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

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Abstract

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. We used transparent expert consultation, comprising modified nominal group technique during a stakeholder workshop, and an online consensus survey. Stakeholders, representing multiple specialities and professions, and patient/carers were invited to participate. Thirty-seven 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, should cut across multiple disciplines and providers and should prioritize 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 policymakers. Overall, stakeholders' recommendations reflect the need for 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.

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Keywords

Advanced disease, breathlessness, consultation, consensus, palliative care

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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, that is, 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 heterogeneous with regard to structure and delivery. As such, there is limited consensus around optimal practices to support people living with chronic breathlessness in advanced 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 advanced disease, it is important to incorporate a range of perspectives in guiding future practice. While commonly used, nominal group and survey techniques have been criticized for lacking transparency, reliability and opportunities for clarification.¹³ The Delphi technique overcomes these issues, but it 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).

Participants

People representing different specialties, professional groups, service providers and service commissioners involved in caring for people living with chronic breathlessness, including voluntary sector organizations 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 organizations 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 above-mentioned methods and invited to complete the online consensus survey.

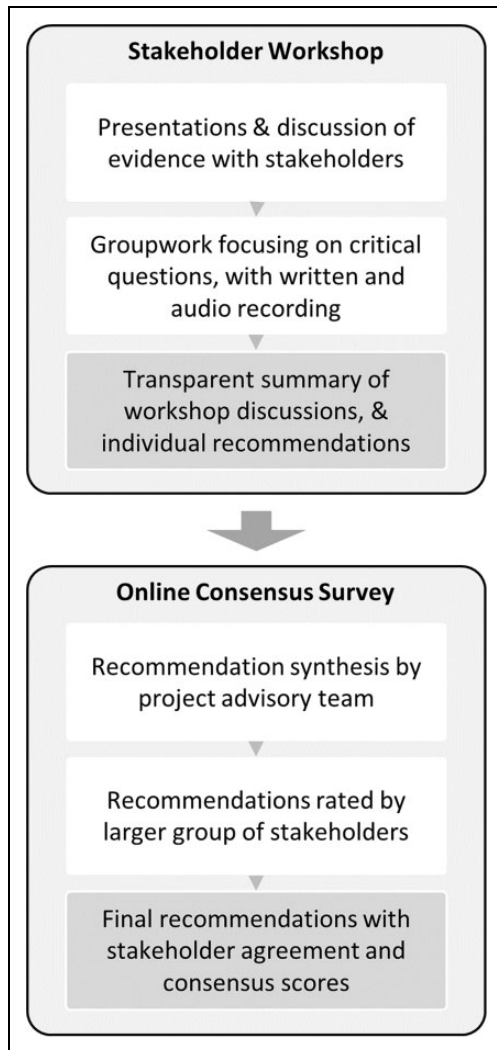


Figure 1. Transparent expert consultation.

Procedure

Identifying critical issues. Critical issues in relation to services for people with chronic breathlessness in advanced disease were identified through a systematic review¹² 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 4 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.

Group discussions were audio-recorded and completed response booklets were collected to provide a record. Scribes noted the top recommendations on flip chart 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 (LJB) reviewed materials generated and summarized the main themes of the discussions throughout the day, including within each parallel group. This primarily involved synthesizing common and salient points from the written notes and referring to the audio recordings where there was lack of clarity. This was summarized 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 anonymized and entered into Microsoft Excel. Recommendations were categorized by two authors (LJB, 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.

Table 1. Structured process for workshop group work.

Step	Process
Written responses	Participants wrote individual answers to 'prompt questions' in response booklets (see the Online 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

After familiarization through multiple readings, two authors (LJB, MM) undertook a process of deduplication and synthesizing similar recommendations within each category. Where further clarity was needed, the graphic recording, flip chart records, scribes' notes and audio recordings were examined. Recommendations were not retained if they were deemed to be: replicating the 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

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

IQR: interquartile range.

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 personalized 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 12 February 2018 to 26 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, interquartile range, range) to determine the levels of agreement and consensus in line with predetermined categories used in a previous study.¹⁶ Classification of agreement and consensus is shown in Table 2.

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

Ethics 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 United Kingdom, 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.

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 standardization. Participants acknowledged successful elements of existing practices, across multiple specialities and disciplines, which should be built upon and not duplicated. The graphic recording summarizing the group discussions is shown in 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

Table 3. Participant characteristics.^a

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 organization	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 ^b	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 ^c	2	5.8	4	5.4

^aWorkshop and survey participants could select more than one option for both sections.

^bMusic and mindfulness therapist.

^cRehabilitation, cognitive behavioural therapist, breathlessness/informal carers and dermatology.

in breathlessness management skills was core to integrating breathlessness services. This should include attention to both the physical and the 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 on ensuring outcomes were patient-led, clearly mapped to service aims and psychometrically robust. Inclusion (and development) of carer-reported outcomes was also discussed. Participants felt strongly that any approach to measurement should be based on existing successful methods, should be consistent, and should be integrated with existing practices.

Table 4. Recommendations and online consensus survey responses.

	Median (IQR) ^a
Clinical recommendations	
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 has 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 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-recognized 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, low cons	
Recognize 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 the current service provision and consider areas for service improvement (P1)	8 (7–9)
Prioritize 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 the 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)

(continued)

Table 4. (continued)

	Median (IQR) ^a
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)

IQR: interquartile range.

^aScores ranged from 1 to 9.

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–R7, R10, R14–R16) or moderate (R2, R8, R11–R13) 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 on how clinicians provide care for

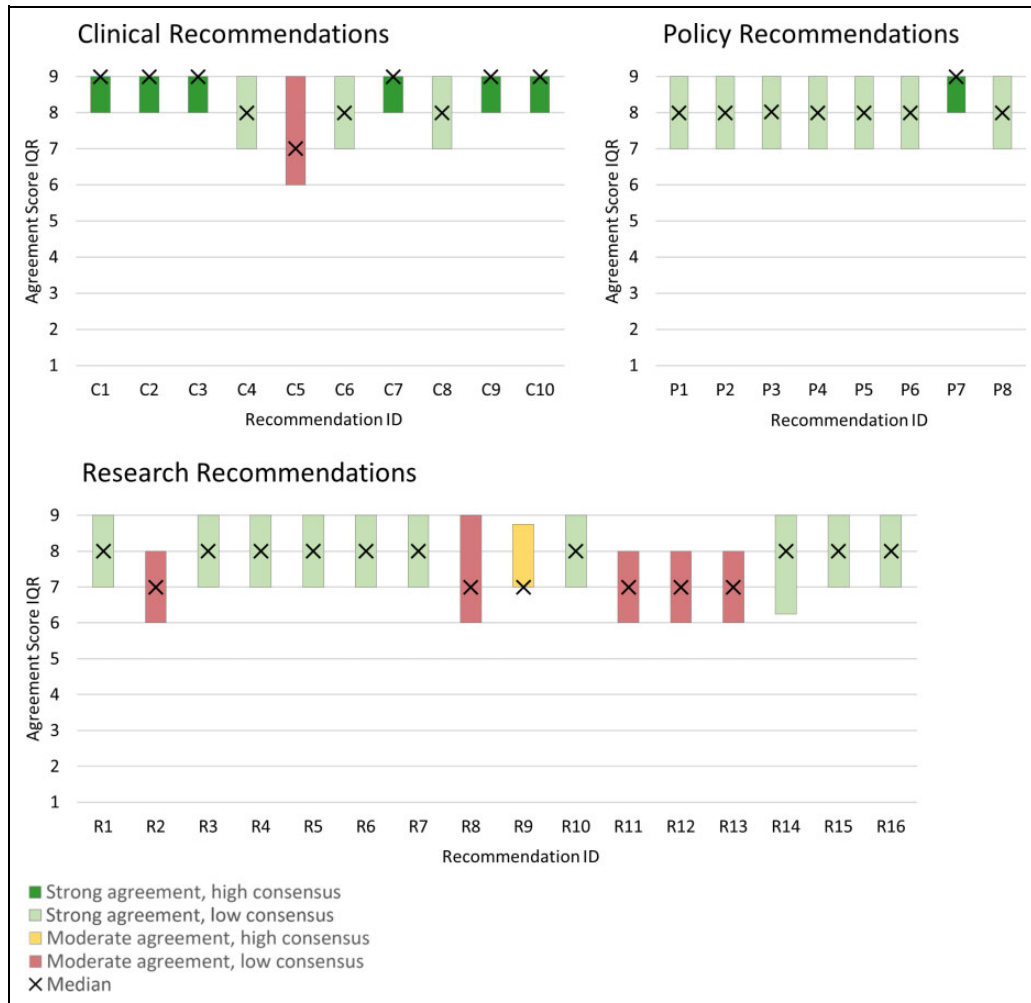


Figure 3. Box plots of online consensus survey scores.

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 healthcare professionals and informal carers. In line with this, participants call for clinicians and policymakers to recognize 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 chronic obstructive pulmonary disease 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, multi-disciplinary 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 breathlessness¹² 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 the inclusion of these recommendations in future priority-setting exercises (e.g. James Lind Alliance partnerships²⁴).

Two of the most supported recommendations were around recognizing 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-based interventions.^{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 organization role). Although these proportions reflect who was invited (rather than differing response rates; see the Online 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 United Kingdom. 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 PAG enabled their input in the design of the workshop and synthesizing 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 the online consensus survey. While following the full Delphi process³⁰ or having additional consultation rounds may have provided more opportunity to refine the recommendations, the TEC technique maximized on the multiple forms of data collected at the workshop (e.g. scribe notes, graphic recording, response booklets, audio recordings) and enabled the multidisciplinary project team to rapidly synthesize and revise the recommendations. Although a substantial part of synthesizing 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 earlier (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 on improved access to person-centred, multi-professional care and the ability of formal and informal carers to provide or access breathlessness management interventions. Stakeholders called for clinical practice

and policy to recognize 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.

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Author contributions

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

Data sharing

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

Declaration of conflicting interests


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Supplemental material

Supplemental material for this article is available online.

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