



A systematic review of self-management programmes for bronchiectasis in adult patients: a realist synthesis of the evidence

Journal:	<i>Disability and Rehabilitation</i>
Manuscript ID	TIDS-02-2021-024
Manuscript Type:	Review
Keywords:	Self-management, Systematic review, Realist review, Bronchiectasis, Chronic condition

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Implications for Rehabilitation

- This evidence synthesis has identified potentially important combinations of interventions to be considered in self-management programmes for adults with bronchiectasis
- A programme theory was developed to describe how three identified self-management programmes work in different contextual circumstances
- Underlying processes including social facilitation and patient empowerment were identified to potentially explain important clinical outcomes



For Peer Review

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3 **A systematic review of self-management programmes for bronchiectasis in**
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5 **adult patients: a realist synthesis of the evidence**
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Abstract

Purpose: Self-management for bronchiectasis patients has been identified as an important component that could potentially empower patients to manage their conditions and improve quality of life. Evidence was reviewed to investigate what self-management programmes work, why and in what circumstances.

Methods: A comprehensive database search was conducted on seven databases for evidence published up to June 2020. Self-management programmes were examined in each study and had to include at least two of the following self-management interventions: education, exercise, adherence to treatment, symptom monitoring, airway clearance techniques (ACT) and action plans. A realist logic of analysis was applied to produce context-mechanism-outcome configurations to provide an explanatory account of which self-management intervention programmes works, for whom and in what circumstances.

Results: Seven eligible articles ($n = 273$) contributed to the development of three context-mechanism-outcome configurations articulated for three self-management programmes that included education and action planning, education and ACT, and education, exercise and ACT. Provision of resources, knowledge, patient empowerment, self-regulation and social facilitation can improve self-efficacy, health-related quality of life and exercise capacity in different contexts.

Conclusions: The findings of this review has identified potentially important underlying processes that contribute to the short-term increase of clinical outcomes in different contexts.

Key words: *self-management, bronchiectasis, realist review, systematic review, chronic illness*

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3 Total word count including abstract: 4,845
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9 Introduction

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11 Bronchiectasis is a long-term respiratory condition characterised by abnormal and
12 permanent dilation of the bronchi [1], impacting significantly on patients' lives with
13 persistent symptoms, repeated infective exacerbations, and reduced quality of life [2,
14 3]. Diagnosis is established through clinical history and confirmed with computed
15 tomography (CT) scan.
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24 Prevalence of bronchiectasis varies considerably due to a number of factors
25 including comorbidity [4] and access (or lack thereof) to CT scanners [5].

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27 Bronchiectasis in the UK has reported to have increased year on year during a
28 period between 2004 to 2013, with an increase in majority of age groups but the
29 most drastic increase was seen in women over the age of 70 [6]. In the US there has
30 been a report of an 8.7% annual increase in the prevalence of bronchiectasis with a
31 higher prevalence in Asian Americans relative to European and African Americans
32 [7]. The economic burden to society is significant with hospitalisation as the major
33 driver of costs, especially in patients with frequent exacerbations [8].
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45 The current aims of therapeutic management are: preservation of lung
46 function, reduction of exacerbations, minimising complications, and improving quality
47 of life. The British Thoracic Society (BTS) Guidelines suggest a five-step plan to treat
48 patients with bronchiectasis, with Step 1 for all patients and Steps 2 to 5 for those
49 with increasingly worse symptoms [1]. Therapeutic strategies and interventions
50 common to most international guidelines include antibiotics, airway clearance
51 techniques (ACT), pulmonary rehabilitation [1, 9]. The aim being to break the vicious
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3 recurrent cycle of infection, inflammation, impaired mucociliary clearance and
4 structural lung damage. In addition to the use of antibiotics to treat acute infections,
5 the use of prophylactic antibiotics, especially macrolides, has been recently
6 endorsed in national UK guidelines [10]. In the context of the increased risk of
7 antibiotic resistance and stewardship [1] the risk-benefit balance needs to be
8 considered and adherence to antibiotic therapy may be especially important for
9 people with bronchiectasis.
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20 Despite the significant costs to healthcare services the available evidence for
21 the management of bronchiectasis remains dearth [1, 12]. The burden of
22 bronchiectasis to patients is significant leading to symptoms of breathlessness,
23 wheezing, cough and chest pain, often resulting to a poorer quality of life [13, 14]
24 and clinical fatigue [15]. Exacerbations are a key feature of patients' disease burden
25 with almost 75% of patients ($n = 1403$) from the BronchUK registry reporting
26 exacerbations in a twelve-month period [16]. Adherence to treatment and prescribed
27 medication, however, may be as low as 20% in up to 50% of bronchiectasis patients
28 [17]. Reasons for low levels of adherence are diverse with research highlighting age
29 and beliefs about negative consequences of therapy as primary factors [18]. It is
30 therefore paramount that patients are supported to use self-management strategies
31 to improve adherence and learn to take control over the management of their
32 condition.
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50 Self-management is advocated by the World Health Organisation [19] and can
51 be defined as the ability to manage a chronic condition including its symptoms,
52 treatment, physical and social ramifications and lifestyle adjustments [20]. There are
53 well-established disease-specific self-management programmes for asthma, chronic
54 obstructive pulmonary disease (COPD) and cystic fibrosis [21-23] but, despite a
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3 recommendation in Step 1 of the BTS guidelines, current evidence regarding self-
4 management for bronchiectasis is very limited. Self-management in other chronic
5 respiratory diseases consists of a variety of components and approaches such as
6 exercise programmes and action plans with evidence showing that it can have
7 significant improvement in health-related quality of life (HRQoL) and a decrease in
8 healthcare utilisation [24-26]. It is imperative therefore to investigate the efficacy and
9 utility of self-management for patients with bronchiectasis; a research
10 recommendation identified by the European Multicentre Bronchiectasis Audit and
11 Research Collaboration (EMBARC) [27], the Association of Respiratory Nurse
12 Specialists UK [28], and the BTS [1].

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27 A Cochrane review of randomised control trials found insufficient evidence to
28 determine whether self-management interventions were effective in patients with
29 bronchiectasis [28]. Self-management programmes need to be developed to meet
30 different contexts and needs of subpopulations of bronchiectasis patients. A realist
31 synthesis acknowledges the variation in self-management program design and may
32 offer practical insights to produce behaviour change by uncovering causal processes
33 involved in participant behaviour. This review therefore extended the work of the
34 Cochrane review by including all study designs. An integrative systematic review
35 using realist synthesis aimed to investigate what self-management interventions
36 works, for whom and in what circumstances. We tested and refined an initial
37 programme theory that posited: a) the interventions that should be considered in
38 self-management programmes, b) the mechanisms that facilitated delivery and
39 uptake of a self-management intervention programme and c) the optimal outcomes
40 for measuring the impact of the intervention.

Method

Reporting and methods of the review followed the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) [29] and Realist and Meta-Review Evidence Synthesis: Evolving Standards (RAMESES) [30] guidelines. The full review protocol (CRD42018110103) is published on PROSPERO (<http://www.crd.york.ac.uk/PROSPERO>).

Search strategy

We searched the following sources: MEDLINE Ovid, EMBASE Ovid, CINAHL (Cumulative Index to Nursing and Allied Health Literature), EBSCO, AMED (Allied and Complementary Medicine), Web of Science Core Collection, Cochrane CENTRAL (Central Register of Controlled Trials), and the clinical trials registries (clinicaltrials.gov and ICTRP, <https://www.who.int/ictrp/en/>). Searches were conducted from inception of each database to June 2020 and were limited to English language. Details of the search strategy are available in an online Supplementary file. Handsearching of grey literature was conducted on OpenGrey (<http://www.opengrey.eu>), Grey Literature Report (<http://www.greylit.org>) and World Health Organisation (<http://who.int/en>).

Database searching was supplemented with CLUSTER (Citations, Lead Authors, Unpublished materials, Scholar searches, Theories, Early examples and Related projects) searching [31]. This maximised identification of evidence either instrumentally linked (i.e. 'sibling studies') or theoretically associated (i.e. 'kinship studies') with eligible studies [32]. This involved identification of the most influential studies (key pearl citations) and backward and forward citation searching to identify all related data. We also contacted lead authors for unpublished data and searched for the project name/identifier where applicable.

Eligibility criteria

Self-management was defined as structured support interventions for individuals with bronchiectasis designed to improve self-health behaviours and self-management skills [23]. As this review is an extension of a recent Cochrane review [28], we defined a self-management programme as one that consists of two or more interventions as illustrated in table 1, along with the full eligibility criteria following the PICO framework [33].



[INSERT TABLE 1 APPROXIMATELY HERE]

Screening and study selection

Search results were compiled using Mendeley (v. 1.19.1). Following removal of duplicates two reviewers (AT and CK) independently screened titles and abstracts for inclusion and screened the full-text of retrieved studies for eligibility. Reasons for exclusion were recorded. Disagreements were resolved by consensus or discussion with a third reviewer (DL). See figure 1 for flow chart.

Data extraction and quality assessment

Study characteristics including participants, methods, interventions and outcomes were extracted by one reviewer (AT) and verified by a second reviewer (CK). There were no disagreements regarding data extraction.

The methodological quality of eligible studies was independently assessed by two reviewers (AT and HM). The risk of bias in randomised controlled trials was assessed using the seven domains comprising the risk of bias (RoB) tool in the Cochrane Handbook of Systematic Reviews [34], with each source of bias rated as

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3 low, high or unclear. The Critical Appraisal Skills Programme Qualitative Study
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5 Checklist [35] was used to appraise qualitative studies. The quality of retrospective
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7 studies were assessed using the National Institutes for Health tool (NIH) for studies
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9 with no control group [36]. Grey literature was appraised using an Accuracy,
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11 Coverage, Objectivity, Date, Significance (AACODS) checklist [37]. We resolved
12
13 disagreements by discussion.
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16 17 *Data synthesis*

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19 All data were synthesised narratively due to the descriptions of the interventions
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21 were too heterogenous to allow for pooling. The leading candidate self-management
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23 programmes from eligible articles became the focus for the realist synthesis. Realist
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25 synthesis allowed the combining of mixed-methods data that fulfilled the needs of
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27 this particular review in which quantitative evidence were the main focus and the
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29 qualitative evidence were used for their explanatory potential. A central element of
30
31 this theory-driven approach was the development of an initial programme theory (i.e.
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33 the ideas and assumptions of how an intervention is expected to achieve particular
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35 outcomes) that described the contextual circumstances (e.g. characteristics of a self-
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37 management programme, condition of participants) where a programme would lead
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39 to particular outcomes of interest (e.g. HRQoL, frequency of exacerbations) and by
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41 which the underlying processes such as the interaction or response being offered by
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43 or embedded in a self-management programme would generate these outcomes.
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50 The programme theory developed in this review were expressed as Context-

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52 Mechanism-Outcome configurations (CMOCs) [38]. The development of our initial
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54 programme theory was achieved through the extrapolation of a previous Cochrane
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56 Review [28], evidence in other respiratory conditions [24, 39, 40] and a clinical
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58 advisory group that included experts by experience. The methods, results and
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3 discussion sections of all eligible articles were used to test and refine our initial
4 programme theory. An evidence table was developed to tabulate and organise the
5 extracted data that enabled theming of emerging patterns of CMOCs that either
6 supported or refuted the initial programme theory. An iterative process of linking of
7 these patterns was then used to formulate hypotheses to determine what works (i.e.
8 at least two interventions that constitute to a self-management programme), for
9 whom and in what circumstances. The hypotheses acted as synthesised statements
10 of findings that produced new and refined CMOCs.
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22 **Results**

23 *Description of included studies*

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25 A systematic search conducted in August 2018 and June 2020 identified 3,099
26 unique records of potentially relevant studies. Following screening of titles and
27 abstracts 3,042 records were considered irrelevant. We obtained the full texts for the
28 remaining 57 records and included 7 articles that met the review eligibility criteria.
29 Two articles were not included in the narrative synthesis as they were reviews [28,
30 41]. However, these reviews were used to refine CMOCs using author
31 interpretations. Five studies were included descriptively summarised and
32 synthesised, two of which were RCTs [42, 43], one qualitative study [44], one
33 retrospective [45] and one prospective cohort study [46]. We contacted the authors
34 of [42] for disaggregated data for the subset of bronchiectasis participants that were
35 reported in this review. Additionally, the authors of two studies [43, 46] were
36 contacted for additional information that was used in this review. The characteristics
37 of included studies are highlighted in table 2.
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3 A total of 271 patients with a confirmed diagnosis of bronchiectasis were
4 included in the review. The mean age of patients were 68.3 with a standard deviation
5 of 42.18. There were 139 females and 112 males, but one study did not report any
6 data on gender [42]. The studies were conducted in four countries including England
7 [42], Northern Ireland [43, 44], Canada [46] and Italy [45]. All patients except the
8 ones that participated in one study [42] were in a stable state condition upon
9 recruitment. The diagnostic criteria varied, four studies used a CT/high-resolution CT
10 scan [43, 44, 46] and two confirmed diagnosis via respiratory physician [42, 43].
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24 [INSERT FIGURE 1 APPROXIMATELY HERE]
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28 *Description of self-management interventions*

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30 There were a total of four interventions (education, exercise, action planning and
31 ACT) that were examined in four unique self-management programmes. One self-
32 management programme included exercise, education and ACT [45], one included
33 action planning, education and ACT [46], another included exercise and education
34 [42] and one included action planning and education [44]. Education was the most
35 prevalent intervention being included in all four self-management programmes
36 whereas, exercise, action planning and ACT were only included in two self-
37 management programmes. Exercise, action planning or ACT was included with all
38 self-management programmes that had an education component. The qualitative
39 study had explored exercise, education, symptom monitoring and ACT within their
40 focus groups.
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3 Patient educational topics ranged from general health education (e.g. nutrition
4 and self-management treatment strategies) [43, 45, 46] to more disease-specific
5 information such as dealing with the signs and symptoms of bronchiectasis and
6 understanding of an exacerbation [43, 46]. Exercise activities included daily
7 supervised volitional strength and non-volitional techniques [42], lower limb
8 endurance training [45] and upper body exercises [45, 46]. Action planning was part
9 of an EPP [43] and a bespoke action plan was used in conjunction with the BTS
10 action plan [46] that provided patients with weekly goals of their choice (e.g. picking
11 an activity) and were asked about their confidence level in achieving their goals. ACT
12 ranged from pursed lip breathing and exhalation on effort [45] to active cycle of
13 breathing and the huff cough technique [46].
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31 The shortest to longest self-management intervention programmes ranged
32 from 3 weeks [45] to 8 weeks [43]. One study is still ongoing [46]. The intensity of the
33 interventions ranged from daily sessions [42] to weekly two- and half-hour sessions
34 to between twelve and fifteen two to three-hour sessions [45]. All four studies had
35 either had a physiotherapist and/or respiratory nurse to deliver and supervise the
36 intervention. Two studies delivered the intervention in groups [43, 45]. One-to-one
37 delivery of the intervention was used in two studies and later transitioned to
38 telephone consultation [42, 46].
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51 *Outcome measures*

52 All four interventional studies reported HRQoL using either SGRQ [42, 43], the EQ-
53 VAS component of the EuroQol-5 dimensions scale [45] or QoL-B and BHQ [46].
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60 Exercise capacity was reported in two studies using either incremental shuttle walk

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3 test and endurance shuttle walk test [42] or 6MWT [45]. One study reported
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5 unplanned readmission rate at 12 months, number of hospital stays and mortality
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7 [42].
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12 Other outcomes unique to individual studies include measuring self-efficacy
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14 using the CDSS, coping as measured by IPQ-R and two EPP questionnaires were
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16 used to assess other factors such as self-rated health, ability to manage the
17
18 condition and self-rated health care use [43]. One study reported fatigue using the
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20 Borg Scale and dyspnoea was assessed by the TDI [45]. Number of clinic visits and
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22 number of patients using an oscillating positive expiratory pressure (OPEP) device
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24 was assessed in one study [46].
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30 *Quality assessment*

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33 There was consistent agreement between reviewers for study quality. The qualitative
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35 study addressing all ten CASP criteria. The unpublished study was considered low
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37 quality based on our assessment of the materials provided and available to us using
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39 the AACODS checklist. The RCTs and the retrospective study were rated as low risk
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41 of bias as they were well reported and of high methodological quality.
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47 [INSERT TABLE 2 APPROXIMATELY HERE]
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51 *Initial programme theory*

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54 Similar to previous realist syntheses [47, 48], ours considered a multiplicity of
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56 different interventions rather than one intervention that composed a self-
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58 management programme. Due to the heterogenous nature of self-management
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3 programmes that can involve combination of several interventions, our development
4 of the initial programme theory can be described as rough. We speculated that SM
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8 programmes should include at least two interventions (e.g. action planning and
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10 education) (C) that would empower bronchiectasis patients to manage their condition
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12 more effectively and to seek care in a timely manner (M) would lead to improved
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14 HRQoL, reduced exacerbation frequency and severity and are cost-effective (O).

17 *Refined programme theory*

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20 A total of three CMOCs were identified across the three leading candidate self-
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22 management programmes building on the initial programme theory. The refined
23
24 programme theory was supported using substantive theory to explain and articulate
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26 the developed CMOCs with supporting excerpts (see online supplementary file). The
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28 different underlying constructs of integrated theory of health behaviour change [49]
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30 was linked with the CMOCs. The fostering and enhancing knowledge and beliefs
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32 impacts on behaviour-specific self-efficacy and influences health behaviour change
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34 is linked with CMOC #1. The construct of self-regulation explains the management of
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36 negative emotions is associated with successful management of chronic conditions
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38 is linked with CMOC #2. The social facilitation in the form of supervision provides the
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40 social support and encouragement for behaviour change that mapped onto CMOC
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42 #3. An overarching realist programme theory in figure 2 illustrates the relationships
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44 between the CMOCs that emerged from the data.
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51 *CMOC #1 Education and action planning*

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53 Three mechanisms were identified through which a self-management programme
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55 consisting of education and action planning leads to short-term increase in self-
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57 efficacy (O₁) and HRQoL (O₂). When there are interactive sessions delivered by
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59 healthcare professionals (HCPs) (C₁), the method of teaching provided visual and
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3 auditory learning resources that enabled different ways to acknowledge new
4 information (M_1). We found evidence of posters and picture handouts depicting a
5 pulmonary exacerbation [44, 46] and the provision of YouTube videos showing the
6 use of an OPEP device [46] was one of the pathways for patients to learn about
7 bronchiectasis. We also identified patterns in the educational sessions being taught
8 that involved educational topics about signs and symptoms, goals of treatment,
9 problem solving and general health promotion (e.g. nutrition). This increased patient
10 knowledge of symptoms and their ability to recognise exacerbations, including
11 commencement of home supply of prescribed antibiotics, (M_2), may explain the
12 increase in self-efficacy.
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27 The clinician-patient partnership in these interactive sessions facilitated
28 patients to actively take part in planning their own goals empowered and increased
29 engagement in patient's own care (M_3). The evidence found indicated patients
30 actively participating in sessions by completing a worksheet about their usual
31 symptoms of bronchiectasis, how they manage them on a daily basis and triggers
32 that make the usual symptoms worse [43]. We found patterns of patients taking
33 responsibility of the status of their bronchiectasis and making their own decisions
34 through weekly action planning and goal setting. Actions plans serve as a treatment-
35 seeking support tool that gives patients the opportunity to set goals that they wish to
36 achieve in a particular week (e.g. exercise, perform a particular ACT), how they will
37 achieve it (e.g. how often they wish to perform their chosen activity) and they would
38 discuss and feedback in the following week. Patients increased engagement in their
39 own self-management would be recorded to gauge their confidence [44, 46] and
40 motivational levels [46] that could explain the positive outcomes.
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CMOC #2 Education and ACT

Education and ACT in a self-management programme for a well-informed population of patients (C₂) raises awareness to psychological and social factors (M₄) that leads to short-term increase in self-efficacy (O₁) and HRQoL (O₂) by. A lack of competence and confidence are perceived psychological factors be associated with performing ACT correctly [44]. HCPs can help address these perceived obstacles by providing relevant educational material that would encourage patients to be more independent with more aspects of their care [44]. Furthermore, feedback from patients have suggested learning how to deal with bronchiectasis socially and psychologically as part of patient education can be of great benefit in addressing these potential issues [28, 43].

Worry, fear and embarrassment are some perceived emotional issues that may affect patients' self-image, particularly when they experience an exacerbation [44]. These negative emotional states may be addressed by being taught how to differentiate symptoms between stable and exacerbation phases. There is some evidence of health-related behaviour from patients that is influenced by their attitudes and social expectations. For example, patients follow advice from family members they believe it to be important [44]. Additionally, family members can have a role in providing emotional support and assisting with performing active cycle of breathing and using the OPEP device correctly to improve HRQoL by reducing the frequency of exacerbations.

CMOC #3 Education, exercise, and ACT

Supervision of bronchiectasis patients with more severe disease (C₃) that engage in a self-management programme consisting of education, exercise, and ACT provides encouragement and support for patients (M₅) leading to short-term HRQoL (O₂) and

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2
3 short-term exercise capacity (O_3). A structured programme of exercise, education
4 and psychosocial support in the form of the SPACE manual [50] has been
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6 implemented for those with bronchiectasis [42]. Self-management strategies that can
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8 be learnt from the manual include problem solving skills, emotional management and
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10 decision making [50].
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15 The improvement in exercise capacity can be achieved when exercise
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17 sessions are individualised and supervised by HCPs. The evidence indicates that the
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19 intensity and duration of exercise sessions were individually tailored based on
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21 patient tolerance and exercise activities included super volitional and non-volitional
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23 techniques [42] and lower and upper limb training [45]. Pursued lip breathing,
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25 exhalation on effort and forward lean position were examples of ACT performed in
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27 relation to patients' needs [45]. Supervision was not always provided, particularly
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29 during the home segments of an intervention. However, telephone support was
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31 provided as an alternative [42]. Patients with more severe cases of bronchiectasis in
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33 terms of the level of airflow obstruction and the number of exacerbations
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35 experienced in previous year before being enrolled on an intervention [45] may have
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37 had an effect on the outcomes [41]. As this particular subset of bronchiectasis
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39 patients are seen as more deconditioned, therefore having a higher magnitude for
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41 improvement in exercise capacity and HRQoL compared to patients with more
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43 preserved lung functioning.
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Discussion

Key findings and comparison to wider literature

This realist synthesis contributes to the literature that acknowledges the wider contextual circumstances and attempts to explain how and why different self-management work programmes work for bronchiectasis patients. As a result of this, the review does not produce evidence of the effectiveness or advantages of self-management programmes, instead it elicits potential underlying processes why these programmes work generating particular outcomes.

In terms of our first research aim, we identified three leading candidate self-management programmes. One self-management programme included education and action planning (CMOC #1), another includes education and ACT (CMOC #2) and finally education, exercise and ACT (CMOC #3). We noted three different contextual circumstances for each CMOC that pertained to the characteristics of an self-management programme such as the interactivity of sessions (CMOC #1) and patient characteristics including being in a well-informed group of patients (CMOC #2) and a more severe diseased sub-population (CMOC #3). For our second research aim, we identified five key mechanisms that facilitated the delivery and uptake of self-management programmes. Three mechanisms generated outcomes pertaining to CMO #1. One of the mechanisms related to the resources offered by the self-management programme and four were related to patient's behavioural or psychological responses.

This review adds to the existing literature by integrating and synthesising quantitative and qualitative research, offering a more nuanced and theoretically informed analysis in a provisional realist synthesis. The overarching programme

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3 theory in this review in part answers the call for further research needed in self-
4 management for adult bronchiectasis patients [1]. In particular, this review has
5 provided explanatory accounts of the potentially most useful interventions in self-
6 management programmes for different subsets of patients. This is line with the
7 assertion that components of self-management programmes may need to
8 accommodate for condition specific patients [51]. For instance, education about the
9 disease may be less useful for a well-informed group of patients and could benefit
10 from learning how to deal with the psychosocial factors of bronchiectasis instead.
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22 The findings contributes a clearer understanding of the mechanisms that
23 emphasise the importance of different components included within self-management
24 programmes. Patient empowerment and the increase of patient engagement in their
25 own care and increasing their knowledge of symptoms are some of the identified
26 underlying processes that can trigger a behavioural change in patients. These
27 findings build on existing evidence that suggests patients become more confident at
28 managing their own health [52] through empowerment provided by self-management
29 strategies [53-57]. Additionally, one of the contexts identified reflect the importance
30 of the clinician-patient relationship where interactive sessions that involve action
31 planning that can lead to an improvement in self-efficacy via patient empowerment.
32
33 A recent study has evaluated the use of a novel action plan, the Bronchiectasis
34 Empowerment Tool [58] to facilitate appropriate treatment-seeking behaviour in
35 patients. Their findings indicated a high demand for such an intervention but
36 according to the authors it required extensive modification due to the arduous nature
37 of the tool for users. This suggests the development of treatment-seeking tools need
38 to be suitable for patients and not contribute to the burden of the disease.
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3 There are similarities between our programme theory and the integrated
4 theory of health behaviour change model [49], which was used to enhance the
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There are similarities between our programme theory and the integrated theory of health behaviour change model [49], which was used to enhance the plausibility and coherence of the patterning of identified CMOCs. Most of our concepts could be categorised using the constructs in the model such as encouragement and support as a form of social facilitation and raising awareness to psychological and social factors as an example of self-regulatory skills and abilities. The use of a realist methodology allowed us to ascertain the mechanisms and identify which contexts need to be influenced by self-management programmes to achieve outcomes of interest. The model lacks in detail regarding specific contexts, which this review fulfils. For example, the enhancement of knowledge in patients was seen interactive sessions delivered by HCPs that increase behaviour specific self-efficacy.

Methodological limitations and implications for further research

One important caveat is the strength of the evidence was limited by a lack of studies and reporting limitations of the specificity of interventions included in self-management programmes. However, we contacted authors where possible for additional information to overcome this shortcoming. There were particular studies that could have further refined and strengthened the theories but were ultimately excluded due to failing to meet our full eligibility criteria. Some noteworthy studies that were excluded include a multi-faceted qualitative study that explored the factors that affected adherence to self-management strategies [59] and three studies that either lacked in a self-management intervention [60] or only one intervention of self-management was evaluated [58, 61, 62]. The addition of these studies may have provided pertinent data to further test and refine the initial programme theory. However, it is important to emphasise this review focused on the trials identified in a

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2
3 previous review [28] and was enriched by inclusion of qualitative and grey literature.
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5 Despite using substantive theory to increase plausibility of our programme theory,
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7 the findings should be viewed as preliminary and non-exhaustive due to limited 
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10 iterative searching.
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13 Psychological aspects and the role of family members were alluded to in the
14
15 refinement of a CMOCs, however, unsubstantiated evidence did not allow us to
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17 identify specific generative causal mechanisms. Empirical evidence has
18
19 demonstrated that chronic airway diseases are associated with depression and
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21 anxiety in patients compared to controls [63]. Family members can help combat
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23 negative emotional factors associated with chronic conditions [64]. Evidence for the
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25 influence and role of social support in COPD patients have been valuable for coping
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27 in COPD patients with a positive social support being related to improved health
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29 outcomes [65] and better disease management behaviours [66].
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34 This review highlighted the paucity of evidence for symptom monitoring and
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36 adherence to treatment as components within a self-management programme.
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38 However, as our understanding of self-management in bronchiectasis is still in its
39
40 infancy, these components may be better perceived as outcomes instead as our
41
42 understanding develops. Additionally, the application of COPD training principles
43
44 was identified as key mechanism despite the significant differences between the
45
46 aetiology and physiological parameters between COPD and bronchiectasis. It is
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48 important therefore that disease-specific self-management programmes are
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50 developed to ensure education provided is appropriate and tailored for patients with
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3 Although we have identified self-management programmes for different sub-
4 populations of bronchiectasis, there is still a distinct lack of sociodemographic data
5 for the patients that participated in the self-management programmes, which may
6 limit the generalisability of our findings. There is no data in any of the reviewed
7 studies regarding patients' lifestyles, their employment status or health literacy.
8 Furthermore, majority of the patients were in a stable state condition and only one
9 study [42] had a particular focus on patients who were currently experiencing an
10 acute exacerbation. Due to the lack of studies to pattern findings, we were unable to
11 develop CMOCs pertaining to this particular subset of patients. We therefore
12 propose that future studies should aim to clearly define and justify the specific nature
13 of self-management, whether the patients are newly diagnosed (i.e. less than a year)
14 and include sociodemographic data as this information will help further refine the
15 programme theory to elicit a better understanding of which self-management
16 programmes are needed for which subset of patients. A future realist evaluation
17 could potentially fill these gaps currently in the literature by using primary data, which
18 could further refine the programme theory developed in this review.
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41 Conclusion

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43 Given the magnitude of the global burden of bronchiectasis and the complexity of
44 self-management interventions, this review has developed a programme theory
45 describing contexts where different self-management programmes work and the
46 underlying processes that generate health-related outcomes, albeit short-term. We
47 have highlighted the pivotal role of self-management for adult bronchiectasis patients
48 and the necessity of testing the multiple self-management programmes identified in
49 this review should be the focus of future research in this area.
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Declaration of interest: The authors report no conflict of interests.

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Table 1. Study selection criteria

	Inclusion	Exclusion
Population	Adults (≥ 18 years old) with bronchiectasis as defined by study authors	Confirmed diagnosis of cystic fibrosis, sarcoidosis, primary ciliary dyskinesia or active allergic bronchopulmonary. Studies with mixed respiratory populations were excluded unless data were available for bronchiectasis alone
Intervention	Experimental studies that examined self-management programmes that included at least two of the following interventions in an interventional group: patient education, symptom monitoring, airway clearance techniques, adherence to treatment including medication, an exercise or physical component and action planning (i.e. treatment-seeking support tools). Pulmonary rehabilitation programmes were eligible when studies explicitly included a self-management support intervention(s) within the programme. Non-experimental studies that explored two or more components were included	Interventions solely comprising patient education or those focused only on exercise, such as pulmonary rehabilitation delivered in a care setting
Comparator	Sole interventions, e.g. education alone and usual care	
Outcome	All primary and secondary outcomes as defined by study authors were included irrespective of follow-up duration	

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Table 2. Characteristics of included studies

Study ID	Study design	Total N (n of bronchiectasis patients) of participants	Mean age (SD), % male	Diagnostic criteria	Severity of condition when recruited	Baseline lung function (SD)	Self-management interventions	Total duration and frequency of intervention	Outcomes	Key findings
Lavery et al. (2007), Northern Ireland	Focus groups	32	64.4 ^a male: 44%	CT scan	Not reported	3 patients had very severe impairment, 12 severe and 17 moderate based on the GOLD guidelines	Education, exercise, symptom monitoring and airway clearance techniques ^b	N/A	N/A	Bronchiectasis had an impact on patients' physical and psychosocial well-being. Patients demonstrated the potential of SM strategies including perceived sources of support and barriers to SM

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3	Lavery	RCT	64,	Intervent	Respirat	Stable	FEV ₁ (%	Action	One	Self-	The
4	et al.		intervention	ion: 60	ory	state	predicte	planning	session	efficacy,	disease-
5	(2011)		: <i>n</i> = 32;	(9);	physicia		d);	and	per week	coping,	specific
6	,		control: <i>n</i> =	control:	n		intervention:	education	(lasting	HRQoL	EPP
7	<i>Northe</i>		32	60 (8),	assessm		59%		(lasting	and two	significantly
8	<i>rn</i>			male:	ent		(20);		2.5	standard	improved
9	<i>Irelan</i>			45%	including		control:		hours)	EPP	self-efficacy
10	<i>d</i>				a CT		65%		for eight	questionn	in 60% of
11					scan		(23)		weeks	aires were	the CDSS
12										used to	subscales
13										assess the	immediately
14										following:	postinterve
15										self-rated	ntion. There
16										health,	was no
17										ability to	improvement
18										manage	in terms
19										the	of coping as
20										condition,	measured
21										adherence	by the IPQ-
22										to	R.
23										medicatio	Intervention
24										n, self-	group
25										rated	reported
26										health	more
27										care use,	symptoms
28										satisfactio	and
29										n and	decreased
30										benefits	HRQoL at 3
31										obtained.	and 6
32										Other	months
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34										included	ntion
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									FEV ₁ and frequency of oral and/or intravenous antibiotic therapy prescribed	
Greening et al. (2014), England	RCT	389 (20 [intervention: $n = 8$; control: $n = 12$] ^a)	Intervention: 78 (7.8); control: 68.8 (11.5), male: 100%	Respiratory physician ^c	Acute exacerbation	MRC dyspnoea grade: intervention group: 4.9 (0.35); control group: 4.4 (0.67) ^a	Exercise and education ^d	Daily strength training over six weeks	Unplanned readmission rate at 12 months, time to first readmission, number of hospital stays, mortality, exercise capacity and HRQoL	There was no significant differences in readmission rates, SGRQ total scores, and in mortality between groups at any time point.

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3	Zanini	Retrospe	108	71 (13),	HRCT	Stable	FEV ₁ (%	Exercise,	15	Exercise	Significant
4	et al.	ctive		male:	scan	state	predicte	education	sessions	capacity,	improvements
5	(2015)	cohort		45%			d) 76	and	(lasting	HRQoL,	in
6	, Italy						(27)	airway	between	fatigue	6MWT and
7								clearance	2-3	and	EQ-VAS
8									hours)	dyspnea	was found
9									over a		after PR in
10									three-		all patients,
11									weeks		$p < .001$ [35
12											(43), 15
13											(12),
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15]. TDI
16											improved
17											significantly
18											by 4 (2.6), p
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4	LeClerc and Muggah (2019), Canada	Prospective cohort	47	72, male: 37%	CT scan	Stable state	Mean score of 6 on the BSI	Education, airway clearance and action planning	Not reported	HRQoL, number of visits, number of patients using OPEP	The use of the OPEP device increased in 72% ($n = 22$) of participants following the first educational session. The SM programme improved patients' quality of life as measured by the QoL-B and BHQ by decreasing the symptoms they experienced and increased their capacity for physical activity. The
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programme encouraged and motivated patients to continue with their ongoing daily therapy

^aThis was reported as a median

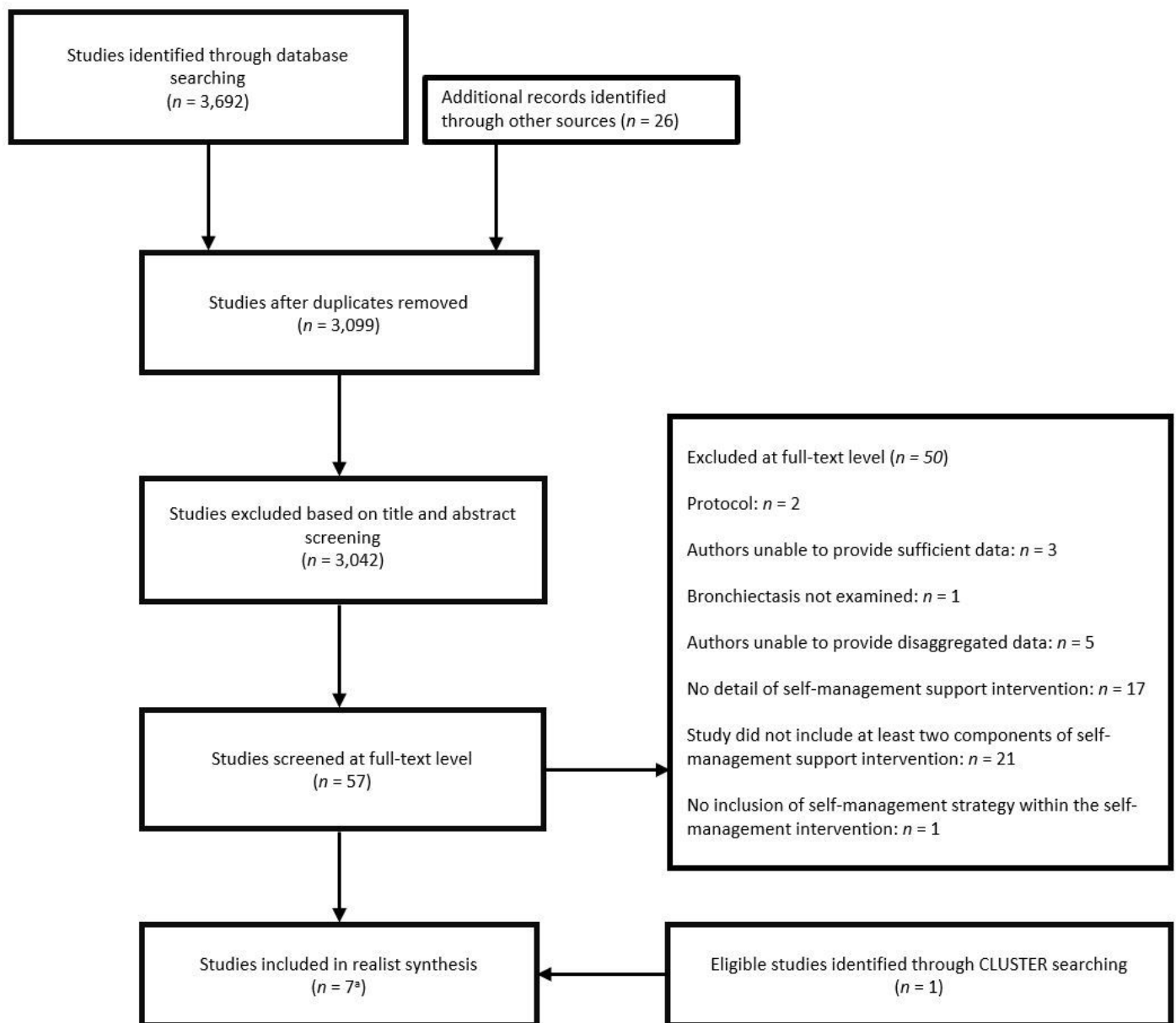
^bThese were self-management strategies discussed in the focus groups

^cData was provided by contacting the author as disaggregated data was not present in study

^dThe education component was delivered using the SPACE manual

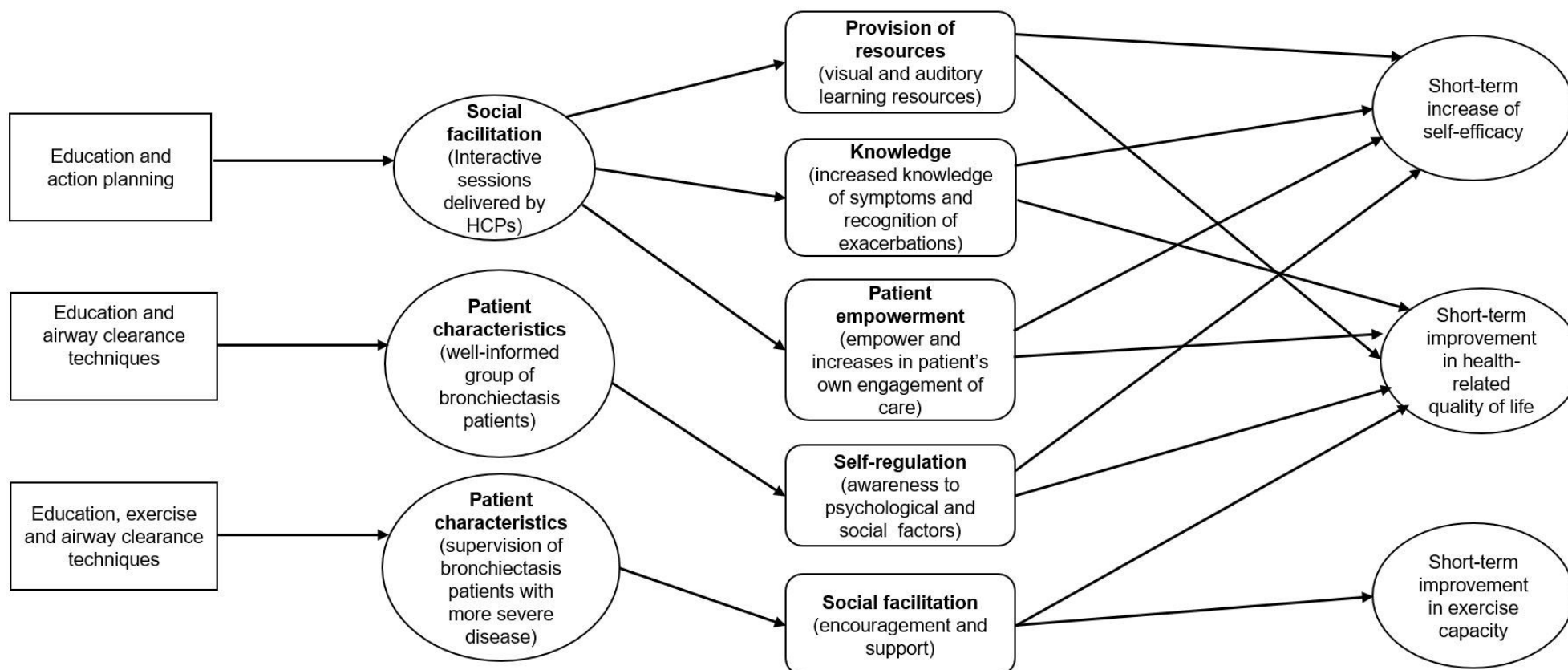
^eAll data was preliminary based on a conference abstract and information provided by the author

Notes: m, mean; SD, standard deviation; CT, computed tomography; GOLD, The Global Initiative for Chronic Obstructive Lung Disease; N/A, not applicable; SM, self-management; RCT, randomised controlled trial; FEV1, force expiratory volume in one second; HRQoL, health-related quality of life; EPP, Expert Patient Programme; CDSS, Chronic Disease Self-efficacy Scale; IPQ-R, Revised Illness Perception Questionnaire; MRC, Medical Research Council; SPACE, Self-management programme of Activity, Coping and Education; SGRQ, St George's Respiratory Questionnaire; HRCT, high-resolution computed tomography; 6MWT, 6 minute walking test; EQ-VAS, EuroQol visual analogue scale; PR, pulmonary rehabilitation; TDI, Transition Dyspnoea Index; BSI, Bronchiectasis Severity Index; OPEP, oscillating positive expiratory pressure; QoL-B; Quality of Life-Bronchiectasis; BHQ, Bronchiectasis Health Questionnaire

Figure 1. Flow chart of summary of study selection

*Two articles were reviews and were not included in the narrative synthesis

Figure 2. Overarching programme theory



Supplementary Data

Supplement to: **A systematic review of self-management programmes for bronchiectasis in adult patients: a realist synthesis of the evidence**

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AMED OVID, CENTRAL, CINAHL EBSCO, EMBASE OVID, MEDLINE OVID, PSYCHINFO, Web of Science Core Collection

Supplementary Table S1: Search strategy in AMED, EMBASE, MEDLINE, PSYCHINFO using OVID search and CENTRAL¹

#	Search terms
1	exp Bronchiectasis/
2	Bronchiect* .mp
3	"bronchoect".mp
4	"kartagener".mp
5	(bronchial* adj3 dilat*).mp
6	1 or 2 or 3 or 4 or 5
7	exp Self Care/
8	exp Education/
9	exp Patient Education/
10	educat*.mp
11	self-manag*.mp
12	self-car*.mp
13	train*.mp
14	Instruct*.mp
15	patient-cent*.mp
16	patient focus* .mp
17	management-plan.mp
18	Behavio\$r.mp
19	(management* adj1 program*).mp
20	(disease* adj2 management*).mp
21	Self-efficac*.mp
22	Empower*.mp
23	(symptom adj2 management*).mp
24	7-23 combined with OR
25	6 AND 24
26	Limit 25 to English language

¹MeSH DESCRIPTOR was used instead of exp due to compatibility issues

Supplementary Table S2: Search strategy in CINAHL using EBSCO

#	Search terms
1	TX: exp Bronchiectasis/
2	TX: Bronchiect*
3	TX: bronchoect*
4	TX: kartagener
5	TX: bronchial* adj3 dilat*
6	1-5 combined with OR
7	TX: exp Self Care/
8	TX: exp Education/
9	TX: exp Patient Education/
10	TX: educat*
11	TX: self-manag* OR "self manag*"
12	TX: self-car* OR "self car*"
13	TX: train*
14	TX: instruct*
15	TX: patient-cent* OR "patient cent*"
16	TX: patient-focus* OR "patient focus*"
17	TX: patient-education OR "patient education"
18	TX: management-plan OR "management plan"
19	TX: behavio\$r*
20	TX: management* NEAR1 program*
21	TX: disease* NEAR2 management*
22	TX: self-efficac*
23	TX: empower*
24	TX: symptom NEAR2 management*
25	TX: 7-24 combined with OR
26	TX: 6 AND 25
27	TX: Narrow by language: English

Supplementary Table S3: Search strategy in Web of Science Core Collection

#	Search terms
1	TOPIC: bronchiectasis or bronchiect* or bronchoect* or kartagener or bronchial NEAR/3 dilat*
2	TOPIC: educat* or self-manag* or "self-manag*" or self-car* or "self-car*" or train* or instruct* or patient-cent* or "patient-cent*" or patient-focus* or "patient-focus*" or patient-education or "patient education" or management* or self-efficac* or empower* or symptom NEAR/2 management*
3	#1 AND #2
4	Refined by languages: English

Supplementary Table S4: Quality assessments for eligible studies ($n = 5$)

Cochrane risk of bias assessment for Greening et al. (2014)

Domain	Authors' judgement	Support for judgement
<i>Selection bias</i>		
Random sequence generation	Low risk	Quote from study: "The clinical trials unit at the

		University of Leicester coordinated randomisation by an automated internet-based service”
Allocation concealment	Low risk	Each participant admitted was randomly allocated using sealed envelopes to reveal which group they were assigned to.
<i>Performance bias</i>		
Blind of participants and personnel	Low risk	Not feasible to blind participants to treatment group. Single blind study. Intervention group unlikely to influence objective outcomes.
<i>Detection bias</i>		
Blinding of outcome assessment	Low risk	Quote, “..all investigators performing the outcome measures were blinded to treatment allocation...”
<i>Attrition bias</i>		
Incomplete outcome data	Low risk	Quote, “multiple imputation by changed equations to replace missing data...”
<i>Reporting bias</i>		
Selective reporting	Low risk	All outcomes were pre-specified
<i>Other bias</i>		
Other sources of bias	High risk	Baseline disease severity and differences in age for the subset of participants with bronchiectasis was not balanced.

Cochrane risk of bias assessment for Lavery et al. (2011)

Domain	Authors' judgement	Support for judgement
<i>Selection bias</i>		

Random sequence generation	Low risk	Quote, "Computed-generated concealed randomisation process conducted by an independent person not involved in the research"
Allocation concealment	Low risk	Quote, "...an envelope corresponding to the patient's number was opened to reveal if they were assigned to the intervention or control group"
<i>Performance bias</i>		
Blind of participants and personnel	Low risk	Due to the nature of the intervention it was not possible to blind participants. It is however, unlikely that the knowledge of being in an intervention group would influence objective outcomes
<i>Detection bias</i>		
Blinding of outcome assessment	Low risk	Quote, "Data were collected by trained health professionals who were blinded to participants' groups and also had no other contact with study patients. Study participants also were requested not to disclose their group assignment"
<i>Attrition bias</i>		
Incomplete outcome data	Low risk	Quote, "Missing data was not replaced for analysis because there were minimal missing values"
<i>Reporting bias</i>		
Selective reporting	Low risk	All outcomes were pre-specified
<i>Other bias</i>		
Other sources of bias	High risk	Power analysis was not performed as it was a proof of concept study. Inadequate sample size may have led to misleading results.

CASP for Lavery et al. (2007)

Criteria	Yes	Can't tell	No
1. Was a clear statement of the aims of the research?	√		
2. Is a qualitative methodology appropriate?	√		
3. Was the research design appropriate to address the aims of the research?	√		
4. Was the recruitment strategy appropriate to the aims of the research?	√		
5. Was the data collected in a way that addressed the research issue?	√		
6. Has the relationship between researcher and participants been adequately considered?	√		
7. Have ethical issues been taken into consideration?	√		
8. Was the data analysis sufficiently rigorous?	√		
9. Is there a clear statement of findings?	√		
10. How valuable is the research?	√		

NIH for Zanini et al. (2015)

Criteria	Yes	No	Other (CD, NR, NA)*
1. Was the study question or objective clearly stated?	√		
2. Were eligibility/selection criteria for the study population prespecified and clearly described?	√		
3. Were the participants in the study representative of those who would be eligible for the test/service/intervention in the general or clinical population of interest?	√		
4. Were all eligible participants that met the prespecified entry criteria enrolled?	√		
5. Was the sample size sufficiently large to provide confidence in the findings?			NR
6. Was the test/service/intervention clearly described and delivered consistently across the study population?	√		
7. Were the outcome measures prespecified, clearly defined, valid, reliable, and assessed consistently across all study participants?	√		
8. Were the people assessing the outcomes blinded to the participants' exposures/interventions?		√	
9. Was the loss to follow-up after baseline 20% or less? Were those lost to follow-up accounted for in the analysis?			NA
10. Did the statistical methods examine changes in outcome measures from before to after the intervention? Were statistical tests done that provided <i>p</i> values for the pre-to-post changes?	√		
11. Were outcome measures of interest taken multiple times before the intervention and multiple times after the intervention (i.e., did they use an interrupted time-series design)?			NA
12. If the intervention was conducted at a group level (e.g., a whole hospital, a community, etc.) did the statistical analysis take	√		

into account the use of individual-level data to determine effects at the group level?			
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*CD, cannot determine; NA, not applicable; NR, not reported

Quality rating: Low risk of bias
Authors' comments: Well reported and of high methodological quality. It was not possible to blind participants to intervention due to the nature of the intervention.

ACCORDS checklist for LeClerc and Muggah (2019)

ACCORDS domains		Yes	No	Can't tell
Authority	Identifying who is responsible for the intellectual content			
	Individual author:			
	Associated with a reputation organisation?	√		
	Professional qualifications or considerable experience?	√		
	Produced/published other work (grey/black) in the field?	√		
	Recognised expert, identified in other sources?			√
	Cited by others? (use Google Scholar as a quick check)		√	
	Higher degree student under "expert" supervision?		√	
Accuracy	Does the item have a clearly stated aim or brief?		√	
	If so, is this met?			√
	Does it have a stated methodology?			√
	If so, is it adhered to?			√
	Has it been peer-reviewed?		√	
	Has it been edited by a reputable authority?		√	
	Supported by authoritative, documented references or credible sources?		√	
	Is it representative of work in the field?	√		
	If No, is it a valid counterbalance?	√		
	Is any data collection explicit and appropriate for the research?	√		
	If item is secondary material (e.g. a policy brief of a technical report) refer to the original. Is it accurate, unbiased interpretation or analysis?		√	
Coverage	All items have parameters which define their content coverage. These limits might mean that a work refers to a particular population group, or that it excluded certain types of publication. A report could be designed to answer a particular question, or be based on statistics from a particular survey. Are any limits clearly stated?		√	

Objectivity	It is important to identify bias, particularly if it is unstated or unacknowledged.			
	Opinion, expert or otherwise, is still opinion: is the author's standpoint clear?			√
	Does the work seem to be balanced in presentation?		√	
Date	For the item to inform your research, it needs to have a date that confirms relevance			
	Does the item have a clearly stated date related to content? No easily discernible date is a strong concern.	√		
	If no date is given, but can be closely ascertained, is there a valid reason for its absence?	√		
	Check the bibliography: have key contemporary material been included?		√	
Significance	This is a value judgment of the item, in the context of the relevant research area			
	Is the item meaningful? (this incorporates feasibility, utility and relevance)	√		
	Does it add context?	√		
	Does it enrich or add something unique to the research?	√		
	Does it strengthen or refute a current position?	√		
	Would the research area be lesser without it?		√	
	Is it integral, representative, typical?			√
	Does it have impact? (in the sense of influencing the work or behaviour of others)	√		

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Supplementary Table S5: Context-mechanism-outcome configurations linked with self-management programmes and substantive theory with illustrative data

Integrated theory of health behaviour change construct	SM programme	Context	Mechanism	Outcome	Supporting excerpts
knowledge and beliefs	Education and action planning	(1) Provision of interactive sessions delivered by HCPs	<p>(1) Visual and auditory learning resources provided by different ways to acknowledge new information</p> <p>(2) Increasing knowledge of symptoms and ability to recognise exacerbations</p> <p>(3) Empowers and increases engagement in patient’s own care</p>	<p>(1) Short-term increase in self-efficacy</p> <p>(2) Short-term increase in HRQoL</p>	<p>“Self-management support empowers the person with the condition by enabling them to modify treatment or behaviour, or to seek professional advice...” [28]</p> <p>“Patients were keen to have a greater knowledge about bronchiectasis and more open access to healthcare services” [44]</p>
Self-regulation	Education and ACT	(2) Well-informed population of bronchiectasis patients	(4) Raises awareness to psychological factors	<p>(1) Short-term increase in self-efficacy</p> <p>(2) Short-term increase in HRQoL</p>	<p>“Improvement in psychological health (e.g., anxiety, depression, well-being) was seen in other studies evaluating the EPP, and patients in this</p>

					<p>study appeared to benefit psychologically, indicated in the satisfaction gained and benefits obtained sections of the EPP questionnaire..." [43]</p> <p>" The added benefit of education, which included instruction in self-management, the importance of airway clearance techniques, and nutritional advice, may account for the greater magnitude of benefit in HRQOL" [41]</p>
Social facilitation	Education, exercise and ACT	(3) Supervision of bronchiectasis patients with more severe disease	(5) Encourage and Support for patients	(2) Short-term increase in HRQoL (3) Short-term improvement in exercise capacity	"Motivational interviewing techniques were used to introduce patients to the manual and to familiarise them with the content. The manual was used throughout the participants' inpatient stay and in

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					<p>the subsequent discussions during telephone calls.” [28, 42]</p> <p>“The self-management programme comprised the SPACE manual, a structured programme of exercise, education, and psychosocial support” [28, 42]</p> <p>“patients with a more severe disease, both in terms of airflow obstruction and hyperinflation and the number of exacerbations in the previous year, showed a higher magnitude of improvement in 6MWD and EQ-VAS score after” [45]</p>
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For Peer Review