Advance Care Planning in Amyotrophic Lateral Sclerosis and other chronic life-limiting conditions: patients' and family carers' perspectives

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ADVANCE CARE PLANNING IN AMYOTROPHIC LATERAL SCLEROSIS AND OTHER CHRONIC LIFE-LIMITING CONDITIONS

patients' and family carers' perspectives

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Nothing in life is to be feared, it is only to be understood.
Now is the time to understand more, so that we may fear less.
Marie Curie

Content

List of chapters

Chapters 1-5 are based on the following publications

Chapter 1:

Vandenbogaerde I, Miranda R, De Bleecker JL, Carduff E, van der Heide A, Van den Block L, Deliens L, De Vleminck A. Advance care planning in amyotrophic lateral sclerosis (ALS): study protocol for a qualitative longitudinal study with persons with ALS and their family carers. BMJ Open. 2022 May 12;12(5):e060451. doi: 10.1136/bmjopen-2021-060451. PMID: 35551085 [2021 IF 3.006; Ranking Q2; ranking 85/172 MEDICINE, GENERAL & INTERNAL]

Chapter 2:

Vandenbogaerde I, Van den Block L, Deliens L, Carduff E, van der Heide A, De Bleecker J, De Vleminck A. Experiences with advance care planning in Amyotrophic Lateral Sclerosis (ALS): Qualitative longitudinal study with persons with ALS and their family carers [submitted]

Chapter 3:

Vandenbogaerde I, Cohen J, Van Audenhove C, Hudson P, Deliens L & De Vleminck A. Support from healthcare professionals in empowering family carers to discuss Advance Care Planning: A population-based survey. Palliat Med. 2022 Nov 9:2692163221135032. doi: 10.1177/02692163221135032. Online ahead of print. PMID: 36349646 [2021 IF 5.713; Ranking Q1; ranking 14/109 HEALTH CARE SCIENCES & SERVICES in SCIE edition]

Chapter 4:

Vandenbogaerde I, De Vleminck A, Cohen J, Verkissen MN, Lapeire L, Ingravallo F, Payne S, Wilcock A, Seymour J, Kars M, Grønvold M, Lunder U, Rietjens J, van der Heide A, Deliens L. Advance care planning-family carer psychological distress and involvement in decision making: the ACTION trial. BMJ Support Palliat Care. 2022 Feb 17:bmjspcare-2020-002744. doi: 10.1136/bmjspcare-2020-002744. Epub ahead of print. PMID: 35177432. *[* SCI impact factor (2021): 4.633 – journal ranking Q1; ranking 25/109 HEALTH CARE SCIENCES & SERVICES in SCIE edition]

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LIST OF ABBREVIATIONS

Abbreviation	Explanation
ACP	Advance Care Planning
ACTION	Advance Care Planning an Innovative Palliative Care Intervention (study)
ALS	Amyotrophic Lateral Sclerosis
BANS-S	Bedford Alzheimer Nursing Severity Scale
CI	Confidence Interval
EM	Estimated Means
EOLD-SWC	End-Of-Life with Dementia – Satisfaction With Care scale
FC	Family Carers
GP	General Practitioner
IES	Impact of Event Scale
MND	Motor neuron disease
NH	Nursing homes
NL	The Netherlands
OR	Odds Ratio
PACE	Palliative Care for Older People in Care and Nursing Homes in Europe
pALS	People with Amyotrophic Lateral Sclerosis
PCU	Palliative Care Unit
RC	Respecting Choices
SD	Standard deviation
SDM	Surrogate decision-maker
STROBE	The Strengthening of the Reporting of Observational studies in Epidemiology
UK	United Kingdom
VOICES-SF	Views of Informal Carers – Evaluation of Services Short Form
WHO	World Health Organization

GENERAL INTRODUCTION

General introduction

Some parts of this general introduction are based on, or to a certain degree copied from the following articles:

Vandenbogaerde I, Miranda R, De Bleecker JL, Carduff E, van der Heide A, Van den Block L, Deliens L, De Vleminck A. Advance care planning in amyotrophic lateral sclerosis (ALS): study protocol for a qualitative longitudinal study with persons with ALS and their family carers. BMJ Open. 2022 May 12;12(5):e060451. doi: 10.1136/bmjopen-2021-060451. PMID: 35551085 [2021 IF 3.006; Ranking Q2; ranking 85/172 MEDICINE, GENERAL & INTERNAL]

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The following paragraphs will provide some background and guide the reader to the aims of this dissertation, followed by the methodology, study design and setting are described, and the outline of this dissertation is listed.

Background

Growing population of people living with a chronic life-limiting illness and with palliative care needs

Over the past century people's life expectancy increased due to improved public health, medical knowledge, and technology[1]. This resulted in a growing population of older people and a fundamental change in death experience by chronic life-limiting illness, especially in the Western high-income countries[1]. Currently, 703 million people are aged 65 years or older and this number is expected to double by 2050[2]. It is estimated that by 2060 48 million people will die from a chronic life-limiting illness[3]. A chronic life-limiting illness is characterized by more prolonged disease trajectories, with a functional decline over months or years and is responsible for more than 73% of the global deaths.[4] Examples of chronic life-limiting illness are cardiovascular diseases, which are the number one cause of death in the general population (17.9 million deaths each year)[5], followed by cancer which is the second leading cause of death (9.6 million deaths each year)[6]. Neurological diseases such as dementia, multiple sclerosis, amyotrophic lateral sclerosis (ALS) or stroke are the third leading cause of death (6.8 million deaths each year).[7] People with a chronic life-limiting illness, such as people living with ALS, dementia, or cancer develop a range of complex needs and symptoms for which palliative care is required[8]. Palliative care is defined by the World Health Organisation (WHO) as *'an approach that aims to improve the quality of life of patients and their families who are facing problems associated with life-threatening illness, through the prevention of suffering by early identification, measurement and treatment of multidimensional problems, symptoms and concerns'*[9]. Early palliative care is recommended, and according to the WHO it is estimated that almost 57 million people need palliative care, but only 14% currently receive it[9], which indicates that palliative care is often not or too late initiated[10,11].

People with Amyotrophic Lateral Sclerosis and their palliative care needs

Amyotrophic Lateral Sclerosis (ALS) is the most common degenerative motor neuron disease in adults, affecting the brain and spinal cord[12]. ALS is incurable and characterized by progressive muscle paralysis involving all voluntary muscles, resulting in difficulties in swallowing, speaking, breathing, and limb paralysis[12]. Respiratory failure is the most common cause of death.[13,14] The prevalence is low, affecting worldwide 4-6/100.000 each year[12]. In Belgium, 220 individuals are diagnosed with ALS each year, whereas an equal amount of people with ALS die annually[15]. The average survival between symptom onset and death is approximately three to four years, which is significantly shorter than the survival of people with other neurological conditions, such as dementia or multiple sclerosis.[12,16–19] Persons are diagnosed with ALS approximately 10 to 12 months after their first symptoms[20,21] which means that they may have already developed severe disability and have a short prognosis[21]. Up to 50% of people with ALS also develop a behavioural and/or cognitive impairment, such as frontotemporal dementia. Persons with ALS often experience physical, emotional, and existential problems that persist until the end of life [22,23]. However, to date, reports show that the complex needs of people with ALS often remain unmet.[21–24] Given the incurable nature of ALS, combined with its rapid progression and unmet palliative care needs an integrated palliative care approach including advance care planning, has been widely advocated for this population[22,23,25].

Multidisciplinary care is the recommended approach for the clinical management of ALS, as research has shown that it improves quality of life in persons with ALS, decrease burden in family carers and leads to a reduction in the number of hospital admissions, and a shortening of hospital stays for patients[26]. In Belgium, this multidisciplinary care is usually organized through neuromuscular reference centres who merely have an advisory role and have expertise in neurology, respiratory care, rehabilitation, as well as in psychology, physiotherapy, occupational therapy, speech and language pathology, nutrition and social work[27,28]. Besides the neuromuscular reference centres, care can also be provided through general practice, community-based services (e.g. home care nurses) and palliative care home teams, but these services are not part of the neuromuscular reference centres[27,28]. Many persons with ALS desire to stay at home for as long as possible, even in the terminal stages of the disease[15], but only 40% die at home[29]. Most often because persons with ALS are admitted to the hospital in the terminal stages with respiratory infections, respiratory failure or because the situation at home is too burdensome for the family carers[15].

People with dementia and their palliative care needs

Dementia is a progressive and incurable neurological disease that affects the brain and that leads to deterioration of cognitive function beyond what might be expected from the usual consequences of biological ageing[30]. Currently, more than 55 million people live with dementia worldwide[30]. This number is expected to increase to 82 million in 2030 and 152 million in 2050[31]. The strongest known risk factor for developing dementia is old age[30,32,33]. Dementia is an overall term for a variety of diseases and injuries, in which Alzheimer's disease is the best-known type of dementia. This type contributes to about 60-70% of all the dementia cases[34]. Dementia is characterized with difficulties in memory, thinking, orientation, comprehension, calculation, learning capacity, language, judgement, and decisional capacity. These difficulties affect the person's ability to perform activities of daily living[30,34]. People with dementia can also experience behavioural and psychological changes such as apathy, depression, aggression, anxiety or trouble sleeping[35]. Thus, people with dementia experience multiple physical, psychosocial and spiritual care needs that persist for month or years until death. However, these care needs often remain unmet. Research has shown that palliative care for people with dementia can improve symptom burden, prevent undertreatment of symptoms and overtreatment with unnecessary and burdensome interventions. Next, it may decrease the burden on family carers and improve the quality of life of both the person with dementia and their families [36,37].

Although most people with dementia wish to stay and die at home, most people with dementia die in a nursing home[38,39]. In Belgium, nearly half of people admitted to a nursing home die with dementia, half of which is advanced dementia[39]. Nursing homes in Flanders are long term care facilities where nursing care is available on-site 24/7, and care from physicians is available off-site[40]. Providing good quality end-of-life care for people with dementia is thus an important aspect of nursing home care[8]. However, different studies have shown that the quality of end-of-life care needs improvement in nursing home residents with and without dementia[41–43]. Providing high quality end-of-life care in dementia is challenging[44], because dementia is often not seen as a terminal condition by health care professional[42], which may lead to poor symptom management or hospital transfers at the end of life[44].

People with advanced cancer and their palliative care needs

According to the WHO one in six deaths are due to cancer[6]. Its incidence and mortality are rapidly growing across the world. The reasons for this are manifold such as growing population, ageing, changing prevalence and distribution of certain cancer risk factors (e.g. physical inactivity)[1,45,46]. Despite progress in diagnosis and treatment, cancer remains a major life-limiting disease, with 18.1 million new cases and 9.6 million deaths worldwide in 2018. Although Europe represents only 9% of the world population, it constitutes almost 25% of the total global cases and 20.3% of the cancer related mortality[45].

People with cancer generally receive oncology care, which aims to cure the cancer, to improve symptom burden, to reduce medical complications related to cancer or to prolong life[47,48], while palliative care in cancer focuses on relieving symptoms[6]. When cure for people with cancer is no longer a realistic option, care needs to be realigned to address their needs and preferences concerning symptom control, psychological support, social, existential and palliative care needs[6,49]. However, these needs often remain unmet. Especially the emotional support was reported as the most commonly unmet need in persons with cancer and their family carers[48,50,51]. Most people with advanced cancer wish to be cared for at home and also prefer to die at home[52–54]. Studies have shown that being able to be cared for at home improves the wellbeing of persons with cancer, reduces healthcare cost, and reduces aggressive treatments at the end of life[55]. Nonetheless, only a small number of people with advanced cancer die in the preferred place[56]. This indicates that timely and effective communication about values, goals, and preferences are important. Thus, advance care planning is widely advocated for this patient population and its families.

Advance care planning as part of good quality palliative and end-of-life care

Advance care planning is an overall term with a variety of definitions and operationalisations[57]. Throughout this dissertation advance care planning is defined as '*a continuous, early-initiated communication process between patients, their family carers and/or health care practitioners that supports adults at any age or stage of health in understanding and sharing their personal values, life goals and preferences regarding future medical care, including end-of-life care'*[58]. The content of such conversations can be documented in an advance directive, a legal document in which a person specifies what actions should be taken for their care if they are no longer able to make decisions for themselves[58].

The way advance care planning is conceptualized and implemented differs between countries, because of the different legal frameworks across countries. In Belgium, there is a specific medicolegal context for advance care planning. Three laws were passed in 2002, all revolving around patient's autonomy[59]:

(1) the law on patient rights in which patients may refuse or withdraw treatment. This can be documented in a legally binding advance directive or 'living will'. This law also states the possibility to assign a surrogate decision-maker. This surrogate decision-maker advocates for one's right in case they can no longer make decisions for themselves[60]

(2) the law on palliative care states that every patient has the right to access palliative care. This law also determines measures for further development of palliative care services[61,62]

(3) the law of euthanasia[63]. Euthanasia is a practice in which the person's life is intentionally ended by the administration of drugs at the patient's explicit request[64]. The law requires that the person who performs euthanasia is a physician, who needs to follow norms and procedures prescribed by the law[63].

Even though people in a healthy condition can have advance care planning conversations, wide consensus exists that particularly patients with a chronic life-limiting illness must have the opportunity to participate in this process**.** Advance care planning is also highlighted by the WHO as an indication of high-quality palliative care[65]. The goal of advance care planning is to provide a timely exploration of a person's underlying values and preferences for future care in order to develop a shared understanding to inform patient-centred care, which becomes particularly important if patients later become cognitively and communicatively incapable of making their own decisions about care[58].

In spite of many discussions about what the right outcome measures in advance care planning ought to be[66], a 2014 systematic review suggests that advance care planning can improve communication about goals of care and overall satisfaction with hospital care[67]. The benefits are most significant if advance care planning is seen as a process with multiple conversations occurring over time, also including the patient's family carers throughout this process[68]. This is crucial as family carers are most often the ones who make decisions at the end-of-life[67]. Studies have shown that when family carers need to make decisions on behalf of the patient without knowing the preferences, it can be an extremely stressful event, leaving them behind with feelings of uncertainty whether they have made the right decision[69].

Although the emphasis of the concept of advance care planning used to be on completing advance directives[70,71], recent recommendations highlight the need to see advance care planning as a process and series of broader conversations about hopes, preferences, and potential care goals, which can also be discussed among patients and family carers themselves[66,72]. Moreover, preferences for current and future care are situational – often related to key events such as symptom progression or multiple hospital admission – and may change over time[72,73]. However, most studies investigate advance care planning at one specific point in time, which only gives a snapshot of the complex reality when engaging in the process of advance care planning. Studies to capture the process of advance care planning over time – such as longitudinal designs – are scarce.

Especially, in a fast-changing disease trajectory such as ALS, it is unclear when and how advance care planning occurs and changes over time. It is recommended that advance care planning is not delayed in persons with ALS[24]. To date, few empirical data is available on how advance care planning is realized in actual practice in ALS. A recent review from 2016 about advance care planning in motor neuron disease, including people with ALS, showed that the uptake of advance care planning seems low in practice and is often delayed until the last months or weeks of life, but less was found about how and when advance care planning should be implemented in the care consistent with the persons' and family carers' needs over time[74]. Most studies on advance care planning in ALS focus on the risks and benefits of life-sustaining interventions (such as invasive or non-invasive ventilatory support or gastrostomy), identification of a surrogate decision-maker and completing an advance directive[21,24,75]. However, studies focusing on the the communication process between the person with ALS, his/her family carer and/or health care professionals in which broader personal values, wishes and preferences for future care are explored, are currently lacking.

Family carers involvement in advance care planning and end-of-life care

Family carers play an important role in end-of-life care, not only by providing physical and emotional or spiritual care to their seriously ill relative, but also by providing information and communication to health care professionals about the wishes of their seriously ill relative[76–78]. Family carers contribution in caring for their relative is often more substantial than for health care professionals[79], which means that family carers have a lot of opportunities to discuss advance care planning with their loved one outside the clinical context. Family carers are willing and welcoming to have advance care planning conversations, but they also admit to having some barriers in performing these conversations such as anxiety, or feelings of being unprepared or feeling uncomfortable[80–82]. Thus, healthcare professionals can play an important role in empowering the family carer to perform advance care planning conversations together with their loved one by having the conversations together or by providing the family carer with tools to initiate these conversations, such as conversation cards[83,84]. However, evidence on the proportion of family carers who performed an advance care planning conversation and received support from a healthcare professional Is currently lacking. Additionally, Involvement of family carers In advance care planning should reduce distress in making end-of-life decisions on behalf of their loved one[85]. Yet, we do not know if an advance care planning intervention would increase the involvement of family carers in decision-making and reduce the family carers' distress.

The perspective of family carers on the quality of end-of-life care is of value. However, only a few studies have so far evaluated the quality of end-of-life care from the perspective of family carers of residents with dementia. Previous cross-sectional studies have indicated that most family carers are grateful for the end-of-life care that the resident with dementia received[86,87] and that the quality of end-of-life care as perceived by family carers is strongly associated with how peacefully their loved one had died[88]. Furthermore, research has also shown that the family carers' experiences with end-of-life care also affects the extent, duration and intensity of their grief[89]. Still, little is known, so far, about which factors related to the resident, their family carers and the care received are associated with the family carers' evaluation of the quality of end-of-life care for nursing home residents.

Study objectives

The purpose of this dissertation is twofold. First, we want to provide in-depth insights in a complex and evolving process such as advance care planning in a disease trajectory of ALS. To investigate how advance care planning topics arise, and if, how and why these change over time. Second, we also want to give an insight into the family carers involvement in advance care planning and their evaluation of end-of-life care, by investigating how these family carers were supported or empowered by healthcare professionals, to know if family carers were involved in decision-making at the end of life, and the level of distress these family carers are experiencing after death of their loved one and to examine the family carers' evaluation of the quality of end-of-life care for nursing home residents with dementia.

The first part of this dissertation is meant to understand in-depth experiences with advance care planning in ALS via patient's and family carers' perspectives. The objectives are:

1. To develop the study protocol of the qualitative longitudinal multi-perspective interview study to understand in-depth experiences with advance care planning in ALS via patient's and family carers' perspectives (**chapter 1**)
2. To understand what the experiences of people with ALS and their family carers are with advance care planning, and to investigate whether these experiences change over time, when, how and why. (**Chapter 2**)

The second part of this dissertation is meant to investigate family carers' involvement in advance care planning and their evaluation of end-of-life care. The objectives are:

3. To investigate how family carers are supported or empowered by health care providers during advance care planning conversations with their seriously ill relatives in the last 3 months of life (**Chapter 3**)
4. To assess the family carers' perspectives about the involvement in decision-making at the end of life, and the level of distress these family carers are experiencing after the death of persons with advanced cancer (**Chapter 4**)
5. To investigate the quality of end-of-life care as perceived by family carers of nursing home residents with dementia in Flanders, Belgium (**Chapter 5**)

Methods

Overview of methods used in the dissertation

In this dissertation, different study designs, and methods are used. To provide in-depth insights into a complex and evolving process of advance care planning throughout the disease trajectory of ALS, we used a qualitative longitudinal research design, whereas we interviewed persons with ALS and their family carers individually three times over a maximum period of nine months (**Chapters 1-2**). To investigate how family carers are supported or empowered by healthcare professionals during advance care planning discussions, we conducted a population-based survey of bereaved family carers sampled from the three largest healthcare insurances in Flanders, Belgium (**Chapter 3**). We used secondary data of the ACTION trial, which was a clustered randomized controlled trial testing the efficacy of the ACTION Respecting Choices (RC) advance care planning intervention in persons with advanced lung or colorectal cancer in six European countries (Belgium, the Netherlands, Italy, Denmark, Slovenia, and the United Kingdom). This secondary data was used to assess the ACTION Respecting Choices (RC) advance care planning intervention on family carers' involvement in decision-making at the end-of-life and the level of distress these family carers are experiencing after the death of persons with advanced cancer (**Chapter 4**). Lastly, to examine the quality of end-of-life care as perceived by family carers of nursing home residents with dementia, we used two retrospective epidemiological studies with comparable research methods from 2010 and 2015 (**Chapter 5**).

Methods used to address the objectives of part 1

A qualitative longitudinal multi-perspective interview study (Chapters 1 - 2)

To address objectives 1-2, we conducted a qualitative, longitudinal, multi-perspective interview design to explore an evolving and complex process such as advance care planning over time from the perspectives of both people with ALS and their family carers[90,91]. This design offers considerable advantages over more typical 'snapshot' techniques in understanding the participants' changing experiences[91]. We used constructionism as an underlying epistemology,[92] as we wanted to know more about the views and meanings constructed by people with ALS and their family carers about experiences with advance care planning and preferences in future (end-of-life) care in the specific disease trajectory of ALS. We included people with ALS and their family carers from three hospitals, all located in Flanders, Belgium, the Northern Dutch-speaking part of Belgium. Inclusion criteria are: 1) the treating neurologist communicated the diagnosis with the person with ALS and their family carer not more than six months ago; 2) person with ALS and their family carer were older than 18 years; 3) both were able to sign a written informed consent. Participants were excluded if they could not speak in Dutch or if the persons with ALS were diagnosed with frontotemporal dementia. We recruited nine dyads of persons with ALS and their family carers. In this study, we interviewed persons with ALS living in the community and their family carers at three timepoints on a three-monthly interval over a period of maximum nine months. Interview guides for the first interviews were developed and cognitively tested with three dyads before the start of our study. The subsequent interviews build upon the previous one and were adapted based on what has been discussed in the previous interview to identify possible changes in their experiences. In each interview participants were asked whether they thought about the future and future (end-of-life) care, what they were thinking about and if they talked about it. In the subsequent interviews, we also highlighted what has been said in the previous interview and asked whether this view has changed and in case of change, why it changed.

Analysis

Qualitative longitudinal analysis is an iterative and multi-dimensional process, which starts with multiple readings of the data. We did a content analysis to obtain an in-depth understanding of the data[93,94]. Codes are constructed in a coding list for each case (a case means person with ALS and their family carer) separately and from each interview round. This coding list was refined within the research team by grouping the codes into categories and themes, which gave us a clear image of the participants' journey and the overarching themes over time. In order to make the data more manageable, a timetable was made to describe changes in experiences with advance care planning. First, we made a timetable for each dyad and each interview to see what has changed over time, how themes overlap and how they interconnect with each other. This gave us a clear image of the participants' journey and the overarching themes over time. Second, we used constant comparison within and between the dyads' timelines to delineate characteristic patterns in the sequences of advance care planning experiences. To limit subjectivity, we discussed the results of this timeline within the research team.

Ethical approval

Ethical approval has been granted by the central ethical committee of the University Hospital of Brussels, (B.U.N. B1432020000128). Ethical approval has been obtained in the other participating hospitals.

Methods used to address the objectives of part 2

A national population-based survey (Chapter 3)

To address objective 3, we conducted a population-based nationwide cross-sectional survey among bereaved family carers, in Flanders, Belgium. Bereaved family carers were recruited via the registers of the three largest health insurers in Flanders. The health insurers maintain records of people who applied for this care budget and the names of their family carers who are also registered during the application process. We randomly sampled 3000 deaths of people with a serious illness who applied for a care budget -indicative of increased informal care needs- from one of the participating health insurers. We only included family carers who had provided care to someone who recently died between two and six months before inclusion to allow sufficient time for grieving while limiting recall bias[95–97].

Analysis

Descriptive statistics were used to summarize the data; therefore we adopted frequency tables and contingency tables with the χ^2-test. To describe which factors were associated with advance care planning discussion, we used multivariable binary logistic regression (advance care planning discussion vs no discussion as dependent variable). The potential predictors included: bereaved family carer characteristics (gender, age in years, educational attainment, medical degree, work status); deceased patient characteristics (serious chronic illness, age in years); and care characteristics (involvement of other family carers, patient lived at home in the final three months, and palliative care services involved) were included in a hierarchical stepwise model selection. We started with a univariable binary logistic regression model and selected, for the stepwise model, only the variables which were (marginally, i.e $p<0.1$) statistically significant. Next, three multivariable binary logistic regression models were constructed: 1) deceased patient characteristics, 2) adding bereaved family carer characteristics, 3) adding care characteristics. Only variables with $p <.05$ were retained for a next step and for the final model. Decisional capacity was left out of the analysis due to multicollinearity with dementia.

Ethical approval

Ethical approval was obtained from the Ethical Review Board of Brussels University hospital of the Vrije Universiteit Brussel (B.U.N. 143201940562)

An international cross-sectional survey (Chapter 4)

To address objective 4, we analysed data of the bereavement questionnaire from the ACTION trial, a multicentre cluster-randomized controlled trial carried out in 23 hospitals in six European countries (Belgium, Denmark, Italy, the Netherlands, Slovenia and the United Kingdom). The hospitals were randomised to either the intervention group or to the control group. Patients with advanced lung- or colorectal cancer were asked to participate in the ACTION study and if they consented, they were followed for one year. If a patient died in the year after inclusion, a bereavement questionnaire was sent to the deceased's address. The bereavement questionnaire was conducted to obtain more information about the last months and weeks of the patient's life as perceived by a bereaved family carer who lost the patient during the one year of inclusion in the study. Involvement in decision-making was measured with a single item of the Views of Informal Carers – Evaluation of Services Short Form (VOICES-SF) questionnaire, which is a 58-item validated questionnaire about health and social services completed by bereaved family carers[98]. Psychological distress was assessed using the impact of event scale (IES) and asked how frequently each item was experienced during the past week after three months of bereavement. The IES measures psychological responses to stress on two subscales: (1) intrusion and (2) avoidance and is mostly used to measure post-traumatic stress syndrome[99]. A total of 390 patients died in the year of inclusion, indicating that 390 bereavement questionnaires were distributed over six European countries. Only 162 returned: 71 for the intervention group; 91 for the control group (response rate: 41.5%).

Analysis

Multilevel analyses were used to determine the effect of advance care planning on involvement in decision-making (via multilevel binary logistic regression) and the effect of advance care planning on psychological distress among family carers (via multilevel linear regression).

Ethical approval

Ethical approval has been obtained from research ethics committees and ethical review boards of all participating hospitals in all countries. Trial registration: International Standard Randomised Controlled Trial Number (ISRCTN), 17231.

Two retrospective epidemiological studies with comparable research methods (Chapter 5)

To address objective 5, we used data from two retrospective epidemiological studies: the Dying Well with Dementia study and the PACE study (Palliative Care for Older People in care and nursing homes in Europe). The Dying Well with Dementia study was conducted in 2010 in Flanders, Belgium, and aimed to describe the clinical characteristics and comfort of home residents with dementia[39]. The PACE study was conducted in 2015 in six European countries (Belgium, Finland, Italy, the Netherlands, Poland and the UK). The aim of this study was to describe and compare those six European countries in terms of comfort in the last week of life of all residents, of whom the presence and severity of dementia could be determined[100]. We identified the family carers of the deceased residents with dementia from the studies in 2010 and 2015 as follows: in the study of 2010, deceased residents with dementia were identified in two steps; first, the administrator was asked to identify all the residents who had died in the predefined period and to include those who had possibly had dementia using the screening criteria of the KATZ scale, used by the Belgian health insurance system to assign financial resources: Category C dementia i.e. 'being completely care dependent or needing help for bathing, dressing, eating, toileting, continence, and transferring plus being disoriented in time and space' OR disorientation in time and space (KATZ scale ≥ 3 or 'having almost daily a problem with disorientation in time and space'). If a resident was identified with one of these two criteria, they were included for data collection in the study of 2010 (n=241). Second, the nursing staff and General Practitioner (GP) were asked whether they thought the resident 'had dementia' or 'was diagnosed with dementia'. If either one indicated 'yes', then the person was considered as someone with dementia. In the study of 2015, questionnaires on all deceased residents were sent to the administrator, nursing staff, general practitioner and family carer (without performing the first step pre-selection in the study of 2010). Residents were considered as having dementia following the same second step procedure of 2010. For objective 5, we are interested in the quality of end-of-life care as perceived by the family carers and therefore only included the cases for whom the family carers had returned a questionnaire.

Analysis

First, we tested for possible differences between the years 2010 and 2015 in the family carers' evaluation on the quality of end-of-life care. The comparability of the data allowed us to combine the data of 2010 and 2015 for the analysis. The assumption of linearity was violated in the variable 'Length of Stay' so this variable was recoded as a categorical variable. We performed univariable multilevel analyses, due to clustering of data in the nursing homes. Multivariable multilevel analyses were tested, whereas we controlled for gender of the resident, length of stay in the nursing home, whether relatives received information on palliative care, and whether they received information on medical care from a care provider. These variables were deemed significant in the univariable multilevel analyses.

Ethical approval

Ethics approval for both studies was obtained from the Ethical Review Board of Brussels University hospital of the Vrije Universiteit Brussel (B.U.N. 14320108163 (2010); B.U.N. 143201422845 (2015)).

Dissertation outline

This dissertation consists of a General Introduction, Part 1, Part 2, and a General Discussion. Parts 1 and 2 contain the findings of all studies presented in this dissertation, that have been published or submitted to A1 or A2 journals.

The General Introduction provided background about chronic illness, amyotrophic lateral sclerosis, cancer, advance care planning, and family carers involvement. Next, the aims of this dissertation, the methodology, study design and setting are described

Part 1 describes in-depth experiences with advance care planning in people with Amyotrophic Lateral Sclerosis (ALS) via patients and family carers' perspectives. **Chapter 1** outlines the study protocol of the qualitative longitudinal multi-perspective interview study. **Chapter 2** reports on the experiences of people with ALS and their family carers with advance care planning and if, how and why these experiences change over time.

Part 2 of this dissertation focuses on the family carers involvement in advance care planning and their evaluation in end-of-life care. **Chapter 3** reports on how family carers are supported or empowered by healthcare professionals during advance care planning discussions with their seriously ill relatives in the last three months of life. **Chapter 4** investigates the family carers' perspectives about the involvement in decision-making at the end-of-life, and the level of distress these family carers are experiencing after the death of persons with advance care. **Chapter 5** investigates the family carers' evaluation on the quality of end-of-life care for nursing home residents with dementia.

The General Discussion consists of a summary of the main findings of this dissertation, reflections on its strengths and limitations, interpretation of the main findings and the implications for research, practice, and policy.

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PART I: In-depth experiences with advance care planning in people with Amyotrophic Lateral Sclerosis (ALS) via patient's and family carer's perspectives

Chapter 1:

Advance care planning in Amyotrophic Lateral Sclerosis (ALS): study protocol for a qualitative longitudinal study with persons with ALS and their family carers

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Abstract

Introduction*:* Amyotrophic lateral sclerosis (ALS) is an incurable motor neuron degenerative disease that has rapid progression and is associated with cognitive impairment. For people with ALS (pALS) and their family carers, advance care planning (ACP) is beneficial, as it can lead to feelings of control/relief and refusal of unwanted treatments. However, evidence concerning the experiences and preferences regarding ACP of pALS and their family carers, especially when their symptoms progress, is scarce. This article describes the protocol for a qualitative longitudinal study that aims to explore: (1) the experiences with ACP and the preferences for future care and treatment of pALS and their family carers, and (2) how these experiences and preferences change over time.

Methods and analysis*:* A qualitative, longitudinal, multi-perspective design. A total of 8 to 9 dyads (pALS and their family carers) will be recruited, and semi-structured interviews administered every 3 months over a 9 month period. Qualitative longitudinal analysis involves content analysis via in-depth reading, followed by a two-step timeline method to describe changes in experiences and preferences within and across participants.

Ethics and dissemination*:* This protocol has been approved by the central ethical committee of the University Hospital of Brussels, and local ethical committees of the other participating hospitals (B.U.N. B1432020000128). The results will be disseminated via the research group's (endoflifecare.be) website, social media and newsletter and via presentations at national and international scientific conferences.

Introduction

Amyotrophic lateral sclerosis (ALS) is the most common degenerative motor neuron disease (MND) in adults, affecting the brain and spinal cord.[1] ALS is incurable and characterized by progressive muscle paralysis. Respiratory failure is the most common cause of death.[2,3] The average survival between symptom onset and death is approximately three to four years, which is significantly shorter than the survival of people with other neurological conditions, such as dementia or multiple sclerosis.[1,4–7] Up to 50% of people with ALS (pALS) also develop a cognitive impairment, such as frontotemporal dementia. Further, they often experience physical, emotional, and existential problems that persist until the end of life. However, to date, reports show that the complex needs of pALS often remain unmet.[8–11] Given the incurable nature of ALS, combined with its rapid progression and unmet palliative care needs, an integrated palliative care approach, including advance care planning (ACP), has been widely advocated for this population[10–12]. ACP is defined as a continuous, early-initiated communication process between patients, their family carers and/or healthcare professionals that enables individuals to define goals and preferences for future end-of-life care. ACP can prepare patients, family carers and healthcare professionals for making the best possible in-the-moment decisions that are consistent with the patients' values, goals and preferences[13].

A 2014 systematic review in geriatric and cancer populations suggests that ACP can improve communication about goals of care and overall satisfaction with hospital care and end-of-life care[14], especially if seen as a process with multiple conversations with patients and their family carers occurring over time[15]. However, the majority of studies have investigated ACP practice or participants' perceptions on ACP at one specific timepoint,[16–18] which merely gives a snapshot of the complex and dynamic reality of engaging in the process of ACP throughout the disease trajectory. A 2016 systematic review of ACP in people with MND, including ALS, also showed important benefits with the uptake of ACP – such as feelings of control/relief and refusal of unwanted treatments – but less was found about how and when ACP should be implemented in the care consistent with the persons' and family carers' needs over time.[19]

Most studies on ACP in ALS focus on the risks and benefits of life-sustaining interventions (such as ventilatory support and gastrostomy), identification of a surrogate decision-maker, and completing an advance directive[8,9,20]. However, recent recommendations highlight the need for ACP to be seen as a series of broader conversations about hopes, preferences, and potential care goals, which can be discussed among patients and family carers themselves in an informal manner[21,22]. Moreover, preferences for current and future care are situational – often related to key events such as symptom progression or multiple hospital admissions – and may change over time.

To our knowledge, only one longitudinal qualitative study about ACP in ALS conducted non-participative observations for 6 months of appointments between pALS and their treating physician, followed by a single in-depth interview with the patients. This study showed the feasibility and acceptability of implementing ACP throughout the pALS disease trajectory.[23] However, only the patients' perspective was considered and not that of their family carers. Up until now, family carers' perspectives about ACP in ALS are mostly explored retrospectively and during bereavement.[24,25] The perspectives of family carers involved in ACP conversations is crucial, as they often have a prominent role in decision-making at the end-of-life[26], and it has been shown that involvement in ACP improves family carers' confidence when making end-of-life decisions on behalf of their relative (if needed)[27] and reduces the family carers' distress and grieving[14,28]. This shows that serial and multi-perspective interviews are ideal in exploring experiences with ACP and preferences for future care and treatments of pALS and their family carers and how these experiences and preferences change over time.

The aim of this article Is to desc”Ibe ’he protocol of a longitudinal and multi-perspective qualitative interview study that aims to explore the experiences of pALS and their family carers with ACP, their preferences for future care and treatments at 3 different timepoints, and to investigate whether these experiences and preferences change over time. This study will allow us to see in-depth if, how and why ACP occurs and changes in a unique and fast-changing ALS patient population and their family carers.

The research questions are:

1) What are the experiences of people with ALS and their family carers with engaging in the process of ACP, and how do these experiences change over time?
2) What are the preferences of persons with ALS regarding their future care and treatments, and how do these preferences change over time?
3) What are the preferences of family carers regarding future care and treatments of persons with ALS, and how do these preferences change over time?

In this article, we outline the research design and methodology developed to answer these research questions. ACP is a complex communication process that requires a research design that is capable of exploring such complexity over time. Longitudinal qualitative research is an emerging methodology, in which time is designed into the research process, making change the focus of analysis. Using this methodology, we aim to investigate lived experience of change with regard to

ACP and future care and treatment of pALS and their family carers; the processes by which this experience is created; and the causes and consequences of this change. With this protocol, we hope to inform future international longitudinal qualitative research in other populations – such as patients with dementia or organ failure – who could also benefit from receiving optimal ACP delivery, which has so far remained understudied[29].

Methods

Study design

This study has a qualitative, longitudinal, multi-perspective interview design to provide rich information about the ACP process over time from the perspectives of both pALS and their family carers[30,31]. This design is most suitable for exploring an evolving and complex process such as ACP[32], as this method is driven by a desire to understand, not just if change happens, but how and why it happens in the socio-cultural context over time[32]. It offers considerable advantages over more typical 'snapshot' techniques in understanding the participants' changing experiences and preferences[31]. Constructionism will be used as an underlying epistemology,[33] as we want to know more about the views and meanings of pALS and their family carers about experiences with ACP and preferences in future (end-of-life) care in the specific disease trajectory of ALS.

Setting

This study will follow pALS living in the community and their family carers interviewed at 3 timepoints on a 3-monthly interval over a period of maximum 9 months. In Belgium, care for pALS is usually organized through neuromuscular reference centers, providing specialist multidisciplinary care comprising expertise in neurology, respiratory care and rehabilitation, as well as in psychology, physiotherapy, occupational therapy, speech and language pathology, nutrition and social work[34,35]. The neuromuscular reference centers are connected to University Hospitals. Care can also be provided by a general practitioner, community-based services (e.g., home care nurses) and palliative home care teams. Almost 90% of the patients stay at home even in the terminal stages of the disease[36]. Voluntary support services, in the form of national or regional ALS associations (e.g., ALS Liga in Belgium), can also deliver care to pALS and their families[3].

Participants and inclusion criteria

We will include patients from 3 hospitals, all located in Flanders, Belgium, the Northern Dutch-speaking part of Belgium. Inclusion criteria are: 1) the treating neurologist communicated the diagnosis with the pALS and their family carer not more than 6 months ago; 2) pALS and their family carers are older than 18 years; 3) both must sign a written informed consent. Participants will be excluded if they cannot speak in Dutch or if the pALS are diagnosed with frontotemporal dementia. Because the study is an explorative multi-perspective study, we decided to include 8 to

9 dyads of pALS and their family carers to explore if, when and how ACP occurs, which can result in a total of 54 interviews to be analysed (if each participant is interviewed individually 3 times). This is a smaller sample compared to other longitudinal interview studies[37,38].

Recruitment

To recruit pALS and family carers, we have purposefully selected 2 academic hospitals (UZ Gent and UZ Brussel) and a non-academic hospital (AZ Maria Middelares). The pALS and their family carers will be approached by their treating neurologist to ascertain willingness to participate in the study. Potential participants who give consent to their neurologist to pass their contact information to the researchers will be contacted by a member of the research team (IV), to address the purpose of the study. We will wait at least 6 weeks after diagnosis to contact the pALS to allow time for grieving after being diagnosed with ALS, which is a life-changing diagnosis[39].

We will recruit new potential participants when a participating pALS and/or their family carer drops out of the study after the first interview – for example, due to severe deterioration of the illness, death, or when they no longer want to participate. Participating in 2 interviews will give us the opportunity to potentially identify changes in experiences and perspectives (if any). Hence, re-recruitment is not necessary. We foresee 1 year of recruitment and the data will be collected through face-to-face semi-structured interviews with persons with ALS and their family carers in Flanders, Belgium, at 3 timepoints from February 2021 onwards

Data collection

The pALS and their family carers are preferably interviewed separately, as this will give us the opportunity to observe similarities and differences in their experiences and preferences in ACP[30]. However, if the pALS or family carers wish to have the interview together, and both agree, we will honour their request.

Three interview guides have been developed for the first interview: one for the pALS, one for the family carers, and one in case the pALS and family carers prefer to be interviewed together. These interview guides focus on: 1) the experience with ALS, 2) the experiences with ACP, and 3) the preferences about future care and treatment. The subsequent interviews will build upon the previous one and will be adapted based on what has been discussed in the previous interview, to identify possible changes in their experiences and preferences. In the case of a possible change in experiences or preferences, we will reflect during the interview what triggered the change (e.g. unexpected hospitalization), and we will also reflect on whether and how the interviewer had an influence on the possible change (see Appendix I).

We aim to interview the participants on a 3-monthly interval, but flexibility in timing is necessary in case of a sudden change in the experiences with ACP and the preferences for future care and treatments. Other studies have shown that it is useful to use telephone contact to assess whether an interview should be brought forward to capture a changing event[37,40]. Therefore, we will conduct short monthly phone calls with the pALS or family carers. During these phone calls, we will ask how the pALS and family carer are doing, how the disease trajectory is evolving and whether a sudden change (e.g. unexpected hospitalisation) has occurred. These monthly phone calls are merely a 'check-in' with the pALS or their family carers to assess whether a subsequent interview needs to be planned sooner than anticipated. These phone calls will not be audiotaped or analysed. Prior to the first interview, the interviewer will ask who should be called for the monthly phone calls. If the pALS prefers to be the contact person and speech deteriorates, we will ask whether the family carer may be contacted. These monthly phone calls also help develop trust between participants and interviewer, and they will also help the interviewer monitor possible distress[29]. In case of distress, the interviewer will advise the participants to talk to their treating neurologist or the psychologist of the neurological department. If the pALS's speech deteriorates, and they still wish to participate, they will have the opportunity to participate via a speech computer or in writing.

Data analysis

Qualitative longitudinal analysis is an iterative and multi-dimensional process, which involves multiple readings of the data. First, content analysis, which involves line-by-line coding, will be used to obtain an in-depth within-case understanding of the data[41,42]. Codes will be constructed in a coding list for each case separately for persons with ALS and family carers and from each interview round. In the case of any discrepancies, the codes will be discussed between 2 researchers until consensus is reached, which results in a coding list. This coding list will be refined within the research team by grouping the codes into categories and themes.

Secondly, we will use a 2-step timeline method to describe changes in ACP experience and preferences within and over all the participants[43]. First, a timeline – with time on the X-axis and the themes on the Y-axis – will be made for each participant of the dyad and each interview to see what has changed over time, how themes will overlap and how they interconnect with each other. Each timeline will give a clear image of the participants' journey and the overarching themes over time. Second, we will use constant comparison within and between the dyads' timelines to delineate characteristic patterns in the sequence of ACP experiences and preferences (both within and between the dyads)[44]. To limit subjectivity, results of this timeline method will be discussed within the research team.

Patient and public involvement

Patients and family carers were involved in the interview guide development. The main results will be disseminated to the study participants. The strategy for the wider dissemination of the study results to pALS and families will be discussed with ALS patient organisations.

Ethical considerations, ethics approval and dissemination

Ethical approval has been granted by the central ethical committee of the University Hospital of Brussels, (B.U.N. B1432020000128), via an amendment. Ethical approval has been obtained in the other participating hospitals. Given the (possible) vulnerability of pALS and their family carers, various safeguards are considered for this study: 1) Serial informed consent will be required in this qualitative longitudinal research. Before the first interview, an informed consent form will be signed. For the other interviews, verbal consent will be obtained by audiotaping. The interviewer will inform the participants of the purpose of the study during the first contact and prior to each interview before audiotaping; 2) The interviewer (IV), who is an experienced clinical psychologist and who is also the main researcher, will take several steps to ensure the participants' comfort prior to and during the interviews (e.g., the interview will be conducted at a location and time of the participants' choosing; breaks will be taken throughout the interviews). Given the extensive educational training and experience, she is capable to capture distress if this arises.; 3) It will be emphasized that, if patients or their family carers would decide not to participate, this decision will by no means influence the quality of their care; and 4) Participants may deteriorate and die during the study – therefore, it is important that the interviewer him/herself has a supportive network to be well-supported in their role as interviewer.[29]

The results of this study will be submitted for publication in peer-reviewed journals and will be presented at national and international research and professional conferences. Furthermore, we will disseminate the results via the research group's (endoflifecare.be) website, social media and newsletter.

Discussion

This will be the first study to provide first-hand, longitudinal, in-depth, and multi-perspective insights into the process of ACP, and this in a unique patient population of ALS and their family carers. Experiences and preferences in ACP may change over time[9,13] – but, so far, no studies have optimally investigated if, how, and why ACP experiences and preferences might change over time in pALS. This study will provide highly valuable information for clinical practice concerning when and how to implement ACP throughout the course of the ALS disease, according to the pALS and family carers' views and preferences, which will improve palliative care and end-of-life care in ALS. Moreover, it will also give us insights into how ACP occurs in the informal context, which is currently understudied in the research field. An important strength of the methodology is that a longitudinal qualitative study, if combined with flexibility, is a less restrictive approach towards studying time and change in complex processes such as ACP[29,45]. ACP is usually measured and described in a single point in time, but the disease's complexity cannot be captured via these snapshot techniques. Another important strength is that longitudinal qualitative studies and multi-perspective interviews are innovative methods in medicine and especially in the palliative care field. Moreover, longitudinal qualitative research is a prospective approach, but experiences and preferences may change with the perspective of time, which allows us to also have a retrospective view, which requires a unique way of interviewing. A final important strength is that the perspectives of both pALS and their family carers will be interviewed to enhance our understanding of the dynamics and relationships between them and the individual needs of persons with ALS and their family carers in ACP, and this approach will allow us to explore similarities and differences in their views about ACP.

This study has several challenges. First, it is common that participants withdraw in these types of studies because of the longitudinal aspect. We aim to tackle this challenge by having monthly phone calls, as studies have shown that this can make the interviewees feel more comfortable[37,40] and develop a trusting relationship with the interviewer,[29] which could limit participant attrition[40]. Also, if the patient's speech deteriorates, we will give the pALS the opportunity to tell their story via writing or the use of a speech computer. Nevertheless, a review did show that pALS need some time to work with a speech computer[46], but before a subsequent interview is planned, we will allow them the sufficient time for working with this speech computer. Another challenge is that recruitment may be difficult since this study does not address cure or treatment. However, previous research has shown that people usually see participating in ACP research as a worthwhile endeavour – and so we consider this challenge to be minor. We will verbally, and with a written informed consent, inform the participants about the purpose of the study in our first contact and before each interview. Discussing ACP is a difficult subject and might be a challenge, especially if the focus is only on end-of-life decisions. However, our focus will be on a broader level of hopes, preferences and potential care goals about the future. Studies have shown that pALS and family carers welcome the opportunity to discuss ACP[9,47] and they regard ACP as something beneficial[48,49]. In this study, we interview pALS and their family carers about if, how and why ACP occurs throughout the disease trajectory. It is possible that, by addressing these topics in the first interviews, the participants will be triggered to think about or discuss ACP, and thus this can influence their views and perceptions on ACP during the subsequent interviews. Therefore, this will be a specific point of attention during the follow-up interviews, and the interviewer will reflect together with the pALS and the family carers on how discussing experiences, assumptions or beliefs about ACP in the previous interviews had an influence on their current experiences with ACP and/or preferences for future (end-of-life) care.

Finally, this study involves a relatively small number of participants (8 to 9 dyads) compared to other longitudinal qualitative studies[37,38]. In Belgium, only 220 new ALS diagnoses occur per year[36], which shows how rare and unique this patient population is. Longitudinal qualitative research inevitably generates a large volume of interviews, for which effective planning is essential to keeping the data manageable. Given the limited timeframe in which to conduct this study, including 8 to 9 dyads was deemed feasible for addressing the aims of this exploratory study. Hence, we will interpret our results with caution in terms of generalization to a larger group of pALS and their family carers.

Additional information

Authors contribution

Conception and design of the work: IV, JDB, EC, AVDH, LVDB, LD, ADV; Ethic approval: IV, JDB, EC, AVDH, LVDB, LD, ADV; Drafting the work: IV; Critical revision for intellectual content: IV, RM, JDB, EC, AVDH, LVDB, LD, ADV; All authors have read and approved the final manuscript.

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Competing interests statement

The authors declare that they have no competing interests.

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Appendix I: interview guides

Interview guide 1: Person with ALS (interview 1)

Introduction

XX months ago you've received the diagnosis of ALS from XX (name neurologist). Can you tell me something more about how it all started, about the process of being diagnosed?

- When did you notice the first symptoms/signals?
- When did you notice that something was wrong?

When you have heard the diagnosis of XX (name neurologist). What was that for you? How did you feel? What did you think?

What were for you the biggest changes since the diagnosis? (*Changes physically, emotionally, socially)*

Thinking about the future

Can you tell me something more about ALS? What impact it has on you?

- Who told you this?
- What did you think/feel when you heard about this?

Do you think about your future? Is it something you think about it more than before the diagnosis or vice versa? How do you see your future now?

Option 1: Patient says (s)he thinks about the future:

- What are your expectations about the future? What do you hope for?
- What is important for you when you think about the future? What would you like to do?
- Is there something you are worried about? What are you worried about?
- Is it something you can discuss with someone? If so, who?

Option 2: Patient says (s)he does not want to think about the future:

- What makes it difficult for you to think about the future?
- Do you know if XX (name FC) thinks about the future? What would (s)he thinks about? Does that keep you awake? Why (not)?

Thinking and talking about the future

Do you think about the care you might need in the future? What do think about? What are your expectations? What are your worries? What do you hope for?

- Who gave you this information?

Option 1: Patient indicates (s)he thinks about it:

- Would that be something you would like to discuss with someone? With whom?
- In case the patient does not wish to discuss future care with FC/HCP:
    - With whom you would like to discuss it? Why not with XX (name FC) or with HCP?
- In case the FC blocks the conversation about future/future care:
    - How do you feel about this?
    - What do you do when that happens?
    - What would you like to tell him/her?
- In case there is open communication about future/future care (see questions hereinafter).
- Patient indicates future or future care is not discussed, but believe the family carer might be open for it:
    - What do you hope to achieve with this conversation? What are your expectations?
    - What would you like to discuss?
    - How would that be for you to discuss your future/future care? Your worries, your ideas, your wishes, etc?
    - When would be the best timing to have this conversation?
    - Who should start the conversation? You, XX (name FC), HCP?
    - Would it be important to you to discuss this first privately with FC? Why (not)?
    - How do you think you would experience this conversation?

Option 2: Patient indicates (s)he does not wants to think about the future:

- What makes it difficult to think about the future?
- Does XX (name FC) has the same feeling?
- How would you react when XX (name FC) starts the conversation? Would you block it? Why (not)?

What are you afraid of that might happen if you talk about this?

Preferences about the future/future care

What is important for you when you think about your future or future care? Do you have specific wishes/ideas/preferences/expectations/hopes/etc + Ask more about the underlying values (What makes you have these wishes/ideas/XX; What is important for your when you think about your future or future care?).

***You may give examples. ATTENTION: give only examples about what you have heard during the interview. How do you feel about a wheelchair, how do you feel about travelling, etc?**

What would be the worst care for you and what would be the best care? Why?

Patient tell his wishes/preferences/ideas during the interview:

- Have you ever discussed these wishes, XX (give some examples about what they have said) with someone? Why (not)?

Option 1: patient indicates wishes/preferences were discussed during a conversation:

- Who was present during this conversation?
- What have you talked about?
- What has triggered the conversation?
    - Was the timing right? Why (not)?
    - In case the timing wasn't right: When would be the right timing for you?
- How did XX (name FC) think about your preferences/wishes?
- Did you talked about it several times after that conversation? Why (not)?
- Which decisions/plans were made about future/future care?
    - How did XX (name FC) felt about this?
    - Do you believe that soms preferences/wishes were difficult for XX (name FC)? Why (not)? What was difficult, what not? How do you cope with that?
- Did you ever consider writing these preferences/wishes down in a living will? Why (not)?
    - Do you believe that people close to you such as XX (name FC) knows which decisions they might need to make in case you cannot say them yourself? Why (not)?
- How did you end the conversation?
- What have you done afterwards?

Option 2: patient tells his/her wishes during interview, but indicates that (s)he did not discuss it with FC or HCP:

- Would you like to discuss your future/future care in the near or distant future? With whom?
- What is for you a barrier to have this conversation?
- What/who could help you to start this conversation? Would you like to discuss it with a HCP? If so, who?
- How would you feel, if you would want a conversation with XX (name FC) and you talk about XX (a preference that came forward during the interview), but XX (name FC) does not want it? OR How would you react if XX (name FC) would start a conversation about your future/future care?
- When would be the best timing to talk about the future or future care (for example to talk about preferred wheelchair)? Would this also be the right time to discuss XX (another preference)? Why (not)?

Option 3: patient makes it clear (s)he does not want to talk about future/future care/preferences/wishes/etc.

- What makes it difficult for you to talk about all this?
- What is the worst that may happen if you would think about it?
- Is there something you are afraid of?

Preferences about the conversation about future/future care

Option 1: In case future/future care has not been discussed yet with FC/HCP:

- When would be the right time for you?
- What makes it difficult to have this conversation?

Option 2: In case future/future care has been discussed:

- What triggered the conversation?
- Who was present? Was everyone present you would have liked to? Why (not)?
- Who supported you during this conversation?
- What made it for you the best time to discuss it? Or would you have preferred another time?

Would you discuss it again in the future? Why (not)?

End of interview

Has everything been discussed what you've had liked to discuss?

Interview guide family carers (FC): first interview

Introduction

XX (name patient) and you received XX months ago/ a while ago the diagnosis of ALS. Can you tell me something more about how it all started, about the process of being diagnosed?

- When did you notice the first signals/symptoms?
- When did you notice that there was something wrong?

When you have heard the diagnosis of XX (name neurologist). What was that for you? How did you feel? What did you think?

What were for you (as carer) the biggest changes since the diagnosis? (*Changes physically, emotionally, socially)*

Thinking about the future

What do you know about the disease/trajectory of the disease? Which impact does ALS have?

- How did you receive this information? Who told you about this?
- What did you think/feel when you've heard this?

Do you think about your future and the future of XX (name patient)? Is it something that you worry about? Are you thinking more about the future than before the diagnosis, or vice versa, do you think about it less? Or is it another way of thinking about your future? Could you tell me something more about this?

Option 1: FC says (s)he thinks about the future:

- What do you expect? What do you hope for – for you and XX (name patient)?
- What is important for you when you think about the future? What do you think is important for the future of XX (name patient)?
- What makes you worry about the future?
- With whom would you prefer to discuss the future?

Option 2: FC thinks about the future, but they do not discuss it with the patient:

- Would you like to discuss it one day with XX (name patient)?
- Do you know what (s)he thinks about? Do you know what (s)he would want or hopes or expects about the future? What do you think is important for XX (name patient)?

Option 3: FC says they do not want to think about the future:

- You say you do not want to think about the future: How come? What makes you to refuse to think about the future?
- Is it something you are afraid of and why?

How do you think XX (name patient) thinks about it? Would (s)he thinks about the future? Would (s)he worries about it?

Thinking and talking about the future

Do you think about the care XX (name patient) might need in the future? What do you think about? What are your expectations? What do you worry about? What do you hope for?

First asking the questions to see the perspective of the family carer AND then asking about how they think the patient thinks about these things.

- From whom did you receive information about his/her care?

Option 1: If FC thinks about future care:

- Is it something you discuss with XX (name patient)? Or with someone else? If so, who?
- If it is not possible to discuss care with patient/HCP:
    - Would you discuss it with friends/family/other professionals/etc?
- FC says (s)he tries to discuss it, but patient does not wish to have a conversation about his/her care:
    - How do you cope with it when you realize XX (name patient) does not want to talk about it?
    - What do you feel/think?
    - What would you like to say to XX (name patient)?
- FC indicates that they communicate openly about future care (go further to questions hereinafter).
- FC indicates they did not discuss future care, but thinks the patient might want to talk about it:
    - What do you hope this conversation would trigger? What are your expectations?
    - What would you like to say in that conversation?
    - How would that be to discuss your worries about XX (name patient) about his/her future?
    - When would it be important – according to you – to discuss it?
    - How would you experience such conversation?
    - Who should ideally start this conversation? Would you start it? Would you like that someone is there to support you? If so, who?
    - Would you prefer to have this discussion alone with XX (name patient) or together with a HCP? Why?

Option 2: FC indicates they do not wish to think about future or future care:

- Why do you not wish to think about future care?
- Does XX (name patient) share this opinion? How do you feel about the future care?
- How would you react/What would you do if XX (name patient) or a HCP wants to discuss future care and starts this conversation? Would you block it or would you talk about it? Why?

Preferences about the future/future care

What is important for you when you think about the future or future care of XX (name patient)? What do you wish for? What are your preferences/ideas? Are there things you know XX (name patient) would want or would not want about care? Are there things about care you do not want?

What is – according to you important about XX's (name patient) future? What is important for you?
***Here we can give examples they have indicated during the conversation or ask how they felt about the wheelchair or walking cane etc → **ATTENTION**: listen carefully what the FC says during the interview → you cannot talk about care wishes they did not talk about. So DON'T talk about a stomach-pump if they did not talk about it): For example you might ask: how do feel about the wheelchair? How do you think XX (name patient) feels about the wheelchair? OR I have heard you've made a lot of travels before your illness, is it something you wish to do with XX (name patient)? Do you think (s)he would enjoy this? Did you discuss it? Did you make plans? ***

OPTION 1: FC knows which preferences/wishes the patient has and they seem to talk about these preferences/wishes with one another:

- I hear that you have discuss it. Was there someone else present during this conversation? If so, who?
- What have you discussed?
- What did the conversation trigger? Did you believe the moment was ideal? Why?
  - In case they did not believe the timing was ideal: When would be the best timing?
- During the conversation, have your feelings been taken into account about certain wishes/preferences/ideas/expectations/hopes/etc XX (name patient) has? Did you say how you felt/thought about it? Why and how?
  - In case not: What makes you did not say how you felt? What might happen if you did?
- Did you make concrete plans about the future/future care?
- Have you considered to write these wishes/preferences/etc down? Why (not)?

    - How do you feel about a living will?
- Imagine you need to make a medical decision instead of XX (name patient), would you believe that you can make this decision? In other words, do you know what XX (name patient) wants or does not want?
- How did the conversation end? What have you done afterwards?

Option 2: FC seems to know about the preferences/wishes the patient have about the future/future care, but they did not discuss it concrete:

- What makes you so sure that XX (give example about a preferences/wish) is what XX (name patient) wants? Have you ever discussed it? Did you ever talk about before diagnosis? Or is it something that you see/feel? Would you like to elaborate this more?
- Would you like to have a conversation about the preferences/wishes with XX (name patient)? Why (not)?
    - What would you say or do if XX (name patient) says a wish/preference you Imagine that during this conversation XX (name patient) tells a wish/preference you cannot support?
    - When would be the ideal moment for you to have this discussion?
    - With whom you would like to discuss it? Only with XX (name patient) or also with HCP/kids/friends/etc?

OPTION 3: FC indicates (s)he does not know which preferences/wishes the patient has, but would like to know them:

- I hear you wish to have a conversation about the wishes/preferences XX (name patient) might have about the future/future care. Would this be something you would like to discuss alone with XX (name patient) or with a HCP or with kids/family/friends/etc?
- Would you initiate the conversation, or would you need help?
- Is there something that would make it difficult to discuss the wishes/preferences? If so, what? And what might help to overcome this barrier?
- What are the preferences you have about the future or future care of XX (name patient)?
- What do you hope to achieve in this conversation? What are your expectations?

OPTION 4: FC indicates (s)he does not know which preferences/wishes/etc the patient has, but FC does not want to know either:

- Why do you not want to know the preferences/wishes of XX (name patient)?

What is the worst possible thing that might happen if you would discuss it? Or if you would know the wishes/preferences of XX (name patient)?

Preferences about the conversation about future/future care

OPTION 1: FCs did not discuss wishes/preferences with patient/HCP/

- When would be the best timing to have this discussion with XX (name patient)?
- What makes it difficult to start this conversation?
- How do you think this conversation might go?
- How would you feel during such conversation?

Option 2: In case they have had this discussion:

- What triggered the conversation? How did it start?
- Who was present during this conversation? Was everyone present you and XX (name patient) would have wanted?
- Did someone help you with this conversation?
- How was the timing of this conversation? Did you feel the timing was right? Why (not)?
- How did you experience this conversation?
- Was everything discussed what you and XX (name patient) wished to discuss? Would you this conversation again in the future? Why (not)?

End of interview

Has everything been discussed what you've had liked to discuss?

Interview patient- FC together: first interview

Introduction

XX (name patient) and you received XX months ago/ a while ago the diagnosis of ALS. Can you tell me something more about how it all started, about the process of being diagnosed?

- When did you notice the first signals/symptoms?
- When did you notice that there was something wrong?

When you have heard the diagnosis of XX (name neurologist). What was that for you? How did you feel? What did you think?

What were for you (as patient and as carer) the biggest changes since the diagnosis? (*Changes physically, emotionally, socially)*

Thinking about the future

What do you know about your disease/trajectory of the disease? Which impact does ALS have?

- How did you receive this information? Who told you about this?
- What did you think/feel when you've heard this?

Do you both think about your future? Is it something that you worry about? Are you thinking more about the future than before the diagnosis, or vice versa, do you think about it less? Or is it another way of thinking about your future? Could you tell me something more about this?

Option 1: Both telling that they think about the future:

- What do you expect? What do you hope for?
- What is important for you when you think about the future?
- What makes you worry about the future?
- With whom would you prefer to discuss the future?
- Do you discuss the future together? Why (not)?

Option 2: Both think about the future, but do not discuss this:

- Would you like to discuss it one day with each other? Why (not)?
- Do you know what (s)he thinks about? Do you know what (s)he would want or hopes or expects about the future? What do you think is important?

Option 3: In case one or both does not think about the future:

- You say you do not want to think about the future: How come? What makes you to refuse to think about the future?

- Is it something you are afraid of and why?

Thinking and talking about the future

Do you think about the care you might need in the future? What do you think about? What are your expectations? What do you worry about? What do you hope for? (patient)

Do you, as a carer think about the care XX (name patient) might need in the future? What do you think about? What are your expectations? What do you worry about? What do you hope for? (family carer)

- From whom did you receive information about the care?

Option 1: Both think about future care:

- Is it something you discuss together? Or with someone else? If so, who?
- Both indicate that they communicate openly about future care (go further to questions hereinafter).
- Both indicate they did not discuss future care, but both think about discussing it together:
  - What do you hope this conversation would trigger? What are your expectations?
  - What would you like to say in that conversation?
  - How would that be to discuss your worries about XX (name patient) about his/her future and vice versa?
  - When would it be important – according to you both – to discuss it?
  - How would you experience such conversation?
  - Who should ideally start this conversation? Would you start it? Would you like that someone is there to support you? If so, who?
  - Would you prefer to have this discussion alone or together with a HCP? Why?

Option 2: Both indicate they do not wish to think about future or future care:

- Why do you not wish to think about future care?
- How do you feel about the future care?
- How would you react/What would you do if an HCP wants to discuss future care and starts this conversation? Would you block it or would you talk about it? Why?

Preferences about the future/future care

What is important for you when you think about the future or future care?

What do you wish for? What are your preferences/ideas? Are there things you would want or would not want about care? Are there things about care you do not want? (patient)

Here we can give examples they have indicated during the conversation or ask how they felt about the wheelchair or walking cane etc* → ***ATTENTION*: listen carefully what the FC says during the* *interview → you cannot talk about care wishes they did not talk about. So <u>DON'T</u> talk about a stomach-pump if they did not talk about it): For example you might ask: how do feel about the wheelchair? OR I have heard you've made a lot of travels before your illness, is it something you wish to do? Did you discuss it? Did you make plans? ***

To family carer:

- What do you think, now you hear these preferences?
- How do you feel?
- Did you know that this was important for XX (name patient)? How did you know? Did you discuss this in the past? Why (not)?

OPTION 1: Patient expresses wishes and they have talked about these preferences/wishes with one another:

- I hear that you have discuss it. Was there someone else present during this conversation? If so, who?
- What have you discussed?
- What did the conversation trigger? Did you believe the moment was ideal? Why?
    - In case they did not believe the timing was ideal: When would be the best timing?
- During the conversation, have your feelings been taken into account about certain wishes/preferences/ideas/expectations/hopes/etc XX (name patient) has? Did you say how you felt/thought about it? Why and how?
    - In case not: What makes you did not say how you felt? What might happen if you did?
- Did you make concrete plans about the future/future care?
- Have you considered to write these wishes/preferences/etc down? Why (not)?
    - How do you feel about a living will?
- Imagine you need to make a medical decision instead of XX (name patient), would you believe that you can make this decision? In other words, do you know what XX (name patient) wants or does not want?
- How did the conversation end? What have you done afterwards?

OPTION 2: FC indicates (s)he did not know which preferences/wishes the patient has, but is glad that the patient gave some wishes and preferences during the interview:

- Do you believe that you would discuss these in more depth? Why (not)? Would this be something you would like to discuss alone with XX (name patient) or with a HCP or with kids/family/friends/etc?

- Who would initiate the conversation?
- What do you hope to achieve in this conversation? What are your expectations?

OPTION 4: Both did not say any wishes or preferences, and both indicate they do not wish to discuss it:

- Why do you not want to know the preferences/wishes of XX (name patient)?
- What is the worst possible thing that might happen if you would discuss it?

Preferences about the conversation about future/future care

OPTION 1: Both did not discuss wishes/preferences with patient/HCP/

- When would be the best timing to have this discussion with XX (name patient)?
- What makes it difficult to start this conversation?
- How do you think this conversation might go?
- How would you feel during such conversation?

Option 2: In case they have had this discussion:

- What triggered the conversation? How did it start?
- Who was present during this conversation? Was everyone present you and XX (name patient) would have wanted?
- Did someone help you with this conversation?
- How was the timing of this conversation? Did you feel the timing was right? Why (not)?
- How did you experience this conversation?
- Was everything discussed what you and XX (name patient) wished to discuss? Would you this conversation again in the future? Why (not)?

End of interview

Has everything been discussed what you've had liked to discuss?

Interview guide for patient with ALS: subsequent interviews (interview 2 and 3)

NOTE: This interview guide only shows the main questions, because the subsequent interviews build upon what has been said in the previous interview.

Introduction

Could you tell me something more about the past 3 months, since our last conversation?

Did you think about our last conversation? Do you wish to clarify something?

What were for you the biggest changes since our last conversation? (*Changes physically, emotionally, socially)*

Thinking about the future

In the previous conversation, we have discussed thinking about the future. You told me XX (what they have told)

- How do you think about it now? Did it change? Why (not)?

Thinking and talking about the future

In the previous conversation, we have discussed talking about the future and then you have indicated you did (not) discuss the future with XX (name family carer) or with someone else

- Did you have this discussion in the past months? Why (not)?

OPTION 1: Patient did not talk about the future, and do not wish to talk about it:

- What makes it difficult to think about the future?
- Does XX (name FC) has the same feeling?
- How would you react when XX (name FC) starts the conversation? Would you block it? Why (not)?
- What are you afraid of that might happen if you talk about this?

OPTION 2: Patient thinks about the future, and wish to talk about the future, but family carer blocks the conversation:

  - How do you feel about this?
  - What do you do when that happens?
  - What would you like to tell him/her?

OPTION 3: Patient thinks about the future, and has discussed it

- What triggered the conversation?
- Who was present? Was everyone present you would have liked to? Why (not)?

- Who supported you during this conversation?
- What made it for you the best time to discuss it? Or would you have preferred another time?
- Would you discuss it again in the future? Why (not)?
- **NOTE:** ask how the previous interview influenced the possible conversation

Preferences about the future/future care

In the previous interview, we have discussed preferences about the future/future care. You said following things XX

- Did wishes/preferences/ideas change during these past months? Why (not)?
- In case wishes/preferences/ideas have changed
    - What triggered the change in your wishes/preferences/ideas?
    - Did you discuss these changes?
        - If so, with whom?
        - If not, why not?
- **NOTE:** ask how the previous interview influenced the possible change

In the previous interview, you told me that:

OPTION 1: you would want to discuss your preferences in the near future:

- Have you done this in the past months? Why (not)?
- In case family carers blocks the discussion:
    - How do you feel about this?
    - What do you do when that happens?
    - What would you like to tell him/her?

OPTION 2: you have discussed your preferences prior to the first interview with XX (the persons they have indicated in the previous interview to whom they have discussed it)

- Have you discussed it again in the past months? Why (not)?

OPTION 3: I hear you have discussed your preferences in the past months

- What triggered the conversation?
- Who was present? Was everyone present you would have liked to? Why (not)?
- Who supported you during this conversation?
- What made it for you the best time to discuss it? Or would you have preferred another time?
- Would you discuss it again in the future? Why (not)?

OPTION 4: you have not discussed your preferences and you do not wish to discuss this

- What makes it difficult for you to talk about all this?
- What is the worst that may happen if you would think about it?
- Is there something you are afraid of?

Preferences about the conversation about future/future care

In the previous conversation you have told me that the ideal timing for this conversation would be XX

OPTION 1: the ideal timing has arrived, but patient did not have this conversation

- Why did you not have this conversation? What stopped you?

OPTION 2: the ideal timing did not arrive, and there was no discussion in the meantime:

- Do you still have the same idea about the ideal timing to have this conversation? Why (not)?

OPTION 3: the ideal timing did not arrive, but patient did have a discussion in the past months:

- What triggered the conversation?
- Who was present? Was everyone present you would have liked to? Why (not)?
- Who supported you during this conversation?
- What made it for you the best time to discuss it? Or would you have preferred another time?
- Would you discuss it again in the future? Why (not)?

End of interview

Has everything been discussed what you've had liked to discuss?

Interview guide for FC: subsequent interviews (interview 2 and 3)

NOTE: This interview guide only shows the main questions because the subsequent interviews build upon what has been said in the previous interview.

Introduction

Could you tell me something more about the past 3 months, since our last conversation?

Did you think about our last conversation? Do you wish to clarify something?

What were for you the biggest changes since our last conversation? (*Changes physically, emotionally, socially)*

Thinking about the future

In the previous conversation, we have discussed thinking about the future. You told me XX (what they have told)

- How do you think about it now? Did it change? Why (not)?

Thinking and talking about the future

In the previous conversation, we have discussed talking about the future and then you have indicated you did (not) discuss the future with XX (name patient) or with someone else

- Did you have this discussion in the past months? Why (not)?

OPTION 1: FC did not talk about the future, and do not wish to talk about it:

- What makes it difficult to think about the future?
- Does XX (name patient) has the same feeling?
- How would you react when XX (name patient) starts the conversation? Would you block it? Why (not)?
- What are you afraid of that might happen if you talk about this?

OPTION 2: FC thinks about the future, and wish to talk about the future, but patient blocks the conversation:

- ○ How do you feel about this?
- ○ What do you do when that happens?
- ○ What would you like to tell him/her?

OPTION 3: FC thinks about the future, and has discussed it

- What triggered the conversation?
- Who was present? Was everyone present you would have liked to? Why (not)?

- Who supported you during this conversation?
- What made it for you the best time to discuss it? Or would you have preferred another time?
- Would you discuss it again in the future? Why (not)?
- **NOTE:** ask how the previous interview influenced the possible conversation

Preferences about the future/future care

In the previous interview, we have discussed preferences about the future/future care. You said following things XX

- Do you believe the wishes/preferences/ideas of XX (name patient) has changed during these past months? Why (not)?
- In case wishes/preferences/ideas have changed
  - What triggered the change in their wishes/preferences/ideas?
- **NOTE:** ask how the previous interview influenced the possible change

In the previous interview, you told me that:
OPTION 1: you would want to discuss the preferences with XX (name patient) in the near future:

- Have you done this in the past months? Why (not)?
- In case patient blocks the discussion:
  - How do you feel about this?
  - What do you do when that happens?
  - What would you like to tell him/her?

OPTION 2: you have discussed with XX (name patient) the preferences prior to the first interview

- Have you discussed it again in the past months? Why (not)?

OPTION 3: I hear you have discussed the preferences with XX (name patient) in the past months

- What triggered the conversation?
- Who was present?
- Who initiated the conversation? You or XX (name patient) or someone else?
- Who supported you during this conversation?
- What made it for you the best time to discuss it? Or would you have preferred another time?
- Would you discuss it again in the future? Why (not)?

OPTION 4: you have not discussed the preferences with XX (name patient) and you do not wish to discuss this

- What makes it difficult for you to talk about all this?
- What is the worst that may happen if you would think about it?
- Is there something you are afraid of?

Preferences about the conversation about future/future care

In the previous conversation you have told me that the ideal timing for this conversation would be XX

OPTION 1: the ideal timing has arrived, but FC did not have this conversation

- Why did you not have this conversation? What stopped you?

OPTION 2: the ideal timing did not arrive, and there was no discussion in the meantime:

- Do you still have the same idea about the ideal timing to have this conversation? Why (not)?

OPTION 3: the ideal timing did not arrive, but FC did have a discussion in the past months:

- What triggered the conversation?
- Who was present?
- Who initiated the conversation? You, XX (name patient) or someone else?
- Who supported you during this conversation?
- What made it for you the best time to discuss it? Or would you have preferred another time?
- Would you discuss it again in the future? Why (not)?

End of interview

Has everything been discussed what you've had liked to discuss?

Chapter 2:

Experiences with advance care planning in Amyotrophic Lateral Sclerosis (ALS): Qualitative longitudinal study with persons with ALS and their family carers

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This chapter is submitted

Abstract

Background: It is unclear when and how people with ALS and their family carers think about their future, what they would prefer in terms of care or treatment, and how their ideas change over time.

Aim: To understand experiences with advance care planning (ACP) of persons with ALS and their family carers – and if, when, how, and why these experiences change over time.

Method: We conducted a qualitative longitudinal interview study with 9 persons with ALS and 9 family carers. We interviewed them every 3 months over a 9-month period. We obtained an in-depth understanding of the data via content analysis, which resulted in overarching themes, and, via timetables, we were able to describe the changing experiences with ACP.

Results: All participants thought about the future and future care, but few talked about it. Over time, ACP experiences were influenced by several intertwined elements: (1) the experienced physical decline and related future care needs; (2) how persons with ALS identify themselves as patients; (3) obtaining information about diagnosis and prognosis; (4) professionals initiating conversations about medical aspects of end-of-life decisions; (5) balancing between hope to remain stable and worry about the future; (6) protecting themselves and each other from worries about the future.

Conclusion: The complexities involved in ACP are highlighted. Various factors play a role in constructing people's ideas, thoughts and feelings about future care and treatment. The patient, their family carer, and the professionals all play an interrelated role in constructing these experiences.

Introduction

Amyotrophic Lateral Sclerosis (ALS) is a degenerative motor neuron disease in adults, affecting the brain and spinal cord.[1] ALS is incurable and characterized by progressive muscle paralysis; the most common cause of death is respiratory failure.[2,3] The average survival rate is three to five years, which is significantly shorter than other neurological diseases, such as dementia or multiple sclerosis.[1,4–7] Over the past decade, research in Europe, Australia and the US has shown that persons with ALS often experience physical, emotional, and existential problems that persist until the end of life – and to date, these complex needs often remain unmet.[8–11] Given the incurable nature of ALS, combined with its sometimes rapid progression and unmet needs, a palliative care approach – in which Advance Care Planning (ACP) plays a prominent role – has been widely advocated for this population[10–12]. ACP is defined as a continuous, early-initiated communication process between patients, family carers and/or healthcare professionals to support patients and family carers in understanding and sharing their personal values, life goals, and preferences regarding future medical (end-of-life) care[13].

Among current discussions of ACP in different populations[14], a 2016 systematic review of ACP in ALS indicated important advantages in the uptake of ACP, such as increased feelings of control and relief, and refusal of unwanted treatments[15]. Most studies on ACP in ALS focus on the risks and benefits of life-sustaining interventions (such as ventilatory support and gastrostomy), the identification of a surrogate decision-maker, or the completion of an advance directive[8,9,16]. However, recent recommendations highlight the need for ACP to be seen as a series of broader conversations and reflections over time about hopes, preferences, and potential care goals[14,17].
Up to now, it is unclear when and how topics about future care arise, how they change over time, and which factors influence them, from the perspective of the patient as well as of the family carer. The aim of this study is to understand the experiences of persons with ALS and their family carers with respect to ACP, and to explore whether these experiences change over time, and, if so, when, how and why.

Methods

Study design

This study has a qualitative, longitudinal, multi-perspective interview design to explore an evolving and complex process such as ACP over time from the perspective of persons with ALS and their family carers[18,19]. This design offers considerable advantages over more typical 'snapshot' techniques in understanding participants' changing experiences[19]. We used constructionism as an underlying epistemology,[20] as we wanted to know more about the views and meanings constructed by persons with ALS and their family carers concerning experiences with ACP.

Setting

In Belgium, specialist care for persons with ALS is usually organized through neuromuscular reference centers that have an advisory role, with expertise in neurology, respiratory care and rehabilitation, as well as psychology, physiotherapy, occupational therapy, speech and language pathology, nutrition and social work[21,22]. The neuromuscular reference centers are connected to university hospitals. To obtain advice from these centers, persons with ALS and their family carer need to be referred by the neurologist from the hospital. In addition to the neuromuscular reference centers, care can also be provided through general practice, community-based services (e.g. home care nurses) and palliative care home teams, but these services are not part of the neuromuscular reference centers[21,22].

Participants and recruitment

We recruited persons with ALS and their family carer via 3 neurologists (1 local and 2 university hospitals) located in Flanders, Belgium. Eligible persons with ALS and their family carers were approached by their treating neurologist to ascertain willingness to participate in the study. Inclusion criteria were: 1) the treating neurologist had communicated the ALS diagnosis to the patient and their family carer not more than 6 months ago; 2) the person with ALS and their family carer were older than 18; 3) both were able to sign a written informed consent. Participants were excluded if they could not speak Dutch or if the person with ALS was diagnosed with frontotemporal dementia. Potential participants who permitted their neurologist to pass their contact information to the researchers were contacted by a member of the research team (IV) to discuss the purpose of the study and, if they wished to participate, to schedule the first interview. Further information on recruitment can be found in the protocol paper[23].

Data collection

We interviewed persons with ALS and their family carers at 3 different times, every 3 months over a period of maximum 9 months between February 2021 and March 2022. Interview guides for the first interviews were developed and cognitively tested with 3 dyads before the start of our study. Each interview built upon the previous one and was adapted according to what had been discussed in the previous interview to identify possible changes in experiences. When changes had occurred, we investigated during the interview what had triggered the change and whether and how the interviewer had influenced the change. Detailed information about the interview guides can be found in the protocol paper and the appendix of the protocol paper[23].

We aimed to interview the participants every 3 months, but the timing remained flexible in case the experiences with ACP suddenly changed. Therefore, we performed short monthly phone calls with the patient or family carer to assess whether an interview should be planned sooner than anticipated due to a changing event (e.g. unexpected hospitalisation). These phone calls were not audiotaped or analysed. Detailed information can be found in the protocol paper[23].

Data analysis

Qualitative longitudinal analysis is an iterative and multi-dimensional process, which starts with multiple readings of the data. First, we did a content analysis – which involves line-by-line coding – to obtain an in-depth within-case understanding of the data[24,25]. A coding list was constructed for each case separately and from each timepoint: that is, timepoint 1: first interviews; timepoint 2: second interviews; and timepoint 3: third interviews. Two researchers (IV & ADV) independently assigned codes to each timepoint and discussed the codes until consensus was reached, which resulted in a coding list. This coding list was refined within the research team by grouping the codes into categories and themes. Second, to describe changes in experiences with ACP among all the participants, we made a timetable for each dyad and each interview to see what had changed over time, how themes overlap and how they interconnect with each other. This gave us a clear image of the participants' journeys and the overarching themes over time. Finally, we did a constant comparison within and between the dyad timetables to delineate characteristic patterns in the sequences of ACP experiences[26]. To limit subjectivity, we discussed the results of this timeline within the research team.

Ethical considerations

Ethical approval has been granted by the central ethical committee of the University Hospital of Brussels, (B.U.N. B1432020000128) and the other participating hospitals. Detailed information about the various safeguards that were considered for this study can be found in the protocol paper[23].

Results

In total, we included 9 dyads (i.e. 9 persons with ALS and 9 family carers) in the study. Re-recruitment was necessary for 1 dyad, as one family carer decided to leave the study after the first interview. One dyad dropped out of the study after two interviews, due to the death of the person with ALS. Most participants were interviewed individually; however, one dyad wanted to be interviewed together for all three timepoints, because the person with ALS experienced difficulties with speech. Two dyads were interviewed together at timepoint 1, but individually at timepoints 2 and 3. Patients' and family carers' characteristics are described in Table 1.

TABLE 1 Characteristics of the respondents (n=18)

Sample Characteristics	**Persons with ALS (n=9)**	**Family carer (n=9)**
Sex		
Male	3	4
Mean age in years	63.78	55.22
Educational level		
Primary school	-	1
Secondary school	7	6
Higher education (college or university)	2	2
Degree in care (e.g. nurse or doctor)		
Yes	3	1
Work status at time of diagnosis		
Full-time	3	5
Part-time	2	2
Retired	4	1
Other (sick leave)	-	1
Change in work status since diagnosis		
Yes	7	2
Mean duration of the interviews in minutes		
Interview 1/Timepoint 1	56'50"	55'66"
Interview 2/Timepoint 2	67'12"	61'63"
Interview 3/Timepoint 3	75'30"	66'15"
Characteristics of the dyads (persons with ALS and FC)		
Spousal relationship	6	
Parent-child relationship	2	
No family	1	

All dyads had some experience with ACP, mostly by contemplating their future trajectory and future care. Only several dyads openly talked to each other about these topics. Some had discussions about specific future medical aspects (e.g. gastrostomy, tracheotomy) or end-of-life decisions (e.g. euthanasia).

Over time, ACP experiences were influenced by several intertwined elements: (1) the experienced physical decline and related future care needs; (2) how persons with ALS identify themselves as patients; (3) obtaining information about diagnosis and prognosis; (4) professionals initiating conversations about medical aspects of end-of-life decisions; (5) balancing between hope to remain stable and worry about the future; (6) protecting themselves and each other from worries about the future.

(1) the experienced physical decline and related future care needs

At each timepoint, participants always started by talking about the physical decline that the patient had experienced in the previous months, and some of them wondered about what would happen next. The most common topics that persons with ALS initially thought about were: how the disease would evolve in the future; whether they would be able to stay at home in the future; what equipment (such as a wheelchair or speech computer) might be needed in the future; possible house renovations; and who would take care of them (e.g. family carer or home care nurses).

"(if I can't use both of my hands) I can't be home anymore, I don't think so. If I have to go to the bathroom, I have to call someone to ask them to help me.(…). And to have someone around constantly from morning to evening, er, I don't know.' (Dyad 7, person with ALS, 74y, male, interview 1)

Family carers worried primarily about how the disease would evolve in the future, and also whether they would be able to handle future care, how their loved one would remain comfortable, and what it would be like after the person with ALS has died. These topics continued to be mentioned throughout the interviews.

'[I lie awake at night thinking] about the care and how it will evolve, and about when he will die, because I will be alone here (…) I don't like being alone (Dyad 9, family carer, 56y, female, interview 3).

However, at timepoint 1, some patients and family carers also felt these worries were for a distant future. Many participants mentioned trying to live with a 'day-by-day' attitude as much as possible, because thinking about the future might bring too much emotional distress or sad feelings.

At timepoints 2 or 3, participants often said that an impactful deterioration in the person with ALS (e.g. not able to go upstairs/not able to walk independently) had led to an immediate reaction in terms of planning, such as renovations for a bedroom and bathroom downstairs or applying for a wheelchair. This physical decline triggered some dyads to think, and a few (3 dyads) to talk, about the future, and for some participants this facilitated planning for equipment or care that might be needed in the future. However, not all patients were ready to discuss these topics with their family carer following physical decline, even though the family carer wanted and tried to talk about them.

"Well, I also sometimes have the feeling, er, when we talk about the illness, that she sometimes says 'pff not now' (…) I then think: I try to help you and it's like you don't care, and that hurts, of course (…) but then she starts to cry and says 'I don't want this disease'." (Dyad 3, family carer, 40y, male, interview 1)

(2) how persons with ALS identify themselves as patients

During the interviews, five persons with ALS also reflected on how they identified themselves in their new role as a patient. More specifically, on not wanting to become dependent, or about having to accept that they would become dependent on others and on equipment (e.g. wheelchair or speech computer) in the future.

"I realize it enough. That I am declining and that I can no longer do things, or that things are not going well, but the hardest part is accepting." (Dyad 3, person with ALS, 44y, female, interview 2)

Most of the patients interviewed expressed their wish to maintain their independence for as long as possible, because they did not want to be a burden on their family carer. Most family carers maintained that they were happy to help and that the care they were giving now was not burdensome. Only one family carer expressed (at timepoint 3) that he found care was difficult, because his wife did not want to use any necessary equipment, such as a walker or speech computer.

"She doesn't like to walk with that [a walker], I understand that. (…) It's easier for us (…) it is. For us it's a convenience, but for her it is still a step backwards." (Dyad 2, family carer, 59y, male, interview 3).

Related to becoming dependent, six persons with ALS found some equipment too confronting to use – such as a walker to facilitate walking – and wanted to postpone it for as long as possible. It seemed that their family carers, or the patient's friends or family, played an important role in either facilitating or hindering the use of equipment. Over time, patients seemed to accept their role as patient in case administration applying for budget or for equipment, but also when family carers helped patients to find peace within their new life, such as being positive about using the wheelchair. Patients accepting their role were more open to thinking – and some even to planning and talking – about the future and future care.

"Our oldest grandson recently said I don't want to look at that [seeing the patient's wheelchair]. Of course, that did not help." (family carer, dyad 8, interview 2)

(3) obtaining information about diagnosis and prognosis

At timepoint 1, almost all participants – except one person with ALS – indicated that they had obtained information about the diagnosis and prognosis either via professionals or the internet. According to the participants, most professionals mentioned shortly after diagnosis that the disease evolves differently for everyone. This often led many participants to have a day-by-day attitude, as they felt that the deterioration was unpredictable. At timepoint 1, most participants said that they searched the internet for information about the disease and its progression, and what the future might hold. However, most of them reported they had stopped searching the internet, because all the devastating stories brought too much negativity.

"I looked it up, but I didn't go far. (…) I know I'm always going to deteriorate, but how? Well, no one knows." (Dyad 3, person with ALS, 44y, female, interview 1).

Over time, some participants expressed information needs about future care (such as applying for supportive equipment). When the interviewer explained the option to contact the psychologist or neurologist from the neuromuscular reference center to discuss these matters, these participants said that they felt their questions were not urgent and the matter could wait until their appointment, which was usually a few months away.

"Yes, I can always contact them, but then I have to go there and I have to go in August anyway, so I can wait a bit. It's not that urgent now." (Dyad 4, person with ALS, 64y, female, interview 2)

One dyad watched a movie about ALS between timepoints 1 and 2. The family carer and patient were shocked to learn that the disease progression could be so fast. Nonetheless, this film helped the patient and her family carer to discuss the future and future (end-of-life) care.

"I thought, you can live with that [ALS] for years and years, but in the film it's only three to five years. (…) No one told me specifically." (Dyad 3, family carer, 40y, male, interview 2)
"That's when I realized how it was going to evolve. I knew what the disease was, but I didn't know how it was going to evolve. And then I made a statement: I decide when it's enough. (…) I've already said that to everyone, yes." (Dyad 3, person with ALS, 44y, female, interview 3)

(4) professionals initiating conversations about medical aspects of end-of-life decisions

Almost all participants thought, and 5 dyads talked among each other, about medical end-of-life decisions, in response to their treating physician or someone from the neuromuscular reference center who initiated a conversation about the possibilities and risks of a gastrostomy, a tracheotomy, and euthanasia. This was most often brought up by professionals shortly after diagnosis and/or in follow-up consultations, which usually led to thinking about these medical aspects. Some patients immediately expressed their preferences to their family carer, while other patients needed more time to open up. In the interviews in which the participants mentioned euthanasia, the patients indicated that they were thinking about euthanasia should they have respiratory failure and would need extra oxygen or a tracheotomy, because they were afraid of suffocation.

"If my breathing gets harder, it's just euthanasia, that's what I've decided." (Dyad 9, person with ALS, 59y, male, interview 1 together with the family carer)

Regarding whether a gastrostomy would be an option, most persons with ALS were in doubt. They explained that the stage of the disease, and how they would feel about this procedure at that moment, would be the deciding factor. For most persons with ALS, their view about this topic remained stable over time. Family carers indicated that they would respect their loved one's choice concerning (not) wanting a gastrostomy, a tracheotomy or euthanasia.

(5) balancing between hope to remain stable and worry about the future

There was a constant balance between hope to remain stable and worry about the future and future care, which was expressed throughout all the interviews. At timepoint 1, all family carers said they hoped that their loved one would remain stable or maintain their quality of life as much as possible or that their loved one would grow old with the disease. The patients also hoped to remain stable or independent for as long as possible, but they also hoped to travel and enjoy the little things in life. Hope about stability, independence and being comfortable remained stable over time, even though worries became more prominent as the disease progressed, and this was a trigger for some family carers and patients to talk about the future and future care with each other.

For some family carers, the worries only increased over time, especially when the family carer could not talk about his or her worries with their loved one.

"I am very, very, very afraid of that deterioration process. (…) You are confronted with it more and more, so you think about it more and more. (…) I used to have a hard time with it, but it's EVEN MORE difficult now." (Dyad 2, family carer, 59y, male, interviews 1 & 2)

Worries about the future were present for most of the patients as well – most often existential or psychological questions, such as feeling that they were being punished or the need to talk to a psychologist – but these did not increase as much over time as compared with the family carers. Interestingly, in each interview, when the person with ALS had previously expressed a wish for euthanasia if they would experience difficulties with swallowing or breathing, the family carer would say that they were glad that swallowing and breathing had not declined in the previous months.

"(name of person with ALS) is a bit deteriorated, especially in the limbs. But I'm glad he can still speak, and swallow. I was most afraid that that would deteriorate." (Dyad 9, family carer, 56y, female, interview 2)

(6) protecting themselves and each other from worries about the future

At each timepoint, it became apparent that persons with ALS as well as family carers tried to protect themselves and each other from their worries about the future and future care, which made it difficult to talk about these things with each other. This protective attitude did not change over time. The majority of family carers tried to be protective – not only for their loved one with ALS but also for themselves (e.g. the emotional distress in talking about euthanasia). The family carers' way of being protective towards the patient and/or themselves was by trying to be optimistic whenever their loved one tried to talk about his or her decline – "try to focus on what you can do" – or by 'blocking' or postponing the conversation when their loved one tried to talk about the future and/or future (end-of-life) care ("you don't need to think about this yet"). Reflecting about this during the interviews made them think about what the person with ALS wanted to say, but they did not want to initiate this conversation.

"I'm not really going to ask. Maybe that's a mistake on my part, that I should [ask]. I always say 'But Dad… you're still here.'" (Dyad 7, family carer, 48y, female, interview 3)

Over time, the patients also tried to protect their loved ones by not talking about having a bad day, feeling sad, or the fact that they fell during that day. For most of the persons with ALS, having a bad day meant that they were in pain or that they were alone and thought negatively about the future and future care, more specifically about dying or what would deteriorate next. Most of the persons with ALS did not want to talk about these things with their family carer, because they did not want to burden or alarm or distress them.

"I'm going to have to talk about how we're going to do it. I also don't know. I'm afraid to talk about it to [name of family carer], she immediately starts to cry. (...) She also has a hard time talking about it. (She says) You're not going to die yet." (Dyad 7, person with ALS, 74y, male, interview 2)

Discussion

Main findings

This longitudinal qualitative study showed that all persons with ALS and their family carers thought about the future and future care to some extent, but only a few dyads talked about it. Over time, experiences with ACP were influenced by several intertwined elements: (1) the experienced physical decline and related future care needs; (2) how persons with ALS identify themselves as patients; (3) obtaining information about diagnosis and prognosis; (4) professionals initiating conversations about medical aspects of end-of-life decisions; (5) balancing between hope to remain stable and worry about the future; (6) protecting themselves and each other from worries about the future.

Strengths and limitations

This study has several strengths. First, this is the first study to provide longitudinal, in-depth and multi-perspective insights into the temporal dynamics of the complex process of ACP in a unique patient population of ALS and their family carers. Second, longitudinal qualitative studies and multi-perspective interviews are innovative methods in medicine and especially in the palliative care field[19]. Longitudinal qualitative studies, when combined with flexibility, are a less restrictive approach towards studying change over time in complex processes such as ACP[27,28]. Finally, monthly phone calls in between the three interviews facilitated a more nuanced understanding of the illness trajectory in case a sudden change appeared. As a result, we were able to conduct an interview that would otherwise have been missed because the patient would have died.

This study also has some limitations. First, there might be some selection bias, as the participants who wanted to participate were possibly more interested in this topic. Nevertheless, we attempted to counteract this bias by not using the term ACP but rather a broader description such as thinking or talking about future and future care. Another limitation is that we were not able to follow up the experiences with ACP until the end-of-life for all dyads. Nonetheless, our sample consisted of people with ALS in different stages of progression in the disease trajectory, and therefore we were able to capture in-depth views of experiences with ACP throughout the disease trajectory.

Interpretation of the main findings

Although most persons with ALS and their family carers in this study indicated that they wanted to live in the now, we found that, at the same time, they were thinking about the future and future care. Most often, they reflected on what would happen next, and whether family carers could handle the future care, rather than thinking about the medical aspects of end-of-life decisions such as euthanasia. Considerations specifically about end-of-life decisions were most often triggered by a professional who initiated such conversations. On the one hand, this indicates that, for patients and their families, ACP is seen as more than a medicalized process about end-of-life decisions or filling out advance directives[29–31], and that ACP also includes psychosocial factors such as relational dynamics and coping with the disease. On the other hand, it shows that professionals often emphasize the medical part of the process over the social or psychological parts of ACP[32,33]. Although the literature states that discussions about gastrostomy, tracheotomy or euthanasia should happen early on in the ALS disease trajectory[8,9,16], it was apparent that these conversations were often perceived – most often by family carers – as being too early or stressful. The need to see ACP as a holistic process in which mental, social and medical processes are intertwined has also been highlighted in other research in other populations[34–36], which indicates that these conversations should focus on the patients' and family carers' hopes, worries and needs.

In this study, experiences with ACP also seemed to be shaped over time by several interrelated factors related to coping with the severity of ALS. Studies on coping in ALS have shown that most patients wish to maintain a sense of control by adjusting to and accepting their new life[37,38], but this is often difficult due to the physical losses[39–41]. This also became apparent in our results. An important theme with regard to coping was the balance between hope to remain stable and worry about what the future might hold. Hope can be helpful in coping with, or controlling, the emotional distress that arises when living with a chronic life-limiting illness[42]. However, the physical decline triggered all participants to think about what the future might hold. Not all of them were prepared to talk about this amongst each other, most often because they wanted to protect one another and themselves from emotional distress during these conversations. Studies have shown that healthcare professionals can play an important role in supporting and facilitating these conversations between family carers and patients[43,44]. Our results show that psychosocial factors, such as coping mechanisms or relational dynamics, are inextricably connected with the ACP process[34] and cannot be simplified to conversations about medical end-of-life decisions or completing advance directives[29–31]. More attention to these ACP complexities is needed in future research, policy and practice[14].

Conclusion

This study has highlighted the interrelated factors shaping the experiences of the ACP communication process in the context of a terminal illness such as ALS. Different factors related to coping with the disease or relational dynamics play a role in constructing people's ideas, thoughts, and feelings about future care and treatment preferences. Psychological, social and medical factors are interwoven and cannot be seen as separate entities in ACP.

Additional information

Authorship

I.V., A.D.V, E.C., L.V.D.B & L.D. designed the study. I.V. conducted the interviews and transcribed the interviews. I.V. & A.D.V. analysed the interviews. All authors contributed to the interpretation of the results. I.V. wrote the manuscript and all authors critically revised the manuscript and approved the final version.

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Conflicts of interest

The authors declare that there is no conflict of interest.

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Part II: family carers’ involvement in advance care planning and their evaluation of end-of-life care

Chapter 3:

Support from healthcare professionals in empowering family carers to discuss Advance Care Planning: A population-based survey

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Abstract

Background: Family carers have a prominent role in end-of-life care for seriously ill persons. However, most of the Advance Care Planning literature is focused on the role of healthcare professionals.

Aims: To investigate (1) what proportion of family carers discussed advance care planning with their relative and associated socio- demographic and clinical characteristics (2) what proportion received support from healthcare professionals for these conversations, (3) what type of support they received and (4) to what extent the type of support received was considered sufficient.

Design/participants: Population-based cross-sectional survey in Belgium of bereaved family carers of persons with a serious chronic illness (N=3000) who died two to six months before the sample was drawn, identified through three sickness funds. The survey explored support from healthcare professionals for family carers during the last three months of the patient's life.

Results: Response rate was 55%. The proportion of family carers that engaged in an advance care planning conversation with their relative was 46.9%. Of these family carers, 78.1% received support from a healthcare professional, mostly by doing the advance care planning conversation together (53.8%). Of family carers receiving support from a healthcare professional, 57.4% deemed the support sufficient.

Conclusion: Many family carers engage in advance care planning conversations with their dying relative. Healthcare professionals often support them by performing the advance care planning conversations together. More insight into how family carers can be supported to conduct these advance care planning conversations, both with and without involvement of healthcare professionals, is necessary.

Introduction

Advance care planning is defined as an early-initiated dynamic communication process between patients, family carers and healthcare professionals that enables individuals to define goals and preferences for future (end-of-life) care[1,2]. The goal of advance care planning is to have a timely exploration of a person's underlying values, wishes, and preferences for future end-of-life care in order to develop a shared narrative and understanding to inform patient-centered care, which becomes particularly important if a patient loses the ability to make their own decisions about care[1,2]. Involvement of family carers is crucial in this process since they often have a prominent role in care decision-making at the end of life[3], and it has been shown that involvement in advance care planning improves family carers' confidence when making decisions at the end of life on behalf of their relative[4].

Recent interventions mostly focus on improving advance care planning between the patient and/or family carers and healthcare professionals[5,6], however qualitative studies have shown that patients and family carers also want to discuss future care and preferences among each other[7–9]. Moreover, previous studies found that healthcare professionals experience many barriers in initiating advance care planning conversations, such as lack of time or not knowing when or how to start these conversations[10,11]. Patients also mostly turn to professional carers for physical and disease-related problems, but do not want to burden them with discussing their psychosocial or existential needs[12].

Family carers' time spent caring for their relative is often more substantial than the time spent by healthcare professionals[13], which means that there are many opportunities to discuss advance care planning within the home setting. Although family carers are mostly willing and welcoming to have these conversations, they often feel unprepared, anxious or uncomfortable to do so[14–17]. Healthcare professionals can play an important role, for instance by facilitating advance care planning conversations between patients and family carers in the home setting; performing the advance care planning conversation together with them or by empowering the family carers to increase their self-efficacy for initiating such conversations by themselves. Providing family carers with tools, such as conversations cards, to initiate advance care planning discussions are practical examples of such approaches[18,19].

The aim of this study was to examine the extent to which family carers of people with a serious illness performed advance care planning conversations with their relative at home in the last 3 months of life and how they were supported by healthcare professionals to conduct these conversations.

Specific research questions are:

1) What proportion of family carers discussed advance care planning with their seriously ill relative in the last 3 months of life and what socio-demographic and clinical characteristics are associated with having such discussion?
2) What proportion received support from healthcare professionals for these conversations?
3) What type of support did they receive?
4) To what extent was the type of support received from healthcare professionals considered as sufficient?

Methods.

Study design and setting

A population-based nationwide cross-sectional survey in Flanders, Belgium was conducted among bereaved family carers. In Belgium, people with long-term extensive care needs can apply for a monthly care budget that helps cover non-medical care. We used registers of the three largest health insurers (accounting for 79% of the population) in Flanders to identify participants. The insurers maintain records people who apply for this care budget and the names of their family carers who are also registered during the application process. The Strengthening of the Reporting of Observational studies in Epidemiology (STROBE)[20] was used to describe all relevant aspects of the study.

Population

We randomly sampled 3000 deaths of people with a serious illness who applied for a care budget -indicative of increased informal care needs- from one of the participating health insurers. For these cases, the person registered in the database as their family carer was selected. We only included family carers who had provided care to someone who recently died between two and six months before inclusion to allow sufficient time for grieving while limiting recall bias[21–23]. Additional inclusion criteria were correct postal address and that both (deceased relative and family carer) were older than 18 years.

Sampling and Selection of the participants

Random sampling with proportional stratification was used to obtain a distribution among the different health insurers reflecting the actual distribution of registered family carers in the total population (Christelijke Mutualiteit n=2,748; Socialistische Mutualiteit n=157; Liberale Mutualiteit n=95). To answer the research questions accurately, family carers were included for analysis in case 1) they had contact with the deceased person during the last three months of life (question 2 in questionnaire); and in case 2) the deceased person had at least one of the listed serious illnesses (question 3 in questionnaire).

Data collection

Questionnaires were sent to the family carers via post between November 2019 and January 2020 by an independent data collector, including an information letter with details of the study, the voluntary nature of the study and the option to complete the questionnaire online. Informed consent was provided as part of the returned the questionnaire. In case of non-response the family carers received two reminders by post (after two and four weeks of non-response). A non-response questionnaire was sent no longer than two weeks after the final reminder.

The questionnaire

As we could not identify an appropriate pre-existing validated instrument(s) to address our research questions we utilized a combination of validated[24] and self-developed items. The questionnaire was tested through two rounds of cognitive interviews with 5 family carers whom were recruited via convenience sampling. The cognitive testing resulted in two additional questions (question 2 and 4), changing the sentence structure of certain items and clarifying certain concepts. The final version of the questionnaire can be found in Appendix 1.

In this study, we focused on the psychosocial task " advance care planning discussions", which was defined in the questionnaire as follows: *'Discussing with my relative about end-of-life care planning (e.g.) the care that my relative wanted or did not want at the end-of-life, advance directives, a living will, wishes about euthanasia or other end-of-life decisions such as palliative sedation'*. For each caregiving task, the family carer was asked: 1) Did you perform this task at least once during the last three months of the patient's life? (yes/no); 2) Which healthcare professionals have supported you in this task? (multiple answers possible including no support was received from healthcare professional); 3) How were you supported in this task by healthcare professionals (multiple answers possible), and 4) To what extent was the support of healthcare professionals sufficient to perform this task (4-point scale).

Statistical analysis

Data were analysed using SPSS Statistics version 26. Descriptive statistics were used to summarize the data, therefore we adopted frequency tables and contingency tables with the χ^2-test. To describe which factors were associated with advance care planning discussion, we used multivariable binary logistic regression (advance care planning discussion vs no discussion as dependent variable). The potential predictors included: bereaved family carer characteristics (gender, age in years, educational attainment, medical degree, work status); deceased patient characteristics (serious chronic illness, age in years); and care characteristics (involvement of other family carers, patient lived at home in the final three months, and palliative care services involved) were included in a hierarchical stepwise model selection. We started with a univariable binary logistic regression model and selected for the stepwise model only the variables which were (marginally, i.e $p<0.1$) statistically significant. Next, three multivariable binary logistic regression models were constructed: 1) deceased patient characteristics, 2) adding bereaved family carer characteristics, 3) adding care characteristics. Only variables with $p <.05$ were retained for a next step and for the final model. Decisional capacity was left out of the analysis due to multicollinearity with dementia.

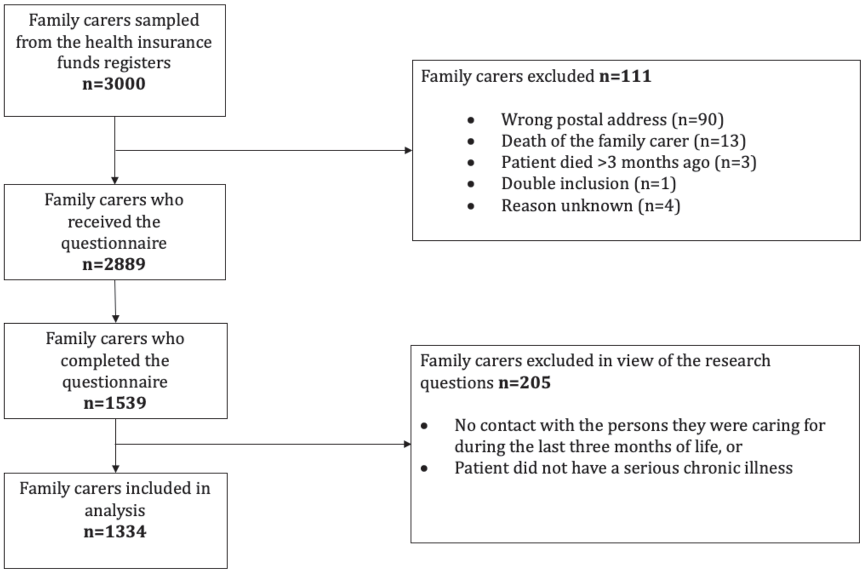
Ethical approval

Ethical approval was obtained from the Ethical Review Board of Brussels University hospital of the Vrije Universiteit Brussel (B.U.N. 143201940562)

Results

Of the sampled 3000 people 2889 received the questionnaire (see figure 1 for the reasons for exclusion). A non-response survey showed that another 89 participants of the sample were non-eligible, resulting in 2800 eligible cases. In total, 1539 participants completed the questionnaire, resulting in a response rate of 55%. For the analysis, a total of 1334 respondents (i.e. family carers of someone with at least one serious illness and had contact with them during the final three months of life) were included.

Figure 1: Flowchart participants



Sample characteristics

Most of the family carers were women (68.2%) and were either the child (51.7%) or partner (34.4%) of the deceased person. Family carers were on average 65.5 years. Approximately 13% of family carers had a medical degree in healthcare (e.g. physician, nurse) (table 1).

Almost 40% of the deceased persons were older than 85 years. The most common chronic illnesses were cancer (31.8%) and dementia (30.7%). In 40.2% of the cases, the deceased person could still make decisions for him/herself in the final 3 months of life and 36.7% of the family carers indicated that specialist palliative care services were involved in the care (Table 1).

Profile of family carers who discussed advance care planning

In 46.9% of the cases, family carers had engaged in advance care planning conversations with their relative three months before bereavement (not shown in table). Advance care planning conversations were more likely in those family carers who were 55 years or younger (54.7%), had a medical degree (e.g. physician, nurse) (57.8%), were the partner of the deceased patient (52.2%) and were the only family carer (52.5%). As for the patient characteristics, advance care planning discussions were more likely when the deceased persons were aged between 66-75 (57.8%); had cancer (44%); had decisional capacity in the final three months (64.8%), lived at home in the final three months (57.4%) and was supported by specialist palliative care services (59.2%) (Table 1).

Table 1: Family carers' and deceased patients' characteristics (total sample and specific for family carers who did have discussed ACP) (n=1334)

	Total number of family carers in the study*	**% of family carers that discussed ACP†**	**p-value‡**
Characteristics of the bereaved family carers	N (%)		
Sex			.911
Male	416 (31.8)	47.2	
Female	892 (68.2)	46.9	
Age in years			**.002**
≤55	238 (18.2)	54.7	
56 – 65	488 (37.3)	42.9	
66 – 75	305 (23.3)	52.0	
76+	278 (21.2)	41.0	
Education level			.361
None	38 (2.9)	30.0	
Primary school	212 (16.2)	45.7	
Secondary school	603 (46.1)	46.6	
Higher education (college)	364 (27.9)	48.4	
University	90 (6.9)	50.6	
Degree in care (nurse or doctor)			**.003**
Yes	167 (13.2)	57.8	
No	1102 (86.8)	45.1	
Work status			.105
Full-time	271 (20.8)	46.4	
Part-time	232 (17.8)	50.2	
Unemployed	30 (2.3)	51.9	
Retired	612 (46.9)	47.8	
Housewife or houseman	103 (7.9)	32.2	
Other	58 (4.4)	45.3	
Relationship towards patient			**<.05**
Partner	457 (34.4)	52.2	
Son(-in-law) or daughter(-in-law)	687 (51.7)	43.7	
Other	185 (13.9)	47.5	
Lived together with deceased person in the last three months of life			**<.001**
Yes	478 (36.7)	56.1	
No	826 (63.3)	42.0	
Other family carers involved in care			**.028**
no other family carers	498 (38.5)	52.5	
others family carers	797 (61.5)	43.5	

Characteristics of the deceased patient			
Age in years			**<.001**
≤65	270 (20.7)	53.9	
66-75	204 (15.7)	57.8	
76-85	318 (24.4)	41.6	
>85	510 (39.2)	41.8	
Serious chronic illness			**<.001**
Cancer	424 (31.8)	44	
Dementia	359 (26.9)	11.9	
Organ failure	329 (24.7)	26.4	
Stroke	60 (4.5)	3.9	
Other	162 (12.1)	13.8	
Decisional capacity of patient throughout last three moths			**<.001**
Yes	529 (40.2)	64.8	
Partly	357 (27.1)	50.6	
No	430 (32.7)	20.8	
Death was surprising			.053
Surprised	402 (30.5)	42.8	
Not surprised	915 (69.5)	48.8	
Did the deceased person live at home in the last 3 months of life			**<.001**
Most of the time	441 (34.3)	57.4	
Sometimes	346 (26.9)	51.2	
Not at all (e.g. nursing home or hospital)	499 (38.8)	35.4	
Palliative Care services involved			**<.001**
Yes	464 (36.7)	59.2	
No	762 (60.3)	40.1	
Don’t know	38 (3)	41.2	

* Percentages are column percentages**;** † Percentages are row percentages

‡ p-value was measured whether the family carer discussed EOL care planning using univariable binary logistic regression analysis

Missing values for sample bereaved family carers: Sex: n=26 (1.9%); Age in years: n=25 (1.9%); Education: n=27 (2.0%); Degree in Care: n=65(4.9%); Work status: n=28 (2.1%); Relationship towards patient: n=5 (0.4%); Living together with deceased relative in the last three months: n=30 (2.2%); Only non-professional: n=39(2.9%); ACP discussions: n=132(9.9%)

Missing values for sample deceased patient: Age in years: n=32 (2.4%); Decisional capacity of patient throughout last three months: n= 18 (1.3%); Death was surprising: n=17(1.3%); Living at home in the last 3 months: n=48 (3.6%); Palliative care services: n=70 (5.2%)

Factors associated with having advance care planning conversations

Multivariable analysis confirmed a higher likelihood of having an advance care planning conversation when family carers were 55 years or younger (OR=1.48, 1.03-2.13), had a medical degree (OR=1.76, 1.20-2.58), and when specialist palliative care services were involved (OR = 2.07

1.55 -2.75). Family carers were less likely to have an advance care planning conversation when the deceased relative was diagnosed with dementia (OR=0.25, .17-.37) (Table 2).

Table 2: Multivariable logistic regression of the association between having ACP discussions and bereaved family carers and deceased patients' characteristics in the last three months (n=1334)

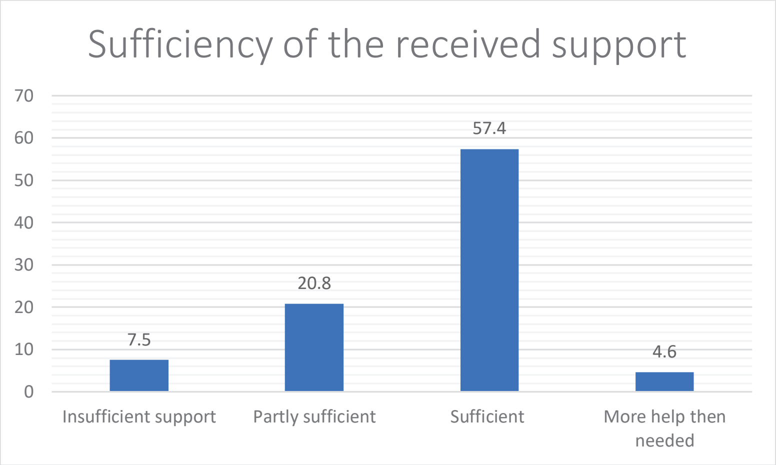
	Did you have had ACP discussions?	
	OR (95% CI)	p-value
Characteristics bereaved family carer		
Age in years		
≤55	1.48 (1.03-2.13)	**.032**
56-65	Ref	
66-75	1.18 (.83-1.67)	.35
>75	.86(.58-1.26)	.43
Medical degree		
Yes	1.76 (1.20-2.58)	**.004**
No	Ref	
Characteristics of the deceased patient		
Serious condition		
Cancer	1.41 (.99-1.20)	.057
Dementia	.25 (.17-.37)	**<.001**
Stroke	.72 (.38 - 1.37)	.32
Other	.96 (.63 - 1.48)	.87
Organ failure	Ref	
Care characteristics		
Patient lived at home during last 3 months of life		
I don't know	1.52 (1.10 - 2.10)	**.011**
Yes	1.14 (.81 - 1.61)	.45
No	Ref	
Family carer was the only informal carer		
Yes	1.36 (1.03 - 1.79)	**.028**
No, there were more	Ref	
Palliative care services involved		
I don't know	1.41 (.66 - 3.04)	.38
Yes	2.07 (1.55 -2.75)	**<.001**
No	Ref	

Multivariable binary logistic regression analysis was used with ACP discussion vs no ACP discussion as dependent variable. We used stepwise modelling to test three models (model 1: bereaved family carer characteristics; model 2: deceased patient characteristics; and model 3: care characteristics). Each candidate predictor per model was either retained or deleted depending on its significance (p <.05), resulting in this final model for the outcome variable.

Support of healthcare professionals and type of support received to conduct advance care planning

Of those family carers who had an advance care planning conversation with their relative 78.1% received support for this from a healthcare professional. Of those that did not receive support (21.9%), 78.3% indicated that they did not need the support of healthcare professionals while 21.7% indicated that they did. Most often, support received was from the GP (66.6%), palliative care nurse or doctor (36.6%), home care nurse (33.2%) or nurse or care worker in hospital or residential care center (32%) (Table 3). The type of support received from a healthcare professional was in 53.8% doing the advance care planning conversation together with the family carer and the deceased person. It was less often by explaining the family carers in how to do advance care planning conversations (17.9%) and rarely by referring them to other useful sources on how to do advance care planning conversations (9.4%) (not in table). The support received was deemed as sufficient by 57.4% of the family carers (see figure 2).

Figure 2: Sufficiency of the received support of the healthcare professionals (n=413)



Percentages are row percentages. Percentages don't add up to 100% as multiple answers were possible. Missing values: Satisfaction with support received: n=40 (9.7%).

Table 3: Healthcare professionals who provided support to the family carers who had ACP-discussions

	Having ACP discussions (n=564)
	%
No support received (n=116)	**21.9**
Needed it	21.7
Did not need it	78.3
Support received (n=448)	**78.1**
Support received of different healthcare professionals	
Home care nurse	33.2
Nurse or care worker in hospital or residential care center	32.0
GP	66.6
Specialist	16.5
Palliative nurse or doctor	36.6
Someone close with experience in healthcare	10.7
Other	0.3

Percentages are column percentages.

Missing values: Talking about ACP: n=35 (6.2%)

Discussion

Main findings

Our findings show that about half of the bereaved family carers indicated to have engaged in advance care planning conversations with their relative in the final three months of life. These conversations were more likely to have taken place when family carers were 55 years of age or younger, had a medical degree or when specialist palliative care services were involved, but less likely when the deceased person was diagnosed with dementia. Seventy-eight percent of the family carers received support from a healthcare professional, usually the GP. The type of support received was most often the healthcare professional doing the conversation together with the family carer and seriously ill person.

Strengths and limitations

This study has several strengths. First, the sampling framework of this study was unique because we used complete registers of health insurances and took a random sample from this. The study therefore suffers less from selection bias compared to most studies that select family carers, for instance, via professional palliative care or other healthcare service use; and statistical generalization to the target population of family carers is improved. Second, the questionnaire was available both on paper and online which seemed beneficial for the response rate (55%), as it was higher compared to other studies with similar populations[25–27]. This study has also some limitations. First, despite the effort of construing a complete sample frame there might still be some selection bias because we only could include registered family carers. As a result, we have no insights into the experience of family carers who were not registered by one of the health insurances. Second, the term “advance care planning” is not always well-known in the general population and therefore, in this survey, we described advance care planning as conversations about end-of-life care and end-of-life decisions or completion of advanced directives or living wills. Recent recommendations highlight the need to approach advance care planning on a broader level, whereas the focus is more on the persons’ values, wishes and preferences, and the need of advance care planning occurring over time with multiple conversations[28,29]. To reduce recall bias, which cannot be entirely excluded, we used a very specific description of advance care planning and the time between advance care planning and the survey completion was a maximum of nine months, which also minimizes the impact of recall bias.

Interpretation of the main findings

Almost 50% of the family carers did have advance care planning conversations with their relative. This result is similar compared to another cross-sectional study about family relationships in advance care planning[30]. We found that family carers who were 55 years of age or younger were more likely to engage in advance care planning conversations with their relative. This finding is in line with other studies showing that in general younger patients and family carers desire more detailed information regarding the illness, and want more involvement and autonomy in decision-making[31,32]. As attitudes towards discussing end-of-life issues might differ in younger family carers compared to older generations, younger family carers might be more open and willing to initiate advance care planning conversations with their relative themselves. To increase normalization of advance care planning, public health campaigns are developed[33] and more self-guided tools to facilitate advance care planning conversations (e.g. websites, conversation cards) are becoming available[34]. Older adults might find it difficult to find their way to these tools, websites or information campaigns of advance care planning, so healthcare professionals have a key role in introducing these tools to older adults[35]. As a lot of these tools has been developed in the recent years, it is highly recommended that these are tested upon the acceptability, comprehensibility, and utility in future studies[36]. Also, a recent review about advance care planning in the community revealed that the COVID-19 pandemic has worsened some known barriers to advance care planning (e.g. issues of trust given fears of rationing of healthcare resources), while others have improved (e.g. lack of consumer knowledge about advance care planning and perceiving advance care planning as irrelevant due to the increased media discussion of advance care planning)[36]. Future research about the impact of the pandemic on the experiences of bereaved family carers now is warranted.

This study focuses on the last 3 months of life, but it is highly recommended to initiate advance care planning in the earlier stages of the disease trajectory[37], and to focus on the process of advance care planning, rather than on its product[38]. Our results showed that when the deceased person was diagnosed with dementia, the likelihood of family carers performing advance care planning conversations with their relative in the final three months decreased, possibly because the person with dementia did not have the mental capacity to participate anymore. Previous studies have shown that only a minority of people with dementia get the opportunity to engage in advance care planning themselves and more often the healthcare professional only involves the family carer[39]. It has been recommended to maximize the opportunity for people with dementia to participate in advance care planning as active agents, in order to highlight their capabilities and to have their voice heard[38]. Furthermore, advance care planning should be reviewed regularly and especially in case of change in health condition, and particularly in people with dementia. In general, it is advocated to consider advance care planning as a holistic, flexible and relational process that is initiated in a timely manner[38].

Our findings seem to indicate that the principles of advance care planning have yet to be adopted more outside the specialized palliative care context. The involvement of specialist palliative care services was strongly associated with engaging in advance care planning in the last 3 months of life. This is not surprising as one of the core tasks of these services is to inform patients and family carers about the severity of the illness and to prepare them for the approaching death[40]. Family carers with a medical degree were also significantly more likely to engage in an advance care planning conversation with their relative in the final three months of life. They might be more aware of the importance of engaging in advance care planning conversations, are more skilled to perform these conversations themselves and find their way more easily to specific professional support and services[41]. In 78%, family carers were supported by a healthcare professional (mostly by the GP) and most of the support by healthcare professionals was given by performing the advance care planning conversations together with the family carers and their relative. This can be considered as an empowering collaborative approach (versus providing information or explaining how to conduct these conversations which were less frequently reported in this study). However, previous studies have shown that family carers and seriously ill persons also are willing to have these conversations alone, without the involvement of a professional[7–9]. Perhaps family carers felt safer to perform these conversations together with a healthcare professional or they might have lacked the necessary tools or skills to perform these conversations themselves. Given that only 57.4% of the family carers perceived the support received as sufficient, more insight is needed into the specific support needs of family carers for conducting these advance care planning conversations with their relative during the disease trajectory, or on how family carers wish to be supported by healthcare professionals in engaging in advance care planning conversations, as openness for communication about end-of-life care and end-of-life decision making might grow during the disease trajectory[15,35]. A possibility for increasing empowering and self-efficacy in family carer is via educational progammes (e.g. FOCUS program)[42,43].

Conclusion

This study shows that about half of the family carers had engaged in advance care planning conversations with their relative in the final three months of life. Most of the family carers who had done so, received support from a healthcare professional, mostly from a GP and mostly by performing the advance care planning conversation together with the family carer and the seriously ill person. Further insight is needed into how family carers can be supported to conduct these advance care planning conversations, both with and without involvement of healthcare professionals.

Additional information

Authorship

J.C., A.D.V., L.D., P.H. and C.V. designed the study. I.V., A.D.V., J.C., C.V., L.D. and P.H. contributed to the interpretation of the results. I.V. wrote the manuscript and all authors critically revised the manuscript and approved the final version.

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Conflicts of interest

The authors declare that there is no conflict of interest.

Data sharing

The datasets are not publicly available to safeguard the privacy of the participants but are available from the corresponding author on reasonable request.

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Appendix: QUESTIONNAIRE

<u>QUESTION ABOUT THE CARE YOU PROVIDED</u>

In this survey we ask you questions about the care for your partner/family member/friend in the period before his/her death. We realize this may bring back painful memories for you, for which we apologise. However, our survey is intended to improve care and your input is very valuable.

Please fill in the questionnaire as completely as possible, although you are of course <u>under no obligation</u> to do so. For each question, please indicate or fill in which answer applies to your situation. If you prefer not to answer a question, you are free to skip it. You may have cared for several loved ones who have died in recent months. In that case, we ask you to keep the care for one close person in mind.

<u>PART 1</u> : The situation in which you took care of your partner/family member/friend

1. What was your relationship with your deceased partner/family member/friend? **I was his/her:**	☐ Partner ☐ Daughter-in-law or son-in-law ☐ Brother or sister ☐ Parent ☐ Other family member: ………………………………. ☐ no family
2. Were you in contact with your partner/family member/friend <u>during the last 3 months of his/her life</u> ?	☐ No→ **<u>GO TO QUESTION 26</u>** ☐ Yes
3. Please indicate whether your partner/family member/friend had one or more of the following chronic illness in the <u>last 3 months before death.</u> *(multiple answers possible)*	☐ Cancer ☐ Heart disease (e.g. chronic heart failure) ☐ Stroke (or its consequences) ☐ Lung disease (not cancer, but e.g. COPD or asthma) ☐ Diabetes ☐ Kidney or liver disease (not cancer) ☐ Dementia (e.g. Alzheimer's disease) ☐ Other nervous system disease (e.g. ALS) ☐ Others: ………………………………………………….. ☐ I don't know ☐ He/she had no chronic condition→ **<u>GO TO QUESTION 26</u>**
4. Was your partner/family member /friend able to make decisions about his/her care during the <u>last 3 months before death?</u>	☐ No ☐ Sometimes or partly ☐ Yes

5. To what extent was the death of your partner/family member/friend surprising to you?	☐ Definitely not surprising ☐ Rather not surprising ☐ Rather surprising ☐ Definitely surprising
6. How long did your partner/family member/friend stay in a home environment during his/her last 3 months of life (by this we mean at home or living with a relative or acquaintance, and not in a hospital or in a nursing home)?	☐ The entire 3 months ☐ Most of the time ☐ About half the time ☐ Only occasionally ☐ Not
7. In the last 3 months before his/her death did you and your partner/ family member/friend live primarily under the same roof?	☐ No ☐ Yes
8. Were you the only non-professional care provider for your partner/family member/friend?	☐ No, there were 1 or more others ☐ Yes, I was the only one
9. Did your partner/family member/friend receive care from a specialized palliative care service?	☐ No→ **GO TO QUESTION 11** ☐ Yes ☐ I don't know
10. Which of these initiatives did you appeal to during the last 3 months of life of your partner/family member/friend ? *(multiple answers possible)*	☐ Palliative home care team ☐ Palliative unit (separate ward) in hospital ☐ Palliative support team in hospital (which provides care in different departments) ☐ Reference person – nurse in the palliative care in residential care center ☐ Palliative day center ☐ I don't know
11. What was your working status at the time that you started to provide care to your partner/family member/friend?	☐ Working full time ☐ Working part-time ☐ Unemployed ☐ Retired ☐ Househusband or housewife ☐ Other: ……………………………………………… …….
12. Have you changed your employment	☐ No, my work status has not changed

during the illness of your partner/family member/friend? *(multiple answers possible)*	☐ Yes, I started working more ☐ Yes, the weekly working time has been reduced ☐ Yes, I changed jobs ☐ Yes, I have completely stopped working ☐ Yes, I have stopped looking for work ☐ Yes, I have retired ☐ Other: ……………………………………………… …….

PART 2: Your activities in caring for your partner/family member/friend

13. Which of the following activities have <u>you undertaken at least once</u> during <u>the last 3 months of your partner/family member/friend's life</u> ?

	YES	**NO**
Preparing food for my partner/family member/friend	☐	☐
Giving food or drink to my partner/family member/friend	☐	☐
Going to consultations with healthcare professionals with my partner/family member/friend	☐	☐
Grocery shopping for my partner/family member/friend	☐	☐
Cleaning for my partner/family member/friend	☐	☐
Washing, ironing or repairing clothes for my partner/family member/friend	☐	☐
Chores in the house or maintenance of the garden for my partner/family member/friend	☐	☐
Follow-up of care provision (e.g. making and planning of appointments with various care providers)	☐	☐
Other: ……………………………………………………………………… …….	☐	☐

PART 3 : Your cooperation with healthcare professional in activities

Below are some questions about different activities. You may have taken these activities upon yourself in the care of your partner/family member/friend, and collaborated with healthcare professionals to do so.

14. GIVING PERSONAL CARE TO MY PARTNER/FAMILY MEMBER/FRIEND (e.g. helping to wash, dress and undress)

a) Did you undertake this activity at least once during the last 3 months before the death of your partner/ Family member/friend?	☐ No→ **GO TO QUESTION 15** ☐ Yes, although I would rather someone else perform this activity ☐ Yes, and I wanted to take this activity upon myself
b) Which healthcare professionals ever supported you in this activity so you could do it by yourself? *(multiple answers possible)*	☐ Home nursing ☐ Nurse or healthcare professional in hospital or nursing home ☐ Family doctor ☐ Specialist (specialist doctor) ☐ Palliative nurse or doctor ☐ Someone from my immediate environment with professional experience in healthcare ☐ Other: …………………………………………. ……. . ☐ Received no help, although I needed it →on **GO TO QUESTION 15** ☐ Didn't receive help but I didn't need it → **GO TO QUESTION 15**
c) In which of the following ways were you ever supported in this activity by healthcare professionals? *(multiple answers possible)*	☐ They explained to me how I could do this ☐ They showed me how to do this ☐ They let me practice this myself under supervision ☐ Other: ……………………………………………. ……. ☐ None of the above
d) To what extent did the above professional support allow you to do this activity by yourself?	☐ I didn't get enough support to do this activity by myself ☐ I partly got enough support to do this activity by myself ☐ I got enough support to do this activity by myself ☐ I got more support than I needed to do this activity by myself

15. FOLLOWING AND ADDRESSING SYMPTOMS IN MY PARTNER/FAMILY MEMBER/FRIEND (to reduce these symptoms or their impact on my partner/family member/friend)

a) Did you undertake this activity at least once during the last 3 months before the death of your partner/ family member/friend?	☐ No→ **GO TO QUESTION 16** ☐ Yes, although I would rather someone else perform this activity ☐ Yes, and I wanted to take this activity on myself
b) Which healthcare professionals ever supported you in this activity so you could do it by yourself? *(multiple answers possible)*	☐ home nursing ☐ Nurse or healthcare professional in hospital or nursing home ☐ Family doctor ☐ Specialist (specialist doctor) ☐ Palliative nurse or doctor ☐ Someone from my immediate environment with professional experience in healthcare Other: …………………………………………… ……. . ☐ Received no help, although I needed it →on **GO TO QUESTION 16** ☐ Didn't receive help but I didn't need it → **GO TO QUESTION 16**
c) In which of the following ways were you ever supported in this activity by healthcare professionals? *(multiple answers possible)*	☐ They explained to me which symptoms I could expect ☐ They explained to me how to deal with certain symptoms ☐ I was shown how to deal with certain symptoms ☐ They let me practice how to tackle certain symptoms, under supervision. ☐ Other: ……………………………………………. ……. ☐ none of the above
d) To what extent did the above professional support allow you to do this activity by yourself?	☐ I didn't get enough support to do this activity by myself ☐ I partly got enough support to do this activity by myself ☐ I got enough support to do this activity by myself ☐ I got more support than I needed to do this activity by myself

16. HELPING MY PARTNER/FAMILY MEMBER/FRIEND IN USING (VARIOUS TYPES OF) MEDICATION

a) Did you undertake this activity at least once during the last 3 months before the death of your partner/ family member/friend?	☐ No→ **GO TO QUESTION 17** ☐ Yes, although I would rather someone else perform this activity ☐ Yes, and I wanted to take this activity on myself
b) Which healthcare professionals ever supported you in this activity so you could do it by yourself? *(multiple answers possible)*	☐ home nursing ☐ Nurse or healthcare professional in hospital or nursing home ☐ Family doctor ☐ Specialist (specialist doctor) ☐ Palliative nurse or doctor ☐ Pharmacist ☐ Someone from my immediate environment with professional experience in healthcare ☐ Other: ……………………………………… ……. . ☐ Received no help, although I needed it →on **GO TO QUESTION 17** ☐ Didn't receive help but I didn't need it → **GO TO QUESTION 17**
c) In which of the following ways were you ever supported in this activity by healthcare professionals? *(multiple answers possible)*	☐ They explained to me how to use it (eg. when to administer which medication, in what dose, information on combination with other medication) ☐ I was shown the method of use (way of administering) ☐ I was allowed to administer medication in a supervised exercise ☐ Other: ……………………………………… ……. ☐ none of the above
d) To what extent did the above professional support allow you to do this activity by yourself?	☐ I didn't get enough support to do this activity by myself ☐ I partly got enough support to do this activity by myself ☐ I got enough support to do this activity by myself ☐ I got more support than I needed to do this activity by myself

17. HELPING WITH SAFE MOVEMENTS OF MY PARTNER/FAMILY MEMBER/FRIEND, INDOOR OR OUTDOOR (e.g. when getting up from a lying or sitting position, walking, sitting down, taking a seat in a car, using a wheelchair and/or walker, …)

a) Did you undertake this activity at least once during the last 3 months before the death of your partner/family member/friend?	☐ No→ **GO TO QUESTION 18** ☐ Yes, although I would rather someone else perform this activity ☐ Yes, and I wanted to take this activity on myself
b) Which healthcare professionals ever supported you in this activity so you could do it by yourself? *(multiple answers possible)*	☐ home nursing ☐ Nurse or healthcare professional in hospital or nursing home ☐ Family doctor ☐ Specialist (specialist doctor) ☐ Palliative nurse or doctor ☐ physiotherapist ☐ Someone from my immediate environment with professional experience in healthcare ☐ Other: …………………………………………… ……. . ☐ Received no help, although I needed it →on **GO TO QUESTION 18** ☐ Didn't receive help but I didn't need it → **GO TO QUESTION 18**
c) In which of the following ways were you ever supported in this activity by healthcare professionals? *(multiple answers possible)*	☐ They explained to me how I could do this by myself ☐ They showed me how to do this by myself ☐ They let me practice under supervision ☐ Other: ……………………………………………… ……. ……………………………… ☐ none of the above
d) To what extent did the above professional support allow you to do this activity by yourself?	☐ I didn't get enough support to do this activity by myself ☐ I partly got enough support to do this activity by myself ☐ I got enough support to do this activity by myself ☐ I got more support than I needed to do this activity by myself

18. HELPING TO PROVIDE MY PARTNER/FAMILY MEMBER/FRIEND WITH PHYSICAL COMFORT (e.g. being able to sit or lie comfortably, skin or facial care, oral care)

a) Did you undertake this activity at least once during the last 3 months before the death of your partner/ family member/friend?	☐ No→ **GO TO QUESTION 19** ☐ Yes, although I would rather someone else perform this activity ☐ Yes, and I wanted to take this activity on myself
b) Which healthcare professionals ever supported you in this activity so you could do it by yourself? *(multiple answers possible)*	☐ home nursing ☐ Nurse or healthcare professional in hospital or nursing home ☐ Family doctor ☐ Specialist (specialist doctor) ☐ Palliative nurse or doctor ☐ physiotherapist ☐ Someone from my immediate environment with professional experience in healthcare ☐ Other: …………………………………………… ……. . ☐ Received no help, although I needed it →on **GO TO QUESTION 19** ☐ Didn't receive help but I didn't need it → **GO TO QUESTION 19**
c) In which of the following ways were you ever supported in this activity by healthcare professionals? *(multiple answers possible)*	☐ They explained to me how I could do this by myself ☐ They showed me how to do this by myself ☐ They let me practice under supervision ☐ Other: ……………………………………………… ……. ☐ none of the above
d) To what extent did the above professional support allow you to do this activity by yourself?	☐ I didn't get enough support to do this activity by myself ☐ I partly got enough support to do this activity by myself ☐ I got enough support to do this activity by myself ☐ I got more support than I needed to do this activity by myself

19. DOING ADMINISTRATION IN CONNECTION WITH CARE FOR MY PARTNER/FAMILY MEMBER/FRIEND (e.g. applying for professional assistance or financial compensation)

a) Did you undertake this activity at least once during the last 3 months before the death of your partner/family member/friend?	☐ No→ **GO TO QUESTION 20** ☐ Yes, although I would rather someone else perform this activity ☐ Yes, and I wanted to take this activity on myself
b) Which healthcare professionals ever supported you in this activity so you could do it by yourself? *(multiple answers possible)*	☐ home nursing ☐ Family doctor ☐ Specialist (specialist doctor) ☐ Palliative nurse or doctor ☐ Social assistant or social worker ☐ Someone from my immediate environment with professional experience in healthcare ☐ Other: ………………………………………… ……. . ☐ Received no help, although I needed it → **GO TO QUESTION 20** ☐ Didn't receive help but I didn't need it → **GO TO QUESTION 20**
c) In which of the following ways were you ever supported in this activity by healthcare professionals? *(multiple answers possible)*	☐ They explained to me how I could manage the administration myself ☐ They explained to me where or whom I could turn to for this ☐ They showed me how to manage the administration so I could do it myself ☐ They showed me where or whom I could go to ☐ They worked together with me to arrange the administration ☐ Other: ……………………………………………. ……. ☐ none of the above
d) To what extent did the above professional support allow you to do this activity by yourself?	☐ I didn't get enough support to do this activity by myself ☐ I partly got enough support to do this activity by myself ☐ I got enough support to do this activity by myself ☐ I got more support than I needed to do this activity by myself

20. ADAPTING THE HOUSE FOR SAFETY AND COMFORT FOR MY PARTNER/FAMILY MEMBER/FRIEND (e.g. in the living room or bathroom)

a) Did you undertake this activity at least once during the last 3 months before the death of your partner/family member/friend?	☐ No→ **GO TO QUESTION 21** ☐ Yes, although I would rather someone else perform this activity ☐ Yes, and I wanted to take this activity on myself
b) Which healthcare professionals ever supported you in this activity so you could do it by yourself? *(multiple answers possible)*	☐ home nursing ☐ Family doctor ☐ Specialist (specialist doctor) ☐ Palliative nurse or doctor ☐ Social assistant or social worker ☐ Someone from my immediate environment with professional experience in healthcare ☐ Other: ………………………………………… ……. . ☐ Received no help, although I needed it → **GO TO QUESTION 21** ☐ Didn't receive help but I didn't need it → **GO TO QUESTION 21**
c) In which of the following ways were you ever supported in this activity by healthcare professionals? *(multiple answers possible)*	☐ They explained what I could adjust at home ☐ They explained the benefits of the proposed changes in the house ☐ They explained how I could do these adjustments by myself ☐ I was referred to available services that performed these adjustments ☐ Other: …………………………………………… ……. ☐ none of the above
d) To what extent did the above professional support allow you to do this activity by yourself?	☐ I didn't get enough support to do this activity by myself ☐ I partly got enough support to do this activity by myself ☐ I got enough support to do this activity by myself ☐ I got more support than I needed to do this activity by myself

21. TALKING TO MY PARTNER/FAMILY MEMBER/FRIEND ABOUT HIS/HER EMOTIONS (e.g. hopelessness, gloom, fear of death)

a) Did you undertake this activity at least once during the <u>last 3 months before the death</u> of your partner/family member/friend?	☐ No→ **<u>GO TO QUESTION 22</u>** ☐ Yes, although I would rather someone else perform this activity ☐ Yes, and I wanted to take this activity on myself
b) Which healthcare professionals <u>ever</u> supported you in this activity so you could do it by yourself? *(multiple answers possible)*	☐ home nursing ☐ Nurse or healthcare professional in hospital or nursing home ☐ Family doctor ☐ Specialist (specialist doctor) ☐ Palliative nurse or doctor ☐ Social assistant or social worker ☐ Psychologist ☐ Someone from my immediate environment with professional experience in healthcare ☐ Other: ………………………………………… ……. . ☐ Received no help, although I needed it → **<u>GO TO QUESTION 22</u>** ☐ Didn't receive help but I didn't need it → **<u>GO TO QUESTION 22</u>**
c) In which of the following ways were you <u>ever</u> supported in this activity by healthcare professionals? *(multiple answers possible)*	☐ I was referred to other resources (e.g. books, brochures, websites) where tips are given ☐ They explained to me how to tackle this ☐ They had these conversations with me and my partner/family member/friend ☐ Other: …………………………………………… ……. ☐ none of the above
d) To what extent did the above professional support allow you to do this activity by yourself?	☐ I didn't get enough support to do this activity by myself ☐ I partly got enough support to do this activity by myself ☐ I got enough support to do this activity by myself ☐ I got more support than I needed to do this activity by myself

22. TALKING TO MY PARTNER/FAMILY MEMBER/FRIEND ABOUT END OF LIFE PLANNING (e.g. the care that my partner/family member/friend did or did not want at the end of life, advance directives, will, wishes regarding euthanasia or other end-of-life decisions such as palliative sedation)

a) Did you undertake this activity at least once during the <u>last 3 months before the death</u> of your partner/family member/friend?	☐ No→ **<u>GO TO QUESTION 23</u>** ☐ Yes, although I would rather someone else perform this activity ☐ Yes, and I wanted to take this activity on myself
b) Which healthcare professionals <u>ever</u> supported you in this activity so you could do it by yourself? *(multiple answers possible)*	☐ home nursing ☐ Nurse or healthcare professional in hospital or nursing home ☐ Family doctor ☐ Specialist (specialist doctor) ☐ Palliative nurse or doctor ☐ Someone from my immediate environment with professional experience in healthcare ☐ Other: …………………………………………… ……. . ☐ Received no help, although I needed it → **<u>GO TO QUESTION 23</u>** ☐ Didn't receive help but I didn't need it → **<u>GO TO QUESTION 23</u>**
c) In which of the following ways were you <u>ever</u> supported in this activity by healthcare professionals? *(multiple answers possible)*	☐ I was referred to other resources (e.g. books, brochures, websites) where tips are given ☐ They explained to me how to tackle this ☐ They had these conversations with me and my partner/family member/friend ☐ Other: ……………………………………………… ……. ☐ none of the above
d) To what extent did the above professional support allow you to do this activity by yourself?	☐ I didn't get enough support to do this activity by myself ☐ I partly got enough support to do this activity by myself ☐ I got enough support to do this activity by myself ☐ I got more support than I needed to do this activity by myself

23. ENSURING THAT MY PARTNER/FAMILY MEMBER/FRIEND MAINTAINED A SOCIAL LIFE (your partner's/family member's/friend's social contact with others)

a) Did you undertake this activity at least once during the <u>last 3 months before the death</u> of your partner/family member/friend?	☐ No→ **<u>GO TO QUESTION 24</u>** ☐ Yes, although I would rather someone else perform this activity ☐ Yes, and I wanted to take this activity on myself
b) Which healthcare professionals <u>ever</u> supported you in this activity so you could do it by yourself? *(multiple answers possible)*	☐ home nursing ☐ Nurse or healthcare professional in hospital or nursing home ☐ Family doctor ☐ Specialist (specialist doctor) ☐ Palliative nurse or doctor ☐ Social assistant or social worker ☐ Someone from my immediate environment with professional experience in healthcare ☐ Other: …………………………………………… ……. . ☐ Received no help, although I needed it → **<u>GO TO QUESTION 24</u>** ☐ Didn't receive help but I didn't need it → **<u>GO TO QUESTION 24</u>**
c) **In which of the following ways were you <u>ever</u> supported in this activity by healthcare professionals?** *(multiple answers possible)*	☐ I was encouraged to go to peer groups with my partner/family member/friend ☐ I was encouraged to visit our relatives or friends ☐ I was reminded that it is important to have a social life ☐ Social activities were organized which my partner/family member/friend and I could participate in ☐ Other: ……………………………………………. ……. ☐ none of the above
d) To what extent did the above professional support allow you to do this activity by yourself?	☐ I didn't get enough support to do this activity by myself ☐ I partly got enough support to do this activity by myself ☐ I got enough support to do this activity by myself ☐ I got more support than I needed to do this activity by myself

PART 4 : Your general experience of collaboration with professionals

24. The statements below are about how healthcare professionals generally worked with you and supported you in the 3 months prior to the death of your partner/family member/friend. We would like to ask you to what extent you find these statements applicable to your contact with the majority of the healthcare professionals who have assisted you. *Please indicate to what extent you agree with each statement.*

	Totally disagree	Disagree	Neutral	Agree	Totally agree	Not applicable
a) I didn't get enough freedom to determine how the care for my loved one was organized and carried out	☐	☐	☐	☐	☐	☐
b) I believe that I and the healthcare professionals were well attuned in the care of my partner/family member/friend	☐	☐	☐	☐	☐	☐
c) The healthcare professionals were always available when I needed them	☐	☐	☐	☐	☐	☐
d) The healthcare professionals did insufficiently let me know that I could ask them questions when I had them	☐	☐	☐	☐	☐	☐
e) I don't have enough information about the available healthcare professionals and services where I could go to ask for help	☐	☐	☐	☐	☐	☐
f) I have received enough information about my partner/family member/friend's condition	☐	☐	☐	☐	☐	☐
g) Information was generally given to me by healthcare professionals in an appropriate manner (e.g. clearly, on time, my feelings were taken into account)	☐	☐	☐	☐	☐	☐
h) Overall, I believed that the healthcare professionals communicated well	☐	☐	☐	☐	☐	☐
i) The healthcare professionals did not have enough expertise to support me in an appropriate way when the end of my partner/family member/friend's life approached	☐	☐	☐	☐	☐	☐

PART 5 : How have you been feeling this past week?

25. For each of the following feelings, please indicate to what extent you have felt this way in your daily life in the past week :

	Hardly or not at all (1)	A little (2)	Average (3)	Quite (4)	Strongly (5)
Interested	☐	☐	☐	☐	☐
Sad	☐	☐	☐	☐	☐
Excited	☐	☐	☐	☐	☐
Depressed	☐	☐	☐	☐	☐
Strong	☐	☐	☐	☐	☐
Guilty	☐	☐	☐	☐	☐
Anxious	☐	☐	☐	☐	☐
Hostile	☐	☐	☐	☐	☐
Enthusiastic	☐	☐	☐	☐	☐
Confident	☐	☐	☐	☐	☐
Quickly irritated	☐	☐	☐	☐	☐
Alert	☐	☐	☐	☐	☐
Ashamed	☐	☐	☐	☐	☐
Full of inspiration	☐	☐	☐	☐	☐
Tense	☐	☐	☐	☐	☐
Determined	☐	☐	☐	☐	☐
Attentive	☐	☐	☐	☐	☐
Nervous	☐	☐	☐	☐	☐
Energetic	☐	☐	☐	☐	☐
Scared	☐	☐	☐	☐	☐

PART 6 : Some extra information about yourself

26. What is your sex?	☐ Male ☐ Female ☐ X
27. What is your age?	.... years

28. What was the age of your partner/family member/friend at the time of death?	………………………… years
29. In which region or province do you live?	☐ Antwerp ☐ Limburg ☐ East Flanders ☐ Flemish Brabant ☐ West Flanders
30. What is your highest diploma obtained?	☐ Primary education ☐ Secondary or secondary education ☐ College (formerly called A1) ☐ University ☐ None of these diplomas obtained
31. Did you ever graduate as a doctor, nurse or health care professional?	☐ No ☐ Yes

If you would like to add something additional to this questionnaire, you can do so below:

……………………………………………………………………… …………

……………………………………………………………………… …………

……………………………………………………………………… …………

……………………………………………………………………… …………

……………………………………………………………………… …………

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……………………………………………………………………… …………

Chapter 4:

Advance care planning—family carer psychological distress and involvement in decision making: the ACTION trial

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Abstract

Context. Facilitated advance care planning (ACP) helps family carers' to be aware of patient preferences. It can improve family carers' involvement in decision-making and their overall experiences at the end-of-life, as well as, reduce psychological stress.

Objectives. To investigate the effects of the ACTION Respecting Choices (RC) ACP intervention on the family carers' involvement in decision-making in the last three months of the patients' life and on the family carers' psychological distress after three months of bereavement.

Methods. Over six European countries a sample of 162 bereaved family carers returned a bereavement questionnaire. Involvement in decision-making was measured with a single item of the VOICES-SF questionnaire. Psychological distress was measured with the impact of event scale (IES).

Results. No significant effect was found on family carers involvement in decision-making in the last 3 months of the patients' life (95%-CI=0.449–4.097). However, the probability of involvement in decision-making was slightly higher in the intervention arm of the study (89.6% vs 86.7%; OR=1.357). Overall, no statistical difference was found between intervention and control group regarding the IES (M = 34.1 (1.7) vs. 31.8 (1.5); (95%CI = -2.2–6.8)).

Conclusion. The ACTION RC ACP intervention showed no significant effect on family carers' involvement in decision-making or on subsequent psychological distress. More research is needed about 1) how family carers can be actively involved in ACP-conversations, and 2) how to prepare family carers on their role in decision-making.

Introduction

Cancer is a common life-limiting disease, with about 4 million new diagnoses and 2 million deaths annually in the European Union[1]. Timely and efficient communication is necessary between clinicians, patients and their family carers, about future care, including care at the end-of-life in patients living with cancer [2]. One approach to facilitate this is advance care planning (ACP) which 'enables individuals to define goals and preferences for future medical treatment and care, to discuss these goals and preferences with family and health care professionals, and to record and review these preferences if appropriate'[3]. In addition to promoting communication, ACP facilitates shared decision-making and, above all, aligns end-of-life care with patients' preferences[4]. This includes the assignment of a surrogate decision-maker (SDM), if the patient loses decisional capacity[5]. Family carers have a prominent role in the process of decision-making at the end of life, but making these decisions without knowledge about the patients' preferences can be extremely stressfull[6]. Studies in Australia and the US have found that facilitated ACP improved the family carer's well-being, satisfaction with hospital care[7] and confidence in making decisions[8].

The ACTION-study was the first and largest European phase III multicenter cluster randomized controlled trial, to test the efficacy of an ACP intervention in patients with advanced lung or colorectal cancer. A systematic review showed that the Respecting Choices (RC) program was one of the most promising ACP-programs with the best evidence for beneficial effects of ACP[4]. The RC was developed in the US and successfully trialled in a geriatric setting in Australia[7,9]. In the ACTION trial, the ACP intervention (ACTION Respecting Choices (RC) ACP intervention), was an adapted and integrated version of RC program, and consisted of a trained facilitator, using scripted conversation guides, to assist the person with cancer and their family carer to discuss goals, values, beliefs and preferences regarding their future treatment and care[9]. However, the ACTION trial found no significant effects on patients' quality of life, symptoms, satisfaction with care, coping or shared decision-making[10]. In this paper we report on the secondary outcomes of the ACTION-study regarding the effect of the ACTION RC ACP intervention on family carer's involvement in decision-making in the last three months of patients' life, and psychological distress after three months of bereavement.

Methods

Study design

We analysed data of the bereavement questionnaire from the ACTION-study which was a multicentre cluster-randomised controlled trial in six European countries (Belgium, Denmark, Italy, the Netherlands, Slovenia and the United Kingdom). Detailed information about the intervention and the trial procedures can be found in the protocol paper[9] or in appendix 1.

Setting and participants

In total, 1117 persons with advanced lung or colorectal cancer participated in the ACTION study. The specific inclusion criteria for patients are described in the protocol paper[9]. The inclusion criteria for bereaved family carers' were: being older than 18 years, being able to complete the bereavement questionnaire in the language of each country and not taking part in another study that is evaluating palliative care services or communication interventions. A bereavement questionnaire was sent with a cover letter, information sheet and prepaid envelope. A reminder was sent when the questionnaire was not returned after two weeks. Return and completion of the questionnaire were considered to indicate that family carers had consented to participate in the study.

Outcome measures

The main purpose of the bereavement questionnaire was to obtain more information about the last months and weeks of the patients' life as perceived by a bereaved family carer who lost the patient during the one year of inclusion in the study. Involvement in decision-making was measured with a single item of the Views of Informal Carers – Evaluation of Services Short Form (VOICES-SF) questionnaire, which is a 58-item validated questionnaire about health and social services completed by bereaved family carers. The item used was: "Looking back over the last three months of his/her life, were you involved in decisions about his/her care as much as you would have wanted?". Possible responses were: 1) I was involved as much as I wanted to be, 2) I would have liked to be more involved, 3) I would have liked to be less involved, 4) Don't know[11].
Psychological distress was assessed using the impact of event scale (IES) and asked how frequently each item was experienced during the past week after 3 months of bereavement. The IES measures psychological responses to stress on two subscales: (1) intrusion and (2) avoidance and is mostly used to measure post-traumatic stress syndrome. Each item could be rated with 1) not at all, 2) rarely, 3) sometimes and 4) often[12]. During the ACTION-study no baseline measurement or other follow-up questionnaire was sent to the family carer.

Statistical analysis

Multilevel analyses were used to determine the effect of ACP on involvement in decision-making (via multilevel binary logistic regression) and the effect of ACP on psychological distress among family carers (via multilevel linear regression).

Ethics

Ethical approval has been obtained from research ethics committees and ethical review boards of all participating hospitals in all countries. Trial registration: International Standard Randomised Controlled Trial Number (ISRCTN), 17231

Results

Sample characteristics

A total of 390 patients died in the year of inclusion, indicating that 390 bereavement questionnaires were distributed over six European countries. Only 162 returned: 71 for the intervention group; 91 for the control group (response rate: 41.5%). For 60 out of 71 patients in the intervention group (84.5%), the family carer had been present during the ACP-conversations. The majority of family carers were female (71.0%), had an average age of 60 (SD: 11.87) and were a partner (70.2%) of the deceased patient. The majority of the deceased patients were approximately 66 years (SD: 9.55), male (64.2%) and died from lung cancer (62,3%). See Appendix 2 for table: sample characteristics.

Family carers' involvement in decision-making and psychological distress

The probability that family carers had been involved in medical decision making with the patient during the last three months of life was slightly higher in the intervention group (89.6%) than in the control group (86.7%), but not statistically significant (OR 1.357, 95%-CI = 0.449 – 4.097).

The intervention group had a slightly higher mean score (M = 34.07; SD = 14.61) on the IES, meaning a higher level of psychological distress, than the control group (M = 31.77, SD= 14.11). However, this difference was not statistically significant either (t(157)= 1.005, 95%-CI = -2,224 – 6.836) (see Table 1).

Table 1: effect of ACP on involvement in decision-making and effect of ACP on psychological distress (measured with IES)

	involvement in decision-making	**Psychological distress (IES)**[a]
	OR (CI-95%) for I was involved as much as I wanted to be vs. I would have liked to be more involved	T-test (CI-95%)
Condition (Control vs. Intervention group)[b]	1.357 (0.449 – 4.097)	1.005 (-2,224 – 6.836)

Multilevel binary logistic regression analysis was used to measure the involvement in decision-making. Not included in the analysis were the responses: I would have liked to be less involved N = 0 (0%) and I don't know N = 4 (2.5%) and missing values N = 1 (0.6%).

Multilevel linear regression analysis was used in order to measure psychological distress, measured with the IES. [a] Possible range impact of events is 0-75. Missing items were replaced by the mean of the other items for that family carer.

Each item was controlled with family carers' age, gender, relationship towards patient; with patients' age, gender, having children & religion; with cancer type and WHO-status. Because no significant effect was found we used the basic regression model with dependent variable 'involvement in decision-making' or 'psychological distress' and independent variable 'condition'

[b] Control group is the reference category

Discussion and conclusion

The ACTION RC ACP intervention did not significantly affect family carers' involvement in decision-making in the last 3 months of patients' life, nor their level of psychological distress after three months of bereavement.

Several hypotheses can explain these results. The ACTION RC ACP intervention was delivered by a trained external facilitator. These facilitators had no contact with the patients' health care professionals and were also not allowed to add information about the ACP-conversation to the medical file of the patient. Participating patients were encouraged by the facilitators to communicate their preferences themselves to their health care professionals, but only few patients reported to have done so[13]. Previous studies have already shown that physicians' awareness of the patients' end-of-life care preferences did not improve when ACP conversations were conducted by nurses or other facilitators when these were not integrated into routine services, and it was suggested that a more interdisciplinary collaboration is needed[14]. Standardization is necessary in a research context in 6 different EU countries. Because of this, the ACTION RC ACP intervention was not integrated with routine services, and thus it might have reduced its effects[10] Probably a whole system-approach is necessary for ACP to be successful, which means that on an individual level, patients and their family carers are provided with the opportunity to have timely conversations, facilitated by skilled staff. It also means that on the system or health care service level appropriate policies and systems are in place to ensure that ACP is offered to patients and their family carers, and that previously discussed wishes and preferences are available to all involved health care professionals[15]. For future research, we would recommend exploring all options for broader involvement at the institutional level.

Moreover, although ACP needs to be patient-centred[15], family carers also need to be empowered and prepared for their role as SDM, which was possibly not sufficiently addressed in the ACTION RC ACP intervention. Little is known about how family carers should be optimally involved in ACP-conversations. Future research should focus on developing a family-focused ACP-model in addition to the current patient-centred ACP models.

Lastly, increasing literature is questioning whether ACP has the capacity to address goal-concordant care[16,17], which leads to a discussion about what the right outcome measures in ACP research ought to be[18]. Rather than focusing on making binding decisions early on, ACP should be seen as a process that facilitates patients, families and professionals to prepare for making better "in-the-moment" decisions. Consequently, outcomes of ACP should concentrate more on its process[18], and thus on the relation domain (e.g. patients and family carers discussing preferences, values and wishes with each other and preparing them for future conversations with professionals[17,18]). However, this was not assessed in the ACTION trial[10]. As for the results of this secondary analysis, other studies in ACP have measured well-being and psychological distress differently and currently, there is no univocal measurement or conclusion on the appropriateness of these outcome measures[4]. Future research should focus on a consistent way of measuring well-being and psychological distress of family carers after ACP. More specifically, qualitative research could enhance our understanding of the well-being and psychological distress of the family carer when engaging in ACP, as well as identify other relevant outcomes for patients and family carers.

This study has several strengths. First, the ACTION study is the first and largest RCT on the effects of ACP for patients with advanced cancer, and their family carers in Europe. Second, to study the effect of ACP for bereaved family carers, we sent out a bereavement questionnaire using items from the VOICES-SF and two validated scales measuring psychological distress, anxiety and depression symptoms.

The study has also some limitations. First, the response rate was modest in each of the 6 countries. As a result, between-country comparisons were not possible due to the low statistical power. Possible causes of non-response were that bereaved family carers were not interested in participating in the study anymore; and/or that the questionnaire was sent to the address of the deceased person, which was not always the address of the family carer. There was no further follow-up of the non-responders, which makes it difficult to understand family carers reasons for not responding. Second, a limited number of characteristics of the bereaved family carers were collected in the ACTION study, therefore we do not know whether the family carer who filled out the bereavement questionnaire was the appointed SDM of the patient.

Additional information

Acknowledgements

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Conflict of interest

The authors declare no conflict of interest

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Appendix 1: Methods

Study design

The ACTION-study was a multicentre cluster-randomised controlled trial in 23 hospitals in six European countries (Belgium, Denmark, Italy, the Netherlands, Slovenia and the United Kingdom). The hospitals were randomised to either the intervention group or to the control group. Patients with advanced lung- or colorectal cancer were asked to participate in the ACTION study and if they consented, they were followed for one year. If a patient died in the year after inclusion, a bereavement questionnaire was sent to the deceased's address. This article follows the CONSORT guidelines extension for cluster-randomized trials[1].

Setting and participants

In total, 1117 persons with advanced cancer participated in the ACTION study. People with lung or colorectal cancer with an estimated life expectancy of at least 3 months were eligible for this study, as both types of cancer have high incidence rates and affect both genders. The specific inclusion criteria for patients are described in the protocol paper[2]. The inclusion criteria for bereaved family carers' were: being older than 18 years, being able to complete the bereavement questionnaire in the language of each country and not taking part in another study that is evaluating palliative care services or communication interventions.

Intervention group vs. control group: ACTION Respecting Choices (RC) ACP intervention vs. care as usual

Patients in the intervention group were given a brochure with a brief description of ACP and questions prompting them to consider factors that may affect their personal goals for care. They were encouraged to read and discuss the information with their family carers. A trained facilitator scheduled facilitated ACP sessions with the patient who was encouraged to also invite a family carer. The intervention could involve either one session - with patient and family carer - or two sessions – the first one only with the patient and second one with patient and family carer. Researchers of the ACTION consortium went to the US and were trained to administer the ACP intervention according to the original US Respecting Choices (RC) programme. Then the RC programme was adapted to the European culture and the facilitators – which were clinical professionals or researchers - were trained to administer the ACTION RC ACP intervention. The following topics could be discussed in the ACP sessions: patient's understanding of the illness, appointing a surrogate decision-maker or personal representative, patient's preferences regarding resuscitation, goals of future care (comfort-focused care or selective treatment), final place of care and other preferences. Patients were encouraged to share their preferences with family members and loved ones. If they wished, patients were supported to document their preferences, including the indication of a "personal representative" (namely, a surrogate decision-maker, further used in text). The intervention group received ACTION RC ACP intervention in addition to 'care as usual'. The control group received care as usual where they were informed that they were included into a study focussing on preparing patients for decision-making about current and future care.[2] If a patient died during follow-up, a family carer was asked to complete a written questionnaire after three months of bereavement.

Procedures

Randomisation

A total of 23 hospitals were randomised, by an independent statistician, to either the intervention group or control group. Cluster-randomisation was used in order to minimize contamination. All eligible patients who consented, were included and followed until one year after inclusion or until death. More detailed information about the trial procedures can be found in the protocol paper[2].

Recruitment of the bereaved family carers

The main purpose of the bereavement questionnaire was to obtain more information about the last months and weeks of the patients' life as perceived by a bereaved family carer. A total of 390 (34.91%) patients died during the year after inclusion (157 in the intervention group and 233 in the control group). Each bereavement questionnaire contained a cover letter, information sheet and prepaid envelope. A reminder was sent when the questionnaire was not returned after two weeks. Return and completion of the questionnaire were considered to indicate that family carers had consented to participate in the study.

Outcome measures

In the bereavement questionnaire, family carers' perception about '*involvement in decision-making in the last three months of the deceased's life'* was measured with a single item of the Views of Informal Carers – Evaluation of Services Short Form (VOICES-SF) questionnaire. The item used was: "Looking back over the last three months of his/her life, were you involved in decisions about his/her care as much as you would have wanted?". Possible responses were: 1) I was involved as much as I wanted to be, 2) I would have liked to be more involved, 3) I would have liked to be less involved, 4) Don't know. The VOICES-SF questionnaire is a 58-item validated questionnaire about health and social services completed by bereaved relatives, who act as proxies to recall the deceased persons' experience in the last three months of life. The VOICES-SF is used as an annual survey to evaluate the quality of end-of-life care in the UK[3,4].

Psychological distress was assessed using the impact of event scale (IES) and asked how frequently each item was experienced during the past week after 3 months of bereavement. The IES measures psychological responses to stress on two subscales: (1) intrusion and (2) avoidance and is mostly used to measure post-traumatic stress syndrome. Each item could be rated with 1) not at all, 2) rarely, 3) sometimes and 4) often[5]. During the ACTION-study no baseline measurement or other follow-up questionnaire was sent to the family carer.

Statistical analysis

Data were analysed using SPSS Statistics version 25. All family carers who returned completed or partially completed questionnaires were included in the analyses, regardless of whether they had responded to all the questions. Descriptive statistics were used to summarize the data, therefore we adopted contingency tables with the χ^2-test for categorical data and univariate independent t-tests for the non-categorical data.

As the data are nested on 3 levels (level 1: family carer – level 2: hospital – level 3: country), the effect of ACP on involvement in decision making among family carers' was determined by multilevel binary logistic regression. The item was recoded into a dummy variable with answer-options: "I was involved as much as I wanted to be" and "I would have liked to be more involved". The other options were coded as missing. The effect of ACP on psychological distress among family carers' was determined by multilevel linear regression. Involvement in decision-making and psychological distress were controlled with family carers' age, gender and relationship towards patient, with patients' age, gender, having children and religion; with cancer type and WHO-status. However, no statistical inferences were found. Statistical inferences were based on two-sided tests at a significance level of $P < 0.05$, using the Bonferroni-correction.

Ethics

Ethical approval has been obtained from research ethics committees and ethical review boards of all participating hospitals in all countries. Trial registration: International Standard Randomised Controlled Trial Number (ISRCTN), 17231

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Appendix 2: table: sample characteristics

Table. Characteristics of bereaved family carers and deceased patients

	Total	**Intervention**	**Control**	**p-value[a]**
Characteristics family carers				
Age (in years), mean (SD)	60.32 (12.39)	60.08 (13.09)	60.50 (11.87)	0.833
Gender, *male* (%)	47 (29.0%)	20 (28.2%)	27 (29.7%)	0.835
Relationship to patient				0.948
Husband/Partner (%)	113 (70.2%)	49 (69.0%)	64 (71.1%)	
Son/Daughter (%)	30 (18.6%)	14 (19.7%)	16 (17.8%)	
Someone else (%)	18 (11.2%)	8 (11.3%)	10 (11.1%)	
Characteristics patients				
Age (in years), mean (SD)	66.11 (10.40)	65.66 (11.43)	66.46 (9.55)	0.632
Gender, *male* (%)	104 (64.2%)	43 (60.6%)	61 (67.0%)	0.394
Children, *yes* (%)	144 (89.4%)	66 (93.0%)	78 (86.7%)	0.197
Religion, *yes* (%)	82 (60.7%)	39 (62.9%)	43 (58.9%)	0.635
Clinical characteristics				
Cancer type				0.061
Lung cancer, stage III or IV	101 (62.3%)	50 (70.4%)	51 (56.0%)	
Colorectal Cancer, stage IV	61 (37.7%)	21 (29.6%)	40 (44.0%)	
WHO performance status[b]				0.398
0-Patient is fully active & more or less as he/she was before his/her illness (%)	37 (23.1%)	12 (16.9%)	25 (28.1%)	

1-Patient cannot carry out heavy physical work, but can do anything else (%)	88 (55.0%)	43 (60.6%)	45 (50.6%)
2-Patient is up for more than half the day and can look after his/herself, but is not well enough to work (%)	30 (18.8%)	14 (19.7%)	16 (18.0%)
3-Patient is in bed or sitting in a chair for more than half the day and needs some help in looking after his/herself (%)	5 (3.1%)	2 (2.8%)	3 (3.4%)

Percentages are column percentages. Not included in table and analyses: missing data in characteristics FCs for gender N = 1 (0.6%); relationship to patient N = 4 (2.5%). Missing data in characteristics patient for gender N = 2 (1.2%), children N = 1 (0.6%); Religion N = 3 (1.8%). Missing data in clinical characteristics: WHO performance status N = 2 (1.2%). 'prefer not to specify'
in religion N = 24 (14.7%). [a] Bivariate differences between intervention and control group are calculated using the Pearson's χ^2-tests.

[b] WHO status is based on the one in baseline

Chapter 5:

Quality of end-of-life nursing home care in dementia: relatives' perceptions

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Abstract

Objectives: Only a few studies have investigated the quality of end-of-life care provided to nursing home residents with dementia as perceived by their relatives. We aim to investigate the quality of end-of-life care as perceived by relatives and to investigate which characteristics of nursing home residents with dementia, their relatives, and the care they received are associated with the evaluation the quality of end-of-life care as perceived by the relatives.

Methods: Data used was from two cross-sectional studies performed in Flanders in 2010 and 2015. Questionnaires were sent to bereaved relatives of nursing home residents with dementia and 208 questionnaires were returned. The quality of end-of-life care as perceived by the relatives was measured with the End-Of-Life with Dementia – Satisfaction With Care scale (EOLD-SWC, scores ranging 10-40).

Results: In total, 208 (response rate^{2010}: 51.05%; response rate^{2015} = 60.65%) bereaved relatives responded to the questionnaire. The quality of end-of-life care as perceived by them was positively associated with the nursing home resident being male ($b = 1.78$, $p < .05$), relatives receiving information on palliative care ($b = 2.92$, $p < .01$) and relatives receiving information about medical care from care providers ($b = 2.22$, $p < .01$).

Conclusion: This study suggests that relatives need to be well-informed about palliative and medical care. Future end-of-life care interventions in nursing homes should focus on how to increase the information-exchange and communication between nursing home staff and relatives.

Keywords: relatives; quality of end-of-life care; information and communication; cross-sectional design; nursing homes

Introduction

The number of people living with dementia today is about 50 million worldwide, and is expected to increase to 82 million in 2030 and 152 million in 2050[1]. Although many people wish to die at home, those with dementia often die in a nursing home[2,3]. In Flanders, Belgium, nearly half of people admitted to a nursing home die with dementia, of which half is advanced dementia[3]. Providing good quality end-of-life care for people with dementia is thus an important aspect of nursing home care[4]. However, different studies have shown that the quality of end-of-life care needs improvement in nursing home residents with and without dementia[5–7]. Providing high quality end-of-life care in dementia is challenging[8], because dementia is often not seen as terminal condition by health care providers[6], which may lead to poor symptom management or hospital transfers at the end of life[8]. Nursing home residents with dementia are usually less able to communicate their needs and symptoms or to provide information of symptom improvement following treatment near the end of life[8]. Thus, palliative care in people with dementia can improve symptom burden, prevent undertreatment of symptoms and overtreatment with unnecessary and burdensome interventions. Next, it may decrease the burden on relatives and improve the quality of life of both the person with dementia and their relative [9,10].

Relatives play an important role in the care of people with dementia, even after admission to a nursing home[11]. They often remain closely involved in their care until the end of life[12] and can provide key information about the resident to the healthcare providers[12,13]. Therefore, the perspective of relatives about the quality of end-of-life care is of value. However, only a few studies have so far evaluated the quality of end-of-life care from the perspective of relatives of residents with dementia. Previous cross-national studies have indicated that most relatives are grateful for the end-of-life care that the resident with dementia received[14,15] and that the quality of end-of-life care as perceived by the relatives is strongly associated with how peacefully their loved one had died[16]. Furthermore, research has also shown that the relatives' experiences with end-of-life care also affects the extent, duration and intensity of their grief[17]. Still, little is known, so far, about which factors related to the resident, their relative and their care are associated with the relative's perception of the quality of end-of-life care received.

Knowing what factors are associated with the quality of end-of-life care in nursing homes as perceived by relatives will help to improve its quality[13], but it may also identify factors that are important in supporting relatives at the end of the resident's life. Therefore, the research questions of this study are: "what is the quality of end-of-life care for nursing home residents with dementia as perceived by their relatives?" and "which characteristics of nursing home residents with dementia, their relatives, and the care they received are associated with the quality of end-of-life care as perceived by the relatives?"

Methods

Study design

This study uses data from two cross-sectional mortality follow-back studies, the Dying Well with Dementia Study conducted in 2010[18], and the Palliative Care for Older People in Care and Nursing Homes in Europe (PACE) study (data collected in 2015)[19]. The study in 2010 was a cross-sectional study of nursing home residents with dementia in Flanders, Belgium. The study in 2015 was a cross-sectional study of deceased nursing home residents in six European countries, of which only the sample of Flanders, Belgium was selected for this study. The methods of both studies are summarized here and are described in depth elsewhere[18,19]. Both studies had an identical design except for the screening procedure to identify the study population and the number of reminders sent to the respondents.

Setting

Both studies identified representative samples of nursing homes in Flanders, Belgium using the same cross-sectional study design to identify relatives of deceased nursing home residents. Nursing homes in Flanders are long term care facilities where nursing care is available on-site 24/7, and care from physicians is available off-site[19]. In both studies, random stratified sampling was conducted of nursing homes by region (five provinces), bed capacity (up to or more than 90, which is the median number of beds in nursing homes in Flanders) and ownership (public, private/non-profit, private/profit).

Data collection and study participants

For this study, we identified the relatives of the deceased residents with dementia from the studies in 2010 and 2015 (see figure 1) as follows: in the study of 2010, deceased residents with dementia were identified in two steps; first, the administrator was asked to identify all the residents who had died in the predefined period and to include those who had possibly had dementia using the screening criteria of the KATZ scale, used by the Belgian health insurance system to assign financial resources: Category C dementia i.e. 'being completely care dependent or needing help for bathing, dressing, eating, toileting, continence, and transferring plus being disoriented in time and space' OR disorientation in time and space (KATZ scale ≥ 3 or 'having almost daily a problem with disorientation in time and space'). If a resident was identified with one of these two criteria, they were included for data collection in the study of 2010 (n=241). Second, the nursing staff and General Practitioner (GP) were asked whether they thought the resident 'had dementia' or 'was diagnosed with dementia'. If either one indicated 'yes', then the person was considered as someone with dementia. In the study of 2015, questionnaires on all deceased residents were sent to the administrator, nursing staff, GP and relative (without performing the first step pre-selection in the study of 2010). Residents were considered as having dementia following the same second step procedure of 2010.

In this study we are interested in the quality of end-of-life care as perceived by the relatives and therefore only included the cases for whom the relatives had returned a questionnaire.

Measurements

Dependent variable

The main outcome, the quality of end-of-life care as perceived by the relatives, was measured using the validated *satisfaction with care – at the end-of-life in dementia (EOLD-SWC) scale*[20]. The EOLD-SWC scale is the most appropriate instrument for measuring quality of end-of-life care as perceived by relatives[21]. The scale consists of ten items (e.g. I felt fully involved in all decision-making) whereby each item can be scored on a 4-point Likert scale: 1) strongly disagree; 2) disagree; 3) agree; and 4) strongly agree. The total score is calculated as the sum score of each item on the 4-point Likert scale and ranges between 10 and 40. The higher the total score on the SWC-EOLD scale, the higher the quality of end-of-life care as perceived by the relatives.

Independent variables

The following characteristics were assessed in the relatives' questionnaire: age of the respondent in years, gender, educational level, relationship with the resident, whether the relative was the most important proxy, whether relatives received information on palliative care or on medical care from a care provider and whether they had discussed preferred medical treatments with the resident.

Questionnaires from the administrator were used to assess characteristics of the residents: age in years, gender, place of death and length of stay in the nursing home. The nursing staff questionnaire was used to measure the resident's functional status one month before death using the *Bedford Alzheimer Nursing Severity Scale* (BANS-S)[22]. This scale consists of seven items (e.g. dressing) whereby each item can be scored from 1 to 4. The total score is the sum of scores, and it ranges from 7 (no impairment) to 28 (complete impairment).

Statistical analysis

Data was analysed using SPSS statistics version 26. Firstly we tested for possible differences between the years 2010 and 2015 in the relatives' evaluation on the quality of end-of-life care, but no differences were found. The comparability of the data allowed us to combine the data of 2010 and 2015 for the analysis. The assumption of linearity was violated in the variable 'Length of Stay' so this variable was recoded as a categorical variable. We performed univariable multilevel analyses (due to clustering of data in the nursing homes). Multivariable multilevel analyses were tested, whereas we controlled for gender of the resident, length of stay in the nursing home, whether relatives received information on palliative care, and whether they received information on medical care from a care provider. These variables were deemed significant in the univariable multilevel analyses. A p-value of <0.05 was deemed significant in all analyses.

Ethics approval

Ethics approval for both studies was obtained from the Ethical Review Board of Brussels University hospital of the Vrije Universiteit Brussel (B.U.N. 14320108163 (2010); B.U.N. 143201422845 (2015)).

Results

Characteristics of the nursing home residents with dementia

In total, 208 (n^{2010} = 97 (response rate: 51.05%); n^{2015} = 111 (response rate: 60.65%) deceased residents with dementia were included in this study (Table 1). At the time of the studies, mean age of these residents at the time of death was 87.27 years. The mean total BANS-S score of these residents with dementia was 20.49. The length of stay of the nursing home was less than six months for 17.5% of the residents with dementia, 6-12 months for 8% and more than 12 months for 74.5% (Table 1).

Characteristics of the relatives

The median age of the relatives of the deceased nursing home resident with dementia was 61 years. Most relatives were a child of the deceased resident (69.7%). In most cases, the relatives considered themselves as the most important proxy (88.5%). The majority had received higher secondary education (37.3%) (Table 1).

Care characteristics

63.6% of relatives indicated they had received information on palliative care from a care provider, either personally or via leaflets; 59.2% had received information about medical care and 76.3% indicated that the resident had not talked about preferred medical treatments with them or with someone else (Table 1).

Table 1: Characteristics of nursing home residents with dementia and their relatives (N = 208, data 2010-2015 combined)[a]

	2010-2015 N=208
Characteristics of the nursing home residents with dementia	
Age at time of death, mean[b] (S.D.)	87.27 (6.9)
Gender, male n(%)[c]	75 (37.9)
Functional/cognitive status one month before death (BANS-S), mean (S.D.)[d]	20.49 (4.1)
Place of death, n(%)[c, e]	
Nursing home	178 (89.0)
Hospital, incl PCU	22 (11.0)
Length of stay in NHs in months, n (%)[f]	
<6 months	35 (17.5)
6 – 12 months	16 (8.0)
>12 months	149 (74.5)
Characteristics of the relatives[g]	
Age (in years), mean (S.D.)	61.07 (10.4)
Gender, male n(%)	74 (36.6)
Relation towards resident, n(%)	
Partner	21 (10.1)
Son or daughter	145 (69.7)
Other	42 (20.2)
Are you the most important proxy? yes n(%)	184 (88.5)
Education level, n(%)	
Primary school	25 (12.4)
Lower secondary education (up to GCSE or equivalent)	59 (29.4)
Higher secondary education (up to A level or equivalent)	75 (37.3)
Tertiary education or higher (higher vocational training or university)	42 (20.9)
Care characteristics[g]	
Relatives received information on palliative care[h]	
Yes (personally or via leaflets)	131 (63.6)
No	75 (36.4)
Relatives received information on medical care from a care provider	
Yes	122 (59.2)
No	84 (40.8)
Relative discussed preferred medical treatments with resident with dementia	
Yes	33 (15.9)
No	158 (76.3)
I don't know	16 (7.7)

Percentages are the valid percentages in frequency tables.

Abbreviations: SD; Standard Deviation; BANS-S: Bedford Alzheimer Nursing Severity-Scale; PCU: Palliative Care Unit; NH: Nursing Homes

[a] Table presents characteristics of the nursing home residents with dementia for whom nursing staff and the administrator filled in a questionnaire and characteristics of the relatives, clinical characteristics, and care processes (relatives' questionnaire):

[b] Calculated by subtracting date of admission from date of death

[c] Missing data reported by administrator: residents' age: 5.8%; residents' gender: 4.8%; and length of stay in the nursing home: 3.8%

[d] Missing data reported by nursing staff: BANS-S: 1.4%; place of death: 3.8%

[e] Place of death was recoded into a dummy variable due to very limited cases present at hospital and palliative care unit (PCU).

[f] Assumption of linearity was violated in the Length of Stay, thus it was recoded into following categories: <6 months, 6-12 months and >12 months

[g] Missing data reported by relatives: relatives' age: 16.8%; relatives' gender: 2.9%; and education level: 3.4%; received information on palliative care: 1%; received information on medical care from a care provider: 1%; and relative discussed preferred medical treatments with resident with dementia: 0.5%

[h] Relatives' receiving information on palliative care was recoded into a dummy variable: options 'yes' and 'not explained personally' were recoded into 'yes', option 'no' remained 'no'.

The evaluation of the quality of end-of-life care as perceived by the relatives

The total mean score of the quality of end-of-life care as perceived by the relatives is 32.66 (SD 5.24). The highest mean score was for the item 'All measures were taken to keep the resident with dementia comfortable' (mean=3,46; SD=0,69), whereas the lowest mean item score was for 'I always knew which doctor or nurse was in charge of the resident with dementia' (mean=2,81, SD=0,94) (Table 2).

Table 2: The quality of end-of-life care as perceived by the relatives (EOLD-SWC scores)

Mean scores (SD)	**2010-2015 combined N=208**
SWC total score: possible range = 10 (worst) – 40 (best)[a]	32.66 (5.24)
SWC item scores: possible range= 1 (worst) – 4 (best)	
1 I felt fully involved in all decision-making	3,30 (0,74)
2 I would probably have made different decisions if I had had more information[b]	3,35 (0,67)
3 All measures were taken to keep the resident with dementia comfortable	3,46 (0,69)
4 The healthcare team was sensitive to my needs and feelings	3,32 (0,70)
5 I did not really understand the resident with dementia's condition[b]	3,28 (0,85)
6 I always knew which doctor or nurse was in charge of the resident with dementia	2,98 (0,93)
7 I feel that the resident with dementia got all necessary nursing assistance	3,42 (0,70)
8 I felt that all medication issues were clearly explained to me	2,81 (0,94)
9 The resident with dementia received all treatments or interventions that (s)he could have benefited from	3,39 (0,67)
10 I feel that the resident with dementia needed better medical care at the end of his/her life[b]	3,26 (0,89)

[a] The total score was calculated on the four responses: 'strongly disagree'; 'disagree'; 'agree' and 'strongly agree'. If the total score was more than 40, we reported it as missing.

[b] Recoded so that higher score means better satisfaction

Missing item values: 1 n=8 (3.8%); 2 n=13 (6.3%); 3 n=7 (3.4%); 4 n=14 (6.7%); 5 n=12 (5.8%); 6 n=10 (4.8%); 7 n=8 (3.8%); 8 n=10 (4.8%); 9 n=11 (5.3%); 10 n=10 (4.8%)

Factors associated with the quality of end-of-life care as perceived by relatives

The multivariable analyses showed that the quality of end-of-life care as perceived by the relatives was evaluated as significantly higher if the resident with dementia was male (EM = 32.9) than if they were female (EM = 31.13) (b = 1.78, p < .05). Relatives who indicated that they had received information on palliative care (EM = 33.47) gave a significantly higher total score for the quality of end-of-life care as perceived by the relatives than those who had not received this information (EM = 30.56) (b = 2.92, p < .01). The quality of end-of-life care as perceived by the relatives was significantly higher when relatives had received information on medical care from a care provider (EM = 33.12) than for those who had not (EM = 30.91) (b = 2.22, p < .01). No other significant associations were found in resident, relative and care characteristics (Table 3).

Table 3: **Multilevel linear regression analysis of the association between the relatives' evaluation on the quality of end-of-life care and nursing home resident with dementia, relative, and care characteristics of two years (N = 208, data from 2010 and 2015 combined)[a]**

	Estimated means (EM)[b]	**Coefficient b (95% CI)**	**p-value[c]**
Characteristics of the nursing home resident with dementia			
Age at time of death	32.15	.04 (-.07 - .15)	.47
Gender			
Men	32.90	1.76 (.30 – 3.26)	**.02**
Women	31.13		
Functional/cognitive status one month before death (BANS-S)	31.98	-.07 (-.26 - .12)	.49
Place of death			
Nursing home	32.10	1.05 (-1.64 – 3.74)	.44
Hospital, incl PCU	31.04		
Length of stay in NHs in months[d]			
<6 months	30.98	-1.20 (-3.19 - .78)	.23
6 – 12 months	32.90	.70 (-2.06 – 3.46)	.62
>12 months	32.18	REF	
Characteristics of the relatives			
Age (in years)	31.77	.05 (-.03 - .13)	.18
Gender			
Men	31.61	-.71 (-2.25 - .82)	.36
Women	32.32	REF	
Relation towards resident			
Partner	31.57	.14 (-2.69 – 2.98)	.92
Son or daughter	32.35	.92 (-.92-2.77)	.33

Other	31.43	REF	
Are you the most important proxy?			
Yes	32.07	.28 (-2.08 – 2.64)	.82
No, that is not me but someone else	31.79	REF	
Education level			
Primary school	31.89	-1.12 (-3.97 – 1.73)	.44
Lower secondary education (up to GCSE or equivalent)	32.03	-.98 (-3.18 – 1.22)	.38
Higher secondary education (up to A level or equivalent)	31.54	-1.48 (-3.5 - .55)	.15
Tertiary education or higher (higher vocational training or university)	33.01	REF	
Care characteristics			
Relatives received information on palliative care			
Yes (personally or via leaflets)	33.47	2.92 (1.24 - 4.59)	**.00**
No	30.56	REF	
Relatives received information on medical care from a care provider			
Yes	33.12	2.22 (.59 – 3.85)	**.01**
No	30.91	REF	
Relative discussed preferred medical treatments with the resident with dementia			
Yes	31.53	REF	
No	32.23	.55 (-1.53 – 2.62)	.60
I don't know	31.69	-.15 (-3.26 – 2.95)	.92

Abbreviations: SD: Standard Deviation, BANS-S: Bedford Alzheimer Nursing Severity-Scale; PCU: Palliative Care Unit; CI: Confidence Interval

[a] Generalised linear mixed models with total score of the EOLD-SWC as dependent variable, years (2010 vs 2015) as fixed factor

[b] Calculated using general linear mixed model; adjusted for correlation withing clusters (nursing homes) and years (2010 vs 2015) as fixed factor. The EOLD-SWC total score ranges from 10 (worst) to 40 (best).

[c] Differences were calculated using generalized linear mixed models, p<0.05 was deemed significant.

[d] Assumption of linearity was violated in the Length of Stay, thus it was recoded into following categories: <6 months, 6-12 months and >12 months

Discussion

This study shows that the quality of end-of-life care as perceived by relatives of nursing home residents with dementia seems relatively high and is associated with several resident and care characteristics. It was higher in cases where the deceased resident with dementia was male, when relatives indicated they had received information on palliative care and when they indicated they had received information on medical care from a care provider.

The strengths of the study are the use of data from large representative nationwide cross-sectional mortality follow-back studies of nursing homes in Flanders, Belgium and the high response rates of the relatives (response rate^{2010} = 51.05%; response rate^{2015} 60.65%)[18,19]. Another strength is the use of a validated scale (EOLD-SWC) with good psychometric properties to measure the quality of end-of-life care as perceived by the relatives[21]. On the other hand, EOLD-SWC is a subjective measurement and the total mean score might be biased by the expectations relatives have about the end-of-life care[14]. Recall bias cannot be excluded, because perceptions were measured three months after the death of the resident with dementia.

Overall, it seems that the relatives are relatively satisfied with many aspects of the quality of end-of-life care that the nursing home resident with dementia had received. The quality end-of-life care as perceived by the relatives was positively associated with the resident with dementia being male. This is in contrast with what we expected since other studies show that male nursing home residents with dementia often receive more burdensome treatments at the end-of-life than female residents with dementia[2,23]. However, these studies did not focus on the perspectives of relatives. Based on our result, we cannot explain why relatives perceive the quality of end-of-life care for male residents better than for female residents with dementia. More research is needed to further explore this result. Relatives who received information on palliative or on medical care tend to evaluate the quality of care better than those who had not received information on palliative or medical care. This finding could be interpreted in two directions. Providing information on palliative and medical care might be an important prerequisite of good quality end-of-life care as assessed by relatives; it might also be that the staff in nursing homes where good quality of end-of-life care is provided automatically provide more information on palliative and medical care to the relatives. This relationship between the provision of information and evaluation of care quality corresponds with the findings from previous studies about the importance for relatives of being informed, not only about medical care, but also about the end of life[24,25]. Studies show that informing relatives about dementia, its prognosis and the dying process, is highly important for good quality of end-of-life care as perceived by the relatives, because lack of information causes them distress about what is going to happen regarding prognosis and the dying process, and about the decision-making process[26]. Furthermore, a qualitative study with relatives of deceased nursing home resident with dementia conducted in the UK also showed that when relatives are informed about and involved in the care process, they will not only perceive the quality of end-of-life care more positively but they will also be better prepared to make decisions on behalf of the resident[27].

Remarkably, our study showed that 36.4% of the relatives did not receive information on palliative care and 40.8% did not receive information about medical care from a care provider. Thus, given that communication and the provision of information are important factors in the relatives perceiving good quality of end-of-life care, and given that a third of them indicated they had not received information on palliative care and/or on medical care, there is room for improvement. Previous findings of qualitative interview studies in the Netherlands showed that relatives valued formal and informal communication with the nursing staff. However, they experienced barriers to initiating communication themselves, e.g. not wanting to burden the staff[28], which led to frustration because they were not informed about the residents' condition[24]. Thus it seems important that nursing staff take the initiative in talking to relatives openly about medical and/or palliative care on a regular basis[24,25]. One possible way to exchange more information could be to organise family conferences in the nursing home or to appoint a care coordinator who can help in enhancing communication between relatives and nursing staff[25]. However, literature has shown that nursing home staff may experience difficulties in recognizing the end of life and dying in residents with dementia, which may hamper open communication with relatives[8]. Organizing regular multidisciplinary meetings in the nursing home to discuss the changes in physical condition of nursing home residents may help staff to better identify dying phase[29]. Future research is necessary to enhance the information exchange between nursing staff and relatives and to learn how much information relatives want or need and on which occasions they believe it is required. More recent studies highlight the importance of involving relatives as equal members of the care team[24,25,27], which may lead to a shared vision about the care that is provided[25]. Future research is necessary on how involvement of relatives in the care team may affect their perception of the quality of end-of-life care.

Conclusion

Relatives receiving information on palliative and medical care from a care provider is associated with them perceiving a better quality of end-of-life care. These results confirm the importance of relatives being well-informed and involved in discussions about palliative and medical care together with the care team. Future end-of-life care interventions in nursing homes should involve the relatives to increase the exchange of information and communication between nursing home staff and relatives.

Additional information

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Conflict of interests

Conflict of interests: none

Authors contribution

All authors gave final approval of the submitted manuscript and agreed to be accountable for all aspects of the work. All authors' specific contributions are listed below.

- Study concept and design: IV, ADV, AVDH, LD, LVDB & TS
- Analysis and interpretation of data: : IV, ADV, AVDH, LD, LVDB & TS
- Preparation of manuscript: : IV, ADV, AVDH, LD, LVDB & TS

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GENERAL DISCUSSION

General discussion

Some parts of this general discussion are based on, or to a certain degree copied from the following articles:

Vandenbogaerde I, Miranda R, De Bleecker JL, Carduff E, van der Heide A, Van den Block L, Deliens L, De Vleminck A. Advance care planning in amyotrophic lateral sclerosis (ALS): study protocol for a qualitative longitudinal study with persons with ALS and their family carers. BMJ Open. 2022 May 12;12(5):e060451. doi: 10.1136/bmjopen-2021-060451. PMID: 35551085 [2021 IF 3.006; Ranking Q2; ranking 85/172 MEDICINE, GENERAL & INTERNAL]

Vandenbogaerde I, Cohen J, Van Audenhove C, Hudson P, Deliens L & De Vleminck A. Support from healthcare professionals in empowering family carers to discuss Advance Care Planning: A population-based survey. Palliat Med. 2022 Nov 9:2692163221135032. doi: 10.1177/02692163221135032. Online ahead of print. PMID: 36349646 [2021 IF 5.713; Ranking Q1; ranking 14/109 HEALTH CARE SCIENCES & SERVICES in SCIE edition]

Vandenbogaerde I, De Vleminck A, Cohen J, Verkissen MN, Lapeire L, Ingravallo F, Payne S, Wilcock A, Seymour J, Kars M, Grønvold M, Lunder U, Rietjens J, van der Heide A, Deliens L. Advance care planning-family carer psychological distress and involvement in decision making: the ACTION trial. BMJ Support Palliat Care. 2022 Feb 17:bmjspcare-2020-002744. doi: 10.1136/bmjspcare-2020-002744. Epub ahead of print. PMID: 35177432. *[* SCI impact factor (2021): 4.633 – journal ranking Q1; ranking 25/109 HEALTH CARE SCIENCES & SERVICES in SCIE edition]

Vandenbogaerde I, De Vleminck A, van der Heide A, Deliens L, Van den Block L, Smets T. Quality of end-of-life nursing home care in dementia: relatives' perceptions. BMJ Support Palliat Care. 2022 Jun 16:bmjspcare-2021-003497. doi: 10.1136/bmjspcare-2021-003497. Epub ahead of print. PMID: 35710707. [2021 IF 4.633; Ranking Q1; ranking 25/109 HEALTH CARE SCIENCES & SERVICES in SCIE edition]

The research aims of this dissertation were twofold: (1) to understand in-depth experiences with advance care planning in a disease trajectory of ALS and (2) to investigate family carers' involvement in advance care planning and their evaluation of end-of-life care. In this part, the main findings of the studies will be summarized, followed by the methodological strengths and limitations of the studies and a general discussion of the most important findings. Finally, we propose some implications for policy, practice, and future research.

Summary of the main findings

To understand in-depth experiences with advance care planning in a disease trajectory of ALS (research aim 1)

In **Chapter 1**, we developed a study protocol for a qualitative longitudinal multi-perspective interview study to understand in-depth experiences with advance care planning in ALS via patients and family carers' perspectives and how these experiences unfold over time throughout the disease trajectory of ALS. The results are described in **Chapter 2**. Over a timespan of about nine months, we were able to interview nine dyads of people with ALS and their family carer (mostly) individually about advance care planning. We interviewed them three times each, resulting in a total of 44 interviews. Overall, all the persons with ALS and their family carers thought about their future and future (end-of-life) care to some extent over time, but not all dyads talked about it amongst each other or with a healthcare professional. Over time, advance care planning experiences were influenced by several intertwined determining factors: (1) the experienced physical decline such as no longer being able to walk or talk and related future care needs that changes over time; (2) how persons with ALS identify themselves as patients; (3) obtaining information about diagnosis and prognosis and learning that everyone evolves differently which makes planning for the future difficult; (4) professionals initiating conversations about medical aspects of end-of-life decisions, even though participants experienced it as stressful or too early; (5) balancing between hope to remain stable and worry about what the future might hold; (6) protecting themselves and each other from the future.

To investigate family carers' involvement in advance care planning and their evaluation of end-of-life care (research aim 2)

In **Chapter 3 and 4** we investigated family carers involvement in advance care planning. We examined the extent to which family carers of people with a serious illness performed advance care planning conversations with their relative at home in the last three months of life and how they were supported by healthcare professionals to conduct these conversations in **chapter 3**. We conducted a population-based survey study. In this survey study, we included bereaved family carers who had provided care to someone who recently died (between two and six months ago) via the registers of three largest health insurers in Flanders, Belgium. Our findings show that about half of the family carers indicated to have engaged in advance care planning conversations with their relative in the final three months of life. These advance care planning conversations were more likely to have taken place when family carers were 55 years of age or younger, had a medical degree (doctor or nurse) or when specialist palliative care services were involved, but it was less likely when the deceased person was diagnosed with dementia. Seventy-eight percent of the family carers received support from a healthcare professional, which was mostly the general practitioner. The type of support received was most often the healthcare professional doing the conversation together with the family carer or seriously ill person, which can be considered as an empowering collaborative approach. Seldomly the more empowering support types to increase self-efficacy in family carers were reported in this study such as providing information or explaining how to conduct these conversations.

In **chapter 4** we reported on the secondary outcomes of the ACTION trial. We investigated the effect of the ACTION Respecting Choices (RC) advance care planning intervention on family carers' involvement in decision-making in the last three months of the patient's life, and psychological distress after three months of bereavement. The ACTION trial was a multicentre cluster-randomised controlled trial in six European countries (Belgium, Denmark, Italy, the Netherlands, Slovenia and the UK) which adapted and tested the efficacy of the ACTION RC advance care planning intervention in patients with advanced lung or colorectal cancer. The intervention group received ACTION RC advance care planning intervention and care as usual, whereas the control group received care as usual. Persons with advanced cancer who were part of the intervention group were invited to have a maximum of two advance care planning sessions together with their family carer and with a trained facilitator. The patients were encouraged to share their preferences with family members and loved ones, and if they wished to document these preferences. For this study, data of the bereavement questionnaire was used. We found that bereaved family carers of patients who participated in ACTION RC advance care planning intervention did not have a significantly higher probability to be involved in decision-making in the last three months of the patient's life, but it was slightly higher in the intervention group (89.6%) than in the control group (86.7%). The intervention group had a slightly higher level of psychological distress three months after bereavement than the control group, but this difference was also not statistically significant.

In **chapter 5,** we examined the family carers' evaluation of quality of end-of-life care for nursing home residents with dementia and the factors that might be associated with this evaluation of the quality of end-of-life care. We found that the family carers' evaluation of the quality of end-life care for nursing home residents with dementia was relatively high. The evaluation was higher, when family carers indicated they had received information on palliative care, and when family carers indicated they had received information on medical care from a healthcare professional.

Strengths and limitations

This doctoral thesis made use of several research methods to address the specific research aims, each having their own strengths and limitations.

A qualitative longitudinal multi-perspective interview study

In **Chapter 1 and 2** we described the methodology and reported the results of a qualitative longitudinal multi-perspective interview study which was set up to understand an evolving and complex process such as advance care planning over time from the perspectives of both persons with ALS and their family carers. Advance care planning is usually measured and described in a single point in time, but the disease's complexity and the complexity of the advance care planning communication process cannot be fully captured via these snapshot techniques such as cross-sectional designs. Therefore, we chose this qualitative longitudinal study design.

One of the major strengths of this study was the combination of a longitudinal design with a multi-perspective interview design. These are innovative methods and are currently understudied in medicine in general as well as in the field of palliative care research. Another important strength was that the perspectives of both persons with ALS and their family carers were analysed to enhance our understanding of the dynamics and relationships between them. This multi-perspective approach also increased our insights of the individual needs of persons with ALS and their family carers in advance care planning. This approach allowed us to explore similarities and differences in their views about advance care planning. A final important strength of longitudinal qualitative study is that it was a less restrictive approach towards studying time and change in complex processes such as advance care planning, if combined with flexibility, such as monthly phone calls in order to grasp change[1,2]. These monthly phone calls in between the three interviews facilitated a more nuanced understanding of the illness trajectory and the implications of the experiences with advance care planning in case a sudden change appeared. In that case a subsequent interview would be planned sooner than anticipated. As a result, we were able to conduct an interview that otherwise would have been missed because the patient would have died[3].

This study also has some limitations. First, there might be some selection bias as the participants who wanted to participate were interested in this topic. Nevertheless, we attempted to counteract this bias by not using the term "advance care planning" but use a broader description such as thinking or talking about the future and future care. A second limitation might be that we do not know how experiences with advance care planning continue or change at the end of life. The average survival rate in ALS is two to three years from symptom onset[4,5], Most persons with ALS are diagnosed 10 to 12 months after their first symptoms, which means they may already developed severe disability or have a short prognosis[6,7]. Therefore, we decided to include persons with ALS in this study when they received their diagnosis no longer than six months ago. Persons with ALS and their family carers were then followed not longer than nine months, in which some participants were still early in their disease trajectory. However, we believe that with the large amount of data that we collected and the different paces in progression in the disease trajectory ALS, we were able to grasp in-depth views of experiences with advance care planning over time[3].

A population-based nationwide cross-sectional survey

We conducted a population-based survey in Flanders, Belgium to investigate the extent to which family carers performed advance care planning conversations with their relative and how they were supported by healthcare professionals to conduct these conversations (**Chapter 3**). The survey was focusing on the conversations at home and were limited to the last three months of life. The survey used to address the objectives was a combination of validated[8] and self-developed items.

A first strength in this study was the sampling framework. This framework was unique because we used complete registers of the three largest health insurances in Flanders, Belgium and we took a random sample from this. The study therefore suffered less from selection bias compared to most studies using convenience samples of family carers, for instance, via professional palliative care or other healthcare service use. Another advantage of using this framework is that statistical generalization to the target population of family carers is improved. Another strength is that the questionnaire was available both on paper and online which seemed beneficial for the response rate (55%), as it was higher compared to other studies with similar populations[9–11].

This study also had some limitations. First, despite the effort of construing a complete sample frame there might still be some selection bias because we only could include registered family carers, hence, family carers who were not registered with the health insurances were excluded from this study. As a result, we have no insights into the experience of family carers who were not registered by one of the health insurances. Second, the term “advance care planning” is not always well-known in the general population and therefore, in this survey, we described advance care planning as conversations about end-of-life care and end-of-life decisions or completion of advanced directives or living wills. Recent recommendations highlight the need to approach advance care planning on a broader level, whereas the focus is more on the persons’ values, wishes and preferences, and the need of advance care planning occurring over time with multiple conversations[12,13]. The design used in this study was a cross-sectional design. These designs make it possible to describe and compare characteristics and outcomes, but cross-sectional data cannot identify cause-effect relations. We tried to minimize recall bias by having a maximum of nine months between identifying a bereaved family carer and survey completion. However, recall bias cannot be entirely excluded[14].

An international cross-sectional survey

In **chapter 4** we used data of the bereavement questionnaire from the ACTION trial to investigate the effect of the ACTION RC advance care planning intervention on family carers' involvement in decision-making in the last 3 months of the patient's life, and psychological distress after 3 months of bereavement.

This study has several strengths. First, the ACTION study is the first and largest multicentre clustered randomized controlled trial on the effects of advance care planning for patients with advanced colorectal or lung cancer, and their family carers in Europe. Second, to study the effect of advance care planning for bereaved family carers, we sent out a bereavement questionnaire using items from the VOICES-SF and two validated scales measuring psychological distress, anxiety and depression symptoms[15].

This study has also some limitations. First, the response rate was modest (on average 41.5%) in each of the 6 countries. Possible causes of non-response were: 1) that the bereaved family carers were not interested in participating in the study anymore; and 2) the questionnaire was sent to the address of the deceased person, which was not always the address of the family carer. Second, a limited number of characteristics of the bereaved family carers were collected in the ACTION study, therefore we do not know whether the family carer who filled out the bereavement questionnaire was the surrogate decision-maker of the patient. Second, we cannot identify cause-effect relations because of the cross-sectional design. It is only possible to describe and compare characteristics and outcomes. Finally, we have no information about the way family carers were involved during the advance care planning conversation, nor how actively the family carer participated into the conversation[15].

The Dying Well with Dementia study and the PACE study

In **Chapter 5**, we used data from two retrospective epidemiological studies: the Dying Well with Dementia study and the PACE study (Palliative Care for Older People in care and nursing homes in Europe) to examine the family carers' evaluation on the quality of end-of-life care for nursing home residents with dementia. Although these were separate studies, both utilised similar study designs, and all variables of interest were measured in the same way.

The strengths of this study were the use of data from large representative nationwide cross-sectional mortality follow-back studies of nursing homes in Flanders, Belgium and the high response rates of the family carers (response rate^{2010} = 51.05%; response rate^{2015} 60.65%)[16,17]. Another important strength of this study was the use of a validated scale (EOLD-SWC) with good psychometric properties to measure the quality of end-of-life care as perceived by the family carers[18,19].

This study also has some limitations. The studies were both cross-sectional study designs. These designs make it possible to describe and compare characteristics and outcomes, but cross-sectional data cannot identify cause-effect relations. Another limitation was that the validated scale EOLD-SWC is a subjective measurement and the total mean score might be biased by the expectations family carers have about the end-of-life care[20]. Due to the retrospective design, the recall bias cannot be excluded. Nonetheless we tried to minimize this by having a timeframe of a maximum of three months between identifying a nursing home resident's death and providing the bereaved family carer with the questionnaire[19].

Discussion of the main findings

Advance care planning is a complex and iterative process

Advance care planning is more than a medicalized process

Our results from **Chapter 2** show that advance care planning is a holistic process in which medical, social and psychological factors are intertwined, and cannot be seen as separate entities. In our longitudinal qualitative study, people with ALS and their family carers thought about the future and future care. Most often, they reflected on what would happen next in terms of how the disease would evolve in the future, rather than thinking about the medical aspects of end-of-life decisions. This was highlighted by other studies in different populations such as dementia, multiple sclerosis and cancer.[12,21,22] In our longitudinal qualitative study, we asked family carers and patients with ALS whether or not they thought about future or future care, and if they did, what they were thinking about, what their worries, hopes, and preferences were. This is in line with the broader concept of advance care planning, in which people with a chronic life-limiting condition become aware of what it means to live well, what is important to them and what it would mean for them if they get sicker[12,23–26]. These conversations about goals and values will eventually help in making the best possible decisions regarding treatment or care that is in line with their core values[12,25–28]. Nonetheless, the dominant focus of advance care planning often remains on medical aspects of end-of-life preferences such as avoiding unwanted treatments that are seen as potentially harmful, and completion of advance directives[29–32]. Studies in the US, Germany and the UK about advance care planning in patients living with ALS have shown that advance care planning in ALS is often delayed until the last months or weeks of life, and therefore recommended that healthcare professionals should initiate conversations about medical aspects of care such as gastrostomy, tracheotomy or medical end-of-life decisions early on in the ALS disease trajectory[7,33–35]. It was apparent that in our study, conversations about these topics were often perceived – most often by family carers– as too early or stressful. Possibly because family carers wished to protect their loved one, but also themselves for these emotionally distressing conversations. This was also found in a qualitative study about advance care planning in persons with dementia and their family carers[36]. One study in which neurologists with a specific clinical interest in ALS were surveyed, showed that 78% of these neurologists believed that discussion and completion of advance directives is useful in ALS. However, only 55% of these neurologists discussed advance directives regularly with persons with ALS, and only 30% of persons with ALS completed such an advance directive[37]. This indicates how emotionally difficult conversations about medical aspects of advance care planning can be[26,38]. These results and our results confirm that it might be better, as suggested by others, to start the process of advance care planning with broader conversations about values, and what it means to live well[25,39].

Transformative and relational complexities in advance care planning

Advance care planning is a complex process which has been highlighted in **chapter 2**. It appears that over time different factors related to coping with the severity of the terminal illness such as ALS and relational dynamics play a role in constructing people's ideas, thoughts, and feelings about future care and treatment preferences. Living with a chronic life-limiting illness has been described as a transformative experience, meaning that one cannot know how they would cope or react until they live in that situation. Therefore planning for the future is difficult and transformative, because choices can change over time due to the unpredictability of the future[40]. However, this does not mean that advance care planning is futile, but rather that it challenges a person to think about what it means to live a fulfilling, valuable life, while simultaneously reflect about what the person really wants or who they really are[40]. Thinking about future and future care decisions is thus complex due to the unpredictability of the future. In our study, the unpredictability of the future often led to a day-by-day attitude in persons with ALS and their family carers, possibly because healthcare professionals indicated that everyone evolves in a different way (**Chapter 2**). People with a chronic illness, try to maintain a sense of control by adjusting to or accepting their new role in life[41,42], but this is often difficult for people living with ALS due to the sometimes rapid physical losses[43–45]. The rapid physical losses in ALS makes it often difficult for person with ALS to learn or re-learn to accept themselves, in which they expressed their wish to remain independent for as long as possible, also because they did not want to be a burden for their family carer. This is also shown in studies on people living with multiple sclerosis[22,46]. Our results show that when persons with ALS were more accepting about their new role in life, they were often more open to think about the future or future care. This was also true when an impactful deterioration in the person with ALS has happened such as not being able to go upstairs/not being able to walk independently. This often led to an immediate reaction in terms of planning such as renovations of a bedroom and bathroom downstairs or applying for a wheelchair. Moreover, this impactful deterioration often led to a reflective process in persons with ALS or their family carers in which discussions about future and future care or treatment was triggered. This has also been found in studies for persons living with multiple sclerosis, suggesting for healthcare professionals that such a turning point might potentially be an optimal timing to initiate conversations about advance care planning[22,46].

An important theme in **chapter 2** regarding coping was the balance between hope to remain stable and worry about what the future might hold. Persons with ALS and their family carers hoped that the person with ALS would stay stable, independent and comfortable over time. Although this hope remained over time, worries became more prominent as the disease progressed. For some family carers, the worries only increased over time, especially when the family carer could not talk about his or her worries with the person with ALS. Studies on ALS have shown that family carers' burden increases over time, because persons with ALS have more care needs over time, and eventually needs permanent care[47–49]. Nonetheless, hope can be helpful to cope or control the emotional distress that arises when living with a chronic life-limiting illness[39,50–55]. Some patients with a chronic life-limiting illness might struggle to talk about the possibility of dying[53,54] and Jacobson and colleagues[39] have described a dual framework which could help healthcare professionals to have discussions about the future with a person who is chronically ill but is reluctant to talk about their future care. It helps patients in the process of living well while also preparing for the possibility of dying, which ultimately leads to conversations about planning for the future and future care[39]. In this way, patients may reflect about their values, and eventually become aware of them in which they might share their preferences with family and healthcare professionals[40].

The mutual protection of patients and family carers towards each other for discussing future and future care, found in our longitudinal qualitative study, also highlights the relational complexities of advance care planning (**Chapter 2**). This has also been found in a 2014 qualitative study in persons with advanced cancer and their family carers[56]. In this study, both patients and family members prioritize each other's wellbeing, and therefore often avoided communication about the disease itself, but also about future and future care[56]. Patients do not want to alarm family carers and the family carers do not want to distress the patient[53,56,57], which is in line with our results in **chapter 2**. Interestingly, our study showed the persons with ALS and family carers also wished to protect themselves, which led to a day-by-day attitude, or positive thinking about the future or even changing the subject in case topics about future or future care would arise. Possibly because they wanted to protect themselves from long-lasting burdensome emotions[36]. However, family carers and patients were still open and willing to have advance care planning conversations, but simultaneously felt uncomfortable or reluctant in doing so, which was also found in studies in other patient populations such as cancer and dementia[36,58–60].

Family carers' involvement in advance care planning and their evaluation of end-of-life care

The importance of involving family carers in advance care planning

In **chapter 4**, we analysed bereavement data from the ACTION trial which was a multicentre cluster-randomized controlled trial carried out in 23 hospitals in six European countries (Belgium, Denmark, Italy, the Netherlands, Slovenia and the United Kingdom). The ACTION trial found positive effects on palliative care use and inclusion of advance directives in medical files, but found no effect on patients' quality of life, symptoms, coping, patient satisfaction or shared decision-making[61]. Also in our study, no significant difference was found between the control group (care as usual) and intervention group (care as usual + advance care planning intervention) in the involvement of family carers in decision-making in the last 3 months of the patient's life. Previous studies have shown that family carers play a pivotal role in end-of-life care, not only by providing physical and emotional or spiritual care to their seriously ill relative, but also by providing information to and communicating with healthcare professionals about the wishes of their seriously ill relative[62–64]. Therefore, family carers' involvement in advance care planning has been seen as crucial as they often make decisions on behalf of their relative at the end of life[65]. The advance care planning intervention in the ACTION trial consisted of a maximum of two advance care planning sessions. This intervention mainly had a patient-centred focus and did not sufficiently address the empowerment and preparedness of the family carer for involvement in decision-making[66]. Most advance care planning interventions often focus solely on identifying a surrogate decision-maker for the patient, but lack to adequately involve and prepare surrogate decision-makers for their role[67–72]. When family carers sufficiently know and understand patients' preferences and the leeway they have in decision-making[27], this could increase the family carers confidence in making end-of-life decisions, as well reducing distress, anxiety or depression during bereavement[38,73,74].

However, in **chapter 4**, no significant differences were found between intervention and control group in family carers' psychological distress three months after bereavement. While different studies on advance care planning have used wellbeing and psychological distress as outcome measures[74–76], there is no univocal conclusion about these outcome measures. Some studies seem to find a significant decrease in wellbeing and stress in family carers, others did not[74]. Previous studies carried out in Australia have shown conflicting findings in regard to advance care planning effects on family members stress, anxiety, and depression. Detering and colleagues[75] for example found that advance care planning had positive outcomes for family carers of older hospitalized patients involved in advance care planning[75]. However, Johnson and colleagues[76] found that family carers of patients with cancer who participated in advance care planning discussions did not show improvements, while their mental wellbeing was negatively affected[76]. They hypothesized that family carers – who were involved in advance care planning conversations – had higher expectations about the care for the patients and probably felt more distressed when patients' preferences were not met during the last months and days of the patient's life[76].

Facilitators for family carers to engage in advance care planning

In **chapter 3** our findings of a population-based survey show that almost 50% of the family carers of people with a serious chronic illness did have an advance care planning conversation with their relative in the last three months of life. The probability of having such conversations increased in case the family carer was 55 years of age or younger. This finding is in line with other studies showing that, in general, younger patients and family carers desire more detailed information regarding the illness, palliative care, and want more involvement and autonomy in decision-making[19,77,78]. As attitudes towards discussing end-of-life issues might differ in younger family carers compared to older generations, younger family carers might be more open and willing to initiate advance care planning conversations with their relative themselves[36,58–60]. Recently more public health campaigns are available to increase normalization regarding advance care planning[79] and more self-guided tools to facilitate advance care planning conversations (e.g. websites, conversation cards) are becoming available[80]. Older adults might experience difficulties to find their way to these tools, websites or information campaigns of advance care planning, so healthcare professionals have a key role in introducing these tools to older adults[38].

The probability of family carers having an advance care planning conversation in the last 3 months also increased in case specialist palliative care services were involved (**Chapter 3**). This result was not surprising as one of the core tasks of specialist palliative care services is to inform patients and family carers about the severity of the illness and to prepare them for the approaching death[81]. It might also be that family carers were more aware about the poor health condition of their loved one because specialist palliative care services were involved. Family carers with a medical degree were also significantly more likely to engage in an advance care planning conversation with their relative in the final three months of life. They might be more aware of the importance of engaging in advance care planning conversations, are more skilled to perform these conversations themselves and find their way more easily to specific professional support and services[82]. Thus, it appears that knowledge about the health condition or receiving information about medical care or palliative care are important prerequisites for family carers to engage in advance care planning conversations[60,82,83] This has also been highlighted in **Chapter 5,** in which our study has shown that when family carers of nursing home residents with dementia were informed about the medical and/or palliative care, these family carers perceived the quality of end-of-life care better compared to family carers who were not informed. Other studies have also shown that informing family carers about dementia, its prognosis and the dying process, is highly important for good quality of end-of-life care as perceived by the family carers[84]. Furthermore, a qualitative study with family carers of deceased nursing home residents with dementia conducted in the UK also showed that when family carers are informed about and involved in the care process, they will not only perceive the quality of end-of-life care more positively but also be better prepared to make decisions on behalf of the resident[85].

Our findings in **chapter 3** also showed that 50% of the family carers did not engage in advance care planning conversation with their relative in the last three months of life. Previous studies have shown that family carers experience difficulties or concerns in raising the topic of advance care planning towards their loved one, such as feeling stressful, uncomfortable or anxious in having these conversations with their loved one[86–88]. This result is also found in chapter 2, in which the family carers preferred that persons with ALS initiated these conversations themselves.

The role of healthcare professionals in supporting family carers in advance care planning

As shown in **chapter 3**, healthcare professionals can play a key role in supporting and facilitating advance care planning conversations by performing the conversations together with the family carer and patient or by referring the family carer to helpful resources or tools (e.g. books or websites) to prepare them for advance care planning conversations. However, in **chapter 4,** the advance care planning conversations were conducted by a trained external facilitator who had no contact with the patient's healthcare professional, and who were also not allowed to add information about the advance care planning conversations to the patients' medical file in the hospital. Instead, the participating patients were encouraged by the facilitator to communicate their preferences themselves to their health care professionals, but only few reported to have done so[89]. Already in 1995, the SUPPORT study, which was a randomized controlled trial conducted in persons with advanced cancer in the US, showed that physicians' awareness of patients' end-of-life care preferences did not improve when advance care planning conversations were conducted by nurses or other external facilitators, who had limited or no contact with other healthcare professionals. The authors suggested a more interdisciplinary collaboration between nurses and physicians, which would have an impact on the quality of care[85]. Another study also using an external facilitator concluded that it might be better if a member of the healthcare team attends these conversations because patients seem to prefer to discuss advance care planning with their treating specialist[70], which our results seem to confirm.

In our study, 78% of family carers who did engage with their loved one in an advance care planning conversation in the last 3 months of life, indicated that they were supported by a healthcare professional in conducting advance care planning conversations and this was most often done by the general practitioner (**Chapter 3**). General practitioners are ideally placed to facilitate these conversations, as they often know the patient and family carer for a considerable length of time, and are often aware of both the medical and the social context[91]. Most of the support by healthcare professionals was given by performing the advance care planning conversations together with the family carers and their relative. Perhaps family carers felt safer to perform these conversations together with a healthcare professional or they might have lacked the necessary tools or skills to perform these conversations themselves.

Interestingly, 22% of the family carers did not receive support from healthcare professionals, of which 78% indicated they did not need support. This suggests that these family carers felt empowered enough to engage in these conversations, without the support from a healthcare professional. However, 22% of these family carers indicated they would have needed the support from a healthcare professional in having advance care planning conversation. It could be that family carers felt hindered to ask healthcare professionals for help or that healthcare professionals were concerned that advance care planning might create anxiety in patients and family carers, and therefore did not initiate an advance care planning conversation.[57,92–94]

Early initiation of advance care planning in neurodegenerative diseases

Our survey also showed that the probability of family carers having an advance care planning conversation in the final three months decreased if their loved one was diagnosed with dementia (**chapter 3**). Probably because the person diagnosed with dementia did not have the mental capacity to participate anymore in these conversations. Nonetheless, it has been suggested in the literature to initiate advance care planning in the earlier stages of the disease trajectory[74], especially in neurodegenerative diseases such as dementia and ALS[7,33,86]. However, previous studies have shown that only a minority of people with dementia get the opportunity to engage in advance care planning themselves. More often, healthcare professionals only involve the family carer, because the fluctuations in cognitive capabilities in dementia are often seen as a barrier to initiate advance care planning with patients themselves[95,96]. It has been recommended to maximize the opportunity for people with dementia to participate in advance care planning as active agents, in order to highlight their capabilities and to have their voice heard[21]. For example, by initiating advance care planning around specific key moments such as changes in health status, or by adjusting the conversation style and content to the person with dementia's cognitive capabilities in the advance care planning conversations.[96]

Our results in **Chapter 2** show that healthcare professionals do initiate conversations early in the disease trajectory of ALS about the possibilities and risks of a gastrostomy, a tracheotomy and euthanasia. This happened most often shortly after diagnosis and/or in follow-up consultations, although other studies have shown that advance care planning in ALS is often delayed until the last months or weeks of life[35]. However, in our study these conversations were often perceived by the family carers and persons with ALS as too early or too stressful. Previous studies in advance care planning in dementia or in multiple sclerosis have shown that it might be better to initiate advance care planning at key trigger points such as diagnosis, but also when an impactful deterioration occurs[22,96], which our results also confirm.

It is important to note that the care for people with ALS and their family carers is often provided at a tertiary level. In Belgium, these are called the neuromuscular reference centres, which merely have an advisory role and have expertise in neurology, respiratory care, rehabilitation, as well as in psychology, physiotherapy, occupational therapy, speech and language pathology, nutrition and social work[97,98]. In Belgium, it is recommended that persons with ALS and their family carers visit a neuromuscular reference center every 2 to 6 months[47]. There are seven neuromuscular reference centres in Belgium and these are always connected to a hospital (mostly university hospitals). To obtain advice from these centres, persons with ALS and their family carers need to be followed by a neurologist from that hospital. Even though people with ALS and their family carers, in our longitudinal qualitative study knew they could contact the healthcare professionals in the neuromuscular reference centres at any given time with questions about care or psychosocial and existential questions, we found that the threshold to contact these centres was often too high which could have hindered the advance care planning conversation (**Chapter 2)**. Studies on ALS have shown that delayed information about care or equipments such as a wheelchair or a speech computer, the limited availability of healthcare professionals and a lack of psychosocial support during advance care planning and end-of-life decisions resulted in a decrease in quality of life in persons with ALS[7,33,99,100]. Moreover, in our study, described in **chapter 2**, participants often felt alone, unsupported and had difficulties in navigating their way throughout the healthcare system. Possibly because primary and palliative care services were often not involved. Previous research in neurodegenerative diseases have shown that a long-lasting, trusting and empathic relationship with a healthcare professional is important and can facilitate advance care planning conversations and primary care professionals ideally placed to have these conversations[46]. In Belgium, persons with ALS and their family carers must contact these primary and palliative care providers themselves, and as shown earlier in **Chapter 3**, these are the healthcare professionals that most often support in advance care planning conversations. A possible approach that could facilitate advance care planning and interdisciplinary collaboration between primary, secondary, and tertiary ALS care is the development of an integrated care model. In Australia, Ireland and the Netherlands such an integrated collaborative care model in ALS has been developed in which persons with ALS and family carers are supported by all healthcare professionals relevant in the care[101–105]. These models have shown an increase in quality of life for persons with ALS, and a decrease in family carers burden[101–105].

Information and communication as important prerequisites for good end-of-life care

Our results in **Chapter 5,** show that family carers are relatively satisfied with many aspects of the quality of end-of-life care that their relative with dementia had received in the nursing home. Moreover, family carers who received information on palliative or on medical care tend to evaluate the quality of care better than those who had not received this information. This finding could be interpreted in multiple ways. Providing information on palliative and medical care might be an important prerequisite of good quality end-of-life care as assessed by family carers. However, it might also be that the staff in nursing homes where good quality of end-of-life care is provided automatically provide more information on palliative and medical care to the family carers. This relationship between the provision of information and evaluation of care quality corresponds with findings from previous studies about the importance for family carers of being informed, not only about medical care, but also about the end of life[82,83]. Information provision is the first important step in advance care planning, because when patients and family carers are informed about the illness or prognosis, it helps them to make informed decisions[106]. Engaging in advance care planning has also shown improvement in the quality of end-of-life care in older adults[107]. Thus, advance care planning is an important prerequisite for good quality of end-of-life care.

Recommendations for research, practice, and policy

Based on the studies in this thesis, important recommendations can be made for future research, practice and policy makers regarding experiences with advance care planning and family carers' involvement in advance care planning.

Recommendations for research

Using research designs that highlight and understand complexities in advance care planning

Our results, described in **Chapter 2**, highlight that advance care planning is a complex process in which interrelated factors such as coping with the severity of the illness (e.g. ALS) or relational dynamics play a role in constructing people's thoughts, ideas, and feelings about future care and treatment preferences. These factors could be identified because we used a research design that supports measuring complexity (**Chapter 1**), instead of using a simplistic, uni-perspective and cross-sectional design, which is commonly used to assess advance care planning effects. We believe that advance care planning needs research approaches which are complexity-informed, focus on relationships and temporal dynamics, and in which causality is viewed as emergent instead of linear. For example, the qualitative longitudinal research design we used to address the first aim of this dissertation (**Chapter 1 and 2**). Besides longitudinal designs, other research approaches such as realist research approaches might also be valuable in understanding complexity. These approaches are theory-driven and investigate why an intervention might work, for whom, in what circumstances and how, in order to identify underlying processes that lead to desired outcomes in a specific context[46,108]. These research approaches are currently underexplored in the field of advance care planning.

Using a consistent way of measuring wellbeing and psychological distress in family carers

Many advance care planning studies have used wellbeing and psychological distress as outcome measures. Our results in **chapter 4** did not show a significant difference between control and intervention group in psychological distress in family carers three months after bereavement. However, there is no univocal conclusion possible, based on these outcome measures. Some studies seem to find a significant increase or decrease in wellbeing and stress in family carers, others did not find significant effects[74–76]. Future research should focus on a consistent way of measuring wellbeing and psychological distress of family carers after advance care planning. More specifically, qualitative research could enhance our understanding of the wellbeing and psychological distress of the family carer when engaging in advance care planning, as well as identify other relevant outcomes for patients and family carers. Additionally, outcome measures such as wellbeing and psychological distress are influenced by several different factors not only related to advance care planning. We suggest using outcomes that are more closely related to the conversation itself. For example Brazil and colleagues used the "decisional conflict scale" to investigate whether advance care planning reduces family carers uncertainty in decision-making in nursing home residents[109]. Thus, "uncertainty in decision-making" might be a better way to evaluate the impact on advance care planning conversations, than an overall wellbeing scale.

More insight is needed into specific support needs of family carers for conducting advance care planning conversations.

Our results in **chapter 3** show that healthcare professionals can support family carers in advance care planning conversations most often by performing the conversations together with the family carer and patient. This can be considered as an 'empowering collaborative approach' versus providing information or explaining how to conduct these conversations which were less frequently reported. More insight is needed into the specific support needs of family carers for conducting these advance care planning conversations with their relative during the disease trajectory, or on how family carers wish to be supported by healthcare professionals in engaging in advance care planning conversations. A possibility to increase empowerment and self-efficacy in family carers might be via different self-guided tools such as websites or conversations cards, but also via psycho-education programs.[80,110,111]

Development of an integrated care model is necessary in people with ALS

The results described in **Chapter 2,** showed that the threshold for persons with ALS and their family carers to contact the neuromuscular reference centres at any given time with questions about care or psychosocial and existential questions was too high. Often, they awaited their appointment which was still months away. This might have hindered advance care planning conversations. Moreover, in our study described in **Chapter 2,** the persons with ALS and their family carers often felt alone, unsupported and had difficulties in navigating their way throughout the healthcare system. Possibly because primary and palliative care professionals were often not involved. As shown in **chapter 3**, these healthcare professionals are most often the ones who give support in advance care planning conversations. A possible approach that could facilitate advance care planning and interdisciplinary collaboration between primary, secondary, and tertiary ALS care is the development of an integrated care model. In Australia, Ireland and the Netherlands, an integrated care model is available in ALS, and this integrated, collaborative approach has shown promising results such as an increase quality of life in persons with ALS, and a decrease family carers burden[101–105]. Developing such an integrated model will support a coordinated approach, in which all relevant healthcare professionals on primary, secondary and tertiary level are involved early and simultaneously in the disease trajectory of ALS. Additionally, this model will provide the best quality of care, including timely advance care planning conversations for people with ALS and their family carers. The first important step in developing such model is identify the core components in these existing care models and to develop a theory-based model in which all the possible end-users in this model are involved (e.g persons with ALS, their family carers and healthcare professionals on all care levels).

Recommendations for practice

Advance care planning should focus on "living well" or "what matters most" now and in the future

Our results in **chapter 2**, highlighted that psychosocial factors such as coping mechanism or relational dynamics are inextricably connected with advance care planning and cannot be simplified to conversations about medical end-of-life decisions. Our results also showed that some people with ALS and their family carers experience advance care planning conversations as too stressful or too early, and thus had difficulties to talk about it among each other or with healthcare professionals. In our qualitative longitudinal study, we asked whether they have thought about the future or future care, and if so, what they were thinking about (in terms of hopes and worries about the future). This broader approach helped people with ALS to become aware of what it means to live well and what is important to them. Therefore, it is recommended for healthcare professionals to start conversations on "living well" or "what matters most" now and in the future. Talking about "living well" or "what matters most" will help patients to clarify their values and goals in the process of living well while also preparing for the possibility of dying and is thus an ideal starting point for advance care planning. When patients know their goals and values, they will make medical care decisions that are aligned with their core values[39]. We recommend that healthcare professionals consistently offer these conversations towards patients and family carers on their own pace and multiple times throughout the disease trajectory. However, healthcare professionals may struggle to have these conversations, because they often lack the necessary communication skills to discuss goals and values with patients and family carers. Therefore, we recommend that healthcare professionals are trained or participate in education programs in which their communication skills are enhanced to engage in advance care planning conversations.

Healthcare professionals can help empowering patients and family carers, and promoting self-efficacy of them to engage in advance care planning

Our results in **Chapter 3,** have shown that almost 50% of the family carers engage in advance care planning conversations and that 78% of these family carers have highlighted that they were supported by healthcare professionals in having these conversations. Most often, the support given was more of collaborative approach versus the approach in which family carers were empowered in having these conversations all by themselves. We recommend that healthcare professionals can help enhancing patients' and family carers' self-efficacy to have advance care planning conversations without their involvement. For example, they might refer patients' and family carers to tools such as books, websites, leaflets, or conversation cards to prepare them to have advance care planning conversations or to continue these conversations at home[112,113]. As our results in **Chapter 2** have shown that family carers and persons with ALS have difficulties with talking to each other about the future and future care partly because of the mutual protection between persons with ALS and their family carers. Possibly, such tools might have helped them in these conversations.

Advance care planning conversations should be facilitated preferably by a healthcare professional who is part of the healthcare team

Our results show that different healthcare professionals can play a role in facilitating advance care planning conversations, by performing the conversations together with the family carer and patient, or by referring the family carer to helpful resources or tools to prepare them for advance care planning conversations (**Chapter 3)**. However, our results also suggests that these conversations should be conducted by a healthcare professional who is part of the healthcare team of the patient and thus more closely involved in the care of the patient. As shown in **chapter 4**, the advance care planning conversations were conducted by a trained external facilitator who had no contact with the patient's healthcare professional. These external facilitators needed to encourage the patients to communicate their preferences themselves to their healthcare professionals, but only a few reported to have done so[89]. The SUPPORT[90] trial and other studies[70] have shown that physicians' awareness of the patient's end-of-life care preferences did not increase when advance care planning conversations were conducted by nurses or other external facilitators, who had limited or no contact with other healthcare professionals. These studies have shown that patients seem to prefer to discuss advance care planning with their treating specialist[70]. In the SUPPORT study, it was suggested that a more interdisciplinary collaboration is needed between nurses and physicians, which would have an impact on the quality of care[90].

Recommendations for policy

Improving advance care planning takes a whole-system approach

The results of this dissertation might encourage policymakers to improve advance care planning practices, by taking a whole system approach. This means that advance care planning should be addressed and coordinated at multiple levels (individual, system or healthcare service level). On an individual level, patients and their family carers are provided with the opportunity to have timely conversations, with or without the support of healthcare professionals. This also means that policymakers are encouraged to advise, support and facilitate training for healthcare professionals to implement advance care planning in routine care, meaning that advance care planning is consistently offered towards patients and their family carers on their own pace and several times throughout the disease trajectory[114]. Policy can also advocate to raise awareness of the importance of empowerment and self-efficacy in patients and family carers engaging in advance care planning conversations. It is important to focus on all people involved: healthcare professionals, patients, family carers and the broader community. Our results have also highlighted that medical, social and psychological factors are interwoven into the process of advance care planning and cannot be seen as separate entities. The development and promotion of public awareness campaigns to inform the community about the broader definition of advance care planning, which indicates that people think about what it means to live well or what matters most.

Implementation of reimbursement for advance care planning should be monitored by policymakers and adjusted if necessary

In 2022, a reimbursement rule for time devoted to advance care planning conversations for general practitioners was approved by the Belgian federal government[115]. This law allows general practitioners to receive a fee in case they have an advance care planning conversation with a patient with a chronic life-limiting illness[115]. In the US, such a reimbursement system for advance care planning conversations in healthcare professionals was implemented in 2016, with the intention to increase the uptake of advance care planning. However, a study has shown that there was limited use of advance care planning services, indicating that this reimbursement rule has not affected actual practice[116]. Nonetheless, when analysing the Belgian policy document about this reimbursement closely, the reimbursement can only be requested once in every patient by the general practitioner. This might encourage the general practitioner to only have one conversation, instead of having multiple conversations over time throughout the disease trajectory. Our results show that advance care planning is a complex and evolving process, in which multiple conversations take place over time. Our results also show that psychosocial factors such as coping or relational dynamics are inextricably connected with the advance care planning process. Thus, advance care planning cannot be simplified to conversations about medical end-of-life decisions or completions of advance directives (**Chapter 2**). Therefore, we recommend that policymakers thoroughly monitor or evaluate the implementation of this reimbursement law regarding advance care planning and adjust it where necessary. Next, it seems that this reimbursement solely focuses on the general practitioners. Our results did show that general practitioners mostly engage in advance care planning conversations together with patients and family carers (**Chapter 3**).

Nonetheless, some patients also wish to have these conversations with their specialist, or other healthcare professionals[76]. Thus, we recommended that this reimbursement should also be available for other healthcare professionals such as specialists, nurses, or psychologists, as they may also be confronted with individuals who want to discuss their future and future care or treatment.

Conclusion

The results of this dissertation have highlighted that advance care planning is a complex communication process in which medical, social and psychological factors are interwoven and cannot be seen as separate entities. Our results also show that interrelated factors such as coping with the severity of the disease or relational dynamics shape the experiences of the advance care planning communication process in the context of a terminal illness such as ALS. However, more attention to these complexities in advance care planning is needed in future research, practice and policy. This dissertation also indicated that about half of the family carers engage in advance care planning conversations. Most of the family carers who had done so, received support from a healthcare professional, in which it is important that the healthcare professional is part of the healthcare team such as the general practitioner. The type of support received was most often by performing the conversation together with the family carer and the patient, which can be seen as collaborative empowering approach. However, more insights are needed on how to support family carers in conducting these conversations, both with and without involvement of healthcare professionals. Our results did not show a significant difference between control and intervention group in psychological distress in family carers three months after bereavement. Many advance care planning studies have used wellbeing and distress, but there is no univocal conclusion about these outcome measures. Family carers who received information on palliative or on medical care tend to evaluate the quality of care better than those who had not received this information. Information provision is also seen as one of the first important steps in advance care planning. Hence, advance care planning is an important prerequisite for good quality of end-of-life care.

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English summary

Introduction

People live longer primarily because of improved public health, medical knowledge, and technology. This resulted in a growing population of older people and a fundamental change in the main cause of death, i.e. due to chronic life-limiting illnesses. Chronic-life-limiting illnesses are characterized by prolonged disease trajectories, with a functional decline over months or years and are responsible for more than 73% of the global deaths. People with a chronic life-limiting illnesses, such as amyotrophic lateral sclerosis (ALS), dementia and cancer, develop a range of complex needs and symptoms for which early palliative care is highly recommended. Yet, palliative care is often initiated too late or not at all.

People with ALS and their palliative care needs

ALS is incurable and characterized by progressive muscle paralysis involving all voluntary muscles, resulting in difficulties with swallowing, speaking, breathing, and limb paralysis. Respiratory failure is the most common cause of death. Persons with ALS often experience physical, emotional, and existential problems that persist until the end of life. However, to date, reports show that the complex needs of people with ALS often remain unmet. Given the incurable nature of ALS, combined with its rapid progression and unmet palliative care needs an integrated palliative care approach including advance care planning, has been widely advocated for this population. Multidisciplinary care is the recommended approach for the clinical management of ALS, as research has shown that it improves quality of life, decrease burden in family carers and leads to a reduction in the number of hospital admissions and a shortening of hospital stays for patients. Many persons with ALS desire to stay at home for as long as possible, even in the terminal stages of the disease, but only 40% dies at home.

People with dementia and their palliative care needs

People with dementia have difficulties in performing activities of daily living and they can also experience behavioural and psychological changes such as apathy, depression, aggression, anxiety or trouble sleeping. Thus, they experience multiple physical, psychosocial and spiritual care needs that persist for month or years until death. However, these care needs often remain unmet. Although most people with dementia wish to stay and die at home, most people with dementia often die in a nursing home. Providing good quality end-of-life care for people with dementia is thus an important aspect of nursing home care. However, providing high quality end-of-life care in dementia is challenging, because dementia is often not seen as terminal condition, which may lead to poor symptom management or hospital transfers at the end of life.

People with cancer and their palliative care needs

Despite progress in diagnosis and treatment, cancer remains a major life-limiting disease, with 18.1 million new cases and 9.6 million deaths worldwide in 2018. People with cancer generally receive oncology care, which aims to cure the cancer, improve symptom burden, reduce medical complications related to cancer or prolong life, while palliative care in cancer focuses on relieving symptoms. When cure for people with cancer is no longer a realistic option, care needs to be realigned to address their needs and preferences concerning symptom control, psychological support, social, existential and palliative care needs. However, these needs often remain unmet, especially the emotional support was reported as the most commonly unmet need in persons with cancer and their family carers. Thus, timely and effective communication about advance care planning is widely advocated for this patient population and its families.

Advance care planning as part of good quality end-of-life care

Advance care planning is defined as '*a continuous, early-initiated communication process between patients, their family carers and/or health care practitioners that supports adults at any age or stage of health in understanding and sharing their personal values, life goals and preferences regarding future medical care, including end-of-life care*'. The goal of advance care planning is to provide a timely exploration of a person's underlying values and preferences for future care in order to develop a shared understanding to inform patient-centred care, which becomes particularly important if patients later become cognitively and communicatively incapable to make their own decisions about care. Recent recommendations highlight the need to see advance care planning to be seen as a series of broader conversations about hopes, preferences, and potential care goals. Moreover, preferences for current and future care are situational – often related to key events such as symptom progression or multiple hospital admission – and may change over time. However, most studies investigate advance care planning at one specific point in time, which only gives a snapshot of the complex reality when engaging in the process of advance care planning. Studies to capture the process of advance care planning over time – such as longitudinal designs - are scarce.

Especially, in a fast-changing disease trajectory such as ALS, it is unclear when and how advance care planning occurs and changes over time. It is recommended that advance care planning is not delayed in persons with ALS. Most studies on advance care planning in ALS focus on the risks and benefits of life-sustaining interventions (such as invasive or non-invasive ventilatory support or gastrostomy), identification of a surrogate decision-maker and completing an advance directive. However, studies focusing on the the communication process between the person with ALS, his/her family carer and/or health care professionals in which broader personal values, wishes and preferences for future care are explored, are currently lacking.

<u>Family carers involvement in advance care planning and their evaluation of end-of-life care</u>

Family carers play an important role in end-of-life care, not only by providing physical and emotional/spiritual care to their seriously ill relative, but also by providing information and communication to health care professionals about the wishes of their seriously ill relative. Family carers contribution in caring for their relative is often more substantial than for health care professionals, which means that family carers have a lot of opportunities to discuss advance care planning with their loved outside the clinical context. Family carers are willing and welcoming to have advance care planning conversations, but they experience some barriers in performing these conversations such as anxiety, or feelings of being unprepared or uncomfortable. Thus, healthcare professionals can play an important role in empowering the family carer to perform an advance care planning conversation. However, evidence on the proportion of family carers who performed an advance care planning conversation and received support from a healthcare professional is currently lacking. Additionally, involvement of family carers in advance care planning should reduce distress in making end-of-life decisions on behalf of their loved one. Yet, it is hitherto unknown if an advance care planning intervention would increase the involvement of family carers in decision-making and reduce the family carers distress.

In case a person is admitted to a nursing home, such as persons with dementia, family carers often remain closely involved in the care of the person with dementia. Family carers can provide key information about the resident to the healthcare professional, most often because nursing home residents with dementia are usually less able to communicate their needs and symptoms or to provide information of symptom improvement following treatment near the end of life. Therefore, the perspective of family carers about the quality of end-of-life care is of value. However, only a few studies have so far evaluated the quality of end-of-life care from the perspective of family carers of residents with dementia.

Study objectives

The purpose of this dissertation is twofold. First, we want to provide in-depth insights in a complex and evolving process such as advance care planning in a disease trajectory of ALS (<u>Part 1 of this dissertation</u>). To reach <u>research aim 1,</u> we first outlined the study protocol of the qualitative longitudinal multi-perspective interview study (**chapter 1**). In **Chapter 2,**_we report on the experiences of people with ALS and their family carers with advance care planning, and if, how and why these experiences change over time. Second, we also want to give an insight into the family carers involvement in advance care planning and their evaluation of end-of-life care (Research aim 2 and <u>Part 2 of this dissertation</u>), by investigating how these family carers were supported or empowered by healthcare professionals (**chapter 3**), to know if family carers were involved in decision-making at the end-of-life, and to explore the level of distress these family carers are experiencing after death of the person with advanced cancer.(**chapter 4**), and to examine the family carers' evaluation of quality of end-of-life care in nursing home residents with dementia (**Chapter 5**).

Methods

We used different study designs and methods to realise our two research aims. To provide in-depth insights into a complex and evolving process of advance care planning throughout the disease trajectory of ALS, we used a qualitative longitudinal research design, whereas we interviewed persons with ALS and their family carers individually three times over a maximum period of nine months (**Chapters 1-2**). To investigate how family carers are supported or empowered by health care providers during advance care planning discussions, we conducted a population-based survey of bereaved family carers sampled from the three largest healthcare insurances in Flanders, Belgium (**Chapter 3**). We used secondary data of the ACTION trial, which was a clustered randomized controlled trial testing the efficacy of the ACTION Respecting Choices (RC) advance care planning intervention in persons with advanced lung or colorectal cancer in six European countries (Belgium, the Netherlands, Italy, Denmark, Slovenia and the United Kingdom). This secondary data was used to assess the ACTION Respecting Choices (RC) advance care planning intervention on family carers' involvement in decision-making at the end-of-life and the level of distress these family carers are experiencing after the death of persons with advanced cancer. (**Chapter 4**). Lastly, to examine the quality of end-of-life care as perceived by family carers of nursing home residents with dementia, we used two retrospective epidemiological studies with comparable research methods from 2010 and 2015 (**Chapter 5**).

Main findings

PART I: In-depth experiences with advance care planning in people with ALS via patient's and family carer's perspectives.

We developed a study protocol for a qualitative longitudinal multi-perspective interview study to understand in-depth experiences with advance care planning in amyotrophic lateral sclerosis via patient's and family carers' perspectives in **chapter 1**. Persons with ALS and their family carers were approached by their treating neurologist to ascertain willingness to participate in the study and were only contacted by a member of the research team if they gave consent to their neurologist to pass their contact information. Eligible persons with ALS were diagnosed not longer than six months ago, were older than 18 years of age, could speak Dutch, were able to sign an informed consent prior to the first interview and were not diagnosed with frontotemporal dementia. Eligible family carers of persons with ALS were older than 18 years of age, were able to speak Dutch and could sign an informed consent prior to the first interview.

In **chapter 2**, we described the results of the qualitative longitudinal multi-perspective interview study in which we tried to understand the experiences with advance care planning for people with ALS and their family carers, and when and how these experiences unfold over time throughout the disease trajectory. Over a timespan of about nine months, we were able to interview nine dyads of people with ALS and their family carer (mostly) individually about advance care planning, three times each, resulting in a total of 44 interviews. Overall, all persons with ALS and their family carers thought about their future and future (end-of-life) care to some extent over time, but not all dyads talked about it amongst each other, or with a healthcare professional. Over time, advance care planning experiences were influenced by several intertwined determining factors: (1) the experienced physical decline such as no longer being able to walk or talk and related future care needs that could change over time; (2) how persons with ALS identify themselves as patients; (3) obtaining information about diagnosis and prognosis, and learning that everyone evolves differently which makes planning for the future difficult; (4) professionals initiating conversations about medical aspects of end-of-life decisions, even though participants experienced it as stressful or too early; (5) balancing between hope to remain stable and worry about what the future might hold; (6) protecting themselves and each other from what the future might hold.

PART II: Family carers involvement in advance care planning and their evaluation of end-of-life care.

We examined the extent to which family carers of people with a serious illness performed advance care planning conversations with their relative at home in the last three months of life and how they were supported by healthcare professionals to conduct these conversations in **chapter 3.** We conducted a population-based survey study. In this survey study, we included bereaved family carers who had provided care to someone who recently died (between two and six months) via the registers of the three largest health insurers in Flanders, Belgium. Our findings show that about half of the family carers indicated to have engaged in advance care planning conversations with their relative in the final three months of life. These advance care planning conversations were more likely to have taken place when family carers were 55 years of age or younger, had a medical degree (doctor or nurse), or when specialist palliative care services were involved, but it was less likely when the deceased person was diagnosed with dementia. Seventy-eight percent of the family carers received support from a healthcare professional, which was mostly the general practitioner. The type of support received was most often the healthcare professional doing the conversation together with the family carer or seriously ill person, which can be considered as an ‘empowering collaborative approach’. Seldomly, some of the more empowering support types to increase self-efficacy in family carers were reported in this study such as providing information or explaining how to conduct these conversations.

In **chapter 4** we reported on the secondary outcomes of the ACTION trial regarding the effect of the ACTION Respecting Choices (RC) advance care planning intervention on family carers’ involvement in decision-making in the last three months of the patients with advanced cancers’ life, and psychological distress after three months of bereavement. The ACTION trial was a multicentre cluster-randomised controlled trial in six European countries (Belgium, Denmark, Italy, the Netherlands, Slovenia and the UK) which adapted and tested the efficacy of the ACTION RC advance care planning intervention in patients with advanced lung or colorectal cancer. The intervention group received ACTION RC advance care planning intervention and care as usual, whereas the control group received care as usual. Persons with advanced cancer who were part of the intervention group were invited to have an advance care planning session together with their family carer and with a trained facilitator. The patients were encouraged to share their preferences with family members and loved ones, and if they wished, to document these preferences. For this study, data of the bereavement questionnaire was used. We found that bereaved family carers of patients who participated in ACTION RC advance care planning intervention did not have a significantly higher probability to be involved in decision-making in the last three months of the patient’s life, but it was slightly higher in the intervention group (89.6%) than in the control group (86.7%). The intervention group had a slightly higher level of psychological distress three months after bereavement than the control group, but this difference was also not statistically significant.

In **chapter 5,** we examined the family carers’ evaluation of quality of end-of-life care for nursing home residents with dementia and which factors might be associated with this evaluation of the quality of end-of-life care. We found that the family carers’ evaluation of the quality of end-life care for nursing home residents with dementia was relatively high. The evaluation was higher when family carers indicated they had received information on palliative care, and when family carers indicated they had received information on medical care from a care provider.

Discussion of the main findings

PART I: Experiences with advance care planning in people with ALS via patients’ and family carers’ perspectives.

<u>Advance care planning is more than a medicalized process</u>

Our results from **Chapter 2** show that advance care planning is a holistic process in which medical, social and psychological factors are intertwined and cannot be seen as separate entities. In our longitudinal qualitative study, people with ALS and their family carers were asked to think about the future and future care. They mainly reflected on what would happen next in terms of how the disease would evolve in the future, rather than thinking about the medical aspects (e.g., gastrostomy, tracheotomy or medical end-of-life decisions) of end-of-life decisions. This is in line with the broader concept of advance care planning, in which people with a chronic life-limiting condition become aware of what it means to live well, what is important to them and what it would mean for them if they get sicker. Advance care planning conversations about goals and values can help people with ALS in making the best possible decisions regarding treatment or care that is in line with their core values. Nonetheless, advance care planning research and practice is predominantly focussed on the medical aspects of end-of-life preferences such as avoiding unwanted treatments that are seen as potentially harmful, and completion of advance directives. It has been recommended in the literature to discuss the medical aspects of care early in the ALS disease trajectory. In our study, it was apparent that these conversations were often perceived – most often by family carers– as too early or stressful. These results confirm that it might be better, as suggested by other literature in the domain of multiple sclerosis, dementia and cancer as well, to start the process of advance care planning with conversations about values, and what it means to live well, instead of starting the conversation about medical aspects of end-of-life care.

Transformative and relational complexities in advance care planning

In **Chapter 2**, our results suggest that over time different factors are related to coping with the severity of a terminal illness such as ALS and that relational dynamics play an important role in constructing people's ideas, thoughts, and feelings about future care and treatment preferences. Living with a chronic life-limiting illness has been described as a transformative experience, meaning that it is difficult for one to imagine how they would cope or react in the future until they live in that situation. Therefore, planning is difficult and transformative because choices can change over time due to the unpredictability of how the disease progresses. An important theme in **Chapter 2** regarding coping was the balance between hope to remain stable and worry about what the future might hold. Hope can be helpful to cope or control the emotional distress that arises when living with a chronic life-limiting illness. However, the physical decline triggered all participants to think about what the future might hold. Not all persons with ALS and family carers were prepared to talk about advance care planning among each other, most often because they wanted to protect one another and themselves in case of emotional distress during these conversations. Studies have shown that healthcare professionals can play an important role in supporting and facilitating these conversations between family carers and patients.

PART II: Family carers' involvement in advance care planning and their evaluation of end-of-life care

The importance of involving family carers in advance care planning

The results described in **Chapter 4** indicated that family carers of patients with cancer, who received the advance care planning intervention were not more involved in decision-making than family carers who were not involved in this intervention. Previous studies in literature have shown that involvement of family carers in advance care planning conversations is crucial as they often make decisions on behalf of their relative at the end of life. The advance care planning intervention in the ACTION trial consisted of a maximum of two advance care planning sessions and mainly had a patient-centred focus and did not sufficiently address the empowerment and preparedness of the family carer for involvement in decision-making. Most advance care planning interventions often focus solely on identifying a surrogate decision-maker for the patient, but lack to adequately involve and prepare surrogate decision-makers for their role. When family carers know and understand patients' preferences and the leeway they have in decision-making, this could increase the family carers confidence in making end-of-life decisions, as well reducing distress, anxiety or depression during bereavement. However, our study described in **Chapter 4** did not find a difference in psychological distress between family carers who were involved in an advance care planning intervention, and family carers who were not involved. Many studies on advance care planning have used wellbeing and psychological distress as outcome measures, however, there is no univocal conclusion about these outcome measures.

Facilitators for family carers to engage in advance care planning

The results described in **Chapter 3**, highlight that about half of the family carers engage in advance care planning conversations. Especially when family carers were 55 years of age or younger, suggesting that younger generations desire more detailed information regarding the illness, palliative care, and want more involvement and autonomy in decision-making. We also found that involvement of specialist palliative care services increased family carers' engagement in advance care planning conversations. This result is not surprising as one of the core tasks of specialist palliative care services is to inform patients and family carers about the severity of the illness and to prepare them for the approaching death. This information might make family carers become more aware of the poor health condition of their loved one. Family carers with a medical degree also seem to engage more in advance care planning conversations than family carer who do not have a medical degree. Presumably because they have more knowledge about advance care planning and perceive these conversations as more important. Thus, it appears that knowledge about the health condition of the loved one or about medical care or palliative care are important prerequisites for family carers to engage in advance care planning conversations.

The role of healthcare professionals in supporting and collaborating with family carers in advance care planning

As shown in **Chapter 3**, healthcare professionals can play a key role in supporting and facilitating advance care planning conversations, mostly by performing these conversations together with the family carer and patient. Another possibility, which was less indicated by family carers was healthcare professionals who referred the family carer to helpful resources or tools such as books or websites for starting advance care planning conversations. Perhaps family carers felt safer to perform these conversations together with a healthcare professional or they might have lacked the necessary tools or skills to perform these conversations themselves. Most often the family carers were supported by the general practitioner, which is not surprising as they know the patient and family carer for a long period of time and are often aware of both the medical and social context. Our results described in **Chapter 4** also suggest that it might be better if a member of the healthcare team attends the advance care planning conversations instead of an external facilitator. Previous studies have shown that external facilitators who are not part of the healthcare team do not ensure improved quality of end-of-life care among patients and family carers, which our results seem to confirm.

Early initiation of advance care planning in neurodegenerative diseases

Based on our study findings in **Chapter 3**, it seems important that advance care planning conversations are initiated early in the disease trajectory, especially in neurodegenerative diseases such as dementia and ALS. However, previous studies in the literature have shown that only a minority of people with dementia get the opportunity to engage in advance care planning themselves and that the healthcare professional only involves the family carer in it. It has been recommended to maximize the opportunity for people with dementia to actively participate to advance care planning, in order to highlight their capabilities and to have their voice heard. Our results in **Chapter 2** show that healthcare professionals do initiate conversations early in the disease trajectory of ALS about the possibilities and risks of a gastrostomy, a tracheotomy and euthanasia. This happened most often shortly after diagnosis and/or in follow-up consultations, although other studies have shown that advance care planning in ALS is often delayed until the last months or weeks of life. However, in our study these conversations were often perceived by the family carers and persons with ALS as too early or too stressful. Previous studies in advance care planning in dementia or in multiple sclerosis have shown that it might be better to initiate advance care planning at key trigger points such as diagnosis, but also when an impactful deterioration occurs, which our results also seem to confirm.

Mostly care for people with ALS and their family carers is provided at a tertiary level. In Belgium, these are called the neuromuscular reference centres, which merely have an advisory role. Even though people with ALS and their family carers, in our longitudinal qualitative study knew they could contact the healthcare professionals in the neuromuscular reference centres at any given time with questions about care or psychosocial and existential questions, we found that the threshold to contact these centres was often too high which could have hindered the advance care planning conversation. However, delayed information about the care or equipments (e.g. a wheelchair or a speech computer), or limited availability of healthcare professionals or a lack of psychosocial support during advance care planning and end-of-life decisions may decrease quality of life in persons with ALS and their family carers. Moreover, in our study, described in **chapter 2**, participants often felt alone, unsupported and had difficulties in navigating their way throughout the healthcare system. Possibly because primary and palliative care services were often not involved. Previous research in neurodegenerative diseases have shown that a long-lasting, trusting and empathic relationship with a healthcare professional is important and can facilitate advance care planning conversations and primary care professionals ideally placed to have these conversations. In Belgium, persons with ALS and their family carers must contact these primary and palliative care providers themselves, and as shown earlier in **Chapter 3,** these are the healthcare professionals that most often support and facilitate advance care planning conversations. A possible approach that could facilitate advance care planning and interdisciplinary collaboration between primary, secondary, and tertiary ALS care is the development of an integrated care model, which has shown promising results regarding quality of life in persons with ALS and decrease in burden in family carers in other countries such as Australia, Ireland and the Netherlands.

Information and communication as important prerequisites for good end-of-life care

Our results in **Chapter 5**, highlight that family carers are satisfied with many aspects regarding the quality of end-of-life care that was provided to the nursing home resident with dementia. Family carers who received information on palliative or on medical care tend to evaluate the quality of care better than those who had not. This finding could be interpreted in multiple ways. Providing information on palliative and medical care might be an important prerequisite of good quality end-of-life care as assessed by family carers. However, it might also be that the staff in nursing homes where good quality of end-of-life care is provided automatically provide more information on palliative and medical care to the family carers. This relationship between the provision of information and evaluation of care quality corresponds with findings from previous studies about the importance for family carers of being informed. Information provision is the first important step in advance care planning, because when patients and family carers are informed about the illness or prognosis, it helps them to make informed decisions. Engaging in advance care planning has also shown improvement in the quality of end-of-life care in older adults. Thus, this might suggest that advance care planning is an important prerequisite for good quality of end-of-life care.

Recommendations for research, practice, and policy

Recommendations for research

- Using research designs that highlight and understand complexities in advance care planning such as longitudinal qualitative studies

- Using a consistent way of measuring wellbeing and psychological distress in advance care planning

- More insight is needed into specific support needs of family carers for conducting advance care planning conversations

- Development of an integrated care model is necessary in people with ALS

Recommendations for practice

- Advance care planning should focus on “living well” or “what matters most” now and in the future

- Healthcare professionals can help empowering patients and family carers, and promoting self-efficacy of them to engage in advance care planning

- Advance care planning conversations should be facilitated preferably by a healthcare professional who is part of the healthcare team

Recommendations for policy

- Improving advance care planning takes a whole-system approach

- Implementation of reimbursement for advance care planning should be monitored by policymakers and adjusted if necessary

Nederlandstalige samenvatting

Inleiding

Mensen leven langer door de verbeterde volksgezondheid, medische kennis en technologie. Dit leidt tot een groeiende populatie ouderen en fundamentele verandering in de sterfte door chronische levensbeperkende ziekten. Een chronische levensbeperkende ziekte wordt gekenmerkt door een langer ziektetraject, met een functionele achteruitgang gedurende maanden of jaren, en is verantwoordelijk voor meer dan 73% van het wereldwijde sterftecijfer. Mensen met een chronische levensbeperkende ziekte, zoals amyotrofische laterale sclerose (ALS), dementia en kanker ontwikkelen een reeks aan complexe behoeften en symptomen waarvoor palliatieve zorg nodig en wenselijk is. Vroegtijdige palliatieve zorg wordt aanbevolen, maar desondanks wordt palliatieve zorg vaak niet of te laat opgestart.

Personen met ALS en hun palliatieve zorgnoden

ALS is ongeneeslijke ziekte en wordt gekenmerkt door progressieve spierverlamming van alle vrijwillige spieren, met als gevolg moeilijkheden met slikken, spreken, ademen en verlamming van ledematen. Ademhalingsproblemen is de meest voorkomende doodsoorzaak bij ALS. Mensen met ALS ervaren vaak fysieke, emotionele en existentiële problemen die tot aan het levenseinde blijven bestaan. Uit rapporten blijkt echter dat de complexe behoeften van mensen met ALS vaak onvervuld blijven. Gezien ALS ongeneselijk is en in combinatie met een snelle progressie én de onvervulde palliatieve zorgbehoeften, is een geïntegreerde palliatieve zorgaanpak, met inbegrip van vroegtijdige zorgplanning van algemeen belang voor deze populatie. Onderzoek heeft aangetoond dat multidisciplinaire zorg in ALS de levenskwaliteit verbetert, het aantal ziekenhuisopnames vermindert en het verblijf in het ziekenhuis voor patiënten verkort. Veel mensen met ALS willen zo lang mogelijk thuisblijven en daar verzorgd worden, zelfs in de terminale stadia van de ziekte, maar slechts 40% sterft thuis.

Personen met dementie en hun palliatieve zorgnoden

Personen met dementie hebben problemen met het uitvoeren van activiteiten van het dagelijks leven en kunnen ook gedrags- en psychologische veranderingen ervaren zoals apathie, depressie, agressie, angst of slaapproblemen. Zij ervaren dus meerdere fysieke, psychosociale en spirituele zorgbehoeften die maanden of jaren aanhouden tot aan de dood. Aan deze zorgbehoeften wordt echter vaak niet voldaan. Hoewel de meeste personen met dementie thuis willen blijven en sterven, sterven de meeste personen met dementie vaak in een woonzorgcentrum. Het bieden van kwalitatief goede zorg aan het levenseinde voor personen met dementie is dus een belangrijk aspect van de zorg binnen een woonzorgcentrum. Het is echter een uitdaging om bij dementie kwalitatief hoogwaardige zorg aan het levenseinde te bieden, omdat dementie vaak niet als een terminale aandoening wordt beschouwd, hetgeen kan leiden tot een slechte symptoombehandeling of overplaatsingen naar een ziekenhuis aan het levenseinde.

Personen met kanker en hun palliatieve zorgnoden

Ondanks de vooruitgang in diagnose en behandeling blijft kanker een belangrijke levensbeperkende ziekte, met 18,1 miljoen nieuwe gevallen en 9,6 miljoen doden wereldwijd in 2018. Personen met kanker krijgen doorgaans oncologische zorg, die erop gericht is om kanker te genezen, de symptoomlast te verbeteren, medische complicaties in verband met kanker te verminderen of het leven te verlengen. Palliatieve zorg bij kanker richt zich eerder op verlichting van de symptomen. Wanneer genezing voor mensen met kanker niet langer een optie is, moet de zorg worden afgestemd op hun behoeften en voorkeuren inzake symptoombestrijding, psychologische ondersteuning, sociale, existentiële en palliatieve zorgbehoeften. Deze behoeften blijven echter vaak onvervuld, vooral de emotionele steun werd gerapporteerd als de meest voorkomende onvervulde behoefte bij mensen met kanker en hun mantelzorgers. Tijdige en doeltreffende communicatie over vroegtijdige zorgplanning wordt dus algemeen aanbevolen voor deze patiëntenpopulatie en hun families.

Vroegtijdige zorgplanning als onderdeel van kwaliteitsvolle zorg rond het levenseinde

Vroegtijdige zorgplanning wordt gedefinieerd als een '*continu, vroegtijdig geïnitieerd communicatieproces tussen patiënten, hun mantelzorgers en/of zorgverleners dat volwassenen op elke leeftijd of in elk stadium van hun gezondheid ondersteunt bij het begrijpen en delen van hun persoonlijke waarden, levensdoelen en voorkeuren met betrekking tot toekomstige medische zorg, waaronder zorg rond het levenseinde*'. Het doel van vroegtijdige zorgplanning is een tijdige verkenning van iemands onderliggende waarden en voorkeuren voor toekomstige zorg om een gezamenlijk begrip te ontwikkelen dat de patiënt centraal stelt. Dit is vooral belangrijk als patiënten later cognitief en communicatief niet meer in staat zouden zijn om hun eigen beslissingen omtrent hun zorg te nemen. Recente aanbevelingen wijzen erop dat vroegtijdige zorgplanning moet worden gezien als een reeks bredere gesprekken over hoop, voorkeuren en mogelijke zorgdoelen. Bovendien zijn voorkeuren voor huidige en toekomstige zorg situationeel en zijn ze vaak gerelateerd aan belangrijke gebeurtenissen zoals symptoomprogressie of meerdere ziekenhuisopnames. Daardoor kunnen die voorkeuren in de loop van de tijd veranderen. De meeste studies onderzoeken echter de vroegtijdige zorgplanning op één specifiek tijdstip, wat slechts een momentopname is van de complexe realiteit dat het proces van vroegtijdige zorgplanning met zich meebrengt. Studies die het proces van vroegtijdige zorgplanning doorheen de tijd in kaart brengen - zoals longitudinale studies - zijn schaars.

Vooral bij een snel veranderend ziektetraject zoals in ALS is het onduidelijk wanneer en hoe vroegtijdige zorgplanning plaatsvindt en verandert in de tijd. Vanuit de literatuur wordt er aanbevolgen om bij personen met ALS vroegtijdige zorgplanning niet uit te stellen tot de laatste levensmaanden of -weken. Vanuit de literatuur merken we op dat de meeste studies over vroegtijdige zorgplanning bij ALS zich vooral focust op de risico's en voordelen van levensverlengende interventies (zoals invasieve of niet-invasieve beademingsondersteuning of gastrostomie), de identificatie van een wettelijke vertegenwoordiger en het invullen van een wilsverklaring. Het is onduidelijk wanneer en hoe deze onderwerpen van zorgplanning aan bod komen bij personen met ALS en hun mantelzorgers en hoe deze onderwerpen in de loop van de tijd veranderen en door welke factoren ze worden beïnvloed.

Betrokkenheid van mantelzorgers bij vroegtijdige zorgplanning en hun evaluatie van levenseindezorg

Mantelzorgers spelen een belangrijke rol in de zorg rond het levenseinde, niet alleen door fysieke en emotionele of spirituele zorg te verlenen aan hun ernstig zieke familielid, maar ook door informatie en communicatie te verstrekken aan zorgverleners over de wensen van hun ernstig zieke familielid. De bijdrage van mantelzorgers aan de zorg voor hun familielid is vaak groter dan die van zorgverleners, wat betekent dat mantelzorgers veel mogelijkheden hebben om met hun dierbare te praten over vroegtijdige zorgplanning buiten de klinische context. Mantelzorgers staan open voor gesprekken over vroegtijdige zorgplanning, maar een aantal barrières houdt hen tegen om deze gesprekken te voeren, zoals angst, of het gevoel onvoorbereid of ongemakkelijk te zijn omtrent het gesprek. Zorgverleners kunnen dus een belangrijke rol spelen om de mantelzorger in staat te stellen een vroegtijdig zorgplanningsgesprek te voeren. Studies over het percentage van mantelzorgers die een vroegtijdig zorgplanningsgesprek hebben gevoerd en daarbij ondersteuning hebben gekregen van een zorgverlener ontbreken momenteel. Vervolgens zou de betrokkenheid van mantelzorgers bij vroegtijdige zorgplanningsgesprekken de psychologische distress moeten verminderen. We weten echter niet of een interventie over vroegtijdige zorgplanning de betrokkenheid van mantelzorgers bij de besluitvorming zou vergroten en de psychologische distress voor mantelzorgers zou verminderen.

Indien een persoon wordt opgenomen in een woonzorgcentrum, zoals personen met dementia, blijven mantelzorgers vaak nauw betrokken bij diens zorg. Mantelzorgers kunnen belangrijke informatie over de bewoner verstrekken aan de zorgverleners in het woonzorgcentrum, meestal omdat bewoners met dementie doorgaans minder goed in staat zijn hun behoeften en symptomen kenbaar te maken of informatie te verstrekken over symptoomverbetering na een behandeling aan het levenseinde. Daarom is het perspectief van mantelzorgers over de kwaliteit van de zorg rond het levenseinde van waarde. Tot nu toe hebben slechts enkele studies de kwaliteit van de zorg aan het levenseinde geëvalueerd vanuit het perspectief van mantelzorgers van bewoners met dementie.

Doelstellingen van het onderzoek

Het doel van dit proefschrift is tweeledig. Ten eerste willen we diepgaande inzichten verschaffen in een complex en evoluerend proces zoals vroegtijdige zorgplanning in het ziektetraject van ALS (deel 1 van dit proefschrift). Om onderzoeksdoel 1 te bereiken, beschrijven we eerst het studieprotocol van de kwalitatieve longitudinale multi-perspectieve interviewstudie (**hoofdstuk 1**). In **hoofdstuk 2** rapporteren we de ervaringen van mensen met ALS en hun mantelzorgers omtrent vroegtijdige zorgplanning, en of, hoe en waarom deze ervaringen veranderen in de tijd. Ten tweede willen we ook inzicht geven in de betrokkenheid van de mantelzorgers bij vroegtijdige zorgplanning en hun evaluatie van de kwaliteit levenseindezorg (deel 2 van dit proefschrift), door te onderzoeken hoe mantelzorgers ondersteund of 'empowered' worden door zorgverleners (**hoofdstuk 3**), om te weten of mantelzorgers betrokken waren bij de besluitvorming rond het levenseinde, en de mate van psychologische distress die deze mantelzorgers ervaren na het overlijden van hun dierbare met kanker. (**hoofdstuk 4**), en om te onderzoeken hoe mantelzorgers de kwaliteit van de zorg rond het levenseinde evalueren bij bewoners met dementie in een woonzorgcentrum (**hoofdstuk 5**).

Methoden

We gebruikten verschillende studieopzetten en -methoden om onze twee onderzoeksdoelen te realiseren. Om diepgaande inzichten te geven in een complex en evoluerend proces van vroegtijdige zorgplanning gedurende het ziektetraject van ALS, gebruikten we een kwalitatief longitudinaal onderzoeksdesign, waarbij we personen met ALS en hun mantelzorgers drie keer individueel interviewden over een periode van maximaal negen maanden (**hoofdstukken 1-2**). Om te onderzoeken hoe mantelzorgers door zorgverleners ondersteund of 'empowered' werden tijdens vroegtijdige zorgplanningsgesprekken, voerden we een bevolkingsonderzoek uit bij nabestaanden. Deze mantelzorgers werden geselecteerd via de drie grootste zorgverzekeringen in Vlaanderen, België (**Hoofdstuk 3**). We gebruikten secundaire data-analyse van de ACTION trial, een geclusterde gerandomiseerde gecontroleerde trial waarin de doeltreffendheid van de ACTION Respecting Choices (RC) vroegtijdige zorgplanningsinterventie werd getest bij personen met gevorderde long- of darmkanker in zes Europese landen (België, Nederland, Italië, Denemarken, Slovenië en het Verenigd Koninkrijk). Deze data-analyse werd gebruikt om de interventie ACTION

Respecting Choices (RC) vroegtijdige zorgplanningsinterventie te beoordelen op de betrokkenheid van mantelzorgers bij de besluitvorming rond het levenseinde en de mate van psychologische distress die deze mantelzorgers eventueel ervaarden na het overlijden van personen met gevorderde kanker (**Hoofdstuk 4**). Ten slotte, om de kwaliteit van zorg aan het levenseinde zoals ervaren door mantelzorgers van bewoners met dementie te onderzoeken, gebruikten we twee retrospectieve epidemiologische studies met vergelijkbare onderzoeksmethoden uit 2010 en 2015 (**Hoofdstuk 5**).

Belangrijkste bevindingen

DEEL I: Diepgaande ervaringen met vroegtijdige zorgplanning bij mensen met ALS vanuit verschillende perspectieven van patiënten en mantelzorgers.

We ontwikkelden een studieprotocol voor een kwalitatieve longitudinale multi-perspectieve interviewstudie om diepgaande ervaringen met vroegtijdige zorgplanning bij personen met ALS te begrijpen via verschillende perspectieven van patiënten en mantelzorgers (**hoofdstuk 1)**. Personen met ALS en hun mantelzorgers werden door hun behandelend neuroloog benaderd om na te gaan of zij bereid waren deel te nemen aan het onderzoek. Zij werden alleen door iemand van het onderzoeksteam benaderd indien zij toestemming hadden gegeven aan hun neuroloog om hun contactgegevens door te geven. De inclusiecriteria voor personen met ALS waren de volgende: personen met ALS werden niet langer dan zes maanden geleden gediagnosticeerd, waren ouder dan 18 jaar, konden Nederlands spreken, konden vóór het eerste interview een informed consent ondertekenen en hadden geen fronto-temporale dementie. De inclusiecriteria voor de mantelzorgers waren de volgende: mantelzorgers waren ouder dan 18 jaar, konden Nederlands spreken en konden voor het eerste interview een informed consent ondertekenen.

In **hoofdstuk 2** beschreven we de resultaten van de kwalitatieve longitudinale multi-perspectieve interviewstudie waarin we probeerden de ervaringen met vroegtijdige zorgplanning voor mensen met ALS en hun mantelzorgers te begrijpen, en wanneer en hoe deze ervaringen zich in de loop van het ziektetraject ontvouwen. Over een periode van ongeveer negen maanden konden we negen dyades van mensen met ALS en hun mantelzorger (meestal) individueel interviewen over vroegtijdige zorgplanning, (meestal) elk drie keer, wat resulteerde in een totaal van 44 interviews. Over het algemeen dachten alle personen met ALS en hun mantelzorgers over de tijd heen, tot op zekere hoogte na over hun toekomst en toekomstige (levenseinde)zorg, maar niet alle dyades spraken hierover met elkaar of met een zorgverlener. Na verloop van tijd, werden de ervaringen met vroegtijdige zorgplanning beïnvloed door verschillende maar met elkaar verweven bepalende factoren: (1) de ervaren lichamelijke achteruitgang bij personen met ALS, zoals het niet meer kunnen lopen of praten en daarmee samenhangende toekomstige zorgbehoeften die in de loop van de tijd veranderen; (2) hoe personen met ALS zichzelf identificeren als patiënt; (3) het verkrijgen van informatie over diagnose en prognose en het leren dat iedereen anders evolueert wat het plannen voor de toekomst moeilijk maakt; (4) professionals die gesprekken initiëren over medische aspecten van beslissingen rond het levenseinde, ook al ervaarden deelnemers dit als stressvol of te vroeg; (5) het balanceren tussen hoop om stabiel te blijven en zorgen over wat de toekomst zou kunnen brengen; (6) zichzelf en elkaar beschermen tegen wat de toekomst brengt.

DEEL II: Betrokkenheid van mantelzorgers bij vroegtijdige zorgplanning en hun evaluatie van de zorg rond het levenseinde.

In **hoofdstuk 3** onderzochten wij de mate waarin mantelzorgers van mensen met een ernstige ziekte thuis vroegtijdige zorgplanningsgesprekken voerden met hun familielid in de laatste drie maanden van het leven en hoe zij daarbij werden ondersteund door zorgverleners. Wij voerden een bevolkingsonderzoek uit en includeerden in dit onderzoek nabestaanden die zorg hadden verleend aan iemand die recent was overleden (overlijden was niet langer twee tot zes maanden geleden) en dit via de registers van drie grootste zorgverzekeringen in Vlaanderen, België. Uit onze bevindingen blijkt dat ongeveer de helft van de mantelzorgers aangaf vroegtijdige zorgplanningsgesprekken te hebben gevoerd met hun chronisch zieke familielid, in zijn/haar laatste drie levensmaanden. Deze vroegtijdige zorgplanningsgesprekken vonden vaker plaats wanneer de mantelzorgers 55 jaar of jonger waren, een medisch diploma hadden (arts of verpleegkundige) of wanneer gespecialiseerde palliatieve zorgdiensten betrokken waren, maar minder vaak wanneer bij de personen waarbij dementie was vastgesteld. Achtenzeventig procent van de mantelzorgers kreeg steun van een zorgverlener, en dat was meestal de huisarts. Het type ondersteuning dat werd ontvangen was meestal de zorgverlener die het vroegtijdig zorgplanningsgesprek samen met de mantelzorger of de ernstig zieke persoon voerde, wat kan worden beschouwd als een 'empowerende-samenwerkende' aanpak. Zelden werd in deze studie de meer 'empowerende' vormen van ondersteuning gerapporteerd om de zelfredzaamheid van mantelzorgers te vergroten, zoals het verstrekken van informatie of uitleg over het voeren van deze gesprekken.

In **hoofdstuk 4** rapporteerden wij de secundaire uitkomsten van de ACTION trial betreffende het effect van de ACTION Respecting Choices (RC) vroegtijdige zorgplanningsinterventie op de betrokkenheid van mantelzorgers bij de besluitvorming in de laatste drie maanden van het leven van de patiënt, en psychologisch distress na drie maanden van rouw. De ACTION-studie was een multicentrische clustergecontroleerde gerandomiseerde studie in zes Europese landen (België, Denemarken, Italië, Nederland, Slovenië en het Verenigd Koninkrijk) waarin de doeltreffendheid van de ACTION RC vroegtijdige zorgplanningsinterventie werd aangepast en getest bij patiënten met gevorderde long- of darmkanker. De interventiegroep ontving de ACTION RC vroegtijdige zorgplanningsinterventie en gebruikelijke zorg, terwijl de controlegroep enkel gebruikelijke zorg ontving. Personen met gevorderde kanker die deel uitmaakten van de interventiegroep werden uitgenodigd om samen met hun mantelzorger en een getrainde facilitator een vroegtijdig zorgplanningsgesprek te hebben. De patiënten werden aangemoedigd om hun voorkeuren te delen met familieleden en naasten, en om deze voorkeuren desgewenst te documenteren. Voor deze studie werden gegevens van de 'bereavement questionnaire' gebruikt. Wij vonden dat nabestaanden van patiënten die deelnamen aan de ACTION RC vroegtijdige zorgplanningsinterventie geen significant hogere kans hadden om betrokken te zijn bij de besluitvorming in de laatste drie maanden van het leven van de patiënt. Gemiddeld bleek de interventiegroep (89,6%) iets hoger betrokken te zijn bij besluitvorming dan in de controlegroep (86,7%). De interventiegroep had drie maanden na het overlijden een iets hoger niveau van psychologische distress dan de controlegroep, maar ook dit verschil was niet statistisch significant.

In **hoofdstuk 5** onderzochten we de evaluatie van de mantelzorgers van de kwaliteit van levenseindezorg voor personen met dementie in een woonzorgcentrum en welke factoren mogelijk samenhangen met deze evaluatie van de kwaliteit van levenseindezorg. De evaluatie was hoger wanneer mantelzorgers aangaven informatie te hebben ontvangen over palliatieve zorg en wanneer mantelzorgers aangaven informatie over medische zorg te hebben ontvangen van een zorgverlener.

Bespreking van de belangrijkste bevindingen

DEEL I: Ervaringen met vroegtijdige zorgplanning bij mensen met ALS vanuit het perspectief van patiënten en mantelzorgers.

Zorgplanning is meer dan een gemedicaliseerd proces

Onze resultaten uit **hoofdstuk 2** tonen aan dat vroegtijdige zorgplanning een holistisch proces is waarin medische, sociale en psychologische factoren met elkaar verweven zijn en niet als afzonderlijke entiteiten kunnen beschouwd worden. In ons onderzoek dachten mensen met ALS en hun mantelzorgers na over de toekomst en de toekomstige zorg. Meestal dachten ze na over wat er zou gebeuren in de evolutie van de ziekte, eerder dan het nadenken over de medische aspecten van beslissingen rond het levenseinde. Dit sluit aan bij het bredere concept van vroegtijdige zorgplanning, waarbij mensen met een chronische levensbeperkende aandoening zich in eerste instantie bewust worden van de betekenis van "goed leven", wat belangrijk is voor hen en wat het voor hen zou betekenen mochten ze zieker worden. Deze gesprekken over doelen en waarden zullen uiteindelijk helpen bij het nemen van de best mogelijke beslissingen over behandeling of zorg en deze zullen ook meer aansluiten bij hun kernwaarden. Desalniettemin blijft de overheersende focus van vroegtijdige zorgplanning nog te vaak gericht op medische aspecten van voorkeuren voor het levenseinde, zoals het vermijden van ongewenste behandelingen die als potentieel schadelijk kunnen worden beschouwd, en het invullen van wilsverklaringen. In studies binnen ALS wordt aanbevolen dat gesprekken over medische aspecten van de zorg, zoals gastrostomie, tracheotomie of medische beslissingen rond het levenseinde, vroeg in het ALS-traject plaatsvinden. In onze studie bleek dat deze gesprekken - meestal door mantelzorgers - vaak als te vroeg of te belastend werden ervaren. Deze resultaten bevestigen dat het waarschijnlijk beter zou zijn om te beginnen met gesprekken over waarden en wat het betekent om goed te leven, in plaats van het gesprek te beginnen over de medische aspecten van de zorg rond het levenseinde. Dit wordt ook gesuggereerd in andere literatuur over vroegtijdige zorgplanning bij patiënten met multiple sclerose, dementie en kanker.

Transformatieve en relationele complexiteiten van vroegtijdige zorgplanning

In **hoofdstuk 2** suggereren onze resultaten dat na verloop van tijd verschillende factoren die verband houden met het omgaan met de ernst van de terminale ziekte (vb. ALS) en de relationele dynamiek, een rol spelen bij het construeren van de ideeën, gedachten en gevoelens van mensen over toekomstige zorg en behandelingsvoorkeuren. Leven met een chronische levensbeperkende ziekte wordt gezien als een transformerende ervaring, wat betekent dat men niet kan weten hoe men ermee zou omgaan of reageren totdat men in die situatie leeft. Daarom is het plannen van de toekomst moeilijk, omdat keuzes doorheen de tijd kunnen veranderen door de onvoorspelbaarheid van de toekomst. Een belangrijk thema in **hoofdstuk 2** met betrekking tot coping was de balans tussen hoop om stabiel te blijven en de bezorgdheden over wat de toekomst zou kunnen brengen. Hoop kan nuttig zijn om het emotionele leed dat ontstaat bij het leven met een chronische levensbeperkende ziekte aan te kunnen of onder controle te houden. De lichamelijke achteruitgang zette alle deelnemers (personen met ALS en hun mantelzorgers) er echter toe aan om na te denken over wat de toekomst zou kunnen brengen (**hoofdstuk 2**). Ze waren niet allemaal bereid hierover met hun mantelzorger te praten, meestal omdat ze elkaar en zichzelf wilden beschermen gezien deze gesprekken vaak als emotioneel belastend worden ervaren. Studies hebben aangetoond dat zorgverleners een belangrijke rol kunnen spelen bij het ondersteunen en faciliteren van deze gesprekken tussen mantelzorgers en patiënten.

DEEL II: Betrokkenheid van mantelzorgers bij vroegtijdige zorgplanning en hun evaluatie van de zorg aan het levenseinde

Het belang van het betrekken van mantelzorgers bij vroegtijdige zorgplanning

De resultaten die in **hoofdstuk 4** werden beschreven, gaven aan dat mantelzorgers van personen met kanker die betrokken waren bij een vroegtijdige zorgplanningsinterventie niet meer betrokken waren bij de besluitvorming dan mantelzorgers die niet van deze interventie hebben genoten. Eerdere studies hebben aangetoond dat betrokkenheid van mantelzorgers bij vroegtijdige zorgplanningsgesprekken cruciaal is omdat zij vaak beslissingen nemen namens hun chronisch zieke familielid aan het levenseinde. De vroegtijdige zorgplanningsinterventie in **hoofdstuk** 4, was vooral gericht op de patiënt en besteedde onvoldoende aandacht aan empowerment en voorbereiding van de mantelzorger op betrokkenheid bij de besluitvorming. De meeste vroegtijdige zorgplanningsinterventies richten zich vaak alleen op het identificeren van een wettelijke vertegenwoordiger voor de patiënt, maar het is ook belangrijk dat die wettelijke vertegenwoordiger voldoende betrokken is bij vroegtijdige zorgplanning zodat hij/zij voorbereid is om indien nodig beslissingen te nemen aan het levenseinde. Wanneer mantelzorgers de voorkeuren van patiënten kennen en begrijpen dat ze ook wat bewegingsruimte hebben in de besluitvorming, kan dit het vertrouwen van de mantelzorgers in het nemen van beslissingen rond het levenseinde vergroten, en ook het verdriet, de stress, de angst of de depressie tijdens de rouwperiode verminderen. In onze studie werd echter geen verschil in psychologische distress gevonden tussen mantelzorgers die betrokken waren bij een vroegtijdige zorgplanningsinterventie en mantelzorgers die niet bij een dergelijke interventie betrokken waren. Verschillende studies over vroegtijdige zorgplanning hebben welzijn en psychologische distress als uitkomstmaten gebruikt, maar er zijn momenteel geen eenduidige conclusies over deze uitkomstmaten binnen het onderzoek van vroegtijdige zorgplanning.

Faciliterende factoren voor mantelzorgers om deel te nemen aan vroegtijdige zorgplanning

In **hoofdstuk 3** tonen de resultaten aan dat ongeveer de helft van de mantelzorgers deelnemen aan vroegtijdige zorgplanningsgesprekken. Vooral wanneer mantelzorgers 55 jaar of jonger zijn, wat mogelijks kan te maken hebben met het feit dat de jongere generaties meer gedetailleerde informatie over de ziekte en palliatieve zorg wensen, alsook meer betrokkenheid en autonomie bij de besluitvorming willen. Betrokkenheid van gespecialiseerde palliatieve zorgdiensten werd ook gezien als een faciliterende factor. Dit resultaat was niet verrassend aangezien een van de kerntaken van gespecialiseerde palliatieve zorgdiensten is om patiënten en mantelzorgers te informeren over de ernst van de ziekte en hen voor te bereiden op het nakende overlijden. Het kan ook zijn dat mantelzorgers zich meer bewust waren van de slechte gezondheidstoestand van hun dierbare omdat er gespecialiseerde palliatieve zorgdiensten bij betrokken waren en ze hierdoor dus een vroegtijdig zorgplanningsgesprek hebben gehad. Mantelzorgers met een medisch diploma lijken ook meer deel te nemen aan gesprekken over vroegtijdige zorgplanning dan mantelzorgers zonder medisch diploma. Vermoedelijk omdat zij zich meer bewust zijn van het belang van deze gesprekken. Het lijkt er dus op dat kennis over de gezondheidstoestand of het ontvangen van informatie over medische zorg of palliatieve zorg belangrijke voorwaarden zijn voor mantelzorgers om deel te nemen aan vroegtijdige zorgplanningsgesprekken.

De rol van zorgverleners bij het ondersteunen van en samenwerken met mantelzorgers bij gesprekken rond vroegtijdige zorgplanning

Zoals uit **hoofdstuk 3** blijkt, kunnen zorgverleners een belangrijke rol spelen bij het ondersteunen en het faciliteren van vroegtijdige zorgplanningsgesprekken bij mantelzorgers. Dit was meestal door het vroegtijdig zorgplanningsgesprek samen met de mantelzorger en de patiënt te voeren. Een andere mogelijkheid was om de mantelzorger door te verwijzen naar nuttige hulpmiddelen of instrumenten die kunnen helpen bij het vroegtijdig zorgplanningsgesprek, zoals boeken of websites. Deze laatste mogelijkheid werd minder aangegeven door de mantelzorgers. Mogelijks voelden mantelzorgers zich veiliger om deze gesprekken samen met een zorgverlener te voeren of misten ze de nodige instrumenten of vaardigheden om deze gesprekken zelf te voeren. Meestal werd de mantelzorger ondersteund door de huisarts, wat niet verwonderlijk is, aangezien deze de patiënt en de mantelzorger vaak al geruime tijd kent en vaak op de hoogte is van zowel de medische als de sociale context van de patiënt. Onze resultaten beschreven in **hoofdstuk 4** suggereren ook dat het beter is dat de zorgverlener deel uitmaakt van het zorgteam van de patiënt om vroegtijdige zorgplanningsgesprekken te voeren, want een externe facilitator die geen deel uitmaakt van het zorgteam zorgt niet voor verbeterde kwaliteit van levenseindezorg bij patiënten of mantelzorgers.

Vroege initiatie van vroegtijdige zorgplanningsgesprekken bij neurodegeneratieve ziekten

Onze resultaten, beschreven in **hoofdstuk** 3, hebben aangetoond dat de kans dat mantelzorgers een vroegtijdig zorgplanningsgesprek voeren met hun naaste in de laatste 3 levensmaanden afnam indien de patiënt gediagnosticeerd was met dementie. Waarschijnlijk omdat de persoon met dementie niet meer de mentale capaciteit had om aan deze gesprekken deel te nemen. In de literatuur wordt er aangeraden om vroegtijdige zorgplanningsgesprekken vroeg in het ziektetraject te laten plaatsvinden, vooral bij neurodegeneratieve ziekten zoals dementie en ALS. Uit eerdere studies is echter gebleken dat slechts een minderheid van de mensen met dementie de kans krijgt om zelf aan een vroegtijdige zorgplanningsgesprek deel te nemen en dat de zorgverlener vaker alleen de mantelzorger erbij betrekt. Er is aanbevolen in de literatuur om mensen met dementie zoveel mogelijk gelegenheid te geven om als actieve personen deel te nemen aan de vroegtijdige zorgplanning, zodat hun capaciteiten naar voren komen en hun stem wordt gehoord. Onze resultaten uit **hoofdstuk 2** laten zien dat zorgverleners wel degelijk vroeg in het ziektetraject gesprekken starten over de mogelijkheden en risico's van een gastrostomie, een tracheotomie en euthanasie, meestal kort na de diagnose en/of in vervolgconsulten, hoewel andere studies hebben aangetoond dat vroegtijdige zorgplanning bij ALS vaak wordt uitgesteld door zorgverleners tot de laatste maanden of weken van het leven. In onze studie ervaarden de mantelzorgers en personen met ALS deze gesprekken echter als te vroeg of te belastend. Eerdere studies over vroegtijdige zorgplanning bij dementie of multiple sclerose hebben aangetoond dat het wellicht beter zou zijn om vroegtijdige zorgplanning te starten op belangrijke triggerpoints zoals de diagnose, maar ook wanneer een impactvolle verslechtering optreedt. Dit blijkt ook uit onze resultaten beschreven in **hoofdstuk 2**.

Het is belangrijk om op te merken dat de zorg voor mensen met ALS en hun familieleden vaak op tertiair niveau wordt verstrekt. In België worden deze de neuromusculaire referentiecentra genoemd, die slechts een adviserende rol hebben en beschikken over expertise in neurologie, ademhalingszorg, revalidatie, alsook in psychologie, fysiotherapie, ergotherapie, logopedie, voeding en maatschappelijk werk. In onze studie, beschreven in **hoofdstuk 2,** merkten we op dat personen met ALS en hun mantelzorgers geen contact opnamen met de zorgverleners van de neuromusculaire referentiecentra. Ondanks dat deze zorgverleners hadden aangegeven dat zij hen op elk moment mochten contacteren met vragen over zorg of psychosociale en existentiële vragen. Dit kan vermoeden dat de drempel te hoog was voor personen met ALS en hun mantelzorgers om contact op te nemen met deze neuromusculaire referentiecentra en hierdoor een vroegtijdig zorgplanningsgesprek mogelijks ook heeft belemmerd. Studies bij ALS hebben echter aangetoond dat uitgestelde informatie over zorg of hulpmiddelen zoals een rolstoel of een spraakcomputer, de beperkte beschikbaarheid van zorgverleners en een gebrek aan psychosociale ondersteuning in vroegtijdige zorgplanningsgesprekken alsook bij beslissingen over het levenseinde resulteerden in een afname van de levenskwaliteit van personen met ALS. In onze studie, beschreven in **hoofdstuk 2**, voelden de deelnemers zich vaak alleen, niet gesteund en hadden zij moeite om hun weg te vinden in het gezondheidszorgsysteem, mogelijk omdat eerstelijns- en palliatieve zorgdiensten vaak niet betrokken waren, wat van invloed heeft op de ervaringen met vroegtijdige zorgplanning bij personen met ALS en hun mantelzorgers. Eerder onderzoek bij neurodegeneratieve ziekten heeft aangetoond dat een langdurige, vertrouwensvolle en empathische relatie met een zorgverlener belangrijk is, zeker bij vroegtijdige zorgplanningsgesprekken. In België moeten personen met ALS en hun mantelzorgers vaak zelf de hulp inschakelen van primaire en palliatieve zorgverleners, en zoals eerder aangetoond in **hoofdstuk 3,** zijn deze hulpverleners vaak diegene die ondersteuning bieden in vroegtijdige zorgplanningsgesprekken. Een mogelijke aanpak die vroegtijdige zorgplanning en interdisciplinaire samenwerking tussen de primaire, secundaire en tertiaire ALS zorg zou kunnen faciliteren is de ontwikkeling van een geïntegreerd zorgmodel. In Australië, het Ierland en Nederland is een dergelijk geïntegreerd collaboratief zorgmodel bij ALS ontwikkeld waarin personen met ALS en hun mantelzorgers worden ondersteund door alle zorgverleners die betrokken zijn in de zorg in ALS. Deze modellen hebben een toename van de levenskwaliteit van personen met ALS en een afname van de belasting van mantelzorgers aangetoond.

Informatie en communicatie als belangrijke voorwaarden voor goede zorg rond het levenseinde.

Onze resultaten in **hoofdstuk 5** laten zien dat mantelzorgers relatief tevreden zijn met veel aspecten omtrent de kwaliteit van levenseindezorg die bewoners met dementie in een woonzorgcentrum hebben ontvangen. Mantelzorgers die informatie over palliatieve of medische zorg ontvingen, beoordeelden de kwaliteit van de zorg doorgaans beter dan degenen die geen informatie over palliatieve of medische zorg hadden ontvangen. Deze bevinding kan op verschillende manieren worden geïnterpreteerd. Het verstrekken van informatie over palliatieve en medische zorg zou een belangrijke voorwaarde kunnen zijn voor kwalitatief goede zorg aan het levenseinde zoals beoordeeld door de mantelzorgers. Maar het kan ook zijn dat het personeel in woonzorgcentra, waar goede kwaliteit van levenseindezorg wordt geleverd, automatisch meer informatie over palliatieve en medische zorg verstrekt aan de mantelzorgers. Deze relatie tussen informatievoorziening en evaluatie van de zorgkwaliteit komt overeen met bevindingen uit eerdere studies over het belang voor mantelzorgers om geïnformeerd te worden, niet alleen over medische zorg, maar ook over het levenseinde. Informatieverstrekking is de eerste belangrijke stap in vroegtijdige zorgplanning, want wanneer patiënten en mantelzorgers geïnformeerd zijn over de ziekte of prognose, helpt dit hen om weloverwogen medische beslissingen te nemen omtrent toekomstige (levenseinde)zorg. Studies hebben ook aangetoond dat vroegtijdige zorgplanning de kwaliteit van zorg aan het levenseinde verbetert. Vroegtijdige zorgplanning is dus een belangrijke voorwaarde voor een goede kwaliteit van levenseindezorg.

Aanbevelingen voor onderzoek, praktijk en beleid

Aanbevelingen voor onderzoek

- Het gebruik van onderzoeksdesigns die de complexiteit van zorgplanning benadrukken en begrijpen, bijvoorbeeld het gebruiken van een longitudinale kwalitatieve studies

- Een consistente manier om welzijn en psychologisch distress te meten in vroegtijdige zorgplanning

- Er is meer inzicht nodig in specifieke ondersteuningsbehoeften bij mantelzorgers in vroegtijdige zorgplanningsgesprekken

- Een geïntegreerd zorgmodel ontwikkelen voor personen met ALS is noodzakelijk

Aanbevelingen voor de praktijk

- Vroegtijdige zorgplanning moet gericht zijn op "goed leven" of "wat belangrijk is", nu en in de toekomst.

- Zorgverleners kunnen patiënten en mantelzorgers helpen om hun empowerment en hun zelfredzaamheid te bevorderen om deel te nemen aan vroegtijdige zorgplanning

- Vroegtijdige zorgplanningsgesprekken worden bij voorkeur gefaciliteerd door een zorgverlener die deel uitmaakt van het zorgteam

Aanbevelingen voor het beleid

- Het verbeteren van vroegtijdige zorgplanning vergt een systeembenadering.

- Beleidsmakers moeten de uitvoering van het terugbetalingssysteem voor vroegtijdige zorgplanning monitoren en indien nodig aanpassen

Curriculum Vitae

Curriculum vitae

Isabel Vandenbogaerde was born on 3 September 1992 in Waregem (Belgium). She obtained her professional bachelor’s degree in Clinical Psychology in 2013 at the Katholieke Hogeschool Kortrijk. In 2016, she obtained her master’s degree in Clinicial Psychology at the Universiteit Gent, and a year later (in 2017) she obtained her master’s degree in Gender and Diveristy, which was an interuniversity educational program. In April 2018, Isabel joined the End-of-Life Care Research Group at the Vrije Universiteit Brussel and Universiteit Gent. Isabel worked on a Research Foundation Flanders (FWO) funded research project under the supervision of Prof dr Aline De Vleminck, Prof dr Luc Deliens, Prof dr Lieve Van den Block and Prof dr Jan De Bleecker. Her doctoral research focusses on the advance care planning from patient’s and family carers’ perspectives.

Professional training

2016-2017	**Master of Arts in Gender & Diversity** Master’s thesis: ‘Clarifications of suicidal thoughts and/or attempts in Flemish LGBT-persons’	Interuniversity (including Gent Universiteit, VUB, Universiteit Antwerpen, Universiteit Hasselt, Katholieke Universiteit Leuven
2013-2017	**Master of Science in Clinical Psychology** Master thesis: ‘“I am sensitive, but does this mean I can control less my feelings”: a physiological study’	Universiteit Gent Faculty of psychology and educational sciences
2010-2013	**Bachelor’s in clinical psychology**	Katholieke Hogeschool Kortrijk (KATHO)

Professional career

April 2018 - 2023	**Doctoral (PhD) Researcher** Involved in 3 projects related to advance care planning and family carers, including the ACTION EU FP7 project, SBO-INTEGRATE and PACE EU FP 7 project.	End-of-life Care Research Group http://www.endoflifecare.be/ Vrije Universiteit Brussel (VUB)
January 2018 – March 2018	**Psychologist** Responsible for counselling and coaching students, teachers, and parents	Egmont & Hoorn (school for adolescents with intellectual disabilities)

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Presentations at (inter)national conferences

2022 Advance Care Planning in ALS. Opleidingsdag ALS Liga **[Invited speaker]**

Ondersteuning van mantelzorgers door professionals bij het voeren van ACP gesprekken: een cross-sectionele studie op populatieniveau. Dutch-Flemish Science-days in Palliative Care 2022, Tiel, The Netherlands **[Oral presentation]**

2021 ACP conversations by family carers: A population-based survey. EAPC World Congress online 2021 **[Oral presentation – live]**

2020 The effect of advance care planning on family carers' involvement in decision-making and their psychological distress: results of the ACTION trial. EAPC World Research Congress online 2020 [Poster presentation]

Collaboration between family carers and professional health carers in home care at the end-of-life: An interview study – International EAPC RN Seminar Public

Health Research in Palliative care: Towards Solutions for Global Challenges **[Oral presentation]**

2019 Wellbeing of family carers of people who died of cancer: Preliminary results of the ACTION advanced care planning (ACP) trial. 7th International Advance Care Planning Conference (ACP-I), Rotterdam, The Netherlands **[Oral presentation]**

The effect of advance care planning on wellbeing of family carers in six European countries: preliminary results of the ACTION – study. 16th World Congress of the European Association for Palliative Care (EAPC), Berlin, Germany [Poster presentation, **rewarded with best abstract in the category of family carers**]

The effect of advance care planning in patients with lung- or colorectal cancer and their family carers in 6 EU countries: de results of the RCT ACTION study. Seminar Thoracic Oncology Group Antwerp (TOGA), Antwerp, Belgium **[Invited speaker]**

ACTION-study: the effect of advance care planning on family carers' involvement in decision-making and their psychological distress. Dutch-Flemish Science-days in Palliative care 2019, Antwerp, Belgium **[Oral presentation].**

Grants or awards received

Wetenschappelijk Fonds Willy Gepts (WFWG) by University Medical Center (VUB), 2021

Award for best abstract in the category of family carers, EAPC World Congress, 2019