

Holistic services for people with advanced disease and chronic or refractory breathlessness: a mixed-methods evidence synthesis

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Declared competing interests of authors: Some of the authors of this report were co-authors on studies included in the systematic review but were not involved in the data extraction and quality assessment for these studies. Specifically, one or more authors have been involved with 17 out of 37 publications, across 4 out of 18 services based in London (Irene J Higginson, Wei Gao and Sara Booth), Hull (Sara Booth), Cambridge (Sara Booth, Morag Farquhar and Irene J Higginson) and Munich (Sara Booth). A committee search showed membership of the Health Technology Assessment (HTA) End of Life Care and Add on Studies that ended in February 2016 for Wei Gao, and membership of the Health Services and Delivery Research (HSDR) Commissioned Board 2009–15, HTA Efficient Study Designs 2015–16, HTA End of Life Care and Add on Studies and Service Delivery and Organisation (SDO) Studies Panel Member for Irene J Higginson.

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Scientific summary

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Scientific summary

Background

Breathlessness is a common, distressing symptom that affects over two million people in the UK. It increases as disease progresses and often becomes chronic or refractory (i.e. not responsive to treatment that is aimed at the underlying disease). It can be frightening for patients and families, and results in reduced life expectancy and high NHS costs.

There are few effective treatments for refractory breathlessness, which suggests that services that combine treatments are needed. Cardiac and pulmonary rehabilitation provide exercise-based approaches to help manage breathlessness, but their reach is limited, particularly with regard to those with the most advanced disease, who become socially isolated and 'invisible'.

Holistic breathlessness services provide pharmacological and non-pharmacological treatments to patients and caregivers, across settings, using multidisciplinary approaches. They emphasise self-management, and target improvements in quality of life (QoL) by reducing the impact of breathlessness and related symptoms on everyday living.

Literature scoping identified a body of primary research related to these services, including randomised controlled trials, but this evidence must be synthesised and understood collectively to inform future practice. The aim of this study was to synthesise evidence on holistic breathlessness services for people with advanced disease and chronic or refractory breathlessness.

Objectives

The objectives were to:

- describe the available evidence for holistic breathlessness services in terms of the intervention format, content, organisation and context, patient characteristics, study design and quality, and outcomes measured
- determine the clinical effectiveness of holistic breathlessness services on symptom burden, health status and QoL
- determine the cost-effectiveness of holistic breathlessness services from patient/caregiver, societal and NHS perspectives
- examine the acceptability of holistic breathlessness services from the perspective of health-care professions and patients, considering rates of referral, uptake and adherence, as well as patient experience and satisfaction
- use individual patient data to examine predictors of treatment response, including characteristics of participants (e.g. level of impairment, symptom burden, multimorbidity) and interventions (e.g. setting, duration, professional input, delivery)
- use stakeholder consultations to elicit stakeholders' priorities for clinical practice, policy and research around holistic breathlessness services, including their role and delivery in relation to cardiac and pulmonary rehabilitation services.

Methods

Systematic review

Nine databases were searched from inception to 2 June 2017 for qualitative, observational and experimental studies. Eligibility, methodological quality and reporting quality were independently assessed

by two authors. Data on service models, health and cost outcomes were synthesised, using meta-analyses as indicated. The quality of the evidence for each outcome was assessed using the GRADE (Grading of Recommendations Assessment, Development and Evaluation) approach. Qualitative data on patient/carer experiences were synthesised thematically. Quantitative experience and patient flow data were tabulated and/or summarised narratively.

Responder analysis

A secondary analysis was conducted of pooled individual patient data from three trials of holistic breathlessness services. The primary analysis considered response as a ≥ 0.5 -point improvement in the Chronic Respiratory Disease Questionnaire (CRQ) mastery score, and secondary analysis considered response as a ≥ 1 -point reduction in Numeric Rating Scale (NRS) distress due to breathlessness. Candidate variables included diagnosis, predicted forced expiratory volume in 1 second (FEV₁), baseline CRQ dyspnoea, fatigue, mastery and emotional domains, EuroQol-5 Dimensions (EQ-5D) Utility Index and visual analogue scale, Hospital Anxiety and Depression Scale anxiety and depression scores, and NRS average breathlessness in the previous 24 hours. Variables significantly related to response in univariate logistic regression models were considered in separate multivariate analyses, comprising backward stepwise logistic regression modelling.

Stakeholder consultation

Transparent expert consultation was used, comprising nominal group technique during a stakeholder workshop and a follow-up online consensus survey. Stakeholders representing multiple specialties and professions, as well as patients/carers, were invited to participate in the stakeholder workshop. Following the presentations covering relevant evidence, participants discussed, and put forward, their own suggestions for future practice during small group sessions. These suggestions were combined and synthesised by the project team into a final list of statements, and circulated to a wider group in the online consensus survey.

The same group of participants, plus additional people from the groups that were under-represented at the workshop, were invited to participate in the online consensus survey from 12 to 26 February 2018. Each rated the final statements from 1 (strongly disagree) to 9 (strongly agree), with opportunities for free-text comments. Scores were analysed descriptively and classified into predetermined categories for levels agreement and consensus.

Results

Systematic review

Of 3239 records identified, 37 papers were included, representing 18 different services. Most services enrolled people with thoracic cancer, involved palliative care staff and comprised 4–6 contacts over 4–6 weeks. Common interventions were breathing techniques (14/18), psychological support (12/18) and relaxation techniques (11/18).

Meta-analyses demonstrated reductions in distress due to breathlessness [$n = 324$; mean difference (MD) -2.30 , 95% confidence interval (CI) -4.43 to -0.16 ; $p = 0.03$] and depression scores ($n = 408$; MD -1.67 , 95% CI -2.52 to -0.81 ; $p < 0.001$), favouring the intervention. Statistically non-significant effects were observed for breathlessness mastery ($n = 259$; MD 0.23 , 95% CI -0.10 to 0.55 ; $p = 0.17$) and anxiety scores ($n = 552$; MD -1.59 , 95% CI -3.22 to 0.05 ; $p = 0.06$). There was no observable effect on overall health status or QoL. Evidence for cost-effectiveness was limited to four studies and was inconclusive.

Patients reported a high level of satisfaction with services. In particular, patients and their informal carers valued the provision of dignified, person-centred care from expert staff; education and information-sharing; and useful self-management interventions that were simple and portable (e.g. handheld fan, breathing techniques).

Responder analysis

The pooled data set comprised 259 participants (118 female) with a mean age of 69.2 [standard deviation (SD) 10.6] years. The most common primary diagnosis was chronic obstructive pulmonary disease (49.8%), followed by cancer (34.7%) and interstitial lung disease (10.4%), with a mean FEV₁% predicted value of 46.2 (SD 21.4).

Controlling for age, sex and trial in the multivariate modelling, baseline CRQ mastery remained the only significant predictor of intervention CRQ mastery response [odds ratio (OR) 0.57, 95% CI 0.43 to 0.74; $p < 0.001$], and baseline NRS distress remained the only significant predictor of intervention NRS distress response (OR 1.64, 95% CI 1.35 to 2.03; $p < 0.001$).

Stakeholder consultation

Stakeholders attending the workshop ($n = 37$) produced 34 statements reflecting their priorities: 10 for clinical practice, 8 for policy and 16 for research. Following ratings by 74 online survey respondents, seven had strong agreement and a high level of consensus.

Stakeholders' priorities for clinical practice were to:

- ensure that breathlessness services are person-centred and flexible in terms of delivery (e.g. appointment location, time, and duration)
- ensure that breathlessness services are cross-cutting, drawing on relevant expertise from multiple disciplines, professions and providers
- work towards ensuring that breathlessness services have the widest possible geographical coverage and access (e.g. travelling communities, people who are homeless, people living in care/nursing homes)
- acknowledge family and/or informal carers within breathlessness services and, when appropriate, actively encourage their participation in education and in management of the patient's breathlessness
- value symptom management in its own right, and be able to deliver (or refer patients for) breathlessness interventions
- share breathlessness management skills with other health and social care professionals, and informal carers.

Stakeholders' priority for health-care policy was to recognise informal carers in terms of their role, importance and support needs.

Conclusions

Holistic services for chronic or refractory breathlessness in people with advanced disease are heterogeneous in their components and delivery, but are acceptable to patients and carers, who value the tailored education, simple and portable breathlessness self-management interventions and psychological support provided in the services. Overall, these services lead to significant improvements in psychological aspects of breathlessness and health. The evidence for cost-effectiveness of these services is inconclusive.

Clinical response to holistic breathlessness services in terms of increased mastery or reduced distress is influenced by baseline scores for these variables, but not by breathlessness severity, patient diagnosis, lung function, health status, anxiety or depression. In the context of limited resources, prioritising patients with low levels of mastery or high levels of distress could be appropriate to direct resources to those most likely to benefit.

Stakeholders showed strongest agreement and consensus around suggestions to improve access to person-centred, multiprofessional care, and support for carers to provide, or access, breathlessness management interventions. Future research in this field should test the optimal models of care and educational strategies to address stakeholders' priorities and understand how best to embed core therapeutic components into routine clinical practice.

Study registration

This study is registered as PROSPERO CRD42017057508.

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