Interventions to Support Informal Caregivers of People with Chronic Obstructive Pulmonary Disease: A Systematic Literature Review

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Abstract

Background

Chronic obstructive pulmonary disease (COPD) imposes tremendous challenges for both patients and informal caregivers. Caregivers are key players in the management of COPD. Recently, COVID-19 further increased reliance on informal caregivers who urgently need specific support. This systematic literature review aimed to systematically describe the content and explore the effects of interventions to support informal caregivers of people with COPD.

Methods

A mixed-methods systematic review was conducted. PubMed, Scopus, Web of Science, and EBSCO were searched. Studies implementing interventions supporting informal caregivers of people with COPD were included. Data were extracted and analysed in outcome domains and categories using framework analysis.

Results

Twenty (14 quantitative, 4 mixed-methods, and 2 qualitative) studies were included. Informal caregivers were mainly female (86%). Caregiving context was poorly/never described. Interventions included patient-caregiver dyads and never caregivers only. Informal caregivers were invited to participate if available. Interventions were delivered across all COPD phases (acute/stable/advanced) and settings (inpatient/outpatient/home), with a wide range of total length,
frequency, and duration of sessions. All included education about the disease and its management. Discharge/action plans (n = 12); adherence to therapy and healthy lifestyles (n = 9); and family concerns and psychosocial issues (n = 7) were also commonly addressed. Only 9 (45%) studies reported caregiver-related outcomes, and overall positive effects were observed in 7/9 outcome domains, using a high variety of qualitative and qualitative methods. Often categories were addressed but not assessed.

**Conclusion**

Interventions have a narrow scope (i.e., education) and have not been specifically designed to support informal caregivers. Current evidence showed positive effects, but high methodological heterogeneity exists. Future studies need to explore caregiver-tailored, taking into consideration gender differences; multicomponent; and flexibly administered interventions to effectively support COPD caregivers.

**Keywords:** Loved ones; Significant others; Carer; Chronic obstructive pulmonary disease; Therapeutics; Caregiving

**Introduction**

Chronic obstructive pulmonary disease (COPD) is a clinically heterogeneous syndrome [1], which imposes tremendous daily challenges not just to patients but also to informal caregivers [2]. Informal caregivers are commonly defined as any relative, partner, friend, or neighbour who has a significant personal relationship with, and provides a broad range of assistance for, a person with a chronic or disabling condition [3].

A large number of people with COPD need informal care [4]. The average number of caregiving hours has been shown to be 9.5 h per day, ranging between 6.7 h in non-dependent to 12.7 h in greatly dependent people [4]. The pandemic COVID-19 has further increased the reliance on informal caregivers who seem to be the forgotten full-time workers [5]. Caregivers are often responsible for medication, appointments, and symptom management; awareness of hygiene, nutritional needs, and exercise regimes; providing physical, psychological, and emotional support; recognizing and managing emergency issues; physical work; handling equipment; managing care recipient behaviour and temperament; handling household- and illness-related finances; and carrying out decision-making and problem-solving needs [6]. It is, therefore, often and wrongly assumed that informal caregivers have the appropriate level of health literacy, disease knowledge, financial security, household conditions, psychological readiness, and medical care abilities [6]. Nevertheless, their multiplicity of roles, together with changes in family dynamics and financial responsibilities, indicates a need for caregivers to have specific knowledge and develop a wide range of skills to support themselves and their loved ones with COPD. It is therefore not surprising they ask for specific support [7]. Information from interventional studies to support informal caregivers is however scarce. Synthesizing such information is of paramount importance to guide the development and encourage the implementation of future support interventions for these key players in the management of COPD, which will also contribute to the better management of future large-scale public health emergencies [5]. This systematic literature review aimed to systematically describe the content and explore the effects of interventions to support caregivers of people with COPD.

**Methods**

A mixed-methods systematic literature review of studies implementing interventions to support informal caregivers of people with COPD was conducted. Prior to commencing the review, searches in the Cochrane Library and the International Prospective Register of Systematic Reviews (PROSPERO) were conducted to exclude the existence of reviews or protocols with similar purpose. This systematic review protocol was registered at PROSPERO (registration no. CRD42020189929). This systematic review was reported according to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines [8].

**Search Strategy**

A systematic electronic literature search was conducted in 4 electronic databases: PubMed, Scopus, Web of Science and EBSCO from their inception to 28 May 2020. The combination of keywords used can be found in online suppl. Table 1 (for all online suppl. material, see www.karger.com/doi/10.1159/000517032). Weekly automatic updates, retrieved from the databases, were conducted until March 2021. Search terms were limited to title, abstract, and
Eligibility Criteria

Studies were included if they implemented an intervention that supported informal caregivers of people with COPD, with or without patients; were randomized controlled trials, quasi-experimental, observational, or qualitative studies; and were written in English, French, Italian, Portuguese, or Spanish languages. We excluded articles which (i) included informal caregivers of patients with respiratory diseases other than COPD, unless COPD data were reported separately; (ii) reported only on psychometric properties of measurement tools to measure outcomes of any domain of the effects of interventions applied to informal caregivers of people with COPD; and (iii) only assessed patients’ perspective of care and/or investigated caregivers’ needs and expectations. Abstracts in conference proceedings were also excluded.

Study Selection

After duplicates were removed, studies were selected based on their titles and abstracts. Those that met the inclusion criteria, or uncertainty remained about its inclusion or exclusion, were retrieved, and the full-text article was read carefully before a decision was made. The selection process (eligibility and screening) was completed by 1 researcher (A.M.); other authors were consulted in cases of uncertainty. A random sample of 10% of abstracts were independently screened by a second author (J.C.) to guarantee consistency.

Quality Assessment

All studies were independently assessed by 2 researchers (A.M. and J.C.) using the QualSyst tool [13], which allows the evaluation of quality of both quantitative and qualitative studies. This tool is composed of 2 scales. The quantitative scale has 14 items with a maximum possible sum of 28 points, and the qualitative scale has 10 items with a total possible sum of 20 points. Each item is scored according to the extent to which criteria are met (“yes”: 2, “partial”: 1, and “no”: 0). Total sum is therefore the number of “Yes”*2 plus the number of “partials”*1. In the quantitative scale, 5 items can however be marked as “not applicable,” when appropriate, and be excluded from the calculation of the summary score. In this case, the total possible sum is then calculated as 28 – (number of “N/A” *2). Summary score is calculated by dividing the total sum by the total possible sum and multiplied by 100 to provide a percentage [13]. Higher percentages indicate better quality but categorization of studies based on their scores is not possible; thus, we have used a previous proposed approach and classified studies with scores >80% as strong, 60–79% as good, 50–59% as adequate, and below 50% as poor [14]. No scale for mixed-methods studies is available; therefore, those studies were assessed according to the more dominant method described, as reported previously [15]. Scoring disagreements were resolved by discussion, until consensus was reached.

The consistency of the quality assessment performed by the 2 reviewers was explored with an inter-rater agreement analysis using the Cohen’s kappa. The value of Cohen’s kappa ranges from 0 to 1 and can be categorized as slight (<0.2), fair (0.21–0.4), moderate (0.41–0.6), substantial (0.61–0.8), or almost perfect (>0.81) agreement [16]. The statistical analysis was performed using IBM SPSS 25.0 (IBM, Armonk, NY, USA).

Data Extraction

Data from included studies were extracted to a structured tabular format comprising the following topics: study (first author, year of publication, country); study design; participants (number of informal caregivers, age, sex; number of people with COPD, age, sex, forced expiratory volume in 1 s percentage predicted); information on caregiving (relationship with the person with COPD, time spent caregiving, and living or not with care recipient); timing (acute, stable, and terminal/advanced); setting (inpatient, home, and outpatient); who delivered the intervention and to whom; intervention description (intervention name, type/content, frequency, and duration); caregiver’s outcomes and tools used to assess each outcome; and caregivers findings (means ± standard deviations, medians [interquartile ranges], frequency, percentages pre- and post-interventions, and effect sizes in quantitative studies when reported; representative quotes in qualitative studies). Data were extracted by 1 researcher (A.M.) and checked by the other 2 researchers (J.C. and D.B.) for accuracy and completeness.

Data Analysis
Intervention outcomes were extracted from each study and analysed using framework analysis [17, 18]. Framework analysis [17–19] was chosen because it is a highly systematic and flexible method of categorizing and organizing data based on commonalities and differences found [18].

First, 1 author, with clinical experience in COPD management, outcome measurement, and interventions with informal caregivers, grouped together verbatim outcomes, from the included studies, with the same semantic meaning, into categories (e.g., Burden Interview and Caregiver Appraisal Scale were both grouped under the burden category based on an evaluation of the content of the scales). Then, using a deductive approach, those categories were mapped into 8 domains (shown in Table 1) that have been previously defined in the literature [15]. If an outcome emerged that could not be mapped into one of the pre-specified domains, a new category or domain was generated as recommended [17–19]. After categorizing all data, the number of studies addressing each domain/category during the intervention and/or assessing caregivers’ outcomes related to the categories of the final framework was counted. Frequent discussions were taken among authors to ensure agreement in the analysis of qualitative data.

<table>
<thead>
<tr>
<th>Domains</th>
<th>Categories</th>
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<tbody>
<tr>
<td>1. Family-related</td>
<td>Communication within the family, family concerns, family coping, and (dys)functional relationships</td>
</tr>
<tr>
<td>2. Health system/family-health professional communication</td>
<td>Access and use, communication between family and health professionals, participation of the family in decision-making, and family preferences of healthcare</td>
</tr>
<tr>
<td>3. Information/education</td>
<td>Adherence to therapy and healthy lifestyles, knowledge about the disease, and strategies to support the patient</td>
</tr>
<tr>
<td>4. Interpersonal/intimacy</td>
<td>Body image, connectedness, and intimacy with the partner</td>
</tr>
<tr>
<td>5. Physical/cognitive</td>
<td>Symptom management and cognitive (dys)function</td>
</tr>
<tr>
<td>6. Practical/daily living</td>
<td>Disease management, equipment handling, exercise, housekeeping, financial strain, funeral preparation, and transportation</td>
</tr>
<tr>
<td>7. Psychosocial/emotional</td>
<td>Anxiety, burden, burnout, depression, despair, end-of-life issues, mood, fear/worry, optimism, satisfaction, and stress</td>
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<tr>
<td>8. Social/societal</td>
<td>Socialization and social support</td>
</tr>
</tbody>
</table>

**Results**

**Study Selection**

A total of 3,913 records were identified. After removal of duplicates, 1,906 remained to be screened for relevant content through title, abstract, and keywords. From these, 1,875 were excluded. The full text of 31 articles was analysed, and 15 were excluded due to the following reasons: (i) not possible to differentiate COPD data from other respiratory diseases \(n = 8\), (ii) intervention was focused on the patient only \(n = 2\), (iii) intervention included formal caregivers \(n = 1\), and (iv) there was no intervention \(n = 4\). Additionally, 1 article was retrieved from weekly updates and 3 others were obtained from manual hand searching of references list. Twenty full-text articles [20–39] were included (shown in Fig. 1).
Quality Assessment

Quality assessment mean score was 74% (95% CI 68–79%), ranging from 54 to 96%. Six (30%) studies were rated as strong, 10 (50%) as good, and 4 (20%) as adequate quality (online suppl. Tables 2, 3). Percentage of agreement between reviewers was 80%, with $k = 0.78$ (95% CI = 0.59–0.97; $p < 0.0001$), rated as substantial.

Study Characteristics

Detailed characteristics of the included studies are shown in Table 2 and online suppl. Tables 4–6. Studies were published between 2002 and 2020 and were conducted in 11 different countries: Australia ($n = 3$), Canada ($n = 1$), Hong Kong ($n = 1$), Iceland ($n = 3$), Iran ($n = 1$), the Netherlands ($n = 1$), Portugal ($n = 3$), Slovenia ($n = 1$), Sweden ($n = 1$), Spain ($n = 1$), UK ($n = 1$), and USA ($n = 3$). Almost 50% ($n = 9$) were published after 2015.
Summary of caregiver domains and categories being addressed with the respective tools used for data collection across studies

<table>
<thead>
<tr>
<th>Caregiver domain and category</th>
<th>No. of studies addressing the domain/category in the intervention</th>
<th>No. of studies assessing the domain/category</th>
<th>Tools used for data collection</th>
<th>Summary of the main findings with representative quotes, whenever available</th>
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<tbody>
<tr>
<td><strong>Domain: family-related</strong></td>
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<tr>
<td>Family adjustment to illness</td>
<td>–</td>
<td>2 [35, 36]</td>
<td>PAIS-SR [35, 36]</td>
<td>Pre 29.9±13.9 versus post 25.8±12.1 (*p = 0.242) [35] EG: Pre 22.7±9.7 versus post 14.9±5.9 [26]</td>
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<td>Communication within the family</td>
<td>4 [26, 32, 35, 36]</td>
<td>–</td>
<td>–</td>
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<tr>
<td>Family concerns and psychosocial issues</td>
<td>7 [25, 26, 32, 33, 35, 36, 38]</td>
<td>–</td>
<td>–</td>
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<tr>
<td>Family coping</td>
<td>5 [26, 30, 35, 36, 38]</td>
<td>4 [26, 33, 35, 36]</td>
<td>F-COPES [26, 35, 36]</td>
<td>Pre 93.1±12.9 versus post 110.4±11.2 (*p = 0.011) [35]</td>
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<td></td>
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<td></td>
<td>Individual interviews [33]</td>
<td>EG: Pre 91.4±15.7 versus post 106.7±12.1 (*p = 0.011) [35]</td>
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<td></td>
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<td>Focus group [26, 35]</td>
<td>&quot;We receive service as a family because I need support as much as my husband. I need to be able to talk about how I feel. That is absolutely necessary... [33]</td>
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<td></td>
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<td>Focus groups [35, 38]</td>
<td>&quot;Our relationship has changed. Before this programme, we did not know how to deal with the disease...&quot; [35]</td>
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<tr>
<td>Family identity</td>
<td>3 [26, 35, 36]</td>
<td>–</td>
<td>–</td>
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<tr>
<td>(Dys)Functional relationships</td>
<td>–</td>
<td>2 [35, 38]</td>
<td>Focus groups [35, 38]</td>
<td>&quot;Even my children were involved in the programme, they asked for information to help us&quot; [35]</td>
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<tr>
<td><strong>Domain: health system/family-health professional communication</strong></td>
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<tr>
<td>Access and use</td>
<td>5 [20, 21, 24, 25, 30]</td>
<td>4 [25–27, 33]</td>
<td>CANHELP Questionnaire [27]</td>
<td>CANHELP questionnaire (points)</td>
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<td>Focus groups [26]</td>
<td>Pre 4.40 versus post 4.80 [27]</td>
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<td></td>
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<td>Individual interviews [25, 33]</td>
<td>&quot;It is such a great comfort knowing I have help from the doctors and nurses if I needed it.&quot; [27]</td>
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<td></td>
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<td>Service use questions [25]</td>
<td>&quot;People with COPD and family members should attend to these programs&quot; [26]</td>
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<tr>
<td>Communication</td>
<td>6 [19, 26, 32, 35–36]</td>
<td>3 [24, 33, 35]</td>
<td>Focus groups [35]</td>
<td>&quot;The receptivity of the professionals was very important, the phone calls that they made us...The</td>
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Individual interviews [24, 33] professionals worried a lot with our family! I received a call from them asking if I was better.” [35] “There are many health care professionals who see us...we never get into trouble anymore if something unexpected happens.” [33]

| Family preferences | 2 [33, 37] | Individual interviews [33] Satisfaction questionnaire [37] “With needs and preferences of the patients and families focal in the interprofessional collaboration, the clinic became an established component of health care for people with COPD and their families.” [33] Carers from EG were significantly more likely than those from CG to prefer domiciliary care (*p = 0.001) [37]

| Participation of the family in decision-making | 1 [33] | Individual interviews [33] “Families ... gained insight into manifestations of the disease and how to interpret them, and they became more assured and certain about what they needed and wanted for themselves” [33]

| Referral to treatment services | 6 [22, 24, 25, 27, 32, 34] | – | – |

**Domain: information/education**

| Adherence to therapy and healthy lifestyles | 9 [20–22, 26, 27, 32, 35, 36, 38] | 1 [26] | Focus groups interviews [26] “I now do some exercise too, and he says ‘You have to do it [the exercise] like that,’ and I follow his commands” [26]

| Coping strategies | – | 3 [20, 26, 33] | Ways of Coping Questionnaire Knowledge [20] Focus groups [26] Individual interviews [33] “He had a severe crisis, and what I’ve learned here was really helpful...” [26]

| Knowledge about the disease | 20 [19–38] | 7 [20, 22, 26, 27, 33, 35, 38] | Adapted Understanding COPD Questionnaire [20] CANHELP Questionnaire [27] Preparedness for caregiving scale [20, 22] PROMIS [22] Focus groups [26, 35] Individual interviews [33, 38] “I must say that I really learnt a great deal” [38] “When we forgot the techniques, we used the handbook at home to remember them” [26] “He had a very strong crisis, and what really helped me was reminding him on how to breathe, the positions to reduce breathlessness.” [35]


**Domain: interpersonal/intimacy**

| Connectedness | 1 [36] | 2 [26, 38] | Focus groups “It helped creating connectedness in our

|
and intimacy

Individual interviews [38]

relationship and in issues related to the disease” [26]

Domain: physical/cognitive

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<td>“He had a very strong crisis, and what really helped me was reminding him on how to breathe, the positions to reduce breathlessness.” [35]</td>
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<td>“Looking back I feel everything has been done so far to the best of all of our ability to make my mother comfortable. She is doing very well, improving a lot, so there is nothing more I can think of to help her other than new lungs” [27]</td>
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Domain: practical/daily living

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<td>EG: pre 21.2 ± 15.5 versus post 18.8 ± 18.6</td>
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Domain: psychosocial/emotional/spiritual

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<tr>
<td>EG: pre 6.2±4.3 versus post 5.2±3.6 (*p = 0.02)</td>
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<tr>
<td>CG: pre 6.7±4.3 versus post 6.7±4.8 (p = 0.98) [28]</td>
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<td>“It is normal that we get involved... and we start to feel anxious. With this knowledge, we have learned to use strategies to overpass problems” [26]</td>
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<td>EG: pre 93.1±12.9 versus post 110.4±11.2</td>
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<tr>
<td>CG: pre 92.5±20.3 versus post 92.5±17</td>
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<td>“... it was good to have the opportunity to talk about this and that and not all next of kin experience things in the same way. We were provided with information and had the opportunity to express our thoughts and I said that it’s more than ‘only’ being a next of kin, as he is not the only one who has a difficult time, but also we as relatives.” [38]</td>
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<tr>
<th>Depression</th>
<th>3 [22, 32, 35]</th>
<th>1 [28]</th>
<th>HADS [28]</th>
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<tbody>
<tr>
<td>EG: pre 3.9±3.6 versus post 3.6±3.6 (p = 0.6)</td>
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<tr>
<td>CG: pre 4.6±4.2 versus post 4.8±4.7 (p = 0.48, p = 0.72) [28]</td>
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<tr>
<td>“It is absolutely hopeless” [24]</td>
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|--------------------------|-----------------|------|-----------------
| EG: pre 21.2±15.5 versus post 18.8±18.6 |
Studies included participants with COPD \((n = 2,022, 52\% \text{ male}, \text{ mean age} = 69.1 \text{ years}, \text{ mean FEV}_{1} = 45.2\% \text{ predicted})\) and their caregivers \((n = 683, 14\% \text{ male}, \text{ mean age} = 57.5 \text{ years})\). None of the studies included caregivers only. Number and characteristics of participating caregivers were not reported in 6 studies \([22, 23, 25, 27, 29, 36]\). Information on caregiving was often \((n = 10)\) not provided \([20, 22–25, 29–31, 36, 38]\). Relationship with the person with COPD was reported in 9 studies \([21, 26–28, 33–35, 37, 39]\), and most caregivers were relatives: spouses \((n = 223; 41\%)\), unspecified relative \((n = 149; 22\%)\), child \((n = 106; 19\%)\); or other relatives \((n = 55; 10\%)\). Only 3 studies provided information about the number of years caregivers had been caring for the person with COPD \([32–34]\), and most were caring between 2 and 4 years \((n = 50; 66\%)\). Information on whether or not the caregiver was living with

<table>
<thead>
<tr>
<th>End-of-life issues and advanced care planning</th>
<th>3 [25, 27, 28]</th>
<th>1 [28]</th>
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<th>–</th>
</tr>
</thead>
<tbody>
<tr>
<td>Goals, values, beliefs, and wishes</td>
<td>1 [28]</td>
<td>–</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td><strong>Stress</strong></td>
<td>4 [23, 35, 36, 38]</td>
<td>3 [20, 22, 25]</td>
<td>Caregiver stress and coping ([20, 22]); PROMIS support measures ([22]); VAS ([25]); Individual interviews ([25])</td>
<td>–</td>
</tr>
<tr>
<td><strong>Quality of death and dying</strong></td>
<td>–</td>
<td>1 [28]</td>
<td>QODD Questionnaire ([28])</td>
<td>QODD questionnaire (points) (\text{EG: Mdiff} = 80\pm8.5 \ (p = 0.17)) (\text{CG: Mdiff} = 74.7\pm11.5 \ (p = 0.17)) ([28])</td>
</tr>
</tbody>
</table>

**Domain: social/societal**

- **Maintaining relationships** | – | 1 [26] | Focus groups \([26]\) | “We have all the contacts and we are trying to organize some group outdoor activities” \([26]\) |
- **No. of times carer goes out** | – | 1 [25] | Self-reported counts \([25]\) | – |

**Domain: quality of life**

- **Health-related quality of life** | – | 2 [25, 27] | SEIQoL-DW \([25]\) | – |

A summary of the quantitative findings is presented and representative quotes are provided, when available, to facilitate interpretation of qualitative findings. Quantitative data are reported as mean ± standard deviation, unless otherwise stated.

CANHELP Questionnaire, Canadian Health Care Evaluation Project Questionnaire; CAS, Caregiver Appraisal Scale; CRQ, The Chronic Respiratory Disease Questionnaire; COPD, chronic obstructive pulmonary disease; CADI, Carers’ Assessment of Difficulties Index; CG, control group; DASS-21, Depression Anxiety and Stress Scales; EG, experimental group; F-COPES, Family Crisis Oriented Evaluation Personal Scales; HADS-A, Hospital Anxiety and Depression Scale – Anxiety; mBorg, modified Borg scale; Mdiff, mean difference; PAIS-SR, Psychosocial Adjustment to Illness Scale – Self-Report; PROMIS, Patient-Reported Outcomes Measurement Information System for the domains of informational and emotional support; QODD, Quality of Death and Dying Questionnaire; SEIQoL-DW, Schedule for the Evaluation of the Individual Quality of Life – Direct Weighting; VAS, Visual Analogue Scale; \(\eta^2\), eta squared – measure of effect size.

* \(p \) value <0.05 considered statistically significant.
the person with COPD was provided by 2 studies [28, 34], and most caregivers were living with the person with COPD (n = 45; 79%).

Fourteen studies followed a qualitative [21–23, 25, 27–29, 31, 33, 35–39], 4 a mixed-methods [20, 24, 32, 34], and 2 a qualitative [26, 30] methodology. Findings for caregiver-related outcome domains were only reported by 9 studies [20, 21, 26, 28, 30, 32–34, 37]: 4 only administered questionnaires [21, 28, 33, 37], 2 used a combination of questionnaires and focus groups [32, 34], and 3 used conversational [26] or semi-structured individual interviews [20, 30] (Table 2; online suppl. Tables 4–6).

Design of the Interventions

Most interventions started during the acute phase of COPD [20–22, 25, 27, 29, 35, 36, 38, 39], often as early as possible after hospital admission [20, 21, 27, 29, 35, 36, 38], although in one of these, the intervention could start either during admission or after discharge [39], and in another, the intervention started few days after discharge [25]. There were, however, also 6 interventions starting during the stable [26, 30–34] and 4 during the terminal/advanced [23, 24, 28, 37] phases of COPD.

Studies were mainly conducted in inpatient settings and at home (n = 7) [20–22, 27, 29, 35, 36] or outpatient settings only (n = 6) [30–34, 39]. Three studies were conducted only at home [23, 24, 28, 37], 2 in outpatient settings and home [25, 26], and 1 in inpatient setting only [38]. A multidisciplinary team composed of nurses, physicians, and physiotherapists delivered the interventions [22–25, 28–34, 38, 39]; however, in 2 studies, dyads (patients-caregivers) [39] and volunteered patients [31] were also part of the team.

Interventions were provided to patients and caregiver as individual dyads (n = 13) [20–29, 35–37], to patients and caregivers integrated in group discussions (n = 6) [30–34, 39], or to patients and caregivers separately (n = 1) [38], but never to caregivers only. In 5 studies, interventions were integrated within pulmonary rehabilitation programmes [23, 30, 32–34]. Comparisons between interventions provided to both patients and caregivers with those delivered to patients only were conducted in 5 studies [21, 31, 33, 34, 38].

Total duration of interventions ranged from 1 session [30, 37] to 6 months [25, 28, 31, 39]. Frequency of sessions ranged from once [30, 37] to daily [21, 27], and in some cases, frequency was undetermined, as it was stated that it varied as needed [25, 26, 35, 36]. Duration of each session varied from 20 min [38] to 2 h [30].

Content and Outcomes of the Interventions

All interventions (n = 20) incorporated a component of education (written and/or oral) about the disease and its management (e.g., management of symptoms, medication, and prevention and management of exacerbations) [20–39]. The 3 other aspects most commonly described within the interventions were discharge/action plans (n = 12) [20, 24, 27–29, 32–37, 39], adherence to therapy and healthy lifestyles (n = 9) [28, 30–36, 39], and family concerns and psychosocial issues (n = 7) [24, 26, 30–34] (shown in Table 2; online suppl. Tables 4–6).

Although all interventions included caregivers, their outcomes were not systematically assessed. In 8 (40%) studies, there was no mention of any caregiver outcomes being assessed [22, 23, 25, 27, 29, 31, 36, 38], and 11 (55%) studies did not report any caregiver finding [22–25, 27, 29, 31, 35, 36, 38, 39]. Among the 9 (45%) studies that reported caregiver findings [20, 21, 26, 28, 30, 32–34, 37], a high heterogeneity of domains/categories was assessed quantitatively with questionnaires [20, 21, 28, 32–34, 37] and qualitatively with interviews [20, 24, 26, 30] and focus groups [32, 34], or combining both methods [20, 32, 34].

Quantitative methods were used to report data on family adjustment to illness (n = 2) [32, 33], family coping (n = 3) [32–34], access to and use of healthcare systems (n = 1) [28], family preferences (n = 1) [21], coping strategies (n = 1) [35], knowledge about the disease (n = 3) [28, 35, 39], disease management (n = 2) [24, 28], anxiety (n = 2) [34, 37], burden (n = 2) [24, 28, 34], depression (n = 1) [37], emotional status (n = 2) [34, 39], stress (n = 3) [23, 28, 32], quality of death and dying (n = 1) [37], and quality of life (n = 2) [24, 28]. Qualitative methods were used to report data on family coping (n = 3) [26, 32, 34], (dis)functional relationships (n = 2) [30, 32], access to and use of healthcare systems (n = 3) [24, 26, 34], family-health professional communication (n = 3) [20, 26, 32], family preferences and their participation in decision-making (n = 1) [26], adherence to therapy and healthy lifestyles (n = 1) [34], coping strategies (n = 2) [26, 34], knowledge about the disease and disease management (n = 4) [26, 30, 32, 34],
knowing what to expect ($n = 1$) [20], complicity and intimacy ($n = 2$) [30, 34], disease management ($n = 1$) [32], equipment handling ($n = 1$) [20], anxiety ($n = 1$) [34], burden ($n = 1$) [30], despair ($n = 1$) [20], stress ($n = 1$) [24], and maintaining relationships ($n = 1$) [34]. Results per domain will now be presented.

**Family-Related**

Eight (40%) studies [24–26, 30–34] addressed this domain within the interventions. Nevertheless, only 5 studies (25%) assessed family-related categories [26, 30, 32–34] and reported their findings on 3 categories: family adjustment to illness, family coping, and (dys)functional relationships. The category most frequently addressed was family concerns and psychosocial issues ($n = 7$) [24, 26, 30–34], and the most assessed was family coping ($n = 4$) [26, 32–34]. Family coping improved in caregivers after participating in an intervention integrated in a pulmonary rehabilitation programme ($p = 0.011$) [32], exceeding the improvements in the control group ($0.01 \leq p \leq 0.048$) [33, 34]. The importance of receiving the intervention as a family within pulmonary rehabilitation was acknowledged by caregivers as an opportunity to talk about their feelings, validate their experiences, and keep them going in their caregiving role [26, 34].

No improvements in total score of family adjustment to illness were observed after pulmonary rehabilitation [32, 33]. Nevertheless, involvement/support of the family increased and relationships changed positively [30, 32]. Family concerns and psychosocial issues [24, 26, 30–34], communication within the family [31–34], and family identity [32–34] were categories addressed but not assessed in the intervention provided.

**Health System/Family-Health Professional Communication**

Thirteen (65%) studies [20, 24, 25, 27–29, 31–36, 39] addressed this domain. Nevertheless, only 7 (35%) [20, 21, 24, 26, 28, 32, 34] assessed and 6 (30%) [20, 21, 26, 28, 32, 34] reported findings on 4 associated categories: access and use, communication, family preferences, and participation of the family in decision-making. Caregivers valued having access to supportive professionals and services and participating in the interventions, and therefore, they would recommend it to others [34]. They felt secure and comfortable knowing they had support from health professionals [26, 28, 34], especially due to the uncertainty that characterizes the disease trajectory [26]. Two other categories, family preferences of healthcare and family participation in decision-making, emerged from the analysis [21, 26], with families feeling more empowered to make informed decisions about what they wanted and needed [21, 26]. Although addressed, referral to treatments [20, 24, 28, 29, 31, 39] was not assessed in the studies.

**Information/Education**

All studies addressed this domain [20–39], but only 8 (40%) [20, 26, 28, 30, 32, 34, 35, 39] assessed and 6 (30%) [20, 26, 28, 30, 32, 34] reported findings on 3 categories: adherence to therapy and healthy lifestyles, knowledge about the disease, and knowing what to expect. “Knowledge about the disease” was addressed by all studies [20–39] and was also the category assessed the most [26, 28, 30, 32, 34, 35, 39]. Adherence to therapy and healthy lifestyles was covered in 9 (45%) studies [28, 30–36, 39], but findings were reported by only 1 study [34]. As a result of these interventions, caregivers started to do some exercise [34] and reported learning a great deal of useful information to better adjust to the demands of the disease [26, 30, 32, 34]. Caregivers also acknowledged having developed coping strategies [26, 34] and emphasized the importance of knowing what to expect from the future [20], after participating in the interventions. Nevertheless, no study addressed “knowing what to expect” in the interventions.

**Interpersonal/Intimacy**

Only 1 study [33] described to have addressed this domain, but 2 studies [30, 34] assessed and reported findings on connectedness and intimacy category. Caregivers considered the intervention to have had a positive impact on creating connectedness [34] and in understanding the intimacy [30] with their loved ones.

**Practical/Daily Living**

All studies addressed this domain in the category disease management [20–39], but only 4 (20%) [20, 24, 28, 32] assessed and 3 (15%) [20, 28, 32] reported findings on disease management and equipment handling categories. Findings showed that after the intervention, caregivers felt more able to handle challenging situations, such as an acute exacerbation [32]. Caregivers also valued learning how to deal with daily equipment [20] and having validated their sense of doing their best when caring for their loved ones at home [28]. Discharge/action plans [20, 21, 24, 27–29, 32–
identification of problems at home [21, 26, 27, 29], and transition of care [35, 36] were categories addressed but never assessed in the interventions.

**Psychosocial/Emotional/Spiritual**

Ten (50%) studies [23, 28, 30–34, 37, 39, 40] addressed but only 8 (40%) [20, 24, 28, 30, 31, 34, 35, 37, 39] assessed and 5 (25%) [20, 28, 30, 31, 34, 37] reported findings on 8 associated categories of this domain. Most studies (n = 6) focused on anxiety [30–32, 39], depression [31, 32, 39], and overall emotional management [30, 32–34, 39]. Nevertheless, end-of-life issues and advance care planning [24, 28, 37] and goals, values, beliefs, and wishes [37] were also addressed in studies conducted with caregivers of people with advanced COPD. Significant decreases (p = 0.02) in levels of anxiety were reported [37] and also perceived by caregivers [34] after participating in the interventions. The acquired knowledge and the opportunity of discussing their thoughts and experiences were identified as main contributors to decrease anxiety levels [34] and burden [30]. No significant differences between groups were found in burden [34], depression [37], overall emotional status [34], or perceived quality of death and dying [37] when measured with the questionnaires. Despair was not addressed in any intervention and caregivers stated to felt hopeless [20].

**Social/Societal**

This domain was never addressed but was assessed by 2 (10%) studies [24, 34] and findings were reported by 1 study [34]. Nevertheless, group discussions, namely integrated within pulmonary rehabilitation, encouraged caregivers to maintain contacts among each other and keep active [34]. The physical and cognitive domain was never addressed or assessed. Caregivers' quality of life was also never addressed. Nevertheless, 2 studies [24, 28] assessed it, but no findings were reported. More categories were addressed than assessed and lack of coherence between what was addressed and assessed within the interventions was observed (shown in Fig. 2).

**Fig. 2.**

Number of studies addressing (columns above the zero line) and assessing (columns below the zero line) each category per domain in the interventions aimed to support informal caregivers of people with COPD. Colours are representative of domains: dark purple, family-related; purple, health system/family-health professional communication; dark blue, information/education; blue, interpersonal/Intimacy; dark green, practical/daily living; green, psychosocial/emotional; orange, social/societal; yellow, quality of life. The difference between the number of studies addressing and assessing the different categories is indicated by the small squares in each column. COPD, chronic obstructive pulmonary disease.
Discussion

This systematic literature review showed that interventions aimed to support informal caregivers have been delivered by multidisciplinary teams to patient-caregiver dyads (never to caregivers only), across all phases of COPD (acute/stable/advance) and settings (inpatients/outpatients/home), with a wide range of total duration as well as frequency and duration of sessions. A very small proportion (14%) of informal caregivers were male. Little or no information on caregiving context has been reported. All interventions included education about the disease and its management, and discharge/action plans, adherence to therapy and healthy lifestyles, and family concerns and psychosocial issues were also commonly addressed. Other equally important aspects (e.g., connectedness, socialization, and quality of life) were rarely or never addressed. Although caregiver-related outcomes were reported by less than half of the studies using a large variety of quantitative and qualitative tools, overall positive effects were observed in various categories related to family, healthcare system/family-health professional communication, information/education, interpersonal/intimacy, practical/daily living, psychosocial/emotional/spiritual, and social/societal domains. Lack of coherence between what was addressed versus assessed within the interventions was also demonstrated.

The natural history of a chronic disease is a dynamic and individual process, especially in such a high complex, heterogeneous, and unpredictable disease such as COPD [1], and therefore, support needs are disease-phase and person-specific. Most of that support is ensured by informal caregivers [4, 41, 42]; however, interventions to support informal caregivers across phases of COPD and settings have been somewhat standardized in their content and style of administration (face-to-face to dyads with telephone monitoring), not taking into account the specific needs of informal caregivers and their loved ones with COPD. Education is an important component of interventions to support informal caregivers of people with COPD as they often have expressed the need to know more about the disease and its management [2, 43, 44]. Delivered to dyads (patient-caregiver), it enhances the competence of the caregiver to provide, and the person with COPD to receive, care; however, interventions to reduce the amount of care provided by caregivers (e.g., respite) and/or to improve the caregiver’s well-being and coping skills (e.g., psychoeducational interventions and support groups) are equally important [45] and were sporadically or never addressed. One-size-fits-all approach to support informal caregivers will be of limited use as they have a multiplicity of roles in COPD management [2, 6, 46] and vastly different needs over time [2, 40, 43, 46–48], which include their own. Moreover, it is already known that symptoms, health status, and coping styles differ among female and male informal caregivers [49, 50]. Therefore, routine assessment of caregiver’s needs in multiple domains and time points seems fundamental to designing multicomponent, tailored, and flexibly administered interventions to effectively support their changing needs over time, as previously acknowledged [10, 11, 40]. Such interventions should not focus only on the negative (e.g., burden and distress) but also on the positive (e.g., satisfaction and personal growth) aspects of caregiving to emphasize their fundamental role and strengths and to build a greater resilience [51]. The type and methodology to deliver such interventions is still unclear [46]. Pulmonary rehabilitation is a multicomponent intervention that has been perceived as a successful and personalized opportunity to support the whole family (not just patients) throughout the chronic lung disease trajectory [2, 30, 32–34, 40, 43, 52, 53], but this topic warrants further investigation [2, 53]. Moreover, a reflection on having a patient-caregiver dyad [39] or caregivers only as members of the multidisciplinary team during the delivery of interventions might be of added value, as it might increase clinical, emotional, and social significance of the interventions to caregivers.

Despite the relative narrow scope of interventions provided, beneficial effects to support caregivers in several outcome domains were found and the qualitative data validated the importance and relevance of such interventions to caregivers’ lives. Similar positive results have been previously reported in those caring for people with lung cancer and other chronic illnesses [51, 54, 55]. Nevertheless, careful interpretation of these benefits is recommended, as interventions do not seem to be designed to support caregivers, instead they were invited to participate if available; therefore, their involvement and information on caregiving were poorly described, and a significant number of studies (55%) [22–25, 27, 29, 31, 35, 36, 38, 39] did not report any caregiver findings. Moreover, the high heterogeneity observed in the timing, setting, designs, type and dose of interventions, measurement tools used, and absence or inconsistent reporting of statistical tests and effect sizes makes comparisons about the effects of interventions difficult and hinders an effective synthesis of evidence [56, 57]. This high methodological heterogeneity, together with the lack of coherence observed between what was being addressed versus assessed in the interventions, constitutes a waste of research opportunity and ultimately delays the development of clinical guidelines. There is, therefore, a need for high-quality research to explore the effectiveness of informal caregiver support interventions.
Nevertheless, results from this systematic review provide clinicians and researchers with guidance on the design of interventions taking into considering gender-specific differences to support informal caregivers of people with COPD on their multiple outcome domains/categories and respective measurement tools. Although much room for improvement exists, approximately half of the studies have been published in the last 5 years, which shows this is an exciting new area of research to improve the lives of all of those living with COPD (patients and informal caregivers).

Strengths of this review include a thorough systematic search and screening according to the well-established PRISMA guidelines [8]; the inclusion of quantitative and qualitative studies with respective quality assessment; and the synthesis of studies conducted across COPD phases and settings to provide a comprehensive overview of the existing interventions to support informal caregivers of people with COPD. Nevertheless, some limitations also need to be acknowledged. First, only peer-reviewed publications included in databases were searched. Additional interventional studies may exist in the unpublished grey literature. Second, the way caregivers were involved in the interventions was often unclear. It is therefore possible that some studies had the involvement of informal caregivers, but poor reporting led to their exclusion. Third, the effect sizes of the interventions cannot be determined from qualitative data; however, quantitative data were scarcely described and qualitative data provide valuable information to guide the development of future interventions. Finally, some degree of interpretative bias may have occurred as qualitative synthesis consists of a researcher’s interpretation. To minimize this risk, a second researcher was used to seek confirmation of the results and interpretation and frequent discussions among research team members were conducted.

Conclusion

This mixed-methods systematic literature review provided a comprehensive synthesis of interventions to support caregivers of people with COPD. Interventions have been provided to patient-caregiver dyads and never to caregivers only, by multidisciplinary teams, across all COPD phases and settings, with a wide range of total length and frequency and duration of sessions, and have mainly focused on education about the disease and its management. Engagement of male caregivers has been scarce. Although caregiver-related outcomes were reported by less than half of the studies, overall benefits have been reported in 7/9 outcome domains/categories. High methodological heterogeneity exists, making comparisons across interventions challenging. Despite this, our findings provide clinicians and researchers with guidance on the design of interventions to support informal caregivers of people with COPD. It also provides recommendations to consider gender-specific differences and routinely assess caregiver’s outcomes to design methodologically robust, caregiver-tailored, multicomponent, and flexibly administered interventions, focused not just on the negative but also on the positive aspects of caregiving, to effectively support informal caregivers over time.

Statement of Ethics

This systematic review was reported according to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines [8]. The protocol was reviewed and approved (it is published) at International Prospective Register of Systematic Reviews (PROSPERO) (registration No. CRD42020189929).

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

Funding Sources

This work was partially funded by Programa Operacional de Competitividade e Internacionalização – POCI, through Fundo Europeu de Desenvolvimento Regional – FEDER (POCI-01-0145-FEDER-007628) and Fundação para a Ciência e a Tecnologia (PTDC/SAL-SER/28806/2017 and UIDB/04501/2020).

Author Contributions

A.M. obtained the funding, conceived the idea, extracted the data, and performed the analysis. A.M., J.C., and D.B. contributed to the design and interpretation of data. A.M. drafted the manuscript. A.M., J.C., and D.B. critically revised the manuscript and approved the final version.

References


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