Recommendations for services for people with living with chronic breathlessness in advanced disease: results of a transparent expert consultation

Authors:

Lisa Jane Brighton[1] – lisa.brighton@kcl.ac.uk
India Tunnard[1] - india.tunnard@kcl.ac.uk
Morag Farquhar[2] - M.Farquhar@uea.ac.uk
Sara Booth[3] - sb628@cam.ac.uk
Sophie Miller[1] - sophiejmiller@googlemail.com
Deokhee Yi[1] - deok_hee.yi@kcl.ac.uk
Wei Gao[1] - wei.gao@kcl.ac.uk
Sabrina Bajwah[1] - sabrina.bajwah@kcl.ac.uk
William D-C. Man[4,5] - W.Man@rbht.nhs.uk
Charles C Reilly[6] - charles.reilly@nhs.net
Margaret Ogden[7] – margaretogden@hotmail.com
Sylvia Bailey[7] - sylviab@talktalk.net
Colleen Ewart[7] - colleen.ewart@btinternet.com
Irene J Higginson[1] - irene.higginson@kcl.ac.uk
Matthew Maddocks[1] – matthew.maddocks@kcl.ac.uk

Affiliations:
1. Cicely Saunders Institute of Palliative Care, Policy and Rehabilitation, King's College London, Bessemer road, London, SE5 9PJ, UK
2. University of East Anglia, School of Health Sciences, Norwich Research Park, Norwich, NR4 7TJ, UK
3. Department of Oncology, University of Cambridge, Cambridge Biomedical Campus, Cambridge, CB2 0XZ, UK
4. National Heart and Lung Institute, Imperial College, London, SW3 6LR, UK
5. Harefield Pulmonary Rehabilitation and Muscle Research Laboratory, Royal Brompton & Harefield NHS Foundation Trust, London, UB9 6JH, UK
6. King's College Hospital NHS Foundation Trust, Department of Physiotherapy, London, SE5 9RS UK
7. Patient/Carer representative, Cicely Saunders Institute of Palliative Care, Policy and Rehabilitation, King's College London, Bessemer road, London, SE5 9PJ, UK

Correspondence to: Lisa Jane Brighton, King's College London, Cicely Saunders Institute of Palliative Care, Policy and Rehabilitation, London, SE5 9PJ, United Kingdom.
Tel: +44 (0)20 7848 5041. Email: lisa.brighton@kcl.ac.uk

Word count: 3368 words

Tables and Figures: 4 tables, 3 Figures

ABSTRACT (200 words)

Background: Chronic breathlessness is highly distressing for people with advanced disease and their informal carers, yet health services for this group remain highly heterogeneous. We aimed to generate evidence-based stakeholder-endorsed recommendations for practice, policy, and research concerning services for people with advanced disease and chronic breathlessness.

Methods: We used Transparent Expert Consultation, comprising modified nominal group techniques during a stakeholder workshop and an online consensus survey. Stakeholders representing multiple specialities and professions, and patient/carers were invited to participate.

Results: 37 participants attended the stakeholder workshop and generated 34 separate recommendations, rated by 74 online survey respondents. Seven recommendations had strong agreement and high levels of consensus. Stakeholders agreed services should: be person-centred and flexible; cut across multiple disciplines and providers; and prioritise breathlessness management in its own right. They advocated for wide geographical coverage and access to expert care, supported through skills-sharing among professionals. They also recommended recognition of informal carers and their role by clinicians and policy-makers.

Conclusion: Recommendations with strongest agreement and consensus centred on improved access to person-centred, multi-professional care, and support for carers to provide, or access, breathlessness management interventions. Future research should test the optimal models of care and educational strategies to meet these recommendations.

INTRODUCTION

Breathlessness affects up to 95% of people with chronic lung disease, around 70% of people with lung cancer, and over 60% of people living with heart disease[1]. Breathlessness generally progresses with disease severity, and becomes chronic, i.e. it persists despite optimal treatment of the underlying condition and results in disability[2]. Chronic breathlessness is highly distressing and associated with considerable anxiety, disability and social isolation[3]. It can result in disrupted sleep, and high levels of stress and burden for informal carers of people with breathlessness[4]. This is often compounded by additional and interacting symptoms experienced alongside breathlessness such as fatigue, anxiety and cough[5, 6], and the frequent presence of co-morbidities.

Rehabilitation services, for example pulmonary rehabilitation, can aid management of breathlessness through exercise, education and behavioural interventions[7, 8]. However, lack of referral[9], potential stigma, restricting symptoms and health deterioration can prevent participation in some patients[10, 11]. Recently, breathlessness-triggered services that focus on holistic assessment, multi-professional care, education and psychosocial support have been shown to improve outcomes for people with chronic breathlessness in advanced disease[12]. However, the operation of such services remains highly heterogenous with regard to structure and delivery. As such, there is limited consensus around optimal practices to support people living with chronic breathlessness in advance disease, and their informal carers.

As multiple specialities (e.g. respiratory, cardiology, oncology, palliative care) and professions (e.g. health and social care, voluntary sector, research) have expertise relevant to supporting people living with chronic breathlessness in advance disease, it is important to incorporate a range of perspectives in guiding future practice. Whilst commonly used, nominal group and survey techniques have been criticised for lacking transparency, reliability, and opportunities for clarification[13]. The Delphi technique overcomes these issues, but can be time-consuming with multiple rounds of consultation, and the initial content can be shaped by a minority. In response, Transparent Expert Consultation (TEC) methods have been developed[13, 14], and used successfully to generate recommendations in palliative and end of life care research[15-19]. The TEC process involves structured opportunities for expression of views at a face-to-face meeting (similar to nominal group technique), followed by consideration of generated recommendations in a wider consensus survey (similar to a single-round Delphi technique), to enable rapid consultation of multiple key stakeholders. We therefore aimed to generate evidence-based recommendations for clinical practice, policy, and research around services for people living with chronic breathlessness in advanced disease, using TEC.

METHODS

Design

TEC[13, 14] methods were used, comprising a modified nominal group technique during a stakeholder workshop, and an online consensus survey (Figure 1).

[INSERT FIGURE 1]

Participants

People representing different specialties, professional groups, service providers and service commissioners involved in caring for people living with chronic breathlessness, including voluntary sector organisations and patient and carer representatives, were purposively invited by email to participate in the stakeholder workshop. Participants were identified through contact lists of people and organisations held by the authors, additional recommendations from these participants, and online literature and website searches. All who were invited to participate in the workshop were also invited to complete the online consensus survey. Additional individuals from groups who were less well represented in the workshop (e.g. patient and carer representatives) were purposefully selected using the methods above and invited to complete the online consensus survey.

Procedure

Identifying critical issues

Critical issues in relation to services for people with chronic breathlessness in advanced disease were identified through a systematic review[12] examining the components, outcomes, and recipients' experiences of holistic breathlessness services. Review findings were discussed within our project advisory group (PAG: comprising researchers, clinicians, and service user representatives), which led to the identification of three critical questions to discuss during the stakeholder workshop:

(1) How do we define and deliver 'holistic breathlessness services'?

(2) How and where can holistic breathlessness services be integrated into current practice?

(3) How should the success of holistic breathlessness services be measured / monitored?

Stakeholder Workshop

The workshop took place on 4th October 2017. On arrival, participants received a pack which included a reminder of the study information (first provided with the email invitation), consent

forms to complete prior to the group work sessions, and the workshop schedule. An artist was present throughout the event to create a live graphic recording of the discussions.

The workshop began with whole-group presentations and discussions on the following topics: defining, acceptability of and experiences of breathlessness services, rehabilitation services, care bundles, and supporting informal carers of people with chronic breathlessness. Following this, participants were purposefully allocated into one of three parallel group sessions (based on expertise, and to ensure diverse roles within each group) to focus on one of the critical questions. These sessions used a modified nominal group technique, facilitated and scribed by members of the research team. Following guidance prepared before the workshop, the facilitators led participants through a structured process (Table 1) and were responsible for chairing the discussion in a way that allowed everyone to contribute.

Table 1: Structured process for workshop groupwork

Step	Process
Written responses	Participants wrote individual answers to 'prompt questions' in response booklets (Supplementary Figure S1). These were tailored to the critical question each group was focusing on, for example "What are the core components of a holistic breathlessness service?" (Group 1); "Where should a holistic breathlessness service be based?" (Group 2); and "What is the ideal set of outcomes to measure for patients?" (Group 3).
Initial reflections	Reflections from this exercise in relation to the critical question were then discussed.
Individual recommendations	Participants wrote their individual recommendations in their response booklets, with a rationale and indication of appropriateness for clinical practice, policy, and/or research.
Ranking	Participants were asked to rank each of their recommendations from highest to lowest.
Discussion	Participants in turn read out their highest ranked recommendation and rationale, which were discussed by the group. This continued until individual lists were exhausted or time was exceeded (approximately 25 minutes).[19]

Group discussions were audio-recorded and completed response booklets collected to provide a record. Scribes noted the top recommendations on flipchart paper in each parallel group to feedback to the whole group. The workshop closed with a summary, and information about the upcoming online consensus survey.

Following the workshop, one researcher (LB) reviewed materials generated and summarised the main themes of the discussions throughout the day, including within each parallel group. This primarily involved synthesising common and salient points from the written notes and referring to the audio recordings where there was lack of clarity. This was summarised in a narrative and checked by other members of the research team to ensure a transparent record of the workshop discussions surrounding the development of individual recommendations.

Online Consensus Survey

Individual recommendations (with their rationale, ranking and grouping) were anonymised and entered into Microsoft Excel. Recommendations were categorised by two authors (LB,MM) into clinical practice, research and policy (assigned to the predominant category where ≥2 selected), and ordered by participants' rankings from most important to least important.

After familiarisation through multiple readings, two authors (LB, MM) undertook a process of de-duplication and synthesising similar recommendations within each category. Where further clarity was needed, the graphic recording, flipchart records, scribes' notes, and audio-recordings were examined. Recommendations were not retained if they were deemed to be: replicating existing recommendations, unclear, outside the scope (e.g. not specific to chronic breathlessness in advanced disease), redundant (e.g. practice recommendations already exist), and/or ranked low priority by the participant who wrote it. Where possible recommendations retained participants' original language, with amendments only to enhance clarity and avoid inflexible statements (e.g. changing 'must' to 'could')[15, 17]. Areas of uncertainty and the final list of recommendations were discussed and revised with the PAG, who had been given a copy of all the original recommendations for transparency. This final list of recommendations was formatted into an online survey, which was piloted by a clinical academic, researcher and patient representative from the PAG to assess and improve user-friendliness and clarity.

Potential participants received a personalised email invitation, followed by two reminders, to complete the online survey. All were offered the option of receiving a hard copy of the survey with a freepost return envelope, if preferred. The survey ran from 12th to 26th February 2018.

Survey respondents were asked to select their profession/role and area(s) of expertise, with a free-text 'other' option if required. Participants were then presented with the three sets of recommendations for research, clinical practice, and policy, and asked to indicate their level of agreement with each recommendation from 1 (strongly disagree) to 9 (strongly agree). An opportunity for free-text comments was presented at the end of each section.[14, 19, 20]

Analysis

Survey responses were analysed using descriptive statistics (frequencies, median, inter-quartile range, range) to determine levels of agreement and consensus in line with pre-determined categories used in a previous study[16]. Classification of agreement and consensus were as shown in Table 2.

Table 2: Classification of agreement and consensus with recommendations:

Median	IQR	Category
≥8	<2	Strong agreement/high consensus
≥8	≥2	Strong agreement/low consensus
<8 to >6	<2	Moderate agreement/high consensus
<8 to >6	≥2	Moderate agreement/low consensus

Free-text comments were collated within each recommendation category, and analysed thematically to aid understanding of the issues raised by the proposed recommendations[21].

Ethics

Ethical approval for this study was obtained from the King's College London research ethics committee (ref. LRS-16/17-4692). Workshop participants provided signed consent prior to the recorded discussions and booklet completion, and were reimbursed reasonable travel costs for attending. Consent for the online survey was presumed through participation.

RESULTS

Participants

Of 117 stakeholders invited, 40 registered for and 37 attended the workshop. Most participants were from the UK, two attended from abroad. Thirty-three stakeholders participated in the group work and completed response booklets (Group 1 n=12; Group 2 n=8; Group 3 n=13). Two patient representatives could not attend the event but completed a response booklet to provide their recommendations. Characteristics of participants who completed the response booklets are shown in Table 3.

Table 3: Participant Characteristics

Characteristic	Workshop Booklets (n=35)		Online Survey (n=74)	
	n	(%)	n	(%)
Profession/Role				
Doctor (clinical)	16	47	30	40.5
Researcher	17	50	29	39.2
Physiotherapist	4	10.8	11	14.8
Patient/Carer representative	3	8.6	9	12.2
Role in charitable organisation	2	5.8	9	12.2
Nurse	2	5.8	7	9.5
Commissioner	2	5.8	4	5.4
Occupational Therapist	1	2.9	0	0
Psychologist	1	2.9	2	2.7
Other[a]	2	5.8	1	1.4
Area of expertise				
Lung disease	16	47	43	58.1
Palliative care	17	50	29	39.2
Research	13	38.2	28	37.8
Cancer	6	17.6	12	16.2
I am a patient/carer	3	8.6	10	13.5
General practice	1	2.9	7	9.5
Heart disease	6	5.8	5	6.8
Psychology	2	5.8	5	6.8
Geriatrics	4	10.8	4	5.4
Other[b]	2	5.8	4	5.4

[a]music and mindfulness therapist
[b]Rehabilitation, Cognitive behavioural therapist, Breathlessness/informal carers, Dermatology
Note: workshop and survey participants could select more than one option for both sections

Of 160 people (workshop invitees and 43 additional people) invited to take part in the online consensus survey, 74 participated, representing a 46% response rate. One invitee declined to participate, three email addresses were no longer valid, six were away until after the survey close date, and 78 did not respond. Of the 74 survey respondents, 26 had previously completed a workshop response booklet.

Stakeholder Workshop

Throughout the event, there were strong themes of the need for improved collaboration, integrated working, and standardisation. Participants acknowledged successful elements of existing practices, across multiple specialities and disciplines, that should be built upon and not duplicated. The graphic recording summarising the group discussions is shown in Figure 2.

[INSERT FIGURE 2]

Group 1: How do we define and deliver 'holistic breathlessness services'? Participants suggested that to define and deliver these services, different models of care needed to be evaluated for clinical and cost-effectiveness. These services need to be evidence-based and integrated, with collection of routine data to review access and outcomes. A key component of delivery should be establishing, and upskilling a range of clinicians in core breathlessness management skills, and supporting them to integrate this into their routine practice.

Group 2: How and where can holistic breathlessness services be integrated into current practice? Participants in Group 2 also felt that upskilling clinicians in breathlessness management skills was core to integrating breathlessness services. This should include attention to both the physical and psychological components of breathlessness, should consider ways to enable self-management, and should not be disease-specific. Challenges with service integration and different approaches across different localities were noted.

Group 3: How should the success of holistic breathlessness services be measured / monitored? Discussions here centred around ensuring outcomes were patient-led, clearly mapped to service aims, and psychometrically robust. Inclusion (and development) of carer-reported outcomes were also discussed. Participants felt strongly that any approach to measurement should be based on existing successful methods, should be consistent, and integrated with existing practice.

In total, 187 individual recommendations were generated for research, clinical practice, and/or policy. Most recommendations had implications for research (n=101), followed by clinical (n=76) and

policy (n=41) implications (multiple categories could be selected). Synthesis of these 187 recommendations resulted in 34 recommendations for the online consensus survey.

Online Consensus Survey

The online consensus survey included 34 final recommendations: 10 for clinical practice, eight for policy, and 16 for research. The recommendations and the scores received in the online consensus survey are shown in Table 4, with boxplots in Figure 3.

Table 4: Recommendations and online survey responses

Clinical recommendations for professionals working with people with chronic breathlessness in advanced disease	Median (IQR)[a]
Strong agreement, high consensus	
Ensure breathlessness services are person-centred and flexible in terms of delivery (e.g. appointment location, time, and duration) (C1)	9 (8-9)
Ensure breathlessness services are cross-cutting, drawing on relevant expertise from multiple disciplines, professions, and providers (C2)	9 (8-9)
Work towards ensuring breathlessness services have the widest possible geographical coverage and access (e.g. travelling communities, people who are homeless, people living in care/nursing homes) (C3)	9 (8-9)
Acknowledge family and/or informal carers within breathlessness services and, where appropriate, actively encourage their participation in education and in management of the patient's breathlessness (C7)	9 (8-9)
Value symptom management in its own right, and be able to deliver, or refer patients for, breathlessness interventions (C9)	9 (8-9)
Share breathlessness management skills with other health and social care professionals, and informal carers (C10)	9 (8-9)
Strong agreement, low consensus	
Define clear referral criteria for breathlessness services (e.g. limiting breathlessness that persists despite optional management of underlying disease), and share these with potential referrers (C4)	8 (7-9)
Use multiple strategies to raise awareness of breathlessness services among potential referrers and the public (e.g. by engaging with professional bodies, charities or patient groups) (C6)	8 (7-9)
Be alert to, and respond to, under-recognised related issues (e.g. sleep, intimacy, etc.) (C8)	8 (7-9)
Moderate agreement, low consensus	
Consider providing the option for patients to self-refer to breathlessness services (C5)	7 (6-9)
Policy recommendations	
Strong agreement, high consensus	
Recognise informal carers in terms of their role, importance, and support needs (P7)	9 (8-9)
Strong agreement, low consensus	
Complete a needs assessment around breathlessness, map it to current service provision and consider areas for service improvement (P1)	8 (7-9)
Prioritise supporting development of breathlessness-triggered services, which span all stages of multiple diseases and conditions (P2)	8 (7-9)

Map how breathlessness services could sit within existing care provision and plans, to avoid duplication (P3)	8 (7-9)
Agree, publish, and review breathlessness service quality standards as new evidence accumulates (P4)	8 (7-9)
Establish an audit programme for breathlessness services to track impact of services nationally or internationally (P5)	8 (7-9)
Increase public awareness and/or education around breathlessness (e.g. as a sign of disease versus normal exertional symptom) (P6)	8 (7-9)
Provide all health and social care staff with education around breathlessness and its management, ideally starting during vocational and/or undergraduate training and continuing throughout professional lives (P8)	8 (7-9)
Research recommendations	
Strong agreement, low consensus	
Explore optimal delivery methods of service provider education for breathlessness assessment and management (R16)	9 (7-9)
Understand the impact of breathlessness and associated factors (e.g. fatigue or isolation) on health and social care service use and costs (R1)	8 (7-9)
Establish a core set of outcome measures for clinical practice and research, incorporating validated patient and carer measures (R3)	8 (7-9)
Determine medium to long-term effects of breathlessness services using follow-up assessments beyond completion of the intervention (R4)	8 (7-9)
Examine and understand models of integrated working between breathlessness services and other providers (e.g. palliative, respiratory, primary, social care) (R5)	8 (7-9)
Assess the clinical and cost-effectiveness of breathlessness services for people unable to engage in cardiac/respiratory rehabilitation services (R6)	8 (7-9)
Assess the clinical and cost-effectiveness of breathlessness services for people who have had their first unplanned hospital admission related to breathlessness (R7)	8 (7-9)
Assess the clinical and cost-effectiveness of the following components within breathlessness services: Carer-focused interventions (R10)	8 (7-9)
Assess need for service provider education around breathlessness (R15)	8 (7-9)
Complete economic modelling (including cost-effectiveness studies) of breathlessness services, which should include health and societal perspectives (R14)	8 (6.25-9)
Moderate agreement, high consensus	
Assess the clinical and cost-effectiveness of the following components within breathlessness services: structured exercise training (R9)	7 (7-8.75)
Moderate agreement, low consensus	
Assess the clinical and cost-effectiveness of breathlessness services for care/nursing home residents (R8)	7 (6-9)
Convene a representative group of funders / commissioners to establish the type of outcomes they would need to see for breathlessness services (R2)	7 (6-8)
Assess the clinical and cost-effectiveness of the following components within breathlessness services: telehealth (e.g. virtual multidisciplinary team meetings, video resources for patients/carers) (R11)	7 (6-8)
Assess the clinical and cost effectiveness of the value of the following variations of breathlessness services: As an adjunct to existing services (e.g. pulmonary rehabilitation) (R12)	7 (6-8)
Assess the clinical and cost effectiveness of the value of the following variations of breathlessness services: Group versus individual delivery (R13)	7 (6-8)

[a]scores ranged from 1 to 9

[INSERT FIGURE 3]

Recommendations for clinical practice

The most strongly supported recommendations were those calling for person-centred care (C1), and drawing on multiple expertise (C2), with widest possible coverage both geographically and demographically (C3). They also included acknowledgement of the role of informal carers (C7), valuing and being able to respond to breathlessness as a symptom in its own right (C9), and sharing these skills with other professionals and informal carers (C10). This was reflected in free-text comments about the importance of holistic care, particularly acknowledging psychological concerns, and the importance of skills-sharing. Participants noted that multidisciplinary working could include multidisciplinary teams, or single-discipline teams with strong links to other specialities.

Despite strong agreement there was low consensus around defining referral criteria (C4), using multiple strategies to raise awareness of breathlessness (C6), and responding to under-recognised related issues (C8). One respondent commented that referral structures should not be too rigid, as this may be a barrier where presentation to the service is atypical. The most contentious recommendation was around the option for patients to self-refer to services (C5): comments highlighted concerns around self-referral and ensuring joined up healthcare, medical record access, and ensuring medications are maximised and reversible conditions ruled out. Overall comments on the clinical recommendations noted the need for better understanding of how these services would sit alongside existing practice, without duplication.

Recommendations for policy

The most strongly supported recommendation for policy was recognition of informal carers in terms of their role, importance, and support needs (P7). The remaining recommendations received high overall agreement but low consensus.

Comments highlighted contention over the utility of mapping (P1, P3) and national audit (P4, P5), questioning their benefit to day-to-day practice. Multiple comments stressed the importance of education (of the public as well as care professionals) as a priority area (P6, P8), particularly to support existing services. However, concerns were also raised around how best to achieve this in a way that learning isn't 'lost' within larger education schemes. Another comment noted that breathlessness-triggered services (P2) should be developed through adapting existing services, rather than introducing something new. One participant highlighted that the policy recommendations had a strong healthcare focus, despite people with breathlessness spending most

of their time outside of health services. Work to understand the role of social care and communities in supporting patients with breathlessness and their carers was suggested.

Recommendations for research

None of the research recommendations received high agreement and high consensus. The only recommendation receiving high consensus (and moderate agreement) was the need for economic modelling of breathlessness services (R9).

The remaining recommendations received low consensus, with strong (R1,R3-7,R10,R14-16) or moderate (R2,R8,R11-13) agreement. Most comments suggested that low consensus resulted, in part, from inadequate definitions of the population of interest (e.g. people having unplanned admissions due to breathlessness: R7), or insufficient justification for the area of research (e.g. effectiveness for care/nursing home residents: R8). Participants commented that they also assigned lower agreement where they felt good understanding or evidence already existed (e.g. the impact of breathlessness: R1). Additional suggestions for research included the role of psychological factors, psychosocial interventions, community support, and the best ways to support informal carers.

DISCUSSION

The strongest recommendations from this stakeholder consultation centre around clinicians providing care for people with advanced disease and chronic breathlessness. Stakeholders recommend care that: is person-centred and flexible; cuts across multiple disciplines, professions, and providers; and focuses on breathlessness management in its own right. This should be developed in the context of wide geographical coverage and access to expert care, supported through a focus on skills-sharing among health care professionals and informal carers. In line with this, participants called for clinicians and policy-makers to recognise the role and potential support needs of informal carers in supporting people with chronic breathlessness.

A focus on the symptom of breathlessness and promotion of joint working was also recommended in a previous consultation exercise focused on breathlessness rehabilitation for people with COPD and heart failure[22], while the need for greater education and skills-sharing was raised in another consensus study defining chronic breathlessness[2]. It is noteworthy that multiple elements of the clinical recommendations are in line with a palliative care approach, including: person centred care, multidisciplinary input, and inclusion of informal carers in the unit of care[23]. These characteristics are commonly observed in holistic services for people with advanced disease and chronic

breathlessness[12] and working to build links with, or learn from, palliative care may be an efficient way to facilitate working in line with recommendations from this and previous consultations. Future work is needed to understand the variation in models of care (including core and optional components) and how best to share breathlessness management skills across individuals, professions, and disciplines. This could be facilitated by inclusion of these recommendations in future priority-setting exercises (e.g. James Lind Alliance partnerships[24]).

Two of the most supported recommendations were around recognising the role and importance of informal carers, and where appropriate supporting and encouraging their participation in care. These recommendations are reinforced by recent evidence demonstrating the substantial contribution of informal carers to people with advanced disease and chronic breathlessness[25], including that their input saves approximately two thirds of what would otherwise be formal care costs[26], and the impact this has on their own health and wellbeing[26][27]. Additional research is necessary to determine optimal methods of supporting informal carers of patients with breathlessness, and work is currently underway to develop evidence-basedinterventions[28, 29].

Methodological reflections

It is a strength that participants in the workshop and online consensus survey represented a wide and relevant range of stakeholders, including patient and carer representatives. However, it may be that this diversity of expertise and knowledge of existing research led to the lower consensus around the research-related recommendations. Most participants were based in UK universities and National Health Service settings within secondary care, and a high proportion of participants were doctors and researchers. The latter partially reflects the tendency for these participants to have dual roles: 69% of the researcher participants were also healthcare professionals; 47% of doctors had additional roles (e.g. researcher, commissioner, charity organisation role). Although these proportions reflect who was invited (rather than differing response rates; Supplementary Table S1), these characteristics may have biased the types of recommendations generated and made them more applicable to health and social care within the UK. The response rate to the online survey was also limited, however it was similar to previous studies using this method[16-18] and all key stakeholder groups were represented.

It is notable that only a small number of patient/carer representatives attended the workshop and completed booklets. This format may be less suitable to people with severe breathlessness and/or caring responsibilities. However, having multiple service user representatives on the project advisory group enabled their input in the design of the workshop, and synthesising and revising the resulting

recommendations. This included working closely together to ensure the recommendations were clear and understandable for people with a range of professional and personal expertise, and incorporating their suggestion to offer the opportunity to speak with a member of the research team if clarification was needed.

Having a clear, structured process incorporating focused discussion and specific questions during the workshop ensured efficient collection of participants' views. Although face-to-face consultation techniques can be subject to bias through some participants contributing more than others in discussions, we mitigated this through providing the opportunity to submit individual written recommendations in a response booklet and primarily focusing on these responses when generating survey content. Whilst following the full Delphi process[30] or having additional consultation rounds may have provided more opportunity to refine the recommendations, the TEC technique maximised on the multiple forms of data collected at the workshop (e.g. scribe notes, graphic recording, response booklets, audio-recordings) and enabled the multidisciplinary expertise of the project team to rapidly synthesise and revise the recommendations. Although a substantial part of synthesising and revising the recommendations was completed by two researchers, the full list of original recommendations had been shared with the PAG for transparency. Importantly, individual recommendations were only removed in line with the reasons stated above (e.g. duplicates, low priority), and not on the basis of controversy or creativity. Moreover, this method still resulted in generally high levels of agreement and consensus, particularly around clinical recommendations. With a growing emphasis on consulting stakeholders as part of the development and evaluation of complex interventions[31, 32], this efficient method of obtaining recommendations and consensus from a diverse group of stakeholders may be increasingly useful.

Conclusions

This stakeholder consultation has generated multiple recommendations for clinical practice, policy and guidance around services for people with chronic breathlessness in advanced disease. The recommendations with strongest agreement and consensus centred around improved access to person-centred, multi-professional care, and the ability of formal and informal carers to know how to provide, or access, breathlessness management interventions. Stakeholders called for clinical practice and policy to recognise the role of informal carers in supporting people with chronic breathlessness in advanced disease. Future research is needed to identify and test the optimal models of care and educational strategies to meet these recommendations.

Author contributions

MM and IJH designed the study, obtained funding, and take overall responsibility for its content. LB, IT, and MM collected the workshop and online survey data, all authors contributed to interpretation of the data and refining the recommendations, LB and IT led the write-up with supervision from MM. All authors contributed to the manuscript and approved the final version.

Competing Interests

The authors declare no competing interests.

Data Sharing

Requests for access to anonymised data should be directed to the corresponding author.

REFERENCES

1. Solano JP, Gomes B and Higginson IJ. A comparison of symptom prevalence in far advanced cancer, AIDS, heart disease, chronic obstructive pulmonary disease and renal disease. . *Journal of Pain and Symptom Management*. 2006; 31: 58-69.

2. Johnson MJ, Yorke J, Hansen-Flaschen J, et al. Towards an expert consensus to delineate a clinical syndrome of chronic breathlessness. *European Respiratory Journal*. 2017; 49.

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

4. Farida AM, Marjolein G and Irene JH. Living with breathlessness: A survey of caregivers of breathless patients with lung cancer or heart failure. *Palliative Medicine*. 2013; 27: 647-56.

5. Bausewein C, Booth S, Gysels M, Kuhnbach R, Haberland B and Higginson IJ. Understanding breathlessness: cross-sectional comparison of symptom burden and palliative care needs in chronic obstructive pulmonary disease and cancer. *J Palliat Med*. 2010; 13: 1109-18.

6. Janssen DJ, Spruit MA, Uszko-Lencer NH, Schols JM and Wouters EF. Symptoms, comorbidities, and health care in advanced chronic obstructive pulmonary disease or chronic heart failure. *J Palliat Med*. 2011; 14: 735-43.

7. McCarthy B, Casey D, Devane D, Murphy K, Murphy E and Lacasse Y. Pulmonary rehabilitation for chronic obstructive pulmonary disease. *Cochrane Database Syst Rev*. 2015; 2: Cd003793.

8. Taylor RS, Sagar VA, Davies EJ, et al. Exercise-based rehabilitation for heart failure. *Cochrane Database Syst Rev*. 2014; 4: doi: 10.1002/14651858.CD003331.pub4.

9. Milner SC, Boruff JT, Beaurepaire C, Ahmed S and Janaudis-Ferreira T. Rate of, and barriers and enablers to, pulmonary rehabilitation referral in COPD: A systematic scoping review. *Respiratory Medicine*. 2018; 137: 103-14.

10. Gysels M and Higginson IJ. Access to services for patients with chronic obstructive pulmonary disease: the invisibility of breathlessness. *J Pain Symptom Manage*. 2008; 36: 451-60.

11. Maddocks M, Kon SSC, Canavan JL, et al. Physical frailty and pulmonary rehabilitation in COPD: a prospective cohort study. *Thorax*. 2016.

12. Brighton L, Miller S, Farquhar M, et al. Holistic services for chronic breathlessness in people with advanced disease: a systematic review and meta-analysis. *Under review*.

13. Raine R, Sanderson C and Black N. Developing clinical guidelines: a challenge to current methods. *BMJ*. 2005; 331: 631.

14.   Yardley L, Beyer N, Hauer K, McKee K, Ballinger C and Todd C. Recommendations for promoting the engagement of older people in activities to prevent falls. *Quality & safety in health care*. 2007; 16: 230-4.

15.   Evans CJ, Harding R, Higginson IJ and Morecare. 'Best practice' in developing and evaluating palliative and end-of-life care services: a meta-synthesis of research methods for the MORECare project. *Palliat Med*. 2013; 27: 885-98.

16.   Farquhar M, Preston N, Evans CJ, et al. Mixed methods research in the development and evaluation of complex interventions in palliative and end-of-life care: report on the MORECare consensus exercise. *J Palliat Med*. 2013; 16: 1550-60.

17.   Gysels M, Evans CJ, Lewis P, et al. MORECare research methods guidance development: recommendations for ethical issues in palliative and end-of-life care research. *Palliat Med*. 2013; 27: 908-17.

18.   Nancy JP, Peter F, Stephen JW, et al. Recommendations for managing missing data, attrition and response shift in palliative and end-of-life care research: Part of the MORECare research method guidance on statistical issues. *Palliative Medicine*. 2013; 27: 899-907.

19.   Higginson IJ, Evans CJ, Grande G, et al. Evaluating complex interventions in End of Life Care: the MORECare Statement on good practice generated by a synthesis of transparent expert consultations and systematic reviews. *BMC medicine*. 2013; 11: 111.

20.   Stevinson C, Preston N and Todd C. Defining priorities in prognostication research: results of a consensus workshop. *Palliat Med*. 2010; 24: 462-8.

21.   Garcia J, Evans J and Reshaw M. ``Is There Anything Else You Would Like to Tell Us'' – Methodological Issues in the Use of Free-Text Comments from Postal Surveys. *Quality and Quantity*. 2004; 38: 113-25.

22.   Man W, Chowdhury F, Taylor R, et al. Building consensus for provision of breathlessness rehabilitation for patients with chronic obstructive pulmonary disease and chronic heart failure. *Chronic Respiratory Disease*. 2016; 13: 229-39.

23.   Maddocks M, Lovell N, Booth S, Man WDC and Higginson IJ. Palliative care and management of troublesome symptoms for people with chronic obstructive pulmonary disease. *The Lancet*. 390: 988-1002.

24.   Best S, Tate T, Noble B, et al. The palliative and end of life care priority setting partnership (PEoLC PSP): Determining evidence uncertainties from the perspective of the end user of research *BMJ Supportive & Palliative Care*. 2014; 4: A42.

25.   Farquhar M. Carers and breathlessness. *Current Opinion in Supportive and Palliative Care*. 2017; 11: 165-73.

26.   Dzingina MD, Reilly CC, Bausewein C, et al. Variations in the cost of formal and informal health care for patients with advanced chronic disease and refractory breathlessness: A cross-sectional secondary analysis. *Palliative Medicine*. 2017; 31: 369-77.

27.   Morag F. Assessing carer needs in chronic obstructive pulmonary disease. *Chronic Respiratory Disease*. 2017; 15: 26-35.

28.   Farquhar M, Penfold C, Benson J, et al. Six key topics informal carers of patients with breathlessness in advanced disease want to learn about and why: MRC phase I study to inform an educational intervention. *PloS one*. 2017; 12: e0177081.

29.   Farquhar M, Penfold C, Walter FM, Kuhn I and Benson J. What Are the Key Elements of Educational Interventions for Lay Carers of Patients With Advanced Disease? A Systematic Literature Search and Narrative Review of Structural Components, Processes and Modes of Delivery. *J Pain Symptom Manage*. 2016; 52: 117-30.e27.

30.   Okoli C and Pawlowski SD. The Delphi method as a research tool: an example, design considerations and applications. *Information & Management*. 2004; 42: 15-29.

31.   Hawkins J, Madden K, Fletcher A, et al. Development of a framework for the co-production and prototyping of public health interventions. *Bmc Public Health*. 2017; 17: 689.

32.   Moore GF, Audrey S, Barker M, et al. Process evaluation of complex interventions: Medical Research Council guidance. *BMJ*. 2015; 350: h1258.
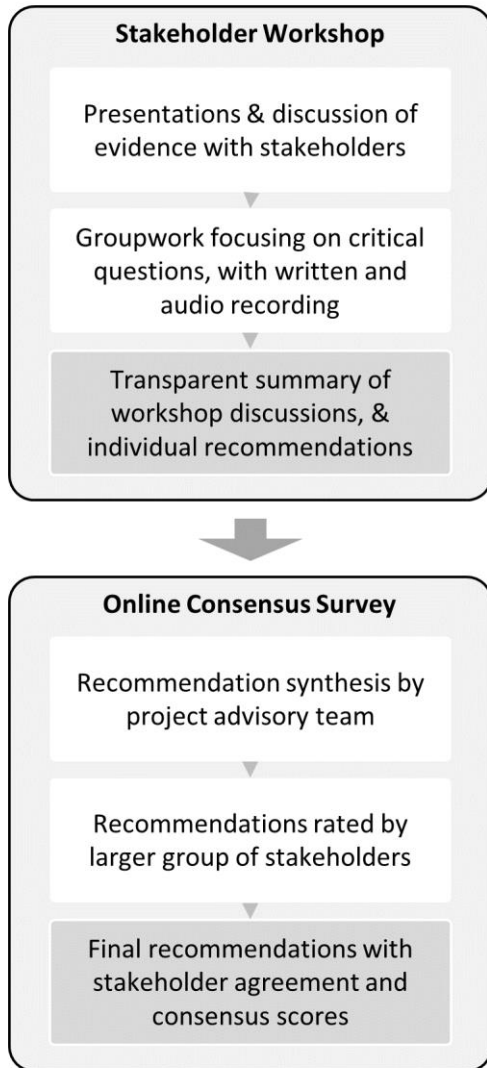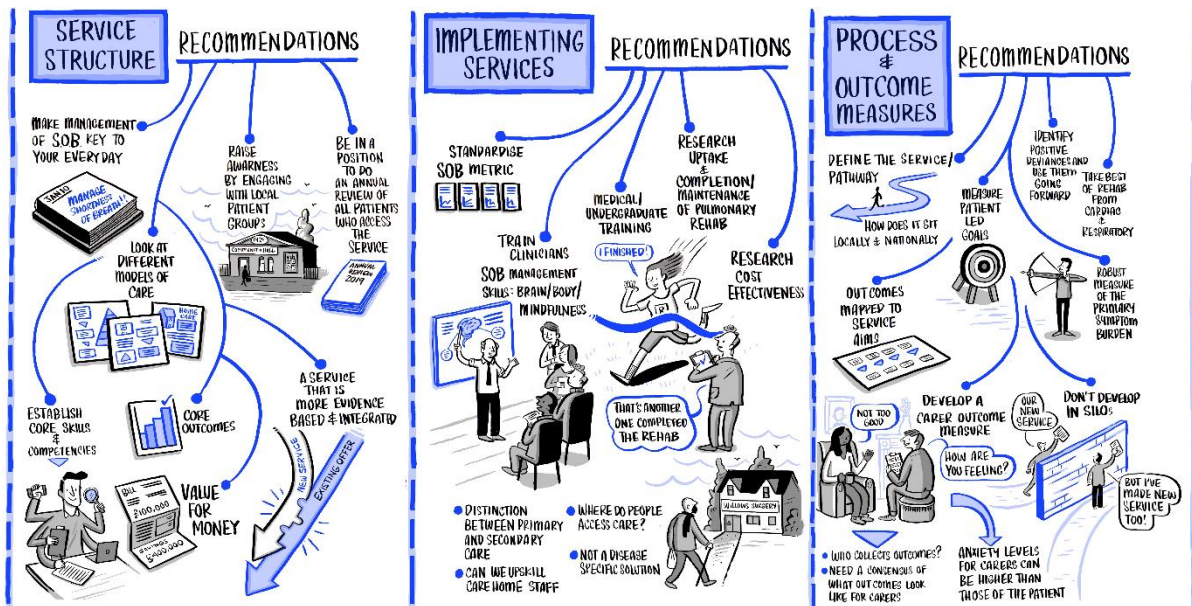
Figure 1: Transparent expert consultation

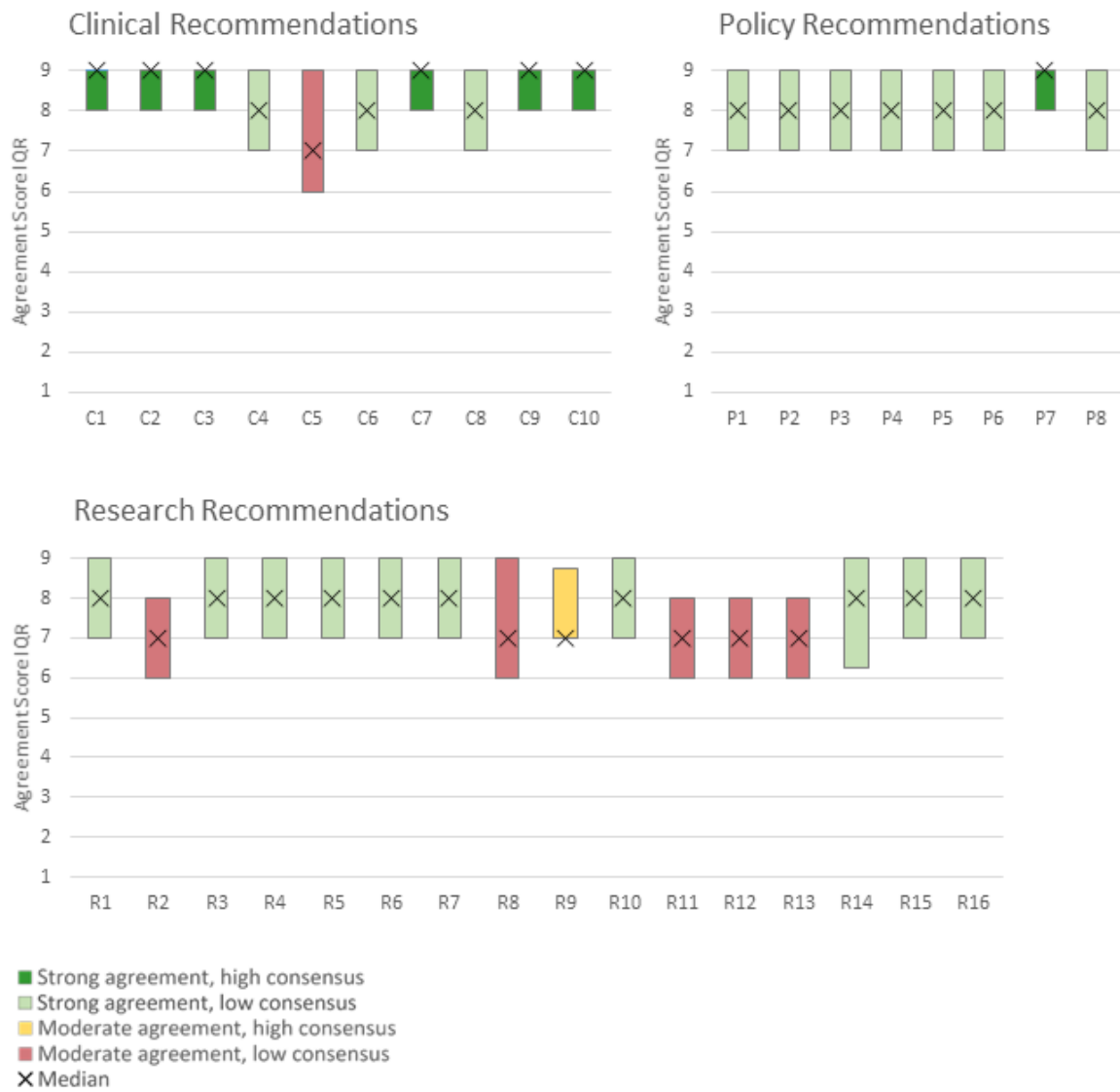**Figure 2: Graphic recording of stakeholder workshop discussions**

Figure 3: Boxplots of online consensus survey scores