsequent effects on function; 2) psychological concerns; 3) social impact; and 4) spiritual distress.

In our model of "total breathlessness," psychological has been changed to emotional to reflect the synthesis findings, and "control" and "context" have been added for completeness. While concerns can be described within a single domain, most are not exclusive to one, and there is a considerable overlap between domains as demonstrated in our model of "total breathlessness."

Participants described how living with breathlessness pervaded every part of their life. The main concerns for people living with breathlessness were comprehensive and wide ranging, and the negative effects of breathlessness on quality of life were evident throughout the data. The experience of breathlessness was entirely unique to the individual and impacted not only them but also those around them. Breathlessness was also described as "invisible," disguised at rest,[29] or hidden due to embarrassment and stigma, as well as a perceived lack of interest and response from clinicians. Participants responded by retreating and not seeking help, and as a result, this distressing symptom remained unacknowledged and undertreated.

Physical Symptom of Breathlessness and Its Effects on Function

Breathlessness was described as affecting all activities of daily living. Breathlessness crept up on individuals and while initially noticed during more strenuous tasks, it soon prevented activities around the home such as cooking and cleaning. As their illness progressed, participants experienced further functional decline and became unable to climb the stairs or walk to the shops, resulting in social isolation. This trajectory of deteriorating physical function affected not only the person but also those around them. Participants became dependent on friends and family to assist with intimate tasks such as washing and dressing.

> … The worst thing I think is the stairs, going up and down the stairs. Ordinary household chores I find difficult. Very restrictive, because of your breathing. And now of late … even simple things like having a shower and getting dressed.
>
> Patient with COPD, Caress et al. 2010.

Participants recognized that they were becoming increasingly breathless, felt frustrated that this caused them to slow down, and meant they were unable to do the things they had previously done. Breathlessness had taken away their independence and left them feeling concerned about the future. Participants felt they were becoming an increasing burden to friends and family.

> … It (breathlessness) is over time slowing down everything.
>
> Female with COPD, Gysels et al. 2011.

Emotional

Participants described a cycle between breathlessness and anxiety, relating not only to acute episodes of breathlessness, but also within the wider context of a person's life affected by chronic breathlessness. The emotional impact of breathlessness extended beyond the individual to those people in a caring role, who were often left feeling helpless and powerless when breathlessness occurred.

> It [shortness of breath] would just take my breath away and just like somebody would grab me and start choking. I couldn't breathe and then when it happened my daughter would take me to emergency.
>
> Patient with Heart Failure, Lowey 2012.

The combination of feeling breathless and not having control over breathing created a frightening scenario. In some cases, it became so severe that the person experiencing it felt it may not resolve

> When you get shortness of breath, you're scared… scared you're gonna take your last breath.
>
> Male with COPD, Leidy et al. 1999.

Table 1

Characteristics of Included Studies

Author	Setting	Participants	Qualitative Approach	Aim of Study	Main Concerns Identified Within Study
Apps et al.[15]	Community setting in U.K.	Fifteen individuals with COPD, six male (aged between 65 and 82 yrs), nine female (aged between 55 and 82 yrs)	Semistructured interviews analyzed through thematic analysis	To understand the self-care experiences of patients with COPD who are primarily managed in primary care, and to examine the challenges of engaging in such behaviors.	Disruption of daily tasks, patients unsure about progression and the future
Bajwah et al.[16]	Outpatient and community setting in U.K.	Eight individuals with a diagnosis of NSIP, IPF, and IIP (five males aged between 65 and 81 yrs, three females aged between 56 and 75 yrs), four informal caregivers (three females aged between 41 and 63 yrs, one male aged 63 yrs), and six health professionals	Semistructured interviews analyzed through thematic analysis	To explore the specialist palliative care needs of people living with end-stage progressive idiopathic fibrotic interstitial lung disease.	Uncontrolled symptom, limitation on social activities, increased reliance on others
Barnett[17]	Outpatient setting in U.K.	Ten individuals with COPD (no other demographic information given)	Unstructured interviews analyzed through hermeneutic analysis	To explore the experience of living with chronic obstructive pulmonary disease by investigating the subjective phenomenon as described by the patient.	Impact on daily activities, feeling frightened, reduced ability to socialize and enjoy a normal life
Booth et al.[18]	Outpatient setting in U.K.	Ten individuals with COPD (six male, four female, aged between 51 and 80 yrs), 10 individuals with cancer (six males, four females aged between 51 and 77 yrs)	Semistructured interviews analyzed through thematic analysis	To investigate the experience of living with breathlessness in those suffering from advanced cancer or COPD and their carers, both to allow comparison of these potentially differing perspectives and to provide insights for future development of both clinical services and research.	Lack of warning signs, overwhelmed by fear, uncertainty and anxiety, increasingly limited mobility, unable to do activities of daily living or socialize, role reversal and loss
Caress et al.[19]	Outpatient setting in U.K.	Fourteen individuals with COPD, eight males, six females, mean age 68 yrs, range 60–80	Semistructured interviews analyzed through thematic analysis	To generate in-depth insights into patients' and family members' understanding of the causation, progression and prevention of chronic obstructive pulmonary disease, and the role of health promotion with this population.	Patients felt frightened and overwhelmed, mobility was affected because of fear of becoming dyspneic, resulting in feelings of helplessness, withdrawal from activities
Clancy et al.[20]	Community setting in U.K.	Nine individuals with COPD, six males, three females, age range 57–78 yrs	Focused-conversation style interviews analyzed through thematic analysis using hermeneutic phenomenological reflection	To explore the existential experiences of 10 patients with chronic obstructive pulmonary disease (COPD) who had been prescribed long-term oxygen therapy (LTOT) and their carers.	Reduced mobility, fear of losing breath, fear of the future, fear of not being in control

Cooney et al.[21]	Community setting in U.K.	Twenty-six individuals with COPD, 15 males (aged between 52 and 86 yrs), 11 females (aged between 49 and 79 yrs)	Semistructured interviews analyzed through grounded theory	Aims to understand the meaning of COPD for people and their response to this disease.	Challenge of day-to-day activities, struggling to live a normal life
Duck et al.[22]	Outpatient setting in U.K.	Seventeen individuals with IPF (seven males, 10 females), median age 67 yrs	Semistructured interviews analyzed through framework analysis	To understand the perceptions, needs, and experiences of patients with idiopathic pulmonary fibrosis.	Loss of independence, change of roles within relationships, increasing dependence
Dunger et al.[23]	Hospital, outpatient, and community setting in U.K.	Eight individuals with cancer (five males, three females, median age 67 yrs), 10 individuals with COPD (four male, six females, median age 66.5 yrs)	Semistructured interviews analyzed through framework analysis	To explore and contrast the experience and meaning of breathlessness in patients with chronic obstructive pulmonary disease (COPD) or lung cancer at the end of life.	Patients adapted and avoided triggers of breathlessness (e.g., physical activity), patients described a loss of independence, and fear of death
Edmonds et al.[24]	Hospital and outpatient setting in U.K.	Twenty-seven individuals with heart failure (20 males, seven females, aged between 38 and 94 yrs [mean 69])	Semistructured interviews analyzed through thematic analysis	To explore patient experience of breathlessness in heart failure.	Uncontrollable breathlessness resulted in a reduction in the number or intensity of activities
Ek et al.[25]	Outpatient setting in Sweden	Four individuals with COPD (one male aged 70 yrs, three females age range 66–75 yrs)	Seventeen interviews (four to five with each participant), 15 phone calls and field notes analyzed through hermeneutic analysis	To illuminate the meaning of living with advanced COPD and LTOT when living alone.	Concerns about self-image, not able to be spontaneous, thoughts about future, fear of dying
Fraser et al.[26]	Outpatient setting in U.S.	Ten individuals with COPD (five males, five females, age range 59–86 yrs (mean 71))	Semistructured interviews analyzed through hermeneutic phenomenology design	To explore the experiences of older adults with severe COPD to gain an understanding of how the disease had affected them and the ways in which they integrated the illness into their lives.	Loss of function, difficulty maintaining a sense of normality, concerns about the future
Gardiner et al.[27]	Community setting in U.K.	Twenty-one individuals with COPD (13 males, eight females, mean age 70.3 yrs)	Semistructured interviews (12 face-to-face, nine telephone), analyzed through thematic analysis	To determine prospectively the needs of patients in the advanced stages of COPD.	Concerns about the future, fear of dying from breathlessness
Gullick et al.[28]	Outpatient setting in Australia	Fifteen individuals with COPD (nine males, six females, age range 55–77 yrs), 14 close family members	Semistructured interviews (18 face-to-face, 40 telephone), analyzed through hermeneutic interpretation	To explore the experience of the person who lives within a body with emphysema—a form of COPD.	Limited ability to mobilize, self-care, or engage with social activities and hobbies, concerns about visibility of breathlessness to others
Gysels et al.[29]	Hospital and community setting in U.K.	Fourteen individuals with COPD in hospital setting (five males, nine females, median age 69 yrs), four individuals with COPD in community (two males, two females, median age 70 yrs)	Semistructured interviews analyzed through grounded theory	To explore the reasons for the disparity between the high needs and the low service use typically reported for breathless patients with COPD and their carers.	Restrictions due to breathlessness requiring adaptations, patients attempted to hide by avoiding contact with the outside world
Gysels et al.[30]	Outpatient setting in U.K.	Ten individuals with COPD (six females, four males, age range 42–78 yrs), six individuals with ILD (three females, three	Semistructured interviews analyzed through thematic analysis	To analyze what constitutes dignity for people suffering from refractory breathlessness with advanced disease, and its	Effect on function and independence limiting every activity, uncertainty regarding future, fear of dying

(*Continued*)

Table 1

Continued

Author	Setting	Participants	Qualitative Approach	Aim of Study	Main Concerns Identified Within Study
		males, age range 72—84 yrs), four individuals with cancer (all males, age range 63—77 yrs)		implications for the concept of dignity.	
Gysels et al.[31]	Hospital, outpatient, and community setting in U.K.	Ten individuals with cancer (five males, five females, age range 52—84 yrs), 14 individuals with COPD (five males, nine females, age range 52—78 yrs), 10 individuals with heart failure (seven males, three females, age range 61—80 yrs), and 10 individuals with MND (nine males, one female, age range 24—77 yrs)	Semistructured interviews analyzed through thematic analysis	To explore and compare the lived experience of breathlessness for patients with four conditions—COPD, heart failure, cancer, and MND	Impaired mobility, increasing dependence, anxiety restricting patients to home, worries about the future
Habraken et al.[32]	Outpatient setting in The Netherlands	11 individuals with COPD, eight males, three females, age range 61—83 yrs	Semistructured interviews analyzed through thematic analysis	To gain insight into why patients with end-stage COPD tend not to express a wish for help.	Fear of not being in control, fear of suffocating and dying
Hallas et al.[33]	Outpatient setting in U.K.	Twelve individuals with chronic respiratory disease (five males, seven females, age range 21—58 yrs)	Semistructured interviews analyzed through interpretative phenomenological analysis	To identify the complex cognitive system of beliefs, appraisals, and perceptions that underpinned patients' experiences of breathlessness and their relationship to the development and maintenance of panic.	Consequences of breathlessness, ability to manage panic, emotional isolation, adjusting to lifestyle changes, avoidance of activities
Harris et al.[34]	Community setting in U.K.	Sixteen individuals with COPD (12 males, four females, mean age 66.8 yrs)	Semistructured interviews analyzed through grounded theory	To identify a strategy for improving the uptake of pulmonary rehabilitation.	Impact on activities of daily living, unable to do domestic tasks or leisure pursuits, requirement to adapt
Hasson et al.[35]	Community setting in Northern Ireland	Thirteen individuals with COPD (10 males, three females, median age 65 yrs)	Semistructured interviews analyzed through thematic analysis	To explore the potential for palliative care among people living with advanced chronic obstructive pulmonary disease (COPD)	Restriction of day-to-day activities, fear that breathlessness could lead to death, concerns about carers
Henoch et al.[36]	Outpatient setting in Sweden	20 individuals with cancer (11 males, nine females, median age 68.5 yrs, range 56—79)	Semistructured interviews analyzed through thematic analysis	To describe lung cancer patients' experience of dyspnea and their strategies for managing dyspnea.	Physical limitations, increased dependence, psychological impact
Jones et al.[37]	Community setting in U.K.	Sixteen individuals with COPD, eight males, eight females, age range 62—83 yrs	Semistructured interviews analyzed through thematic analysis	To determine the needs of patients dying in primary care from chronic obstructive pulmonary disease	Lack of mobility resulting in difficulties around the house and with social contact, fear of dying
Jonsdottir[38]	Outpatient setting in Iceland	Ten individuals with COPD, six males, four females, mean age 61 yrs	In-depth interviews analyzed according to Newman's hermeneutic dialectic method	To explore the life patterns of people with COPD	Loss of control, impact on mobility, isolation, impact on ability to work
Jørgensen et al.[39]	Hospital setting in U.K.	Twelve individuals with COPD (six males [age range 45—79 yrs], six females [age range 64—80 yrs])	Video-based narrative analyzed through grounded theory	To explore how people with moderate to most severe COPD predominantly cope with breathlessness during daily living.	Concerns and anxiety about triggering breathlessness, meaning that activities need to be carefully regulated, feeling of defeat

Lai et al.[40]	Inpatient palliative care unit in Hong Kong	Eleven individuals with lung cancer (eight males, three females, age range 54–75 yrs)	Semistructured interviews analyzed through thematic analysis	To describe the experience of dyspnea and helpful interventions in Chinese patients with advanced lung cancer admitted in the palliative care unit in one region in Hong Kong	Loss of control, impact on activities of daily living, not able to do as much physically, negative perception of self, loss of independence, isolation
Leidy et al.[41]	Outpatient setting in U.S.	Twelve individuals with COPD, six males, six females, mean age 66.8 yrs, range 50–76	In-depth interviews analyzed according to Colaizzi's phenomenological method and consensus dialogue approach	To describe the meaning of functional performance from the perspective of patients themselves	Loss of control, fear of dying
Lowey[42]	Community setting in U.S.	Twenty individuals with heart failure or COPD (nine males, 11 females, mean age 73 yrs, age range 52–93 yrs)	Semistructured interviews analyzed through thematic analysis	Describe the care preferences of individuals living with advanced cardiac and respiratory illnesses about their current and future health, understanding about options for care at the end of life, and expectations from health care providers	Lack of control, unpredictable, fear of breathlessness and the consequences, unable to do hobbies
Luthy et al.[43]	Hospital setting in Switzerland	Thirty-two individuals with COPD, 19 males, 13 females, mean age 64.2 yrs	Interview including drawing task and comments analyzed through thematic analysis	To explore the perception of dyspnea in patients with severe chronic obstructive pulmonary disease	Fear of dying, no control over breathlessness
Nicholls[44]	Outpatient setting in New Zealand	Ten individuals with chronic respiratory disease, five males, five females, age range 60–78 yrs	Narrative interviews analyzed through thematic analysis	To explore how a person's personality or "self" affected the way his or her chronic breathlessness was expressed, and conversely, how the experience of breathlessness affected one's notions of "self"	Unpredictability of breathlessness, fear of dying, loss of independence, loss of normality, unable to do social activities
O'Driscoll et al.[45]	Outpatient setting in U.K.	Fifty-two individuals with lung cancer, 30 males, 22 females, mean age 60 yrs, age range 33–76 yrs	Assessment notes recorded by nurses during conversations with patients analyzed through thematic analysis	Understand the experience of breathlessness and the restrictions it imposes on daily life.	Fear of dying, unable to do activities around the house, restrictions on personal life, disruptions to social activities, unable to fulfill role in society
Oliver[46]	Hospital and community setting in U.K.	Sixteen individuals with COPD, 12 males, four females, mean age 65 yrs, age range 59–75 yrs	Semistructured interviews analyzed through thematic analysis	To explore the perceptions and needs of chronic obstructive pulmonary disease (COPD) patients	Unpredictable, fear of not being able to breathe, fear of dying, loss of self-esteem
Roberts et al.[47]	Community setting in Canada	Ten individuals with cancer, four males, six females, age range 62–80 yrs	Semistructured interviews analyzed through thematic analysis	Understand how patients and nurses interpret nature and meaning of breathlessness	Fear of dying, restriction on daily activities
Robinson[48]	Community setting in U.K.	Ten individuals with COPD, six males, four females, mean age 65.4 yrs, age range 51–74 yrs	Narrative interviews analyzed through a mind map	To describe the experience of living with severe oxygen-dependent chronic obstructive pulmonary disease (COPD)	Impact on physical activities including personal care, fear of breathlessness
Victorson et al.[49]	Outpatient setting in U.S.	Fifteen individuals with COPD, 12 males, three females, mean age 81 yrs, age range 72–92 yrs	Semistructured interviews analyzed through grounded theory	Identify important patient-reported concepts of dyspnea and associated activities to develop a dyspnea-specific conceptual model for chronic obstructive pulmonary disease (COPD).	Impact on physical activities, unable to do activities around the house, fear of not being able to breathe

(*Continued*)

Table 1
Continued

Author	Setting	Participants	Qualitative Approach	Aim of Study	Main Concerns Identified Within Study
Walthall et al.[50]	Outpatient setting in U.K.	Twenty-five individuals with heart failure (15 males, 10 females, mean age 72.66 yrs, age range 53–86 yrs)	Semistructured interviews analyzed through thematic analysis	To explore how patients with chronic heart failure describe their experiences of breathlessness, how daily life is affected and how they adjust to and manage these symptoms.	Impact on ability to undertake activities of daily living, fear of dying, feeling isolated and lonely, unable to fulfill societal role or undertake social activities
Williams et al.[51]	Outpatient and community setting in U.K.	Six individuals with COPD, four males, two females, age range 64–83 yrs	Semistructured interviews analyzed through thematic analysis	To explore what is most important to people living with COPD.	Impact on physical ability and mobility, therefore making it difficult to leave the house
Wortz et al.[52]	Outpatient setting in U.S.	Forty-seven individuals with COPD, 25 males, 22 females, mean age 68.4 yrs	Semistructured interviews analyzed through thematic analysis	To address gaps in the literature on self-management support by examining patients' responses to questions about goals, needs, and expectations regarding self-management using qualitative methods in a broadly representative sample of patients with moderate to severe COPD	Fear of dying, impact on mobility

COPD = chronic obstructive pulmonary disease; NSIP = nonspecific interstitial pneumonia; IPF = idiopathic pulmonary fibrosis; MND = motor neurone disease.

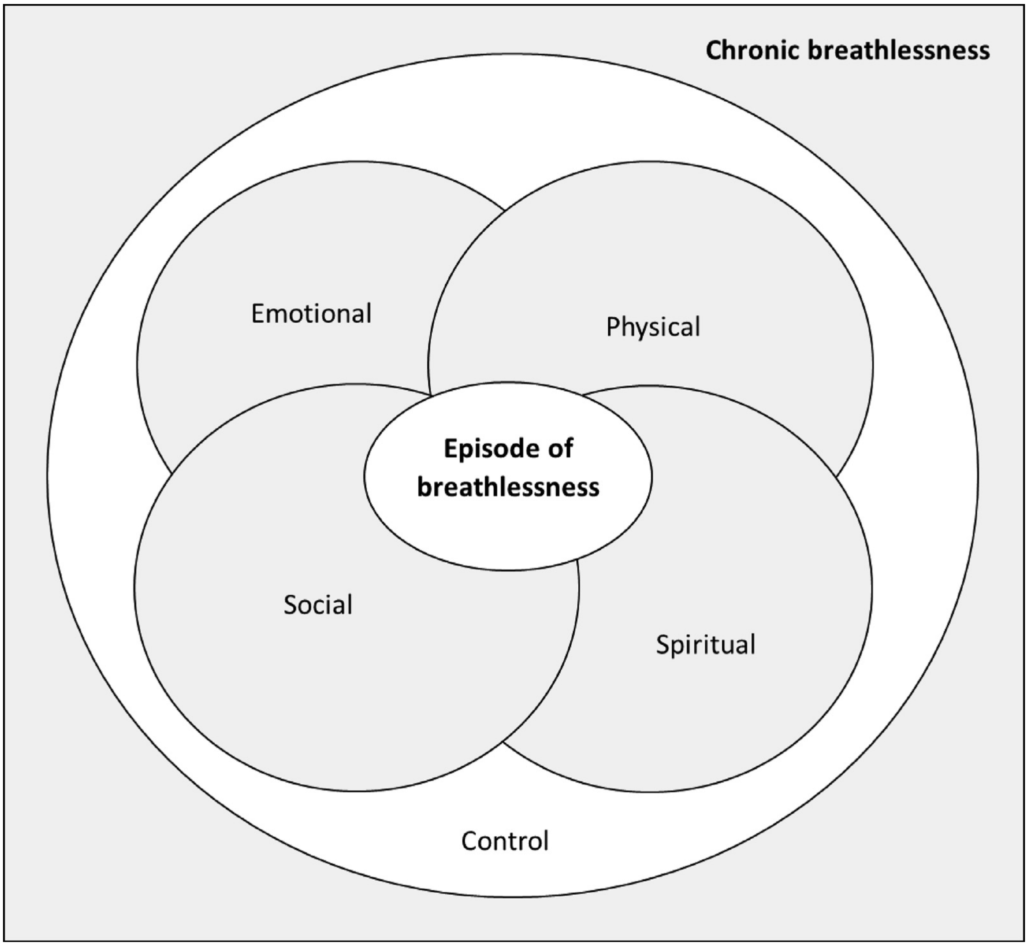

Fig. 2. Total breathlessness. Adapted from Abernethy and Wheeler (2008).

Participants described feeling vulnerable and perceived that an episode of breathlessness might become life threatening and result in death, often describing the sensation of drowning or suffocating.

> I had extreme shortness of breath and I felt I couldn't breathe at all. I felt rather as if I was drowning. I really thought that I was not, you know, going to survive without some form of treatment …
>
> Male with Heart Failure, Walthall et al. 2017.

Fear was also described in the wider context of an individual's life and included fear of triggering an episode of breathlessness, fear of deterioration, and fear of the future.

> It's just stops your life, stops you from living.
>
> Patient with COPD, Caress et al. 2010.

Fear of triggering breathlessness led to activity withdrawal, and participants were less likely to plan activities outside of the home resulting in social isolation.

> I'd like to go for walks but I can't. I just get too breathless. When I sit in a chair I'm fine. In the last six months I've only been out once.
>
> Patient with COPD, Barnett et al. 2005.

The consequences of activity avoidance have been described in the literature as the "downward spiral of disease" whereby breathlessness leads to inactivity and subsequent muscle deconditioning, therefore increasing ventilation and respiratory drive.[54] The vicious circle of dyspnea inactivity is a conceptual model recently developed and validated to explain the clinical course of COPD and emphasizes the importance of exercise capacity and exacerbations as drivers.[55]

Social

Breathlessness affected people's social lives, often excluding them from activities they previously enjoyed. The lost opportunity for routine social interactions was significant to the individual yet often unreported due to a lack of perceived relevance by health care professions. To provide high-quality care, health care professionals need to adopt a person-centered approach and invest the necessary time to understand an individual's values and priorities, in so doing ensuring that patients feel their concerns are valid and important.

> If I went to a party people would say: "Oh come on, let's dance, its New Year's Eve," and I'd say, "No I don't want to dance," they'd say, "Oh come on, don't be so boring," coz I used to love dancing. I said: "No it's not worth that, we'll dance for five minutes and I'll be sitting down for the next hour, so I'd rather just sit down and watch you dancing." So it's gradually, little things were in my head but not enough to go to the doctor. You can't go to the doctor and say "I can't dance." It's a strange thing to say to the doctor.
>
> Patient with COPD, Gysels and Higginson, 2008.

Breathlessness also had impact significantly on relationships, both physically and emotionally. Physically, living with breathlessness resulted in reliance on family or friends to support and assist with activities of daily living, and as a consequence, participants worried about being a burden to those acting in a caring role.

> I have to rely on my family to cook for me. I don't have the ability to do so but I really don't wish to impose too much on them.
>
> Patient with Lung Cancer, Lai et al. 2007.

Furthermore, illness and increasing dependence often necessitated changes within relationships, for example, between a husband and wife where one becomes the patient and the other the carer. These changes were often significant and could include a loss of intimacy and difficulties maintaining a sexual relationship.

> It has an awful effect on my life. Making love, I can't make love very often because I can't breathe.

Changes in relationships also occurred within the wider family, sometimes associated with a loss of role or inability to maintain a previously assumed responsibility, for example, as a mother.

> I can't walk. [I can't] go running with the kids or play with them because I'm out of breath after 5 minutes. They're only young and they keep saying 'Will you play table tennis with me mammy?' 'I'm not able to, honey.' I'm drained all the time.
>
> Female with COPD, Cooney et al. 2013.

Spiritual

Spiritual distress relating to breathlessness included references to self-identify, sense of purpose, and connection to others. Participants described how living with breathlessness had altered their perception of self, resulting in a feeling of loss. Not being able to fulfill family roles and responsibilities that were once enjoyed left participants lacking in purpose, and they found it difficult to maintain a sense of identity.

> I try not to let it be seen… it's this role reversal which I find very hard… once a Mother Hen always a Mother Hen. Do you ever see the chicks look after the hen? No, you don't. And I haven't been able to do it.
>
> Female with Cancer, Booth et al. 2006.

Participants also described the impact of breathlessness on their perceived ability to live as a "normal person." Low self-worth resulted in a loss of confidence, and participants questioned their ability to cope with everyday tasks and challenges.

> It has changed my life considerably because I hate seeing someone come in … I just feel so useless and helpless … when you go to do something and you realise you can't do it and you mustn't do it … I feel mainly frustrated and disappointed. It's mainly the lack of the normal life I suppose and not being able to do, looking after myself properly and the housework and the cooking.
>
> Female with Heart Failure, Walthall et al. 2017.

Control

The impact of control was evident across domains, both in relation to acute episodes of breathlessness and within the wider context of a person's life affected by chronic breathlessness. Participants described a loss of control over breathing during an episode of breathlessness. The sensation of losing control left participants feeling vulnerable and overwhelmed, powerless to the sensation of breathlessness, and some participants questioned whether they would be able to regain control.

> It is nasty, it is an unpleasant feeling. It is something I did not have control over.
>
> Female with COPD, Ek et al. 2011.

The unpredictability of breathlessness also created a sense of loss of control in other aspects of participant's lives. Living with breathlessness not only prevented one from being spontaneous but also made it difficult to plan; participants did not know how they were going to feel next week, or tomorrow, or even in a few hours' time, reflecting the uncertainty of the symptom. Participants described feeling exposed and helpless, as though the breathlessness was controlling them. It interrupted the daily pattern of their lives and was exhausting.

> It just starts all of a sudden, and you just never know when it's coming on.
>
> Patient with COPD, Fraser et al. 2006.

> It [shortness of breath] always just all of a sudden, it just comes on.
>
> Female with COPD, Lowey et al. 2012.

Context of Concerns (Acute Episode and Chronic Breathlessness)

The context in which breathlessness occurs emerged as important, with concerns either relating directly to an episode of breathlessness or more broadly as a consequence of chronic breathlessness. Context influences how an individual experiences and responds to breathlessness and is important for health care professionals to consider when agreeing management strategies. In our model, this is presented as concerns within an "episode of breathlessness," compared to concerns due to "chronic breathlessness."

During an episode of breathlessness, participants described an immediate feeling of fear, with concerns relating to a loss of control.

> When I am out of air everything gets out of order.
>
> Patient with COPD, Jonsdottir 1998.

However, participants also described fear and control in the wider context of chronic breathlessness, including fear of triggering breathlessness, fear of the future, and the concept that breathlessness was taking control over their life as a whole.

> It [dyspnea] controls me. It controls my life.
>
> Patient with Lung Cancer, Lai et al. 2007.

Similarly, concerns relating to the physical impact of breathlessness are described within an episode and also in the wider context as a consequence of chronic breathlessness. Participants described how they may have to pause mid activity (e.g., walking) to enable an episode of breathlessness to resolve in the short term.

> I have to sit down and take a proper break to make my body work again, and that is not easy.
>
> Patient with Lung Cancer, Henoch et al. 2008.

The longer term physical impact included avoidance of activities resulting in physical deconditioning and social isolation.

> I don't walk much now because I know that it would [make me breathless] I avoid anything that would.
>
> Patient with COPD, Cooney et al. 2013.

Many of the concerns identified within the social and spiritual domains extended and impacted far beyond an episode of breathlessness. Concerns included changes to role both as an individual but also within society, strain on relationships, and a loss of perceived purpose in life as a whole. These wider concerns are significant and important to people experiencing breathlessness yet remain less commonly assessed in routine clinical practice.

> It's just stops your life, stops you from living.
>
> Patient with COPD, Caress et al. 2010.

Discussion

Although earlier work has highlighted the experience of living with breathlessness,[10,56] this is to our knowledge the first attempt to systematically identify and synthesize the main concerns for people with advanced illness experiencing breathlessness, to guide the choice of outcome measures in clinical practice and research. We consider these concerns within a model of "total breathlessness" that incorporates six domains: 1) the physical symptoms of breathlessness and subsequent effect on function; 2) emotional features; 3) the spiritual distress experienced; 4) the social impact of breathlessness; 5) concerns relating to aspects of control; and 6) the context of breathlessness (acute episode or chronic). The main concerns identified are complex and multifaceted and commonly impact across more than one domain, making breathlessness challenging to assess, measure, and research. Results from this synthesis highlight just how broad and extensive the main concerns for people living with breathlessness are, extending far beyond a single episode of breathlessness, encompassing multiple domains and impacting significantly on those around them.

Two additional domains—control and context—were required to fully encompass the findings of this qualitative synthesis and produce our model of "total breathlessness." Participants described the importance of control during an immediate episode of breathlessness. A lack of control left them feeling frightened and vulnerable and often resulted in crisis help-seeking. Control was also described in the wider context of a person's life affected by chronic breathlessness, the impact often resulting in disengaged coping for individuals. Participants described how the uncertainty and unpredictability of breathlessness meant they felt unable to make definitive plans, or be spontaneous. The concept of control was a recurring theme across domains within the "total breathlessness" model, and owing to its prominence within this synthesis, we decided it should be considered as a domain in its own right.

We also added context as a domain of "total breathlessness." Participants described concerns relating to the immediate episode of breathlessness and also in the wider context of chronic breathlessness. Existing literature shows that patients with advanced disease experience distinguishable types and patterns of episodic breathlessness relating to different contextual triggers.[57] Recent qualitative work has shown that unpredictable episodes are experienced as unpleasant with a higher intensity when compared to predictable episodes.[58] The context of a concern is important and can shape an individual's response including how they cope and seek help as a consequence. The way a person copes and seeks help during an unpredictable episode of breathlessness is likely to be different to how they cope and seek help with the long-term physical impact of their illness. An understanding of the context within which a concern is positioned can help clinicians to tailor management strategies and enhance coping for patients.

This synthesis provides new in-depth understanding of the concerns for people experiencing breathlessness and again emphasizes the significant impact of breathlessness on the social and spiritual domains within a person's life. Synthesis of the included studies combined has highlighted the importance of social participation, demonstrating the significant impact of breathlessness on relationships and loss of perceived role within the family.[17,51] The challenge to meet existential distress and preserve personal integrity is identified as significant in this review,[36,41] yet these are aspects that are less frequently acknowledged and measured within clinical practice.

Recent work proposes that the concept of "breathing space" can be used by clinicians to assess the impacts of breathlessness and provide guidance to patients on coping, help-seeking behavior, and treatment.[10] This framework can be helpful in considering the underlying theory with implications mostly on a staff and systems level. The findings of our work are complementary but distinct, providing clinicians and researches with a practical framework that can identify concerns at an individual level and ensure that appropriate coping strategies and help-seeking behavior are adopted.

This review also provides new evidence to support the choice of outcome measures in clinical trials of interventions for breathlessness. Our model of total breathlessness demonstrates that people with advanced illness express concerns across multiple domains, supporting the use of multiple-domain outcome measures in clinical practice and research. An increasing number of multiple-domain measures have been developed and validated to assess breathlessness. Examples include the Multidimensional Dyspnea Profile, the Dyspnoea-12, and the Chronic Respiratory Disease Questionnaire.

The Multidimensional Dyspnea Profile was designed for use in laboratory and clinical research and assessed sensory and affective dimensions of breathlessness at a specific time or during a specific activity.[59] The Dyspnoea-12 is often used in clinical practice and measures breathlessness severity, incorporating physical and affective aspects. It does not depend on a reference level of activity or specific time period and refers to how breathing feels "these days".[60] The Chronic Respiratory Disease Questionnaire is a health-related quality of life questionnaire that measures breathlessness experienced in the past two weeks across several domains including emotional function and mastery.[61] The context in which breathlessness occurs emerged as important in this review, and these measures consider context by incorporating different questions about the timing of, or triggers of breathlessness. Context should be carefully considered to choose the most appropriate multiple-domain measures in clinical practice and research.

Although multiple-domain measures are increasingly used in clinical practice, they are less commonly used in interventional research, and in particular drug trials. Cochrane reviews have recently been conducted to determine the effectiveness of oxygen,[62] benzodiazepines,[63] and opioids[64] in the management of chronic breathlessness. For each of these reviews, the selected primary outcome was breathlessness as measured using one of several single-domain measures (Numerical Rating Scale, Visual Analogue Scale, and Modified Borg). These outcome measures have the advantage of being short and straightforward to complete and can be pooled across studies for purposes of comparisons. However, they are limited by their simplicity and do not assess all the domains of total breathlessness that have been identified in this review. Future trials in breathlessness should consider a combination of breathlessness assessment (using a single-domain measure), in conjunction with a multiple-domain measure to ensure comprehensive assessment of total breathlessness, including the concerns and problems that matter to patients.

Strengths and Limitations

The qualitative methodology is a strength of this systematic review because it has enabled an in-depth understanding of the main concerns for those experiencing breathlessness. Use of an established framework in the model development has ensured that the results are embedded within the current knowledge base.

However, the majority of studies included in this synthesis were of participants with a diagnosis of COPD, based in the outpatient/community setting, in the U.K. Although there are many similarities in terms of the symptom burden of breathlessness across disease groups in advanced illness, there are also differences in terms of patient experience. For example, the length of diagnosis and the speed of onset of symptoms are different for people with cancer or ILD, when compared to other chronic lung disease, and this is likely to have impact on the concerns expressed by these patients.[31,65,66] Further research is needed to explore the concerns of people experiencing breathlessness in populations other than COPD and should build on the proposed model, to examine whether findings fit within these domains. This review also identifies the significant impact of breathlessness on those close to the people experiencing it. Further research should explore this impact and consider how it can be measured and addressed in routine clinical practice.

The original data were not available for analysis, and therefore, the synthesis relies on the quotes and results as interpreted and presented in the published papers.

Conclusions

People with advanced illness living with breathlessness have concerns in multiple domains, supporting a concept of "total breathlessness," which includes the original four domains of "total dyspnea," as well as two new domains (control and context) identified in this review. Control and context have been newly identified as important, particularly in their influence on coping and help-seeking behavior. The importance of social participation, impact on relationships, and loss of perceived role within social and spiritual domains also emerged as significant to the individual. Our model of "total breathlessness" provides a practical framework to guide comprehensive clinical assessment and optimize clinical interactions. Future trials in breathlessness should consider a combination of breathlessness assessment (using a single-domain measure), in conjunction with a multiple-domain measure to ensure comprehensive assessment of total breathlessness.

Disclosures and Acknowledgments

The authors declare that they have no conflict of interest.

N. Lovell is completing a training fellowship funded by Cicely Saunders International and Marie Curie (grant number A18859). M. Maddocks is supported by an NIHR Career Development Fellowship (CDF-2017-10-009) and NIHR Health Services & Delivery Research grant (HSDR 16/02/18) and NIHR CLARHC South London. I.J. Higginson is an NIHR Emeritus Senior Investigator and is supported by NIHR CLARHC South London. This research was supported by the Collaboration for Leadership in Applied Health Research and Care (CLAHRC) South London, which is part of the National Institute for Health Research (NIHR), and is a partnership between King's Health Partners, St. George's, University London, and St George's Healthcare NHS Trust. The views expressed are those of the authors and not necessarily those of the NHS, the NIHR, or the Department of Health.

References

1. Parshall MB, Schwartzstein RM, Adams L, et al. An official American Thoracic Society statement: update on the mechanisms, assessment, and management of dyspnea. Am J Respir Crit Care Med 2012;185:435–452.

2. Johnson MJ, Yorke J, Hansen-Flaschen J, et al. Towards an expert consensus to delineate a clinical syndrome of chronic breathlessness. The Eur Respir J 2017;49. https://doi.org/10.1183/13993003.02277-2016.

3. Simon ST, Weingartner V, Higginson IJ, et al. Definition, categorization, and terminology of episodic breathlessness: consensus by an international Delphi survey. J pain symptom Manag 2014;47:828–838.

4. Solano JP, Gomes B, Higginson IJ. A comparison of symptom prevalence in far advanced cancer, AIDS, heart disease, chronic obstructive pulmonary disease and renal disease. J pain symptom Manag 2006;31:58–69.

5. Moens K, Higginson IJ, Harding R. Are there differences in the prevalence of palliative care-related problems in people living with advanced cancer and eight non-cancer conditions? A systematic review. J pain symptom Manag 2014;48:660–677.

6. Bajwah S, Higginson IJ, Ross JR, et al. Specialist palliative care is more than drugs: a retrospective study of ILD patients. Lung 2012;190:215–220.

7. Young J, Donahue M, Farquhar M, et al. Using opioids to treat dyspnea in advanced COPD: attitudes and experiences of family physicians and respiratory therapists. Can Fam Physician Medecin de famille canadien 2012;58:e401–e407.

8. Dorman S, Jolley C, Abernethy A, et al. Researching breathlessness in palliative care: consensus statement of the National Cancer Research Institute Palliative Care Breathlessness Subgroup. Palliat Med 2009;23:213–227.

9. Bausewein C, Farquhar M, Booth S, et al. Measurement of breathlessness in advanced disease: a systematic review. Respir Med 2007;101:399–410.

10. Hutchinson A, Barclay-Klingle N, Galvin K, et al. Living with breathlessness: a systematic literature review and qualitative synthesis. Eur Respir J 2018;51. https://doi.org/10.1183/13993003.01477-2017.

11. Moher D, Liberati A, Tetzlaff J, et al. Preferred reporting items for systematic reviews and meta-analyses: the PRISMA statement. Ann Intern Med 2009;151:264–269. w64.

12. Reid C, Seymour J, Jones C. A thematic synthesis of the experiences of adults living with hemodialysis. Clin J Am Soc Nephrol: CJASN 2016;11:1206–1218.

13. Garcia-Rueda N, Carvajal Valcarcel A, Saracibar-Razquin M, et al. The experience of living with advanced-stage cancer: a thematic synthesis of the literature. Eur J Cancer Care 2016;25:551–569.

14. Thomas J, Harden A. Methods for the thematic synthesis of qualitative research in systematic reviews. BMC Med Res Methodol 2008;8:45.

15. Apps LD, Harrison SL, Williams JE, et al. How do informal self-care strategies evolve among patients with chronic obstructive pulmonary disease managed in primary care? A qualitative study. Int J Chronic Obstructive Pulm Dis 2014;9:257–263.

16. Bajwah S, Higginson IJ, Ross JR, et al. The palliative care needs for fibrotic interstitial lung disease: a qualitative study

of patients, informal caregivers and health professionals. Palliat Med 2013;27:869–876.

17. Barnett M. Chronic obstructive pulmonary disease: a phenomenological study of patients' experiences. J Clin Nurs 2005;14:805–812.

18. Booth S, Silvester S, Todd C. Breathlessness in cancer and chronic obstructive pulmonary disease: using a qualitative approach to describe the experience of patients and carers. Palliat Support Care 2003;1:337–344.

19. Caress A, Luker K, Chalmers K. Promoting the health of people with chronic obstructive pulmonary disease: patients' and carers' views. J Clin Nurs 2010;19:564–573.

20. Clancy K, Hallet C, Caress A. The meaning of living with chronic obstructive pulmonary disease. J Nurs Healthc Chronic Illness 2009;1:78–86.

21. Cooney A, Mee L, Casey D, et al. Life with chronic obstructive pulmonary disease: striving for 'controlled co-existence'. J Clin Nurs 2013;22:986-995.

22. Duck A, Spencer LG, Bailey S, et al. Perceptions, experiences and needs of patients with idiopathic pulmonary fibrosis. J Adv Nurs 2015;71:1055–1065.

23. Dunger C, Higginson IJ, Gysels M, et al. Breathlessness and crises in the context of advanced illness: a comparison between COPD and lung cancer patients. Palliat Support Care 2015;13:229–237.

24. Edmonds PM, Rogers A, Addington-Hall JM, et al. Patient descriptions of breathlessness in heart failure. Int J Cardiol 2005;98:61–66.

25. Ek K, Sahlberg-Blom E, Andershed B, et al. Struggling to retain living space: patients' stories about living with advanced chronic obstructive pulmonary disease. J Adv Nurs 2011;67:1480–1490.

26. Fraser DD, Kee CC, Minick P. Living with chronic obstructive pulmonary disease: insiders' perspectives. J Adv Nurs 2006;55:550–558.

27. Gardiner C, Gott M, Small N, et al. Living with advanced chronic obstructive pulmonary disease: patients concerns regarding death and dying. Palliat Med 2009;23:691–697.

28. Gullick J, Stainton MC. Living with chronic obstructive pulmonary disease: developing conscious body management in a shrinking life-world. J Adv Nurs 2008;64:605–614.

29. Gysels M, Higginson IJ. Access to services for patients with chronic obstructive pulmonary disease: the invisibility of breathlessness. J pain symptom Manag 2008;36:451–460.

30. Gysels M, Reilly CC, Jolley CJ, et al. Dignity through integrated symptom management: lessons from the breathlessness support service. J pain symptom Manag 2016;52:515–524.

31. Gysels MH, Higginson IJ. The lived experience of breathlessness and its implications for care: a qualitative comparison in cancer, COPD, heart failure and MND. BMC Palliat Care 2011;10:15.

32. Habraken JM, Pols J, Bindels PJE, et al. The silence of patients with end-stage COPD: a qualitative study. Br J Gen Pract 2008;58:844–849.

33. Hallas CN, Howard C, Theadom A, et al. Negative beliefs about breathlessness increases panic for patients with chronic respiratory disease. Psychol Health Med 2012;17:467–477.

34. Harris D, Hayter M, Allender S. Improving the uptake of pulmonary rehabilitation in patients with COPD:: qualitative study of experiences and attitudes. Br J Gen Pract 2008;58:703–710.

35. Hasson F, Spence A, Waldron M, et al. I can not get a breath: experiences of living with advanced chronic obstructive pulmonary disease. Int J Palliat Nurs 2008;14:526–531.

36. Henoch I, Bergman B, Danielson E. Dyspnea experience and management strategies in patients with lung cancer. Psycho-oncology 2008;17:709–715.

37. Jones I, Kirby A, Ormiston P, et al. The needs of patients dying of chronic obstructive pulmonary disease in the community. Fam Pract 2004;21:310–313.

38. Jonsdottir H. Life patterns of people with chronic obstructive pulmonary disease: isolation and being closed in. Nurs Sci Q 1998;11:160–166.

39. Jørgensen LB, Dahl R, Pedersen PU, et al. Four types of coping with COPD-induced breathlessness in daily living: a grounded theory study. J Res Nurs 2012;18:520–541.

40. Lai YL, Chan CW, Lopez V. Perceptions of dyspnea and helpful interventions during the advanced stage of lung cancer: Chinese patients' perspectives. Cancer Nurs 2007;30:E1–E8.

41. Leidy NK, Haase JE. Functional status from the patient's perspective: the challenge of preserving personal integrity. Res Nurs Health 1999;22:67–77.

42. Lowey SE. Care preferences of individuals living with advanced cardiac and respiratory illness. Dissertation Abstr Int Section B: The Sci Eng 2012;72:5885.

43. Luthy C, Cedraschi C, Pasquina P, et al. Perception of chronic respiratory impairment in patients' drawings. J Rehabil Med 2013;45:694–700.

44. Nicholls DA. The experience of chronic breathlessness. Physiother Theor Pract 2003;19:123–136.

45. O'Driscoll M, Corner J, Bailey C. The experience of breathlessness in lung cancer. Eur J Cancer Care 1999;8:37–43.

46. Oliver SM. Living with failing lungs: the doctor–patient relationship. Fam Pract 2001;18:430–439.

47. Roberts DK, Thorne SE, Pearson C. The experience of dyspnea in late-stage cancer. Patients' and nurses' perspectives. Cancer Nurs 1993;16:310–320.

48. Robinson T. Living with severe hypoxic COPD: the patients' experience. Nurs times 2005;101:38–42.

49. Victorson DE, Anton S, Hamilton A, et al. A conceptual model of the experience of dyspnea and functional limitations in chronic obstructive pulmonary disease. Value Health: J Int Soc Pharmacoeconomics Outcomes Res 2009;12:1018–1025.

50. Walthall H, Jenkinson C, Boulton M. Living with breathlessness in chronic heart failure: a qualitative study. J Clin Nurs 2017;26:2036–2044.

51. Williams V, Bruton A, Ellis-Hill C, et al. What really matters to patients living with chronic obstructive pulmonary disease? An exploratory study. Chronic Respir Dis 2007;4:77–85.

52. Wortz K, Cade A, Menard JR, et al. A qualitative study of patients' goals and expectations for self-management of

COPD. Prim Care Respir J: J Gen Pract Airways Group 2012; 21:384–391.

53. Abernethy AP, Wheeler JL. Total dyspnoea. Curr Opin Support Palliat Care 2008;2:110–113.

54. Donaldson AV, Maddocks M, Martolini D, et al. Muscle function in COPD: a complex interplay. Int J chronic obstructive Pulm Dis 2012;7:523–535.

55. Ramon MA, Ter Riet G, Carsin A-E, et al. The dyspnoea-inactivity vicious circle in COPD: development and external validation of a conceptual model. Eur Respir J 2018;52.

56. Gysels M, Bausewein C, Higginson IJ. Experiences of breathlessness: a systematic review of the qualitative literature. Palliat Support Care 2007;5:281–302.

57. Simon ST, Higginson IJ, Benalia H, et al. Episodes of breathlessness: types and patterns–a qualitative study exploring experiences of patients with advanced diseases. Palliat Med 2013;27:524–532.

58. Linde P, Hanke G, Voltz R, et al. Unpredictable episodic breathlessness in patients with advanced chronic obstructive pulmonary disease and lung cancer: a qualitative study. Support Care Cancer 2018;26:1097–1104.

59. Banzett RB, O'Donnell CR, Guilfoyle TE, et al. Multidimensional Dyspnea Profile: an instrument for clinical and laboratory research. Eur Respir J 2015;45:1681–1691.

60. Yorke J, Moosavi SH, Shuldham C, et al. Quantification of dyspnoea using descriptors: development and initial testing of the Dyspnoea-12. Thorax 2010;65:21–26.

61. Guyatt GH, Berman LB, Townsend M, et al. A measure of quality of life for clinical trials in chronic lung disease. Thorax 1987;42:773–778.

62. Ekstrom M, Ahmadi Z, Bornefalk-Hermansson A, et al. Oxygen for breathlessness in patients with chronic obstructive pulmonary disease who do not qualify for home oxygen therapy. Cochrane database Syst Rev 2016;11:CD006429.

63. Simon ST, Higginson IJ, Booth S, et al. Benzodiazepines for the relief of breathlessness in advanced malignant and non-malignant diseases in adults. The Cochrane database Syst Rev 2016;10:CD007354.

64. Barnes H, McDonald J, Smallwood N, et al. Opioids for the palliation of refractory breathlessness in adults with advanced disease and terminal illness. Cochrane database Syst Rev 2016;3:CD011008.

65. Bajwah S, Ross JR, Peacock JL, et al. Interventions to improve symptoms and quality of life of patients with fibrotic interstitial lung disease: a systematic review of the literature. Thorax 2013;68:867–879.

66. Kreuter M, Bendstrup E, Russell AM, et al. Palliative care in interstitial lung disease: living well. Lancet Respir Med 2017;5:968–980.

Appendix

Appendix 1
Search Strategy: Medline Search Strategy

Concept 1: Exposure	Concept 2: Outcome	Concept 3: Population
exp dyspnea/	exp patient preference/	exp Palliative Care/
dyspn*.tw.	exp health priorities/	exp Terminal Care/
short* of breath.tw.	experience*.tw.	exp Terminally Ill/
breathless*.tw.	expectat*.tw.	exp Hospices/
acute exacerbation*.tw.	prefer*.tw.	palliat*.tw.
	priorit*.tw.	terminal care.tw.
	concern*.tw.	end of life.tw.
	narrative.tw.	hospice*.tw.
		dying.tw
		chronic respiratory disease.tw.
		chronic lung disease.tw.
		chronic obstructive pulmonary disease.tw.
		end stage*.tw.
		life limit*.tw.
		advance*.tw.
		progressive*.tw.
		severe*.tw.
		chronic.tw.
		adj2
		illness.tw.
		condition*.tw
		disease.tw.
		diagnosis.tw.
		chronic obstructive pulmonary disease.tw.
		interstitial lung disease.tw.
		heart failure.tw.
		cancer.tw.

Appendix 2

Critical Appraisal Skills Program Checklist Appraisal

Author	Clear Statement of Aims	Appropriate Methodology	Appropriate Design	Appropriate Recruitment Strategy	Appropriate Data Collection	Relationship Between Researcher and Participants Considered	Ethical Issues Considered	Rigorous Data Analysis	Clear Statement of Findings	Value of Research
Apps et al.[15]	Yes	Yes	Yes	Yes	Yes	No	Yes	Yes	Yes	Yes
Bajwah et al.[16]	Yes	Yes	Yes	Yes	Yes	No	Yes	Yes	Yes	Yes
Barnett, M.[17]	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Booth et al.[18]	Yes	Yes	Yes	Yes	Yes	No	Yes	Yes	Yes	Yes
Caress et al.[19]	Yes	Yes	Yes	Yes	Yes	No	Yes	Yes	Yes	Yes
Clancy et al.[20]	Yes	Yes	Yes	Yes	Can’t tell	No	Yes	Can’t tell	Yes	Yes
Cooney et al.[21]	Yes	Yes	Yes	Yes	Yes	No	Yes	Yes	Yes	Yes
Duck et al.[22]	Yes	Yes	Yes	Yes	Yes	No	Yes	Yes	Yes	Yes
Dunger et al.[23]	Yes	Yes	Yes	Yes	Yes	No	Yes	Yes	Yes	Yes
Edmonds et al.[24]	Yes	Yes	Yes	Yes	Yes	No	Yes	Yes	Yes	Yes
Ek et al.[25]	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Fraser et al.[26]	Yes	Yes	Yes	Yes	Yes	No	Yes	Yes	Yes	Yes
Gardiner et al.[27]	Yes	Yes	Yes	Yes	Yes	No	Yes	Yes	Yes	Yes
Gullick et al.[28]	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Gysels et al.[29]	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Gysels et al.[30]	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Gysels et al.[31]	Yes	Yes	Yes	Yes	Yes	No	Yes	Yes	Yes	Yes
Habraken et al.[32]	Yes	Yes	Yes	Yes	Yes	No	Yes	Yes	Yes	Yes
Hallas et al.[33]	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Harris et al.[34]	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Hasson et al.[35]	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Henoch et al.[36]	Yes	Yes	Yes	Yes	Yes	No	Yes	Yes	Yes	Yes
Jones et al.[37]	Yes	Yes	Yes	Yes	Yes	No	Yes	Yes	Yes	Yes
Jonsdottir, H.[38]	Yes	Yes	Yes	Yes	Yes	No	Can’t tell	Can’t tell	Yes	Yes
Jørgensen et al.[39]	Yes	Yes	Yes	Yes	Yes	No	Yes	Yes	Yes	Yes
Lai et al.[40]	Yes	Yes	Yes	Yes	Yes	No	Yes	Yes	Yes	Yes
Leidy et al.[41]	Yes	Yes	Yes	Yes	Yes	No	Can’t tell	Yes	Yes	Yes
Lowey, S.[42]	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Luthy et al.[43]	Yes	Yes	Yes	Yes	Yes	No	Yes	Yes	Yes	Yes
Nicholls, D.[44]	Yes	Yes	Yes	Yes	Yes	No	Yes	Yes	Yes	Yes
O’Driscoll et al.[45]	No	Yes	Yes	Yes	Yes	Yes	Can’t tell	Yes	Yes	Yes
Oliver, S.[46]	Yes	Yes	Yes	Yes	Yes	Yes	Can’t tell	Yes	Yes	Yes
Roberts et al.[47]	Yes	Yes	Yes	Yes	Yes	No	Yes	Yes	Yes	Yes
Robinson, T.[48]	Yes	Yes	Yes	Yes	Yes	No	Yes	Yes	Yes	Yes
Victorson et al.[49]	Yes	Yes	Yes	Yes	Yes	No	Can’t tell	Yes	Yes	Yes
Walthall et al.[50]	Yes	Yes	Yes	Yes	Yes	No	Yes	Yes	Yes	Yes
Williams et al.[51]	Yes	Yes	Yes	Yes	Yes	No	Yes	Yes	Yes	Yes
Wortz et al.[52]	Yes	Yes	Yes	Yes	Yes	No	Can’t tell	Yes	Yes	Yes