Development and evaluation of a palliative rehabilitation blended learning program to support family caregivers of people with ALS and cognitive and/or behavioral impairments

- a complex intervention study focusing on acceptance and perspectives

PhD dissertation

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List of original articles

This PhD dissertation is based on the following original articles which will be referred to by their Roman numbers.

Article I **Reflections of family caregivers and health professionals on the everyday challenges of caring for a person with amyotrophic lateral sclerosis and cognitive impairments: a qualitative study**

Olesen, L.K; la Cour, K.; With, H.; Handberg, C.

Published in: Palliative Care and Social Practice. 2022, February.

Article II **A cross-sectional evaluation of acceptability of an online palliative rehabilitation program for family caregivers of people with amyotrophic lateral sclerosis and cognitive and behavioral impairments.**

Olesen, L.K; la Cour, K.; With, H.; Mahoney, A.; Handberg, C.

Published in: BMC Health Services Research. 2022, April.

Article III **Experienced benefits and challenges of an online palliative rehabilitation program for family caregivers of people with amyotrophic lateral sclerosis.**

Olesen, L.K; la Cour, K.; Nimmon, L.; With, H.; Handberg, C.

In preparation. Planned submitted to: Journal of Evaluation and the Health professions.

Article IV **Online peer-support among family caregivers of people with amyotrophic lateral sclerosis and cognitive impairments in a palliative rehabilitation blended learning program**

Olesen, L.K; la Cour, K.; Thorne, S.; With, H.; Handberg, C.

In review: Journal of Evaluation in Clinical Practice. September 5th, 2022.

The articles are presented at the end of the dissertation

Abbreviations

Family caregivers	Caregivers (spouses/partners)
PALS/CIs	Person(s) with ALS and cognitive and/or behavioral impairments
ALS	Amyotrophic Lateral Sclerosis
PALS	Person(s) with ALS
ALS/CIs	ALS and cognitive and/or behavioral impairments
WHO	World Health Organization
RCFM	The national Rehabilitation Center for Neuromuscular Diseases, Denmark
MRC	The British Medical Research Council
ID	Interpretive Description methodology
SOC	Sense of Coherence
TFA	Theoretical Framework of Acceptability
HW	Heidi With, group facilitator
MBSR	Mindfulness-bases Stress Reduction

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Preface

Family caregivers, like spouses and partners of persons with amyotrophic lateral sclerosis and cognitive and/or behavioral impairments are highly affected and burdened by the rapidly progressive and devastating neurodegenerative disease of their partners. They struggle with multiple challenges and experience various needs which call for a palliative rehabilitation intervention that supports their comprehensibility, manageability, and meaningfulness of everyday challenges related to and the loss of the persons with amyotrophic lateral sclerosis and cognitive and/or behavioral impairments. Interventions that meet and support these multiple and complex needs are still missing from exiting literature.

The aim of this PhD dissertation was thus to develop and evaluate a new palliative rehabilitation blended learning program targeting and supporting family caregivers of persons with amyotrophic lateral sclerosis and cognitive and/or behavioral impairments in dealing with everyday challenges and needs related to the affected partner.

Background

The dissertation focuses on family caregivers (hereafter referred to as caregivers), i.e., spouses and partners, of persons with amyotrophic lateral sclerosis and cognitive and/or behavioral impairments (PALS/CIs). To ease the reading and set the context of the PhD project, a brief presentation of ALS and cognitive and behavioral impairments is provided initially followed by an introduction to the caregiver role and burden and the challenges and needs experienced by caregivers in everyday life. This is followed by a description of a palliative rehabilitation approach and finally, previous support initiatives.

Amyotrophic lateral sclerosis (ALS)

ALS is an incurable heterogeneous neurodegenerative disorder that affects upper and lower motor neurons [1]. The symptoms comprise muscle weakness, spasticity, dysarthria, dysphagia, respiratory failure, and subsequent paralysis of the muscles [2]. ALS often has a focal onset and later relentlessly spreads to different body regions [1]. Respiratory failure mostly attributes to death within 2-5 years from disease onset [1]. The ALS-incidence in Europe is increasing, ranging from 2.1 – 3.8 per 100,000 [3]. Because there is no cure, most medical care focuses on symptom management, slowing down progression, prolonging respiratory function and addressing dysphagia [4] as well as offering palliative care to maximize quality of life for the persons with ALS (PALS) and their caregivers [5]. PALS often stay at home with their spouse or partner becoming the primary caregiver and thereby serving as an important component in the provision of care [6].

ALS and frontotemporal dementia

ALS is described as a multisystem disorder because of the clear clinicopathological relationship with frontotemporal dementia [7, 8]. The extra motor-manifestations in relation to ALS vary in degree and forms and concern changes in behavior, language impairments, and executive dysfunctions [9]. In approximately 50 % of the ALS-cases, the cognitive and behavioral impairments occur at a mild to moderate degree [9, 10]. In 10-15%, the symptoms are severe enough to fulfill the diagnostical criteria for frontotemporal dementia [11-14]. Most frequent changes in cognition concern the executive dysfunctions and language impairments [15]. The behavioral impairments concern apathy, stereotyped behavior, disinhibition, loss of sympathy and empathy, and dietary changes [15]. Recognizing and paying attention to the behavioral and/or cognitive impairments is crucial since they are related to significantly shorter survival time [16-18]. In fact, persons with ALS and frontotemporal dementia have a nearly three times higher risk of early death than people who “only” have ALS [19], which might be explained by noncompliance in the use of medical interventions [18-20]. In this dissertation the abbreviation “PALS/CIs” is used to describe the person(s) with ALS regardless of whether the impairments are related to the cognitive or behavioral impairments or both.

Caregivers

Living with a partner who suffers from the devastating and fatal ALS disease is a complex and difficult experience [21, 22]. Research shows that caregivers are highly affected by the deterioration of their partners who have ALS as they struggle with a very demanding role and high burden [23, 24]. ALS is described as a family illness because partners often assume primary responsibilities for the comprehensive and complex care that inevitably will increase as ALS progresses [21]. For instance, they are expected to provide physical and emotional support and assist in decision-making [23] without being trained [25]. Furthermore, a recent study found that caregivers experience difficulties with practical issues related to the extra responsibilities that come with the caregiving role [26-28], such as managing the household and finances [4, 29]. Nevertheless, they frequently refrain from seeking or accepting support due to difficulties in balancing personal time with caregiving responsibilities [30].

The heavy caregiving tolls decrease the caregivers' quality of life [31, 32] as they struggle with limited time and a restricted social life [26, 27] which, in turn, cause feelings of social isolation [33]. ALS results in many transitions for the caregivers, one being the change in their relationship from being a spouse/partner to becoming a caregiver [4, 29]. Such transitions are inescapable with ALS and cognitive and/or behavioral impairments (ALS/CIs), and a study found that psychological distress in caregivers of PALS may be related to lower perceived control over caregiving [34]. Caregivers experience difficulties with emotional issues from witnessing their partner's health deteriorate, role changes, and their own distress [28]. A scoping review on supportive care needs of PALS and their caregivers found that the caregivers experience emotional problems like hopelessness, despair, frustration, sadness, feeling drained, depression, and fear [35]. In fact, psychological distress is higher in caregivers than in PALS, and the caregiver burden increases consistently throughout the trajectory [6].

The presence of cognitive and behavioral impairments (e.g., apathy, disinhibition, and executive impairments) in PALS is known to cause a high caregiver burden [36-40]. Lillo et al. (2012) found that caregivers are more burdened by behavioral impairments in the PALS than the physical disability caused by ALS [39]. Further, behavioral impairments like disinhibition, impulsivity, apathy, and changes in eating habits, of the PALS are negatively associated with caregiver burden and quality of life [25, 32, 34, 37, 41]. Research shows that caregivers of PALS and frontotemporal dementia are three times more likely to report a higher burden, assessed by the Zarit Burden Interview, than those caring for PALS without frontotemporal dementia [42], which underlines the negative consequences of the cognitive and behavioral impairments on the caregivers' everyday lives and leisure [42-45]. Thus, caregivers of PALS/CIs have been placed in the highest group of all caregivers for stress [46]. Major and minor depression in ALS caregivers have been reported [24, 47] and anxiety and depression are long-term predictors for burden [48]. Lack of information for caregivers can evoke negative emotional effects such as anxiety, depression, and uncertainty [33, 35]. It is therefore not surprising that they experience a significant need for information earlier in the trajectory in order to prepare for and handle the disease as it progresses [4]. Nor that they express a need for practical, social, informational, psychological, physical, emotional and spiritual support [35]. Further, they express a need to share feelings, emotions, and experiences with people who are in similar situations [49]. Research shows that ALS causes lasting impact on the caregivers after the death of their spouse, and involvement of caregivers of deceased PALS is therefore important when trying to create awareness of the caregivers' needs [50].

Palliative rehabilitation

The World Health Organization (WHO) states that all people should have access to universal health coverage, which includes prevention, promotion, treatment, rehabilitation and palliative care [51]. In this dissertation, palliative rehabilitation constitutes the framework for the supportive intervention for the caregivers of PALS/CIs. Palliative care is associated with care of persons with a life-threatening illness focusing on relief of suffering and "total pain" *per se* [52, 53]. Total pain was first described by Cicely Saunders in the early 1960s and recognizes pain as multi-facetted and related to suffering, e.g., comprising physical, mental, material, social and spiritual aspects [54]. While rehabilitation is associated with recovery and full functioning, focusing on physical capability *per se* [52, 53]. Nevertheless, palliative care and rehabilitation have convergences such as both being interdisciplinary fields and having quality of life as an overall goal [52, 55]. Palliative rehabilitation focuses on enabling people to participate as fully as possible in all aspects of everyday life whereas the holistic perspective of palliative care may support caregivers in maintaining dignity, autonomy, and self-care [55-57]. The aim of palliative rehabilitation is to empower people to adapt to their new life situation through constructively coping with losses resulting from deteriorating health while at the same time maintaining dignity [53, 58]. Palliative rehabilitation often focuses on the needs of the affected person's [53, 58-60]. However, caregivers of PALS/CIs must go through a parallel process of having to adapt to a new life situation just like the affected person [61, 62], which is why this PhD project focused on the caregivers' needs for alleviation of grief, mourning and support functioning. Caregivers constantly have to adjust and deal with changing needs as ALS/CIs progresses and the combination of palliative care and rehabilitation of the caregivers is therefore important during their partners' unpredictable and chaotic illness trajectories [52]. Research on palliative rehabilitation interventions for caregivers is sparse [52, 63]. Nevertheless, palliative rehabilitation seem important because ALS and the cognitive and/or behavioral impairments are complex diseases that entail continual losses and challenges for the caregivers that could be accommodated by different intervention initiatives [52]. A recent literature review on rehabilitation and palliative care suggest that the two approaches could be regarded as a continuum of care, as both assuming that a person can benefit from their combined support [52]. Further, the review shows that the combination of palliative care and rehabilitation works within and across different organizational contexts [52].

Supportive initiatives

Different supportive interventions that aim to reduce maladaptive coping strategies as a means to improve the well-being of caregivers have been developed during the last decade [30, 64-67] and may even enhance caregiver coping with ALS [23]. However, a recent study points to the importance of timely, problem-focused coping strategies to caregivers provided by a multidisciplinary ALS-team in order to alleviate their caregiver burden [42]. Caregivers need more supportive interaction and information about their partners' disease and end-of-life phase [21, 35]. Furthermore, information on cognitive and behavioral impairments provided by professionals may help caregivers of PALS understand and deal with symptoms [46]. Active planning within a multidisciplinary care setting provides an avenue for caregivers of PALS and frontotemporal dementia to proactively cope with cognitive/behavioral impairments that will induce improved care and reduce the risk of caregiver burnout [42]. One way of providing such support is to facilitate peer-to-peer contact and a space within which they can share experiences [68]. Peer-support creates solidarity and mutual understanding which lead to a sense of hope and gratitude for life [69]. However, because of the limited time available to caregivers in daily life and the sometimes rapid deterioration associated with ALS, it is necessary to develop caregiver support that is specific to this population and easily integrated and generalizable across ALS care settings [42]. Telemedicine is one way of offering accessible support to caregivers [23, 70]. Online media removes some of the strains of travelling and thereby lowers stress in ALS-caregivers [71]. Online support enhances accessibility and provides a flexible platform that enables caregivers to participate where and whenever they have a moment [65, 72]. For caregivers with a magnitude of responsibilities and limited time, online support is the only option [65, 72].

In Denmark, ALS-care is provided by both public and private healthcare services, nationally or locally based. Multidisciplinary ALS-teams at the hospitals are responsible for the diagnosis, treatment, and continuous ambulant follow-ups of PALS. Palliative care is offered by public institutions located in or outside hospitals. The PALS's daily need for help, support and rehabilitation is mainly placed within local care settings. PALS are offered referral to the National Rehabilitation Center for Neuromuscular Diseases (RCFM), a highly specialized outpatient hospital financed by the Danish government [73]. RCFM's role is to support the public social and health care system with specialized rehabilitation of people with different kinds of neuromuscular diseases, including ALS [74]. RCFM cooperates with the person having a neuromuscular disease, their families, and professionals in public and private care settings and at hospitals. All services provided by RCFM are free of charge and covered by the Danish welfare system [75]. Neurological hospital departments refer about 95% of all PALS to RCFM, whose professionals are organized in multidisciplinary teams, consisting of occupational therapists, physiotherapists, nurses, medical doctors, psychologists, and social workers [76]. This PhD project is carried out in affiliation with RCFM which constitutes the project setting.

Rationale for this study and aim for the PhD

As outlined in the introduction, there was a profound need for accessible support interventions for caregivers dealing with everyday challenges related to PALS/CIs.

- Caregivers often take great part in the care of their relatives without being trained.
- Caregivers lack information on potential consequences of ALS including the expectancies of cognitive and/or behavioral impairments.
- Caregivers ask for support but often end up declining or dropping out of interventions due to heavy caregiving responsibilities, lack of time, and a high caregiver burden.
- Research shows that caregivers are more burdened by the behavioral impairments than by ALS alone and that their psychological distress is worse than that of the PALS.
- Accessible palliative rehabilitation interventions for caregivers of PALS/CIs seem to be missing.

Overall, there is an unmet need for investigating how palliative rehabilitation interventions that support caregivers of PALS/CIs in dealing with everyday challenges and reduce the negative consequences of the disease could best be designed. The introduction section mentions methods and strategies designed to support the caregivers in dealing with these complex challenges, however, interventions are highly dependent on the context in which they are implemented. It was therefore crucial to tailor the support intervention for caregivers of PALS/CIs to the setting of RCFM [77, 78].

Overall aim and research questions

The overarching aim of this PhD project was to develop and evaluate an online targeted palliative rehabilitation blended learning program named EMBRACE to support caregivers of PALS/CIs in dealing with everyday challenges and needs.

Study-specific aims

The four sub-studies in this dissertation will address the following aims:

To explore the reflections of family caregivers and professionals of deceased PALS/CIs on everyday challenges and needs related to the PALS/CIs.

To investigate the acceptability of a new online palliative rehabilitation blended learning program (EMBRACE) for family caregivers of PALS/CIs.

To explore the experienced benefits and challenges of a new palliative rehabilitation blended learning program (EMBRACE).

To understand what goes on in online peer-support in a new palliative rehabilitation program (EMBRACE) among caregivers of PALS/CIs.

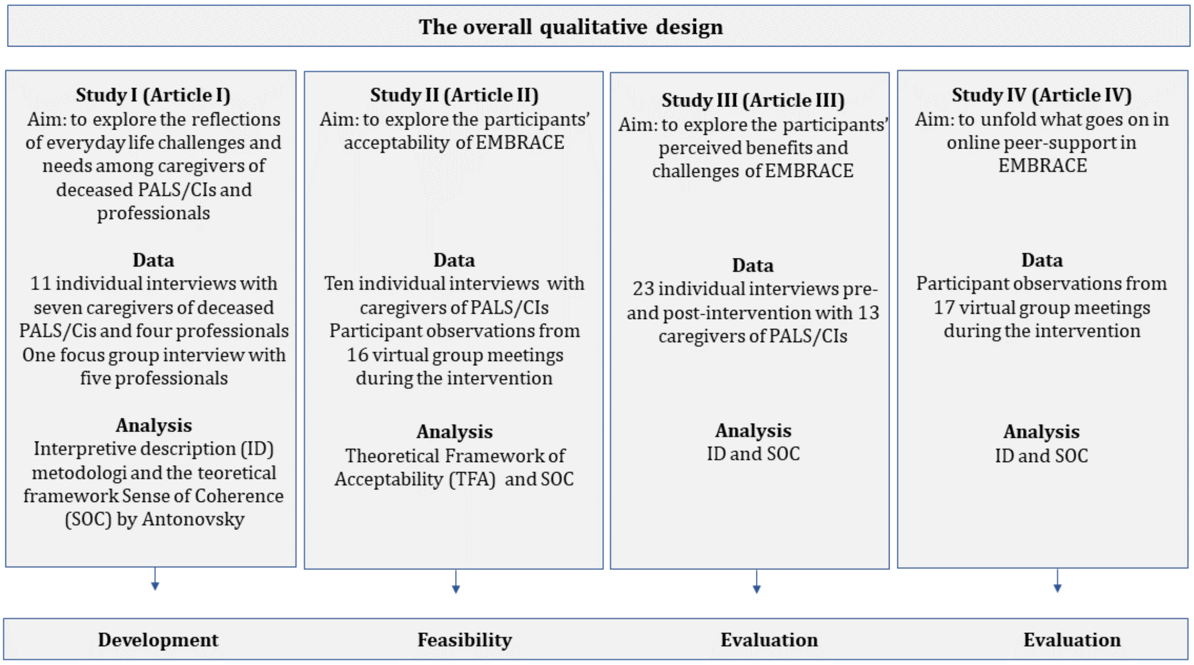
Methods

First, I will briefly introduce the design and elaborate on the overall methodological framework for the dissertation. Second, I will present the methodology and the two theoretical frameworks. Third, a program theory and a logic model used to develop, guide, and evaluate the intervention are illustrated. Fourth, methods employed in the four studies are briefly presented. Fifth, each study is presented by design, sampling/ recruitment, data, and analysis. Finally, the ethical considerations and the researcher's role are discussed.

Design

Due to the nature of the study aims, a qualitative design was chosen for **study I, II, III and IV** (Figure 1).

Figure 1. Presentation of the overall study design, data, and analysis.



The overall qualitative design

Study I (Article I)
Aim: to explore the reflections of everyday life challenges and needs among caregivers of deceased PALS/CIs and professionals
Data
11 individual interviews with seven caregivers of deceased PALS/Cis and four professionals
One focus group interview with five professionals
Analysis
Interpretive description (ID) metodologi and the teoretical framework Sense of Coherence (SOC) by Antonovsky

Study II (Article II)
Aim: to explore the participants' acceptability of EMBRACE
Data
Ten individual interviews with caregivers of PALS/CIs
Participant observations from 16 virtual group meetings during the intervention
Analysis
Theoretical Framework of Acceptability (TFA) and SOC

Study III (Article III)
Aim: to explore the participants' perceived benefits and challenges of EMBRACE
Data
23 individual interviews pre- and post-intervention with 13 caregivers of PALS/CIs
Analysis
ID and SOC

Study IV (Article IV)
Aim: to unfold what goes on in online peer-support in EMBRACE
Data
Participant observations from 17 virtual group meetings during the intervention
Analysis
ID and SOC

The overall methodological framework - The British Medical Research Council's (MRC)

The British Medical Research Council's (MRC) framework on developing and evaluating complex interventions in health guided the methodology of the whole PhD project [77-79].

This PhD project was initially guided by the MRC guidance from 2008 and modified according to the updated 2021 MRC framework.

A complex intervention is defined as interventions containing several interacting components [80]. The components concern the number of difficulties in behavior changes, number of groups or organizations participating, variability of outcomes and degree of flexibility and tailoring [80]. The MRC guidance from 2008 consisted of four non-linear phases including a development, feasibility/piloting, evaluation and implementation phase [80]. Going through each phase, researchers must assess and address main uncertainties such as the study design, the intervention, and/or implementation procedures [79, 81]. In 2021, the MRC published the latest update on the framework (Figure 2) [77, 78]. The phases now concern: 1) developing or adapting an existing intervention or identifying an already existing intervention and exploring its options for evaluation. 2) feasibility and acceptability. 3) evaluation assessing if the appropriate methods to address the research question are being used and 4) implementation. Six core elements have been defined and should be assessed and addressed before moving to the next phase. They are a) *context* (any feature of circumstances in which the intervention is conceived, developed, evaluated and implemented); b) *develop, refine, and (re)test program theory*; c) *engage stakeholders* (patients and members of the public, or those who are targeted by the intervention or policy or involved in developing or delivering the intervention); d) *identify key uncertainties* (that is already known from either the program theory, stakeholder, or the research team); e) *refine intervention* (fine tuning or making changes to the intervention once the prototype has been developed); f) *economic consideration* (determining the comparative resource and outcome consequences of the intervention for the people and organizations affected)[77, 78]. The updated framework is defined as a framework and not a guideline, enabling a more flexible and pragmatic use of different designs to develop and evaluate complex interventions [77, 78]. Further, it emphasizes the need for greater attention to understanding the "how" and under which circumstances an intervention brings changes [82-84]. The MRC now underpins the necessity of using qualitative methods to explore and fully grasp the complexities involved when developing and evaluating complex interventions and of being able to make the needed refinements of the intervention along the phases described within the framework [77, 78].

The MRC framework was used in the project because of its structured approach which is suitable to address several uncertainties regarding the target population, the intervention, and the context [77, 78]. Regarding the caregivers of PALS/CIs there were uncertainties about what their actual challenges and needs were and how an intervention could best fit in and support them in their complex everyday lives. As for the intervention, there were uncertainties about how to modulate and deliver the content needed in the most suitable manner for this highly burdened group of caregivers; how the blended learning format, including the length of the intervention, size and composition of the groups, and the number of videos and other intervention components would be relevant, beneficial, and accepted by the participants?

The approach was pragmatic indicating that the intervention setting was the "real" world [85]. The overall focus was to systematically develop an acceptable and beneficial practice-oriented supportive intervention targeting caregivers of PALS/CIs and to make the intervention sustainable to fit well into the real world setting and the clinical practice at RCFM.

The PhD project was divided into four sub-studies, covering three phases of the MRC framework (Figure 2). Phase one in the MRC framework concerned development and modelling, phase two investigated acceptability, and phase three evaluated the benefits and challenges of the intervention. Furthermore, the third phase also entailed an investigation of what goes on in online peer-support among caregivers of PALS/CIs in EMBRACE. Figure 3 portrays the chronology of the four sub-studies.

Figure 2: The MRC framework from 2021 in relation to each sub-study [77]. Derived from Skivington, Matthews et al. 2021, an open access article distributed under the terms and conditions of the Creative Commons Attribution (CC BY) license Creative Commons - Attribution 4.0 International - CC BY 4.0. This figure is also reproduced with kind permission from Dr. Kathryn Skivington.

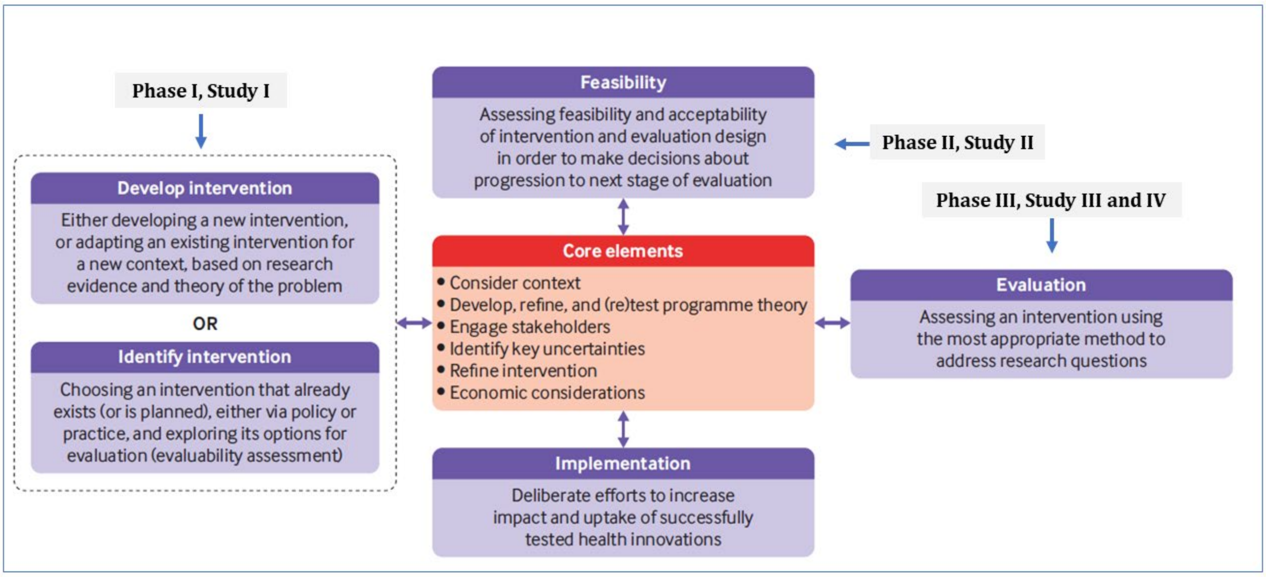


Figure 3. Timeline illustrating the chronology of the four articles and their work process.

<table>
<tr><th></th><th>2019</th><th colspan="2">2020</th><th colspan="2">2021</th><th colspan="3">2022</th></tr>
<tr><td>Article I</td><td>Ongoing analysis</td><td colspan="2">Empirical</td><td colspan="2"></td><td colspan="3"></td></tr>
<tr><td>Article II</td><td></td><td>Ongoing analysis</td><td colspan="2">Empirical</td><td colspan="4"></td></tr>
<tr><td>Article III</td><td></td><td colspan="3">Ongoing analysis</td><td colspan="3">Empirical</td><td></td></tr>
<tr><td>Article IV</td><td></td><td colspan="5">Ongoing analysis</td><td colspan="2">Empirical</td></tr>
</table>

The 2021 MRC framework systematically addresses new core elements, that I have not directly addressed in the dissetation. Nevertheless, all the core elements have been assessed and addressed indirectly throughout the development and evaluation process of the intervention. To estimate the economic aspects of the implementation of EMBRACE in the clinic at RCFM the following cost were included: hours spend on preparation, recruitment, and execution of EMBRACE by a group facilitator, secretarial help with the aforementioned, and annual payment to the online hosting platform Simplero.

To be able to modulate and refine the intervention through all phases of the MRC framework, the intervention setting, and design and the included components were continuously and systematically assessed and addressed by the research team [77, 78]. In line with the 2021 framework awareness of the importance of context was considered, e.g., when the COVID-19 pandemic forced me to convert the intervention into an entirely virtual setting.

Methodology – Interpretive Description (ID)

In consistence with the explorative aims of the studies in this dissertation, ID informed the methodology in **study I, III and IV** [85]. ID is a qualitative inductive analytical methodology that aims to develop new inquires that are translated into practice for the benefits of informing and advancing practice [85]. ID seeks to unravel and gain a deeper understanding of human phenomena by taking a point of departure in problems and challenges raised in the real world practice [85]. The research question must therefore always be practice-oriented [85]. ID seeks to construct a coherent description and in-depth interpretation of the relations and patterns within the phenomenon studied by moving beyond the self-evident and not controlled by the evidence-based knowledge [86]. Although ID draws on an interpretive explanation of data like ethnography, grounded theory and phenomenology, it was developed because of a need to go beyond the descriptive level to understand the "so what" of the research phenomenon and to avoid the problems related to classic methodologies [85]. The classic methodologies seek to develop theories or understandings about a phenomenon but not necessarily always to identify applicable and useful findings to be used in practice [85]. In contrast, ID has an applied health research focus throughout the research process [85]. ID is a pragmatic methodology that allows the researcher to use and combine various methodologies and/or theories necessary to help fully capture the phenomenon under study [85]. This pragmatic and eclectic approach is useful when exploring new fields because the flexibility allows for unexpected findings and for making the necessary adjustments before moving forward [85, 87]. However, ID strives to maintain sufficient rigor to ensure credibility by providing an organizing logic during the different study phases [85, 87, 88]. The methodology draws on social constructivism as well as critical realism [86]. The epistemology of ID entails that human experiences are constructed and impacted by social and contextual factors, which create multiple, subjectively constructed realities to be explored inductively [85]. A way of exploring such realities is by asking people about their experiences and entering the field to observe them in order to discover the impact of contextual and social factors on the matter being studied [85]. ID's epistemology is different than the hermeneutic one which focuses on the experiential context in which the individual's actions evolve and become meaningful, and the inquiry lies within a dialectic known as the hermeneutic circle [89]. In ID, everything is explored through an interpretive lens that is influenced by the epistemology of the methodology applied (or theory if such one is applied), which may be used to structure and guide the study and analyze data, for instance, to qualify and develop an interview guide [85].

ID was chosen as the methodology because of its applicability to inform and advance the support of caregivers of PALS/CIs in the RCFM practice through this PhD project [85]. ID seemed particularly useful for this purpose because of its practice-oriented approach, the research's origin in practice, and the inductive and pragmatic approach, that allowed for methodological freedom to design the project that was most suitable for the research purposes [85]. ID allowed for a conceptual and epistemological framework and strategy which was beneficial in guiding the methodological path [85]. Finally, ID is compatible with the epistemology of the frameworks (the MRC and Sense of Coherence) used in this dissertation, with regard to the individual's interaction with its environment which is constantly changing and it prescribes that research focus should be on the individual in its context [77, 85, 90].

Theoretical framework – Sense of Coherence (SOC)

To accompany ID, SOC was employed as both a methodological framework to inform the development of the intervention and as a theoretical framework to help understand the participants' health and actions as they go through the stressful and chaotic phase of life as caregivers of a PALS/CIs [90, 91]. According to the founder of SOC, Aaron Antonovsky, SOC is not a theory that aims at keeping people well, it is the answer to the Salutogenesis question [92], i.e. what are the origins of health [91]?

> *"Rather, in than it derives from studying the strengths and weaknesses of promotive, preventive, curative and rehabilitative ideas and practices, it is a theory of the health of that complex system, the human being" (Antonovsky, 1996, p.13) [92].*

SOC refers to a life-orientation that is structured, manageable and meaningful, or coherent [93]. SOC rests on three core elements: comprehensibility, manageability and meaningfulness [92]. The elements reflect the individual's way of thinking, being, and acting [90], thereby representing the individual's view of life as well as their ability to maintain health in spite of stressful situations [90, 93]. *Comprehensibility* is the cognitive component that refers to the extent to which the individual perceives the stimuli that they are confronted with from both internal and/or external environments as ordered, consistent, structured and clear, and that the information make sense cognitively [90]. *Manageability* is the instrumental/behavioral component and refers to the extent to which the individual perceives that resources that are at their disposal are adequate to meet the demands posed by the stimuli they are faced with [90]. The final element, *meaningfulness* is the motivational component that refers to the extent to which an individual feels that life makes sense emotionally, and that problems and demands are worth investing energy in and that they are regarded as challenges more than burdens [90].

Antonovsky assumes that chaos and change is a normal state of life but that humans cope with these individually and therefore experience different impact on their health [90, 91]. Humans interact with the constantly changing surrounding environment and is affected by it [90]. Antonovsky sees a close connection between mental health and SOC [93] and describes health as a continuum between ease and dis-ease [94]. A high SOC is related to a better perceived health (ease) while a lower SOC is related to a larger consumption of medications (dis-ease) [95]. A high SOC could therefore be considered a resilience factor because it reduces stress and decrease internal and external problems [95].

SOC was suitable for this PhD project because caregivers of PALS/CIs face chaos and stress during everyday life, and their health is naturally impacted by the fatal and devastating situation of their partner with ALS/CIs. As a methodology SOC informed the development and tailoring of the EMBRACE intervention as means to enhance the participants' comprehensibility, manageability, and meaningfulness, and thereby their sense of coherence. As a theoretical framework SOC helped to understand why and how some participants in EMBRACE cope well with life challenges while others do not which is important as means to target health promoting initiatives better in the future [90, 91, 95, 96]. The SOC elements helped understand the components in EMBRACE and how these either promoted or restrained the participants' overall sense of coherence. Further, SOC indirectly guided the design, interview and participant observation guide, analysis and writing of findings for all four studies (Figure 1).

Theoretical Framework of Acceptability (TFA)

The TFA was used as a theoretical framework to help investigate the participants' acceptability of the intervention. Acceptability of an intervention is key to successful implementation [97, 98] and necessary but not enough for the effectiveness of an intervention [99]. Content, context, and quality of care may impact participants' acceptability [99], and research shows how participants who accept an intervention are more likely to adhere to treatment recommendations and thereby benefit from clinical outcomes [100, 101]. In line with the MRC framework, predefined progression criteria related to the participants' acceptability of the intervention were used to investigate their acceptance of EMBRACE [77, 78]. The aim of assessing acceptability was to clarify whether an evaluation of EMBRACE was feasible [77, 78]. Lack of and failure to provide clear definitions of acceptability when evaluating and implementing complex interventions contributed to the development of the TFA [99]. The TFA consists of seven concept-driven constructs: affective attitude, burden, ethicality, intervention coherence, opportunity costs, perceived effectiveness, and self-efficacy that can support investigation of acceptability prospectively, concurrently and retrospectively (Figure 4) [99]. The TFA is a multi-faceted framework representing the extent to which people delivering and receiving the healthcare intervention find it appropriate based on anticipated or experiential cognitive and emotional reactions to the intervention [99]. The TFA was chosen to strengthen the investigation of the participants' acceptability of EMBRACE through the nuanced deductive constructs of acceptability and was applied in **study II** (Figure 1).

Figure 4. The Theoretical Framework of Acceptability with its seven constructs [99]. The framework illustrates the periods of data generation and methods used. Inspired by Sekhon, Cartwright et al. 2017, an open access article distributed under the terms and conditions of the Creative Commons Attribution (CC BY) license Creative Commons - Attribution 4.0 International - CC BY 4.0

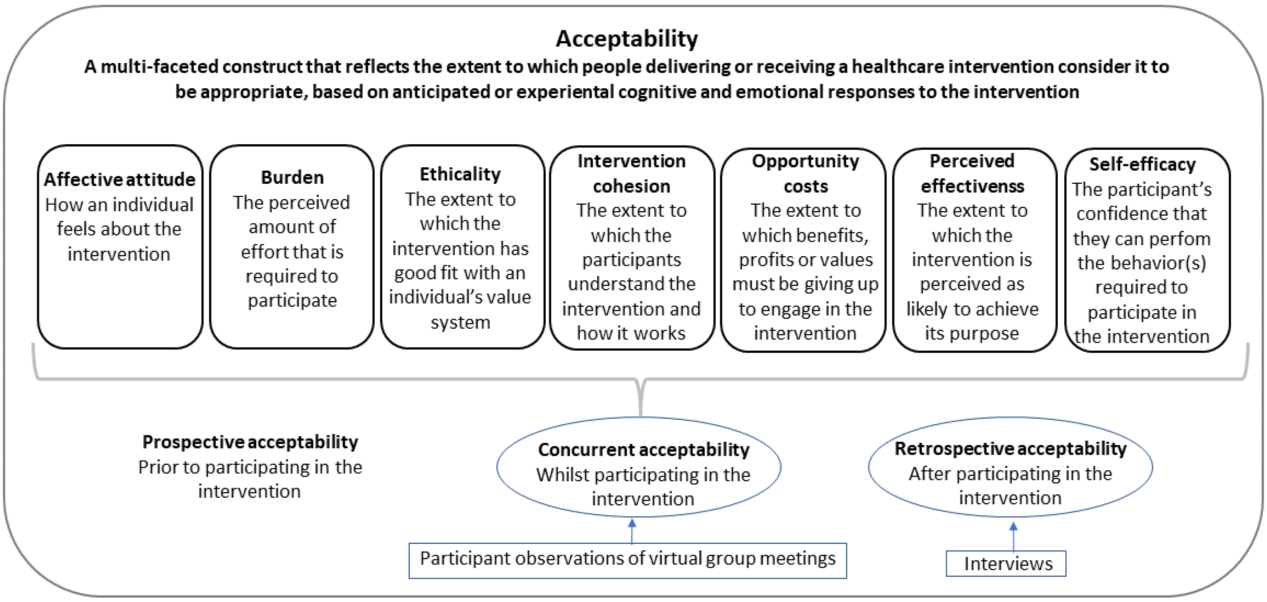
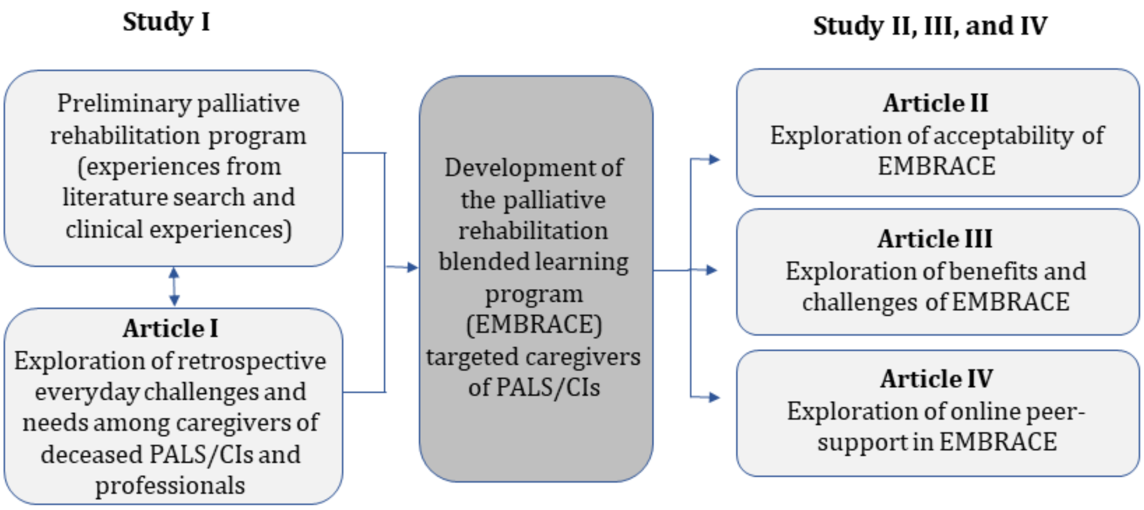


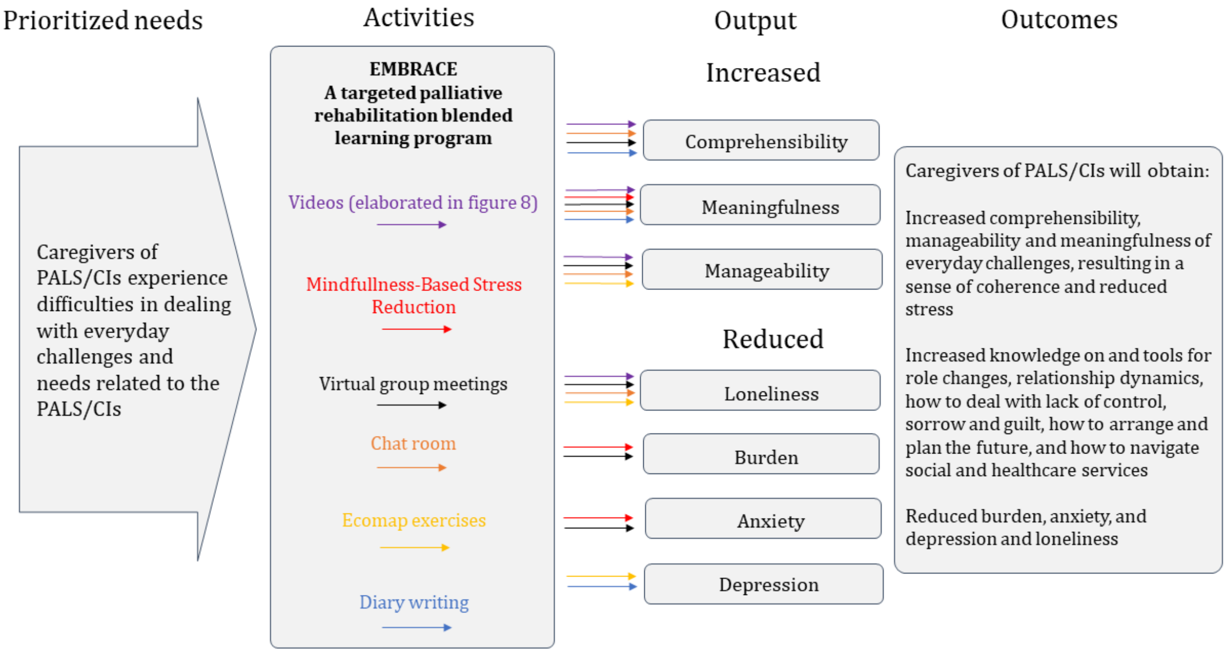
Figure 5. An overview of the PhD process and its sub-studies.



Program theory

As recommended by the MRC framework, a program theory was developed initially to illustrate the prioritized needs and expected outcomes related to specific intervention components as well as the interaction between these (Figure 6) [77, 78]. The focus of the project was overlapping, looking on both the inputs, processes, and outcomes, and illustrated by an effectiveness, system and theory-based approach which is often used in conjunction within complex interventions [77, 102]. Using a visual representation, the program theory articulates the multiple ways it may generate outcomes and the complexity that premises the EMBRACE intervention. Through the PhD project and the first three phases of the MRC framework, the program theory was iteratively adjusted according to learnings and new guidance was developed for the next steps (see figure 13 p. 52).

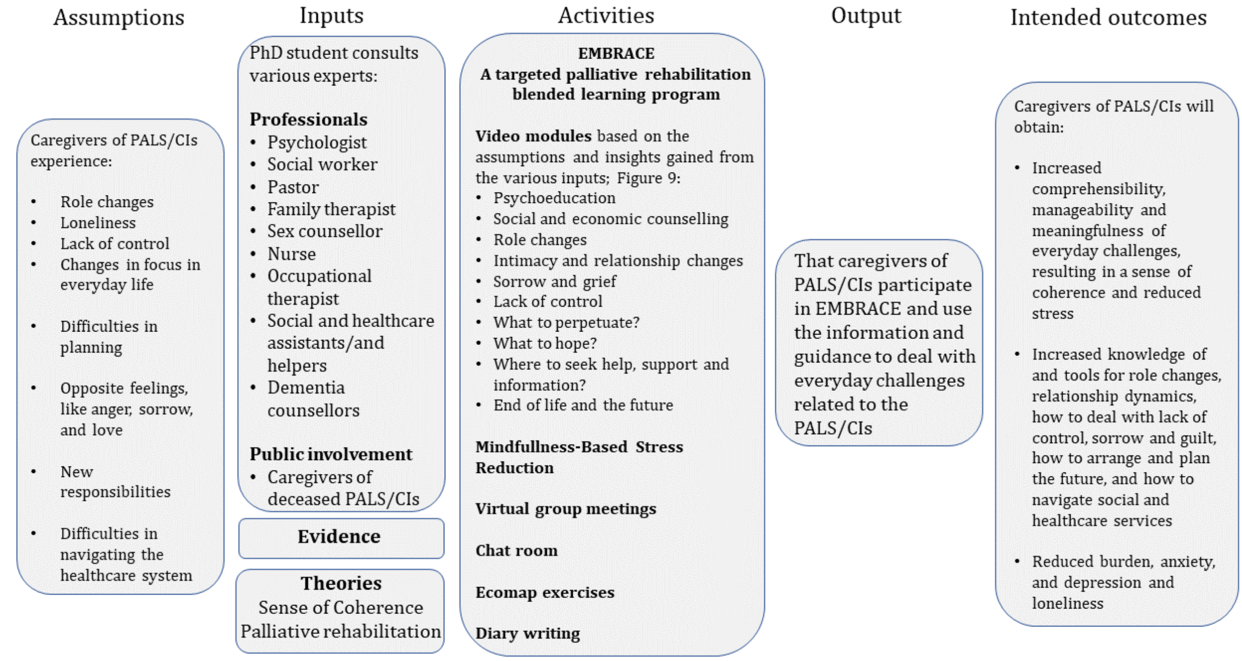
Figure 6 Initial program theory of the complex palliative rehabilitation blended learning intervention named EMBRACE. Version 1.0.



Logic model

A logic model was developed during the initial phase of the development process of EMBRACE to illustrate the assumptions, inputs, activities, outputs and intended outcome of the intervention (Figure 7) [78]. The complex processes of EMBRACE is presented as a linear process in the logic model, however, this should not be misinterpreted as 'simple' [103].

Figure 7. Logic model of the EMBRACE intervention.



Project setting

The PhD project was affiliated with RCFM in Denmark (described on p. 14). EMBRACE was delivered online using Microsoft Teams and Zoom and the online hosting platform Simplero.

Methods

Individual interviews

Interviews were employed as a method to gain access into the participants' lifeworld, their experiences, and thoughts [104]. One-on-one interviews were preferred to facilitate an intimate environment where the participants would feel safe to share and elaborate on their deepest thoughts and reflections on everyday challenges and needs (**study I**), acceptance of EMBRACE (**study II**) and the benefits and challenges related to EMBRACE (**study III**) (Figure 1). One-on-one interviews were also chosen for ethical reasons with sensitive topics being touched upon in the interviews and to ensure that focus was on one specific person and their experiences. Because of the sensitive topics and the potential emotional impact on the participants, a debriefing was made after the interviews to ensure that they were in a balanced state when finishing the interview [105]. I collected all the interviews as part of the development process **(study I)** and pre- and post-intervention **(study II and III)**. The post-intervention interviews allowed for more profound questions, deriving from the stories told during virtual group meetings and from participant observations of the virtual group meetings that needed to be further explored and elaborated in the post-intervention interviews.

All interviews (individual and focus group, see below) were performed as semi-structured to ensure a certain focus on the specific research questions for **study I, II, and III** [104]. Interview guides were developed and consisted of open-ended questions related to the research questions (appendix 1-4). All interviews were audio recorded to ensure authenticity of the participants' terminology and expressions and for subsequent transcription in verbatim to support data analysis. Interviews were performed either in-person at RCFM, at the caregivers' home, or at their workplace, or by phone, or via Skype/Microsoft teams.

Focus group interviews

Interviews with professionals in **study I** were conducted as focus group interviews to facilitate group discussions, elaborations and foster a recall of experiences around everyday challenges and needs related to supporting PALS/CIs and their caregivers (Figure 1). Purposive sampling was applied to ensure maximum variation e.g., different job functions, professional backgrounds, and experiences with PALS/CIs. Attention during the interview was on the potential power-relation or hierarchy between the participants and how this might impact on the group dynamics, e.g., visitation consultants vs healthcare assistants/helpers, nurses vs healthcare assistants/helpers, and professionals with profound experience vs limited experience. Furthermore, attention was on the familiarization between some of the participants but not among all of them. This awareness meant that I, as the interviewer, could help facilitate the dialogue when needed, when, for instance, the same person talked all the time. Heidi With (HW), ALS-consultant, nurse, and family therapist from the research group, attended the focus group interview as an assistant moderator.

Participant observations

Participant observations were chosen as a method in **study II and IV** with the aim to generate data on and understandings of the participants' acceptability of EMBRACE **(study II)** and what goes on between the participants in the online peer-support format of EMBRACE **(study IV)** (Figure 1). Participant observations enabled focusing on the social practice, interactions, verbal and nonverbal communication as well as the relations between the participants in the virtual group meetings [106]. To grasp the group discussions, the participants' perspectives and behaviors and to reduce interference with group discussions, the observer role was moderate, i.e., my presence and role during the virtual group meetings were known by the participants and the group facilitator (HW), but I did not actively participate or only occasionally interacted with the group during meetings [107]. My researcher role differed from that of the participants and group facilitator as I aimed to become explicitly aware of the behaviors and interactions of the participants observed and to record these using field notes [106].

I interacted with the participants on the first meeting where I welcomed and introduced them to the intervention. Participants were reminded that the meetings would be recorded and that I would observe every meeting with my camera turned off. After replying to potential questions from the participants, I then turned my camera off, "leaving" the participants with the group facilitator. At the end of the final meeting, the group facilitator invited me "back" into the meeting, where I turned my camera on and thanked them for their willingness to participate and informed them about the future process of the PhD project including the articles that would be based on the video-recordings.

Participant observation guides were developed and applied to strengthen and focus the participant observations to the area of research (appendix 5-6). The participant observations consisted of observations and field notes from the virtual group meetings which enabled explorations of interactions, behaviors, and perspectives without indirectly interrupting the participants. Field notes were collected using a wide-focus and descriptive approach to the observations [107, 108]. After a virtual group meeting had ended, the group facilitator and I individually spend 15 minutes on a non-stop writing exercise. The following day we met and discussed our notes from the exercise to capture and understand the participants' interactions, perspectives, and reflections, but also to ensure that the intervention was proceeding and delivered as planned. If not, small adjustments were made, e.g., a reminder to stop sharing slides while the participants were making their take-away message. I collected the participant observations during the two rounds of the EMBRACE intervention, and the main supervisor (Charlotte Handberg) retrospectively watched all the recording to enhance reliability of the coding.

Questionnaires

In **study III**, two questionnaires were used as background characteristics on the participants' self-reported pre- and post- intervention levels of burden, anxiety, and depression (appendix 7 and 8). The questionnaires were the Zarit Burden Interview (ZBI) and the Hospital, Anxiety and Depression Scale, commonly applied in ALS-research. The questionnaire versions in Danish were used as the participants were native Danish speakers. The ZBI has undergone a linguistic validation methodology [109], and the HADS is validated in Danish [110]. The ZBI measures the experienced burden in caregivers [24, 111, 112]. Questions are scored from 0–4 points, and the total score ranges from 0 to 88 points, higher scores reflect greater burden [113]. A score ≥24 indicates a clinically high burden and helps to identify caregivers of PALS who are at risk of caregiver burden [37]. HADS measures the levels of anxiety and depression and consists of 14 items, divided into two 7-item subscales of anxiety and depression [114]. Scores are divided into three sub-groups, 0–7, 8–10, and 11–21, with a cut-off ≥8 indicating possible cases of anxiety or depression [114]. The questionnaires were attached in emails to all the enrolled participants and again forwarded by email right after finishing the intervention. One reminder to fill out and return the questionnaires were forwarded to the relevant participants who had not replied within a period of two weeks. Participants' characteristics were separated into a drop-out group and an intervention group with characteristics being represented by median sum score and range of burden, anxiety, and depression pre-intervention for the drop-out group and pre- and post-intervention for the intervention group.

Studies

The four studies are described briefly below with references to the previous sections on methodology, theoretical frameworks, and methods.

Study I – article I

Design

A qualitative design employing individual and focus group interviews were applied (p. 26). The study was guided by the MRC framework, ID and SOC (p. 16-20)(Figure 1) [77, 78, 85, 90].

Samling and recruitment

Caregivers of deceased PALS/CIs and professionals were included (described separately below).

Caregivers

Participants were eligible if they had either lived with the deceased or had been a close relative to the deceased within the last six month, and they were not eligible if they had previously been cooperating with me. Participants were asked to describe their partner or parent's cognitive and/or behavioral impairments and were included based on their perceptions.

Caregivers were recruited through Facebook groups for caregivers of PALS and on RCFM's website and Facebook page. Thirteen caregivers wished to participate and seven were included (Table 1). Reasons for exclusion were PALS/CIs were still alive or the caregiver did not perceive their partner or parent as cognitively and/or behaviorally impaired.

Table 1. Demographic characteristics of the caregivers that participated in study I.

Participants – caregivers		**(n = 7)**
Gender	Male	1
	Female	6
Age	<22	1
	23–45	1
	47–55	2
	56–65	2
	>66	1
Relation	Married/spouse/partner	5
	Adult child	2
Occupational status	working	4
	Retired	3
Years of ALS-trajectory	<1	0
	2–3	6
	4–5	1

Professionals

The professionals were eligible if they had formal experience of caring and supporting PALS/CIs and were either a healthcare professional or trained and employed in private healthcare services. The term 'professionals' will be used for both the healthcare professionals and trained employees. Professionals, known by HW or me, were excluded. Key leaders from different healthcare departments in two municipalities were identified through the municipality administration, including a leader from a private healthcare service. The leaders selected the participants who fulfilled the inclusion criteria (appendix 12). Nine professionals were included (Table 2).

Table 2. Demographic characteristics of the professionals that participated in study I.

Participants (professionals)		(n = 9)
Gender	Male	1
	Female	8
Residence	Urban (>40,000 Citizens)	5
	Rural (<40,000 Citizens)	4
Years of ALS experience	1	3
	2–3	4
	4–5	1
	>6	1
Professional background	Healthcare professional*	8
	Unskilled	1
Healthcare profession*:(n=8)	Nurses	3
	Occupational therapists	2
	Social assistants/helpers	3

Data

Caregivers

Seven caregivers were interviewed (p. 26); five caregivers in one-on-one interviews, and two caregivers who were family related were interviewed together. The caregivers were introduced to a draft of the EMBRACE intervention and encouraged to comment and suggest alternative components to include in the intervention (appendix 13).

Professionals

One focus group interview was conducted in-person with five professionals (p.26). Focus group interviews had been planned for the other professionals but were converted into individual interviews due to the COVID-19 pandemic. Instead, four individual interviews were conducted (p. 26).

Analysis of qualitative data - Study I, III and IV

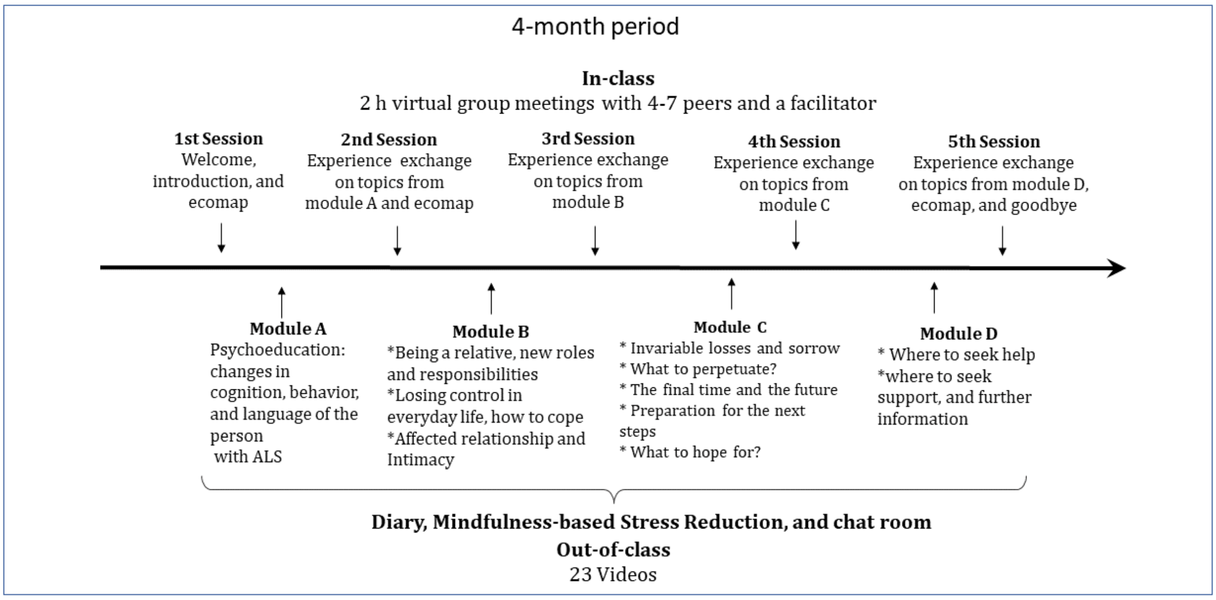
The analytic process was the same for **study I, III and IV** and followed four iterative steps suggested by ID and indirectly related to the SOC elements [85, 86, 90]. First, interviews were transcribed in full length and uploaded into the qualitative analysis program NVivo^{TM}12. For **study IV** video-recordings of virtual group meetings and field notes from participant observations were uploaded in NVivo^{TM}12. Secondly, all transcripts were intensively read and for **study IV**, Charlotte Handberg and I watched the video-recordings while taking additional field notes. Individually, Charlotte Handberg and I read and did an initial coding in a broad manner of the transcripts and notes for insights related to the phenomenon studied in **study I, III and IV** and indirectly to the SOC elements. Thirdly, results of the initial coding were considered, and patterns and relationships among the data groupings were explored and discussed by Charlotte Handberg and I. The research team behind each of the studies then critically examined patterns and relationships within the data. This process generated tentative themes which led to the primary categorization. Working iterative, the categorization of data and exploring how they were related to the SOC elements – comprehensibility, manageability and meaningfulness – led to the final condensation that identified the overarching themes [85, 90]. Fourthly, an interpretive thematic and conceptual description of the relationship among the themes developed by the research team led to an illustrative depiction capturing the main understandings of the phenomenon studied in **study I, III and IV** [85].

Development phase – the EMBRACE intervention

To target the design and content of EMBRACE the plan was to include caregivers of PALS/CIs as partners in the initial process of developing the EMBRACE intervention [115]. A panel of caregivers of PALS affiliated with RCFM was invited by email to comment on the project protocol and a draft of evidence-based components to include/exclude in the intervention. However, no caregivers replied to the two requests. The development of EMBRACE occurred after study I and before study II. The principles behind the development were creative, iterative, and dynamic processes that were flexible and adaptive to changes and strived towards future evaluation and implementation [115]. The process was also systematic and inspired by evidence, theoretical knowledge, clinical experience and empirical data (findings from **study I**), indicating that the intervention was both inductively and deductively developed [116]. An alternative strategy of user-involvement was applied to gain the caregivers' perspectives and comments on a prototype of EMBRACE which was to present a graphic illustration of the design, components, and timeframe at the end of the interviews with the caregivers of deceased PALS/CIs from **study I** (appendix 13). The caregivers thereby acted as consultants rather than co-producers, indicating that the decisions were made by the research team [115].

A Word document was used to log reflections, arguments for decisions and feedback from the supervisors and experts as a way of keeping track and systematizing the development process. A large board was also applied to draw, write and brainstorm on as part of the development. In the final phase of the development, all the knowledge and inputs from **study I**, insights from supervisors and experts from RCFM, evidence from research and theories were gathered and organized into a coherent and beneficial set of components/activities. Next, all perspectives, ideas, and evidence of each intervention component was modeled leading to the final design of EMBRACE (Figure 8). However, despite critical reflection and profound knowledge on which components to include in EMBRACE, it was not until a feasibility evaluation had been undertaken, that I could be sure of whether the right selections were made during the design and modelling phase [79]. According to the MRC framework, there is no criteria for when to proceed to the next phase of the development and evaluation process, however, distributing reports or articles on the initial development process is recommended as a way of knowledge sharing [115].

Figure 8. The EMBRACE intervention. A 4-month palliative rehabilitation blended learning program to support family caregivers of persons with amyotrophic lateral sclerosis and cognitive and/or behavioral impairments. Inspired by Olesen, la Cour et al. 2022 [117].



The EMBRACE intervention was developed and modelled by me in close collaboration with the supervisors and healthcare professionals from RCFM. An experienced ALS-counsellor from RCFM (HW), who is a nurse and certified family therapist facilitated the virtual group meetings. The group facilitator has in-depth knowledge about everyday life with ALS from over 15 years of working with families affected by ALS. From clinical practice and participation in previous research projects, the group facilitator has also gained profound insights into the caregiver's challenges and needs and is experienced in facilitating groups of PALS and their caregivers. The group facilitator participated in most of the videos recorded and provided to the participants.

To assist with building a webpage on an online hosting platform named Simplero, an IT-technician at RCFM helped construct the layout and pictures, and group facilitator and I provided the video materials, texts, and the caption for each video. The research team tested the webpage to make sure that functions such as the chat room, evaluation modules, comment boxes and video modules worked properly and that navigating the webpage was easy and intuitive for the participants. Healthcare professionals at RCFM helped pilot test the webpage and a few adjustments were made.

The EMBRACE intervention, components, and delivery

To prevent EMBRACE from being a ‘black box’, an intervention manual was developed together with the research team to make it possible for other clinicians to replicate the intervention [118, 119]. The intervention comprises specific and non-specific approaches with the specific components being the active ingredients that are included in the intervention, like mindfulness or ecomap exercises [120]. The non-specific components are those used to deliver or facilitate the components in the intervention, like the hosting platform [120]. In EMBRACE, the intervention comprised a supportive program with various components like videos on topics identified in **study I**, theory, and evidence, Mindfulness-Based Stress Reduction, virtual group meetings, chat room, ecomap exercises (which is a diagram of social relations), and diary writing. Components included are described below and divided into ‘in-class components’ and ‘out-of-class components’ due to the blended learning format [121]. EMBRACE was executed twice, in the fall 2020 and again in the fall 2021. To further reduce the burden on the participants, each component in EMBRACE was voluntary, meaning that participation in group meetings, etc. did not require that the participants had watched the videos beforehand (or later).

Online support

An online format was chosen for several reasons. Firstly, online support is known to enhance accessibility and is acceptable among caregivers of PALS [71, 122]. Secondly, online support reduces the burden of travelling and logistics and the caregiver’s level of stress [71]. Thirdly, in online interventions participants are not physically present [123], which was important during the COVID-19 pandemic and to avoid the risks of spreading the virus [124]. Because of the COVID-19 pandemic, EMBRACE was delivered solely as online with virtual group meetings instead of physical meetings as originally planned.

Blended learning

The intervention was delivered through a blended learning design containing both in- and out-of-class components [121]. In-class components consisted of virtual group-facilitated meetings and ecomap exercises (Figure 8). Out-of-class components entailed targeted videos on topics related to the caregivers’ challenges and needs, mindfulness-based stress reduction, and diary writing.

In-class-components

Group meetings

Five virtual group meetings were held on the same weekday every 4 weeks during the four-month intervention period. The meetings lasted from 2.30 PM to 4.30 PM and were facilitated by HW. Every meeting followed the same structure: first a round of catching up with what had happened since the last meeting for each participant, followed by an open discussion on the topic of the day and a take-away message from each of the participants (appendix 14). The topics of the day were presented on a power point slide and was based on topics from the released videos on the website. The group facilitator received ongoing supervision from an external supervisor and debriefed with me after each meeting as a way of coping and preparing mentally for the discussions and interactions during the group meetings. The meetings were run on Microsoft Teams in 2020 and changed to Zoom meetings in 2021 due to technical issues, e.g., not being able to see everyone on the screen at the same time or screens freezing.

Ecomap

Individual ecomap exercises were carried out three times during the intervention to help the participants explore whether their social and professional relations were either supportive or burdensome in their daily lives. The participants were encouraged to draw their network in the first, second and fifth meeting. At the first meeting they drew their current network, at the second meeting their dream scenario, i.e., an image of how they wished their network should be like, and on the fifth their present network again. The participants started the exercise during the meetings and were encouraged to finish them before the next meeting. If they had finished the exercise during the meeting, they would then share their ecomap with the group members, and if not, they would follow up on these at the following meeting (appendix 15).

Out-of-class-components

Website

The online hosting platform Simplero was used to deliver the videos, mindfulness-based stress reduction exercises and the chat room. Participants were assigned to their specific peer-support group. After each of the first four-group meetings, a module containing between 3-6 videos was released, which the participants were encouraged to watch in preparation for the following meeting, forming the meeting agenda. The participants were asked to mark the videos that they had watched as '*completed'* and next evaluate them using a scale of 0-9, indicating their desire to recommend the video to people who were in a similar situation. They could also write comments to the research team on each video in a free-text box (appendix 16-19).

Chat room

The participants had access to a private chat room on Simplero (appendix 20). The chat room made it possible for them to post, read and comment on statements and questions written by fellow group-members. They could address open comments or questions to the group facilitator by writing her name in the chat message. Private messages were not possible.

Mindfulness-based stress reduction (MBSR)

Seven targeted MBSR videos were developed by a psychologist and mindfulness therapist for the participants. The videos were available on Simplero throughout the intervention and three months post-intervention. The videos contained an introduction to MBSR, breathing exercises, a body scan and two yoga videos. The participants were also encouraged to mark and evaluate the videos after watching them (appendix 21).

Diary writing

The participants received a physical notebook in their letterbox before the first group meeting. It contained an introduction with inspiration on how to use it (appendix 22) and an overview of the intervention program with exact dates for the group meetings (appendix 23). Four A-3 papers for the ecomap exercise were included with an example of how to draw different types of lines to help illustrate their relations (appendix 15). Throughout the intervention the group facilitator asked the participants whether they used the notebook, and if they said 'yes', she asked if they wanted to share anything from their book.

Study II – article II

Design

After the development of EMBRACE, the next phase in the MRC framework was the feasibility and acceptability phase (Figure 2) [77]. A feasibility study examines if an intervention is appropriate for further testing, and assessing acceptability is, thus, an important element of the study [77, 78, 99, 125]. A qualitative cross-sectional design was employed to assess acceptability of EMBRACE. The study was guided by the MRC framework, SOC, and the TFA (p. 16, 20-21) (Figure 1)[77, 78, 90, 99].

Samling and recruitment

Participants who participated in EMBRACE in 2020 were eligible. Twelve participants were included (Table 3). For EMBRACE the inclusion criteria were (a) partners or spouses who live with a relative with ALS referred to RCFM and who has received an initial visit from a professional at RCFM, (b) caregivers who understand and speak Danish, and (c) caregivers of persons with ALS with a cut-off score ≥22 on the Amyotrophic Lateral Sclerosis-Frontotemporal Dementia-Questionnaire (ALS-FTD-Q), a validated questionnaire containing 25 items, with a total score ranging from 0-100, higher scores indicating more behavioral changes (appendix 24)[126]. Recruitment took place at RCFM where healthcare professionals sampled the participants by identifying 208 persons with ALS referred to RCFM up to September 8, 2020. Invitations to participate were sent by letter to each of these caregivers (appendix 25). They were encouraged to contact HW or me if they were interested. They were then asked to score the behavioral impairments of their affected partner using the ALS-FTD-Q [126]. Thirty-one caregivers wished to participate and fifteen fulfilled the inclusion criteria and were enrolled, two of whom never got to participate in the intervention due to the death of their affected partner and parent. For further descriptions on sampling and recruitment see article II [117]. The welcoming letter for EMBRACE can be seen in (appendix 26).

Table 3. Demographic characteristics of the participants in study II.

Participants		**(n=12)**
Gender	Male	3
	Female	9
Age	18-25	1
	39-50	3
	51-55	4
	56-67	4
Relation	Married/partner	11
	Adult child of a PALS/CIs [1]	1
Occupational status	Working	7
	Early retirement/retired	4
	Studying	1
Years of ALS-trajectory	0-2	4
	2-4	2
	4-8	4
	8-12	1
	12-14	1
ALS-FTD-Q score	22-30	2
	31-35	5
	36-40	1
	41-46	3
	47-55	1
Urban	≥ 80.000	1
Rural	≤ 80.000	11

Data

Data was generated using triangulation with participant observations (concurrently - during the intervention) and interviews (retrospectively-post-intervention) to assess the participants' acceptance of EMBRACE (Figure 4) (p. 25-27) [99]. Interview questions focused on the experiences, attitude, feelings, and preferences of the components in EMBRACE. The participant observation guide extended to the TFA constructs (appendix

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[1] Person with amyotrophic lateral sclerosis and cognitive and/or behavioral impairments

5) [99, 117]. Participant observations of video-recordings of 16 virtual group meetings were carried out. The interview guide is provided in appendix 3.

Analysis

All data were organized and analyzed deductively according to the TFA constructs in NVivo12™ [99]. The deductive approach was applied to search specifically for aspects of acceptability. Four of the five authors (except the second author) carried out the participant observations. As part of piloting the participant observations guide, the team individually watched the same two recordings, while collecting field notes. Next, we meet to discuss whether the guide was understandable and applicable and to clarify potential uncertainties, e.g., which TFA construct was the most suitable for a certain action/behavior or saying from the recordings. The four authors then individually, systematically, and deductively collected participant observations on every recording using the participant observations guide. Next, we systematically went through each meeting discussing what was being said and whether observations were organized correctly according to the constructs to ensure reliability (appendix 5). The field notes from each of the four authors were then condensed by me and presented in an overview within each of the TFA constructs. The overview was then discussed with all the authors of the article.

Interview data were also deductively coded according to the TFA constructs by Charlotte Handberg and me and subsequently discussed with all authors. Further descriptions on the interview analysis are provided in article II [117].

Study III – article III

Design

A qualitative interpretive study design using interviews pre- and post-intervention was applied. The ZBI and HADS were employed to obtain population characteristics on burden, anxiety, and depression pre- and post-intervention (p. 28).

Samling and recruitment

Participants included in EMBRACE were eligible (p. 36). The ZBI and HADS were employed to obtain population characteristics on burden, anxiety, and depression pre- and post-intervention (Figure 9). Thirty-one interested caregivers opted to participate. Thirteen participants were included (Table 4).

Figure 9. Participant flowchart in study III

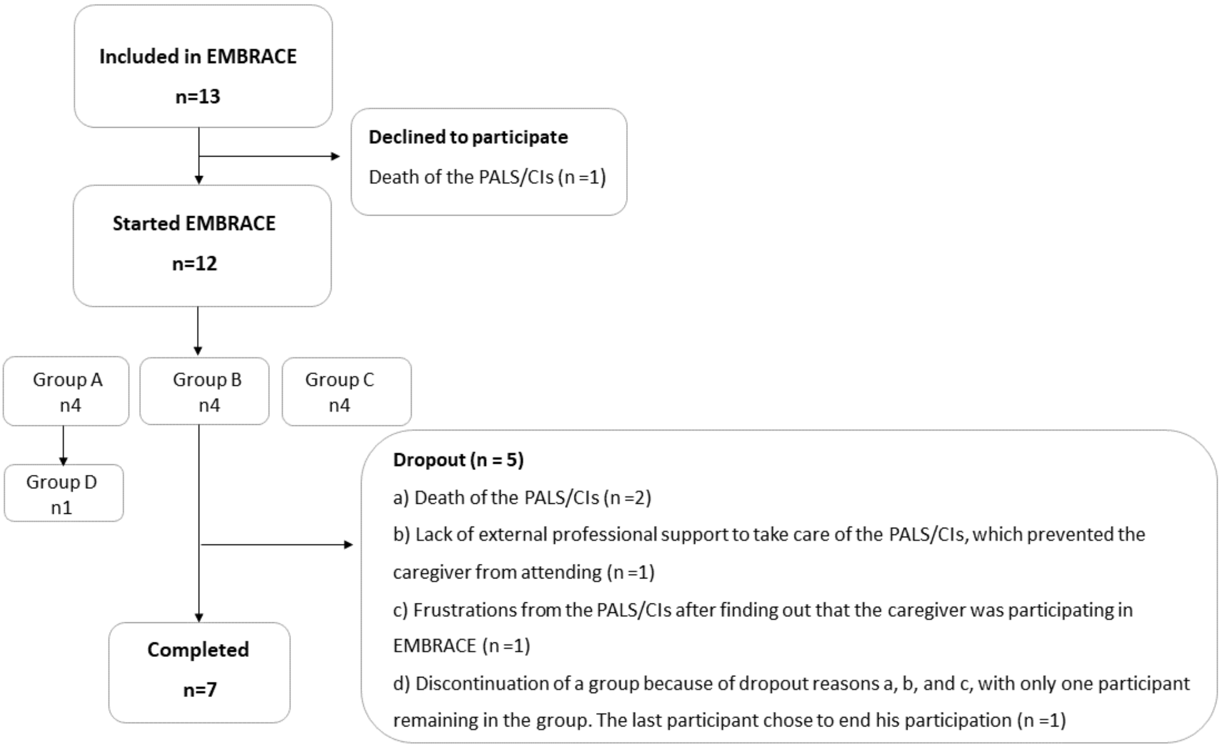

Table 4. Characteristics of participants, including pre-and post-intervention characteristics on participants' levels of burden, anxiety, and depression using the Zarit Burden Interview (ZBI) and Hospital, Anxiety and Depression Scale (HADS)[127, 114]. Scores are separated into participants dropping out and the intervention group. Post-intervention scores were collected right after finishing the 4-month EMBRACE intervention. ZBI ranges from 0-88 points with higher scores indicating greater burden [127]. Scores ≥24 indicate clinical high burden[37]. HADS cut-off score ≥8 indicate possible cases of anxiety and/or depression [114].

Study population at baseline (n = 13[2])		
Gender, n (%)	Male	4 (30,77%)
	Female	9 (69,23%)
Age (years), median (range)		58 (39–70)
Relation, n (%)	Married	13 (100%)
Occupational status, n (%)	Working	8 (61,54%)
	Early retirement/retired	5 (38,46%)
Trajectory of ALS as a caregiver of a PALS/CIs[3] (months) median (range)		25 (2–173 months)
ALS-FTD-Q score of the person with ALS, median (range)		35 (24–55)

	Background characteristics of the caregivers		
	Study population pre-intervention (n = 12[4])		**Study population post-intervention**(n = 7)
	Baseline (n =5) **Drop-out group** median (range)	**Baseline** (n = 7) **Intervention group** median (range)	**Post-intervention** (n = 7) **Intervention group** median (range)
Burden	46 (33–64)	38 (26–56)	42 (34–54)
	Baseline (n = 4[5])	**Baseline** (n = 7)	**Post-intervention** (n = 7)
Anxiety	9,5 (4–14)	5 (4–14)	10 (5–12)
Depression	6.5 (4–11)	8 (1–12)	7 (1–12)

Participants who were not eligible for EMBRACE were offered support from relevant healthcare professionals from RCFM.

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[2] Total number of included participants. One participant lost her spouse with ALS and cognitive and/or behavioral impairments between the pre-intervention data generation and start of the EMBRACE intervention.

[3] Abbreviation for person with amyotrophic lateral sclerosis and cognitive and/or behavioral impairments

[4] One participant did not return the questionnaires.

[5] One participant did not return the questionnaire.

Data

Interviews were carried out with participants pre- and post-intervention and with non-completers post-intervention to capture potential barriers related to participation (p. 26). The participants received the ZBI and HADS pre- and post-intervention (p. 28) [114, 127].

Analysis

For the qualitative analysis process see page 31.

Study IV – article IV

Design

A qualitative design employing participant observations were applied (p. 27).

Sampling and recruitment

Participants from the first and second round of EMBRACE were eligible (p. 36). Nineteen participants were included and divided into four groups (Table 5).

Table 5. Demographic characteristics of the participants in study IV.

Participants		**(n = 19)**
Gender	Male	5
	Female	14
Age	39–50	3
	51–55	5
	56–67	7
	68-74	4
Relation	Married	19
Occupational status	Working	9
	Early retirement/retired	10
Trajectory of ALS (years)	0–2	10
	3–4	3
	5–8	4
	9–14	2
ALS-FTD-Q score	22–30	6
	31–35	5
	36–40	4
	41–46	2
	47–55	1
	56–74	1

Data

Participant observations were collected on 17 video-recordings of virtual group meetings between August to December in 2020, and again between August to December in 2021 (p. 27).

Analysis

A secondary analysis of the 12 recordings from the first round of EMBRACE was performed as the primary analysis [117] gave rise to an additional question regarding online peer-support that this study sought to further investigate. A primary analysis of raw data was performed for the five recordings from the second round of EMBRACE in 2021. For descriptions of the analyses of participant observations see page 31.

The researcher's role

As a former ALS-counsellor at RCFM, I have obtained clinical knowledge on everyday life with ALS and the challenges that caregivers are faced with and struggle with. Working at RCFM allowed for easy access to the expertise of the multi-professional ALS-team [128] that I used to be part of but also to families in which a person had ALS/CIs. I was familiar with the ALS symptoms and cognitive and behavioral impairments. I also had prior knowledge about how these impairments impacted on the caregivers' lives and the caregivers need of an intimate environment, where they could speak freely, where their needs were given attention, and where they could get support. As part of introducing myself to the potential participants, I intentionally signed the invitation for EMBRACE as an ALS-counsellor and PhD student from RCFM as a way of stating that I possessed a specific ALS knowledge, and that I was affiliated with a highly specialized rehabilitation center. Because I used interviews as a method to generate some of my data, I interacted with the participants and used myself as an instrument to gain knowledge [104]. My knowledge on everyday life with ALS and the profound burden on the caregivers made it easier for me to get the participants to share perspectives and elaborate on their thoughts during interviews. I was explicit about my role as a PhD student and why I wanted to interview them. Occasionally, the participants asked a question related to ALS, which I answered to the best of my knowledge and asked if they needed further information and support, I offered to reach out to their ALS-counsellor at RCFM or their clinician at the hospital.

Ethical considerations

All participants were informed about the project orally and in writing, about its aim and their rights such as their ability to withdraw from the study at any time without risking consequences on their affiliation with RCFM. All participants signed an informed written consent before the intervention started. Data were anonymized and participants referred to by IDs to ensure confidentiality. The study was registered retrospectively on clinicaltrials.gov [ID no. NCT04638608] on November 20, 2020, due to my unawareness of pre-registration requisites. The sub-studies followed the Declaration of Helsinki [129]. According to the adjudications made by the Central Denmark Region Committees on Health Research Ethics [File no. 1-10-72-1-19] and by the Danish Data Protection Agency [File no. 2019-521-0144] the project was not liable to notification.

Ethical considerations related to the EMBRACE intervention (Study II-IV)

Caregivers who were not eligible for EMBRACE were offered support from relevant healthcare professionals at RCFM. Invitations for EMBRACE were sent by letter addressed to the PALS and the caregiver, clearly stating that the intervention was targeted caregivers of PALS/CIs. By sending a physical letter I envisioned that it most often would be the caregivers who picked up letters because of the physical impairments of the PALS/CIs. This would leave the caregivers with the opportunity to decide whether to share the information in the invite with their partner or not, depending on personal preferences. I knew from clinical practice that some caregivers experienced cognitive and/or behavioral impairments in their partner, and that the invitation might cause conflicts or sorrow. I also knew that PALS/CIs do not always have insights in their own disease. Participation in the EMBRACE intervention did not require approval from the PALS/CIs, and the caregivers' participation was kept confidential among the research team and the participants and was not disclosed to healthcare professionals at RCFM.

For the first round of EMBRACE, participants were encouraged to invite a designated person (a friend or family member) to participate alongside them in the virtual group meetings to support them now and later in the illness trajectory. Only two participants chose to invite one of their adult children. The designated persons, in this case adult children, were regarded as support for their healthy parent and were therefore not given the same attention and support as the other spouse-participants during the virtual group meetings. However, taking part in EMBRACE as a child of a fatal ill parent with ALS/CIs was challenging and unethical, causing me to change the procedure for the second round of EMBRACE. Participants for the second round were, thus, no longer allowed to invite a person to participate alongside them. Instead, they were encouraged watch the videos in EMBRACE and to share their own experiences and thoughts with someone outside the intervention as a means to support them during and after the illness trajectory.

The virtual group meetings enabled the participants to share experiences and concerns, which entailed that other participants and their designated person[6] would overhear and get personal insights from their fellow group-members. To ensure confidentiality, the group facilitator initially and explicitly stated that things shared within virtual group meetings (or the chat) between the participants were not to be shared elsewhere. When participants dropped out due to the death of their partner, lack of support from professionals or frustration from their partner, they were asked by the group facilitator or me whether this information should be provided to their group-members during virtual group meetings, which they always gave permission. A moment of silence was held after sharing the news of the death of a partner. Those who had lost their PALS/CIs were welcome to participate in the following meeting to say goodbye to the group. One participant chose to do this.

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[6] The participants in the first round of EMBRACE were allowed to invite a person from their family or network to participate along with them, except the PALS/Cis. The designated persons were not regarded as participants and therefore not interviewed. Their role were to support the caregivers of PALS/CIs through the illness-trajectory of the affected relative. (appendix 27).

Findings

The main findings from the four sub-studies are presented separately and with further descriptions and examples of participant quotes and questionnaire outcomes provided in article I-IV.

Article I: Reflections of family caregivers and health professionals on the everyday challenges of caring for a person with amyotrophic lateral sclerosis and cognitive impairments: a qualitative study.

The aim of this study was to explore the everyday challenges and needs of caregivers and professionals supporting the PALS/CIs.

Findings showed the reflections on everyday challenges retrospectively as perceived by seven caregivers of deceased PALS/CIs and nine professionals working in the homes of the families (Figure 10) [29]. Reflecting perspectives of 14 women and two men.

Three themes related to the caregivers were identified

Adjusting to new roles while balancing, demonstrated that the caregivers continuously had to adapt to new roles and learn new skills as ALS/CIs progressed. They described that having a partner with ALS was a project in which they struggled with multiple roles, being either a spouse or child of a parent with ALS/CIs while at the same time taking on the work of 'nurse', coordinator, or project manager.

Accepting that nothing else matters, demonstrated how the caregivers put their own lives on hold to support their partners. They described how ALS/CIs had put life into perspective emphasizing that it was 'now or never'. They were conflicted about both wanting to make the most of the little time left, but also sometimes hoping that it would all come to an end because of the burden and struggle related to the ALS/CIs.

Realizing different values in relationships revealed that families, friends, and professionals could be both supportive and burdensome. While the supportive relations were perceived as a lifeline when dealing with everyday challenges, the burdensome relations to the professionals were frustrating, causing increased burden and insecurity.

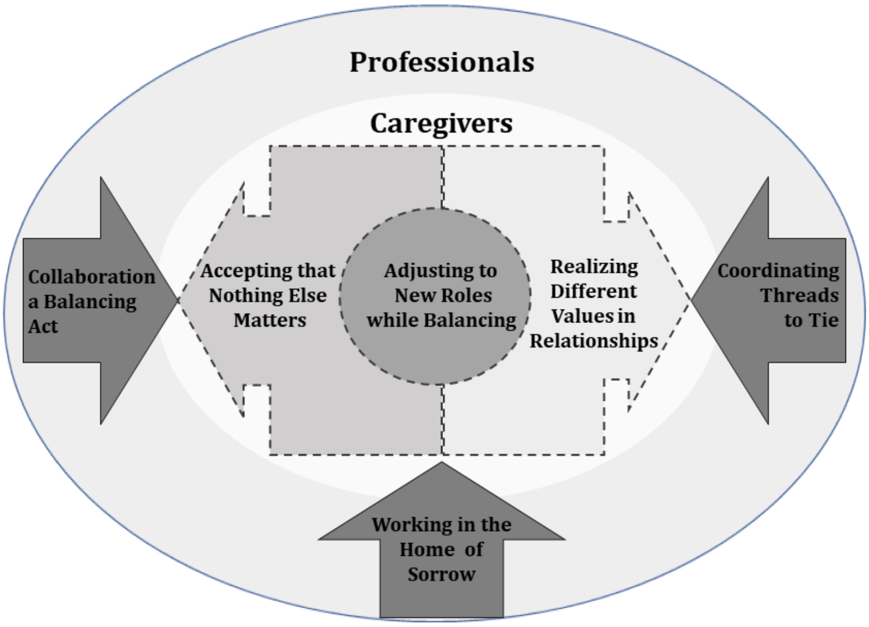
Three themes related to the professionals were identified

Working in the home of sorrow reflected how the professionals sensed the caregivers' sorrow, frustration, and hopelessness and how the professionals tried to support the caregivers in various ways.

C*ollaboration a balancing act* emphasized how the professionals had to be careful using the right words, and how disagreements between the caregivers and the professionals were often caused by failure to align expectations for the levels of support and help in advance.

C*oordinating threads to tie* described how the professionals belonged to different authorities, departments and professionals that supported the families, and how coordination was imperative when trying to support the complex care needs, but not always easy or without consequences for the professionals [29].

Figure 10. Reflections on everyday challenges of family caregivers and health professionals in coping with a person with amyotrophic lateral sclerosis and cognitive and/or behavioral impairments. The findings represented the caregivers (illustrated in the inner circle) interacting with the health professionals, who were seen as intruders in the everyday lives of the families [29].



Article II: A cross-sectional evaluation of acceptability of an online palliative rehabilitation blended learning program for family caregivers of people with amyotrophic lateral sclerosis and cognitive and behavioral impairments.

The aim was to examine the participants' acceptance of the EMBRACE intervention.

Findings reflected the participants' acceptance of EMBRACE guided by the seven TFA constructs [99]. Findings were based on participant observations of 12 participants during virtual group meeting and interviews post-intervention with 10 of the 12 participants. Nine women and three men were part of the participant observations. Perspectives from the same nine women and one of the men were obtained in the post-intervention interviews.

Affective attitude primarily concerned the virtual group meetings and the importance of engaging with peers. The intervention was experienced as a welcoming brake from everyday stress, and the participants were grateful for being offered the support and for the intervention's focus on their needs, challenges, and experiences. The meetings were perceived as a free forum where they could share anything they worried about. The video content reminded them of their own situation and challenges, and it felt nice to be mirrored in others and acknowledged for their feelings, thoughts, and concerns.

Burden referred to technical issues, interruptions during meetings, lack of time and difficulties in reading body language. Because of the technical issues and interruptions, the participants experienced how their attention shifted and how it was difficult to prioritize themselves due to high caregiving responsibilities.

Ethicality dealt with how group meetings became a place for the participants to share detailed descriptions of intimate and sorrowful situations, that they had not shared with anyone else. However, because of loyalty to the PALS/CIs and not wanting to expose their partners unnecessarily, sharing entailed careful consideration for the participants. They generally felt that no one in their network understood what they were going through and that dealing with and sharing concerns with people in their network was difficult. They furthermore feared being frowned upon or seen as unlikable.

Intervention coherence represented the participants' understanding of the purpose of EMBRACE. Observations revealed that they had to be reminded of the purpose, whenever their focus shifted to the PALS/CIs instead of themselves. Targeted knowledge from the videos and discussions from the virtual group meetings enhanced the awareness of their needs and situation and how to handle them. The video content was regarded as relevant and engendered emotional conversations that they were not able to have outside the group. Being in the same situation enabled them to relate, share and support each other.

Opportunity costs described how participants had to take time off from work or cancel a study group to be able to participate. Despite the lack of time, they highly prioritized participating in the virtual group meetings, sometimes gaining time by not watching the videos beforehand.

Perceived effectiveness described how the EMBRACE intervention was found relevant and useful, the most favored components being peer-support and the videos. EMBRACE provided a safe environment for like-minded others and offered an accessible supportive intervention with the opportunity to discuss sorrowful topics. The virtual group meetings were regarded as invigorating, removing some of the frustrations, burden, and loneliness, and even causing bodily relief and easier breathing.

Self-efficacy concerned how a flexible and non-demanding setting of EMBRACE made it easy for the participants to engage without feeling guilty for not watching the videos beforehand. The online format was crucial for their ability to participate despite their lack of time. Lack of concentration and poor memory made it challenging to focus and remember things. The virtual group meetings helped change some of the participants way of thinking as they gained insights which supported them in coping with everyday challenges.

Article III: Experienced benefits and challenges of an online palliative rehabilitation program for family caregivers of people with amyotrophic lateral sclerosis.

The aim was to explore the participants' experiences of benefits and challenges of the EMBRACE intervention before and after the intervention.

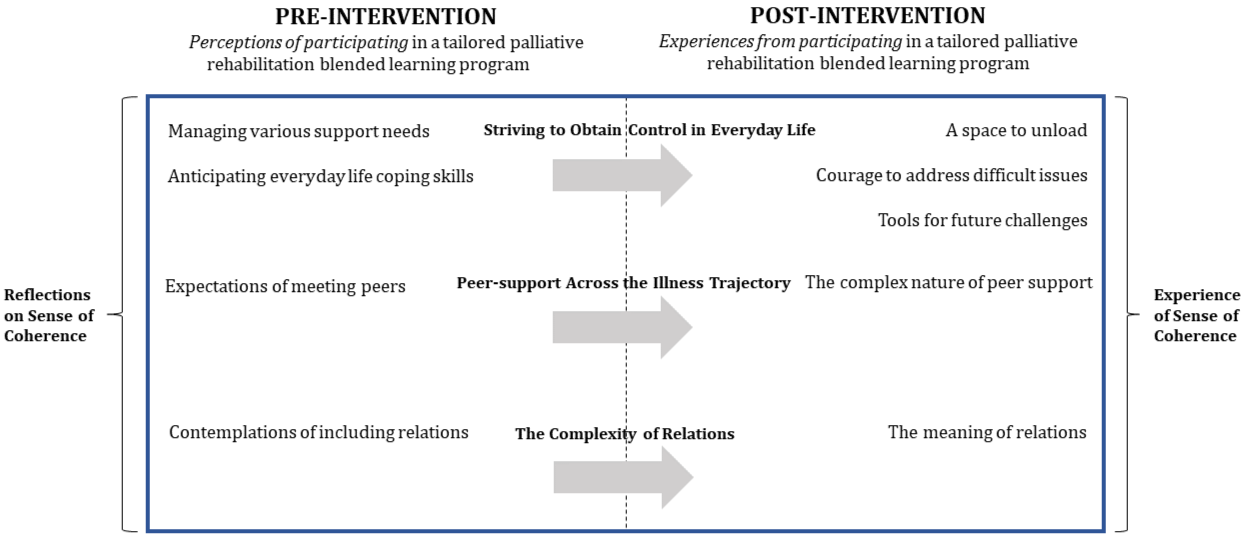
Findings represented the participants' expectations for and perceptions of benefits and challenges of EMBRACE (Figure 11). Thirteen participants were interviewed pre-intervention, and 10 of them post-intervention. The findings comprised three overarching themes described below.

Striving to Obtain Control in Everyday Life involved the support needs and coping skills that the participants anticipated that EMBRACE could provide to support them in dealing with a PALS/CIs. It demonstrated how they, post-intervention, felt they had encountered a space for unloading feelings and thoughts, and how they had gained new insights in dealing with everyday challenges. Finally, it showed how they had found the courage to address difficult issues which was necessary when trying to get support.

Peer-support Across the Illness Trajectory concerned their expectations for meeting peers who represented various stages of ALS and how this entailed both advantages and inconveniences. It appeared that some of their anticipations for the intervention ended up matching their perceptions of the intervention. For instance, the heterogeneity of the groups turned out to be confronting but also important and instructive for the way they would comprehend and manage everyday life.

The Complexity of Relations revolved around the participants' struggling to include other people in everyday life with all the challenges it entails, such as balancing the private sphere and supporting the complex care needs. The theme also reflected the experiences of estranged relations to people part of their network, who were reluctant to deal with or at unease with the PALS/CIs and the concomitant sorrow.

Figure 11. A visual depiction of expected benefits and challenges before the EMBRACE intervention and experiences of benefits and challenges after the intervention. (In preparation).



Article IV: Peer-support among family caregivers of people with amyotrophic lateral sclerosis and cognitive impairments in a palliative rehabilitation blended online learning program

The aim was to explore what goes on in online peer-support among the participants in EMBRACE.

Findings were based on participant observations during 17 virtual group meetings of 19 participants in EMBRACE, reflecting 14 women and five men.

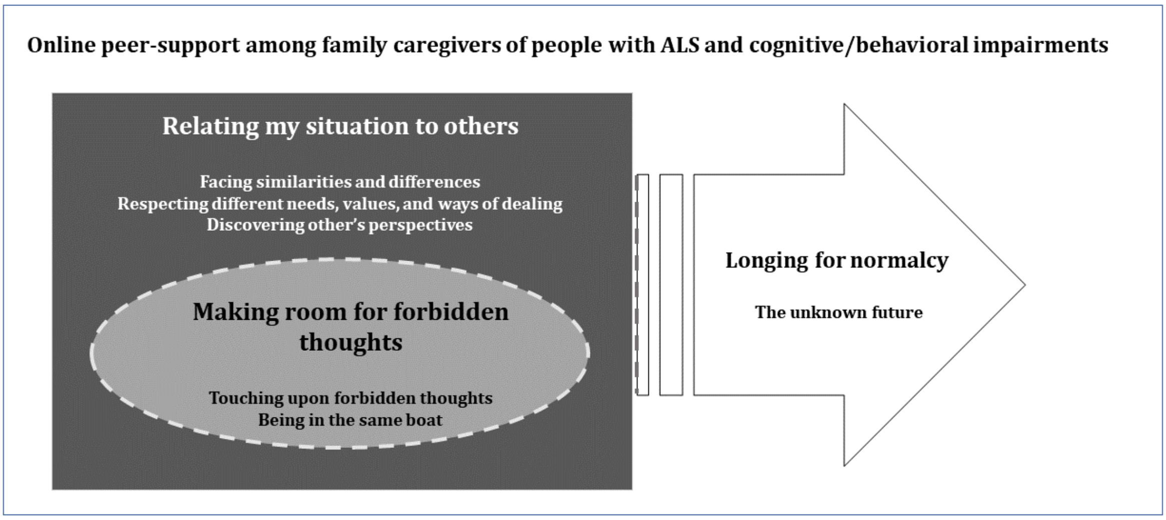
Findings revealed three themes elaborated below (Figure 12).

Relating my situation to others represented the various ways caregivers interacted in the virtual group meetings and how the interactions initially and throughout the intervention revolved around practical and emotional issues. It became apparent that there was no ‘one size fits all’ and that the participants embraced and respected different ways of dealing with challenges and needs, something which seemed to be related to their personal values, stages of disease and everyday situation. The participants shared their arduously learned lessons as a way of supporting and preparing each other for the illness trajectory.

Making room for forbidden thoughts entailed touching upon sensitive topics related to the caregiver role. Sharing personal and sorrowful concerns and frustrations engendered feelings of trust and a sense of being in the same boat. The participants could relate to each other’s stories and acknowledged the experiences shared between them. The online setting and the participants’ approach created an environment where they could speak candidly and without risking being frowned upon. It appeared that the familiarization empowered them to address their genuine wishes for a normal future with all the trivialities that ALS/CIs had robbed them of.

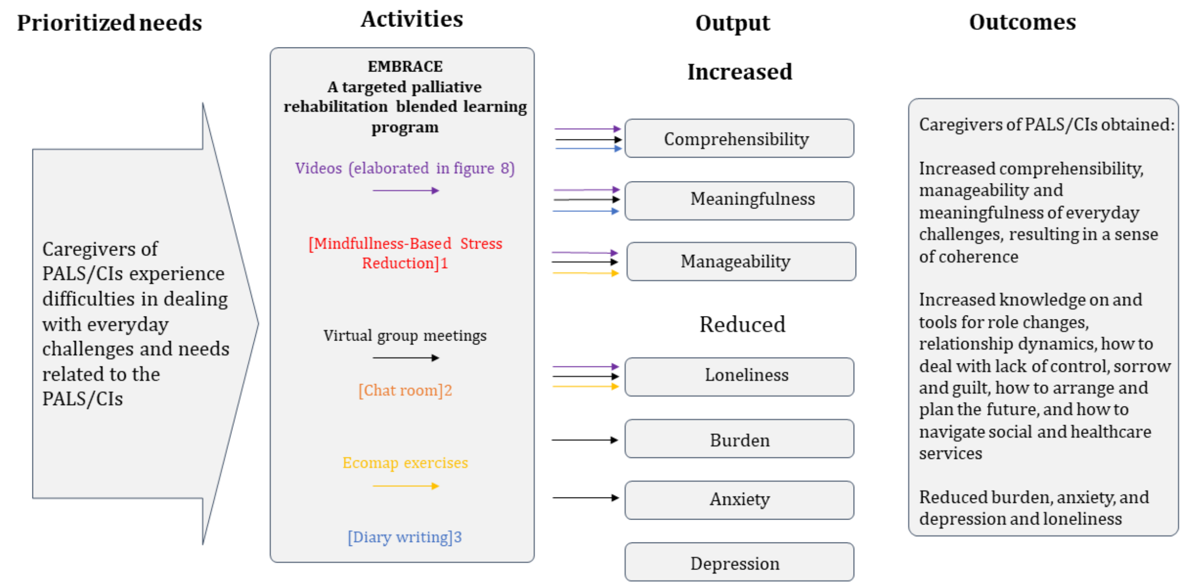
Longing for normalcy concerned the uncertainties related to the unknown future of their partners' illness trajectory, and their longing for reverting to everyday life post-mortem. It seemed like they had prepared themselves for a fast trajectory at the time of diagnosis but had realized that the ALS/CIs continued for good and bad. The unknown future kept the participants in suspense, enhancing feelings of impotence and restraint because ALS/CIs overruled everything and stood in the way of their future dreams.

Figure 12. Understanding of what goes on in online peer-support among caregivers of people with ALS and cognitive and/or behavioral impairments in the palliative rehabilitation blended learning program, EMBRACE. (*In review with the Journal of Evaluation in Clinical Practice).*



In summary, a refined program theory was developed based on the findings from the evaluation of EMBRACE (**study II-IV**) illustrating with precaution the overall assumptions drawn on the qualitative empirical findings with refinement needs highlighted at the bottom for a version 2.0 of the EMBRACE intervention (Figure 13).

Figure 13. The refined program theory of the EMBRACE intervention illustrating with precaution the overall assumptions drawn on the empirical findings from study II-IV.



1. Mindfulness: Not usefull. Alternative exercises were used by those who had been familiar with relaxation exercises before EMBRACE
2. Chatroom: Not used. Reasons: not nessesary between meeting, prefered face-to-face communication, not a 'chat-kind-of-person'
3. Diary writing: Beneficial for those who had been using a diary before the intervention

Discussion of findings

First, I will elaborate on the different options for online peer-support in EMBRACE and discuss the different relevance of peer-support. Second, findings from all sub-studies relating to the cognitive and/or behavioral impairments of the PALS will be discussed. Third, I will discuss the illness related uncertainties of the affected partner and its influence on the participants' struggle during everyday life.

The meaning of online peer-support

The participants had two options for engaging with peers: in virtual group meetings and through written messages in a private chat room. It appeared that only one of the options for peer-support was preferred and considered important for the participants. The goal of EMBRACE was to reduce the magnitude of adverse impact on the caregivers' everyday life of the disease of their partner [130]. Findings showed that the virtual group meetings were the most favored and beneficial component in the intervention. The chat room, on the other hand, was not used by the participants; only one participant shared a chat message about the loss of her relative, which two participants responded to. Contrary to the findings of other studies [65, 131, 132], it seemed obvious that the type of peer-support offered through written messages in a closed chat room was not relevant for this group of caregivers. In the studies mentioned, online peer-support was provided through chat messages with which the sharing of experiences facilitated recognition and acknowledgement among caregivers of children with chronic asthma [131], caregivers of PALS [65] and people with depression [132]. Contrary to findings of positive aspects of online peer-support provided through chat messages, findings from **study II and III** indicated that the participants did not perceive it necessary or relevant because they would be meeting in virtual groups with short intervals of time and preferred to pose their questions and share experiences "face-to-face". Some participants expressed that it was difficult to start the chat conversation because they did not know what to write or because they did not know their peers that well. This was also found in another study on caregivers of PALS and progressive muscle atrophy [65]. Additionally, the study found that messages tended to be too generic and not useful because everyone had different needs and challenges [65]. Regarding the generic messages, this aspect remains unclear within the EMBRACE intervention as the participants did not even start writing chat messages.

Different categories of support have been encountered in online support, with informational and emotional support being the most frequent ones [133-135]. This was also the case with my findings. In line with a recent study, I found that the norm of giving and receiving support usually followed a reciprocity where confiding with someone in a similar situation was important during a period of personal crisis [130]. People need to connect in times of crisis to reestablish a sense of personal self-efficacy along the path from disillusion and despair to reconstruction of the psychological growth and improvement of the quality of life [130]. With the design of EMBRACE, the participants were able to connect virtually in groups every 4th week for four months but also between group meetings in a chat room. Despite their becoming closer with each other as the intervention progressed, and continuous emphasizing the importance of meeting and discussing with peers, they did not feel the need for or had the courage to write a post on the chat. These findings contradict findings from a systematic review on chat-based hotlines for health promotion provided by health professionals [136]. The review showed that instant message applications were preferred over other modes of services like, face-to-face counselling, voice calls, emails, and text messaging [136]. Further, it indicated that chat-based hotlines using messaging services had mostly positive and statistically significant effects on mental health, e.g., anxiety, depression and well-being with user satisfaction being moderately high [136]. Contrary to the review findings, some of our participants explicitly stressed that they were not a “chat-person”, implying that they never wrote messages on social medias.

The virtual group meetings were regarded as both: important, relevant, and necessary but also challenging and confronting for the participants who were new to ALS/CIs. The reason for the ambivalence was the potential risk of getting glimpses of future situations and challenges related to more severe stages of ALS/CIs. Despite the heterogeneity in disease stages and physical, cognitive, and behavioral impairments of their partner with ALS/CIs, I found that the participants were still able to give and receive peer-support during the virtual group meetings. Being of value for others bolstered the participants’ eagerness to continuously share and support each other in spite of being at different stages of the disease, which created a safe environment for them for sharing experience and gaining new insights in dealing with everyday life. In line with these findings, Smit et al. (2021) also found that people with depression sensed a feeling of belonging, emotional growth, self-efficacy, and empowerment from corresponding in writing in an online community for like-minded others [132]. Furthermore, a study on caregivers of people with Alzheimer’s showed that caregivers found respite in online communities where the written support mainly pertained to caregiver well-being and was provided in a positive tone [137].

In summary, EMBRACE offered an accessible platform for peer-support with the possibility to exchange experiences and receive support that was crucial in continuing to deal with everyday life as a caregiver of a PALS/CIs. Interestingly, findings showed the preference for online peer-support provided through virtual group meetings over chat messaging. This indicates that peer-support offering “face-to-face” contact through a virtual group meeting was important in terms of giving and receiving relevant support from like-minded others.

Reflections of cognitive and behavioral impairments of the PALS on study findings

As research indicates that caregivers of PALS/CIs are more burdened than caregivers of PALS [39, 42], the focus on EMBRACE was on the caregivers’ challenges and needs in everyday life related to the affected partner. Interestingly, in all study findings in the PhD project, ALS seemed to overshadow the entailing cognitive and/or behavioral impairments and was the terminology that the participants referred to when sharing everyday experiences in the virtual group meetings and in pre- and post-intervention interviews. Apparently, they did not differentiate between ALS and ALS/CIs. Nevertheless, research clearly shows that the presence of cognitive and/or behavioral impairments in PALS have a negative influence on caregiver burden and quality of life [25, 34, 37, 40, 41]. The fact, that the participants did not seem to differentiate between ALS and ALS/CIs in their statements could be explained by the fact that it is easier to say *"ALS"* than to say, "*cognitive impairments"* and/or *"behavioral impairments"* which are not always easy for laypersons to pronounce. This was seen from the participant observations in EMBRACE. Again, the reason for the participants not differentiating may also be explained by the reason that they were included in EMBRACE based on their partner's illness, and that the focus of the intervention was on their needs. Conversations concerning the illness progression and practical aspects such as applying for a car was therefore guided back to the needs of the caregivers by the group facilitator. It might also have been a more or less active coping strategy not to mention the cognitive and/or behavioral impairments during the group meetings [138]. Research shows that caregivers may feel ashamed because of the stigma experienced in relation to dementia [139] and try to conceal it from others [140]. Internalizing public stigma is seen in adult children of parents with dementia when they feel ashamed or embarrassed about their affected relative [141]. A recent systemic review of caregiver coping strategies in ALS and frontotemporal dementia showed similar coping strategies with problem-solving and positive emotion-focused coping being the most utilized strategies [138]. Furthermore, the review also found that no coping strategies were used conclusively at different times and in specific stress situations [138].

In summary, findings should be interpretated with caution in terms of being representative of caregivers of PALS with cognitive and/or behavioral impairments as they might not represent the complex and multiple challenges related to the specific cognitive and/or behavioral impairments seen in relation to ALS, but perhaps "only" represent ALS-caregivers. Furthermore, caregivers of PALS and caregivers of persons with frontotemporal dementia may benefit from the same problem-oriented interventions [138] and thereby potentially also from the EMBRACE intervention, giving that the participants in EMBRACE did not seem to highlight nor pay any attention to the cognitive and/or behavioral impairments [142].

Navigating the illness related uncertainties as a caregiver of a PALS/CIs

Findings across the sub-studies indicated that the participants constantly had to struggle with practical as well as emotional aspects of being a caregiver of a PALS/CIs and that this included adjusting to new roles and responsibilities. These findings are in line with what Conroy et al. (2021) found in a multi-center, exploratory study of caregivers of PALS [28]. Furthermore, like my findings on caregivers of PALS/CIs, Conroy et al. also found that the caregivers of PALS delt with feelings such as sadness, sorrow, worry, anger, frustration, and stress caused by the uncertainties related to the future illness trajectory. Uncertainty is a common psychological reaction to serious illness [143] and is defined as the *"inability to determine the meaning of illness-related events"* (p.225) [144]. Not being able to accurately envisage or predict how an illness will proceed causes uncertainty [143, 145]. Research shows how struggling with complex, unfamiliar, and potential threatening situations has a negative influence on physical and psychological functioning and enhances feelings of losing control, fear, emotional distress, and diminishes quality of life in cancer patients [146-149]. Findings in this PhD project reflect the experiences of caregivers representing the full ALS trajectory of their partner, from few months to 14 years of experience with ALS. Compared to Mishel's (1988) *uncertainty in illness theory*, which consists of four major component; antecedents generating uncertainty, appraisal of uncertainty, coping with uncertainty and adaptation to the illness [144], these components or stages of coping with uncertainty may reflect what I found. For instance, I found that caregivers with years of experience with ALS had already adapted to the illness and their "new" everyday life situation in many ways, while those who were new to ALS were, naturally, at the earlier stages of appraisal or coping with the illness related uncertainties of their partner. Navigating and managing the uncertainties related to being a caregiver of a PALS/CIs was challenging for the participants and entailed lack of control in everyday lives and longing for normalcy. Furthermore, their needs seemed to be related to alleviation of grief and mourning as well as functioning. This could be explained by a theory review that show that uncertainty can be associated with lack of control both mentally and psychically [143]. Participants in EMBRACE received information on the illness related uncertainties of their partner and on how to comprehend, manage and find meaning in these uncertainties in relation to their reduced ability to control everyday life [90-92]. The participants' longing for normalcy can, according to a recent study on the complexity of normalcy, be explained by the important role that it plays for patients' illness experiences and quality of life [150].

This indicates that caregivers of PALS/CIs go through a parallel process of having to face and struggle with illness related uncertainties of their partner. Furthermore, the uncertainties have a negative influence on the participants' well-being and cause feeling of grief, sorrow and frustration which underlines their needs for alleviation of suffering and mourning and support of functioning in everyday life as a caregiver of a PALS/CIs. Palliative rehabilitation thereby seem important to this vulnerable group of caregivers [53].

Discussion of methods

This section outlines my critical considerations regarding the four frameworks used in this PhD project. First, I will discuss how the MRC framework from 2021 influenced on the evaluation of EMBRACE given the changes made from the 2008-guidance. Second, I will discuss the use of the ID methodology followed by a discussion of the credibility of the PhD studies which is assessed by the four quality criteria in ID; epistemological integrity, representative credibility, analytic logic and interpretive authority [85]. Third, I will discuss the relevance of using SOC to guide the design, development of EMBRACE, analysis and writing of findings. Fourth, I will discuss the TFA and how the deductive approach was beneficial and challenging when investigating the participants' acceptance of EMBRACE.

Discussion of MRC frameworks

Since its publication, the 2021 MRC framework was used to strengthen the reporting of the acceptability **(study II)** and evaluation **(study III and IV)** of EMBRACE. The framework's broader focus on the contextual influence was a strength because it enabled non-linear associations and potential side effects to emerge through the evaluation of complex social systems within EMBRACE [77, 78]. As prescribed in the updated framework I found that outcomes were not always possible to estimate due to the nature of complex social systems that could be unpredictable [77, 78]. For instance, the estimated benefits of mindfulness and diary writing turned out to be mostly on the individual level with participants having multiple reasons for accepting or not accepting each of the components and the combination of them. The more pragmatic approach outlined in the updated framework provided a flexibility to choose the optimal study design within the PhD project to best investigate and understand the complexities [77, 78]. If informed by the 2008-guidance, focus would have been more narrowly on an individual level, the effectiveness or cost-effectiveness of EMBRACE [80]. This would have left us with a "black box" and missing insights into the contextual associations and an inability to unravel the complexities of EMBRACE, including the participants' experiences of the intervention [78]. At worst, this could have led to research waste with misinterpretations or a knowledge gap [152] e.g., if the reasons for not performing the mindfulness exercises had not been uncovered within the evaluation of EMBRACE. The 2021 framework guided the evaluation of EMBRACE in the direction of focusing on what works for whom and under which circumstances. This was clearly a strength as it helped us understand the "black box" of EMBRACE and expanded our understanding of the program theory in a non-linear way (Figure 13) [77, 78].

As recommended by the 2021 MRC framework, various stakeholders were involved across the phases of developing, testing, and evaluating EMBRACE [77], e.g., caregivers of deceased PALS/CIs, participants in EMBRACE, healthcare professionals at the RCFM and the supervisors. The recommendation on involving stakeholders throughout the project was a strength as it enhanced the chances of gaining all relevant perspectives and addressing and incorporating these in developing and evaluating EMBRACE [77, 115].

Research shows that involving participants from an intervention [78] and those with lived experiences (**Study I-IV**) in research is beneficial as they possess an expertise that no one else could bring to the team [153]. In retrospect, the involvement of stakeholders during the development, test and evaluation phases of EMBRACE have not been addressed much in the articles but it was implicitly executed in each of the first three phases of the MRC framework [77]. Involving stakeholders in research has been emphasized as important to improve the quality of research, moving towards effective interventions [154]. This PhD project focused on the participants' perspectives of EMBRACE and future attention should also include perspectives of the distributers, e.g., the professionals delivering the intervention [78, 99, 115].

Overall, the use of the 2008 and especially the 2021 MRC framework was a strength as they provided useful guidance and support for the development, test, and evaluation of the intervention [77, 78, 80]; however, the evaluation could have been strengthened if supplemented by a SOC questionnaire as described by Akbari et al. (2021) [155]. Assessing participants' self-reported SOC pre- and post-intervention using a SOC-questionnaire would have helped clarify whether EMBRACE had enhanced the participants' comprehensibility, manageability, and meaningfulness and thereby their SOC. Furthermore, the program theory could have been strengthened if informed by a learning theory like Lave and Wenger's on situated learning in communities of practice [156] or Knud Illeris on a comprehensive understanding of human learning [157]. Employing a learning theory to guide the design, components, and content of EMBRACE may have strengthened the learning outcomes. Such theories regard learning as an interplay between the human (the participants) and its environment (the EMBRACE setting, group-members, and the group facilitator) [156, 157], which may have helped understand the learning mechanism among the participants. For instance, how to provide and tailor information to participants who experience reduced ability to concentrate and remember because of the stressful situation which influences negatively on their learning ability [158].

Discussion of the ID methodology

According to ID, human reality is individually constructed, contextual and complex, and research within this qualitative inquiry therefore involves human beings (i.e., caregivers and professionals) as well as context (the EMBRACE intervention), which made ID suitable for my studies [85]. Overall, ID provided a methodological path for me based on the applied practice orientation with the research questions founded in practice with the purpose of investigating and understanding practice and finally to inform and improve practice at RCFM through reapplying the knowledge gained in the PhD studies [85, 159]. As previously stated, (p. 19), ID claims to be an unconventional methodology applying a more pragmatic approach that allows the researcher to choose between the optimal methods and theories [85, 87]. However, in the spite of its free approach, ID also include requirements and elements that must be followed such as providing a structured and logic guidance from the initial development of research questions, to scaffolding, framing and strategizing the study, to entering the field and conceptualizing findings in a structured, yet iterative, analysis process leading to practice implications relevant in the "real world" [85]. Questions concerning the methodological freedom within ID may therefore not be as free as intended [85], something which has been challenged and addressed by other researchers [160, 161]. ID also recognizes the opportunities within conventional approaches and therefore encourages researchers to go beyond the disciplinary standpoints and borrow from different approaches in the relevance and benefits of the research purpose [85]. This, however, was never an issue in this PhD project as it was originally designed as an ID study and followed the elements of ID while also including the theoretical lens of SOC to help understand the areas in focus [90]. The stepwise analysis description in ID was a help as it visualized and structured the process, making it easy to follow while simultaneously enhancing the transparency of the analysis process [85, 86]. ID also challenged me as a new researcher, as it does not provide clear descriptions of when to stop the interpretation in order not to drag the interpretations too far [85]. The quality assessment criteria proved to be beneficial in enhancing the studies credibility in a transparent and systematic way [85].

Overall, the ID methodology served as a strength as it offered structure and guidance on how to design the whole research project. Still, I experienced some challenges, especially during the interpretation of findings.

The credibility of the PhD dissertation was assessed by the four evaluation criteria in ID; epistemological integrity, representative credibility, analytic logic, and interpretive authority [85]. They are elaborated on and discussed below.

Epistemological integrity

Epistemological integrity concerned the consistency between the basic assumptions about the essence of knowledge and the research question [85]. I pursued epistemological integrity by enhancing the consistency between the epistemological viewpoints and research aims. In line with ID, my epistemological standpoint was aligned with the interpretive naturalistic understanding that concedes the contextual and constructive nature of human experience [85]. ID does not have one specific knowledge standpoint, and the researcher therefore has the responsibility to explicitly account for, argue and form the line of reasoning throughout the research process and centrally to the epistemological integrity [85]. As a novice researcher, I assumed the responsibility of carefully choosing elements that were relevant for the project and respectfully considered how they might influence on each other and the findings gained [85]. As an occupational therapist with a master's degree in educational psychology I acknowledge human experience as individualized, complex, and socially constructed and influenced by the contextual setting. Therefore, interviews and participant observations seemed like credible tools to explore the participants' perspectives on their own challenges and needs and the EMBRACE intervention. The alignment between epistemological groundings and the research aims assisted me in focusing and understanding the "real world" while at the same time acknowledging the social and contextual influence that surround human experiences [85].

Representative credibility

Representative credibility is obtained when the findings made are coherent with the methods applied to study the specific area of research [85]. The pragmatical approach of ID allowed different methods, such as interviews, participant observations, and questionnaires to be employed in no prearranged order [85]. This was a strengths as triangulating data sources offered different angles, breadth, and depth to explore and elucidate the study aims which enhanced representative credibility [85]. Furthermore, the different data sources provided audio, visual and texted types of data which nuanced the understandings and potential discrepancies in what was said and done in the virtual group meetings and what was said directly to the interviewer pre- and post-intervention [85].

ID recognizes the maximum variation in data before knowledge claim can be attempted within qualitative findings [85]. In line with this, I strived to gain insights from a broad range of participants, both caregivers and professionals in **study I**, as well as from caregivers representing various characteristic like age, sex, years of living with ALS, social status, etc. in **study I-IV** [85]. I strived for purposive sampling for the caregivers in **study I**, but ended with convenience sampling, since it turned out to be difficult to recruit participants [85]. The sample provided first hand perspectives representing various characteristics and thereby helped me gain nuanced and in-depth insights into the retrospective everyday reflections of challenges and needs among caregivers of deceased PALS/CIs [29]. A potential limitation might have been their ability to recall memories [162], or their wish to leave their partner/parent with a good reputation [163]. However, I was not relying on their specific information *per se*, I was interested in how they reflected upon their caring experiences. The participants openly shared sorrowful and shameful episodes from their past life with the deceased PALS/CIs and they did not express difficulties in recalling everyday life with the PALS/CIs (**study I**). Findings from **study I** may reflect caregivers of deceased PALS/CIs who felt that they were able to give an interview, e.g., not in severe grief or under great stress, which could be a limitation and affect the representative credibility by representing more positive reflections on the past everyday life with the PALS/CIs. Moreover, findings represent some participants who had lost their partner or parent years ago and some who had lost recently. Although this broad representation is a strength, it may constitute a risk of diluting the findings by not representing characteristics such as short span since death of the PALS/CIs, which might impact on the level of grief and stress and thereby retrospective reflections. However, these aspects were never the intention for **study I**.

The online format made participant observations of the virtual group meetings difficult e.g., sensing the atmosphere, interactions and picking up expressions and behavioral signs of the participants because of the two-dimensional format with everyone looking in same direction (into the camera). Findings based on participant observations in **study II and IV** may therefore lack nuances in the descriptions on behavior and interactions between the participants.

As previously discussed, it appeared that the cognitive and/or behavioral impairments of the PALS were not often explicitly addressed by the participants and, thus, not reflected in most findings. However, the aim of the PhD project was to address and support the everyday challenges and needs of caregivers of PALS/CIs and not those of the PALS/CIs. In Denmark it is not yet common practice at all neurological hospital departments to perform formal extensive (neuro)psychological assessments for cognitive and/or behavioral impairments of the PALS. Furthermore, it is not always possible to do a broad assessment in the clinical practice because of fatigue in PALS, time, and shortage of qualified personal [164]. I strived to ensure and optimize the inclusion of caregivers of PALS/CIs by applying a commonly used behavioral screening tool, the ALS-FTD-Q, which is sensitive to all the behavioral domains that are often affected in PALS [164]. I knew that such screening could be biased, not necessarily reflecting the actual levels or the whole behavioral spectrum of impairments in the PALS [165]. Various instruments, like the Edinburgh Cognitive and Behavioral ALS Screen (ECAS), or the amyotrophic lateral sclerosis-frontotemporal dementia cognitive screen (ALS-FTD-Cog) measuring the cognitive impairments might have strengthened the reliability of the findings [166, 167]. Conversely, a review on screening instruments for cognitive and behavioral impairments in PALS shows, that the cognitive instruments may be too concise to detect language impairments [164]. Using separate instruments to detect behavioral impairments could probably provide more detailed and/or reliable findings [164] and thereby be more suitable for screening for behavioral impairments in PALS [168]. Various screening instruments have been developed with some being more promising than others, including the ALS-FTD-Q [164]. The various instruments result in PALS ranging widely in level of cognitive and behavioral impairments between the different instruments [142, 164]. I was aware of this potential bias and still chose to apply only one screening instrument to assess the behavioral impairments to reduce the burden on the caregivers. A questionnaire carried out by caregivers was applied because it is important and effective to include their insights as they may be better at observing and detecting changes in behavior and cognition from ALS-onset [14]. Attention should be drawn to the questionnaire score that may be influenced by and representing the caregiver's own burden which, in turn, could cause higher scores of their affected partner on each item [169]. Challenges and bias have been found in different instruments employed in screening for cognitive and/or behavioral impairments [142, 164, 170]. Moreover, I envisaged that several questionnaires as part of the inclusion process for EMBRACE might prevent some caregivers from reaching out to be included because of the extra “workload”, [171].

Findings from the sub-studies may lack a men's perspectives, which might have provided more specific knowledge on potential sex-related differences in perceived challenges and needs, thereby enhancing the representative credibility, although this was not the purpose of the PhD project. The vast majority of participants were female spouses, which is commonly seen in caregiver research [172], ALS-caregiver research [24, 26, 28, 173, 174], and in research of caregivers of PALS with behavioral impairments [25]. Our sample size in all studies may be regarded as a limitation due to few participants. Findings from **study II-IV** were nevertheless based on the participants included in EMBRACE. This was a strength because interview data represent pre- and post-intervention experiences from participants included in the first round of EMBRACE, as well as perspectives from those who dropped out, which enabled me to explore and understand both benefits and potential barriers for participation in the intervention (**study III**).

In summary, findings can, to a certain extent, be representative of (female) caregivers of PALS, but not necessarily of the influence of the specific cognitive and/or behavioral impairments seen in relation to ALS. The study population represents caregiver-spouses of people with rare and devastating disease(s), ALS (and cognitive and/or behavioral impairments). They represent a burdened and vulnerable group of caregivers between thirty and seventy years of age. Our findings and practical implications can, to a large extent, be generalized to other groups of vulnerable caregivers who struggle to deal with everyday life as a caregiver of a partner with a progressive and fatal disease, like incurable cancer and dementia.

Analytic logic

Analytic logic entailed the existence of a clear rationale infiltrating all stages of the study, from design, data generation and analyses, to the final interpretations and conclusions [85]. To enhance credibility of the analytic logic in this dissertation, I have attempted to explicate my positioning in the field as a researcher and a healthcare professional and my choices of methodological and theoretical frameworks that served as “scaffolding” for the studies [85]. Within ID there is no clear answer to the balance between description and interpretation in the analysis, and as a new researcher it was challenging to find the right balance. I therefore cooperated with my supervisors and the founder of the ID methodology (Professor Thorne in **study IV**) to increase credibility of the findings [88, 175]. The iterative ID analysis process was beneficial as it ensured a profound and in depth coherent interpretation of data, relating data to the findings [85, 86]. To achieve analytic logic and enhance transparency of the analysis process, a model reflecting the findings with its joint relations was created [85, 86].

Interpretive authority

Interpretive authority concerned the trustworthiness of the findings made by me as the researcher [85]. In line with ID, I acknowledge that I, like any other individual, will be under the influence of others including the surrounding environment and structures [85]. I continuously remained attentive to and reflected on my presumptions as a healthcare professional within ALS rehabilitation, a researcher and person. As an “insider” in the field of research, I applied reflexivity throughout the process of preparing and carrying out the studies to pursue trustworthiness [85, 176]. For instance, I explicitly attempted to identify and consider my own presumptions and biases, such as my initial “know how” in relation to caregivers’ challenges and needs from my previous clinical practice, and I continuously confronted my presumptions with questions like “what else could this mean?” [85, 176]. Furthermore, to strengthen the interpretive authority and reduce the potential researcher subjectivity of the studies, I continuously discussed my perceptions and reflections with the research team, which included researchers conducting the intervention and researchers who did not [85]. By being an “insider” and using myself as the “instrument” in the project, I was aware of the potential risk of seeing what I wanted to see [85]. According to ID, my "insider" position was also an advantage in applied practice research, as the researcher has clinical knowledge on the matter studied, and this may be useful when entering the field or in preparation of e.g., interview guides [85]. On the other hand, because I had only been working with ALS for two years prior to the PhD project, I also consider myself partly an outsider and not too shrouded in the ALS-practice. Additionally, I did not have any former relations with the participants prior to the studies, except from two related participants that I had met in a hospital prior to the interview in **study I**. Nor was I part of the execution of EMBRACE and thereby not indirectly in contact with the participants during the intervention. To enhance transparency of the analysis process and to reduce the risk of extending the interpretation too far from the data, I documented the process from initial interpretations to the development of coherent and overarching themes (Article IV, figure 3). When presenting the findings in the articles, I documented these with quotes from the participants to illustrate my interpretation and conclusions and thereby my interpretive authority [85].

Discussion of SOC

SOC was beneficial as a methodological framework to inform the development of EMBRACE as it helped focus on health promotion and participants' well-being as they faced health undermining stressors as a consequence of their partner's illness [92]. Guided by the SOC elements, I strived to promote the participants' health by shaping and enhancing their SOC through indirectly tailoring the intervention components to enhance comprehensibility, manageability, and meaningfulness [90, 92]. SOC thereby served as a strategic advantage in strengthening focus and providing SOC elements helpful to uncover and understand the complex interactions between the participants and the intervention components in EMBRACE [90, 92].

SOC also acted as a powerful theoretical framework to guide the research questions, study designs, and analytic direction. SOC was used to uncover the reflections on everyday challenges and needs (**study I**) and evaluate EMBRACE (**study II-IV**) by inductively exploring the participants' SOC. I acknowledge that this way of indirectly conceptualizing SOC and its elements through the processes of empirical studies may not be within the "spirit" of SOC [92, 177]. Antonovsky intended to investigate SOC using SOC questionnaires [92, 177]. In this PhD project, SOC was explored qualitatively using interviews and participant observations as methods to understand the participants' comprehensibility, manageability, and meaningfulness. The SOC elements were used indirectly throughout the analysis process, alongside ID or the TFA. The reason for this was the more explorative study aims. The qualitative methods provided nuanced subjective insights into the participants' SOC and thereby broadened the understandings in relation to evaluating EMBRACE. Investigating SOC using a SOC questionnaire might have provided outcome scores that were comparable to other study results. However, questionnaires would not have provided and expanded the complex understandings of EMBRACE in terms of clarifying the intervention components impact on the participants' comprehensibility, manageability, and meaningfulness. By employing and assessing SOC indirectly as an translation tool, I reduced some of the limitations associated with overreliance on self-reporting and observations and minimized the likelihood of general simplistic findings and interpretive mistakes [85]. A recent scoping review on the SOC questionnaire showed that the SOC concept has expanded but is still employed to identify people's level of SOC or when planning an intervention to promote people's SOC [178]. Retrospectively, including a SOC questionnaire pre- and post-intervention as a supplement to the qualitative data may have expanded insights into the estimates of effectiveness of EMBRACE on the participants' SOC and each of the SOC elements, giving a larger study population. A scoping review emphasized that future research, on rigorous interventions, should determine if recognizing people's level of SOC through a SOC questionnaire score enables more tailored care of the person struggling with illness-related conditions or not [178]. In line with the epistemology of Antonovsky, I perceived the participants as interacting with the constantly changing environment [90, 179]. Informed by SOC, the focus of the studies was on the participants in their context, which was a strength as it enhanced the understanding of the dynamic and interrelated aspects, e.g., the participants' experiences of EMBRACE and how this indirectly influenced on their perception of SOC [90, 179].

In summary, SOC provided a framework with elements useful in both designing, developing and evaluating EMBRACE which expanded my possibilities of translating their experiences into knowledge useful in the practice of RCFM but also in a broader perspective in the applied world [90].

Discussion of the TFA

Guided by the TFA, I deductively investigated the participants' acceptance of EMBRACE through seven theory-driven constructs [117]. The TFA provided constructs with anticipated definitions representing broad aspects of acceptability [99]. In EMBRACE, this broad and clear understanding of acceptability, based on the TFA, seemed like a strength before the investigation. On one hand, during and after the investigation of acceptability, the nuanced aspects of acceptability may have fragmented data because of the seven constructs and this, in turn, could limit a coherent interpretation of acceptability of EMBRACE. On the other hand, the specific constructs may also have provided insights into acceptability that could have been overlooked if not explicitly highlighted in the framework. For instance, the interview guide was not informed by the TFA constructs, which caused some constructs like "ethicality" and "opportunity costs" to be overlooked resulting in a lack of insight into aspects of acceptability as defined by Sekhon et al. (2017) [99]. Further, the deductive coding of data was sometimes challenging as some of the constructs seemed to cluster, which is acknowledged by the research group behind the TFA [99]. A limitation of deductive coding in relation to the TFA may also be that not all data fit into the constructs, and the question is then, what to do about these data?

According to Sekhon et al. (2017), acceptability is a subjective evaluation made by the individuals (participants in EMBRACE) who experience (or expect to experience) an intervention [99]. In EMBRACE, data triangulation employing participant observations and interviews was used to assess the participants' acceptability of EMBRACE. However, Sekhon and colleagues state that both qualitative and quantitative methods can be applied for this purpose. They highlight individual and focus group interviews as well as reflective diary entries [99]. I collected participant observations during virtual group meetings without interrupting the participants with questions related to their acceptability. Still, I performed a deductive analysis guided by an observation guide developed on the basis of the TFA constructs on the participant observations. The structured deductive guide was surprisingly easy for the members of the research group to apply, and this strengthened the focus on the seven constructs. Nevertheless, this way of assessing acceptability may not fit within the TFA, as it can also be challenging to sort out what is observed or is not observed and why [85]. Still, I regard data from the participant observations as a strength because they provided knowledge of the participants' doings and saying during the virtual group meetings, which was then supplemented, compared, and broadened by perspectives shared in the individual post-intervention interviews. A limitation may be that the pre-intervention interviews were not included. Thus, the TFA suggests that assessing prospective intervention acceptability can help explore the full course of the participants' experiences of acceptability of EMBRACE [99]. Recently, a theory-driven generic TFA questionnaire was developed to evaluate acceptability of interventions and the use of such questionnaire may have strengthened the reliability of the findings [180]. The questionnaire contains a total of 10 questions, with two questions to choose from within the constructs of "affective attitude" and "ethicality" [180]. In total, eight questions are scored, one for each of the seven constructs and an extra question related to the "generalized acceptability" [180]. Sidani et al. (2011) have also developed a framework focusing on acceptability in terms of adherence, appropriateness in addressing the presenting problem, convenience, risks and effectiveness [120]. This framework has overlaps with the TFA, but I chose the TFA because it nuanced the understanding of the participants' acceptability by adding constructs such as burden, self-efficacy, ethicality, and opportunity costs which were not part of the framework from Sidani et al. [99, 120].

Overall, the use of the TFA was a strength as it helped focus and nuance the participants' acceptability of EMBRACE; however, it was also challenging sometimes to deductively choose the construct that fitted best based on empirical data. Luckily, agreement within the research team was always reached.

Conclusion

The overall aim of the PhD project was to develop, test and evaluate the palliative rehabilitation blended learning program EMBRACE to support caregivers of PALS/CIs in dealing with everyday challenges and needs related to their affected partner. Based on the findings, the following conclusion can be drawn:

Firstly, this PhD study contributed to profound and important insights into caregivers of PALS/CIs' experiences of everyday challenges and needs related to a deceased PALS/CIs. In depth understandings were obtained on caregivers of PALS/CIs' experiences on acceptance, benefits, and challenges related to the EMBRACE intervention. The findings demonstrated how the participants constantly had to adjust to and struggle with changes in their everyday lives caused by the deterioration of their partners functioning. Furthermore, the findings revealed how the illness of a partner gave rise to uncertainties that had a negative impact on the participants' well-being and ability to control everyday life. To comprehend, manage and find meaning in a complex situation as a caregiver of a PALS/CIs, the participants had a need for being alleviated of their mourning and grief as well as support for functioning in everyday life. These challenges emphasized the relevance and importance of palliative rehabilitation to support this fragile group of caregivers throughout the illness trajectory of their partner.

Secondly, an important finding was that the disease ALS seemed to overrule the entailing cognitive and behavioral impairments which seemed to be less relevant to the participants during their participation in the EMBRACE intervention. This may reflect the fact that they were included in the EMBRACE intervention based on their affected relative with ALS/CIs and therefore had no need to differentiate between illnesses among like-minded others.

Thirdly, findings illustrated that the participants perceived the EMBRACE intervention as acceptable and beneficial. Clearly, peer-support was the most important aspect because it enabled online face-to-face interaction and sharing of experiences with people who recognized and acknowledged the difficult feelings and concerns that penetrated everyday life as a caregiver of a PALS/CIs. The EMBRACE intervention facilitated a safe environment for the participants where they could share intimate, sorrowful, and frustrating stories which was not possible outside the group meetings. Peer-to-peer support across all stages of ALS provided the participants with beneficial insights and knowledge that could be used in their management of everyday challenges and needs as a caregiver of a PALS/CIs.

Fourthly, findings demonstrated how the EMBRACE intervention, guided by SOC, promoted the participants' health by tailoring intervention components that would enhance their comprehensibility, manageability, and meaningfulness. Furthermore, I found that adding SOC elements helpful to unravel and understand the interactions between the participants and their context to evaluate the benefits and challenges of the EMBRACE intervention was a strategic advantage. SOC also helped illuminate the intervention components that could be strengthened to reduce the challenges perceived by the participants.

Finally, the EMBRACE intervention provided an environment for the participants to engage, relate and share personal experiences, concerns and frustrations related to being a caregiver of a PALS/CIs. This knowledge should be seen in combination with existing evidence on the positive influence of peer-support, tailored components and information provided through an accessible online intervention to support caregivers of PALS/CIs during the illness trajectory of their relative.

Implications for online caregiver support

Based on the findings in this dissertation, I suggest that attention should be paid on the following implications for online caregiver support to challenge and enhance the benefits of an online palliative rehabilitation blended learning program.

Focus should be on online support interventions that aim to address and support caregivers' needs of being alleviated of their mourning, grief, and support their functioning in everyday life. This is important as caregivers go through a parallel process of having to adjust to a new everyday life like their affected partner. Tailored interventions based on a palliative rehabilitation approach should provide insights, understandings, and tools to enhance caregiver management of everyday life, in terms of how to deal with illness related uncertainties, lack of control and what to hope.

The fact that no illness differentiation between ALS and the entailing cognitive and/or behavioral impairments was made by the caregivers may indicate that the EMBRACE intervention could act as an online support intervention to caregivers of PALS with or without cognitive and/or cognitive impairments, supporting their dealing with everyday challenges and needs related to the affected partner.

It is further suggested that online support interventions should incorporate virtual face-to-face peer-support groups facilitated by a healthcare professional. To enhance learning outcomes, groups of caregivers representing different stages of ALS/CIs is beneficial, as both the experienced and the less experienced caregiver can reflect and potentially learn and adjust their ways of dealing based on beneficial insights gained from one another. Regular meetings every month through a minimum of four months is recommended as development of relations take time online and is necessary for the caregivers to feel comfortable to share aspects from their lives.

It was demonstrated that the EMBRACE intervention, informed by SOC, was beneficial in enhancing the caregivers' comprehensibility, manageability, and meaningfulness during a chaotic life situation. SOC should be regarded and included as a resource, strengthening the focus in the development, and tailoring of a health promotive support intervention for caregivers to enhance their SOC and thereby reduce their experience of stress.

These above-mentioned initiatives and suggestions on the development and tailoring of an accessible online support intervention may enhance interventions targeting caregivers in dealing with everyday challenges and needs related to PALS/CIs.

Future research

This PhD dissertation presents knowledge on caregivers of PALS/CIs' experiences of the EMBRACE intervention. Still, there are areas to explore in future research. Suggestions on areas for future research to confirm, nuance and broaden the insights gained in this project are outlined below.

I. *Transferability of the EMBRACE intervention*
   The fact that the caregivers did not seem to differentiate between ALS and ALS/CIs it could be beneficial to explore and clarify whether EMBRACE is beneficial for caregiver of PALS without cognitive and/or behavioral impairments or not. This can lead to insights into the interventions' transferability to caregivers of PALS and potentially also other groups of caregivers struggling with progressive and fatal illnesses of a partner, such as cancer and dementia.

II. *Perspectives of distributers*
   Focus on the project was on the recipients e.g., the caregivers and their perspectives of the EMBRACE intervention. To enhance a coherent understanding of the EMBRACE intervention, it is important to assess the perceptions of benefits and challenges of the distributers e.g., the professionals executing EMBRACE.

III. *Additional support intervention for professionals*
   The findings demonstrated that relations with professionals supporting the families were complex and entailed conflicts due to lack of knowledge and clearing expectations. The collaboration between the professionals and the families might be better if the professionals were informed about how to address and support the complex needs of the families. It could therefore be beneficial to deliver the already developed support intervention targeted professionals and explore their experiences of intervention.

IV. *Perspectives of the PALS/CIs*
   The findings documented the caregivers' challenges and needs related to PALS/CIs and the caregivers' experiences of the EMBRACE intervention. It could, however, be beneficial to explore the perspectives of PALS/CIs to provide insights on how it is to be the indirect cause of the need for a support intervention for the caregiver.

English summary

Family caregivers of persons with amyotrophic lateral sclerosis and cognitive and/or behavioral impairments (PALS/CIs) are highly affected by the deterioration of their partner's functioning causing high caregiver burden, anxiety and depression. Amyotrophic lateral sclerosis (ALS) is a progressive and fatal neurodegenerative illness with no cure. Around half of the people with ALS are at risk of developing cognitive and/or behavioral impairments. The cognitive and behavioral impairments in persons with ALS are known to increase caregiver distress. This PhD project is affiliated with the Danish National Rehabilitation Center for Neuromuscular Diseases.

The overall aim of the PhD project was to develop and evaluate the palliative rehabilitation blended learning program named EMBRACE, by involving caregivers of PALS/CIs and professionals. The dissertation consists of four articles based on four empirical studies, employing individual and focus group interviews, participant observations and questionnaires to generate data. In all, 28 caregivers and nine professionals participated in this study.

In article I [29], the aim was to explore the reflections of family caregivers of deceased PALS/CIs and professionals on retrospective everyday challenges and needs related to supporting PALS/CIs. The analysis on family caregiver data revealed three interrelated themes: *Accepting that nothing else matters, Adjusting to new roles while balancing*, and *Different values in relations*. The themes represented the caregivers' constant struggle to comprehend, manage, and to find meaning with their relative's illness in everyday life, and furthermore revolved around promotive or restraining relations with family, friends, and professionals. Three themes were identified in data on the professionals: *Collaboration a balancing act, Working in a home of sorrow*, and *Coordinating threads to tie.* The themes illustrated how the professionals acted as an outside interference on the families lives because of the family's dependency on support but also how the complex care of the PALS/CIs required coordination with different departments, organizations, and people.

In article II [117], the aim was to investigate the participants' acceptability of EMBRACE. A deductive analysis showed that *affective attitude* referred to the importance and meaning of peer-support. *Burden* concerned the technical challenges and frequent interruptions. *Ethicality* concerned the exposure of the affected partner while sharing own experiences. *Intervention coherence* referred to the shared destinies among the participants. *Opportunity costs* concerned work-related costs. *Perceived effectiveness* addressed the usefulness of engaging with peers to learn how to deal with challenges now and later in the illness trajectory. *Self-efficacy* represented their willingness to learn about the illnesses.

In article III (in preparation), the aim was to explore the participants' perceptions of benefits and challenges of EMBRACE. Three overarching themes were identified from the analysis. The themes represented an interrelated parallel process containing both individual and interpersonal factors that influenced the participants' experiences: *Striving*

to Obtain Control in Everyday Life, Peer-support Across the Illness Trajectory, and The Complexity of Relations.

In article IV (in review), the aim was to investigate what goes on in online peer-support among participants in EMBRACE. Three themes were identified: *Relating my situation to others'* concerned the different ways caregivers interacted and how these initially evolved around practical topics. Later, as the participants began to familiarize, they started *Making room for forbidden thoughts* such as personal and sorrowful thoughts and frustrations that they could not share elsewhere. Feelings of trust and a sense of belonging empowered them to share their genuine wish and *Longing for normalcy* with all the trivialities that ALS/CIs had robbed of them.

The overall conclusion in this dissertation is that caregivers of PALS/CIs struggle with illness related uncertainties of their partner causing lack of control and feelings of no sense of coherence in everyday life. The caregivers have needs of being alleviated of their suffering and mourning as well as supported in their daily functioning. A palliative rehabilitation approach guided by SOC was beneficial in enhancing the caregivers' comprehensibility, manageability, and meaningfulness in a chaotic everyday life. Caregivers in the EMBRACE intervention did not seem to differentiate between ALS and ALS/CIs, which may indicate that EMBRACE is transferable to caregivers of persons with ALS without cognitive and/behavioral impairments.

Dansk resumé

Pårørende til personer med amyotrofisk lateral sklerose og kognitive og adfærdsmæssige forandringer (PALS/CIs) er i høj grad påvirket af deres partners sygdomssvækkelse, hvilket belaster den pårørende og fører til angst og depression. Amyotrofisk lateral sklerose (ALS) er en uhelbredelig, progressiv og dødelig neurodegenerende sygdom. Omkring halvdelen af populationen med ALS risikerer kognitive og/eller adfærdsmæssige forandringer. De kognitive og adfærdsmæssige forandringer ved personer med ALS er associeret med lidelse blandt pårørende. Dette Ph.d.-projekt er affilieret ved RehabiliteringsCenter for Muskelsvind i Danmark.

Det overordnet formål med Ph.d.-projektet var at udvikle og evaluere den palliative rehabiliteringsintervention, kaldet FAVN, gennem brugerinvolvering af pårørende til PALS/CIs og professionelle. Afhandlingen omhandler fire artikler baseret på fire empiriske studier, der anvender individuelle- og fokusgruppe interviews, deltager observationer og spørgeskemaer til at generere data. I alt deltog 28 pårørende og ni professionelle i projektet.

Formålet med artikel I [29] var at undersøge pårørende til afdøde PALS/CIs og professionelles' refleksioner over hverdagsudfordringer og behov relateret til PALS/CIs. Tre sammenhængende temaer blev identificeret i data fra de pårørende: *Accept af at intet andet betyder noget*, *Tilpasning til nye roller, mens man balancerer* og *Relationers forskellige betydninger.* Temaerne repræsenterede de pårørendes konstante kamp for at begribe, håndtere og finde mening med deres pårørendes sygdom i dagligdagen og omhandlede samtidig de fremmende og hæmmende relationer til familie, venner og professionelle. Tre temaer blev identificeret i data om de professionelle: *Samarbejde en svær øvelse*, *Arbejde i sorgens hjem* og *Koordinerede tråde at binde*. Temaerne illustrerede hvorledes de professionelle agerede som udefrakommende forstyrrelser på familiernes liv grundet deres afhængighed af professionel støtte samt hvorledes den komplekse pleje af PALS/CIs krævede koordinering med forskellige afdelinger, organisationer og personer.

Formålet med artikel II [117] var at undersøge de pårørendes accept af FAVN. En deduktiv analyse viste, at *Følelsesmæssig attitude* refererede til vigtigheden og betydningen af peer-støtte. *Belastning* omhandlede tekniske udfordringer og hyppige afbrydelser. *Etik* omhandlede eksponeringen af den sygdomsramte, når man delte egne oplevelser. *Intervention sammenhæng* refererede til de fælles skæbner blandt deltagerne. *Lejligheds omkostninger* omhandlede arbejdsrelaterede omkostninger. *Oplevet effektivitet* adresserede gavnligheden af at engagere sig med ligesidede for at lære at håndtere udfordringer nu samt senere i sygdomsforløbet. *Tiltro til egne evner* repræsenterede deres villighed til at lære om sygdommene.

Formålet med studie III (under udarbejdelse) var at undersøge deltagernes oplevelser af fordele og udfordringer ved FAVN. Tre overordnede temaer blev identificeret i analysen. Temaerne repræsenterede en sammenhængende parallel proces, der indebar såvel individuelle som interpersonelle faktorer, der influerede på deltagernes oplevelser:

Stræben efter at opnå kontrol i hverdagen, *Peer-støtte på tværs af sygdomsforløbet*, og *Kompleksiteten i relationer.*

Formålet med artikel IV (i review) var at undersøge, hvad der foregår i online peer-støtte blandt deltagerne i FAVN. Tre overordnede temaer blev identificeret: *Sammenholde min situation mod andres* indebar de forskellige måder de pårørende interagerede på og hvorledes disse i starten omhandlede praktiske foranstaltninger. Senere, da deltagerne havde lært hinanden bedre at kende, begyndte de at *Skabe plads til forbudte tanker* som for eksempel personlige og sorgfulde tanker og frustrationer, som de ikke kunne dele andre steder. Følelsen af tillid og af at høre til gjorde deltagerne i stand til at dele deres oprigtige ønske og *Længsel efter normalitet* med alt det, som ALS/CIs havde frarøvet dem.

Den overordnede konklusion i afhandlingen er, at pårørende til PALS/CIs kæmper med sygdomsrelaterede usikkerheder, hvilket fører til tab af kontrol og en manglende følelse af sammenhæng i hverdagslivet. De pårørende har brug for lindring af deres lidelser og sorg samt støtte af deres gøren og laden i hverdagslivet. En palliativ rehabiliteringstilgang, guidet af teorien om *oplevelse af sammenhæng*, synes gavnlig i forøgelsen af deltagernes begribelighed, håndterbarhed og meningsfuldhed i en kaotisk hverdag. Pårørende i FAVN synes ikke at differentiere mellem ALS og ALS/CIs, hvilket kan indikere, at FAVN er overførbar til pårørende til personer med ALS uden kognitive og/eller adfærdsmæssige forandringer.

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Articles

This PhD dissertation is based on the following original articles which will be referred to by their Roman numbers.

Article I **Reflections of family caregivers and health professionals on the everyday challenges of caring for a person with amyotrophic lateral sclerosis and cognitive impairments: a qualitative study**

Olesen, L.K; la Cour, K.; With, H.; Handberg, C.

Published in: Palliative Care and Social Practice. 2022, February.

Article II **A cross-sectional evaluation of acceptability of an online palliative rehabilitation program for family caregivers of people with amyotrophic lateral sclerosis and cognitive and behavioral impairments.**

Olesen, L.K; la Cour, K.; With, H.; Mahoney, A.; Handberg, C.

Published in: BMC Health Services Research. 2022, April.

Article III **Experienced benefits and challenges of an online palliative rehabilitation program for family caregivers of people with amyotrophic lateral sclerosis.**

Olesen, L.K; la Cour, K.; Nimmon, L.; With, H.; Handberg, C.

In preparation. Planned submitted to: Journal of Evaluation and the Health professions.

Article IV **Online peer-support among family caregivers of people with amyotrophic lateral sclerosis and cognitive impairments in a palliative rehabilitation blended learning program**

Olesen, L.K; la Cour, K.; Thorne, S.; With, H.; Handberg, C.

In review: Journal of Evaluation in Clinical Practice. September 5th, 2022.

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Appendix 27. **Information letter for the co-participant**

Appendix 28. **Informed consent for participation in a health science research project**

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Article I

Palliative Care & Social Practice *Original Research*

Reflections of family caregivers and health professionals on the everyday challenges of caring for persons with amyotrophic lateral sclerosis and cognitive impairments: a qualitative study

Lene Klem Olesen, Karen la Cour, Heidi With and Charlotte Handberg

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Abstract

Aims and objectives: To explore reflections of family caregivers and health professionals regarding the challenges involved in caring for persons with amyotrophic lateral sclerosis and cognitive and/or behavioral impairments (PALS/CIs).

Background: Family caregivers of PALS/CIs are highly burdened and at great risk of psychological sequela. Professionals working with these families can be negatively affected on their well-being and are at risk of burnout.

Design: The design was a qualitative interview study.

Methods: One focus group and 10 individual semi-structured interviews were conducted with seven family caregivers and nine professionals after the death of a PALS/CIs. The analysis was guided by the interpretive description methodology and the theory of sense of coherence. This study adheres to the COREQ guidelines and the ICMJE recommendations.

Results: The family caregivers' challenges regarding coping with everyday needs related to the sick person were associated with 'Accepting that nothing else matters', 'Adjusting to new roles while balancing', and 'Realizing different values in relationships'; whereas the professionals' challenges were related to 'Collaboration a balancing act', 'Working in a home of sorrow', and 'Coordinating threads to tie'.

Conclusion: Family caregivers found coping with the complexity of the diseases a challenge, and their everyday life needed constant adjustment to new roles, coping with inappropriate behavior, and navigating through the progression of the diseases of their sick relatives while collaborating with numerous professionals. The professionals struggled with coordinating and collaborating with the families and with other colleagues due to the severeness and complexity of diseases.

Relevance to clinical practice: Findings point to the importance of relationships for caregivers and professionals and a need to provide support through an online palliative rehabilitation program that encompass coping strategies in relation to the diseases.

Trial registration details: Id no. NCT 04638608.

Keywords: amyotrophic lateral sclerosis, caregiver, cognitive impairment, healthcare professional, interpretive description, multidisciplinary, palliative care, qualitative research, rehabilitation, sense of coherence

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Introduction

Amyotrophic lateral sclerosis (ALS) is a progressive neurodegenerative disease that worsens relentlessly, with death occurring after a median of 3.2 years.[1] A review showed that the number of people diagnosed with ALS is increasing, ranging in Europe from 2.1–3.8 per 100,000.[2] There is now a clear clinicopathological relationship between ALS and frontotemporal dementia.[3] Studies show that about 50% of the people with ALS may also suffer from extra-motor signs and symptoms, such as cognitive and/or behavioral impairments, and 10–15% meet the diagnostic criteria for frontotemporal dementia.[4–6] Family caregivers, like spouses, partners, children, and siblings (hereafter termed caregivers) are described as the pillars of ALS patient care in the literature and research concludes that the consequences of ALS have a profound impact on the psychosocial well-being of these caregivers.[7–9] Two studies show that behavioral changes are the strongest predictor for psychosocial distress in caregivers of persons with ALS.[8,10] Caregivers experience decreased well-being due to the more cognitive and/or behavioral impairments than to the physical disability of the person with ALS.[8,11] Also, health professionals supporting and helping people with ALS are at high risk of experiencing decreased well-being.[12] The presence of executive and behavioral impairments in ALS has also been found to negatively affect the relationship between the ALS patient, family, and the health professionals.[13] Connelly *et al.*[14] conclude that health professionals need psychosocial support that addresses the moral distress and the ethical challenges related to the terminal neurological illness and the communication performance and focuses on a supportive attitude of acceptance of the inevitability of death.

Background

A review of the cognitive, emotional, and psychological manifestations in ALS found impairments in perception and processing of emotions, social cognition, and alterations in certain cognitive functions, such as executive functions, verbal fluency, language, and verbal memory.[3] Moreover, behavioral changes seen in relation to ALS concern disinhibition, apathy, loss of sympathy or empathy, perseverative, stereotyped or ritualistic behavior, hyperorality, or dietary change.[15] Cognitive and behavioral impairments in ALS are associated with more rapid progression and poorer prognosis, with a risk of death being 2 to 2.53 times higher than unimpaired controllers, equal to a median survival of 19–20 months compared to 46 months in unimpaired controllers.[16]

Caregivers of people with ALS are often without any previous experience with the role of a caregiver.[17] Caregivers need to accommodate themselves to the significant needs of the person with ALS as well as to a myriad of associated cognitive and behavioral symptoms within a short period of time.[18] Caregivers are known to neglect their own needs and jeopardize their well-being in order to take care of the person with ALS.[8,9] Rehabilitation assists ALS patients and caregivers to adapt to the psychical and psychological challenges of living with ALS[19,20] and the palliative care that is essential from the debut of ALS[21] because it seeks to alleviate psychical, psychological, and existential distress among the ALS patients and the caregivers and improve their quality of life.[10,22]

Existing research is sparse on which challenges caregivers and health professionals experience in everyday life with the person with ALS particularly in regard to cognitive and/or behavioral impairments (PALS/CIs) and how these can be addressed and supported to enable coping.[23] The aim of this study was to explore reflections of caregivers and health professionals regarding coping with the everyday challenges involved in caring for a PALS/CIs. This effort was undertaken in order to better target future supportive interventions for both caregivers and health professionals.

Methods

Design

The design was qualitative, and the methodology Interpretive Description (ID) was used. ID was chosen as a means to explore the challenges faced by caregivers and health professionals through an inductive analytical methodology.[24] ID aims to create knowledge that can help solve challenges that arise in clinical practice.[24] The theoretical lens of the sense of coherence (SOC) by Aron Antonovsky was applied to explore the participants' ability to cope with life stressors and how three core components, comprehensibility, manageability, and meaningfulness, need to be present in order to cope with and experience life as coherent and thereby reduce stress.[25] Combined ID and SOC guided the whole study from the planning phase, developing of interview guides, and analysis to the writing up of findings by offering an organizing logic with focus on a practice-based problem, methods to use, and analytical interpretive descriptions within the SOC.[24,25]

This study was carried out in Denmark, where public healthcare, including assistive devices, is part of the taxpaid National Health System.[26] A PALS/CIs can receive help and support at home from either community or private healthcare services without being charged. This study was embedded in the PhD project EMBRACE (FAVN) and affiliated with the National Rehabilitation Center for Neuromuscular Diseases (RCFM).[27] RCFM is a highly specialized private outpatient hospital financed by the National Health System in Denmark that supports the public social and healthcare system in rehabilitation of people with different kinds of neuromuscular diseases, including ALS.[28] The professionals are organized into multidisciplinary teams consisting of nurses, occupational therapists, physiotherapists, medical doctors, psychologists, and social workers.[27] The professionals who work with persons with ALS are called ALS counselor and are provided with the possibility to get access to highly specialized knowledge of ALS and rehabilitation of everyday life with the disease.

Data collection

A total of 16 persons participated: caregivers ($n = 7$) and health professionals ($n = 9$), representing approximately 35 PALS/CIs. The caregivers were two adult children and five partners or spouses. The health professionals were nurses, social and healthcare assistants/helpers, visitation and aid consultants, dementia consultants and occupational therapists, and trained people without a professional education employed in private healthcare services. Hereafter, all are referred to as health professionals. Caregivers were eligible if they were a spouse, partner, or an adult child of a deceased person with ALS and if the caregivers described the deceased as having had cognitive impairments as defined in *International Classification* of Diseases 11th Revision.[29] The caregivers were included if they had either been living with the person with ALS until death or had been in a close relationship with the deceased within the last 6 months. Convenience sampling was applied in selecting the caregivers,[24] who were asked to contact the first author after an information letter about the purpose of the study was posted on the websites of the patient organization The Danish Muscle Dystrophy Foundation and the RCFM. The first author then assessed the eligibility of the caregivers according to the inclusion criteria before enrollment. A further two caregivers were recruited through a neurological department at a hospital and one through an ALS counselor at the RCFM (Table 1). Caregivers known to the first author (former ALS counselor at RCFM) or third author (ALS counselor at RCFM) were excluded.

Professionals were eligible to be included if they had a formal experience of caring and supporting PALS/CIs and had either a healthcare education or if they were trained people employed in private healthcare services. Professionals were excluded if they were known by the first or third author. Purposive sampling of the health professionals was applied, aiming for variety among participants in regard to professional background, years of seniority, and experience with PALS/CIs, and their in-depth knowledge by virtue of their equipped experience with the topic investigated.[24] Key leaders from specific departments within community health and social care were identified through the municipal administration of one large and one medium-sized community in Denmark, and one leader from a private healthcare service was selected and invited by phone and e-mail. The key leaders chose the health professionals that fulfilled the inclusive criteria (Table 2).

Between March and May 2020, 10 individual semi-structured interviews were carried out by the first author with caregivers and health professionals by in-person meetings in the homes of the caregivers, by phone, or via Skype interviews. Seven caregivers participated in six individual interviews and two related caregivers were interviewed together. Interviews lasted between 57 minutes and 2 hours 7 minutes.

Focus group interview was used with the health professionals in order to allow a dialogue, rethinking, and discovery of unexpected experiences between health professionals from the same community healthcare services in regard to ALS and cognitive impairments.[24] One semi-structured focus group interview with five participants and four semi-structured individual interviews were conducted with the health professionals by the first author. The focus group interview took place at local health care center, while the third author observed. Focus group interviews with all the health professionals had been planned but was

Table 1. Demographic characteristics of the caregivers that participated in the study.

Participants – caregivers		**($n = 7$)**
Gender	Male	1
	Female	6
Age	<22	1
	23–45	1
	47–55	2
	56–65	2
	>66	1
Relation	Married/spouse/partner	5
	Adult child	2
Occupational status	working	4
	Retired	3
Years of ALS-trajectory	<1	0
	2–3	6
	4–5	1

ALS, amyotrophic lateral sclerosis.

changed to individual interviews due to the COVID-19 lockdown. The individual interviews with the health professionals lasted between 42 minutes and 1 hour 10 minutes and the focus group interview 2 hours 15 minutes. Ten health professionals were invited, but one nurse dropped out due to extensive workload caused by the COVID-19 pandemic. Two semi-structured interview guides were developed for caregivers (Table 3) and health professionals (Table 4). Interviews were digitally recorded and conducted in Danish, and quotes were subsequently translated into English. The research group consisted of four female researchers: PhD student (master's degree in educational psychology, OT, former ALS counselor), professor (PhD, OT), Senior Researcher (PhD, Master of Public Health, RN) both with experience within the fields of rehabilitation and palliative care, and RN (clinical nurse specialist, family therapist, and ALS counselor at RCFM).

Analysis

The analysis was inspired by the three core elements in SOC: comprehensibility, manageability, and meaningfulness, which guided an inductive analysis of the participants' way of coping with everyday challenges related to the PALS/CIs.[25,30] Furthermore, the analysis followed the four-step process guided by the ID methodology.[24] First, all interviews were transcribed in verbatim and uploaded into the electronic software program NVivo™ 12. Second, the transcripts were read and initially coded separately by the first and last author, and subsequently the codes were compared according to circumstances found in the data. Third, preliminary relationships were distilled as categories and a critically appraisal of relationship within all data across subgroups and relevant thematic options lead to the primary categorization. In an iterative process of scrutinizing the codes, the whole research team critically discussed the relationships and patterns within the data, and this led to the final extraction of the main messages and capture of the overarching categorical themes. Finally, the last analytic step resulted in the interpretive thematic and conceptual description that formed a model capturing the main findings regarding the caregivers' and health professionals' challenges in their everyday life with the deceased PALS/CIs.[24]

Ethics

The study was registered on clinicaltrials.gov (Id no. NCT 04638608) and adheres to the COREQ checklist,[31] the Declaration of Helsinki[32] and follows the ICMJE recommendations.[33] The study was not under obligation to notification to the Danish Data Protection Agency (File no. 2019-521-0144), or the Central Denmark Region Committees on Health Research Ethics (File no. 1-10-72-1-19). Participants provided informed consent and both oral and written consent were obtained from all participants prior to data collection.

Results

The analysis with the theoretical lens of SOC gave an overall understanding of the challenges among our participants, the caregivers and health professionals, through six categorical themes.[25] The interrelating themes of the caregivers, 'Accepting that Nothing Else Matters', 'Adjusting to New Roles while Balancing', and 'Realizing Different Values in Relationships', revealed how the caregivers were in a constant struggle to understand and capture the meaning and impact of their situations. The themes unfold the caregivers' difficulties in comprehending and managing the everyday challenges of the PALS/CIs, which caused an overall sense of there being no coherence in everyday life. The relationships with family, friends, and health professionals varied in value and indicated how they either promoted or restrained an SOC among the caregivers. The health professionals acted as an outside interference in the families' everyday lives due to their dependency on help and support, which was represented in the themes: 'Collaboration a Balancing Act', 'Working in a Home of Sorrow', and 'Coordinating Threads to Tie' (Figure 1).

Adjusting to new roles while balancing

The caregivers described how the consequences of their relative falling ill with ALS and cognitive impairments changed the roles in the family and how they had to constantly adjust to these changes.

> Yes, but I was still a wife, but at the same time I was also the wife who was doing an ungrateful job . . . I was also the nurse, and all that. Because it was me who was in charge of everything. (ID 8)

As the PALS/CIs became increasingly impaired, the caregivers explained how they had to take greater and greater charge of the everyday chores, taking on new responsibilities in the housekeeping and learning craftsman skills, such as changing tires and fixing things in the home. The caregivers explained how they had to support and help the relative getting dressed, being groomed, guided through daily activities, and moreover help organize and carry out things during everyday life.

> I pushed his wheelchair into the bathroom, where his commode was, and then, we went into the bedroom, and I got him undressed and tucked him in. And then in the morning, when we got up, it was all about getting him out of bed and into the wheelchair. And getting him bathed and dressed and getting him downstairs and then we started . . . and I had to give him his medicine and his protein supplement and whatever else he needed in the feeding tube. (ID 8)

They explained how they supported and guided the PALS/CIs during public and social arrangements, where they, in addition, had to deal with provocative outbursts and inappropriate behavior by their partner with ALS, which was caused by the cognitive impairments. One participant explained how she was confronted by the manager of a grocery shop.

Table 2. Demographic characteristics of the health professionals that participated in the study.

Participants (professionals) in total		**($n = 9$)**
Gender	Male	1
	Female	8
Residence	Urban (>40,000 Citizens)	5
	Rural (<40,000 Citizens)	4
Years of ALS experience	1	3
	2–3	4
	4–5	1
	>6	1
Professional background	*Healthcare professional	8
	Unskilled	1
*Healthcare profession	Nurses	3
	Occupational therapists	2
	Social assistants/helpers	3

ALS, amyotrophic lateral sclerosis.
*Elaborates which professions the healthcare professionals revolves around.

> '[Name of the affected partner] is no longer allowed here because he steals'. And I remember how sad I felt . . . And when I came home, I saw . . . [name of the affected partner] had also taken candy from the plastic bins, you know, the ones that are placed at a child's level, and I could also see when he came home, that he was sitting on some candy . . . And I remember being very sad when this happened because there were a lot of people, I knew who were just standing there and staring. (ID 10)

The caregivers spoke about how they had to assume many different roles, informal carer, nurse, project leader, all-round secretary, and caretaker, and at the same time being emotional involved as a spouse, partner, or child.

> Yes, she did come to help, and it was partly to let me have time with [the name of the affected partner], but also to be able to have time to do the things that needed to be done besides that . . . Yes, you have many roles in that situation. Yes, but you became a

Table 3. Interview guide for caregivers of deceased persons with amyotrophic lateral sclerosis and cognitive and/or behavioral impairments. (PALS/CIs)

Key questions	Elaborative questions
Try and tell me about yourself	How long has it been since you lost your relative/loved one?
What was your relationship with the person with amyotrophic lateral sclerosis and cognitive and/or behavioral impairments (ALS/CIs) you have lost?	How was your relationship with your family?
Looking back, try to describe what have been most burdensome for you in the whole ALS/CIs disease trajectory?	How did you experience your own situation? What did you do to manage the disease trajectory? What was important for you in this period of life? How did your network react toward you?
How did you become aware of your relative's ALS/CIs?	How did the cognitive impairments (CIs) of your relative affect your relationship? What was your reaction toward the CIs of your relative? How did handle you handle the challenges related to the CIs?
How would you describe everyday life with your relative with ALS/CIs?	How did the ALS/CI trajectory of your relative affect your family? What was your role during the disease trajectory? What kind of losses did you experience during the period? How did you cope with these losses? Who supported you during the trajectory?
Try to explain at which point in the ALS/CIs trajectory of your relative you realized you needed support?	What kind of help and support did you receive? What did you miss in order for you handle the situation? What was the biggest challenge for you in your everyday life? Who was important for you during this period?

ALS, amyotrophic lateral sclerosis; CI, cognitive impairments.

Table 4. Interview guide for health professionals caring for people with amyotrophic lateral sclerosis and cognitive and/or behavioral impairments (PALS/CIs).

Key questions	Elaborate questions
Try to elaborate on when you felt most challenged in your work with the families of a person with amyotrophic lateral sclerosis and cognitive and/or behavioral impairments (PALS/CIs)?	What was stressful for you in your work with the families? How did you work with the person with CIs? How did you work with the relative to the person with CIs? What would have been supportive for you in this situation? How were you able to take care of yourself? How did you experience your ability to support the carer?
How did you become aware of the cognitive and/or behavioral impairments (CIs) of the person with amyotrophic lateral sclerosis (ALS)?	How would you describe the person with CIs? How did you experience the personality and behavior impairments? How did you experience the language impairments? When did the CIs appear during everyday life? What was your reaction toward the person with CIs? How did the CIs affect your relationship toward the person with ALS? How did you handle the challenges related to the CIs? What was the biggest challenge in regard to CIs? What did you miss in regard to your handling of the situation? What kind of support could have beneficial for you? What was the biggest support for you in this situation? What kind of role did you have in the disease period? When have you felt alone with your challenges?

ALS, amyotrophic lateral sclerosis; CI, cognitive impairments; PALS/CIs, persons with amyotrophic lateral sclerosis and cognitive and/or behavioral impairments.

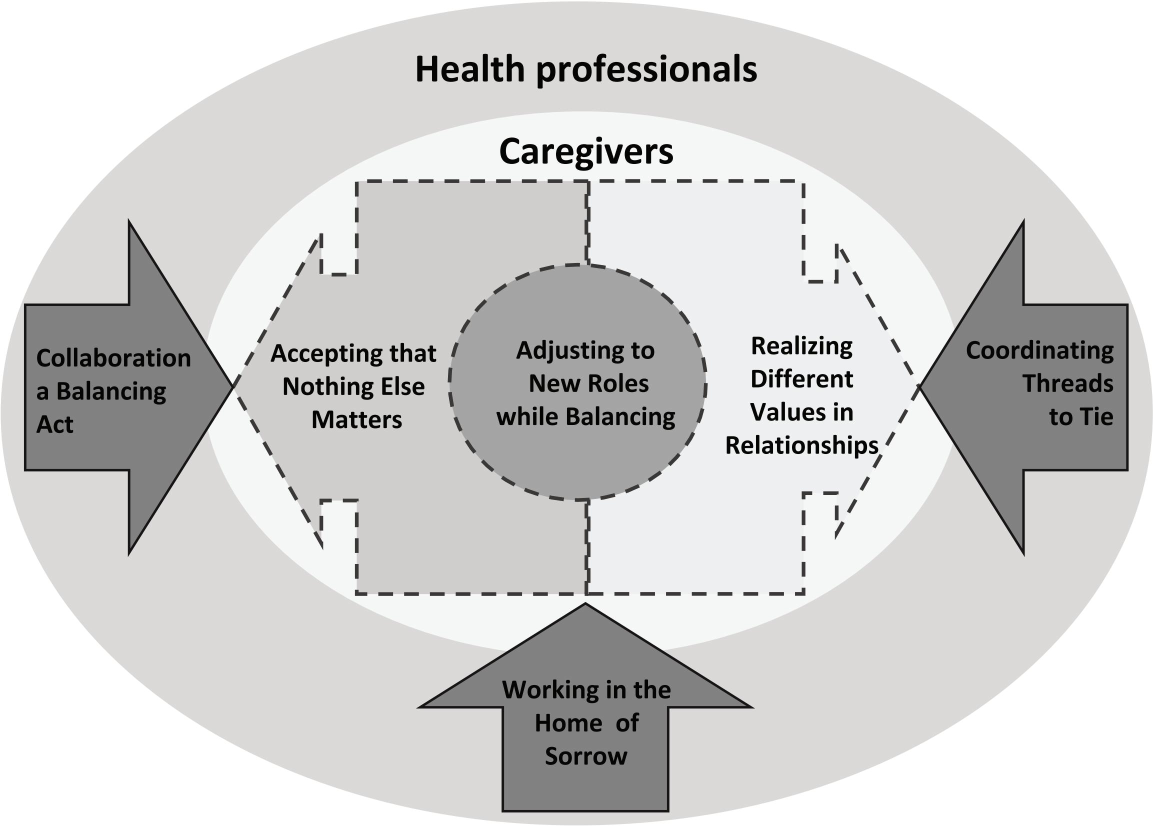

Figure 1. Reflections on everyday challenges of family caregivers and health professionals regarding coping with a person with amyotrophic lateral sclerosis and cognitive and/or behavioral impairments. The findings represent the caregivers illustrated in the inner circle interacting with the health professionals, who act as outsider interference in the everyday lives of the families.

> project manager, that's how it was, it became project ALS . . . It did. (ID 13)

The coordinating role in the 'ALS project' was described as a way to get things 'fixed' and as a project that affected every aspect of their daily lives. For instance, they became responsible for the personal as well as the practical care of the PALS/CIs, providing support, coordinating appointments with different departments and people in the hospital and in the community healthcare services, and taking care of the home, kids, economy, and gardening.

The caregivers emphasized how physical impairments affected their relationship and intimacy. They could kiss, caress, and hug and the PALS/CIs only responded passively.

> Well, I must say, it does something to the relationship when you have to wipe the saliva off his month before you can give him a kiss. No matter whether you like or not, it does matter! . . . I don't think I really realized it [missing physical contact]. Well, I sometimes gave him a kiss on the cheek, but then I sometimes felt that it was a duty at the end. (ID 13)

Some caregivers expressed how they tried to hold on to the relationship by continuing to sleep next to the person, holding hands, hugging when they helped the PALS/CIs' arm around themselves, but at the same time explained how different and difficult it was.

> Actually, right from day 1 when he was diagnosed, it was like that . . ., I thought 'From now on you can't be committed to him as much as you once were a. You may as well start to say goodbye, because he's not staying and whether it lasts 2 years or 5 years or whatever'. I probably didn't expect it to only last [number of] years. It was like that, when he gave me hugs and stuff like that, I did not feel as intensely effected as I once had, and I'm sure, that's because I was starting to let go. (ID 11)

Accepting that nothing else matters

The symptoms from ALS and cognitive impairments overshadowed the caregivers' everyday lives.

> I think it's often difficult not to get upset, so I tried all the time to think: 'You're here now and you're trying to make [the affected relative] happy and also make your other relatives happy . . . and then you can wait and get upset when you get back home'. (ID 14)

They argued how time became crucial and limited because they along the course of the development of the disease quickly realized the irreversibility of the condition and that they were about to lose their relative, which emphasized the need to prioritize the time left together. Doing things that mattered to the PALS/CIs, like visiting old haunts, going on weekend trips, and daily excursions became a prioritization of the feeling that nothings else mattered. They described that knowing that it was 'now or never' made them endure their relatives' behavior and sharpen their focus on living in the present moment while putting their own lives on hold.

> As my mother said, we were living in the present, it was my father who was top priority, and everyone else had to push everything else aside. It wasn't in his spirit the way things turned out, but that's how it was. And we could only do that because we loved him. (ID 9)

Relinquishing spare time and in general repressing your own needs to support and help the PALS/CIs became a way of coping and making the last period together precious.

> Yes, it affected me deeply [when the partner became terminal ill], it did. But I tried as best I could not to show it and then . . . the last 3 weeks [the partner] lived; I almost didn't sleep. (ID 8)

The caregivers expressed that they needed to process feelings like grief, sorrow, and frustration and repress their need to relax, plan their days like before, and sleep properly. The caregivers reflected on their constantly conflicting feelings toward the PALS/CIs. They described how they were divided by their love for their relative, but at the same time, sometimes hoped that it all would come to an end.

> I used to say that he died a little every day, and I broke down a little more every day. So, I was just so . . ., had come to grips with the sorrow before he died, because I'd lost him slowly along the way after all. (ID 11)

They explained how they sometimes wanted to get away from home because their lives were turned upside down and filled with uncertainty, frustration, and sorrow, but as soon as they went to visit friends, they wanted to go back home and not waste the precious time they had left with the relative. The caregivers elucidated how they were torn by conflicting feelings like uncertainty about the future and wanted to get the most out of the little time left rather than being frustrated about not being able to deal with the ALS and the cognitive impairments and take control of their everyday lives. As part of these inconsistent feelings, the caregivers explained that they felt frustrated, scared, angry, and sad when they began to notice the changes in behavior and language and how the lack of knowledge about the disease caused even more conflicts in the families.

> I think it was very scary. And I was like It's really weird' but it was also when I learned something about it, I think it helped me a lot, but I still feel it was very unpleasant. (ID 14)

Changes in behavior and language became a known and recognizable condition that the caregivers explained they had to endure and, for instance, stopped confronting their relative regarding the cognitive impairments related to uninhibited behavior and language. Everyday life was emphasized by the caregivers as full of losses and sorrow because of the constant progression of the disease. Being a caregiver of PALS/CIs was described as a struggling to manage the challenges and new roles without any prior experience of such a complex situation. This caused a sense of loneliness in a situation characterized by chaos.

Realizing different values in relationships

The caregivers described various relationships as a continuum containing both positive and negative aspects, indicating that some relationships became important regarding being able to cope with everyday life with the PALS/CIs and were therefore considered a lifeline in contrast to adverse relations being burdensome, frustrating, but also sometimes necessary. Relationships with family members, friends, and professionals spanned the whole range of these feelings. The caregivers described how some professionals became important to the families because they helped and supported them during the trajectory.

> We have a care coordinator, [name of the coordinator], and SHE IS SIMPLY WORTH HER WEIGHT IN GOLD. I LOVE HER MORE THAN ANYTHING ELSE IN THE WORLD. And she simply stood by us right from day 1 . . . she took care of everything. Everything that had to be applied for and all that stuff. She got the needed signatures, filled out the papers, made sure to send them, and before I could say, Jack Robinson, she was here with it. (ID 8)

The professionals were the nurse, dementia consultant, general practitioner (GP), neurologist, and health coordinator, who kept calling, asking if they needed any help, prioritized extra time to talk with the caregivers despite having little time. The caregivers described how these health professionals became a lifeline, highlighting the major and vital role the health professionals played regarding the caregivers' ability to deal with changed life conditions. But some had negative experiences, and one caregiver said,

> Well, every time we got a new application that had to be submitted, you had to start all over. And you know that as the diseases progresses. You get new social workers who must process the application for speech impairments, for example, or who must take care of the assistive aids or . . . We also had to deal with a new category when he began to need a personal assistant. So, we kept meeting new people . . . And that is also very annoying when you are dealing with a terminal patient. (ID 13)

The caregivers described how the collaboration sometimes became a burden which consisted of hard and sometimes very frustrating work when, for instance, they had to draw the line in relation to how they wanted the professionals to act in their homes, or how they had to fight for the needed help from the community healthcare consultants. The caregivers sometimes felt an increased workload when community or private healthcare services entered the homes because the caregivers had to deal with a large turnover of staff coming into their homes and spend time and energy instructing health professionals, translate the wishes of the PALS/CIs, or rearrange the positions in bed or wheelchair after the health professionals had left the home. They elaborated how this caused insecurity and frustration for both the caregivers and the sick relative because it was crucial that the health professionals knew the PALS/CIs and how the person should be helped and supported despite their lack of ability to express themselves. The caregivers described how persons in their family and friends played an essential part in relation to their experiences of burden and ability to deal with the disease trajectory.

> She [a friend] helped me with many legal things, like applying for disability cards, and, or, you know . . . a disability car, and for compensation for critical illness, and my own critical illness, and . . . And my sister too, and our boys. They've been nothing but amazing, our boys . . . They sometimes kindly said 'Mom, go to a café and we'll take care of Dad!'. (ID 10)

Also, these relationships became a lifeline for the caregivers who often felt lonely despite having people – often health professionals – around them all day. The caregivers also described how other acquaintances surprised them by taking responsibility and action independently and supported them, while others disappeared.

> And that's a thing in all of this, you lose friendships . . . So, we knew, that we would now be separating the sheep from the goats again. So, that's the way it is! And you know what . . . I don't have anything left over for people who can't figure out whether to call or come by or something, I frigging don't! (ID 9)

Results of the professionals

Working in a home of sorrow

The professionals explained how the homes were characterized by hopelessness because of the ALS diagnosis and how the cognitive impairments caused sorrow in the family and made the professionals' work more difficult.

> What does it mean for a human being to be in this psychological state they're in when they have cognitive impairments? A lot of people find it hard to understand that you can't get back to who you used to be. The fact that you can't just get some medication for it or treatment so that you can get your parent or spouse back. When you have changed so radically that you actually become physically and verbally aggressive or without inhibitions. And you lose your understanding of the disease and try to stand up, and also become scarred of everyone

> around you because they don't help you. It's really difficult. These are the biggest challenges I face. (ID 6)

The professionals experienced how caregivers were distressed, anxious, and highly burdened due to their changed life condition as a result of ALS/CIs' condition. The health professionals described how they tried to support family members in coping with this transformation of the PALS/CIs, but at the same time, were confronted with a trajectory of dilemmas and diverse feelings between the couple like hope and hopelessness, in which they constantly had to navigate without erasing hope or causing further sorrow or grief.

> Well, I feel most challenged where I can see that the PALS/CIs is simply getting worse and worse and the caregivers in the family won't acknowledge it. You see the grief in the person's eyes due to being so fatally ill, and the spouse doesn't understand that the person has become this ill or refuses to accept it . . . I think that is so terrible to witness. (ID 15)

The professionals explained that they exposed their ears, bodies, and souls to a lot of tears, frustrations, and helplessness from the PALS/CIs and caregivers even though they were often pressured for time or lacked knowledge on how to support the families. Several of the professionals found it frustrating not to be able to ease the caregivers' sorrows, which caused a self-doubt among them. Some professionals explained how they were able to distance oneself from the emotional commitment.

> We get so affected by it [ALS/CIs] because it is so hopeless, and the hopelessness . . . it becomes such a heavy burden to carry. We must, although it may sound a little cynical, we have to take two steps back sometimes to see it all in a larger perspective. We cannot involve ourselves as if we were closely related. We need to maintain a professional approach. (ID 7)

Others explained how they often ended up engaging themselves a little extra because they became so affected by the severity of the situation that it made it hard for them to leave their jobs behind them when they were not working. The professionals nonetheless expressed how relieved they were that they were able leave the home compared to the caregivers who were always there.

Collaboration a balancing act

The professionals described the collaboration with the families as a balancing act due to constant potential dilemmas and conflicts lurking in the family and in the collaboration.

> There was this woman with dementia-like behavior, and it was very difficult because, well, she had a friend who had the idea that she could just set up a private helper arrangement, but she couldn't, because she couldn't be both the boss and take part in the other arrangements where she could delegate that role, that wouldn't be relevant because the friend couldn't undertake the task to the degree it was necessary . . . and that was actually a great frustration for particularly the relatives. I really don't think the woman [PALS/CIs] had the ability to understand it anymore, but there was a great feeling of dissatisfaction, and the thing about us is that we simply didn't speak the same language, and the relative, she had some expectations about how she just had to put the employees in the system, and then it went completely, well hotchpotch. (ID 7)

They explained how they had to be careful to use the right words and how a wrong choice in actions or attitudes could create an instant negative and tense atmosphere. Disagreements were often related to a lack of balancing expectations between what the professionals could offer and how this could be matched with the family's needs. Working in a private home while adhering to the health and safety at work act and the relevant legislation also sometimes caused an unequal and difficult collaboration with the families.

> . . . for us it is a workplace that must be able to function as such, and for them it is a home and must be able to function as such as well, so I sometimes have to compromise, and sometimes they have to be able to compromise. (ID 15)

The collaboration with the families was also affected and dependent on the personal and professional competences and relations to the family, where the professionals had to navigate while delivering a professional service. The professionals talked about revealing or not revealing their personalities and private lives for the families while still struggling with staying professional in relation to the families.

> That is something we talk a lot about. When do we open ourselves up a little bit and take something

> more personal into it [the relationship], and when do we keep ourselves completely closed up in order to be able to distance ourselves from some of the difficult things that also are there. But it is also important that we sometimes open up a little and allow something personal into it [the relationship], because otherwise the person you face can't relate to you if you are too professional, then you have to open up for some things and bring something personal into them. You just must remember to close up again when you do not need to be open anymore. (ID 6)

The professionals had to reduce frustrations and avert dangerous situations by supporting the caregivers in handling the inappropriate behavior and language of the PALS/CIs.

> And it's from experience, the more experience you have, the easier you get into the family, the easier you can 'talk these symptoms down' and talk about what can be done about the symptoms instead of making them worse. Then it becomes something we can work on, and in that way, we can come up with solutions instead of just seeing barriers all the time. And if his language is inappropriate and becomes very offensive 'well, how do you balance it? And how do you respond to these actions? And if you stay, if he's aggressive and lashes out at someone, how do you position yourself by the bed, so he doesn't hit you?' But he can still have his outburst without it hurting anyone. (ID 6)

Various coordinating threads to tie

The professionals referred to a union of different authorities, departments, and professionals that supported the PALS/CIs. They mentioned internal collaborators in the community healthcare settings, like nurses, social and healthcare helpers/assistants, visitation and aid officers, and dementia consultants. Moreover, they mentioned external collaborators like neurological departments, pulmonary departments, GPs, the palliative units, and the private healthcare services. Working harmoniously with the other disciplines was explained by the professionals as both important and useful in regard to fulfilling the complex care needs of the PALS/CIs.

> I have experienced, when you have these collaborative meetings where both patient and caregivers are present, and you represent a professional skill, that it can calm down these families, because then they know that everyone has heard the same thing. And if there is a relative who is so frustrated and would like to split the professionals, then you can say, 'Well, at the meeting where we were all present, so and so and so . . .' (ID 3)

When collaboration succeeded, the professionals described how they supported the families despite different areas of focus, places of location, responsibilities, and lack of acquaintances. For instance, authorities and providers working together to formulate schemes that will secure the ability of the PALS/CI to stay as independent as possible or professionals reaching out to spar with collaborators.

> That's where the collaboration works really well with caregivers and with the home care, that's a good support, indeed! Then you experience that, yes, that there is a unified whole and we share common ground, and we can see that we can do this together. We can deliver good support and care for the family, despite all things. (ID 7)

Respect for each other professional expertise, statements, and heterogeneity was also addressed as an important aspect in the interdisciplinary collaboration. The many different professionals helping the families initiated a need to coordinate actions for the professionals to co-operate in and across sectors instead of working in parallel or counteracting each other, which unfortunately was described to be the case in some homes.

> Yes, there were several things where I had to make ends meet here and wondered, 'is it me or what is happening?' But the external collaborator simply felt it was a competition regarding who should get to know this PALS/CIs best. Where I finally said to a colleague, 'Well, I do! Because I've been there the most'. Because I'd been there for 4 months before she came, and we have figured it all out together. So, that was MY biggest challenge down there, the respiratory team. (ID 2)

Collaboration became challenging when responsibilities were unclear or when lack of knowledge about or respect for each other's job area was unclear, which affected their job satisfaction negatively. The professionals stated that a coordinator was crucial for both collaborators and the families when trying to cope, plan, and make everyday life as manageable as possible because such person had a close collaboration with and knowledge of how the PALS/CIs and the families functioned.

Discussion

The everyday challenges of the caregivers and health professionals in our study were compounded by the spiraling of losses because the diseases and adaptation were ongoing processes. The challenges initiated inappropriate behavior and constant need for adjustments in roles and support and consequently with a high workload in everyday life. The caregivers in this study were without any previous experience regarding supporting their relative with ALS/CIs. De Wit *et al.*[34] found that dealing with behavioral changes without an understanding of symptoms and causes may be most challenging for caregivers because behavioral changes are the strongest predictor of psychological distress in caregivers of people with ALS and are also associated with lower perceived control over caregiving. Research has shown that caregivers of people with ALS have unmet needs regarding support, help, and understanding of their situations.[34] This relates well with our findings that showed how the caregivers struggled with adjusting to new roles and realizing the meaning of relations, which could be explained by the lack of comprehensibility of the situation among caregivers.[25] In order to cope with the constantly reoccurring challenges during everyday life, the caregivers accepted that nothing else mattered, which seemed to add to their comprehension of coherence and provided meaningfulness and a way to manage and comprehend their complex situation.[25] Burke *et al.*[35] argue that reconfiguring existing roles within the family could reduce frustration or irritation if cognitive impairments are present. This was also highlighted in a study on the burden of caregivers, where adjustments of roles and responsibilities, interaction style, and activities of daily living may be beneficial and reduce the experience of burden and increase the quality of life for the caregivers.[36] A study on the experiences of caregivers of people with ALS regarding care and support concluded that caregivers often stop seeking or accepting support.[37] This was also described by the caregivers in this study, but they nevertheless realized the importance of relations with health professionals and their other relatives. Online services could accommodate the need for support among burdened participants, who had difficulties in leaving their relative at home alone due to the extent of caregiving responsibilities. A previous report highlight online services, like telehealth as a way to support caregivers of people with ALS.[38] The use of social media have shown to increase the connection between caregivers of people with ALS, and the attendance and socialization.[39] Caregivers expressed trouble with managing cognitive and/or behavioral impairments because they had no knowledge of the disease but nonetheless emphasized how they acquired the needed skills along the way. These findings are in line with a study where caregivers described a reduction in unmet needs as they became more confident and gained more experience as time went on.[40] This emphasized how the caregivers' experiences improved along the ALS/CIs' trajectory and moreover their comprehensibility that made them capable of managing different tasks and thereby becoming more confident and finding a meaning in the situation.[25] De Witt *et al.*[8] concluded in a study on care demands among caregivers of people with ALS and progressive muscular atrophy that caregivers might feel that they were fully capable of fulfilling care tasks while at the same time experiencing that they have no control over their lives in general. Being emotionally involved as a carer and a novice within such a complex situation has a major negatively impact on the well-being of caregivers of PALS/CIs.[8,35] Due to the complexity and lack of time for these caregivers, telehealth could meet their need for information and support, and Geronimo *et al.*[41] found that caregivers of people with ALS and professionals generally view telehealth as favorable. Targeting educational programs with specific variables to manage behavioral problem is advocated for caregivers of people with dementia.[42,43] Blended psychosocial support for caregivers of people with ALS also shows a significant effect on self-efficacy related to control over thoughts.[44]

In relation to the health professionals, it was emphasized how working in a home of a PALS/CIs was filled with sorrow and mourning due to constant loss and how it affected their work–life balance because of the hopelessness and the effect on the caregivers. Oxford neurologists, Brian Matthews (1920–2001) likewise recognized a balancing act between the needs of the ALS patient, the carer, and the physician, and he stated that the suitability of specialized neurology practice was about being able to continue to support a case of ALS.[45] According to the SOC theory, it could be argued that the professionals experienced the sorrow in the family as unmanageable, making it difficult for them to cope with it.[25] The professionals in our study explained how they struggled with the mixed emotions, and for some, it was difficult to leave their jobs behind when they went home. These findings are underlined by the results from a study on professionals working in the palliative field that concluded that professionals are at risk of burnout, moral distress, fatigue, and affected clinical decision-making due to unexamined emotions and unprocessed grief.[46] Some of the professionals in our study described different ways of addressing the families' need for support, but also highlighted this as a complex and difficult challenge. The professional's explained that their interactions in the homes appeared to matter regarding how the families experienced everyday challenges and their ability to cope with them. These findings are supported by a review on health services that suggests staff training and service delivery models as strategies to remedy problems because skill deficits in professionals can reduce the well-being of the caregivers.[47] Oliver and Turner[12] likewise found education in the palliative approach useful for professionals caring for a person with ALS and supporting the caregivers. They also stated how a strong multidisciplinary team is important because of each health professional's expertise in the team contributing to a robust internal support to serve the person with ALS and the families more effectively.[12] This is in line with the findings in this study stressing the challenges the health professionals were burdened with in their daily practice – trying to collaborate and coordinate in a home filled with grief and sorrow – a task that needs specially acquired skills and support.

Methodological considerations

The study included 16 participants, which is a relatively small sample size, but the participants had special expertise and knowledge within the research field and the interviews were conducted in depth, which helped broadening the perspectives on challenges in everyday life with a PALS/CIs.[48] However, due to convenience sampling of the caregivers, results may not necessarily reflect the distinctions in perspectives between caregivers and this could therefore be a limitation.[31] The caregivers were included based on their personal description of cognitive and/or behavioral impairments of the deceased PALS/CIs, and no disease-specific cognitive screening measure was applied. Having used a specific cognitive and/or behavioral screening measure may have ensured caregivers of PALS/CIs with a higher degree and presence of the cognitive and/or behavioral impairments seen in the ALS spectrum.[5] However, defining and diagnosing ALS/FTD is widely discussed and international guidelines from Strong *et al.*[49] found the term *frontotemporal spectrum disorder* (ALS-FTSD) as most appropriate to characterize the breath and severity of the frontotemporal dysfunction that can be encountered in relation to ALS. Neuropsychological testing is time-consuming, and a variety of simple tools have been developed to assess cognitive and/or behavioral impairments, and some of these are administered by carer.[50] A review on screening instruments on cognition and behavior in ALS patients in clinical practice showed that not all patients were able to complete a full neuropsychological battery due to motor and/or language impairments,[51] and thus screening in regard to inclusion would be difficult and uncertain. The participants were included based on their specific experience with cognitive and behavioral impairments in the person with ALS who not necessarily had a confirmed clinical assessment or diagnose. Since diagnosing and defining cognitive and/or behavioral impairments in relation to ALS is complex, our results should be seen in this light. In addition, our participants were specifically asked to present challenges related to cognitive and/or behavioral impairments which may reflect a larger amount of these challenges.

Using focus group interviews for the professionals facilitated the rethinking and discovery of unexpected perspectives regarding the health professionals' own challenges in relation to supporting the families.[24] The professionals had different experiences of caring for PALS/CIs and different interactions with the families, which caused one especially experienced participant in the focus group interview to be more outspoken, which could have reduced the value of the group discussion and the credibility of the results.[52] Nevertheless, the interviewer made sure that everyone got the possibility to express an opinion throughout the interview. Having to replace the second planned focus group interviews with the health professionals with individual interviews gave an opportunity to get more in-depth knowledge from participants. Furthermore, only one professional from a private healthcare service was included, and this could be another limitation because these health professionals have different qualifications due not having a health-professional education but being personally trained and often working alone for many hours. However, the participants in our study had many years of experiences with different PALS/CIs and could therefore elaborate on everyday challenges based on a wide perspective.

The results mirror everyday reflections of caregivers and health professionals in a Danish context, which may affect the external credibility owing to a tax-financed healthcare system in Denmark; however, similar results are expected to be found among caregivers and health professionals in other Western countries delivering healthcare services to PALS/CIs. Appling the SOC theory and the ID methodology during all processes in the study helped explore the SOC among participants coping with everyday challenges related to the PALS/CIs in a systematic and stringent way that ensured transparency and internal credibility.[24,25]

Conclusion

Everyday challenges of caregivers and health professionals of PALS/CIs were found to be affected by the complexity of constant progression of the diseases and the various needs to fulfill. Being emotional involved as a carer and having to struggle with cognitive and/or behavioral impairments without prior experience was found to be the strongest predictor for psychosocial destress among caregivers. The caregivers are heavily burdened by the constant need to adjust to new roles while dealing with unclear responsibilities. Supportive educational interventions, telehealth, and social medias are beneficial among caregivers and health professionals who look after people with ALS because they increase self-efficacy, socialization, accessibility, and reduce burden. The health professionals struggled with mixed emotions and their ability to support and engage adequately with the families without taken home the sorrow from the families. Professional's interaction with families of PALS/CIs had a major influence on the family's ability to cope with everyday challenges and remedy problems. Multidisciplinary approaches and education in palliative care would be useful in creating a robust internal support for these families and at the same time support the health professionals' ability to create a positive work-life balance. This study adds to an understanding of the challenges and needs of caregivers and professionals and the relationship between these. It also highlights elements of support and knowledge that might be important and necessary to incorporate into the development of a targeted online intervention for caregivers of PALS/CIs and professionals who help them. Relevant elements to include in palliative rehabilitation for carers and health professionals could be psychoeducation, peer-support, and knowledge on change in roles, loss of control, intimacy, preparation for the future, coping with loss and sorrow, and on how to take care of oneself. Future research should address the need to develop and test the outcomes of a targeted online palliative rehabilitation intervention for caregivers and health professionals in order to support their coping with challenges in everyday life with a PALS/CIs.

Relevance for clinical practice

- Caregivers experience imbalance in their relationship due to increasing loss of their partner and challenging needs to fulfill without any previous experience within this role.
- Health professionals struggle to support families while balancing their professional relation toward these and need specialized support and knowledge to fulfill this challenge.
- Caregivers and health professionals are challenged by the trajectory of ALS and cognitive and/or behavioral impairments and are in need of support and knowledge from other health professionals recommended through a targeted online palliative rehabilitation intervention.
- Elements to include in such an intervention could be palliative rehabilitation, psychoeducation, peer-support, and specialist knowledge on changes in roles, loss of control, intimacy, preparation for the future, coping with loss and sorrow, and taking care of oneself.
- Online interventions could increase the accessibility for caregivers and health professionals and thereby meet the gap between the participants' need for support and their lack of time.

What does this paper contribute to the wider global clinical community?

- Knowledge on the need for both a palliative and rehabilitation intervention to help caregivers of people with ALS and cognitive and/or behavioral impairments confront and cope with their challenges.
- Knowledge on important elements to address when developing a targeted online palliative rehabilitation intervention for caregivers and health professionals.
- Knowledge on the importance of strong personal relationships for caregivers and

professionals when being a part of the challenging care and support for a person with ALS and cognitive and/or behavioral impairments.

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Author contributions

Lene Klem Olesen: Conceptualization; Data curation; Formal analysis; Funding acquisition; Investigation; Methodology; Project administration; Resources; Software; Validation; Visualization; Writing – original draft; Writing – review & editing.

Karen la Cour: Conceptualization; Data curation; Formal analysis; Methodology; Project administration; Resources; Supervision; Writing – review & editing.

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Data availability

The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

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Article II

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RESEARCH

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A cross-sectional evaluation of acceptability of an online palliative rehabilitation program for family caregivers of people with amyotrophic lateral sclerosis and cognitive and behavioral impairments

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Abstract

Background: Amyotrophic lateral sclerosis (ALS) is a progressive fatal neurodegenerative disease. Around half of the population with ALS develop cognitive and/or behavioral impairment. Behavioral changes in persons with ALS are perceived as the strongest predictor of psychosocial distress among family caregivers. Interventions aiming to support family caregivers are emphasized as important in relation to reducing psychological distress among family caregivers. Successful healthcare interventions depend on the participants' acceptance of the intervention. Therefore, this study aims to evaluate the acceptability of a new online palliative rehabilitation blended learning program (EMBRACE) for family caregivers of people with ALS and cognitive and/or behavioral impairments.

Methods: A qualitative cross-sectional design using the theoretical framework of acceptability to evaluate acceptance of the intervention based on data collected through individual in-depth interviews and participant observations. Individual interviews were conducted in 10 participants post-intervention and participant observations were recorded during virtual group meetings among 12 participants. A deductive retrospective analysis was used to code both datasets in relation to the seven constructs of the theoretical framework of acceptability: affective attitude, burden, ethicality, intervention coherence, opportunity costs, perceived effectiveness, and self-efficacy. The theory of sense of coherence by Antonovsky informed the development and design of the intervention and interviews. The study adheres to the COREQ (consolidated *criteria* for reporting qualitative research) guidelines.

Results: Within the seven constructs we found that affective attitude addressed the meaning and importance of peer support and focused on the participants' needs and challenges. Burden referred to technology challenges, time pressure, and frequent interruptions during meetings. Ethicality concerned transparency about personal experiences and the exposure of the affected relative. Intervention coherence referred to a shared destiny among participants when they shared stories. Opportunity costs primary concerned work-related costs. Perceived effectiveness referred to the usefulness and relevance of peer support and the meetings that brought up new ideas on how to approach

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current and future challenges. Self-efficacy involved the motivation to learn more about ALS and ways to cope that were accommodated by the convenient online format.

Conclusions: The findings showed that the participants favored peer support and the videos that reduced feelings of loneliness and frustration but also confronted them and provided knowledge on future challenges. Further research should explore the benefits of the program and the meaning of online peer support among caregivers of people with ALS and cognitive and/or behavioral impairments.

Trial registration: Retrospectively registered on November 20th, 2020. ID no. NCT04638608.

Keywords: Family caregiver; ALS; cognitive impairments, Behavioral changes, Support, Palliative rehabilitation, Acceptability, Intervention, E-health, Feasibility

Background

Amyotrophic lateral sclerosis (ALS) is a devastating progressive neurodegenerative disease that has prominent non-motor manifestations like cognitive and behavioral impairments [1]. The discussion of the ALS and frontotemporal dementia (FTD) continuum has been retold and are now described as two distinct entities [2]. Cognitive and behavioral impairments in ALS are associated with more rapid progression and poorer prognosis and risk of death that is 2 to 2.53 times higher than in unimpaired controls [3]. Cognitive, emotional, and psychological impairments in ALS may cause alterations in certain cognitive functions such as executive functions, verbal fluency, language, and verbal memory [4]. Moreover, impairments and abnormal and inappropriate behavior, like apathy, loss of manners, aggression, and being tactless, are not uncommon in persons with ALS with the cognitive/behavioral variant of FTD [5, 6].

Research shows that behavioral changes are the strongest predictor for psychosocial distress in family caregivers (hereafter caregivers) of people with ALS [7, 8]. Not only do the cognitive and behavioral impairments increase the burden and the anxiety on caregivers, but they also affect their well-being [9–11].

Caregivers of people with ALS and FTD provide care with a tremendous resilience, compassion, and devotion [12], which is why caregivers need individual time-targeted psychosocial support, containing education and management of challenging symptoms [13]. However, the burdened caregivers frequently refrain from seeking or accepting support due to the difficulty of balancing their personal time with their caregiving responsibilities [14, 15]. There is currently no cure for ALS or the cognitive/behavioral impairments, and two reviews on palliative care in motor neuron diseases (like ALS) therefore advocate for structured support of caregivers in the form of counseling, support groups, and a crisis management system (before and after death of their relative) [16, 17]. Caregivers of people with ALS are likely to experience greater psychological well-being and quality of life from combined psychoeducational support and mindfulness [18]. Similarly, active planning within a multidisciplinary care setting provides an avenue for caregivers of people with ALS and FTD to proactively cope with cognitive/behavioral impairments that will induce improved care and reduce the risk of caregiver burnout [1]. A rehabilitation program for people with ALS and their caregivers has been shown to have a positive effect on the participants' incentive to understand the disease and benefit from peer support [19]. However, due to the heavy burden and demands caregiving of people with ALS and FTD places on the caregivers [13], it is important to take the caregivers' time into consideration by using videoconferencing [20]. Research suggests that blended care in the form of combined face-to-face and online healthcare can help bridge the gap between the need for support, information, and lack of time among caregivers of people with ALS [21]. However, some challenges remain because successful implementation of healthcare interventions depends on the recipients' acceptance of the intervention [22–24]. For recipients to adhere to the intervention and benefit from the improved clinical outcomes [22, 25], it is necessary to develop intervention programs that are accepted by caregivers of people with ALS and cognitive and/or behavioral impairments (PALS/Cis). Hence, we developed the EMBRACE intervention, a 4-month online program aimed at supporting the ability of caregivers of PALS/Cis to handle everyday challenges related to the care of PALS/Cis (Fig. 1). The aim of the present cross-sectional study was to evaluate the acceptance of a new online palliative rehabilitation program (EMBRACE), a blended learning program developed for caregivers of PALS/Cis.

Methods

Theoretical framework

The framework on developing and evaluating complex interventions in healthcare from the UK Medical Research Council, the theoretical framework of acceptability (TFA) and the theory of sense of coherence by Antonovsky were used to evaluate the acceptance of EMBRACE [26–29]. According to the updated

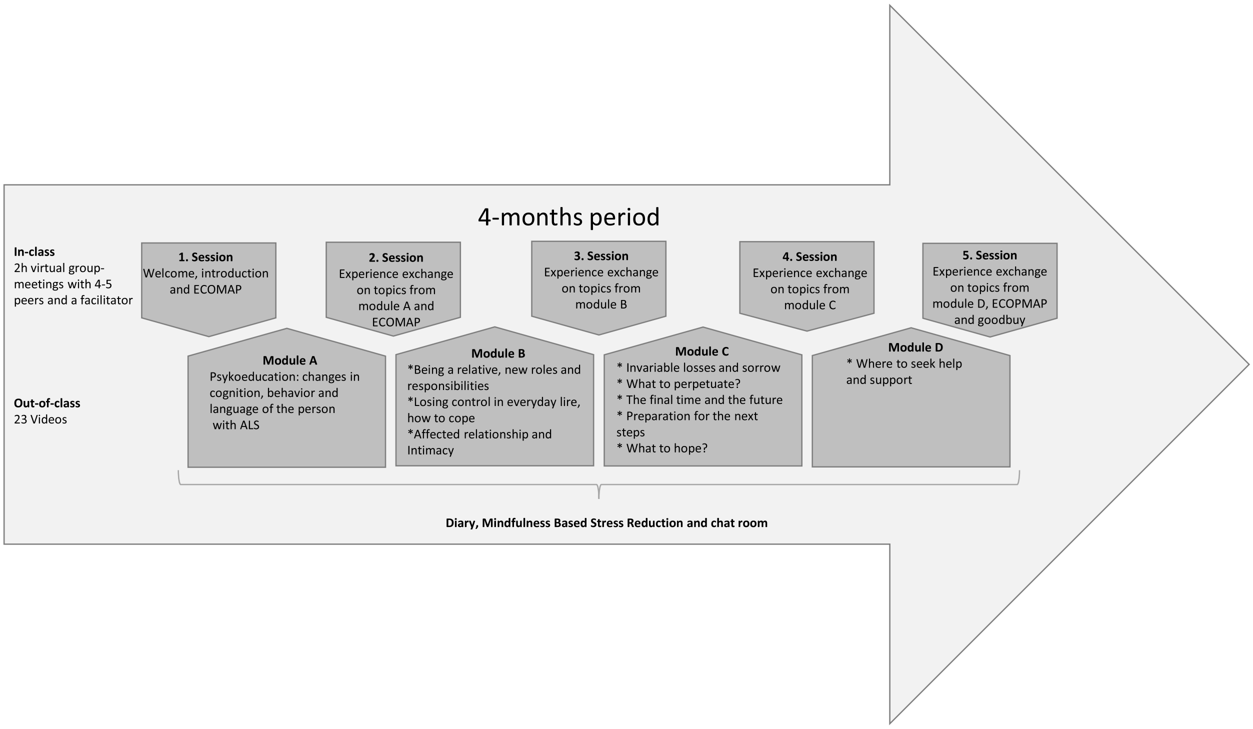

Fig. 1 The EMBRACE intervention. A 4-month online palliative rehabilitation blended learning program for family caregivers of people with ALS and cognitive and/or behavioral impairments. The intervention was facilitated by an experienced healthcare professional from the Danish National Rehabilitation Center for Neuromuscular Diseases, who is a registered nurse and certified family therapist with 15 years of experience working with persons with ALS and their families

guidelines from the Medical Research Council, acceptability is important to address during the initial stage of the intervention development [30]. The TFA framework consists of seven constructs: affective attitude, burden, ethicality, intervention coherence, opportunity costs, perceived effectiveness, and self-efficacy that were used to evaluate acceptability in a concurrent and retrospective view [27]. The TFA informed the development of an observation guide, the analysis, and writing up the findings based on the seven constructs [27]. The theory of sense of coherence guided the development and design of the intervention and interviews [29]. The theory focuses on three core components, comprehensibility, manageability, and meaningfulness, that should be present in order to cope and experience life as coherent, thereby reducing stress [29].

Design and setting

A qualitative cross-sectional design using the theoretical framework of acceptability to evaluate acceptance of the intervention based on data collected through individual in-depth interviews and participant observations [27].

The study was carried out online through the platform Simplero, and group meetings were run with Microsoft Teams. The study was embedded at the National Rehabilitation Center for Neuromuscular Diseases (RCFM) in Denmark [31]. RCFM is a national, highly specialized private outpatient hospital financed by the government, with rehabilitation services free of charge for its patients [31, 32]. RCFM offers highly specialized advice and counseling to persons with neuromuscular diseases, their families, health professionals, professional caregivers, and rehabilitation specialists [31]. Public neurological hospital departments refer about 95–97% of people with ALS to RCFM [31]. The professionals at RCFM are organized in multidisciplinary teams consisting of occupational and physiotherapists, nurses, doctors, psychologists, and social workers [31]. To provide rehabilitation on the patients' terms and to get as much insight into the patients' everyday lives as possible, most of palliative rehabilitation by the professionals at RCFM is performed in the homes of the persons with ALS [31].

Intervention

EMBRACE had a blended learning format, combining both videos and virtual group meetings. The content rests on evidence- and experience-based topics identified in a qualitative study on challenges and needs among caregivers of deceased PALS/Cis [33]. We developed and recorded 23 videos based on topics associated with caregivers' challenges and needs. The participants received a diary before starting the invention and were encouraged to take notes and write down their thoughts during the intervention. The participants were asked to make ecomaps three times during intervention as a means to explore potential supportive relations that could be beneficial during the disease-trajectory and after the death of the PALS/Cis. They were also offered customized Mindfulness Based Stress Reduction videos. The diary, ecomaps, and the mindfulness videos were not used as data. In addition to the empirical evidence and experience base, the theoretical lens of sense of coherence strengthened and targeted the content in EMBRACE to meet the caregivers' need for comprehensibility, manageability, and meaningfulness [29]. EMBRACE consisted of three groups, each of which included 4–5 participants, facilitated by the third author. EMBRACE was developed and carried out by the first and third authors, who had extensive knowledge of the field under research due to working as healthcare professionals at RCFM. This team received regular professional group supervision during the delivery of the intervention.

Characteristics of participants and sampling

Participants were sampled based on the following inclusion criteria: [1] caregivers (partners and spouses) living with a person diagnosed with ALS referred to RCFM who had received an initial visit from healthcare professionals from RCFM, [2] caregivers who were able to speak and understand Danish, and [3] caregivers of persons with ALS with a cut-off score ≥ 22 on the Amyotrophic Lateral Sclerosis-Frontotemporal Dementia-Questionnaire (ALS-FTD-Q), a validated questionnaire containing 25 items, the total score ranging from 0 to 100, with higher scores indicating more behavioral changes [34]. A cut-off score ≥ 22 on the ALS-FTD-Q indicated mild behavioral change of the person with ALS [34]. Caregivers were encouraged to invite a relative to accompany them throughout the intervention. The companion could not be an affected relative. Two caregivers chose to invite an adult relative to accompany them.

A two-step sampling process was performed for the intervention. First, healthcare professionals from RCFM helped identify persons with ALS referred to RCFM up to September 8, 2020, who met the first and second inclusion criteria. Next, invitations containing information about the intervention program and the research project were sent to persons with ALS and caregivers, 208 in total. Thirty-one caregivers contacted the first or third author, wishing to participate. The interested caregivers participated in screenings by phone where they scored their affected relative using the ALS-FTD-Q [34]. A total of 15 participants were included in the intervention (Fig. 2). Participant observations during the interventions in 16 virtual group meetings were obtained from 12 of the 13 participants who started the intervention (11 partners and 1 adult child of a parent with ALS) (Table 1). All 15 included participants were invited to participate in interviews about their expectations for EMBRACE prior to the intervention. Eleven of the 12 participants (including non-completers) were invited to participate in post-interviews (Fig. 3). The person who was not invited had just lost a relative who had died of ALS. For this study, we draw on the post-interviews and participant observations. The inclusion for post-interviews was ongoing from September 14, 2020, to February 25, 2021. Ten caregivers out of 11 participated in post-interviews. One did not respond to the invitation.

Data generated

Data were generated by using data triangulation with individual in-depth interviews post-intervention and participant observations during the group meetings. We chose to include participant observations in this dataset to obtain an objective point of view on the intervention [35]. Retrospective participant observations in 16 recorded virtual group meetings were carried out individually by the first, third, fourth (an external health anthropologist), and fifth authors. Each of the 16 meetings lasted for around 2 h and were run with Microsoft Teams. Participant observations were carried out according to a predefined participant observation guide (Table 2).

This evaluation focused on the execution process and retrospective experiences of acceptability to accentuate the participants' perceptions and experience of EMBRACE. Therefore, the pre-intervention interviews will be reported elsewhere. The post-interviews are reviewed in the present study. Interviews were carried out by the first author with the seven participants completing the intervention and three non-completers. Interviews with non-completers were carried out to learn about their reasons for withdrawing and potential barriers regarding acceptability. Nine interviews were generated online using Microsoft Teams, and one interview was conducted in-person at the caregiver's workplace. Interviews were carried out by the first author and were digitally recorded. Interviews lasted between 58 min and 1 h 41 min.

Observation guide

A participant observation guide composed of seven constructs from the TFA [27] was used (Table 2).

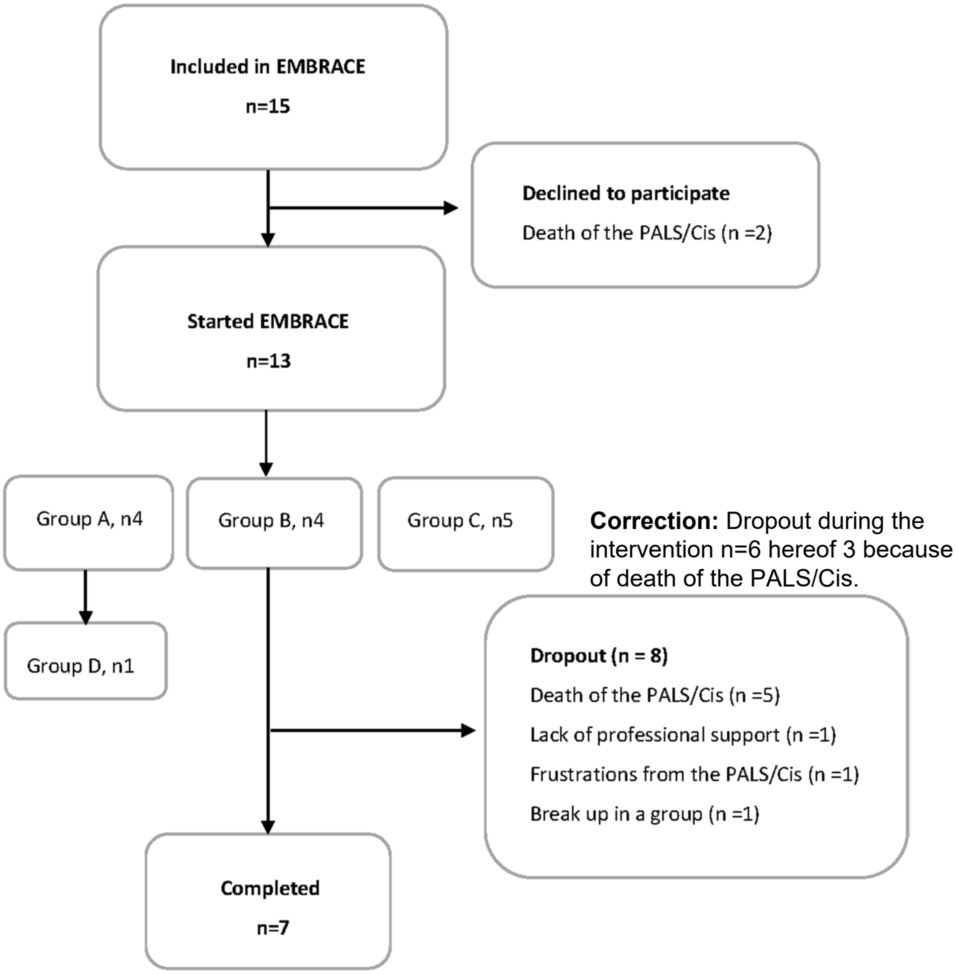

Fig. 2 Participant flow diagram. Overview of allocation and numbers of participants in each group, including numbers of and reasons for participants dropping out

Interview guide

A semi-structured interview guide composed of open-ended questions was used. The questions focused on the participants' experiences, attitudes, feelings, preferences, and boundaries regarding the intervention and recommendations for improvements [See Additional file 1].

Data analysis

All data were organized and analyzed retrospectively and deductively according to the seven constructs of the TFA [27]. Participant observations were carried out individually by the first, third, fourth, and fifth authors. Each person watched all 16 recorded videos from the group meetings and filled out the predefined participant observation guide for each video. Next, the whole group systematically went through each construct for each video, allowing each person to present their organization of data within the construct. The group then discussed what had been said and whether it was the correct organization according to each construct. Data extraction and condensation related to each of the seven constructs across the 16 meetings was subsequently undertaken by the first author, and the extract was discussed with the entire group (Fig. 4).

Interviews were transcribed verbatim, then read and individually deductively coded according to the seven constructs in NVIVO^{12} by the first and fifth authors [27]. The selected codes and phases were then discussed in the whole research group in terms of which codes were most fitting according to the TFA constructs [27].

Results

Feasibility results on acceptance of the EMBRACE intervention are structured by the TFA constructs and presented below [27].

Table 1 Demographic data on the participants based on the dataset from the participant observations and interviews

Participants		**($n = 12$)**
Gender	Male	3
	Female	9
Age	18-25	1
	39-50	3
	51-55	4
	56-67	4
Relation	Married/partner	11
	Adult child of a PALS/Cis[1]	1
Occupational status	Working	7
	Early retirement/retired	4
	Studying	1
Years of ALS-trajectory	0-2	4
	2-4	2
	4-8	4
	8-12	1
	12-14	1
ALS-FTD-Q score	22-30	2
	31-35	5
	36-40	1
	41-46	3
	47-55	1
Urban	$\geq$ 80.000	1
Rural	$\leq$ 80.000	11

[1] Person with amyotrophic lateral sclerosis and cognitive and/or behavioral impairments

Affective attitude

Affective attitude concerned the participants' feelings toward EMBRACE mainly centering around the group meetings and the impact of the attitude of the other group members. Observation showed that the participants expressed positive thoughts toward the EMBRACE intervention and were grateful for having been given the opportunity to participate. In general, they were positive about the intervention and described the development of relationships with group peers throughout the meetings and the importance of peer support as meaningful.

> *"I've liked the closed forum where everything has been allowed. It's been pretty liberating to be able to talk about what you are struggling with." (ID 2G)*

> *"Yes, I also found support in listening to each other's stories, and I thought 'Ah, I'm not the only one who feels like that. In a way, it's a good thing. Not that you want it for other people, but it's nice to know that you're not alone." (ID 2C).*

Participant observations showed that the participants talked about looking forward to the meetings, which they said were a welcoming break from their everyday lives, which were otherwise filled with various activities, work, care, and support for the PALS/Cis. Generally, the participants found that the meetings were

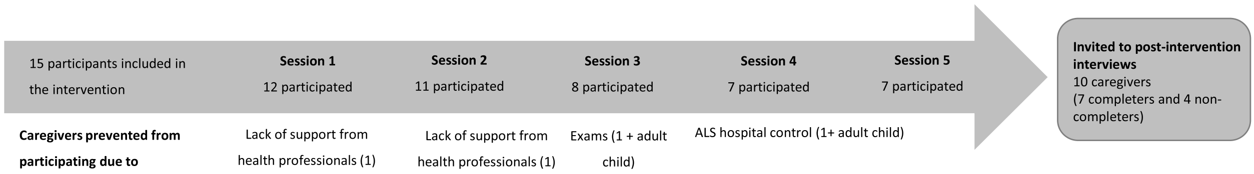

Fig. 3 Overview of participants in group session, participants invited to interviews post-intervention, and reasons for non-participation

Table 2 Participant observation guide for caregivers of PALS/Cis

The TFA constructs	**Elaborative participant observation questions**
Affective attitude	How do the participants show and express their feelings about the intervention?
Burden	How do the participants show and express their perceived amount of effort required to participate?
Ethicality	How do the participants show and express the intervention's fit with their individuals value system?
Intervention cohesion	How do the participants show and express their understanding of the intervention and how it works?
Opportunity costs	How do the participants show and express their opportunity costs, like benefits, values, or profits that must be given up to engage in the intervention?
Perceived effectiveness	How do the participants show and express their experience of perceived effectiveness/or the opposite with the intervention?
Self-efficacy	How do the participants show and express their confidence that they can perform the behavior(s) required to participate in the intervention?

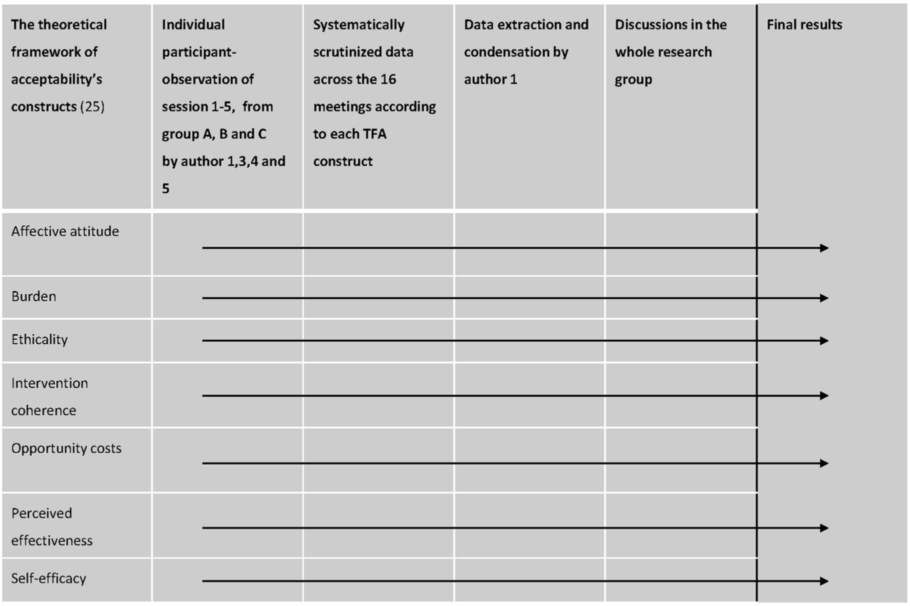

Fig. 4 Analysis process of participant observations. The seven constructs of the theoretical framework of acceptability were used: affective attitude, burden, ethicality, intervention coherence, opportunity costs, perceived-effectiveness, self-efficacy [27]

characterized by a special atmosphere and that there was a mutual understanding and sympathy for each participant's everyday challenges. The meetings were perceived as a common ground where the participants felt free to ask questions regarding things they were worried about. For instance, the less experienced participants embraced the lived experiences of the more experienced participants by expressing the importance of and their appreciation for learning from peers, which they found useful as preparation for the difficult time ahead of them. The more experienced participants showed consideration for the feelings of the less experienced participants, who they knew would soon learn the harsh realities of living with a spouse in the advanced stage of the disease, which they expressed by showing their sympathy for and deep understanding of the everyday challenges these participants faced. Directing their focus from the well-being of the PALS/Cis to their own needs was also seen as a major benefit of the intervention because everything around them usually concerned the PALS/Cis.

"When I watched some of the videos, I thought 'oh my God, it's me in that video.' It's me talking. And it's been like, I know it's strange to use the word 'nice', because there is nothing nice about it, but, well it gives you peace of mind. You know, relief and peace because like 'well, there's actually something I've got under control.'" (ID 2E).

Although, several participants described feelings such as tension or having stomach cramps before and during the first meeting, these feelings were later replaced by feelings of relief, peace, thankfulness, and being less lonely and less frustrated.

> *"I feel relieved when I leave the meeting. It's something about the way I breathe. There is room to breathe." (ID 2B).*

One participant did, however, find the meetings exhausting and causing stomachache, which made it difficult for her to listen and open up to the other participants. On the other hand, she said she was comfortable with just listening to the other participants because she recognized what they were saying.

Burden

Burden contained the perceived amount of effort that was required to participate. In general, participant observations revealed that the challenges the participants faced during the intervention were related to technology issues, interruptions during meetings, lack of time, and difficulties reading body language during the online meetings. Technology problems included unstable internet connections causing the screen to freeze, missing images of anyone but the person speaking, echoes, and overheated devices. Although the participants had secluded themselves from their surroundings during meetings – in bedrooms, private offices, the workplace, a car, or children's or parents' houses – they were sometimes interrupted by phones ringing, children entering the room, requests to assist the PALS/Cis, finding a charger for the computer, and having to change location in the home.

> *"And then again, with a poor internet connection, and then one thing happens after another. Well, and then there's just the thing about having my husband in the house, right? Well, you can't just … I've had to close three doors and turn on the TV in the living room downstairs to make sure that he can't hear me." (ID 2K).*

The interruptions shifted the attention of the participants from the meetings to the situation in the home. Because of time pressure, the participants had difficulties giving priority to themselves and talked about finding it hard to settle down to participate in the meetings and watch the videos between the meetings.

> *"Well, at that point I thought that maybe an hour would be enough, because I actually felt exhausted. You had to be ready for it, and you had to compose yourself and find the time, and it had to fit into your daily schedule." (ID 2F).*

The meetings were described as intense, which fatigued some participants, but on the other hand, they did not want to reduce the length of the meetings.

> *"It's difficult to deal with such emotional themes for 45 min, and get everyone to say something. On the other hand, it's also extremely difficult to set two hours aside when you are at home with a sick spouse and a care team. Your presence is frequently required, so you must go back and forth during the meeting. You're interrupted. And then something else happens, and you have return to the subject being discussed, but can you do that mentally? It probably can't be done any other way when you're in this situation." (ID 2I).*

Mutual apprehension between the participants was also referred to as an important factor.

> *"I think it was because we hadn't picked each other. Because we all know that if you, like, know the others, then you know who you get along with. So, it was like being together with people who were forced on you, because you hadn't chosen them." (ID 2F).*

Despite finding the virtual platform convenient and easy to operate, some participants said they would have preferred physical meetings because the virtual format made it difficult to read body language and have casual conversations.

Ethicality

Ethicality concerned the extent to which EMBRACE had a good fit with the participants' value systems. The meetings became an intimate room for asking other participants direct and confronting questions as well as a room for telling their personal experiences about everyday challenges regarding the PALS/Cis, such as how to do deal with apathy from PALS/Cis or deal with great frustrations due to living with PALS/Cis who had no recognition of their disease.

> *"It's really like you're asking people; 'Why has your husband chosen to live?' It's really a difficult and big question. That's what I've been struggling with for a long time." (ID 2K).*

For several participants telling the truth involved many ethical issues which they had not discussed with anyone else.

> *"You expose your spouse in a completely different way, right? And the thoughts you share are not something other members of the family should hear." (ID 2K).*

For instance, this could be a wish for a quick disease trajectory because of the degrading situation for the PALS/Cis, but at the same time not wanting to lose a spouse. However, this was difficult for one participant to relate to.

> *" It's been hard. Because members of my group were quite negative, and it drained my energy. I think they had a hard time finding something positive to say. And I couldn't relate to how they somehow wanted it to come to an end. It was a completely different world for me (giggles). Yes. I felt they almost blamed their spouse for falling ill. Presumably, leaving them as the strong ones. For me it was unfamiliar land, I didn't understand them." (ID 2F).*

Several participants described having no one else to share such thoughts with, as they did not expect people without personal experience with a PALS/Cis to understand their situation and feared they would be judged as unsympathetic.

> *"Well, I think that the honesty – that honesty – that you don't have to beat around the bush because you're scared, you know... That it's actually okay to say 'right now it really sucks,' you know, 'because so and so and so'. People know what it means, it's not just because I use bad language, it's because I'm being honest. You don't have to be afraid to tread on someone's toes or eh … People understand you and they accept it, right. But as I said before, I wish the intervention would have been longer." (ID 2O).*

Some participants felt that sharing personal stories from everyday life in the group meetings would expose the PALS/Cis in a negative but nevertheless truthful way. During meetings the participants shared details about private dilemmas and challenging situations even though it meant exposing themselves and their partners. When asked to think of a dream scenario of how everyday life could be, some participants found it difficult because they had trouble putting themselves first and said that they were not the one that was soon to die.

Intervention coherence

Intervention coherence concerned the extent to which the participants understood EMBRACE and how it worked. The participants expressed an understanding of the purpose of EMBRACE, by underlining the meaning of the intervention targeting their needs as relatives, but participant observations showed that sometimes they had to be reminded to focus on their own needs and challenges and not on those of the PALS/Cis.

> *"I think EMBRACE is really good because it offers information. It prepares you for everything that's going to happen. I think that's important. You become prepared for what you're probably going to face. Well, so you're prepared to act." (ID 2I).*

> *"Getting a forum as a relative and gaining this knowledge. Because I wouldn't have gotten any knowledge if I hadn't searched for it myself. So, what turns up as a structured offer in such a course is really good, because the disease IS serious!" (ID 2I).*

The participants exemplified how the intervention had worked, for instance, by pointing to the supportive element of peer support and insight into various experiences on how to handle or prepare for possible future challenges.

> *"And group meetings, that's the thing when you hear from other people, that they are … I'm reassured that I'm not alone in the world, that there are others whose lives are as hard as mine. I also get, I also discover that there are others that are just getting started." (ID 2A).*

> *"No, but just talking to someone who knows how it is, and how it can be, and how much the disease takes up your life and how you sometimes feel like throwing up and think 'I don't want to do this anymore, can it please just end.' Sometimes you just feel like that. Of course, people don't understand you when they're not in the middle of it, so you don't say it aloud. But it's actually okay to speak out to someone who's in the same situation, because we've all felt like that now and then." (ID 2C).*

The participants thereby gained a better understanding of their own situation and challenges and how to handle these. Participants emphasized the common thread between relevant topics in the videos and the group discussions that prompted emotional conversations that they could not have had with family and friends. Despite being different in terms of personalities, values, challenges, and stages of their partner's disease, the participants' common situation of living with a PALS/Cis made it possible for them to better understand, relate to, and support one another.

Opportunity costs

Opportunity costs were related to the extent to which benefits, profits, or values must be given up to engage in EMBRACE. This construct was not one of the main focus areas, but two conditions were brought up. These concerned having to take time off from work and cancelling a study group meeting to participate. The participants generally gave high priority to the meetings although their busy lives made it hard for them to find the time.

> *"It suited me fine. Because of the COVID-19 pandemic I was working from home, so I could fit the meetings into my schedule and work flexible hours." (ID 2I).*

> *"When it's busy at work, the driver [a colleague] walks around singing. But that's the way it is. That's the only way for me to participate. I couldn't participate from home. That's not possible. Well, that's nonsense, because I could have said to myself; 'I'll go to another room and close the door and the care team can yell and scream as crazily as they want." (ID 2A).*

Perceived effectiveness

Perceived effectiveness concerned the extent to which EMBRACE was perceived as likely to achieve its purpose. The participants found the intervention useful and relevant, especially stressing the importance and benefits of peer support and targeted videos.

> *"I've learned something every time. I really have. Also, my understanding of the disease and all the issues it raises. Well, in a way, I wouldn't say, I've calmed down inside, but I think I'm more prepared for what's going to happen. Emotionally, too. Because you have seen other group members who are at a more advanced stage of the disease and how they have handled it. However, we are all different*

> *and deal with such situations in different ways. You must remember that. But it has certainly helped me, because I have begun to search for who I am and to be better prepared emotionally as things happen." (ID 2H).*

> *"I think that the thing about us being at different stages of the disease, I think that's really good. I don't think there would be anything to learn from it if all our relatives had just been diagnosed, because what would we talk about? I think that (being at different stages) is really good, and I think that those of us that are new learn a lot from hearing the stories. A great deal actually." (ID 2K).*

They found the topics, format, and discussions so useful that the did not want the intervention to end. They said that they felt included in a community of shared destinies where sympathy for each other's everyday life challenges was emphasized.

> *"Well, to be seen, heard, and understood. I think that means a lot. I mean what I learn from it. You know, you can – I have a huge network – and you can talk to them, but it's in a different way, and they have another frame of reference than the one you have, as a relative. So, meeting others means a lot to me." (ID 2B).*

In spite of difficult and sorrowful conversations and an initial lack of energy, participants said that they felt the meetings were invigorating; removed some of their burden, frustrations, and loneliness; and provided them with new ideas on how to approach current or future challenges. Additionally, the videos gave rise to reflections and understanding of targeted topics, thereby intensifying the focus on the participants' needs and challenges.

> *"I think it was good, and that it (EMBRACE) covered many different things – both practical and emotional things – and well, all the different challenges that you have faced or will face." (ID 2H).*

For some, writing notes and reflections in their diary was a way to reduce stress by helping them to "get things out of their mind" and not constantly having to remember everything. From participant observations, we noted that several participants talked about experiencing bodily relief, feeling calmer, more peaceful, and being able to breathe easier.

Self-efficacy

Self-efficacy concerned the participants' confidence that they could perform the behavior required to participate. They described different behaviors and how these either enhanced or hindered their participation in EMBRACE, like having difficulties in asking confronting questions or figuring out how to express oneself.

> *"I haven't done anything wrong, right? I'm really bad at that. I mean I'm really bad at blaming myself for everything. But I've also become better at realizing and accepting it, and I'm working on doing something about it. It's a huge process, and I'm not sure that I'll ever cross the finishing line; I know that, but it's a relief to know that it's there." (ID 2E).*

The virtual format made it possible and easy to attend the meetings and watch the videos, which enhanced participation. The flexible and non-demanding nature of the meetings helped the participants attend without having a guilty conscience about not being "prepared", not having watched the videos beforehand, etc. The diary made it easy to take notes for those who found this valuable. Motivation for wanting to learn more about how to handle challenges related to living with a PALS/Cis as well as contributing to research to support future caregivers also enhanced the participants' engagement in the intervention.

> *"It was great having the opportunity to talk, but I also found it difficult. Although I spoke very bluntly in that context, it was hard. It was hard for me to assess what was the right thing to say. It was very difficult because I wanted to give something to the others, but did I do that? Or was it a scare story, or what was it, right?" (ID 2I).*

Lack of concentration and poor memory were mentioned as hindering factors for their ability to focus and remember things said in the videos.

> *"Then I will try to download them, because I think that the one with the preacher – there were so many, many, many things that you – well that were hard to take in all at once. And that's exactly what each video is – how do I put this – it's unique, right, but's also consuming. First you must watch it, then work through it and then again convert it into something you can use. So, it's not done in just one afternoon, is it?" (ID 2O).*

Their desire to help the members of their group caused them to share their personal stories in order to prepare these members for the future. However, participant observations revealed that this sometimes involved talking about the affected relative instead of their own personal challenges. Some participants praised others for their eloquent way of describing their problems while not holding back their own thoughts. Moreover, they became more courageous during the series of meetings, asking each other more personal questions and discussing serious issues.

> *"Well, I could listen and then I could ask. When we'd meet once or twice it was okay to ask those questions – about practical matters but also about difficult things. And one thing I could also really use it for was that I could use it to, like, think about 'how am I as a person in this (situation)' compared to 'how are the others." (ID 2H).*

Some participants talked about gaining new personal insights during the meetings and how the meetings changed their ways of understanding and dealing with different situations.

Discussion

This study sheds light on the acceptance of the EMBRACE intervention from the perspectives of caregivers of PALS/Cis. We found factors related to all constructs of the TFA, but some were more prominent than others. The discussion is structured according to the TFA constructs.

Regarding affective attitude, the participants generally reported very positive experiences about participating in EMBRACE and for the opportunity to engage with peers. Our study showed that the participants made use of their peer's different perspectives regarding caring for a PALS/Cis to prepare for future challenges. Similar findings were observed in a recent study on a psychoeducational intervention for persons with ALS and their caregivers [36]. They found that peer-support was one of the two main reasons for utility of the intervention [36]. In other studies, peer-support has been shown to lead to camaraderie, comparisons, and hope [37]. Comparisons with people who are dealing with things that are experienced as worse or more difficult than what ALS patients dealing with has further been demonstrated to be helpful for ALS-patients to feel better about themselves and their situation [37]. Trying to balance between handling everyday challenges and not knowing what will come next seemed to use up a lot of the participants' resources. This is in line with prior studies showing that caregivers face the conflict of trying to be prepared for the future while being overwhelmed by the issues of caring throughout the progression of the illness and coping with uncertainty [38, 39]. However, the participants in the present study embraced the stories from the more experienced participants despite this opening a potential black box regarding the later stages of the disease. Locock and Brown [37] found that some caregivers and ALS-patients chose isolation as a deliberate defense strategy to protect themselves from facing a potential future situation while others valued social interaction with peers. For our participants sharing thoughts on hopes and sorrows with peers during group meetings broke down some of the barriers and fears concerning the future. A report on caregivers' preparation for the death of their relative found that caregivers were plagued with a guilty conscience when thinking about the future [38]. They found that caregivers might be cognitive and behaviorally prepared for the future but not emotionally, due to the situation of living with both hope and fear [38]. The participants in our study looked forward to the meetings and appreciated the focus on their needs and challenges, despite sometimes finding it difficult not to talk about the PALS/Cis. This might be because caregivers tend to regard their own needs as secondary compared to the needs of the PALS/Cis [15].

Burden concerned the technology issues, caregiving responsibilities, and lack of time that affected the participants' attention toward the elements of the intervention. We found that the blended virtual format created an accessible opportunity for the caregivers to participate despite lack of time, intense meetings, and problems with the technology. Our findings on the benefits of using an online blended learning format showed that it enhanced accessibility and could perhaps bridge the gap between the needs of the caregivers and their lack of time due to caregiving responsibilities and practical tasks. In line with our findings, another study showed that accessibility of the support given was crucial for the increase in self-efficacy among caregivers of ALS-patients [21]. Our findings demonstrate that the participants prioritized attending the meetings because they felt related in a special way to the other group members, who understood their situation and meet their needs for support. Mazanderani et al. (2012) also found that similarities in diagnosis was an important reason for valuing other's experiences as knowledge [40]. The use of social media has also been shown to increase the connection among caregivers of people with ALS, as well as their attendance and socialization [41]. The sense of distance that can occur between people when communicating through social media can furthermore for some people enable particular forms of computer-mediated distal empathy and still enable interactions and sharing of experiences with peers [40]. However, timely provision of problem-solving coping strategies is important to take into account when mitigating caregiver burden in PALS/Cis [1].

Ethicality was identified as dealing with feelings of guilt regarding the sharing of private challenges and exposing the PALS/Cis, but at the same time not wishing to be judged by peers. The participants placed themselves in vulnerable positions by being transparent about their everyday challenges. However, talking with peers about challenges and future concerns seemed to reduce feelings of guilt, which is consistent with a previous study on caregivers of people with ALS [15]. The authors found that caregivers experienced cohesion when sharing personal experiences and tips with peers who understood their situation and what they were going through, which nobody else in their social network could [15]. Contrary to that study, participants in our study shared intimate challenges with peers, and did not feel that topics like these were too private to discuss [15].

Intervention coherence concerned to what degree the participants found the topics relevant, useful, and empowering in relation to understanding and dealing with their personal challenges. To offer the participants knowledge on the disease, existential factors, resilience, and potential future challenges empowered them to change or moderate their interactions with the affected relative, which was also found in a previous report [18]. Effective caregiving requires that the caregivers receive emotional and practical support which allow them to better manage the different needs of their sick relatives, thereby reducing the overall burden and increasing empowerment [18]. Our study showed that the participants engaged with peers and supported each other in a way that family and friends were not able to do. Despite exposing a vulnerable side of oneself and risking potential tough comments from peers, the participants found the courage to speak up in order to receive advice and support. Reports confirm the benefits of peer support as encouraging mutuality and overcoming feelings of social isolation [42, 43]. We found that the participants were willing to open up and share concerns, which contradicts what De Witt et al. (2019) found in caregivers of people with ALS, where the majority of participants indicated that they would be passive partakers in group sessions and would only read the information and not share personal stories [15]. Studies show that being in the same situation as ALS-patients or caregivers was experienced as beneficial in relation to comparison of progression and challenges, but also confronting in terms of facing reality [19, 37]. A study illustrated that involvement in groups of carers or ALS-patients could change over time as they struggled with their changing needs and fears [37].

Opportunity costs were related to how the participants had to give up work or study groups to participate and were not something that they paid a lot of attention to. This might be influenced by the setting in which the research was conducted, because in Denmark, health and social care is free of charge, and the participants therefore did not experience financial costs in relation to participation. In contrast, studies on caregivers of people with ALS have found that caregivers perceive the uncertainty about their financial futures as stressful, because care responsibilities often compete with work and/or other family commitments [39, 44]. The majority of published studies stem from developed countries, and many studies do not take socioeconomic variables into account, like individuals wealth or national healthcare systems, which makes it difficult to extrapolate results to all countries [45].

Perceived effectiveness concerned the participants' feeling that group meetings and peer support were invigorating, encouraged mutuality, and removed some of their burden, frustrations, and loneliness, providing them with ideas on how to approach and deal with challenges. The intervention thereby seemed to fulfill its purpose. However, a report by Weisser et al. (2015) shows that caregivers of people with ALS express a need to be encouraged to seek support, timely information, and education, based on personalized care, in order to foster resilience [46]. Nevertheless, we found that targeting information on cognitive and behavioral impairments not only offered an intimate and reflexive environment but was also useful to emphasize the shared destinies and to learn from peers. Caga et al. (2021) also found it particularly important to offer information on ALS and cognitive impairments and problem-solving strategies as part of supporting caregivers of PALS/Cis [1]. Our results showed that some participants found it beneficial to keep a diary during the intervention. Offering caregivers of critical ill persons a diary is important as a means to gain understanding and to cope, and it may also reduce post-traumatic stress disorder, anxiety, and depression among caregivers [47].

Self-efficacy involved how the participants found it convenient and easy for them to participate. The blended learning format and the non-demanding participation seemed to be imperative to accommodate the heavily burdened participants who found it difficult to leave their relative at home alone. These findings are consistent with a previous report highlighting online services, like telehealth, as a way to support caregivers of people with ALS [48]. Telehealth in ALS is often well-received by caregivers, but finances and legislation may hinder telehealth implementation in ALS care [49]. Our results showed that the participants were motivated to learn more about the disease and how to deal with it, but that they found it difficult to assimilate knowledge due to stress, fatigue, and poor memory. However, a recent report showed that knowing too much about the disease trajectory could have a negative effect on caregivers' experience of burden [44]. Nevertheless, our study adds to the importance and meaning of gaining insight and knowledge from peers to understand and manage the diseases as a caregiver [19]. Our study also adds to the success of complimenting group-based peer support with psycho-educational interventions [50].

Overall, the participants' acceptance of EMBRACE was related to the opportunity to meet and share experiences with peers, which is in line with the TFA's assumption that acceptability may impact the behavioral engagement in the intervention [27].

Strengths and limitations

This study has several strengths. The TFA framework and the theory of Antonovsky proved useful for guiding the interviews, the intervention, and the analysis of data as the TFA offered pre-defined constructs to address a complex phenomenon as acceptability. This helped us design the intervention to increase the sense of coherence and reduce the stress of caregivers of PALS/Cis. The data triangulation of interview and observation data provided us with a rich and nuanced perspective of the participants' level of acceptance and thereby strengthened insights gained [35]. The participants were interviewed within two weeks after the intervention, which meant their experiences were still on top of their minds. It might have strengthened our findings if the participants were interviewed about their acceptance of the intervention during one of the group meetings, as it would have provided the participants with the opportunity to discuss, share, and elaborate on their perceptions of acceptance of EMBRACE. A potential limitation could be that we did not include the pre-interviews in the present study, which could have given insight into the participants' expectations regarding the EMBRACE intervention before enrollment. However, we aimed to evaluate the participants' acceptability of EMBRACE, not their perception of the intervention. Additionally, questions in the interview guide for the post-interviews were not fully analyzed in this study due to the deductive TFA analysis, which was used as an alternative to thematic analysis and could therefore have comprised the empirical data [51]. In the same way that the guide lacked specific TFA questions which might have revealed further perspectives on ethicality and opportunity costs, some constructs were only represented briefly and therefore perhaps not fully portrayed in the current study. However, by using the TFA in the analysis, we were able to access both enhancing as well as restraining issues regarding evaluation of the acceptance of EMBRACE [27].

As to representative credibility, the relatively small sampling of 12 participants reflects firsthand perspectives of 10 participants interviewed on acceptance of EMBRACE but with an overrepresentation of women's perspectives (Table 1). However, the ratio of men to women with ALS is reported to be between 1 and 2 [52]. Most participants in the present study were between 51 and 67 years of age, and thereby represented the general family caregiver [52]. They represented the full trajectory of ALS, with experiences ranging from months to 14 years. Despite this sampling, there is always more to study, and according to Thorne, there are no such notion as data saturation [35].

According to analytical logic and interpretive authority [35], the first author generated all the data while also being an "insider" with experience of working within the research field. The fact that the first author played a central role before and after the intervention might have entailed the risk of the participants not speaking freely and honestly. The first author did, however, not facilitate the group meetings and therefore were not in direct contact with the participants during the intervention. Furthermore, none of the participants knew her beforehand and during the interviews, they did not hesitate to express pros and cons of their perception of EMBRACE. Finally, the "insider" position made preunderstandings unavoidable which could have increased the risk of missing aspects or misinterpretations in relation to what an "outsider" would find [35]. However, to avoid these risks, the research team was a combination of researchers conducting the intervention and researchers who did not contribute to carrying out the intervention. Moreover, to reduce the risk of "blind spots", we included an external health anthropologist in the research group, who performed the initial observations. In collaboration the whole team coded the participant observations along with scrutinizing data according to the TFA constructs. Interviews were coded by the first and fifth author and then discussed in the whole research team.

Conclusion

This study evaluates the acceptance of the online palliative rehabilitation blended learning program, EMBRACE, from the perspectives of caregivers of PALS/Cis. Results indicate that the intervention supported caregivers of PALS/Cis in dealing with everyday challenges in relation to a PALS/Cis and reduced their experience of guilty conscience, fear, loneliness, uncertainties, and gave insights into ways of dealing with everyday challenges now and in the future that they could not have gained elsewhere. A special atmosphere in the group meetings fostered greater social connectedness and feelings of belonging to a group among the participants, thereby reducing feelings of loneliness. The results demonstrate facilitators as well as barriers to consider when offering targeted online group-based interventions for caregivers of PALS/Cis. Attention toward the participants' experience of affective attitude, burden, ethicality, opportunity costs, and self-efficacy should be especially considered when targeting caregiver support in order to develop an acceptable and useful supportive intervention. The use of the TFA helped identify issues within the seven constructs of acceptability that were useful for informing modifications in the design of EMBRACE. Future research should investigate the perceived impact from participating in EMBRACE and the effect of online peer-support for caregivers of PALS/Cis. Moreover, future studies should evaluate the EMBRACE intervention through a process evaluation, exploring contextual factors, implementation processes, and mechanisms of impact. Finally, it would be important to design an intervention for healthcare professionals to ALS-families as they experience decreased job satisfaction and are at risk of burn-out.

Supplementary Information

The online version contains supplementary material available at https://doi.org/10.1186/s12913-022-07986-4.

Additional file 1.

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Authors' contributions

All authors have made a substantial contribution to all the following: (a) conceptions and design of the study, or acquisition of data, or analysis and interpretation of data; (b) drafting the article or critical revision for important intellectual content; (c) final approval for the submitted.

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Availability of data and materials

The datasets generated and analyzed during the current study are not publicly available because they contain information that could compromise research participant privacy/consent, but they are available from the corresponding author on reasonable request.

Declarations

Ethical approval and consent to participate

On 20/11/2020 the study was retrospectively registered on clinicaltrials.gov [ID no. NCT04638608] and follows the COREQ checklist [53] and the Declaration of Helsinki [54]. Ethical approval for this study was deemed unnecessary according to national regulations by the Danish Data Protection Agency [File no. 2019-521-0144] and the Central Denmark Region Committees on Health Research Ethics [File no. 1-10-72-1-19]. Participants were informed orally and in writing about the project and the aim of the study. Oral and written informed consent were obtained from all participants and data were anonymized and participants referred to by identifying initials. No relationship between the participants and authors 1, 2, 4, and 5 was established prior to study commencement. Author 3 knew two of the participants from her work as an ALS counsellor.

Consent to publication

Not applicable.

Competing interests

The authors declare that they have no competing interests.

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Publisher's Note

Article III

Title page

Experienced benefits and challenges of an online palliative rehabilitation program for family caregivers of people with amyotrophic lateral sclerosis

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Clinical Trial Registration Number: Trial register: Danish Rehabilitation Centre for Neuromuscular Diseases Protocol Record DanishRebCND Lene Klem Olesen, ID number: NCT04638608. Registration was done until after the interviews had been conducted because the need to register was not known beforehand.

Abstract

Purpose: The purpose was to explore the benefits and challenges experienced by family caregivers of persons with amyotrophic lateral sclerosis and cognitive and/or behavioral impairments.

Materials and methods: A qualitative interpretive study using individual semi-structured interviews pre- and post-participation in a palliative rehabilitation blended learning program was applied. In total, 13 family caregivers were interviewed pre-intervention and 10 of them post-intervention.

Results: Three overarching themes were identified: *Striving to Obtain Control in Everyday Life, Peer support Across the Illness Trajectory, and The Complexity of Relations*. Information provided in targeted videos and sharing experiences with peers in virtual group meetings were beneficial to comprehend, manage, and find meaning in everyday challenges related to being a caregiver.

Conclusion: Participants were presented with information from videos and peers that made them more aware of their present situation and what to expect in the future, which they considered beneficial. Challenges concerned the information flow with regard to facing difficulties and the impact of the illness in terms of fatigue and lack of time.

Keywords: family caregivers, amyotrophic lateral sclerosis, cognitive impairments, behavioral impairments, palliative rehabilitation, blended learning, qualitative design

Clinical trial registration: This study was registered on clinicaltrials.gov [ID no. NCT04638608]. A Complex Intervention Study on a Palliative Rehabilitation Blended Learning Program to Support Relatives and Health Care Providers of People with ALS and Cognitive Impairments in Coping with Challenges – Full Text View – ClinicalTrials.gov

Main text

Experienced of benefits and challenges of an online palliative rehabilitation program for family caregivers of people with amyotrophic lateral sclerosis

Introduction

Amyotrophic lateral sclerosis (ALS) is a fatal, progressive, heterogeneous neurodegenerative disease with no cure [1] with death occurring after a median of 3.2 years [2]. Cognitive and behavioral impairments are prominent non-motor manifestations of ALS akin to frontotemporal dementia (FTD) [3]. Behavioral impairments in ALS are associated with poorer prognosis and risk of death that is 2 to 2.53 times higher than in unimpaired controls [4]. Cognitive and behavioral impairments contribute to the burden of family caregiver (hereafter caregivers) of persons with ALS [5, 6]. In fact, the presence of cognitive and behavioral impairments in these persons are a major source of high caregiver burden, and caregivers of persons with ALS and FTD are three times more likely to report higher burden than are those caring for persons with ALS without FTD [3]. Caregivers express a need for psychosocial support [7] such as a need to share feelings, emotions, and viewpoints with people who are in similar situations [8]. Caregivers of persons with ALS struggle with limited time, restricted social life, and extra responsibilities related to ALS [9, 10]. A review concluded that caregivers caring for a person with ALS and cognitive and/or behavioral impairments (PALS/CIs) and caregivers caring for a person with a severe clinical ALS profile may experience reduced benefits of interventions because of the caregiver's adaptive problem-solving and coping mechanisms [11]. De Wit et al. developed a blended program consisting of in-person and e-health elements based on the Acceptance and Commitment Therapy to support caregivers of person with ALS and progressive muscle atrophy [12, 13]. Their findings showed that the caregivers undergoing the program became more aware of their own situation and found they were in more control which, in turn, empowered them to make choises related to their needs [13]. Findings from the RCT of de Wit et al. showed that almost half of the participants dropped-out mainly because of lack of time [12]. Online programs enhance accessibility and provide a flexible platform that enables caregivers of persons with ALS to work on program elements where and whenever they have a moment [13]. A study on psychoeducation groups for persons with ALS and their caregivers showed that the intervention was useful because it was delivered by professionals and because it offered the possibility of sharing experiences with peers [8]. Supportive interventions for caregivers can be complex to evaluate because their effect depends on the synergictic effect between the intervention elements, and therefore conclusions tend to be ambiguous [14]. Focusing on identifying and refining the active elements in an intervention is therefore important to fully understand how an intervention works [14]. Gathering knowledge on the caregivers' experiences of benefits and challenges of a specific intervention is necessary to fully tailor a palliative rehabilitation program supporting caregivers of PALS/CIs. We therefore developed a 4-month palliative rehabilitation blended learning program, named EMBRACE to support caregivers of PALS/CIs in comprehending, managing, and finding meaning in everyday challenges related to the affected relative (Figure 1, inspired by [15]). The blended learning format consisted of in- and out-of-class-activities. The in-class-activities were five virtual group facilitated meetings every fouth week and three ecomap exercises, which is a graphic portrayal of personal and family relationships [16]. The out-of-class-activities were seventeen targeted videos, a chat room, a diary, and seven customized videos on mindfulness-based stress reduction. The targeted videos were developed on the basic of topics identified in research evidence and findings in a previous study on the challenges and needs among caregivers of deceased PALS/CIs (for further details, please see [17]). The initial videos distributed to the participants in the first out of four modules concerned psychoeducation as a means to create a mutual understanding of the cognitive and behavioral impairments related to ALS. In-class-activities were executed on Microsoft Teams and out-of-class-activities were provided on the online hosting platform Simplero. The participants received a blank notebook to use as their private diary and were free to write whatever they felt was worth noting. Vitual group meetings were facilitated by the fourth author who had extensive knowledge of the target group from working as a healthcare professional at RCFM. The intervention was conducted in 2020.

INSERT FIGURE 1 ABOUT HERE

The aim of this study was to explore the benefits and challenges experienced by family caregivers of PALS/CIs before and after participation in the palliative rehabilitation blended learning program EMBRACE.

Materials and methods

Design and setting

This qualitative interpretive study is embedded in a larger research project the EMBRACE intervention. The study is guided by the British Medical Research Council (MRC) framework for developing and evaluating complex interventions in health [18, 19]. As a means to understand the mechanisms of the impact of EMBRACE, a qualitative interpretive study design was applied to explore the experienced benefits and challenges of EMBRACE. The present study was conducted at the Danish National Rehabilitation Center for Neuromuscular Diseases (RCFM), for further details, please see [20].

Participants

Thirteen participants were included (Figure 2, Table 1). Four men and nine women. The inclusion criteria were: (a) partners or spouses of a relative diagnosed with ALS, but not necessarily diagnosed with FTD or cognitive and/or behavioral impairments, referred to RCFM and who had received an initial visit from a professional at RCFM. The partner or spouse had to be co-habiting with the person with ALS, (b) caregivers who understood and spoke Danish, and (c) caregivers of persons with ALS with a cut-off score ≥22 on the Amyotrophic Lateral Sclerosis-Frontotemporal Dementia-Questionnaire (ALS-FTD-Q), a validated questionnaire containing 25 items, with a total score ranging from 0–100, higher scores indicating more behavioral impairments [21]. The cut-off-score ≥22 indicated mild behavioral impairments [21].

Participants were encouraged to invite a person from their network or family to participate alongside them in the virtual group meetings to support them throughout the illness trajectory of their relative. The co-participants could not be the PALS/CIs. The co-participants were not considered participants and therefore not interviewed. Their role during meetings was passive indicating that they were not the primary focus of the meetings.

INSERT FIGURE 2 AROUND HERE

INSERT TABLE 1 AROUND HERE

Recruitment

Recruitment took place at RCFM. Healthcare professionals from RCFM sampled the participants by identifying 208 persons with ALS, referred to RCFM up to September 8, 2020 who met inclusion criteria a and b. Due to various or different screening procedure at neurological departments in Danish hospitals, the research team send each of the 208 caregivers an information letter about the EMBRACE intervention. The intention was to get the caregivers, who perceived their partner or spouse as behaviorally impaired, to contact the first or fourth author for further screening using the ALS-FTD-Q with a healthcare professional (inclusion criteria c). In total, 31 interested caregivers contacted the first or fourth author by phone and were then informed about each question at a time with score possibilities and provided with additional information if needed to score their affected relative [21]. Participants who were not eligible for EMBRACE were offered extra support from relevant healthcare professionals at RCFM.

Data generation

Data on the study group was generated using individual semi-structured interviews with the participants enrolled pre- and post-intervention and with non-completers post-intervention. Interviews with participants who did not finish were analyzed to explore the participants' potential barriers for participation. Interviews were conducted in Danish by the first author and quotes were later translated by two interpreters, one a native Dane and one a native American. Semi-structured interview guides with open-ended questions were used [appendix 1]. The questions focused on the participants' experiences of benefits and challenges in relation to EMBRACE with specific attention to the intervention elements, e.g., the diary, mindfulness, group meetings ect. The participants were introduced to the intervention elements during the pre-intervention interviews as a means to explore their expectation towards the elements (appendix 1). Seventeen of 23 (73,91%) interviews were completed online using Microsoft Teams. There was also one telephone interview and three in-person interviews at the RCFM and two at the caregiver's workplace. Interviews were digitally recorded and lasted between 27 minutes and 1 hour and 41 minutes. Background characteristics of the participants' experience of burden, anxiety, and depression were obtained using the Zarit Burden Interview (ZBI) and the Hospital, Anxiety and Depression Scale (HADS) pre- and post-intervention [22-25] for further details on the questionnaires please see Tabel 1.

Data analysis

Methodology and theoretical framework

Data was generated from the same group of participants. The study was founded in an interpretive description (ID) methodology. A qualitative inductive analytical methodology was used to gain insight into the participants' experiences of benefits and challenges [26]. We thus analyzed empirical data that went beyond the self-evident content analysis. Our interpretation involved inductive reasoning and pattern recognition to reveal the underlying complexity of practice as a means to improve palliative rehabilitation services in the future [26].

The theoretical framework of Sense of Coherence (SOC) was used indirectly and inductively to explore the participant's way of coping with life stressors such as the disease and the consequences of it in relation to their participation in EMBRACE [27]. SOC consists of the three concepts comprehensibility, manageability, and meaningfulness that should be present to cope with and experience life as coherent and thereby reduce the experience of stress [27]. SOC helped derive knowledge that could be essential when designing health-promoting interventions for caregivers of PALS/CIs and to increase the intervention's capability to strengthen caregivers' sense of coherence [27]. SOC indirectly permeated the entire study from the development of EMBRACE to the design, data collection, analysis, and writing of the findings [27, 28].

The analysis was guided by four steps according to the ID methodology and indirectly and inductively by the SOC concepts [26, 27, 29]. First, interviews were transcribed and uploaded into NVivo^{TM}12 to manage data analysis. Second, transcripts were read intensively for the pre- and post-intervention interviews respectively and coded for insights related to benefits and challenges. Third, patterns and relationships within and between the data groupings in the pre- and post-interviews were explored and finally across the whole data set. The whole research team critically discussed the relationships and patterns within data, and relevant tentative thematic insights leading to the primary categorization of themes were negotiated and defined. An iterative process of going back and forth between raw data and tentative themes lead to the main concepts derived from the key insights in data and captured the overarching themes which were then indirectly associated with the SOC concepts [27]. Fourth, a visual depiction of main findings and the relationship among these was developed to represent a coherent interpretive thematic and conceptual description [26].

Ethics

The study was registered on clinicaltrials.gov [ID no. NCT04638608] and adheres to the COREQ check-list [30], the Declaration of Helsinki [31] and conform to the ICMJE. Ethical approval for this study was deemed unnecessary according to national legislations by the Danish Data Protection Agency [File no. 2019-521-0144] and the Central Denmark Region Committees on Health Research Ethics [File no. 1-10-72-1-19]. Informed written and oral consent were obtained from all participants and data were anonymized and participants referred to by IDs.

Findings

Our findings represent the participants' expected benefits and challenges before the EMBRACE intervention and experiences of benefits and challenges after the intervention (Figure 3). Findings indirectly show how the participants, before starting the intervention, anticipated that EMBRACE would help them comprehend, manage, and find meaning with everyday life and how they, after finishing the intervention, had enhanced their comprehensibility, manageability and meaningfulness through the knowledge gained (elaborated below). An interrelated parallel process that entailed both individual and interpersonal factors had an influence on the participants' experience of benefits and challenges of EMBRACE. The individual factors included a striving to obtain control in everyday life, followed by the interpersonal factors involving meeting peers across the illness trajectory, and finally an appreciation of the complexity of relations. Three parallel themes emerged inductively represented by nine subthemes. The three overarching themes were: *Striving to Obtain Control in*

Everyday Life, *Peer support Across the Illness Trajectory, and The Complexity of Relations*. To provide the readers with an overall context, we first present our findings derived from pre-intervention interviews by unfolding the subthemes, followed by a presentation of the findings from the post-intervention interviews.

INSERT FIGURE 3 ABOUT HERE

Pre-intervention interviews

Striving to Obtain Control in Everyday Life

Managing various support needs

The participants talked about the dilemma of being indirectly burdened by the severity of ALS, meanwhile grappling with the realization that they were not the one who was going to die. They said that they envisioned and hoped that EMBRACE would help them comprehend and manage the various needs they experienced and help them relax and process grief and a bad conscious.

> *"My expectations are perhaps that I'll learn to understand myself better and just be with it, instead to trying to push it away…I am not sick, so perhaps learn how to live with it, myself and my sorrow." (ID 1E)*

Being able to understand and learn how to live with the consequences of ALS/CIs seemed to foster less burden and stress. They imagined the videos could be a medium for mirroring everyday lives, feelings, and ways of dealing with the challenges.

> "*In relation to the videos [presented in the interview] I somehow hope that they will give me some peace and show me that it's quite normal to react the way I do. Because I am sad, and I react angrily, sometimes because of impotence." (ID 1O)*

They thought the videos would help find meaning and peace now and later and thereby provide a means to understand and sense themselves again.

Anticipating everyday life coping skills

The participants talked about their anticipation of the intervention's focus and looked forward to the time given them to talk about themselves instead of the PALS/CIs. They expressed a motivation and wish to gain new knowledge about ALS and its cognitive and behavioral impairments as well as insights and tips on how to comprehend and manage challenges in the present and future course of the disease.

> *"To get as much knowledge as possible will help me later, yes makes things easier. Well, I become more autonomous and not that dependent on other people…and that's what I think this [the intervention] could help me with." (ID 1H)*

They described an anticipated hope that knowledge gained from EMBRACE could help them find meaning and ways of dealing with challenges, like the changed behaviors and changes in their own behavioral patterns, help them avoid scolding their partner.

> *"It's not up until now that I have realized how I have missed being informed, for a long time, about all these topics and being able to talk about them and have someone to share my views on them." (ID 1F)*

Targeted information was seen as way of clarifying issues through discussions with peers, and participants in the intervention expressed a wish that EMBRACE could help them clarify whether they were on the right track in terms of how they managed everyday challenges.

They argued that touching upon sensitive and taboo topics such as death, sorrow, and intimacy could in advance seem harsh but nevertheless relevant.

> *”If you had asked me one month ago, it would properly have been the other video, the one about difficulties in communication [presented with the topics in the videos]. But that’s not of relevance anymore. Now it’s about the future, right? Well how is it going to go.” (ID 1M)*

Offering support and information at the beginning of the disease was emphasized as important. Participants described a struggle to decide what was the right or wrong thing to do when trying to manage everyday challenges.

Peer support Across the Illness Trajectory

Expectations regarding meeting peers

The participants expressed how they believed that meeting peers would be the most important and meaningful element in the intervention. They talked about how they imagined that meeting peers would imply that they did not have to explain, excuse, or argue about their experiences, frustrations, and sorrowful thoughts.

> *”I’m looking forward to it [meeting people in the same situation], I really am. Because I hope...we don’t have to sit and gaze at our navels, it’s just that you get to talk to someone who knows. Well, you don’t have to explain things, they know exactly what you mean, if you say ‘Yes, how are things going with X and X or how do you experience X and X, and what do you do?’.” (ID 1O)*

The participants who were new to caring for a person with ALS talked about being anxious about meeting caregivers with experience of the later stages of the disease because they anticipated that these caregivers would present them with information and challenges about what could occur. The participants with years of experience with ALS expressed an urgent altruistic need to prepare caregivers new to ALS for what will come.

> *”We are our own little knowledge bank, right? And why not share with others what you have figured out or what you know is a practical way to handle problems here and now.” (ID 1O)*

They all believed and expressed that it was important and necessary to engage with peers with the aim of sharing experiences because they considered every peer to in *‘the same boat’*, as one participant said.

The complexity of relations

Contemplations on including relations

The participants talked about the balance of trying to navigate the relationships with families and friends. They described how they imagined that bringing a family member or a friend into the virtual group meetings could provide support.

> *”First, I thought it was a bit weird. But then after I had thought it through and talked to my friend about it, then it seemed like a good idea because you can continue to discuss the things afterwards without having to repeat everything. And it’s very nice that other people know about your problems so you can lean on them for support in everyday life.” (ID 1K)*

However, they envisioned that involving a family member or friend in everyday life through participation in EMBRACE could potentially scare or worry them, and therefore their involvement prompted careful consideration.

> *“Honestly, I must say I don’t actually think that anyone from our network could bear participating in the intervention. I don’t think they can, well they can’t.” (ID 1H)*

They explained how they pondered whether participation from people in their network would reduce their own interaction, honesty, and openness in the meetings because they also wished to shield family members and friends from their concerns and frustrations.

> *"I've decided that it was – that this was a road I wouldn't want them to go down because it would be too difficult: I mean I'm not sure that I would want them inside my 'headquarters' in this." (ID 1I)*

Post-intervention interviews

Striving to Obtain Control in Everyday Life

A space to unload

The participants talked about how EMBRACE had helped them to prioritize time for themselves, and instead of mindfulness, most participants prioritized activities like walking the dog, running, doing training exercises, or handicrafts.

> "*I have started to walk all the way down to the beach and it's really nice. And I practice continuing with that because it makes me feel good to take a walk in the woods by myself, no doubt about it! Well, just to walk and be by myself." (ID 2E)*

They highlighted the peer support given in the group meetings and the topics in the videos as helpful in term of feeling recognized, being able to breathe easier, and feelings of relief. One participant described the group meetings like this:

> *"Well, I think there's room to whine [in the group meetings]. That's all there's to it, if you could say it like that…Because I have said it before, you have a need to talk about yourself but also about the sick relative. And to hear yourself talk and the other members talk means that you are part of the process all the time." (ID 2B)*

Being able to reflect and discuss topics with peers or perhaps write about them in the diary was regarded as a way of processing stress and doubt. Stress made it difficult for them to remember things, and they reported how notes in the diary or on a computer or piece of paper had helped them structure and memorize information. Others experienced and described how they were too tired to write things down, while other expressed that keeping a diary was not something they would do.

Courage to address difficult issues

The participants talked about how EMBRACE had provided them with new strategies and a courage to talk to the PALS/CIs about difficult things at home and with the peers in the virtual group meetings. They described how the videos gave various insights into existential topics like ethics, morals, values, and dilemmas.

> *"I actually think that I've been good at separating the person from the disease, but also you learn about communicating clearly in relation to the person and in relation to the disease. That's one of the things that I feel I've gotten help with. The pastor [in the video] has helped a lot, you know, ethics, morals, and values. Eh, yes, it's the pastor's way of addressing it that I think I've found useful. You know all the intangible things." (ID 2B)*

They explained how topics from the videos were later brought up for discussion in the virtual group meetings. This meant that they were given an opportunity to reflect on topics they had not considered or were suppressing because they were seen as challenging.

> *"You get to touch upon various topics. Otherwise, you'll just be sitting and talking about 'this is my life, my life is like this, and what do you think about that?'…But when XXX [name of the group facilitator] is there and asks questions then you get to talk*

> *about topics that you haven't brought up yourselves…Perhaps you start to think of some other things instead." (ID 2C)*

Tools for future challenges

The participants talked about their everyday lives and their future concerns and dreams in relation to topics presented in the videos. They explained how knowledge gained from EMBRACE had made them feel more capable of comprehending and managing future disease progression and challenges because thoughts and concerns had been demystified.

> *"It's a nice change…Well, I have tried to look back and realized that the way I acted was more unconscious, and I'm glad to see that some things stick in my mind. Then it's not something that I have to concentrate on and say 'Now I must do this'…And now when I look back, I see that I have actually used it [knowledge reviewed in the intervention] without thinking. And I think that's cool." (ID 2E)*

Information from the videos and perspectives from the discussions in virtual group meetings on what to do or say gave them a ballast and helped them be on guard in terms of putting their foot down and setting boundaries.

> *"I was reaffirmed that it was okay, well that you try to get your life working. I said at one of the meetings, that it feels like I've been withdrawing a bit from XXX and that I had a bad conscience about that, right? And then I was told that it was important, right? I was told that it was okay to do that without a feeling of bad conscience. Well, it was a little like being set free." (ID 2G)*

They talked about how the videos gave rise to new insights, such as specific behavioral and psychological patterns of reaction when being a partner of a fatally ill person and that this knowledge was useful to put the "*puzzle*" together.

> *”Like the many arms of an octopus, all the thoughts that are present, but how do they find their way to where they belong? ‘These thoughts belong on THAT arm’… Well, it’s been nice like that. I don’t know how to describe it. Well, a relief. It’s nice because the thoughts are there anyway.” (ID 2E)*

Peer support Across the Illness Trajectory

The complex nature of peer support

The participants described how meeting peers was the most important and meaningful element of EMBRACE because they experienced a special sense of community, honestly, and support. They explained how heterogeneity in the groups was important:

> *“Well, we are four totally different people, and at totally different places in our lives, more or less. Hmm, but I think the first time it was kind of hard to meet XXX [another participant] and hear her story. But then again, what I am left with now, then I think, well I wouldn’t have gotten the same benefit out of it if she hadn’t been there. I wouldn’t!” (ID 2H)*

Generally, they experienced and expressed a need to speak candidly and how this was special and possible in the group despite participants being at different stages of the disease and having different backgrounds.

> *“Well, it’s people who are in same situation. They are just at different places in their lives and in trying to deal with the situation. But there’s an understanding for the thoughts and feelings you have and how you are doing today.” (ID 2O)*

The disease was described as the steppingstone to the establishment of meaningful relations in the virtual group meetings and was the binding experience that connected them in different ways. This mutual understanding made it easy to listen, relate, and share personal experiences.

> *"No, but to sometimes have someone to talk to who knows how it is and how it can be and how much effort the disease takes and how you're just about to give up occasionally and think 'I can't bother anymore, can it just end'. That's how it can be sometimes. And of course, people don't understand that if they are not in the middle of it, so you don't say it out loud, but you can say it out loud to someone who is in the same situation because that's how we have all feel occasionally." (ID 2C)*

The shared destinies made it legitimate for participants to ask sensitive questions in the virtual group meetings, which they described as a safe place for feelings and forbidden thoughts. For instance, expressing the frustrations involved in caring for a PALS/CIs or concerns about the long course of the disease.

The Complexity of Relations

The meaning of relations

The participants spoke of their networks and how they contained both positive and negative relations with feelings like disappointment, sorrow, anger, and love. They explained how the exercises with ecomaps in the intervention had been an eye-opener and confronted them with intense, complicated, and strained relations. One participant said:

> *"Because there's always a dream scenario when someone's missing. Then I think of my sisters. Well, for instance some of those who used to be close by, well, they are not there…yes because then I realized that you are quite alone. It sorts of gets onto the paper. 'Wow', right?" (ID 2F)*

They had come to realize during the disease trajectory how people reacted differently, with some staying and supporting them while others vanished from their lives.

> *"Luckily, I was also surprised by how many colleagues of mine were on the ecomap. Then I began 'Well, hallo they are also there'. Not because they call all the time, but they are here when I need them. And the same goes for friends and so on. But there are also people where I think 'Come on get going', right?" (ID 2K)*

They stressed how some family members and friends were reluctant to deal with the PALS/CIs because they found it difficult to communicate or be around the sorrow associated with PALS/CIs. One participant also expressed her reluctance to share her experience in her social networks:

> *"I have a good network and talk a lot with them, but the difference is that I don't want to talk too much because then they say 'Phew, I feel sorry for you, and can you cope with it, and are you going to sell the house' and things like that. And I don't feel the need to talk about that and I don't feel the need to share those very awful details". (ID 2I)*

Participants were aware that many individuals in their social networks were not ready to be confronted with the information about the disease.

Discussion

Exploring the benefits and challenges of EMBRACE was not simply a matter of either/or because the participants perceived them as inseparable and individualized, with findings centering around three themes: *Striving to Obtain Control in Everyday Life; Peer support Across the Illness Trajectory,* and *The Complexity of Relations*.

Our findings show that the participants strived to obtain control by gaining insights and perspectives that could help them deal with everyday life now and in the future course of the disease. Information on various topics provided through different intervention elements helped clarify and demystify concerns and frustrations and helped them to put the puzzle together. An RCT study showed that offering caregivers of people with dementia a multi-component psycho-educational intervention was beneficial for them with regard to coping with everyday life [32]. The RCT results show that the intervention group, n = 20, significantly improved in relation to depression level, positive appraisal of fulfillment in caregiving, self-growth, and coping skills compared with the control group, n = 24, who only received leaflets with information on a few topics [32]. Our findings indicate that the structured psychoeducational information in EMBRACE helped the participants to better understand the disease and the potential challenges that may be encountered along the disease trajectory. Similarly, Gonella et al. (2022) also found that structured and targeted psychoeducational information enhanced caregivers of persons with advanced dementia's understanding of their relative's prognosis, their acceptance of the approaching death, and belief in their inner strengths and potential [33]. This indicates that EMBRACE supported the participants' needs for information and provided beneficial knowledge that would help them clarify the uncertainties related to the disease of their relative, and thereby enhance their comprehensibility of the chaostic situation [27, 34].

Our findings on peer support illustrate that the participants gained important and new perspectives, and that they valued being able to share and discuss issues without having to explain or argue. Meeting peers reduced their feeling of loneliness and created a feeling of community. Peer support is highlighted as important in building meaningful connections and in relation to feelings of empowerment among caregivers of persons who survived a stroke [35]. Another benefit of our findings was that the participants felt acknowledged and described feeling safe to share frustrations, sorrows, and "forbidden" thoughts. A recent study showed that being part of a group allowed caregivers of persons with ALS to feel that their negative emotions were common among their peers who delt with the same difficult situations [8]. Being able to be authentic and being recognized by others in the same situation are valuable aspects of peer support [13, 36, 37]. However, we also found that seeking help from others entailed challenges for the participants with less experience of living with PALS/CIs because of the potential of having to face difficulties related to the information about future stages of the disease. A study on caregivers of persons with ALS also found benefits and challenges within peer support, with some finding it beneficial in terms of sharing experiences while others found it distressing [36]. The downside of meeting peers was the possible confrontation with future challenges [36]. Overall, this indicates that peer support in EMBRACE is regarded as a meaningful and important aspect that prepares and supports caregivers of PALS/CIs in managing and comprehending everyday life as a caregiver [27, 34].

In regard to complex relations, the participants experienced that their interpersonal relations were strained because some family members and friends feared the PALS/CIs and had withdrawn from contact, which was also found in a newly published study on caregivers of persons with ALS [38]. This study of Poppe et al. (2022) showed that strained relations affected the caregivers of persons with ALS because they were regarded as very important in their everyday lives [38]. Goldstein et al. (2006) found that the best predictors of caregivers' distress were early reduction in social activities and dissatisfaction with social relationships [39]. This is not surprising, since a study shows that social environment has a dramatic impact on our feeling of life satisfaction and well-being, especially in times of distress, crisis, or disaster [40]. Human resilience depends on the strength and richness of social connections and affiliations with groups [40], which emphasizes the importance of EMBRACE in supporting caregivers of PALS/CIs, so that they can maintain and nurture relations to support them through the disease of their relative.

Implications for research

Future studies should explore how online peer support unfolds among caregivers of PALS/CIs participating in EMBRACE and investigate if EMBRACE is effective in reducring stress in caregivers. Furthermore, such studies should explore if and how quality of life findings and skills learned in the online intervention are translatable into meaningful supportive initiatives in the participants' everyday lives.

Strengths and limitations

A strength of this study was the use of semi-structured interviews that allowed the participants to elaborate on their experienced benefits and challenges. SOC indirectly guided the analysis of participants' comprehensibility, manageability, and meaningfulness through facilitating their reflections and empowerment of everyday challenges in EMBRACE [27]. Being able to understand and describe the benefits and challenges of (potentially inherent in) EMBRACE through SOC seems essential when designing health-promoting initiatives because it could accumulate usable generalized resistance resources and eliminate burden related to participation [41].

As to representative credibility, the findings from the 23 interviews reflect 13 participants' firsthand perspectives, and these helped to understand the complex phenomena of benefits and challenges of EMBRACE. According to Thorne (2016), there is no specific endpoint with an assumption of qualitative data saturation, because this implies that one would have obtained and interpretated sufficient data to fully understand all that is potentially relevant to the phenomenon under investigation, which is not possible according to our epistemological orientation [26]. Prior to the pre-intervention interviews, none of the participants knew the first author who conducted the interviews. However, in the post-intervention interviews she had met briefly with all the participants during the first and fifth group meeting, welcoming and thanking them for their participation, which might have influenced the degree of honesty of the participants' statements. In this way the interviewer was an "insider" with an increased risk of having blind spots, e.g., missing aspects or interpretations in relation to what an "outsider" would find [26]. To avoid this risk, the research team consisted of a combination of researchers who conducted the intervention and researchers who had not been part of the intervention.

Conclusion

The EMBRACE intervention provided caregivers of PALS/CIs insights into ways of comprehending, managing, and finding meaning with everyday life now and in the future course of the disease. Supportive interventions for caregivers of PALS/CIs should offer multifaceted information and knowledge related to the caregivers' needs and challenges in everyday life and prepare them for potential future consequences of ALS/CIs. The elements of EMBRACE offered the participants the opportunity to share experiences with peers and gave them targeted information and knowledge from videos. Together the peer support and gained information and knowledge were meaningful, important, and valuable aspects regarding helping participants to comprehend and manage challenges related to PALS/Cis. The participants strove to obtain control in everyday life, which entailed meeting peers across the illness trajectory but also complex relations with family and friends. These findings contributed to an overall understanding of the benefits and challenges within EMBRACE and how they, in different ways, facilitated the participants to obtain control and manage everyday life as caregivers of PALS/CIs. When developing targeted supportive initiatives for vulnerable and burdened caregivers of PALS/CIs, it is important to include targeted information that enhances comprehensibility, manageability, and meaningfulness in everyday life with a PALS/CIs. Such initiatives should also offer an opportunity to engage and share experiences with peers in similar situations across the illness trajectory. Supportive initiatives should take accessibility into account and yet make elements within the intervention supportive and informative, but this support should be optional since the caregivers are burdened.

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The authors report that there are no competing interest to declare.

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Appendices

Appendix 1. Interview guide pre-intervention for caregivers of people with amyotrophic lateral sclerosis and cognitive and/or behavioral impairments. Inspired by [15]

Key questions	Elaborative questions
Tell me about your expectations regarding participation in the EMBRACE intervention.	What made you want to participate? What to you hope to gain from participating? What considerations do you have in relation to participating? Which parts of the intervention are must beneficial to you, the support videos or meeting peers?
What do you think about the virtual format of the intervention?	Which benefits do you see in relation to the online format? Which challenges do you see in relation to the online format?
How do you feel about meeting peers in the virtual group meetings?	What considerations do you have in regard to sharing personal experiences with peers? Which reflections do you have in relation to share personal experiences with the person who was invited by the peer to participate in the group meetings? How do you feel about sharing private stories from your everyday life when your relative is not present? Where do you plan to be seated during the virtual group meetings? And why? What expectations do you have to the group facilitator?
What are your expectations regarding the videos?	Based on the titles of the videos and your situation, which topic(s) do you find most beneficial? And why? What do you think about the combination of topics in the videos in relation to your needs and challenges? What are your reflections about the topics in general? Which title(s) made the greatest impression on you? And why? Do you have a strategy on how to approach or not approach each video?
What do you think about inviting a person from your network to participate with you?	Who would you like to accompany you during the intervention? Why exactly this person? What are your expectations to this person? What are your considerations about watching the videos with your chosen person? Which benefits do you see in relation to participating with a friend/or family member? Which challenges do you see in relation to participating along with a friend or family member?

What are your thoughts on writing a diary during the intervention?	What would be beneficial for you to write in your diary? How do you imagine the diary would support you in dealing with the challenges you face in everyday life? What are your considerations on potentially sharing diary notes with peers during group meetings? What are your considerations on potentially sharing diary notes during group meetings with the person invited to participate by the peer? How would you feel about sharing your diary notes with people from your private network?
What are your throughs on practicing mindfulness during the intervention?	How willing are you to give the mindfulness a chance? What are your expectations in regard to mindfulness? What benefits do you see in regard to mindfulness? What challenges do you see in regard to mindfulness?
What are your reflections regarding making an ecomap?	How do you think making an ecomap could support you in everyday life? What benefits do you see in relation to making three ecomaps (on entry, during, and after finishing the intervention)? What challenges do you see in relation to making three ecomaps?
What is your stance on the total intervention?	Which topics do you miss in the intervention?

Interview guide post-intervention for caregivers of persons with amyotrophic lateral sclerosis and cognitive and/or behavioral impairments.

Key questions	Elaborative questions
Tell me about how it has been for you to participate in the EMBRACE intervention?	What did you gain from participating? Was there anything you experienced that was beneficial to you? If yes, what? What prompted you to participate? Do you feel that something was lacking in the intervention?
How were you able to incorporate the things you learned into your everyday life?	How was EMBRACE beneficial to you in your everyday life? How did you use the things your learned in EMBRACE in coping with everyday challenges? Which challenges did you experience when trying to incorporate the learned skills into your everyday life? In general, what did you think of EMBRACE?
(Looking at a picture of every element in the intervention) **Which elements were the most meaningful for you?**	Please elaborate why?
What did you think about the videos?	How many videos did you watch? What was your impression of the content of the videos? How did you use the videos during the intervention? How did you use the videos as preparation before the meetings? How did the topics fit your needs? What did you think of the number of videos?

	What did you think of the lengths of the videos? What did you think of the way the videos were distributed to you after each meeting as preparation for the next meeting?
How was it to meet the other participants?	What you think of the group meetings? How did you use the other participants? What influence did the group composition have on your openness? What was beneficial about the meetings? What did you think about the content of the meetings? What did you learn? What was challenging about the meetings? How did the virtual meetings work out? What did you think about the size of the group? What would you have liked to see being done differently in the meetings? What did you think about the facilitator? How did the length of the meetings fit you? How did the time of day fit you? How did the frequency fit you? How did the number of meetings fit you?
What is your opination of the blended learning format, combining videos and virtual meetings?	What did you think of the online format? What did it mean to you that EMBRACE was online? Which challenges did you experience with the online format? How did you experience the coherence between the videos and the meetings? How did you prioritize your time between watching the videos and participating in the meetings?
How did you use the diary?	What prompted you to use/not use the diary? How did your dairy reflections support you during the intervention? How will you use the diary in the future? Would you like to share the content of your diary with family and friends?
How did you use the mindfulness exercises?	What made you use/not use the exercises? What impact did the exercises have on you? In which situations did you use mindfulness?
What is your opinion of the ecomaps?	What prompted you to draw/not draw the ecomaps? How did the ecomaps effect you? What were the benefits of making ecomaps? What was challenging about making ecomaps? What did you think about making three ecomaps?
What did you think about the opportunity to write messages to the other group members?	Why did you use/not use the chatroom? What was beneficial about the chatroom? What was challenging about the chatroom?
If you could change anything in EMBRACE, what would it be?	What did you like most? What did you not like? What was most beneficial to you? What was most challenging for you? All things considered, how has it been for you to participate?

Table 1. Characteristics of participants, including pre-and post-intervention background characteristics on participants' levels of burden, anxiety, and depression using the Zarit Burden Interview (ZBI) and Hospital, Anxiety and Depression Scale (HADS)[25, 42]. Scores are separated into participants dropping out and the intervention group. Post-intervention scores were collected right after finishing the 4-month EMBRACE intervention. ZBI ranges from 0-88 points with higher scores indicating greater burden [42]. Scores ≥24 indicate clinical high burden [5]. HADS cut-off score ≥8 indicate possible cases of anxiety and/or depression [25].

Study population at baseline (n = 13[1])		
Gender, n (%)	Male	4 (30,77%)
	Female	9 (69,23%)
Age (years), median (range)		58 (39–70)
Relation, n (%)	Married	13 (100%)
Occupational status, n (%)	Working	8 (61,54%)
	Early retirement/retired	5 (38,46%)
Trajectory of ALS as a caregiver of a PALS/CIs[2] (months) median (range)		25 (2–173 months)
ALS-FTD-Q score of the person with ALS, median (range)		35 (24–55)

Background characteristics of the caregivers

	Study population pre-intervention (n = 12[3])		**Study population post-intervention** (n = 7)
	Baseline (n =5) **Drop-out group** median (range)	**Baseline** (n = 7) **Intervention group** median (range)	**Post-intervention** (n = 7) **Intervention group** median (range)
Burden	46 (33–64)	38 (26–56)	42 (34–54)
	Baseline (n = 4[4])	**Baseline** (n = 7)	**Post-intervention** (n = 7)
Anxiety	9,5 (4–14)	5 (4–14)	10 (5–12)
Depression	6.5 (4–11)	8 (1–12)	7 (1–12)

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[1] Total number of included participants. One participant lost her spouse with ALS and cognitive and/or behavioral impairments between the pre-intervention data generation and start of the EMBRACE intervention.

[2] Abbreviation for person with amyotrophic lateral sclerosis and cognitive and/or behavioral impairments

[3] One participant did not return the questionnaires.

[4] One participant did not return the questionnaire.

Figures; figure captions

Figure 1: The EMBRACE intervention. A 4-month palliative rehabilitation blended learning program to support family caregivers of persons with amyotrophic lateral sclerosis and cognitive and/or behavioral impairments. Inspired by [15]

Figure 2: Participants included and reasons for drop-out.

Figure 3: A visual depiction of expected benefits and challenges before the EMBRACE intervention and experiences of benefits and challenges after the intervention.

Implication for palliative rehabilitation

- Interventions should be developed to support partners and spouses at different stages of the disease and give them the opportunity to share experiences through group facilitated meetings.
- Health professionals should be aware of the risk of facing difficulties, such as
    - Risk of confrontations with what might be ahead
    - Some participants may be more talkative and having wider boundaries related to confrontations, sharing information that everyone might not be ready to hear yet.
    - Health professionals should therefore possess group facilitating competencies and knowledge about the disease and caregiver coping to accommodate such issues.
- Future palliative rehabilitation programs targeted caregivers of person with amyotrophic lateral sclerosis and cognitive and/or behavioral impairments should include information on ways to manage everyday challenges such as reducing stress and finding meaning in the caregiver role.
- Intervention elements should be optional and easily accessible because caregivers are highly burdened.

Figure 1. The EMBRACE intervention. A 4-month palliative rehabilitation blended learning program to support family caregivers of persons with amyotrophic lateral sclerosis and cognitive and/or behavioral impairments. Inspired by (Olesen, la Cour et al. 2022).

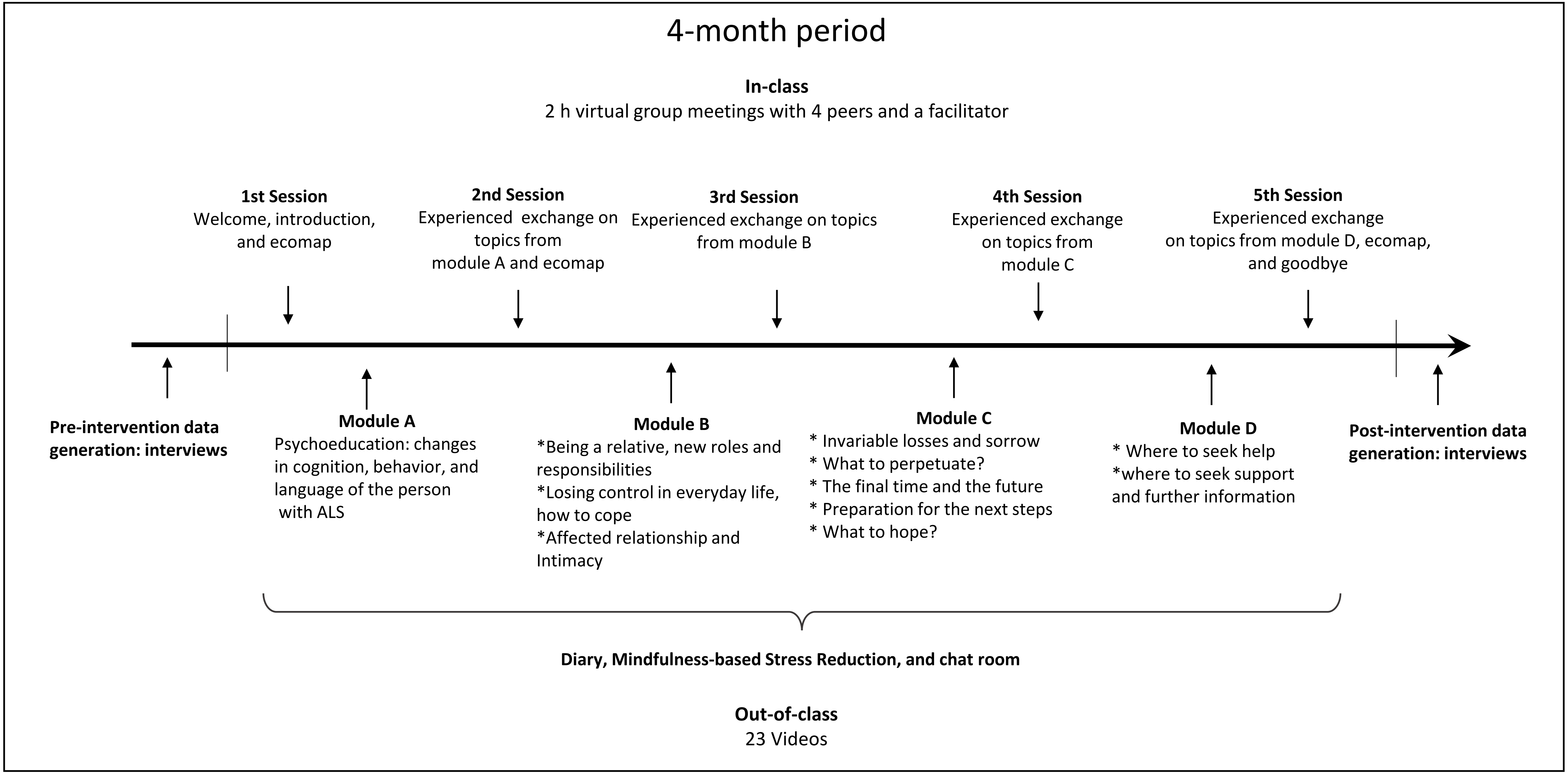
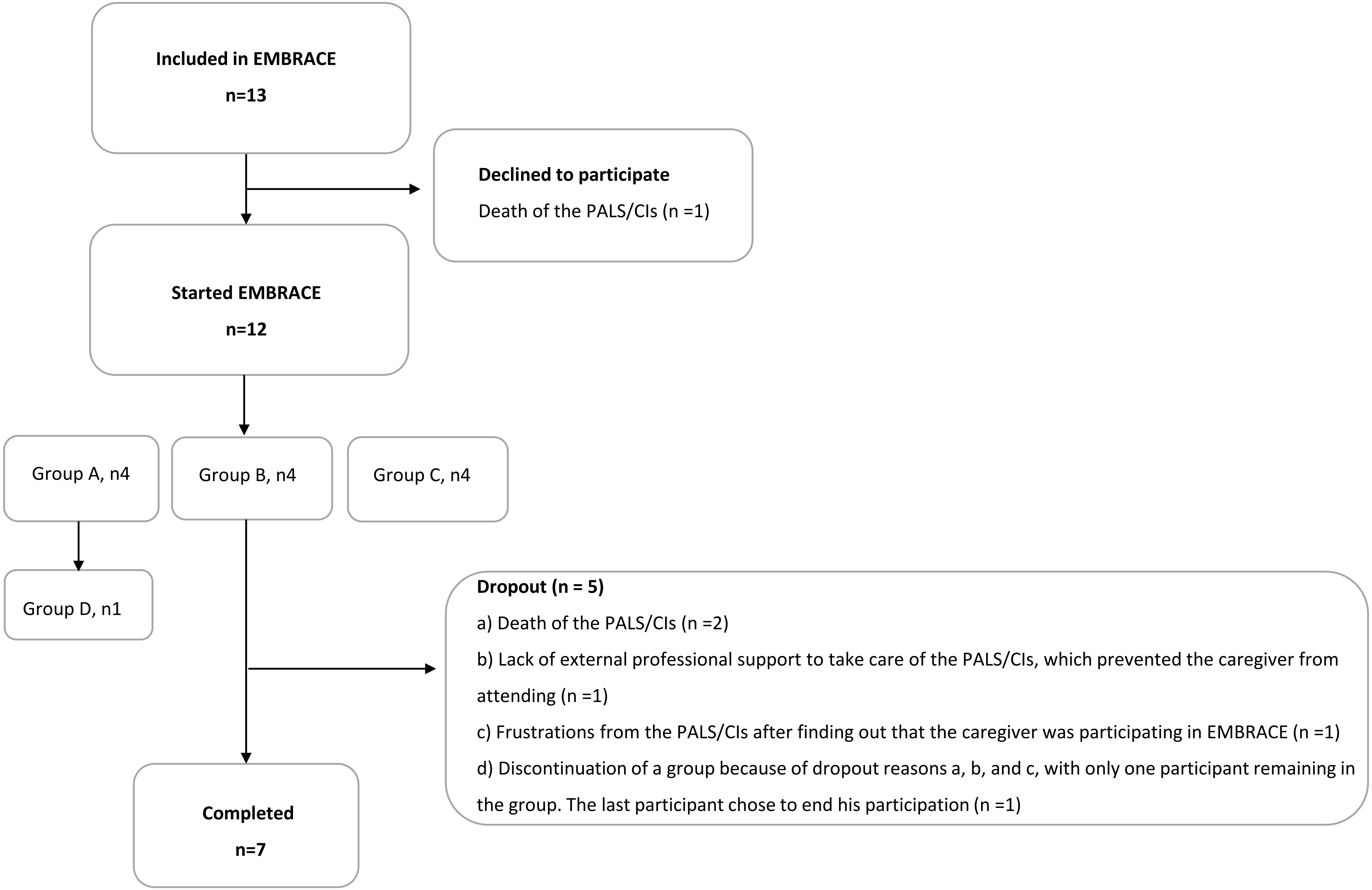


Figure 2. Participants included and reasons for drop-out



Dropout (n = 5)

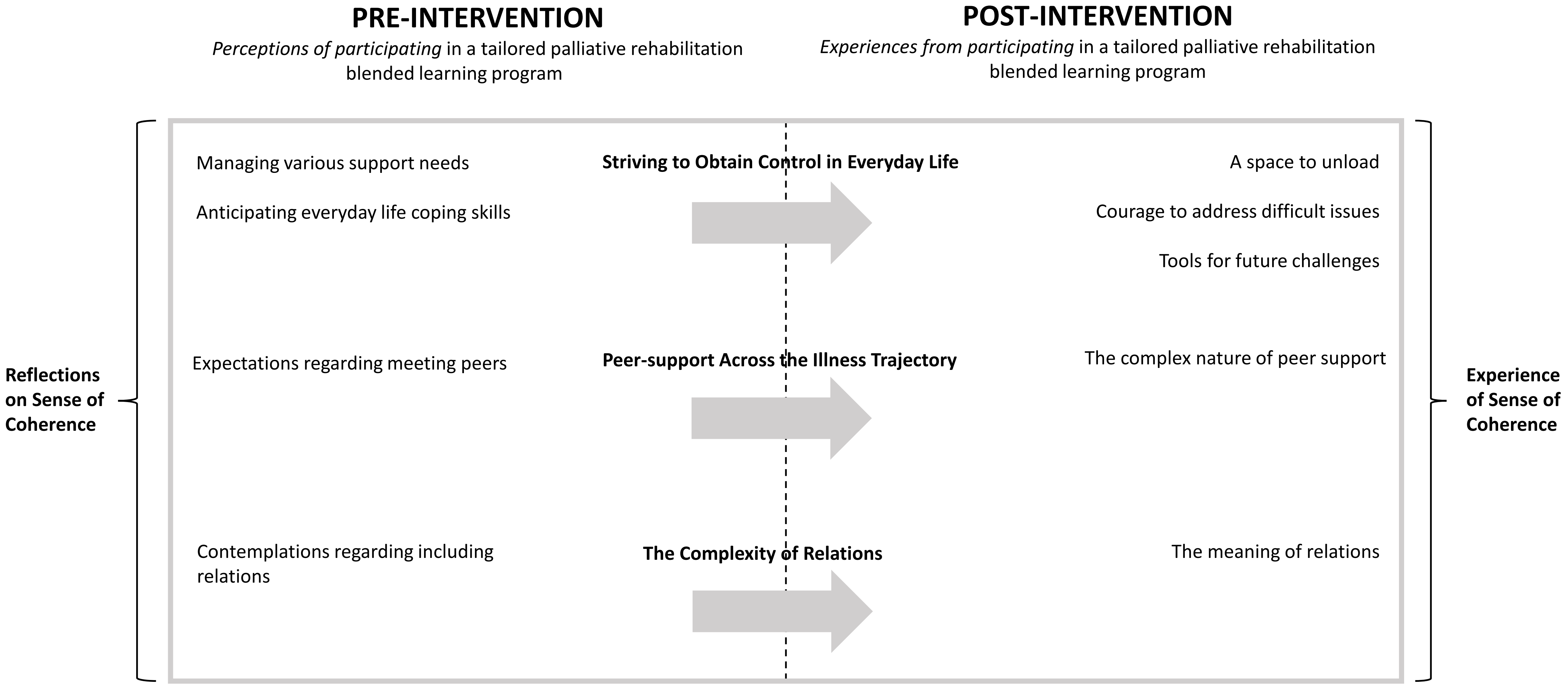
a) Death of the PALS/CIs (n =2)

b) Lack of external professional support to take care of the PALS/CIs, which prevented the caregiver from attending (n =1)

c) Frustrations from the PALS/CIs after finding out that the caregiver was participating in EMBRACE (n =1)

d) Discontinuation of a group because of dropout reasons a, b, and c, with only one participant remaining in the group. The last participant chose to end his participation (n =1)

Figure 3. A visual depiction of expected benefits and challenges before the EMBRACE intervention and experiences of benefits and challenges after the intervention.



Article IV

Peer-support among family caregivers of people with amyotrophic lateral sclerosis and cognitive impairments in a palliative rehabilitation blended online learning program

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Clinical Trial Registration Number: Trial register: Danish Rehabilitation Centre for Neuromuscular Diseases Protocol Record DanishRebCND Lene Klem Olesen, ID number: NCT04638608. Registration was done after the interviews had been conducted due to lack of knowledge on the need to register beforehand.

Authors contributions

All authors have made a substantial contribution to all the following: (a) conceptions and design of the study, or acquisition of data, or analysis and interpretation of data; (b) drafting the article or critical revision for important intellectual content; (c) final approval for the submitted paper.

Abstract

Rationale: Family caregivers of persons with amyotrophic lateral sclerosis and cognitive and/or behavioral impairments (PALS/CIs) experience various challenges and needs, including emotional and practical support from peers.

Aims and Objectives: The aim was to understand what goes on in an online peer-support in order to better promote targeted palliative rehabilitation initiatives for family caregivers of PALS/CIs.

Method: A qualitative design using participant observations of 17 recorded virtual group-facilitated meetings from two rounds of the four-month intervention was performed. The inductive interpretive description methodology and the theoretical framework of Sense of Coherence guided the study. Nineteen participants were included and divided into four groups.

Results: Three themes emerged: '*Relating my situation to others*'', '*Making room for forbidden thoughts*' and '*Longing for normalcy*'. The themes reflected the various ways caregivers interacted in online group meetings and how the interactions evolved around practical, emotional, and forbidden thoughts. Sharing personal and sorrowful concerns and frustrations engendered feelings of trust and a sense of belonging, which empowered the participants to address their genuine wish and longing for normalcy with all the trivialities that ALS/CIs had robbed of them.

Conclusion: Online peer-support enables caregivers of PALS/CIs to share experiences with everyday life challenges that cannot always be shared elsewhere. Being able to relate to and learn from other's experiences alleviated feelings of loneliness, frustration, and concerns and thereby enhanced comprehensibility, manageability, and meaningfulness. Palliative rehabilitation interventions should offer

caregivers regular online face-to-face meetings since familiarization takes time online and is necessary in order to make them feel safe to share their deepest concerns and frustrations. Online group interventions should be facilitated by trained healthcare professionals as means to support dynamic group interactions and discussion of sensitive topics.

Keywords: family caregivers, amyotrophic lateral sclerosis, cognitive impairments, behavioral impairments, palliative rehabilitation, blended learning, participant observation

Clinical trial registration: This study was registered on clinicaltrials.gov [ID no. NCT04638608]. A Complex Intervention Study on a Palliative Rehabilitation Blended Learning Program to Support Relatives and Health Care Providers of People With ALS and Cognitive Impairments in Coping With Challenges - Full Text View - ClinicalTrials.gov

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Introduction

Amyotrophic lateral sclerosis (ALS) is regarded as one of the most devasting diagnoses across the range of human illnesses due to the loss of personal independence caused by progressive muscle weakness [1]. ALS also has prominent non-motor manifestations akin to frontotemporal dementia, including cognitive and behavioral impairments [2]. Family caregivers (hereafter caregivers) of people with ALS face significant detriments to their own mental and physical health related to the ALS [3]. While ALS caregiving itself places a significant burden on the caregivers [4], the cognitive and/or behavioral impairments often create more far-reaching and burdensome consequences for them [2, 5-7]. Caregivers of people with frontotemporal dementia who claimed to have been provided a greater level of knowledge and information found it to be helpful in raising their awareness of the symptoms faced by their affected partner [8]. However, they struggle with multiple practical, social and psychological needs [9] as well as limited time, restricted social life and burdened by extra responsibilities [10, 11]. It is well recognized that caregivers of people with frontotemporal dementia indicate feelings of being alone which influences their existential well-being [12].

Understandably, ALS-caregivers typically express a need for psychosocial support [13]. One mechanism for such support is peer contact, and a space within which they can share experiences with people with whom they can relate [14]. Peer-support has been found to be crucial for affected caregivers since it creates a connection and mutual understanding which lead to a sense of hope and gratitude for life [15]. One way of offering accessible support to caregivers is through online media because the format removes some of the strains of travelling and thereby lower stress in ALS-caregivers [16]. A recent study showed that the aspects of Sense of Coherence (SOC), like comprehensibility, manageability, and meaningfulness, enhance caregivers of stage IV lung cancer patients' awareness of the disease and help them cope with everyday life [17]. Further, being aware of the situation (comprehensibility), accepting it (meaningfulness) and knowing how to deal with it (manageability) may lead caregivers to act in concordance with the challenging situations and limited prognosis, and thereby enhance their sense of coherence and reduce their stress [17, 18]. Understanding what goes on in online peer-support groups for caregiver of PALS/CIs seems missing from the existing literature. The aim of this study is to understand what goes on in online peer-support among caregiver of PALS/CIs within a palliative rehabilitation blended learning program in order to better promote targeted online peer-support rehabilitation initiatives for this population.

Methods

Design, methodology, and theory

This study was conducted as an observational study and employed video-recorded participant observations of virtual group meetings. The study was guided by interpretive description (ID) methodology and the theoretical framework of SOC [18-20]. ID was applied to unravel the complexity of online peer-support by facilitating an interpretation that contains inductive reasoning, pattern recognition, and concept relating [21]. SOC guided the development of the palliative rehabilitation blended learning program named EMBRACE and helped understand what goes on in online peer-support and how this indirectly affected the participants' sense of coherence through its three core elements; comprehensibility, manageability and meaningfulness

[18]. EMBRACE takes place over a four-month period and combines targeted videos, virtual group facilitated meetings, diary writing, mindfulness-based stress reduction and ecomap exercises (diagram of social and personal relationships) (Figure 1).

INSERT FIGURE 1 ABOUT HERE

Sample

Nineteen caregivers of PALS/CIs were included (Table 1). The inclusion criteria were: (a) being a caregiver who lives with a relative with ALS referred to the National Rehabilitation Center for Neuromuscular Diseases (RCFM) in Denmark and who had received a visit from professionals at RCFM, (b) being able to understand and speak Danish, and (c) having a relative with ALS with a cut-off score ≥22 on the Amyotrophic Lateral Sclerosis-Frontotemporal Dementia-Questionnaire (ALS-FTD-Q) [22]. Recruitment took place at RCFM, in 2020 for the first round of EMBRACE and in 2021 for the second round. A list of ALS-patients referred to RCFM was assessed by the first author (LKO) and the fourth author (HW) along with healthcare professionals from RCFM to see who meet the inclusion criteria. The procedure resulted in 208 caregivers identified for the first round and 221 caregivers for the second round (Figure 2). Invitations were sent to each of these caregivers. Fifty-one interested caregivers contacted the team by phone to be included and were then asked to score their relative using the ALS-FTD-Q [22].

INSERT TABLE 1 ABOUT HERE

INSERT FIGURE 2 ABOUT HERE

Data generation and analysis

Data collection was conducted between August and December 2020, and again between August and December 2021. It included participant observations of video-recordings of 17 virtual group facilitated meetings using Microsoft Teams and Zoom. Three groups completed all five group meetings, one group completed two out of five meetings. An observation guide was developed and used to focus on what was going on in the online peer-support meetings, such as interactions and communication among the participants, and field notes were collected. Field notes consisted of a) sketch notes of actions and behavior (e.g., obvious signs of distress or happiness; relational dynamics, b) descriptions of what was going on (e.g. discussions), and c) reflexive and analytical notes [23]. LKO conducted participant observations throughout the two intervention periods and all video-recordings were reviewed by the last author (CH) to enhance reliability of the coding.

Data consists of the 12 videos recorded in 2020 and the five videos recorded in the second round of EMBRACE in 2021 (Figure 2). The analytical process was guided by four steps suggested by ID and indirectly by the SOC-elements [18, 19]. First, all video-recordings and field notes were uploaded into the analysis program NVivo^{TM}12. Secondly, video-recordings were watched while taking field notes. Notes were read and initially coded in a broad manner for insights related to what goes on in online peer-support and indirectly to the SOC-elements. Thirdly, results of initial coding were considered and patterns and relationships among the data groupings were explored and discussed. The research team critically examined patterns and relationships within the data. This process generated tentative themes which led to the primary categorization (Figure 3). Working iteratively, the categorized data and exploring how they were related to comprehensibility, manageability and meaningfulness led to the final condensation that identified the overarching themes [18]. Fourthly, an interpretive thematic and conceptual description of the relationship among these led to an illustrative depiction capturing the main understandings what goes on in online peer-support among the participants in EMBRACE. (Figure 4, result section) [19].

INSERT FIGURE 3 ABOUT HERE

Ethics

The study was registered on clinicaltrials.gov [ID no. NCT04638608] and adheres to the STROBE guidelines [24] and the Declaration of Helsinki [25]. Ethical approval was deemed unnecessary by the Danish Data Protection Agency [File no. 2019-521-0144] and the Central Denmark Region Committees on Health Research

Ethics [File no. 1-10-72-1-19]. Informed consent was obtained from participants. No identifying information is presented in the article that could jeopardize confidentiality.

Results

Three themes were identified: “Relating my situation to others”, “Making room for forbidden thoughts” and “Longing for normalcy”. Relating situations in terms of similarities and differences was the initial and continuous interaction between the participants which led to feelings of trust with a genuine wish to share forbidden thoughts with someone who could relate. Sharing forbidden thoughts fostered a sense of belonging which empowered them to talk about their longing for normalcy with all the trivialities that they had lost because of ALS/CIs (Figure 4).

INSERT FIGURE 4 ABOUT HERE

Relating my situation to others

Facing similarities and differences

Facing similarities and differences reflected the way the participants understood and related their situation to others; it centered around stages of disease, levels of physical, cognitive, and behavioral impairments, degree of support from professionals and life situation. They seemed to comprehend what their peers talked about, which was apparent in the way they nodded and expressed interest toward the person talking. Sometimes they seemed surprised or worried when they were told something unexpected as to how things had progressed in other families, as they opened eyes widely and lifted their brows. The participants who were new to ALS/CIs appeared overwhelmed when presented with future challenges, like having their home *“invaded”* by professionals, which caused them to roll their eyes or clutch their heads. As a way of comprehending, managing, and finding meaning with their own situation they asked their peers to draw a parallel to their own experiences.

> *C: I really think it’s tough right now because it [ALS/Cis] progresses so quickly.*

> *D: It's completely understandable, C, the way you feel. I have been through it myself. All of it! [Takes a deep breath].*
>
> *C: I think it [the ALS/Cis deterioration] progresses very fast.*
>
> *D: It does progress very fast.*
>
> *C: [Crying]*
>
> *D: I've been in the same situations as you C. I was so skinny and lost one kilo after another, and in the end, I had to let the professionals take over [to care for PALS/CIs]. I completely understand the way you feel, and it's very hard! I could tell you a lot of things, but you'll figure them out yourself step by step.*
>
> *(C: female caregiver; D: male caregiver).*

Being able to share concerns with someone who show sympathy and understanding seemed meaningful, as if it reduced the participants' feelings of being alone.

Respecting different needs, values, and ways of dealing

Respecting different needs, values and ways of dealing reflected the participants' manner of welcoming various approaches to everyday challenges without judging things as "right" or "wrong". It appeared that they listened with curiosity to their peer's stories by quietly looking into the camera and nodding while others spoke. It became apparent that there was "no size fits all" and a participant stated that there are no "*easy choices"* (ID P, 2. meeting), indicating the struggle that they had to deal with when making decisions. The group environment seemed non-judgmental and enabled them to share whatever they wanted without being frowned upon. As a way of comprehending and managing ALS/Cis, it appeared that those who were new to ALS/CIs asked elaborative questions, like 'How do you deal with this…?' to the those who were at later stages of the disease.

> *C: So T, don't you think it [referring to a talk about if PALS/CIs should go to a nursing home] would give you more freedom or peace of mind?*
>
> *T: Yes, for sure.*

> *C: Well, cause your partner is very sick and he has told you that he can't cope with this anymore, right? Well, it wears him out because he feels and senses that you're sad and deeply frustrated.*
>
> *T: Yeah, that's right. We text each other about how we're feeling down, long distance.*
>
> *C: Yeah, that's one of the things that I have thought about too, because you're actually running away from home because you can't stand being there, and that's not fair! Your home should be your safe base, where you feel good…*
>
> *T: Yeah, but it's a very well-organized running away that makes me feel good*
>
> *C: Yes, but I don't think I would be able to do that because I would constantly be wondering: are they [the helpers] doing what they're supposed to do?*
>
> *(C: female caregiver; T: female caregiver).*

It seemed legitimate for the participants to ask questions, comment honestly and share each other's point of view in a respectful manner as a way of comprehending and finding meaning with everyday life as a caregiver. However, some with years of experience with ALS/CIs posed more direct and persistent questions related to how they felt bound but also prioritized and acted in a manner that allowed them to manage everyday life.

> *T: My fingers are itching to grab onto D and make him do something else besides "being on vacation" [working]. Because I think what we can do is to help each other in a constructive manner.*
>
> *D: I don't know what to say about T wanting to grab me. Well, there're only 24 hours in a day, and I struggle with a lot of thoughts. But time will probably solve it all [when PALS/CIs dies]. I'm doing it my way.*
>
> *(T: female caregiver; D: male caregiver).*

The wish to support and help each other was strong, but it was clearly permissible to act differently in terms of trying to comprehend and manage difficult situations. Prioritization and finding ways of dealing appeared to be related to different stages of ALS/CIs, everyday situation, and personality.

Discovering other's perspectives

Discovering other's perspectives represented how sharing arduous learned lessons between the participants was a way of supporting and preparing each other for the course of disease and thereby hopefully enhancing comprehensibility and manageability and reducing stress.

> *J: I really hope that she can use my experience and advice because [H] must be going through the same things as we [J and S] have… I'm hoping and crossing my fingers that H will make some good decisions for her and her family based on the knowledge she's gained. That's my hope. I'm not participating in this [intervention] for my own sake, it's too late for me now, but it's not too late for other people [tears in her eyes and biting her lip]. (Female caregiver).*

They used body language to signal the importance of different assistive aids and keenly shared advice on practicalities like when and where to apply for such aids. Those who were new to ALS/CIs seemed grateful for the advice from the more experienced participants as the new knowledge was meaningful, enhanced their comprehensibility of challenges, and prepared them to manage everyday life. This was seen in the way they constantly smiled and leaned into the conversation.

> *K: I have gained a lot of new information that I hope I'll remember to use when I need it. I think it was the perfect time for me to start this in program, and I would like to say to you H, that the experience you have shared has been indispensable for us because where else would we find that kind of knowledge... I think it's fantastic that we're all at different stages of the disease because we can all contribute with different insights. I also appreciate H's very honest recount of everyday life with a breathing machine and where to go and who to contact for information and support. (Female caregiver)*

It appeared that learning from others was important and meaningful and especially to those who were new to ALS/Cis, which emphasized the importance of being at different stages and mentoring each other on "dos and don'ts" as preparation to manage ALS/CIs. It seemed that sharing experiences on practicalities was the first natural and easy step to take in their process of familiarizing and became the steppingstone to addressing more sensitive topics.

Making room for forbidden thoughts

Touching upon sensitive topics

Touching upon sensitive topics represented the participants' deepest thoughts, concerns and frustrations related to being a caregiver of a PALS/CIs. It appeared that frustrations occurred from having to go on and not being able to do anything about the disease progression, the longing for death of the PALS/CIs and the troubled conscience that followed from having such thoughts.

> *T: I'm also getting some thoughts that I'm not sure if I can say out loud. That is, when someone is this ill [referring to T's PALS/CIs], I become really envious of the other members of this group whose partners will die before mine because their disease is not dragging on forever like I feel is the case with my partner. I think waiting is agony. Let's say that the disease trajectory is two years. Because, from my perspective, happiness for sick person or the caregiver is not that it [life of PALS/CIs] continues. It's absurd to sit here and say it out loud. (Female caregiver).*

It appeared that their peers agreed and supported the statement despite representing different stages of disease as they nodded and verbally expressed that there *"was no need to drag it out".* They talked about making hard decisions like choosing a breathing machine, which could prolong the life of their relative, and how these decisions would cause further burden on them as well as their family. It was obvious how they struggled with having to make constant adaptions to manage disease progression and how they felt torn about future hopes.

> *S: You prepare yourself for doing the best you can for your partner and set yourself aside because xxx [PALS/CIs] has a short lifespan. So, I set my own needs aside, all the things that I can do without, to please xxx [PALS/CIs], but I can't keep doing that forever. I have a life too. I sometimes think: dammit, how many years is this going to take, you know! (Female caregiver).*

Readjusting the timeframe of the disease was confusing and boundless and caused the participants to feel ashamed for wishing for a faster trajectory. Those who were at more advanced stages of ALS/CIs looked exhausted, with pallid faces, when they described how they felt indefinitely tied hand and foot. It was clear that they had lost a lot because of ALS/CIs, such as friends, families, leisure time and their life partner; these losses were recognized and comprehended by their peers. who nodded and some looked downcast and sad. Talking to like-minded others prevented the bubble from bursting and made them able to breathe more freely as peers comprehended and embraced their deepest thoughts.

Being in the same boat

Being in the same boat represented how being a caregiver of a PALS/CIs created a mutual understanding and connected them in a way that overruled anything else, like sex, age, or stage of disease. Outside the group, they had not experienced comprehending everyday life as a caregiver for a PALS/CIs, and this joint point of departure enhanced a feeling of group belonging.

> *P: I appreciate that everybody knows about the challenges you're facing. You understand these challenges when you're in the middle of them, you can't share this with outsiders. Where else would you meet someone who is in the exact same situation as you? (Female caregiver).*

It seemed like the implicit knowledge of all participants was important in terms of shared comprehension of what it was like to be a caregiver of a PALS/CIs. They took a deep breath and seemed calmed, while repeatedly expressing how talking honestly with peers was liberating, meaningful and not possible elsewhere.

> *S: It's been wonderful for me to be able to say that on one hand I want xxx [PALS/CIs] to die, but on the other hand I want to keep him. You can't share such ambivalent feelings with anyone else, but it was so nice to be allowed to say it here [in the group]. Because that's how I feel. And as you said K, to be able to express how you are feeling without being frowned upon. (Female caregiver).*

Having a 'room' to share the deepest concerns and frustrations without having to worry about the implications created a new lifeline for the participants. They poured out their hearts by sharing personal concerns and frustrations which empowered them to look into the future and express their longing for normalcy.

Longing for normalcy

T*he unknown future*

The unknown future concerned the uncertainty related to the partners' trajectory, and their inability to toward reverting to everyday life in the post-mortem. They expressed awareness of their partners' inescapable death, but without knowing how long it would take or how it would progress, which limited their comprehensibility and manageability.

> *K: I'm just waiting, which is a terrible feeling, because when does it [ALS/Cis] get worse? I can be seized by that feeling and I want it to go away, because I must enjoy what we have. But I'm constantly looking for signals and what is happening now. What is xxx [name of the PALS/CIs] saying and doing now, because all the time you hear about how fast it will progress…and you feel like being in a waiting room… It's awful to feel this way, but the thoughts are still there. (Female caregiver).*

They explained having mentally prepared themselves for a fast trajectory at the time of diagnosis but later coming to realize that the disease continued for better or worse. A participant stretched her flexed arms into the air with the palms turned upward, indicating that the unknown future raised many questions and caused frustration. The great uncertainty was also related to how they would be able to revert to everyday life after the death of the PALS/CIs. Being in suspense seemed to enhance feelings of impotence and reduce manageability because ALS/CIs overruled everything and influenced their future dreams.

> *M: I dream about rewinding life a little, until the disease vanishes completely, uhmm, but hey, I can't do that! But [if I could] xxx [name of the PALS/CIs] and I were going to have a good time and enjoy all the things we never got to do. We could travel more and stuff like that. I miss that, our active life together. (Female caregiver).*

The dreams entailed all that ALS/CIs had robbed of them, like their life partner, relations to family and friends, having time alone, and being able to do whatever they wanted. From their bodily rocking movements, it was obvious that a tension inside their body emphasized a dilemma of not being able to live out these wishes. They longed for normalcy with all the trivialities of everyday life it implied.

Discussion

We found that online peer-support among caregivers of PALS/CIs participating in EMBRACE centered around relating situations to others, making room for forbidden thoughts and longing for normalcy. It appeared that differences related to everyday life and the disease became less important as they began to familiarize and instead the conversation revolved around forbidden thoughts, concerns and frustrations and longing for normalcy. Regarding being able to relate to other's situation we found that the participants felt recognized and acknowledged by the peers and did not risk being frowned upon. A study of online peer-support in caregivers of persons with Alzheimer's also found a positive tone between the caregivers and that the support mainly pertained to the well-being of the caregivers [26]. This might indicate that peer-support revolves around embracing and empathizing with one another. We found that discovering others' perspectives helped the participants to comprehend and manage challenges and was meaningful in preparation for dealing with the future course of disease. Previous studies have likewise demonstrated how peer-support, where caregivers shared experiences, information, practical advice, emotions, adaptations, and family relations, had a positive influence on caregivers' incentive to understand and cope with challenges related to ALS [27, 28]. Bilenchi et al. (2022) found that the most important function in a group was the ability and need to share perspectives and learn from peers who are in the same situation [28]. However, tips and advice shared from one caregiver to another in a written form on an online platform is not necessarily transferable and useful since they tend to be too generic, and each individual situation is different [29]. Overall, it seems like sharing experiences and gaining other's perspectives is important and useful when peers are familiar with each other and can target the perspectives to support others comprehensibility, manageability, and meaningfulness as means to deal with everyday life with a PALS/CIs.

In the present study, we found that the participants shared forbidden and sorrowful thoughts that they could not share elsewhere, and that this caused a sense of belonging. This might be explained by a study by De Wit et al. (2019) that showed how being able to share how you feel with people who are in similar situation is valuable in relation to feeling recognized and acknowledged [29], which our findings also showed. Our participants explicitly stressed how they felt being among like-minded people where it was legitimate to ask and share sorrowful and forbidden thoughts. Perceiving oneself as being, at some level, "*the same*" as others means that people can engage and benefit from experiences shared by people who are in the same situation [30]. In keeping with our findings, it has been illustrated that peer-support provided through online media can foster recognition, acknowledgement, and self-disclosure [31]. Locock et al. (2010) also found that peer-support can lead to camaraderie, hope, mutual understanding, and comparisons because of joint characteristics of ALS which create feelings of being with people who are like me [32]. Our findings revealed that the participants found the courage to share for the first time their wishes for a fast disease trajectory and how they faced common conflicts in relation future hopes. A study showed that group interactions can enable caregivers of PALS to understand that negative emotions and feelings are common among people struggling with difficult experiences [28] which is consistent with our findings. Moreover, among people with multiple sclerosis or depression, being among like-minded others can help overcome feelings of social isolation and encourage mutuality [33, 34]. Overall, this emphasizes the importance of providing caregivers a non-judgmental environment where they can share their deepest thoughts without being frowned upon, as research has also shown that the risk of burnout increases in caregivers if cognitive impairments are present in the PALS [35].

We found that our participants struggled with having to make difficult decisions because these often would prolong the agony and thereby increase their burden. This “spillover effect” is described as the measurable effect of a patient’s illness on the surrounding individual, including the caregiver [36]. In a recent study, Lin et al. (2021) also found that caring for a family member can result in physical, emotional, and psychological conditions, like anxiety and depression [36]. As our findings showed, initiatives that prolong the agony may be associated with very high caregiver burden [37].

Longing for a normal life, as before their partner fell ill, reflected how the participants dealt with the great uncertainty related to the disease and their ability to revert to everyday life after the death of their partner. Further, they experienced a guilty conscience from having thoughts about the future. This aligns with findings from a recent study showing that caregivers often felt angry because of ALS, which led to feelings of guilt [28]. One way of adapting to new life situations is through palliative rehabilitation, which assists families to maintain resilience and adapt to losses from their loved one’s illness [38]. Additionally, sharing perspectives with peers about future concerns is also found to reduce feelings of guilt and create a sense of coherence because no one else can comprehend what the caregivers are struggling with [14] (+in review).

Strengths and limitations

A strength was the use of SOC and ID that offered theoretical elements and methods useful to help understand what goes on in online peer-support, enhancing the study’s integrity by informing the entire process from study aim, analysis and interpretation of findings to writing up the findings [18, 19, 39]. Another strength was the meeting recordings that helped to capture the participants’ choice or words and intonation, while offering limitless viewings, which supported the analysis process as well as enabled the use of quotes from group meetings to ensure credibility and substantiate this process.

By consistently using SOC we strived to make our interpretive authority more transparent and consistent by way of relating codes and themes to the SOC elements. We performed an inductive analysis guided by the iterative process described by ID, and not predetermined by the SOC elements to prevent overlooking important issues related to what goes on in online peer-support [18, 19, 39]. Another strength was that the analysis was performed individually by two researchers and later discussed within the research team, including researchers who conducted the intervention and those who did not, to reduce the potential influence of researcher subjectivity [19].

As to representative credibility, we acknowledge that our sample size was limited [19]. Nevertheless, was it a strength that the data set reflected all participants included in EMBRACE, representing differences in sex, age, and years of experience with ALS/CIs through a four-month intervention period, and thereby allowed for an understanding of what goes on in online peer-support among caregivers of PALS/CIs. A potential limitation may be that the data mainly represents aspects of ALS generally and not all aspects of its cognitive and/or behavioral impairments. Because the focus of the intervention was on the caregiver’s needs and not those of their partner, whether the participants’ scoring of their partner using the ALS-FTD-Q survey was related to or impacted by their own burden, or if the PALS was cognitively and/or behaviorally impaired remains unclear.

Conclusion

This study emphasizes the importance of enabling online face-to-face peer-support for the purpose of sharing experiences of everyday life challenges and forbidden thoughts with someone who can relate across stages of the disease. Being able to relate to and learn from others’ situations and experiences may alleviate feelings of loneliness, frustration, and concerns and thereby enhance comprehensibility, manageability and meaningfulness and support a sense of coherence. Online peer-support facilitated a non-judgmental ‘room’ for the participants to share advice and experiences on practicalities around what to do within the difficult situations but also in terms of sharing forbidden thoughts, concerns and longings related to the future course of disease and the post-mortem period. Palliative rehabilitation interventions should offer caregivers regular online meetings because feelings of belonging take time online and require the caregivers to feel safe to share concerns. Online group interventions should be facilitated by trained healthcare professionals to support dynamic group interactions and respectful discussion around sensitive topics. Overall, this indicates that there is good reason to include peer-support in supportive interventions within an online palliative rehabilitation program for this population.

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Conflict of interest:

The authors report that there are no competing interests to declare.

Tables

Table 1. Demographic on the participants

Figure legends

Figure 1. The EMBRACE intervention. A 4-month palliative rehabilitation blended learning program to support family caregivers of persons with amyotrophic lateral sclerosis and cognitive and/or behavioral impairments. Inspired by (Olesen, la Cour et al. 2022).

Figure 2. Flow diagram of participants included in the EMBRACE intervention during 2020 and 2021, including reasons for dropout.

Figure 3. Analytic steps illustrating how codes and themes were extracted.

Figure 4. Understandings of what goes on in online peer-support groups among family caregivers of people with ALS and cognitive/behavioral impairments in the palliative rehabilitation blended learning program, EMBRACE.

Tables

Table 1. Demographic on the participants

Participants		**(n = 19)**
Gender	Male	5
	Female	14
Age	39–50	3
	51–55	5
	56–67	7
	68-74	4
Relation	Married	19
Occupational status	Working	9
	Early retirement/retired	10
Trajectory of ALS (years)	0–2	10
	3–4	3
	5–8	4
	9–14	2
ALS-FTD-Q score	22–30	6
	31–35	5
	36–40	4

	41–46	2
	47–55	1
	56–74	1

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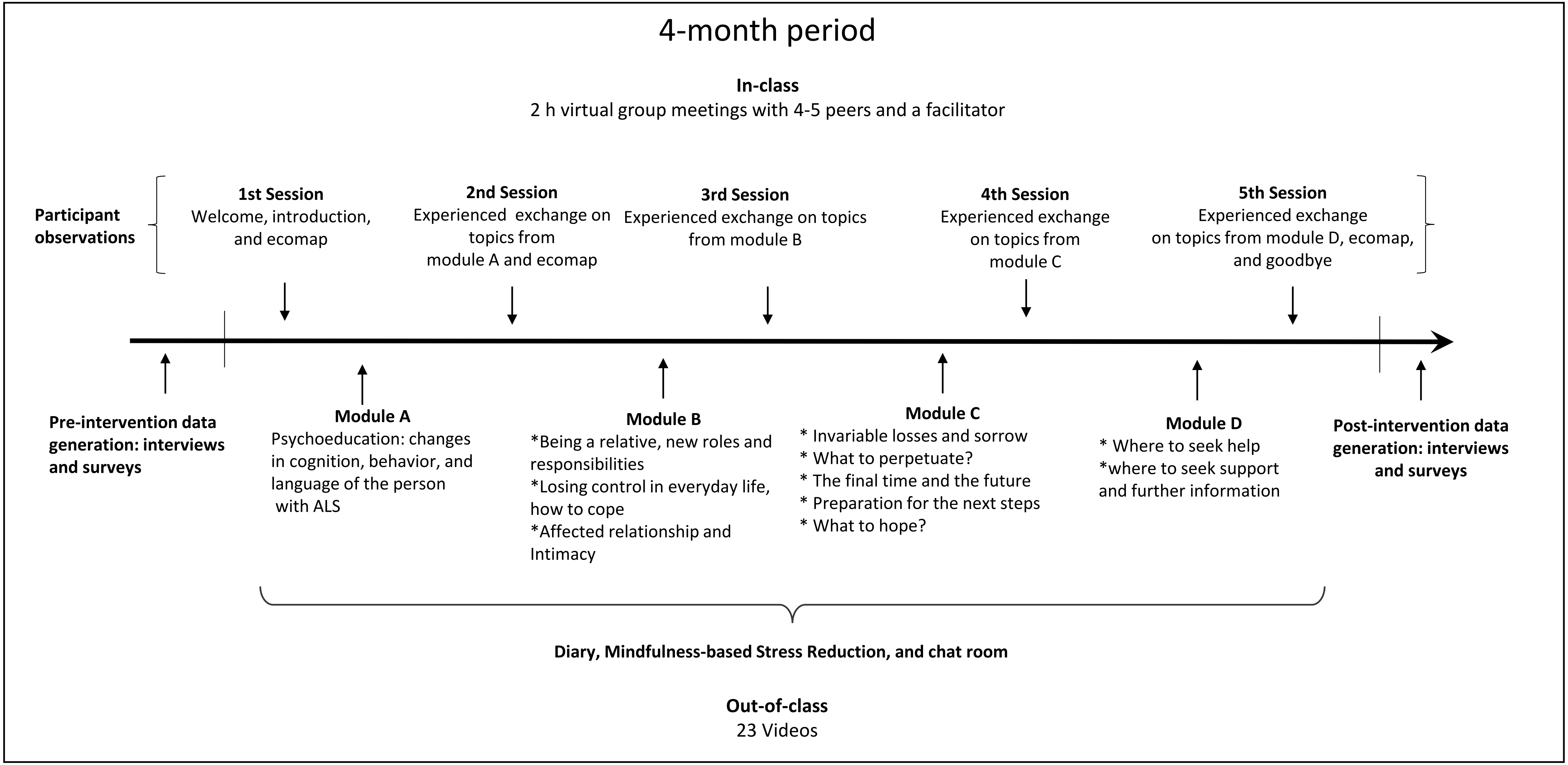

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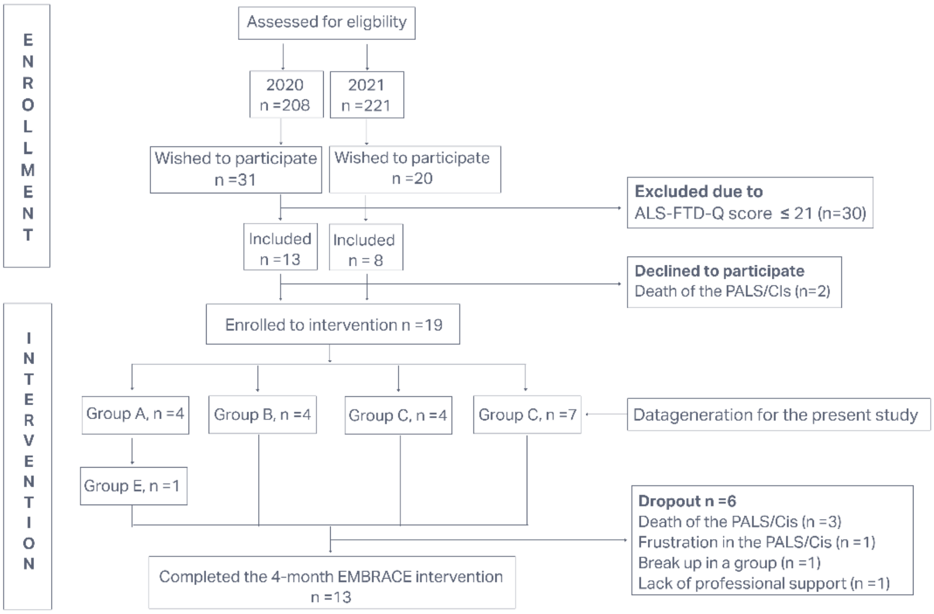


Figure 3. Analytic steps illustrating how codes and themes were extracted.

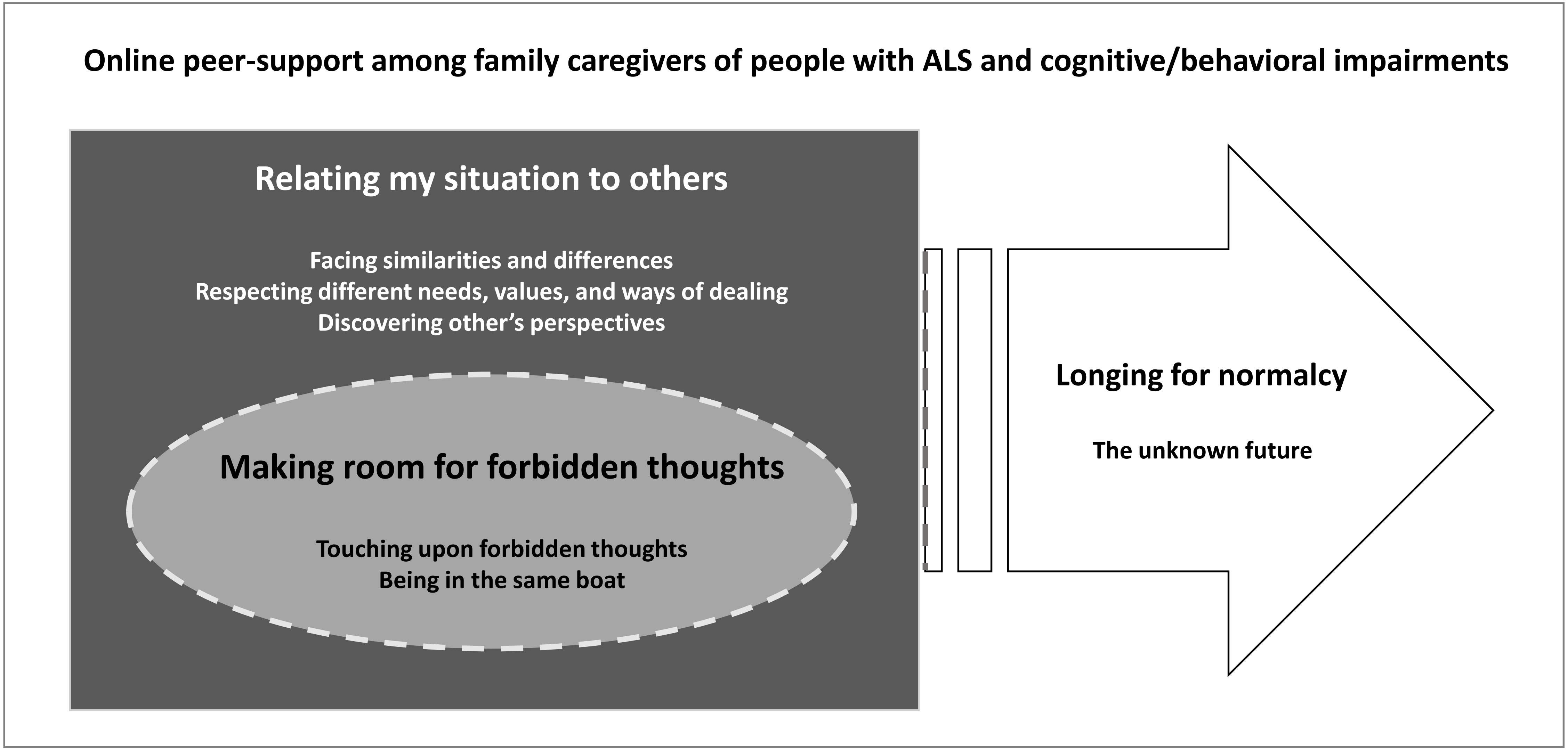
Step 1 Initial coding	Respecting different needs, values and ways of dealing 1	New perspectives rise new insights 2	Being in the same boat 3	Facing similarities and differences 4	The long and inevitable goodbye 5	Feeling torned about future hopes 6	Longing for normalcy 7	The unknown future 8	Touching upon sensitive topics 9	Sense of liberation when sharing 10

Step 2 Generalized patters	Making room for various thoughts (1, 2) A	Emulating me in relation to others (3, 4) B	Running out of time (5, 6) C	Wishing for normalcy (7, 8) D	Making room for forbidden thoughts (9, 10) E

Step 3 Primary categorization	Comparing my situation against others (A, B)	Making room for fobidden thoughts (C, E)	Longing for normalcy (D)

Step 4 Envisioning thematic findings with subthemes	**Relating my sitation to others** Facing similarities and differences Respecting different needs, values, and ways of dealing Discovering other's perspectives	**Making room for forbidden thoughts** Touching upon sensitive topics Being in the same boat	**Longing for normalcy** The unknown future

Figure 4. Understandings of what goes on in online peer-support groups among family caregivers of people with ALS and cognitive/behavioral impairments in the palliative rehabilitation blended learning program, EMBRACE.



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Appendix 1. Interview guide for caregivers of deceased persons with amyotrophic lateral sclerosis and cognitive and/or behavioral impairments (PALS/CIs). Study I

Key questions	**Elaborative questions**
Try and tell me about yourself	How long has it been since you lost your relative/ loved one?
What was your relationship with the person with amyotrophic lateral sclerosis and cognitive and/or behavioral impairments (ALS/CIs) you have lost?	How was your relationship with your family?
Looking back, try to describe what have been most burdensome for you in the whole ALS/CIs disease trajectory?	How did you experience your own situation? What did you do to manage the disease trajectory? What was important for you in this period of life? How did your network react toward you?
How did become aware of your relative's ALS/CIs?	How did the cognitive impairments (CIs) of your relative affect your relationship? What was your reaction toward the CIs of your relative? How did handle you handle the challenges related to the CIs?
How would you describe everyday life during the whole period of your relative's ALS/CIs?	How did the ALS/CI trajectory of your relative affect your family? What was your role during the disease trajectory? What kind of losses did you experience during the period? How did you cope with these losses? Who supported you during the trajectory?

Try to explain at which point in the ALS/CIs trajectory of your relative you realized you needed support?	What kind of help and support did you receive? What did you miss in order for you handle the situation? What was the biggest challenge for you in your everyday life? Who was of importance for you during this period?

Appendix 2. Interview guide for professionals caring for people with amyotrophic lateral sclerosis and cognitive and/or behavioral impairments (PALS/CIs). Study I.

Key questions	Elaborate questions
Try to elaborate on when you felt most challenged in your work with the with families with a person with amyotrophic lateral sclerosis and cognitive and/ or behavioral impairments (PALS/CIs)?	What was stressful for you in your work with the families? How did you work with the person with CIs? How did you work with the relative to the person with CIs? What would have been supportive for you in this situation? How were you able to take care of yourself? How did you experience your ability to support the carer?
How did you become aware of the cognitive and/ or behavioral impairments (CIs) of the person with amyotrophic lateral sclerosis (ALS)?	How would you describe the person with CIs? How did you experience the personality and behavior impairments? How did you experience the language impairments? When did the CIs appear during everyday life? What was your reaction toward the person with CIs? How did the CIs affect your relationship toward the person with ALS? How did you handle the challenges related to the CIs? What was the biggest challenge in regard to CIs? What did you miss in regard to your handling of the situation? What kind of support could have beneficial for you? What was the biggest support for you in this situation? What kind of role did you have in the disease period? When have you felt alone with your challenges?

Appendix 3. Interview guide post-intervention for caregivers of persons with amyotrophic lateral sclerosis and cognitive and/or behavioral impairments. Study II and III.

Key questions	Elaborative questions
Tell me about how it has been for you to participate in the EMBRACE intervention?	What did you gain from participating? Was there anything you experienced that supported you? If yes, what? What prompted you participate? What did you miss from participating?
How were you able to adapt the things you learned into your everyday life?	How was EMBRACE useful to you in your everyday life? How did you use the things your learned in EMBRACE in coping with everyday challenges? Which challenges did you experience when trying to adapt the learned skills into your everyday life? Generally, what did you think of EMBRACE?
(Looking at picture of every element in the intervention) **Which elements were the most meaningful for you?**	Please elaborate why?
What did you think about the videos?	How many videos did you watch? What was your impression of the content of the videos? How did you use the videos during the intervention? How did you use the videos as preparation before the meetings? How did the topics fit your needs? What did you think of the number of videos? What did you think of the lengths of the videos? What did you think of the way the videos were distributed to you after each meeting as preparation for the next meeting?
How was it to meet the other participants?	What you think of the group meetings? How did you use the other participants? What influence did the group composition have on your openness? What was great about the meetings? What did you think about the content in the meetings? What did you learn? What was not great about the meetings? How did the virtual meetings work out? What did you think about the size of the group? What would you have liked differently about the meetings? What did you think about the facilitator? How did the length of the meetings fit you? How did the time of the day fit you? How did the frequency fit you? How did the number of meetings fit you?
What is your opination of the blended learning format, combining videos and virtual meetings?	What did you think of the online format? What did it mean to you that EMBRACE was online? Which challenges did you experience with the online format? How did you experience the coherence between the videos and the meetings? How did you prioritize your time between watching the videos and participating in the meetings?

How did you use the diary?	What prompted you to use the diary/or not? How did your dairy reflections support you through the intervention? Prospectively, how would you use the diary? How would you like to share the content of your diary with family and friends?
How did you use the mindfulness exercises?	What made you use the exercises/or not? What impact did the exercises have on you? In which situations did you use mindfulness?
What did your opination regarding use of the ecomaps?	What prompted you to draw the ECOMAPs/ or not? How did the ECOMAPs effect you? What was great about making ECOMAPS? What was not that great about making ECOMAPs? What did you think about making three ECOMAPs?
What did you think about the opportunity to write messages to the other group members?	Why did you use the chatroom/ or not? What was great about the chatroom? What was unfavorable about the chatroom?
If you could change anything in EMBRACE, what would it be?	What did you like most? What did you not like? What was the easiest thing? What was the hardest thing? All things considered, how has it been for you to participate?

Appendix 4. Interview guide pre-intervention for caregivers of people with amyotrophic lateral sclerosis and cognitive and/or behavioral impairments. Inspired by [1]. Study III.

Key questions	Elaborative questions
Tell me about your expectation for participating in the EMBRACE intervention.	What made you want to participate? What to you hope to obtain from participating? What considerations have you in relation to participate? Which part of the intervention are must beneficial to you, the support videos or meeting peers?
What do you think about the virtual format of the intervention?	Which benefits do you see in relation to the online format? Which challenges do you see in relation to the online format?
How do you feel about meeting peers in the virtual group meetings?	Which considerations do you have in regard to share personal experiences with peers? Which reflections do you have in relation to share personal experiences with the person, who was invited by the peer to participate in the group meetings? How do you feel about sharing private stories from your everyday life, when your relative is not present? Where do you plan be situated during the virtual group meetings? And why? Which expectations do you have to the group facilitator?
What are your expectations to the videos?	Based on the headlines of the videos and your situation, which topic(s) do you find most beneficial? And why? What do you think about the combination of topics in the videos in relation to your needs and challenges? What are your reflections about the topics in general? Which headline(s) made the greatest impression on you? And why? Do you have a strategy on how to approach or not each video?
What do you think about inviting a person from your network to participate with you?	Who would you like to accompany you during the intervention? Why exactly this person? What are your expectations to this person? What are your considerations about watching the videos with your chosen person? Which benefits do you see in relation to participate with a friend/or family member? Which challenges do you see in relation to participate along with a friend or family member?
What are your thoughts on writing diary during the intervention?	What would be beneficial for you to write in your diary? How do you imagine the diary would support you in dealing with the challenges you face in everyday life? What are your considerations on potentially sharing diary notes with peers during group meetings? What are your considerations on potentially sharing diary notes during group meetings with the person invited to participate by the peer?

	How would you feel about sharing your diary notes with people from your private network?
What are your throughs on doing mindfulness during the intervention?	How willing are you to give the mindfulness a change? What are your expectations to mindfulness? What benefits do you see in regard to mindfulness? What challenges do you see in regard to mindfulness?
What are your reflections on making an ecomap?	How do you think making an ecomap could support you in everyday life? What benefits do you see in relation to making three ecomaps (when entering the intervention, during and when finishing the intervention)? What challenges do you see in relation to making three ecomaps?
What is your stance on the total intervention?	Which topics do you miss in the intervention?

1. Olesen, L.K., et al., *A cross-sectional evaluation of acceptability of an online palliative rehabilitation program for family caregivers of people with amyotrophic lateral sclerosis and cognitive and behavioral impairments.* BMC Health Serv Res, 2022. **22**(1): p. 697.

Appendix 5. Participant-observation guide for caregivers of persons with Amyotrophic Lateral Sclerosis and cognitive and/or behavioral impairments. Study II.

The TFA constructs [1]	Elaborative participant-observation questions
Affective attitude	How does the participants show and express their feelings about the intervention?
Burden	How does the participants show and express their perceived amount of effort that is required to participate?
Ethicality	How does the participants show and express the intervention's fit with their individual's value system?
Intervention cohesion	How does the participants show and express their understanding of the intervention and how it works?
Opportunity costs	How does the participants show and express their opportunity costs, like benefits, values or profits that must be given up to engage in the intervention?
Perceived effectiveness	How does the participants show and express their experience of perceived effectiveness / or the opposite with the intervention?
Self-efficacy	How does the participants show and express their confidence that they can perform the behavior(s) required to participate in the intervention?

1. Sekhon, M., M. Cartwright, and J.J. Francis, *Acceptability of healthcare interventions: an overview of reviews and development of a theoretical framework.* BMC health services research, 2017. **17**(1): p. 88-88.

Appendix 6. Participant observation guide on virtual group facilitated meetings in the EMBRACE intervention [1]. Study IV.

Areas of observations	**Sketch notes**	**Thick descriptions**	**Reflexive and analytical notes**
Behavior and action: **(What, by whom, when)** **e.g. obvious signs of distress or happiness**			
Context: **(what else is going on, discussions, dynamics)**			
Relational dynamics: **(gestures on interesse or disinterest, recognition)**			

1. DeWalt, K.M. and B.R. DeWalt, *Participant observation: a guide for fieldworkers, second edition*. 2nd ed. 2011: AltaMira Press.

Appendix 7. The Zarit Burden Interview, (ZBI-22)

SPØRGESKEMA OM BELASTNING

VEJLEDNING: I det følgende finder du en række udsagn, som viser, hvordan man kan have det, når man skal passe og pleje en anden person. Efter hvert udsagn skal du markere, hvor ofte du har det sådan: Aldrig, Sjældent, Af og til, Ret ofte eller Næsten altid. Der findes ingen rigtige eller forkerte svar.

1. Føler du, at din pårørende beder om mere hjælp, end han/hun har brug for?

   0. Aldrig 1. Sjældent 2. Af og til 3. Ret ofte 4. Næsten altid

2. Føler du, at du ikke har nok tid til dig selv på grund af den tid, du tilbringer sammen med din pårørende?

   0. Aldrig 1. Sjældent 2. Af og til 3. Ret ofte 4. Næsten altid

3. Føler du dig stresset, fordi du både skal passe din pårørende og forsøge at leve op til dit ansvar i familien eller på arbejdspladsen?

   0. Aldrig 1. Sjældent 2. Af og til 3. Ret ofte 4. Næsten altid

4. Bliver du flov over den måde, din pårørende opfører sig på?

   0. Aldrig 1. Sjældent 2. Af og til 3. Ret ofte 4. Næsten altid

5. Bliver du vred på din pårørende, når du er sammen med ham/hende?

   0. Aldrig 1. Sjældent 2. Af og til 3. Ret ofte 4. Næsten altid

6. Synes du, at din pårørende for tiden påvirker dit forhold til andre familiemedlemmer eller venner i en negativ retning?

   0. Aldrig 1. Sjældent 2. Af og til 3. Ret ofte 4. Næsten altid

7. Er du bange for, hvad fremtiden vil bringe for din pårørende?

   0. Aldrig 1. Sjældent 2. Af og til 3. Ret ofte 4. Næsten altid

8. Føler du, at din pårørende er afhængig af dig?

   0. Aldrig 1. Sjældent 2. Af og til 3. Ret ofte 4. Næsten altid

9. Føler du dig anspændt, når du er sammen med din pårørende?

   0. Aldrig 1. Sjældent 2. Af og til 3. Ret ofte 4. Næsten altid

10. Føler du, at det er gået ud over dit helbred, at du skal tage dig af din pårørende?

0. Aldrig 1. Sjældent 2. Af og til 3. Ret ofte 4. Næsten altid

11. Føler du, at du har mindre privatliv, end du kunne ønske på grund af din pårørende?

0. Aldrig 1. Sjældent 2. Af og til 3. Ret ofte 4. Næsten altid

12. Føler du, at dit sociale liv har lidt under, at du passer og plejer din pårørende?

0. Aldrig 1. Sjældent 2. Af og til 3. Ret ofte 4. Næsten altid

13. Generer det dig at have venner på besøg på grund af din pårørende?

0. Aldrig 1. Sjældent 2. Af og til 3. Ret ofte 4. Næsten altid

14. Føler du, at din pårørende synes at forvente, at det er dig, der skal tage dig af ham/hende, som om du var den eneste, han/hun kan regne med?

0. Aldrig 1. Sjældent 2. Af og til 3. Ret ofte 4. Næsten altid

15. Føler du, at du ikke har råd til at passe og pleje din pårørende oven i dine øvrige udgifter?

0. Aldrig 1. Sjældent 2. Af og til 3. Ret ofte 4. Næsten altid

16. Føler du, at du ikke vil være i stand til at tage dig af din pårørende ret meget længere?

0. Aldrig 1. Sjældent 2. Af og til 3. Ret ofte 4. Næsten altid

17. Føler du, at du har mistet kontrollen over dit liv, siden din pårørende blev syg?

0. Aldrig 1. Sjældent 2. Af og til 3. Ret ofte 4. Næsten altid

18. Ville du ønske, at du bare kunne overlade pasningen af din pårørende til andre?

0. Aldrig 1. Sjældent 2. Af og til 3. Ret ofte 4. Næsten altid

19. Føler du dig usikker på, hvordan du skal håndtere pasningen af din pårørende?

0. Aldrig 1. Sjældent 2. Af og til 3. Ret ofte 4. Næsten altid

20. Føler du, at du burde gøre mere for din pårørende?

0. Aldrig 1. Sjældent 2. Af og til 3. Ret ofte 4. Næsten altid

21. Føler du, at du burde kunne klare pasningen af din pårørende bedre, end du gør?

0. Aldrig 1. Sjældent 2. Af og til 3. Ret ofte 4. Næsten altid

22. Hvor belastende er det alt i alt for dig at passe og pleje din pårørende?

0. Slet ikke belastende 1. Lidt belastende 2. Noget belastende 3. Ganske belastende 4. Yderst belastende

Appendix 8. The Hospital, Anxiety and Depression Scale, HADS.

Spørgeskema om angst og depression, til brug på hospital (HADS)

Navn: ____ Dato: _______________

FOLD HER

Læger er opmærksomme på, at følelser spiller en stor rolle i de fleste sygdomme. Hvis din læge kender til disse følelser, vil han eller hun bedre kunne hjælpe dig.

Dette spørgeskema er udformet med henblik på at hjælpe din læge med at finde ud af, hvordan du har det. Læs hvert spørgsmål herunder og **sæt streg under det svar**, som bedst beskriver, hvordan du har haft det de seneste 7 dage. Ignorer de tal, som er skrevet ud for hvert spørgsmål.

Tænk ikke for lang tid over dine svar, idet din umiddelbare reaktion på hvert spørgsmål sikkert vil være mere præcis end en lang, gennemtænkt besvarelse.

FOLD HER

Jeg har følt mig anspændt eller stresset

A	D	
3		Det meste af tiden
2		En stor del af tiden
1		En gang imellem
0		Aldrig

Jeg har fortsat fundet glæde ved det, jeg plejede at glæde mig over

A	D	
	0	Helt som før
	1	Ikke helt som før
	2	Kun en smule
	3	Næsten ikke

Jeg har haft en slags forskrækket følelse, som om noget frygteligt var ved at ske

A	D	
3		Helt afgjort og ganske slemt
2		Ja, men det er ikke så slemt
1		En smule, men det bekymrer mig ikke
0		Slet ikke

Jeg har kunnet le og se det morsomme i mange ting

A	D	
	0	Lige så meget som altid
	1	Ikke helt som før
	2	Helt afgjort ikke som før
	3	Slet ikke

Jeg har haft bekymrende tanker, der er faret igennem hovedet på mig

A	D	
3		Næsten hele tiden
2		En stor del af tiden
1		En gang imellem, men ikke så ofte
0		Kun sjældent

Jeg har følt mig i godt humør

A	D	
	3	Aldrig
	2	Ikke ofte
	1	Somme tider
	0	Det meste af tiden

Jeg har kunnet sidde behageligt og føle mig afslappet

A	D	
0		Altid
1		Som regel
2		Ikke så ofte
3		Aldrig

Jeg har følt det, som om at jeg fungerede langsommere

	A	D
Næsten hele tiden		3
Meget ofte		2
Somme tider		1
Aldrig		0

Jeg har haft en slags forskrækket følelse, som om jeg havde "sommerfugle" i maven

	A	D
Aldrig	0	
En gang imellem	1	
Ganske ofte	2	
Meget ofte	3	

Jeg har mistet interessen for mit udseende

	A	D
Helt afgjort		3
Jeg er ikke helt så omhyggelig, som jeg burde være		2
Måske er jeg knap så omhyggelig som før		1
Jeg er lige så omhyggelig som altid		0

Jeg har følt mig rastløs, som om at jeg skulle bevæge mig hele tiden

	A	D
Virkelig meget	3	
En hel del	2	
Ikke så meget	1	
Slet ikke	0	

Jeg har med glæde set frem til, det jeg skulle lave

	A	D
Lige så meget som altid		0
Knap så meget som jeg plejede		1
Helt afgjort mindre end jeg plejede		2
Næsten ikke		3

Jeg er pludselig blevet grebet af panik

	A	D
Meget ofte	3	
Ganske ofte	2	
Ikke særlig ofte	1	
Aldrig	0	

Jeg har kunnet glæde mig over en god bog eller et godt radio- eller TV-program

	A	D
Ofte		0
Somme tider		1
Ikke ofte		2
Meget sjældent		3

Kontroller nu, at du har besvaret alle spørgsmålene

	A	D
TOTAL		

Spørgsmålene i dette skema er oprindeligt publiceret i *Acta Psychiatrica Scandinavica*, 67, 361–70,
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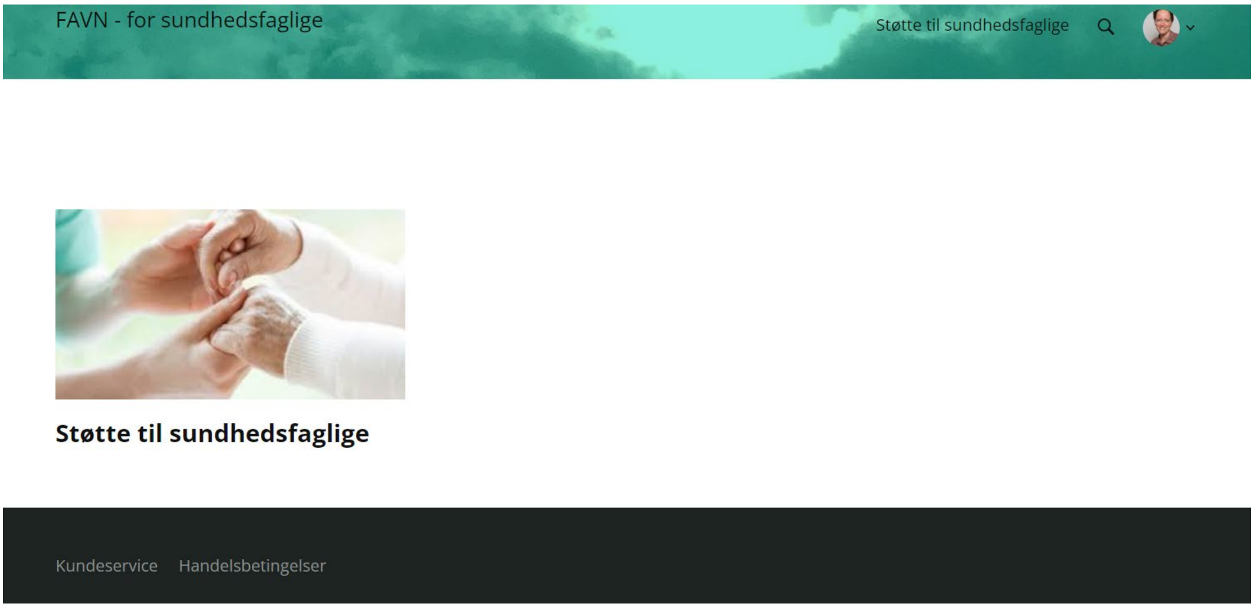
GL Assessment er en del af Granada Group.

HADS - Denmark/Danish - Version of 13 Jan 15 - Mapi.
ID4776 / HADS_AU3.0_dan-DK.doc

Appendix 9. Simplero to professionals – frontpage



Appendix 10. Simplero to professionals – overview of the module and text to one of the videos

Kursusindhold

Støtte til sundhedsfaglige 0 / 8

○ **ALS**
19:39

○ Evaluering 1

○ Kognitive forandringer
17:37

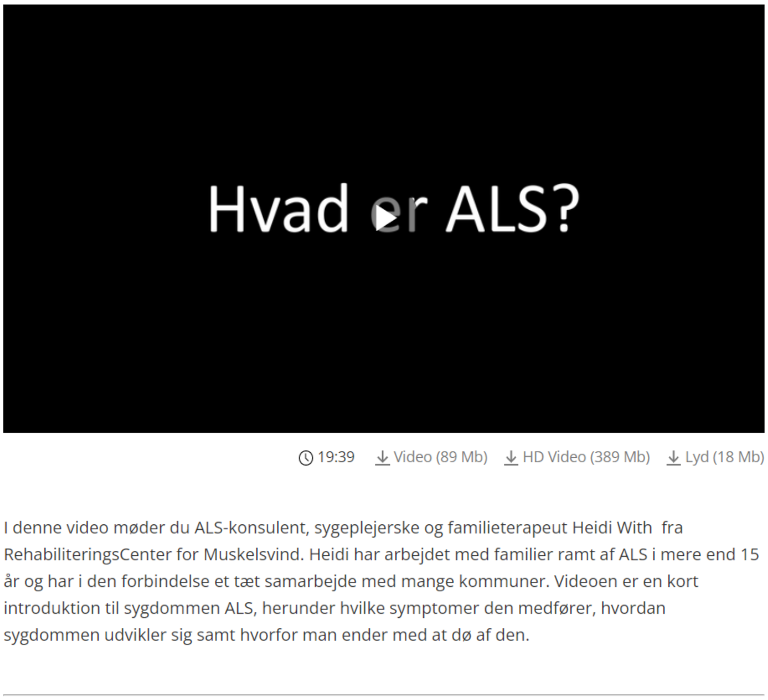
○ Evaluering 2

○ Samarbejdet med familien
13:43

○ Evaluering 3

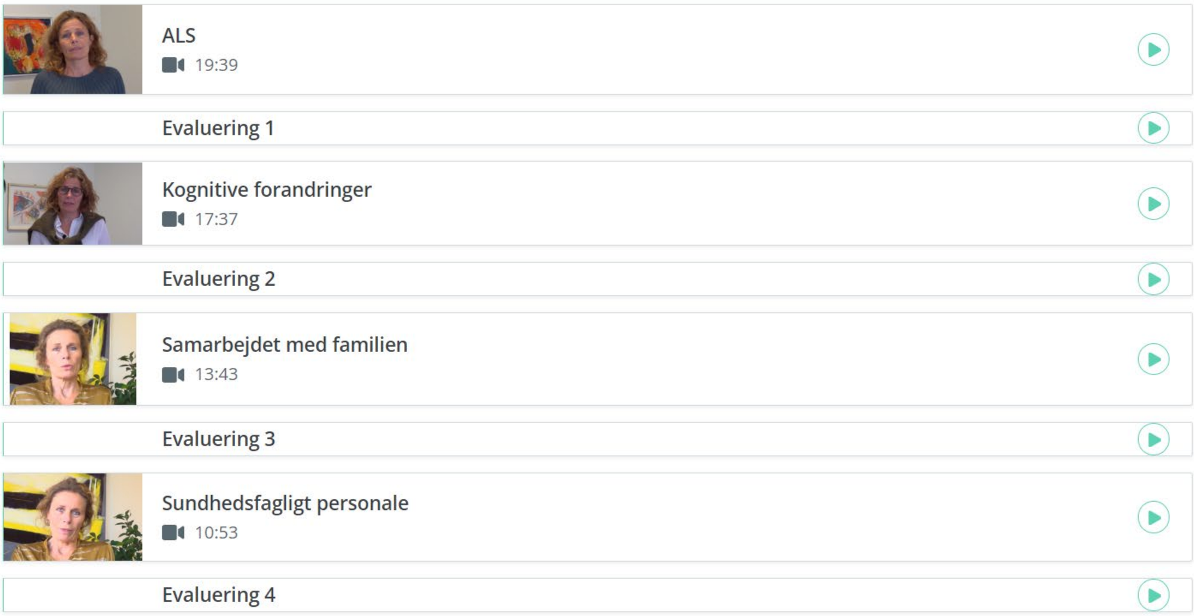
○ Sundhedsfagligt personale
10:53

○ Evaluering 4



I denne video møder du ALS-konsulent, sygeplejerske og familieterapeut Heidi With fra RehabiliteringsCenter for Muskelsvind. Heidi har arbejdet med familier ramt af ALS i mere end 15 år og har i den forbindelse et tæt samarbejde med mange kommuner. Videoen er en kort introduktion til sygdommen ALS, herunder hvilke symptomer den medfører, hvordan sygdommen udvikler sig samt hvorfor man ender med at dø af den.

Appendix 11. Simplero to professionals – overview of topics in the module



ALS
19:39

Evaluering 1

Kognitive forandringer
17:37

Evaluering 2

Samarbejdet med familien
13:43

Evaluering 3

Sundhedsfagligt personale
10:53

Evaluering 4

Appendix 12. Confirmation letter to the professionals enrolled in study I

RehabiliteringsCenter for Muskelsvind

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Kongsvang Allé 23
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Øst for Storebælt
Blekinge Boulevard 2, 1. sal
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Tlf. +45 8948 2222
Fax +45 8948 2212

www.rcfm.dk

Kære XXX

Tak fordi du har sagt ja til at deltage i 1. del af i et 3-årige ph.d.-projekt ved Rehabiliteringscenter for Muskelsvind (RCFM).

Projektets formål er at udvikle et **læringsprogram, som støtte til pårørende og sundhedsprofessionelle til personer med ALS og hvor vedkommende har ændret personlig/adfærd/sprog. Læringsprogrammet indeholder vigtig viden og råd til, hvordan man kan håndtere de daglige udfordringer og samtidig passe på sig selv i relationen til familier ramt af ALS og hvor vedkommende har ændret personlig/adfærd/sprog**.

Projektet har to dele

- Et lærings- og støtteprogram til pårørende til nulevende personer med ALS/ændret personlighed/adfærd/sprog.
- Et lærings- og støtteprogram til sundhedsprofessionelle i kommunerne.

Man ved at både pårørende og sundhedspersoner kan blive udfordret af kompleksiteten i sygdommene og vi ønsker derfor at understøtte begge grupper med en målrettet indsats.

Som sundhedsprofessionel, der arbejder tæt sammen med familier ramt af ALS og hvor vedkommende har ændret personlig/adfærd/sprog, har du en vigtig viden om at hverdagens udfordringer og samarbejdet med familien/r. Jeg ønsker med dit bidrag at få sundhedsprofessionelles erfaringer og oplevelser fra hverdagslivet, da denne viden skal anvendes til at udvikle og målrette støtteprogrammet til sundhedsprofessionelle, der står midt i disse udfordringer nu og i fremtiden. Dine erfaringer er således med til at hjælpe andre i den situation du selv står/stod i.

Viden og erfaringer fra samarbejdet og de arbejdsmæssige udfordringer sundhedsprofessionelle står overfor med familier ramt af disse to sygdomme vil forhåbentligt kvalificere RCFM's ydelser til pårørende og sundhedsprofessionelle på sigt.

RCFM samarbejder tværfagligt med brugeren, oftest i brugerens hjem, hvor der arbejdes på at identificere hvilke behov for rehabiliteringsindsatser og viden brugerne har. RCFM indtager en central position i det neuromuskulære faglige vidensfelt, og samarbejder i det tværsektorielle felt i hele Danmark med både hospitaler, kommuner og almen praksis. RCFM's indsats har ofte en afgørende betydning for hvilken rehabiliteringsindsats, der iværksættes hos den enkelte bruger og dermed brugerens muligheder for at klare sig i hverdagslivet med en neuromuskulær sygdom.

Når du deltager, vil jeg som nævnt bede dig om at skrive under på en samtykkeerklæring, hvori du samtykker til, at jeg må bruge data opnået i forbindelse med projektet. Det er helt frivilligt at deltage i projektet, og alle der deltager er anonyme. Man kan til enhver tid vælge ikke at deltage eller trække sit samtykke tilbage. Det er vigtigt at understrege at dit tilbud fra og samarbejde med RCFM på ingen mådes ændres, hvis du vælger at trække dit samtykke tilbage.

Appendix 12. Confirmation letter to the professionals enrolled in study I

Hvis du har nogle spørgsmål om projektet eller til mig er du meget velkommen til at kontakte mig på mail leol@rcfm.dk eller ringe til mig på tlf. 22652435

Jeg ser frem til at høre fra dig.

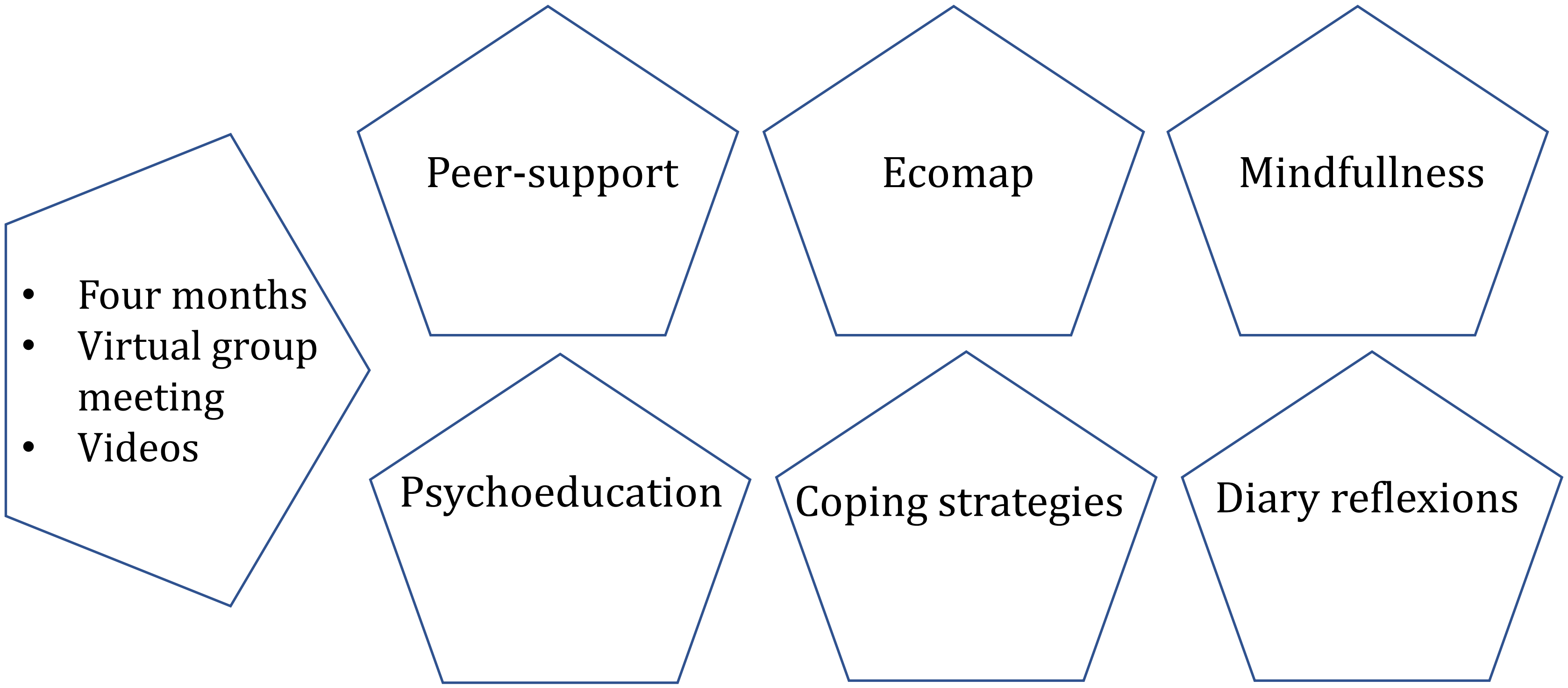
Med venlig hilsen

Lene Klem Olesen

Ph.d.-studerende, Cand.pæd.pæd.psyk og ergoterapeut

Mobil: 22652435
Mail: leol@rcfm.dk

Appendix 13. Evidence-based components in the EMBRACE intervention



Appendix 14. Intervention manual on the first virtual group meeting.

	Tid	Mål	Indhold/stikord	Form	Leveret/ikke leveret Og deres reaktioner på indholdet
Velkommen	10 min	Skabe tryghed og give deltagerne tid til at lande og føle sig mødt.	Præsentation af mig selv. Erfaring fra projekt 'Mere hverdag, mindre sygdom'. Hvordan er det at være landet her, trådt ind i et rum? Hvilke tanker er der tænkt? Hvordan er det at afsætte/prioritere tiden, er der ro? Hvor sidder du? Er der støj omkring dig.? Nervøs/spændt/glæde? Der kan være signaler jeg har svært ved at opfange, hvorfor jeg måske afstemmer med deltagerne løbende. Ikke altid øjenkontakt, skærmbillede/kamera placeret forskelligt. Her må man være som man er. Et rum for **jer**, med fokus på **jer**. Hvorfor jeg af og til vil bringe jer tilbage på sporet af jer selv. Hvad kan deltagerne forvente af i dag.	Gruppefacilitator Heidi With (HW) taler, med fokus på at beskrive modsatrettede følelser Program: (delt skærm) Velkommen Kort information om projektet v/ projektleder Lene Klem Olesen (LKO) Hvordan er vi sammen med respekt for hinanden. 'Tavshedspligt'/fortroligt rum Præsentation Netværksbillede Dagbog Hvad sker der på de næste møder (Simplero) Hvordan har det været for dig at deltage.	
Info om projektet	5 min	Sikre at deltagerne er fuldt oplyste om at de deltager i forskningsprojekt	Beskrivelse af projektet Information om støttetilbuddet til sundhedsprofessionelle og at deltagerne modtager en samtykkeformular til PALS ift.	LKO informerer	

			kontakt til kommune/bureauer (Annulleret pga Corona) Møde optages LKO lytter med, men deltager ikke. Tekniske eller formelle spørgsmål vedr. projektet og programmet rettes til LKO Deltagerne skal selv lave en kontaktliste over oplysninger, hvis de ønsker at dele deres mail og telefonnummer med de andre – kan foregå via chatten.		
Hvad har I til fælles og hvordan er vi der bedst for os selv og hinanden.	10 min	At give deltagerne en mulighed for at danne sig et overblik over hvem de er i gruppen, som leder hen imod formen at præsentere sig på.	Beskrive hvad deltagerne har tilfælles (fælles tredje) og modsat, med alt det der er forskelligt. Vigtigheden af at rumme hinandens forskelligheder. Tydelige opfordringer til hvordan vi er sammen med det vi bringer ind i gruppen. Hvordan er deltagerne og meddeltagernes rolle forskellige.		
Præsentation	45 min	Målet er at der allerede ved præsentation skabes en spejling og startende gruppefølelse., samt at det bliver klart hvordan rollerne er fordelt mellem deltagere og meddeltagere. En oplevelse af at der er plads til at være mig. Den enkelte deltager skaber en plads til sig selv, via sin præsentation. For at sikre deltager/meddeltager ikke er usikre på deres roller	Deltagerne præsenterer sig Navn By Hvem bor du sammen med Hvor længe har ALS/CIs været en del af din hverdag. (Afgrænsning af at sygdomshistorik ikke får plads her) Hvad laver du, når du ikke er hjemme. Hvem har du evt. taget med. Hvad har fået dig til at deltage i FAVN/EMBRACE Meddeltagerne præsenterer sig (barn) Navn By	5 min til hver deltager.	

			Hvad har fået dig til at takke ja til at deltage i FAVN/EMBRACE Meddeltagerne præsenterer sig(veninde/nabo) Navn By Relation Hvordan er det for dig at være inviteret med. Evt. Spørgsmål til rollefordelingen mellem deltager og meddeltager. Veninde/voksent barn.		
Netværksbillede	20 min	At give deltagerne et visuelt overblik over deres netværk, så de får et billede af, hvor de kan hente ressourcer henne og et billede af, hvor de måske bruger unødvendige ressourcer. Og et ønske om at give dem forståelsen af at kunne påvirke de ressourcer, der er i netværket i en positiv retning.	Hvorfor lave netværksbillede. Der henvises til de 4 ark i dagbogen. Der pointeres at centrum er den pårørende og ikke dem som par. Der tegnes et netværksbillede på mødet, ved at dele skærmbillede. Og hvor instruktionen af tegnemulighederne giver undervejs af HW. Der gives 5 min til at starte eget netværksbillede Opfordres til at lave det færdigt til næste gang, hvor det er frivilligt at dele, men der tales om, hvordan det var at lave det. Høre deltagernes tanker om opgaven/muligheden	Fotoeksempel af netværksbillede.	
Dagbog	5 min	At skabe en bevidst refleksion over hverdagen, dens udfordringer og de tanker og følelser den vækker. At give deltagerne en mulighed for at udtrykke sig på skrift med henblik på at tydeliggøre tanker og følelser omkring rollen som pårørende til en person med ALS/CI. Gennem dagbogsrefleksioner øges fokus og begribeligheden af de oplevelser hverdagen byder og dermed åbner en mulighed for at kunne agere på	Præcisere at det er deres personlige bog, der ikke skal deles med nogen. Kan måske hjælpe med til at se hvilken udvikling de selv gennemgår, eller ændret blik og tilgang til nogen problematikker. Høre deltagernes tanker om opgaven/muligheden.	Vise dagbogen frem.	

		de ting man oplever. Ligeledes giver det deltagerne en mulighed for at lave et tilbageblik på hvilken proces/udvikling de har været igennem			
Chatrum	5 min	At informere deltagerne om muligheden for at mødes i chatten mellem møderne og skabe et rum for de pårørende, hvor de kan blive mødt og forstået	Henlede til introduktionsvideo i Simplero, der viser hvordan chatten fungerer Muligt at stille spørgsmål til HW	Deltagerne kan i Simplero se en video, der kort præsenterer chatrummet og hvordan man opretter og kommenterer på beskeder.	
Hvad kan I forvente af de næste møder	5 min	Hjælpe deltagerne til at holde fokus på dem selv. Give dem mulighed for at forberede sig til næste møder	Om to timer vil du modtage en mail, der giver adgang til Simplero platformen, hvor du vil blive præsenteret for diverse film. På næste møde vil der være fokus på kognitive forandringer og når det er svært at tale sammen. Er der særlige behov eller ideer fra deltagerne, som jeg skal tage med eller være opmærksom på.		
Hvordan har det været? Take away	15 min	At få deltagerne til at forholde sig til hvad de tager med fra mødet, samtidig med at de via hinanden får et resume af indholdet.	Hvad tager du med dig fra mødet i dag, hvad gjorde indtryk? Hvordan har det været at deltage? 2 min til hver deltager HW afslutter med personlig melding	Power point, minimeret som dele skærm.	

Appendix 15. Example of the ECOMAP exercise

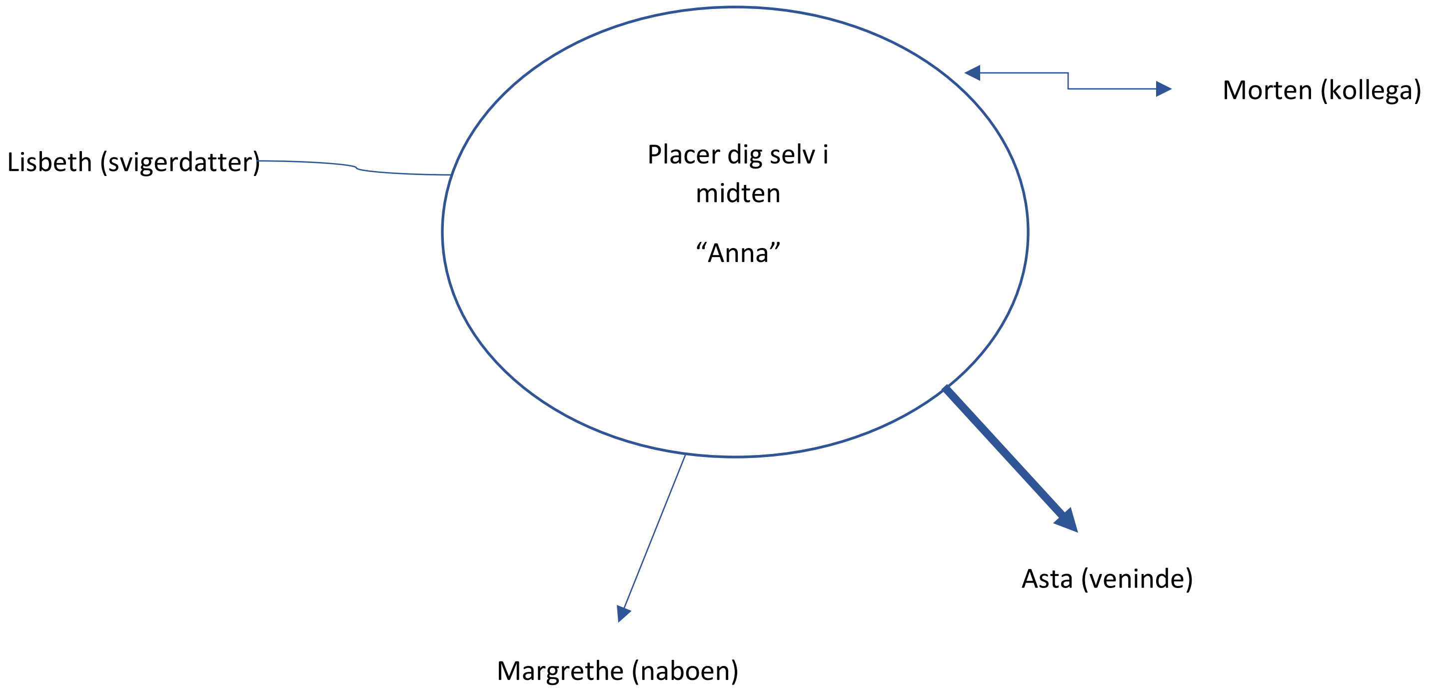
Tæt og god relation = ➡

Du giver omsorg for person = →

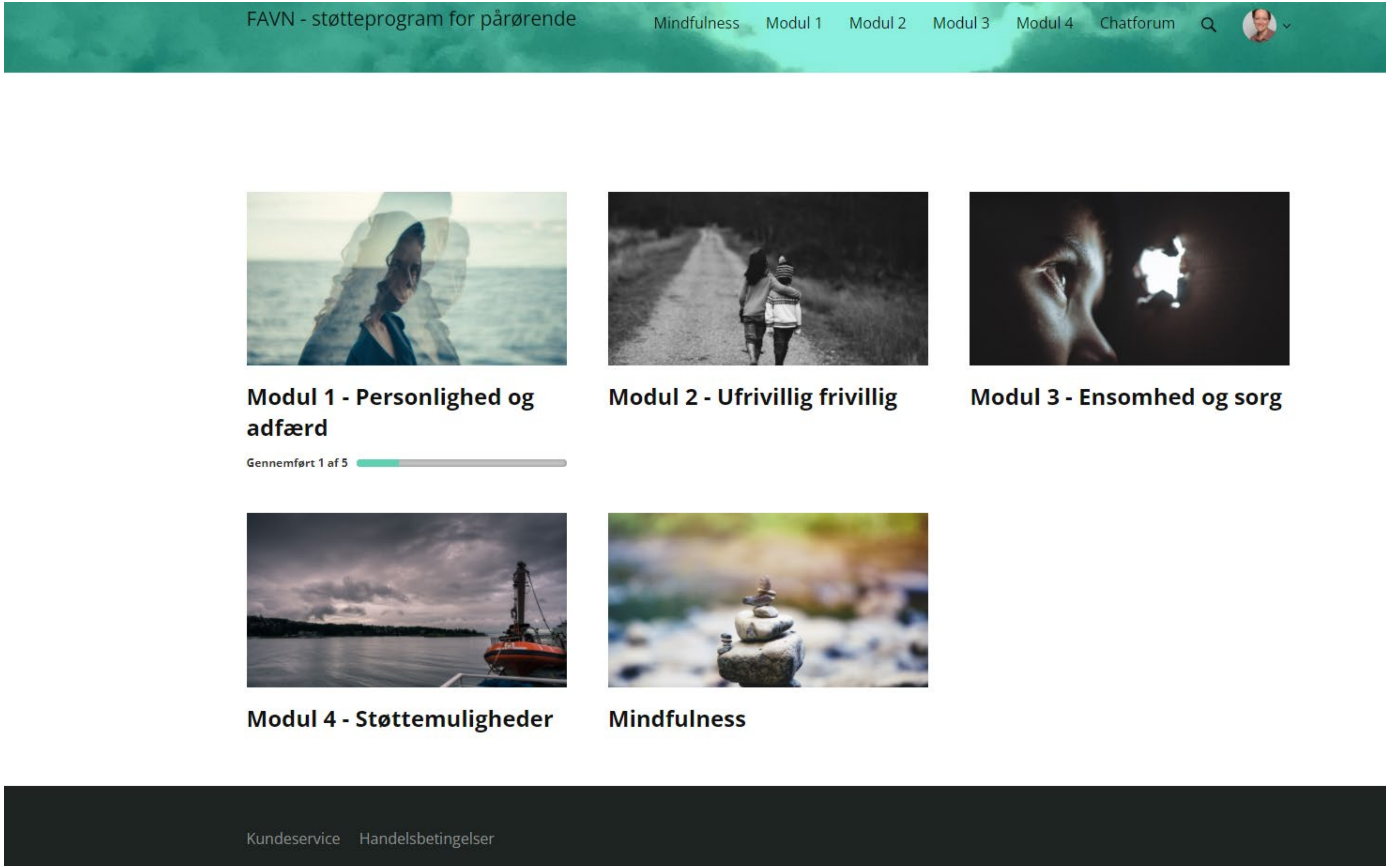
Omsorgen går begge veje = ↔

Relation er udfordrende = ~

EKSEMPEL PÅ ECOMAP



Appendix 16. Simplero – overview of the four modules, including mindfulness



FAVN - støtteprogram for pårørende

Modul 1 - Personlighed og adfærd

Gennemført 1 af 5

Modul 2 - Ufrivillig frivillig

Modul 3 - Ensomhed og sorg

Modul 4 - Støttemuligheder

Mindfulness

Appendix 17. Simplero – overview of module 1, including the topic presented in videos

FAVN - støtteprogram for pårørende

Modul 1 - Personlighed og adfærd

1 af 5 lektioner afsluttet

Kursusindhold

Modul 1 - Personlighed og adfærd 1 / 5

- ○ Introduktion til støtteprogrammet 7:02
- ○ Introduktion til chat 6:01
- ○ Pårørendeambassadører 1:00
- ✓ **Forandringer i personlighed og adfærd 31:52**
- ○ Når det er svært at tale sammen 14:00

Forandringer i personlighed og adfærd

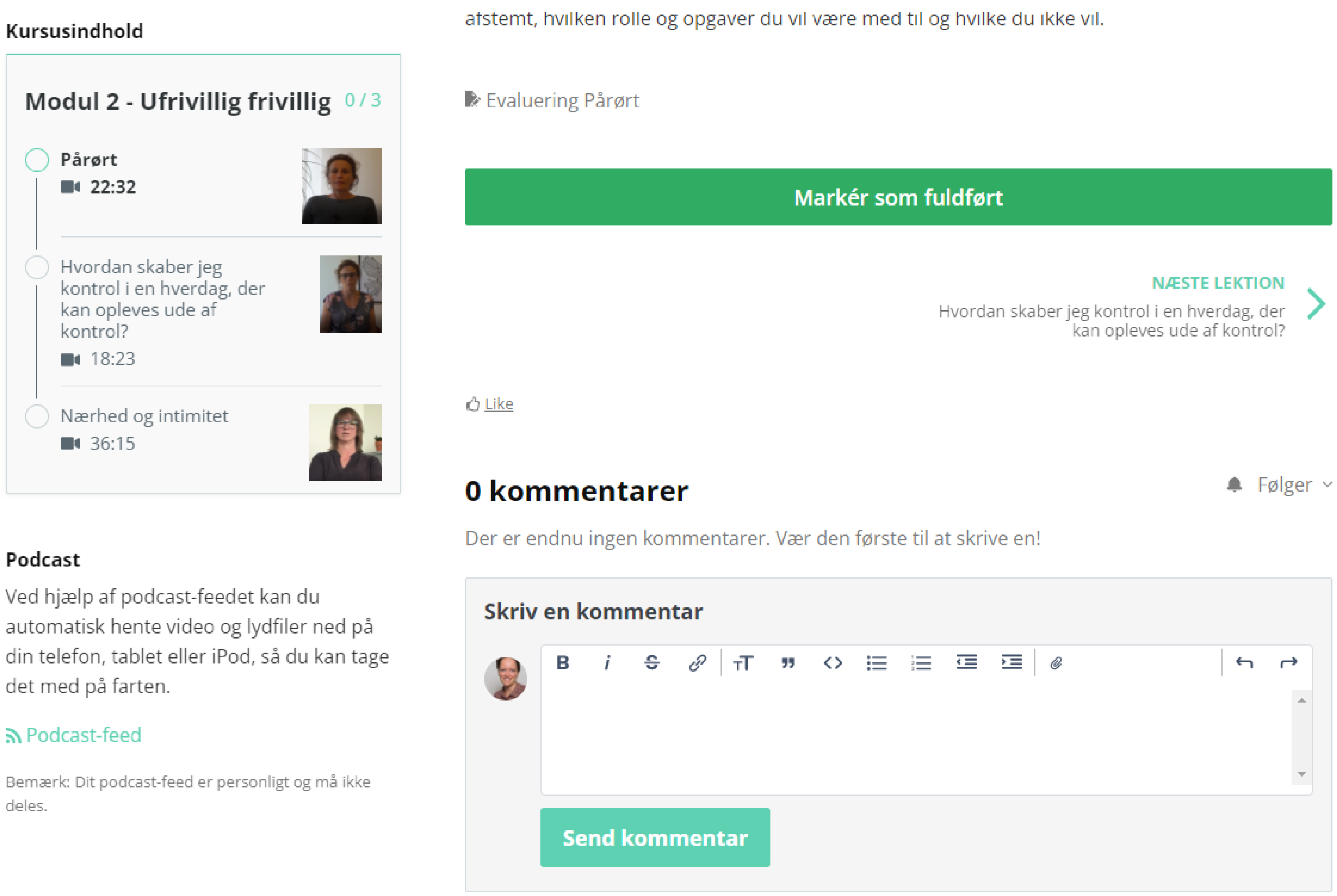
I denne video taler Heidi om kognitive forandringer i forbindelse med ALS. Der er her tale om nogle særlige forandringer i hjernen, som kan påvirke personligheden, adfærden og sproget.

Du vil undervejs i videoen opleve, at nogle symptomer er genkendelige, mens andre er ukendte.

Podcast

Ved hjælp af podcast-feedet kan du

Appendix 18. Simplero – overview of how to mark a video as completed and write comments



Kursusindhold

Modul 2 - Ufrivillig frivillig 0 / 3

- ○ **Pårørt** 22:32
- ○ Hvordan skaber jeg kontrol i en hverdag, der kan opleves ude af kontrol? 18:23
- ○ Nærhed og intimitet 36:15

Podcast

Ved hjælp af podcast-feedet kan du automatisk hente video og lydfiler ned på din telefon, tablet eller iPod, så du kan tage det med på farten.

Podcast-feed

Bemærk: Dit podcast-feed er personligt og må ikke deles.

afstemt, hvilken rolle og opgaver du vil være med til og hvilke du ikke vil.

Evaluering Pårørt

NÆSTE LEKTION

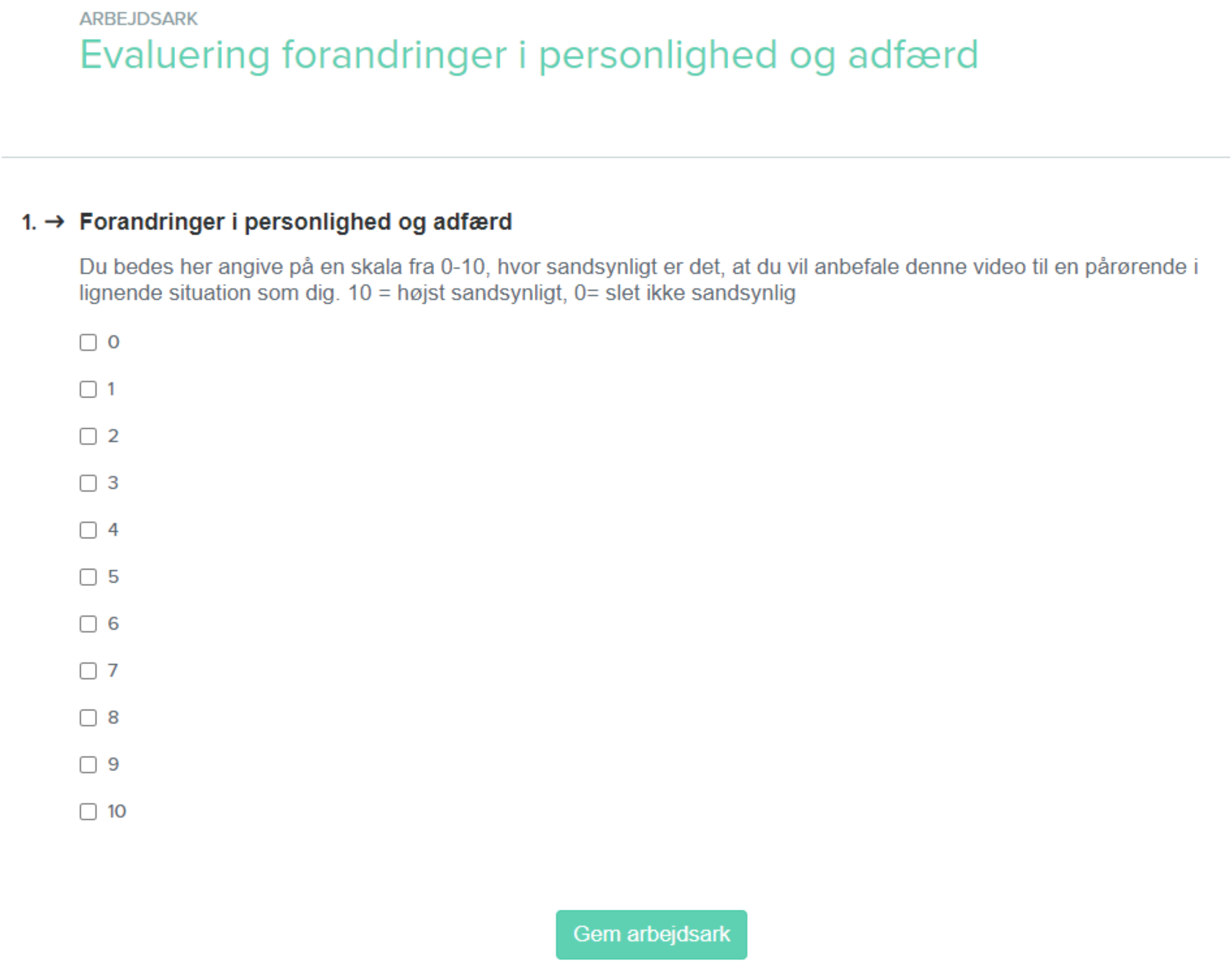
Hvordan skaber jeg kontrol i en hverdag, der kan opleves ude af kontrol?

0 kommentarer

Der er endnu ingen kommentarer. Vær den første til at skrive en!

Skriv en kommentar

Appendix 19. Simplero – overview of the evaluation scale for each video watched



ARBEJDSARK

Evaluering forandringer i personlighed og adfærd

1. → **Forandringer i personlighed og adfærd**

Du bedes her angive på en skala fra 0-10, hvor sandsynligt er det, at du vil anbefale denne video til en pårørende i lignende situation som dig. 10 = højst sandsynligt, 0= slet ikke sandsynlig

- ☐ 0
- ☐ 1
- ☐ 2
- ☐ 3
- ☐ 4
- ☐ 5
- ☐ 6
- ☐ 7
- ☐ 8
- ☐ 9
- ☐ 10

Appendix 20. Simplero – overview of the chat forum

Chatforum

Velkommen til et lille lukket forum, hvor du har mulighed for at udveksle erfaringer med andre i samme situation som dig - ordet er frit, men forventer naturligvis, at vi alle udviser gensidig respekt for hinanden og holder en god tone :-)

Velkommen

Kære deltager

Her har du mulighed for at skrive, hvad der måtte fylde for dig, i forhold til det at være pårørende. Det kan både være gode råd, nye ideer, spørgsmål til de andre i gruppen, eller det at dele en tanke med de andre. Jeg vil opfordre til, at der også her, holdes et særligt fokus på at "snakken" er med udgangspunkt i jer som pårørende.

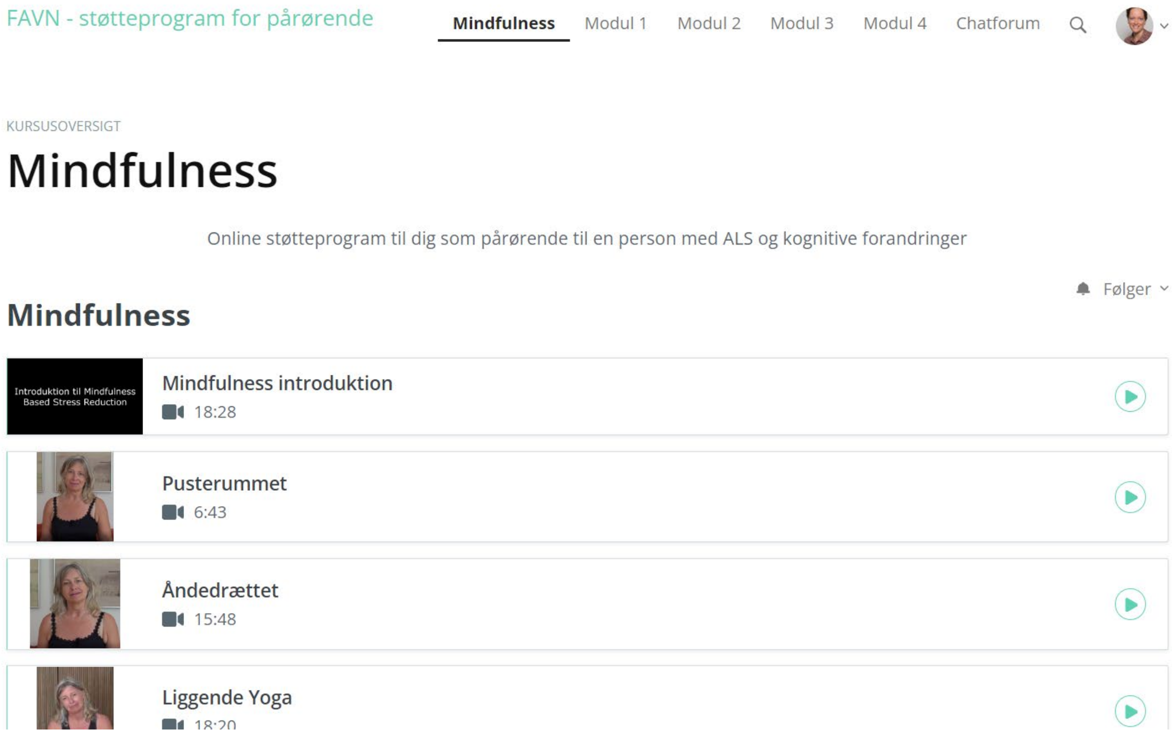
Som gruppeleder vil jeg følge med her på chatten, for at få en fornemmelse af hvad der fylder, har interesse og måske hvad der er særligt behov for.

Du ...

Læs mere

Af Heidi With den 14 okt '20 08:22 i Åbent forum · 0 kommentarer

Appendix 21. Simplero – Mindfulness-based stress reduction (MBSR)



KURSUSOVERSIGT

Mindfulness

Online støtteprogram til dig som pårørende til en person med ALS og kognitive forandringer

Mindfulness

Mindfulness introduktion
18:28

Pusterummet
6:43

Åndedrættet
15:48

Liggende Yoga
18:20

Appendix 22. Introduction to the diary

Kære Navn

Denne dagbog er sendt til dig som pårørende, der deltager i støtteprogrammet FAVN.

Dagbogen er tænkt som en bog du fx kan have liggende ved din side, når du ser de film, der løbende vil blive tilgængelige for dig via platformen Simplero. Ligeledes vil du med fordel kunne have dagbogen fremme til de virtuelle gruppemøder, evt. for at kigge tilbage og se hvad du har noteret eller til at tage notater i under mødet. Du er naturligvis også velkommen til at gøre brug af dagbogen på andre tidspunkter.

Hvordan du bruger dagbogen, er helt valgfrit. Om det er noter fra det du hører og ser, tanker om din egen situation, spørgsmål, opgaver du skal huske, om det er via tegninger, lange sætninger, stikord eller udklip er alt sammen op til dig.

I bogen er ilagt 4 ark papirer, som jeg vil introducere dig til ved det første virtuelle gruppemøde mandag d. 26/10 kl. 14:30

Måske du vil finde det brugbart, et stykke henne i forløbet at se tilbage på, hvad der fyldte for dig, da du startede i FAVN sammenlignet med, hvad du er optaget af i slutning af støtteprogrammet.

Vigtigt er det, at dagbogen ikke kommer til at føles som en byrde. Formålet er, at levne en plads og et rum for dig og dine tanker, et sted hvor du kan notere, tegne eller indsætte det som kan være hjælpsomt for dig.

Med venlig hilsen

Heidi With

Appendix 23. The EMBRACE program

FAVN

4 måneders støtte-program til pårørende

2. november kl. 14.30-16.30
Velkomst og introduktion

- Ændring af personlighed
- Når det er svært at tale sammen

23. november kl. 14.30-16.30
Opsamling og erfaringsudveksling

- PÅRØRT
- Hvordan skaber jeg kontrol i en hverdag, der kan opleves ude af kontrol?
- Nærvær & intimitet

14. december kl. 14.30-16.30
Opsamling og erfaringsudveksling

- Tab, sorg & tabuer
- Hvad skal foreviges?
- Den sidste tid og fremtiden
- Hvordan sikres fremtiden bedst muligt?
- Hvad skal jeg håbe?

18. januar kl. 14.30-16.30
Opsamling og erfaringsudveksling

- Hvor kan jeg få støtte og hjælp?

22. februar kl. 14.30-16.30
Afrunding

RehabiliteringsCenter
for Muskelsvind

Appendix 24. Amyotrophic lateral sclerosis - Frontotemporal dementia - questionnaire

Amyotrofisk lateral sclerose - Frontotemporal demens - Spørgeskema (ALS-FTD-Q)

I det følgende spørgeskema vil din partners, dit familiemedlems eller din vens adfærd blive evalueret. Hvor der står “ham” eller “hans”, kan det til enhver erstattes med “hende” eller “hendes”. Det tager ca. 10 minutter at udfylde spørgeskemaet, og det skal helst foregå i et lokale, hvor patienten ikke er til stede. Spørgsmål besvares ved at afkrydse i feltet ud for det svar, der passer. Spørgeskemaet består af to dele, A og B.

Dato . . - . . - . . . .   Udfyldt af (fx partner, søskende, barn) . . . .

Patientens navn . . . .

Patientens fødselsdato . . - . . - . . . .   Patientens køn   M / K

Patientens højest gennemførte uddannelsesniveau . . . .

Del A De følgende 13 udsagn sammenligner din partners ***nuværende*** adfærd med hans adfærd ***for tre år siden***.

Der er følgende svarmuligheder:
- helt uenig
- overvejende uenig
- overvejende enig
- helt enig

	Helt uenig	**Overvejende uenig**	**Overvejende enig**	**Helt enig**
1. Din partner er mindre interesseret i sine omgivelser	☐	☐	☐	☐
2. Din partner går mindre op i sin personlige hygiejne	☐	☐	☐	☐
3. Din partner sætter oftere sig selv først	☐	☐	☐	☐
4. Din partner bliver lettere irriteret eller vred	☐	☐	☐	☐
5. Din partners koncentrationsevne er blevet dårligere	☐	☐	☐	☐
6. Din partners adfærd er mere rastløs	☐	☐	☐	☐
7. Din partner er blevet mere indesluttet	☐	☐	☐	☐
8. Din partners aktiviteter forekommer mere planløse	☐	☐	☐	☐
9. Din partner har flere problemer med hukommelsen	☐	☐	☐	☐
10. Din partner henvender sig oftere til fremmede	☐	☐	☐	☐
11. Din partner har øget lyst til sex (Undlad at svare, hvis spørgsmålet ikke er relevant)	☐	☐	☐	☐

Nogle ALS-patienter oplever tvungen latter eller gråd uden en logisk grund.
De følgende to udsagn beskriver IKKE dette fænomen.
De beskriver generelle, følelsesmæssige ændringer hos din partner.

	Helt uenig	**Overvejende uenig**	**Overvejende enig**	**Helt enig**
12. Din partner er ikke så følelsesmæssig stabil	☐	☐	☐	☐
13. Din partner er oftere ekstremt glad	☐	☐	☐	☐

Del B De følgende 12 udsagn omhandler din partners adfærd i ***den seneste måned.*** Bemærk! Nogle udsagn beskriver normal adfærd, mens andre beskriver unormal adfærd. Læs derfor udsagnet grundigt, inden du svarer.

Du har følgende svarmuligheder:
- Aldrig
- Nogle gange
- Ofte
- Altid

	Aldrig	**Nogle gange**	**Ofte**	**Altid**
14. Din partner er mistænksom	☐	☐	☐	☐
15. Din partner gentager de samme håndbevægelser eller sætninger	☐	☐	☐	☐
16. Din partner føler ikke skam	☐	☐	☐	☐
17. Din partner ved, hvor han befinder sig	☐	☐	☐	☐
18. Din partner udviser fornærmende adfærd	☐	☐	☐	☐
19. Din partner er god til at bedømme en situation	☐	☐	☐	☐
20. Din partner hamstrer mad eller er stærkt optaget af mad	☐	☐	☐	☐
21. Din partner forstår, hvad hans sygdom indebærer	☐	☐	☐	☐
22. Din partner ser eller hører ting, som ikke er der	☐	☐	☐	☐
23. Din partner udviser barnlig adfærd	☐	☐	☐	☐
24. Din partner ved, hvilken tid på dagen det er	☐	☐	☐	☐
25. Din partner efterligner dig eller andre	☐	☐	☐	☐

Scoring af ALS-FTD-Q

Spørgsmål 1-13:

	Helt uenig	Overvejende uenig	Overvejende enig	Helt enig
Værdi =	0	1	3	4

Spørgsmål 14, 15, 16, 18, 20, 22, 23 og 25:

	Aldrig	Nogle gange	Ofte	Altid
Værdi =	0	1	3	4

Spørgsmål 17, 19, 21 og 24:

	Aldrig	Nogle gange	Ofte	Altid
Værdi =	4	3	1	0

ALS-FTD-Q-score: score for hvert spørgsmål lægges sammen for at beregne ALS-FTD-Q-scoren.

Cut-off- værdier: milde ædfærdsforstyrrelser ≥ 22

svære adfærdsforstyrrelser (inden for bvFTD-området) ≥ 29

Disse er foreløbige cut-off-værdier, der afventer fremtidig validering

Appendix 25. Invitation to the EMBRACE intervention

RehabiliteringsCenter for Muskelsvind

Vest for Storebælt
Kongsvang Allé 23
DK - 8000 Århus C

Øst for Storebælt
Blekinge Boulevard 2, 1. sal
DK - 2630 Taastrup

Tlf. +45 8948 2222
Fax +45 8948 2212

www.rcfm.dk

Xxxx xxxxx
Xxxxxxx
xxxxxxxxxxxxxxxxx

FAVN – Et online støtteprogram for pårørende til personer med ALS og kognitive forandringer

Jeg henvender mig til jer, da jeg er i gang med et ph.d.-projekt ved Rehabiliteringscenter for Muskelsvind (RCFM). Projektet har fokus på pårørende til personer med ALS, hvor den sygdomsramte også har kognitive forandringer (forandret personlighed, adfærd og/eller sprog).

Formålet med projektet er at udvikle et målrettet **støtteprogram til pårørende til personer med ALS og kognitive forandringer. Støtteprogrammet indeholder vigtig viden og råd til, hvordan man som pårørende kan håndtere de daglige udfordringer, og samtidig finde en mere skånsom og mindre belastende vej i sygdomsforløbet**.

Forskning viser, at pårørende til personer med ALS og kognitive forandringer kan blive særligt udfordret og belastet af kompleksiteten i sygdommene og vi ønsker derfor at understøtte pårørende med en målrettet online støtteindsats.

Tilbuddet er til dig som pårørende
Støtteprogrammet foregår online og løber over fire måneder, **fra uge 44 i 2020 til uge 9 i 2021**. Dem der deltager, vil få tilsendt en række målrettede film samt en invitation til at deltage i fem online gruppemøder med 5-6 andre pårørende, der står i lignende situation som dig. Gruppemøderne guides af ALS-konsulent, sygeplejerske og familieterapeut Heidi With, som har 14 års erfaring i samarbejdet med familier med ALS.
Du har mulighed for at invitere et andet familiemedlem eller ven med til at deltage sammen med dig igennem hele støtteprogrammet, dog ikke den sygdomsramte.

Gruppemøderne ligger fast om eftermiddagen hver 3. uge og varer to timer. Dato for de fem gruppemøder vil blive tilsendt efter d. 29.9.20. I ugerne mellem videomøderne vil du få tilsendt film à max 25 minutters varighed, som du kan vælge at se, hvor og hvornår det passer dig. Filmene indeholder forskellige emner, som kan være informative og hjælpsomme til dig som pårørende. Du kan se emnerne på folderen der ligger med i brevet.

I forbindelse med evalueringen af projektet, vil du som deltager du blive bedt om at udfylde to korte spørgeskemaer før og efter programmet. Ligesom du evt. vil blive inviteret til et individuelt interview. Efter hver set film vil du blive bedt om at score filmens relevans ud fra 1 spørgsmål. Gruppemøderne vil blive optaget og selve optagelserne vil indgå som en del dataindsamlingen. Optagelserne ses kun af forskningsgruppen og slettes, når de ikke længere skal bruges.
Deltagelse i projektet forudsætter, at man har en smartphone, tablet eller computer samt at man har internetforbindelse.

Formålet er at du via støtteprogrammet, får et fleksibelt og trygt rum, hvor du opleve at blive støttet og styrket i sygdomsforløbet. Viden og erfaring fra projektet vil efterfølgende blive anvendt til at hjælpe andre pårørende, der kommer til at stå i lignende situation.

Appendix 25. Information to the EMBRACE intervention

Projektet er anmeldt til Videnskabsetisk Komité og Styrelsen for Patientsikkerhed. Når du deltager, vil jeg bede dig om at skrive under på en samtykkeerklæring, hvori du samtykker til, at jeg må bruge data opnået i forbindelse med projektet. Det er frivilligt at deltage i projektet, og alle der deltager er anonyme. Du kan til enhver tid vælge ikke at deltage eller trække dit samtykke tilbage.

Hvis du har nogle spørgsmål til projektet eller ønsker du at deltage er du meget velkommen til at kontakte mig på nedenstående kontaktoplysninger eller **Heidi With** på hewi@rcfm.dk /tlf. 22652436 **senest den 21.9.20.** Vi vil herefter aftale et tidspunkt, hvor vi kan ringe dig op og drøfte, hvilke udfordringer du oplever som pårørende.

Du vil senest få besked **den 23.9.20** om, hvorvidt vi vurderer at tilbuddet dækker dine behov.

Jeg håber meget, at du har lyst til at deltage, og ser frem til at høre fra dig.

Med venlig hilsen og forhåbentlig gensyn

Lene Klem Olesen
Ph.d.-studerende, Cand.pæd.pæd.psyk, ergoterapeut og ALS-konsulent
RehabiliteringsCenter for Muskelsvind og Aarhus Universitet
Mobil: 22652435
Mail: leol@rcfm.dk

Appendix 26. Welcome letter to the caregivers enrolled in the EMBRACE intervention

Aarhus d. 29/9-20

Kære XXXX

Vi takker for din interesse i støtteprogrammet FAVN, og er glade for at kunne bekræfte at du er inkluderet i projektet og dermed er deltager i FAVN.

Du vil være en del af en gruppe på 4-5 pårørende, som ligeledes har en partner der har ALS og er mentalt forandret. På de virtuelle møder vil Heidi With deltage som gruppeleder og være medvirkende til at mødet forløber bedst muligt.

Fokus på dig som pårørende

Det at være pårørende kan for mange være en stor belastning, bl.a. når fokus ofte hviler på den sygdomsramte, og man som pårørende ikke kan finde overskud eller tid til at have fokus på egne behov.

I projekt Favn vil fokus være på dig som pårørende.

Du vil blive støttet i at samtaler tager udgangspunkt i dig som pårørende. Hvordan det er at være dig, hvordan du finder vej i hverdagen, hvad du kan have brug for, hvilke tanker og bekymringer du går med, hvilke tanker du gør dig om fremtiden, etc. Det kan være sårbart og uvant at have fokus på sig selv, og samtalen ledes let hen på den sygdomsramte. Som gruppeleder vil jeg hjælpe med til, at du sammen med resten af gruppen, holder fast i et måske tiltrængt fokus på dig selv.

Første virtuelle møde foregår xxxxxdag d. kl. 14:30 – 16:30 (se vedhæftede oversigt)

Få dage forinden får du tilsendt et link på din mail, så du på dagen kan tilgå Teams. Teams er den forbindelse vi bruger til at afholde vores virtuelle møder.

Simpleo

Efter det første virtuelle møde vil du på din mail få tilsendt et log in til platformen Simpleo.

På Simpleo kan du tilgå små film, med relevante temaer, som danner grundlag for snakke til næste virtuelle møde. Desuden er der en chatfunktion på Simpleo, der gør det muligt for dig, at skrive sammen med de andre gruppedeltager i et lukket forum.

Meddeltager

Som beskrevet i tidligere udsendte invitationen, har du mulighed for at invitere en ven eller et familiemedlem med til at deltage i de virtuelle møder, dog ikke din ALS-ramte partner.

Appendix 26. Welcome letter for the caregivers enrolled in the EMBRACE intervention

Vi har vedhæftet et brev, som du skal læse og herefter udlevere til den meddeltager du evt. har valgt skal være med til de virtuelle møder. Hvilket naturligvis er frivilligt. Det skal være den samme person der er med hver gang. Altså er det ikke muligt at have forskellige meddeltagere med fra møde til møde. Vi vil gerne have oplyst navn og relation på evt. meddeltager inden mødestart.

Dagbog

Inden mødestart vil du med posten, få tilsendt en dagbog som er din. I dagbogen er der et forord der informerer dig om hvad du har mulighed for at bruge dagbogen til, så længe du deltager i FAVN.

Har du tekniske problemer, spørgsmål eller kommentarer til ovenstående er du meget velkommen til at henvende dig til os.

Med venlig hilsen

Lene Klem Olesen, Ph.d.- studerende, mail leol@rcfm.dk tlf. 22652435
Heidi With, projektansat, ALS-konsulent og familieterapeut, mail hewi@rcfm.dk tlf. 22652436

Appendix 27. Information letter for the co-participant

Til dig som meddeltager

FAVN

4 måneders støtteprogram til pårørende.

Du er af en ven eller et familiemedlem blevet inviteret til at være meddeltager i et støtteprogram, for pårørende til en partner med ALS og mentale forandringer, kaldet FAVN.

Forskning viser, at pårørende til personer med ALS og mentale forandringer, kan blive særligt udfordret og belastet af kompleksiteten i sygdommene, og vi ønsker derfor at understøtte pårørende med en målrettet online indsats.

Programmet FAVN består af 5 virtuelle møder med 6-7 andre pårørende/meddeltagere i lignende situation. Derudover vil den pårørende, løbende få tilsendte korte film med relevante temaer.

De nærmere oplysninger om hvad FAVN indeholder og tilbyder, vil du have mulighed for at spørge ind til, hos den person, der har inviteret dig til at være meddeltager.

Vi ved fra tidligere undersøgelser, at deltagere i lignende tilbud, kan opleve en form for ensomhed samt et behov for, i hverdagen, at dele de oplevelser der har fundet sted i forbindelse med deltagelse i et støtteprogram. Derfor giver vi mulighed for, at hver deltager i støtteprogrammet FAVN, kan invitere en person til at følge processen, hvilket i dette tilfælde er dig. Velkommen.

Som meddeltager har du en rolle som ”vidne”, til de snakke og oplevelser der foregår i gruppen. På den måde kan du være en sparringspartner og støtte for den pårørende. Det vil være de pårørende der er primære deltagere, du som meddeltager vil have en “mindre stemme” i gruppen.

Det at have været til det samme møde, efterfølgende udveksle tanker og refleksioner og evt. fortsætte snakken, kan i nogle tilfælde mindske ensomhedsfølelsen og dermed belastningen hos den pårørende.

Vi forventer at din meddeltagelse foregår med respekt for de følsomme emner, der kan blive delt på de virtuelle møder, og at indholdet af samtalerne bliver mellem deltagerne og dig som meddeltagere.

Har du spørgsmål til ovenstående, er du meget velkommen til at kontakte undertegnede.

Med venlig hilsen

Lene Klem Olesen, Ph.d.- studerende, mail leol@rcfm.dk tlf. 22652435
Heidi With, projektansat, ALS-konsulent og familie- og psykoterapeut, mail hewi@rcfm.dk tlf. 22652436

DET VIDENSKABSETISKE KOMITÉSYSTEM

(S1)

Informeret samtykke til deltagelse i et sundhedsvidenskabeligt forskningsprojekt.

Forskningsprojektets titel: FAVN - Et komplekst interventionsstudie med et online palliativt rehabiliteringsprogram til støtte for pårørende og fagpersoner til personer med ALS og kognitive forandringer

Erklæring fra forsøgspersonen:

Jeg har fået skriftlig og mundtlig information og jeg ved nok om formål, metode, fordele og ulemper til at sige ja til at deltage.

Jeg ved, at det er frivilligt at deltage, og at jeg altid kan trække mit samtykke tilbage uden at miste mine nuværende eller fremtidige rettigheder til behandling.

Jeg giver samtykke til, at deltage i forskningsprojektet, og har fået en kopi af dette samtykkeark samt en kopi af den skriftlige information om projektet til eget brug.

Forsøgspersonens navn: ____

Dato: _______________ Underskrift: ____

Ønsker du at blive informeret om forskningsprojektets resultat samt eventuelle konsekvenser for dig?:

Ja _____ (sæt x) Nej _____ (sæt x)

Erklæring fra den, der afgiver information:

Jeg erklærer, at forsøgspersonen har modtaget mundtlig og skriftlig information om forsøget.

Efter min overbevisning er der givet tilstrækkelig information til, at der kan træffes beslutning om deltagelse i forsøget.

Navnet på den, der har afgivet information: Lene Klem Olesen

Dato: _______________ Underskrift: ____

Projektidentifikation: (Fx komiteens Projekt-ID, EudraCT nr., versions nr./dato eller lign.)

1-10-72-1-19