ROMY VAN RICKSTAL

10th of May 2023

ADVANCE CARE PLANNING IN YOUNG-ONSET DEMENTIA

Perspectives of People with Dementia, Family Caregivers and Physicians

Advance care planning is a process of communication between patients, family caregivers and professionals to explore patients' preferences for future care, including at the end of life. Nonetheless the potential significance of advance care planning in dementia is agreed upon, its uptake is specifically low in this population. Although up to 9 procent of people with dementia develop symptoms before the age of 65 years old and, as such, are diagnosed with young-onset dementia, there is a dearth of studies including these younger patients and their family caregivers.

This dissertation gives insight into their own perspectives on advance care planning, as well as into those of physicians, through several interview-studies with people with young-onset dementia and their family caregivers themselves and a focus group study with physicians. The results of this work show that especially the content of advance care planning might have nuances for these younger persons confronted with dementia. Moreover, several challenges for engaging in advance care planning were identified from the perspective of both patients and family caregivers, as well as physicians. This dissertation led to several recommendations for practice, policy and future research.

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9 789461 174901

ISBN 978 94 6117 490 1

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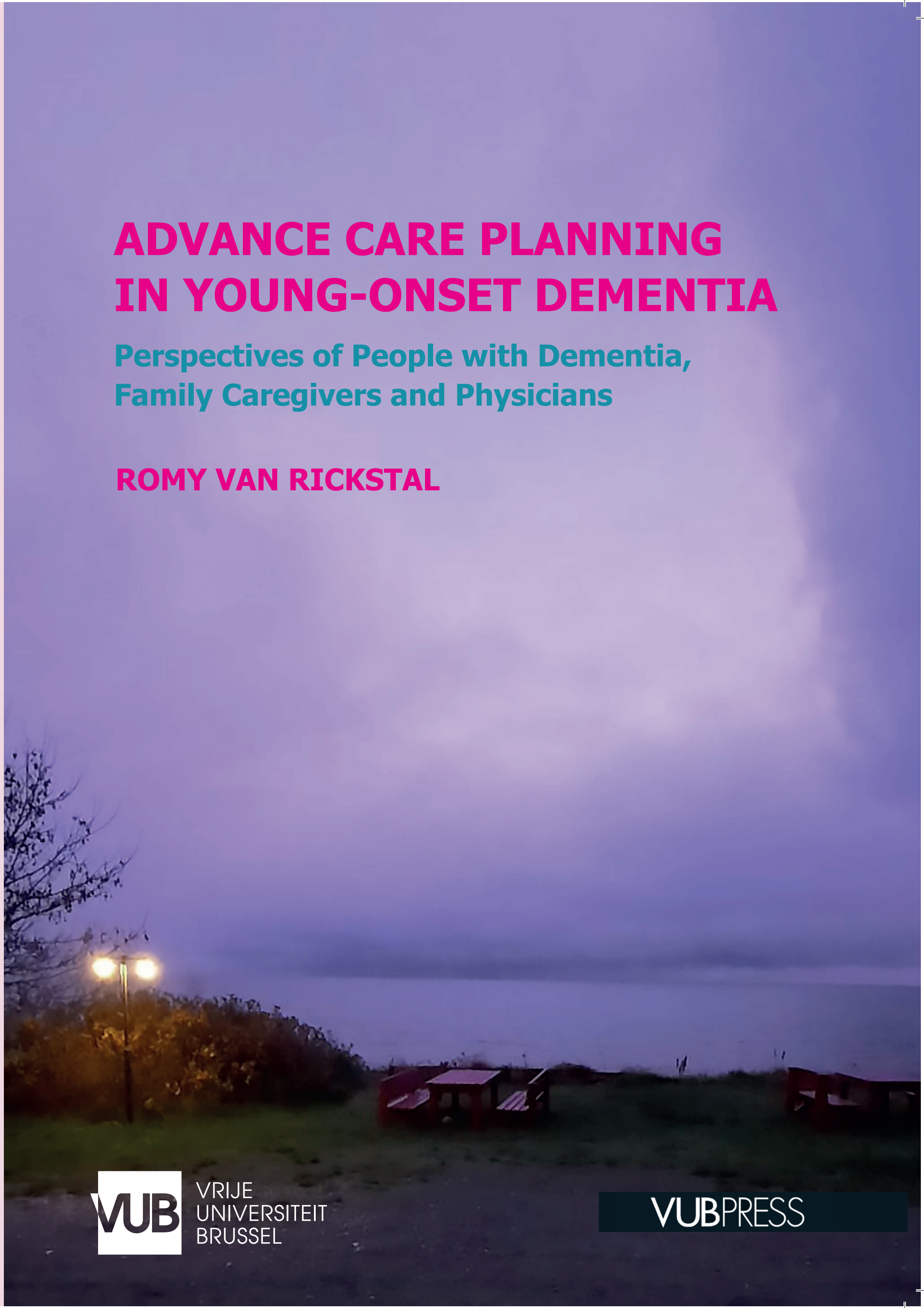

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Romy Van Rickstal

Thesis submitted in fulfilment of the requirements for the degree of

Doctor of Philosophy (PhD) in Social Health Sciences

10th of May 2023

Vrije Universiteit Brussel (VUB)

Faculty of Medicine and Pharmacy

End-of-Life Care Research Group

This dissertation is supported by a PhD Fellowship from the Research Foundation - Flanders (FWO)

SUPERVISORS

Promotor:
Prof. Dr. Lieve Van den Block
End-of-Life Care Research Group
Department of Family Medicine and Chronic Care

Co-promotors:
Prof. Dr. Aline De Vleminck
End-of-Life Care Research Group
Department of Family Medicine and Chronic Care

Prof. Dr. Sebastiaan Engelborghs
Department of Neurology, Universitair Ziekenhuis Brussel, Brussels, Belgium.
Center for Neurosciences (C4N), Vrije Universiteit Brussel (VUB), Brussels, Belgium.
Department of Biomedical Sciences, University of Antwerp, Antwerp, Belgium

EXAMINATION COMMITTEE

Prof. Dr. Johan Bilsen *(Chair*)
Department of Public Health, Vrije Universiteit Brussel
Mental Health and Wellbeing Research Group

Prof. Dr. Peter Paul De Deyn
Laboratory of Neurochemistry and Behavior, Institute Born-Bunge, University of Antwerp
Universiteit Groningen

Prof. Dr. Liesbeth De Donder
Department of Educational Sciences, Vrije Universiteit Brussel
Society and Ageing Research Lab

Prof. Dr. Eva Dierckx
Department of Clinical and Lifespan Psychology, Vrije Universiteit Brussel
Society and Ageing Research Lab

Prof. Dr. Chris Gastmans
KU Leuven
Centre for Biomedical Ethics and Law

Prof. Dr. Mathieu Vandenbulcke
KU Leuven Brain Institute (Director)
Department of Geriatric Psychiatry, University Hospital Leuven

Opgedragen aan mijn mooie grootmoeder,

Mevrouw Mirèse Vanrusselt

VOOR MIJN OUDERS

Alleen hetgeen niet weerkeren kon,
wilde ik fotograferen, al moest ik
er mijn nek voor breken.

Wat eeuwig was zette ik buiten beeld.
Het was niet veel. Twee mensen. Niet meer.

Hen had ik steeds bij de hand.
Kerken en kastelen stonden in een ver land.

Thans, als na een brand, zoek ik
in elke stip hun gezicht.

Eddy van Vliet

DANKWOORD (*Acknowledgements*)

Er zijn veel personen die ik graag wil danken voor hun hulp en steun de afgelopen jaren. Ik heb er evenwel voor gekozen om dit dankwoord aan slechts enkelen onder hen te richten.

Vooreerst wil ik diegenen die hebben deelgenomen aan mijn onderzoek bedanken en in het bijzonder de **personen met jongdementie en hun mantelzorgers**. Ik hoop bovenal dat dit werk ten dienste kan staan van jullie en, mettertijd, kan bijdragen aan de zorg die jullie krijgen.

Verder wil ik graag de **juryleden** oprecht bedanken voor hun interesse, expertise en scherpzinnige vragen en bedenkingen. Jullie maakten voor mij van deze laatste horde een leerzaam moment met waardevolle inzichten om mee te nemen naar de toekomst.

Bijzondere dank gaat uiteraard uit naar jou, **Lieve**, als mijn promotor. Toen ik je inmiddels meer dan zes jaar geleden hoorde spreken tijdens de PALM-opleiding, wist ik ‘bij haar moet ik zijn’. Mijn intuïtie bleek meer dan terecht. De weg die we samen hebben doorlopen, heeft mijn hoge verwachtingen nog weten overtreffen.

Aline, ook jij als mijn co-promotor en dagelijks begeleider, hebt me erg geïnspireerd. Altijd stond je paraat om advies te geven, te motiveren, maar ook om me uit te dagen.

Kunnen opkijken naar vrouwen als jullie, Lieve en Aline, heeft me niet alleen als onderzoekster, maar ook als persoon mee gevormd.

Bas, reeds aan de start van dit doctoraat mocht ik rekenen op je hulp, kritische kijk en klinische inzichten. Uw bijdrage maakte van dit doctoraat een mooier geheel.

Lieve, Aline, en Bas, dank jullie wel voor jullie geloof in mij en voor de kansen die jullie me gaven. Ten slotte wil ik graag uiten hoezeer ik jullie betrokkenheid gewaardeerd heb. De empathische en zorgzame wijze waarop jullie me steeds hebben begeleid, siert jullie als promotoren. Ik kijk dan ook naar wat de toekomst mag brengen.

Mama, dankzij u heb ik altijd het gevoel gehad te mogen staan op de schouders van reuzen. Jij hebt me geleerd om zo min mogelijk te vluchten voor het leven of voor wat het me biedt, integendeel. Lachen (zij het soms met een traan), het glas heffen en odes brengen aan het leven. Wat zijn wij daar samen bedreven in.

Vele rollen nam je op ten aanzien van mij en je vervulde ze stuk voor stuk glorieus. Liefde en bescherming in overvloed. Twee handen op één buik. Ik had het niet beter kunnen treffen.

Mijn diepste dank gaat uit naar u, Mamzeltje, voor zoveel meer dan ik kan verwoorden en op zoveel meer dagen dan vandaag. Met u als reisgezel, toch eender waar en wanneer aan mijn zijde, geraak ik er wel.

Proost op ons en grazie di tutto cuore.

Romy

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Abbreviations: Advance care planning (ACP) & young-onset dementia (YOD)

CHAPTERS ARE BASED ON THE FOLLOWING PUBLICATIONS

CHAPTER 1

Van Rickstal, R., De Vleminck, A., Aldridge, M. D., Morrison, S. R., Koopmans, R. T., van der Steen, J. T., Engelborghs, S., & Van den Block, L. (2019). Limited engagement in, yet clear preferences for advance care planning in young-onset dementia: An exploratory interview-study with family caregivers. *Palliative medicine*, *33*(9), 1166–1175. doi:10.1177/0269216319864777. [2018 SCI impact factor 4.956; Ranking D1; ranking n° 6 of 98 in health care sciences & services]

CHAPTER 2

Van Rickstal, R., De Vleminck, A., Morrison, S. R., Koopmans, R. T., van der Steen, J. T., Engelborghs, S., Neugroschl, J., Aldridge, M. D., Sano, M., & Van den Block, L. (2020). Comparing Advance Care Planning in Young-Onset Dementia in the USA vs Belgium: Challenges Partly Related to Societal Context. *Journal of the American Medical Directors Association*, *21*(6), 851–857. doi: 10.1016/j.jamda.2020.01.007. [2018 SCI impact factor 4.899; Ranking Q1; ranking n° 6 of 53 in geriatrics & gerontology]

CHAPTER 3

Van Rickstal, R., De Vleminck, A., Engelborghs, S., & Van den Block, L. (2023). Experiences with and perspectives on advance care planning in young- and late- onset dementia: A focus group study with physicians from various disciplines. *Frontiers in aging neuroscience*, *15*, 1130642. https://doi.org/10.3389/fnagi.2023.1130642 [2021 IF 5.702; Ranking Q1; ranking 16/109 neurosciences in SCIE edition]

CHAPTER 4

Van Rickstal, R., De Vleminck, A., Engelborghs, S., Versijpt, J., & Van den Block, L. (2022). A qualitative study with people with young-onset dementia and their family caregivers on advance care planning: A holistic, flexible, and relational approach is recommended. *Palliative medicine*, *36*(6), 964–975. doi: 10.1177/02692163221090385. [2020 IF 4.762; Ranking Q1; ranking 18/108 in health care sciences & services in SCIE edition]

CHAPTER 5

Van Rickstal, R., De Vleminck, A., Chambaere, K., & Van den Block, L. (2022). People with young-onset dementia and their caregivers discussing euthanasia: a qualitative analysis of their consideration. Submitted.

PART I

GENERAL INTRODUCTION

General background

Palliative care is defined by the World Health Organization as "an approach that improves the quality of life of patients and their families facing the problems associated with life-threatening illness, through the prevention and relief of suffering by means of early identification and impeccable assessment and treatment of pain and other problems, physical, psychosocial and spiritual"[1]. Given that dementia has a progressive trajectory with no current treatments to halt or reverse the condition, the European Association of Palliative Care emphasizes that a palliative care approach is recommended[2]. According to experts in a Delphi study by the European Association for Palliative Care, one of the highest research priorities related to palliative care in dementia is advance care planning (ACP)[2]. ACP is a process of communication between patients, family and professional caregivers to explore patients' preferences for future (medical) care, including care at the end of life[3]. The overarching goal of such communication process is to increase the chance that patients receive care that is consistent with their wishes and values, even at times when they no longer have decisional capacity and are unable to express or make their own choices[4]. Regardless of recent discussions regarding ACP, and more specifically the research resources invested in it[5,6], it is argued that ACP can be especially relevant in dementia as the disease eventually precludes people from making their own treatment decisions[7]. A precondition for appropriate care for people with dementia is efficient and timely communication between patients and their caregivers[8]. In case of Alzheimer's disease, diagnosis is possible during the stage of mild cognitive impairment. This broadened timeframe for planning care, resulting from earlier diagnosis, increases the opportunity for and the importance of ACP[9]. Providing the opportunity for ACP is regarded as an essential element of sound palliative care[10] and of ethically responsible long-term care after a diagnosis of dementia[11].

The history and conceptual evolution of ACP

The concept of ACP arose in the United States of America (USA) in the late 1960s and started as a movement for establishing advance directives, also known as living wills, as legal guiding mechanisms for patients to refuse treatment[12]. Back then, ACP focused mainly on documenting an agreement between patient and physicians in an advance directive[3].

Following these developments in the USA, other countries gradually introduced advance directives, and as such, for the first time allowed persons to specify wishes for medical treatment prior to losing cognitive capacity to do so. However, the pitfalls of focusing exclusively on documenting preferences became clear, and as such, the importance of communication was emphasized[13].

In recent times the recognition has grown that solely completing documents is not likely to be sufficient for enabling people to receive the care they wish for[3]. ACP has by now broadened to a process of communication which can result in, but doesn't merely consist of, documenting care preferences. The conceptual evolution that ACP has undergone over time, and particularly in recent years, has shifted the focus to a process of ongoing support in communication and shared decision making, which also helps prepare people for "in the moment decision making" if necessary[6,14]. A review from 2021 on ACP intervention studies showed that solely 4% of recent, as opposed to older, studies equated ACP to advance directive completion[15]. However, this review simultaneously showed that there is no unity within the clinical and scientific field on what exactly constitutes ACP: studies vary in terms of ACP definitions, the aspects of ACP they target and the outcome types they put forward[15].

Three laws defining the legal framework for ACP in Belgium

The conceptualization or implementation of ACP differs between countries, partly due to different legal frameworks across nations and/or a nation's states. Physician-assisted dying laws (such as euthanasia and physician-assisted suicide) are globally established, considered and debated[16,17]. Euthanasia is the act in which a health care practitioner intentionally ends a patient's life by active drug administration at this patient's explicit request[18]. The debate regarding physician-assisted dying typically focusses on two conditions that are usually put forward for making physician-assisted dying justifiable, namely the presence of unbearable and irremediable suffering on the one hand, and a patient's competent request to end his/her life on the other[19]. Both these justifications might be challenged when a person with dementia makes a request for euthanasia[19].

Belgium has a specific medico-legal context for end-of-life care. Three laws, all passed in 2002, reflect an increase of patient autonomy as a societal value[20]: the laws on patient rights[21], on palliative care[22] and on euthanasia[18] shape the Belgian legal framework of ACP. Under Belgian law, people can complete several types of written advance directives: it is possible to refuse certain medical treatments or interventions for the event one would lose decisional capacity[20], and it is possible to make an advance euthanasia request for the event one would be in an irreversible vegetative state[18]. Additionally, it is possible to appoint a durable power of attorney to be a person's surrogate in decision-making[20].

The Belgian euthanasia law puts forward several substantive criteria: one must suffer unbearably from an incurable condition, without any prospect of improvement, and must express a current, well-considered, voluntary and repeated request for euthanasia[18]. These conditions make that in Belgium people with dementia are eligible for euthanasia in those stages of their condition where they still have cognitive competence to voice a current, oral request. The ongoing public and political debate in Belgium focuses on the legal status of an advance euthanasia directive for patients in advanced stages of dementia, where it might be difficult to substantiate a former request[23].

Potential high relevance, yet low uptake of ACP in dementia

Although relevant for all people with a chronic condition, ACP can be particularly relevant for persons with dementia[11]. Alzheimer's disease and related dementias comprise of a severe set of neurological disorders which are characterized by cognitive impairment and memory loss[24]. The progressive accumulation of disability will cause deterioration in several cognitive domains and will lead to interference with daily functioning[25]. In general, dementia leaves people with a relatively long timeframe of diminishing cognitive function and loss of ability to self-manage care, since it usually progresses slowly over many years. Nevertheless, at some point patients may quickly lose decision-making capacity[26]. Particularly towards the end of patients' life, caregivers often find themselves making medical decisions on behalf of patients, without patients' preferences having been discussed. Such decision-making has been shown to elicit distress in family caregivers and does not always lead to decisions in accordance with patients' wishes[27]. Providing people with dementia the opportunity to engage in ACP could provide them with a chance to adequately prepare for the future at a time they are still capable to do so[28].

Despite consensus of its possible significance in dementia, the average uptake of ACP is particularly low in this patient population[29]. Globally, it is estimated that less than 40% of persons living with dementia engage in ACP[30]. The most recent numbers, dating back to 2012, from a nationwide study in Belgium, indicate that patients with dementia who had died, had discussed treatment preferences with their general practitioner in only 7.8% of cases and that legal representatives were appointed in only 10.8% of cases. Moreover, this study indicated that several end-of-life care issues (such as diagnosis and prognosis) and preferences (such as options for palliative care) were discussed significantly less with patients who had dementia compared to patients without dementia[31]. Research focussing on nursing home residents in Flanders, Belgium shows that written advance directives were present in just 13.4% of residents with moderate dementia and in 8.4% of those with severe dementia[10].

ACP in dementia is shown to be challenging

It is indicated that there are specific challenges related to ACP engagement in dementia, as research showed that having dementia, when compared to other conditions, is negatively associated with discussing preferences for treatment[32]. Studies show that physicians lack the confidence, skills and knowledge about how to initiate ACP in practice[33], or how to explain ACP to their patients with dementia[4]. They are uncertain about how to share prognostic information[34], such as the terminal nature of dementia, how to sensitively ascertain and respond to patients' information preferences[35], and how to avoid or manage patients' potential feelings of anxiety[36]. The World Alzheimer Report of 2016 concludes that physicians, despite acknowledging the value of ACP, struggle with its implementation in practice, among others, because of moral concerns (fear to upset people they care for) and because of communication challenges during interaction with patients with dementia and their caregivers[33]. Evidence on ways of optimally communicating about ACP, is both wanted and needed by clinicians[37,38,39].

Prevalence, clinical characteristics and social features of YOD

Nonetheless dementia is typically associated with older age, up to nine percent of people with dementia is estimated to suffer from the young-onset variant, meaning that persons develop symptoms before the age of 65 years old[40]. Recently a systematic review and meta-analysis shed more accurate light on estimates of prevalence and showed the importance of YOD worldwide, as well as the need to organize sufficient care for this population. It is estimated that globally 3.9 million people live with YOD, with an estimated age-standardized prevalence of 119 per 100 000 population[41].

As with late-onset dementia, neurodegenerative disorders are the most common cause of dementia in YOD[42,43,44]. Dementia of the Alzheimer type is the most common dementia subtype in both young- and late-onset dementia, but proportionally has a higher prevalence within late-onset dementia[42,44]. In addition to behavioral changes and decreasing cognition, Alzheimer's disease is characterized by a progressive deterioration in performing activities of daily living which are required for independent living and personal maintenance[45]. Anosognosia, a patient's unawareness of a neurological deficit, is a frequent phenomenon in Alzheimer's dementia and, for people with Alzheimer's dementia, most commonly applies to deficits in activities of daily life and behavioral changes[46].

The stage and the subtype of dementia, as well as the patient's disease-awareness are related to certain neuropsychiatric symptoms. Apathy and agitation are most common in YOD and are suggested to increase more in the disease trajectory of YOD when compared to late-onset dementia[47]. In nursing home residents, higher rates of apathy were found in YOD than in late-onset dementia, irrespective of dementia subtype[48]. However, when comparing YOD and late-onset dementia, findings point to lower neuropsychiatric symptoms overall in YOD, at least in a community dwelling population[48]. A study comparing quality of life in people with young- vs. late-onset dementia, found that the group of people younger than 65, to which the authors referred as very young YOD, experienced less quality of life than their older peers. More specifically these young patients were more sad, less emotionally calm and content, more fearful and agitated and they more frequently suffered from malnutrition[49].

The basis for YOD's age cut-off is found in psychosocial rather than in neurobiological reasons: for instance, the age of 65 years old signals a boundary between a life phase of labor and of retirement[43]. Early-onset dementia, as YOD is sometimes also referred to, might affect people in their prime of life. Younger persons with dementia are often still working, may have significant financial commitments, might have children living at home and might themselves be caregivers for their parents[50,51,52]. Additionally, it is suggested that this phase of life leads to more preserved disease awareness in younger persons with dementia: as their environment is more demanding, the loss of roles and status related to the condition is clearer in comparison to older people with dementia[53].

The lack of evidence-based insights for ACP in the context of YOD

Based on their phase of life, it has been hypothesized that younger patients and their family caregivers have distinct preferences and needs in terms of palliative care and specifically in terms of ACP: they are suggested to desire more autonomy and to request more involvement in the decision-making process[54]. Despite this hypothesis, the views and experiences of these patients in terms of ACP remain uncovered. Given that family caregivers face a high likelihood of having to make difficult care decisions for their loved one with dementia, it is recommended to involve them in ACP as early as possible[55,56]. Nonetheless, in current literature, also the perspectives of family caregivers of people with YOD remain poorly understood.

The development of a clinical guideline on ACP in dementia clarified that available evidence is very limited; recommendations for ACP in dementia are rooted in studies of low quality and expert consensus[55], a problem that was also highlighted in the World Alzheimer Report[33]. Additionally, studies inquiring physicians regarding their perspectives on and experiences with ACP in dementia focus exclusively on older people with dementia, and mainly include general practitioners[7,34,57]. Evidence-based best practice guidelines for ACP in dementia are in any way scarce and limited[55], and for YOD they are simply ‘undiscovered territory’. The specific population of people with YOD and their family caregivers remain an underrepresented group in (end-of-life care) research[54]. Moreover, studies including people with YOD themselves are nearly non-existent.

Necessary gaps to address

- Gathering an in-depth understanding of patients’ and caregivers’ insights and views is indispensable for conceptualizing ACP in a manner that is adjusted and accommodated to them. It is of utmost importance to optimally identify if, how and why people with YOD and their family caregivers possibly wish to engage in ACP.

- In the light of an internationally evolving landscape for physician-assisted dying legislation, it is informative to shed light on the views of people with YOD and their caregivers regarding euthanasia.
- Inquiring physicians from various disciplines that are crucial in dementia care regarding their perspectives on ACP, with a focus toward both young- and late-onset dementia, is needed to gain a more inclusive understanding.

Aims and research questions of the current doctoral dissertation:

PART I aims to identify the experiences with, the perspectives on and the preferences for ACP according to both family as well as professional caregivers of persons with YOD

CHAPTER 1: To explore - from the perspective of family caregivers - (1) the extent to which patients, family and/or professional caregivers had already engaged in ACP and the reasons for doing so or not and (2) their preferences for how to ideally engage in ACP with the patient, family and/or professional caregiver(s).
CHAPTER 2: To explore - from the perspective of family caregivers – (1) the engagement in and the conceptualization of ACP of family caregivers of persons with YOD and (2) to inquire which potential similarities and differences exist in this area between American and Belgian persons with YOD and their family caregivers.
CHAPTER 3: To inquire physicians from various disciplines about their experiences with and perspectives on discussing ACP with people with young- and/or late-onset dementia.

PART II aims to identify the experiences with, the perspectives on and the preferences for ACP according to people living with young-onset Alzheimer’s disease and their family caregivers

CHAPTER 4: To explore the experiences with and the views on advance care planning of people with young-onset Alzheimer’s disease and their respective family caregivers.
CHAPTER 5: To describe what people with young-onset Alzheimer’s disease and their family caregivers spontaneously express regarding euthanasia and communication about euthanasia when discussing the broader topic of ACP.

Methods

Due to the exploratory and sensitive nature of our research topic, a qualitative approach was deemed most appropriate throughout this doctoral study[29]. Four out of 5 chapters in the current dissertation are studies based on individual interviews with patients and/or family caregivers (Chapter 1, 2, 4, 5), one is a study based on focus group interviews with physicians (Chapter 3). Below we outline the commonalities in methodology for the individual interview studies, while also addressing the specificities per chapter. Subsequently, we will discuss the methodology of our focus group study. Lastly, we will describe the method of constant comparative analysis, which was used for analyzing the data of all chapters.

Individual interview studies

In-depth, semi-structured interviews were most suitable, since they are based on an interview guide developed by the researchers yet provide room for participants to elaborate on unanticipated topics.

All respondents were recruited via organizations or physicians. American family caregivers were enrolled by their loved-one's treating physician. In Belgium, we had four intermediate persons helping with recruitment: two neurologists, a coordinator of a day care centre for people with YOD, and the founder of a non-profit organization that organizes activities for people with YOD and their families. Eligible respondents were approached by them, were briefly explained about the study and were asked for permission to give their contact information to the researchers. Participants were then contacted by phone or email (by RVR or ADV), addressing any questions and ascertaining willingness to take part in the study. Conversely to the interviews with American family caregivers (Chapter 2) which took place over the phone, each interview with Belgian respondents (including people with YOD) was conducted face-to-face. A mutually convenient time was agreed upon to conduct the interview, and Belgian respondents could choose a location of their preference. With respondents' consent, the interviews were audio-recorded to allow for *ad verbatim* transcription.

People with dementia who wished to participate in an interview, had to meet six inclusion criteria:

1. Being formally diagnosed with young-onset probable Alzheimer's disease (based on strictly applied standard diagnostic criteria: e.g. NIA-AA criteria)
2. Having a score of 16 or more on the MMSE
3. Being diagnosed for 6 months or more (for reasons of sensitivity to grieving stage after diagnosis)
4. Being 18 years of age or older
5. Speaking Dutch
6. Signing written informed consent (themselves + family caregiver's consent as witness is required)

Interviews were standardly conducted individually, yet as an ethical safeguard, persons YOD dementia were given the possibility of having their caregiver present if it made them feel more comfortable.

Family caregivers were defined as 'persons who play a crucial role in the life and care of patients and who are co-responsible in decision making'[58] and had to meet four inclusion criteria for participation:

1. Being the primary caregivers of a person formally diagnosed with probable young-onset dementia
2. Being 18 years of age or older
3. Speaking Dutch (for American respondents: speaking English)
4. Signing written informed consent

For family caregivers who were recruited as dyads, namely together with their loved-one with dementia (Chapter 4), the criterium for signing written informed consent was supplemented with the criterium of also providing written informed consent for the patient's participation (as a witness).

Overall, we have included 48 respondents for individual semi-structured interviews. Of those, 38 were family caregivers (of which 25 Belgian and 13 American) of people with YOD and 10 were people living with YOD. Of our 38 family caregivers, 32 were spouses, 2 were siblings and 4 were adult children of patients. For more details on participant characteristics, please see the individual Chapters.

Chapter 1	15 Belgian family caregivers
Chapter 2	*Caregivers of chapter 1* + 13 American family caregivers
Chapter 4	10 people with YOD + 10 of their respective family caregivers
Chapter 5	*Caregivers of chapter 1 & patients and caregivers of chapter 4*

Ethics

For our interviews with family caregivers (Chapter 1 & 2), studies were approved by the Ethics Commission of the Brussels University Hospital (B.U.N. 143201732034) as the central commission and the Institutional Review Board of Hospital Network Antwerp (ZNA, EC approval no 4939) and of Hospitals GasthuisZusters Antwerp (GZA, 170407ACADEM) as local commissions. For Chapter 2, approval was additionally obtained by the Institutional Review Board of Icahn School of Medicine at Mount Sinai, New York (GCO# 16-1046), for the study with American family caregivers. For interviews with people with YOD and their family caregivers, ethics approval was obtained from the University Hospital Brussels (B.U.N. 143201939497) as the central commission and from Hospital Network Antwerp (ZNA, approval n°5208) and Gasthuiszusters Antwerp (GZA, approval n°190304ACADEM) as local commissions. Our secondary analysis described in Chapter 5, was in line with the initial ethics approval for Chapters 1 and Chapter 4.

For interviewing persons with young-onset Alzheimer's disease (as described in Chapter 4), additional measures were taken throughout recruitment to ensure ethically sound inclusion:

1. Potential participants are informed about the study by an intermediate person
2. After giving consent to the intermediate person for sharing contact information, potential respondents were contacted by RVR
3. If people expressed interest in participating, a copy of the informed consent form was sent by mail in order to timely provide people with all relevant information

4. If someone other than a physician initially recruited the patient, respondents were asked permission for the first author to contact the patient's treating physician (ascertaining inclusion criteria 1/2/3 for people with YOD)
5. If respondents chose to participate, a date, time and place of their choice was agreed upon for data-collection
6. Before the actual interviews, people with YOD were asked a short yes- or no- survey to guarantee that they were familiar with the study and their rights therein *(the survey is included in the current dissertation in Appendix of Chapter 4)*
7. Prior to starting the interview, written informed consent needed to be provided. For people with YOD, we adhered to a double consent procedure: caregivers were asked to also sign the informed consent form of their loved-one with dementia, as a "witness" that patients were well informed about the study and their rights throughout the interview, and that they voluntarily and consciously chose to participate.

Focus group study

The qualitative research design most suitable for exploring physicians' perspectives was that of a focus groups, as this approach allows for open discussion and interaction between participants[59]. We recruited 21 physicians through a personal email (sent by RVR) or through a general recruitment mail spread within several organizations (Belgian Dementia Council, and the Flemish Associations for Psychiatry, Geriatrics and Neurology). After physicians expressed their willingness to participate, they were sent a doodle in which they could indicate suitable moments for the focus group to take place. Online focus groups[60] were chosen due to Covid-19 regulations at the time and for minimizing participation burden for already challenged physicians. We strived for heterogeneous focus group composition in terms of physicians' background as this increases the range of positions taken on a specific subject[61].

An interview guide consisting of open-ended questions was developed within the research team. Each focus group was moderated and observed by two researchers (four by RVR & ADV, one by RVR & LVdB). They were conducted in Dutch, video- and audiotaped with participants' consent and subsequently transcribed verbatim. Researchers reached consensus that data-saturation had been reached by the fifth focus group.

Ethics

Ethics approval was obtained from the University Hospital Brussels (B.U.N. 143201939497) as central commission and from Hospital Network Antwerp (ZNA, approval n°5208) and Gasthuiszusters Antwerp (GZA, approval n°190304ACADEM) as local commissions.

Data analysis of the interviews and focus groups

All audio-recordings of the individual interviews were transcribed verbatim by the first author, those of the focus groups by a transcription agency. Within an iterative process of line-by-line reviewing, we assigned a descriptive code to a discrete text-fragment representing a certain idea or concept. Such codes were then grouped in concepts of a higher hierarchy based on their similarities or differences. In the last phase, these concepts were gathered in a limited number of overarching themes, ultimately resulting in a 'coding tree'. Transcripts, or minimally a subset of transcripts were reviewed and coded independently by two researchers (RVR and ADV). The construction of coding trees was always done collaboratively until consensus was reached. As such, our work method corresponds to a way of thematic analysis, and specifically the constant comparative method. As stated previously by Braun and Clarke[62,63], thematic analysis can be best thought of as a family of or an umbrella term for a set of approaches for the analysis of qualitative data that have a shared focus on developing themes *(patterns of meaning)* from qualitative data. In the broadest sense, every form of qualitative analysis involves considering whether a specific extract of data is, for example, best coded with this or that code, or is evidence of this or that process, etc. As such, the 'idea' encapsulated by constant comparative analysis is core in good qualitative practice generally[62]. However, as a name and as a systematized technique, constant comparative analysis is specifically associated to Grounded Theory. Our constant comparative method was not aimed at developing a theory (as is the case in grounded theory), rather our goal was to identify patterns in data, to describe them, and to provide an interpretation/ explanation of those patterns and their mutual relationships[62,64]. In doing so, we were inspired by a guide that offers support in qualitative data-analysis and which was -in turn- inspired by the constant comparative analysis of the Grounded Theory Approach[64]. Not all steps/ procedures within this guide were incorporated in our work process, for instance, no analytic software was used due to the personal preference of the researcher (RVR).

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PART II

IDENTIFYING THE EXPERIENCES WITH, PERSPECTIVES ON AND PREFERENCES FOR ACP IN YOD OF FAMILY CAREGIVERS AND PHYSICIANS

CHAPTER 1

Limited engagement in, yet clear preferences for advance care planning in young-onset dementia: An exploratory interview-study with family caregivers

Van Rickstal, R., De Vleminck, A., Aldridge, M. D., Morrison, S. R., Koopmans, R. T., van der Steen, J. T., Engelborghs, S., & Van den Block, L. (2019).

Published in: *Palliative medicine*, *33*(9), 1166–1175.
https://doi.org/10.1177/0269216319864777

ABSTRACT

Background:
The significance of advance care planning in dementia is widely acknowledged. Despite the suggestion that younger people with dementia and their family might have distinct needs and preferences in this area, studies on advance care planning in young-onset dementia are absent.

Aim:
We aim to explore 1) whether family caregiver(s) had already engaged in advance care planning with patients and/ or professionals and the accompanying reasons, and 2) family caregivers' preferences for how to ideally engage in the process with patients, family and professionals.

Design:
A qualitative study was conducted: we analyzed semi-structured interviews (n=15) through the method of constant comparative analysis.

Setting/ participants:
We included Flemish family caregivers of persons with young-onset dementia.

Results:
Plans for the future typically concerned non-medical affairs. Participants' limited engagement in advance care planning was clarified through several reasons: not considering it useful, hindering patient behaviour, adopting a day-to-day attitude, caregivers emotionally protecting themselves and uncertainty about patients' cognitive competence. However, endorsement for advance care planning showed from respondents' preferences: it should be initiated timely, by a third party and emphasize patients' remaining capacities. Lastly, the need for information and high-quality care emerged.

Conclusions:
A gap of knowledge, of information, and in care intertwiningly hinder advance care planning. In young-onset dementia, engaging in advance care planning is not an option equally accessible as not doing so. Policy makers, institutions and professionals could reflect on their responsibility in providing these patients and caregivers an actual choice to engage in advance care planning or not.

INTRODUCTION

Alzheimer's Disease and related dementias are a severe set of neurological disorders characterized by memory loss and cognitive impairment [1]. Deterioration in several cognitive domains, due to the progressive accumulation of disability, leads to interference with daily functioning [2]. At present, there is no available treatment to halt or reverse underlying pathologies [1]. Although typically associated with older adults, six to nine percent develop the condition before the age of 65. Such young-onset dementia, also referred to as early-onset dementia, affects approximately 54.0 per 100 000 people between ages 30 to 64 [3]. Young-onset dementia may affect people in their prime of life: patients may have children living at home, significant financial commitments, are often still working and might themselves be family caregivers for their parents [4,5,6].

Advance care planning is defined as a process of communication that supports adults at any age or stage of health in understanding and sharing their personal values, life goals, and preferences regarding future care, including end-of-life care [7]. The aim of advance care planning is to help ensure people receive care that is consistent with their wishes, including at times when they have lost decisional capacity [8]. Advance care planning is particularly applicable for persons with dementia given the certainty of cognitive decline and impaired decision making that accompanies disease progression. The concept of advance care planning has broadened from the completion of advance directives to refuse medical treatments to a dynamic and continuous process that consists of communicating values and preferences for care. Both discussions and documentation are important within advance care planning [7]. By Belgian law two types of advance directives can be completed: one to refuse medical interventions if people are unable to decide for themselves and one to request euthanasia in case of irreversible coma [9,10].

A recent clinical review on communication between health care professionals and persons living with serious illness [11], identified advance care planning as a research priority that could substantially improve practice, increase involvement of people with dementia and contribute to the quality of patient-centered care. Additionally, advance care planning is seen as one of the highest research priorities related to palliative care in dementia according to experts in a Delphi study by the European Association for Palliative Care [12]. The importance of advance care planning for persons with dementia was also emphasized by the World Health Organization (WHO) [13]. However, existing guidelines and recommendations regarding advance care planning for people with dementia are based on research concerning older patients, low-quality studies and expert consensus [14]. Evidence for best practice advance care planning for people with dementia is generally limited and scarce, and for people with young-onset dementia it is simply ‘undiscovered territory’. Nevertheless, it has been suggested that people with young-onset dementia might have distinct needs and preferences for advance care planning based on their active phase of life, desire for more autonomy and a more active role in decision-making [15]. Perspectives of their family caregivers are also poorly understood, despite the previously highlighted importance of involving them in the advance care planning process as early as possible [16]. For clinical practice, it is of utmost importance to optimally inquire if, how, and for which reasons people with young-onset dementia and their family caregivers want to engage in advance care planning. therefore, this study explored - from the perspective of family caregivers - 1) the extent to which patients, family and/ or professional caregivers had already engaged in advance care planning and the reasons for doing so or not and 2) their preferences for how to ideally engage in advance care planning with the patient, family and/or professional caregiver(s).

METHODS

Study design

Qualitative data were collected through face-to-face semi-structured interviews.

Participants

We purposefully sampled a heterogeneous sample of family caregivers of people with young-onset dementia in terms of how much time had passed since diagnosis (including bereaved caregivers). Family caregivers were defined as ‘persons who play a crucial role in the life and care of patients and who are co-responsible in decision-making’ [17]. Eligible respondents needed to be: 1) the main family caregiver of a person formally diagnosed with young-onset dementia (regardless of dementia subtype), 2) aged 18 or older, 3) Dutch-speaking and 4) needed to have given written informed consent to participate.

Recruitment

Participants were recruited from three Flemish sites: 1) a day care centre (De Toren); 2) a volunteer organization (Het Ventiel), both specifically aimed at people with young-onset dementia and their families, and 3) the Department of Neurology's Memory Clinic at Hospital Network of Antwerp (ZNA, Middelheim & Hoge Beuken). Coordinators at each site approached eligible subjects, asking for participation and for permission to give their contact information to the researchers. Participants were contacted by phone or email by RVR or ADV, addressing any questions and ascertaining willingness to participate. After agreement, interviews were scheduled at a mutually convenient time, at a place of respondents' choice. No potential respondent contacted by the researchers refused participation.

Data collection

All interviews were conducted face-to-face by two investigators (RVR, ADV), audio-taped and transcribed verbatim. An interview guide was developed and reviewed within the research team *(see figure 1)*. Since we applied a 'broad' conceptualization of advance care planning, we (also) used the term 'planning for the future' in the topic guide.

Figure 1: Interview Guide

Knowledge of and information received from professionals or gathered themselves about:

- Diagnosis and disease trajectory of young-onset dementia
- Advance care planning*

Previous engagement in planning for the future (extent, manner, reason):

- Plans for the future (in general)
- Advance care planning and planning for (medical) care
    - ° Documentation of advance care planning plans?

Preferences for how to 'ideally' engage in advance care planning

- Person to initiate advance care planning
- Timing for advance care planning
- Content of advance care planning

*If respondents were not familiar with the concept, advance care planning was explained to them at the beginning of the interview as: 'a process of communication that supports adults at any age or stage of health in understanding and sharing their personal values, life goals, and preferences regarding future care, including end-of-life care[7]. The goal of advance care planning is to help ensure people receive care that is consistent with their values, goals and preferences, including at times when they lose decisional capacity and are no longer able to decide for themselves[8]'

Data analysis

Despite our heterogeneous sample, 15 interviews with an average duration of 60 minutes allowed us to reach data-saturation, as agreed upon by consensus between RVR and ADV. We used the constant comparative method of qualitative data analysis to develop and implement consistent and comprehensive coding of open-ended data [18]: transcripts were reviewed line-by-line and discrete sections of text representing a given idea or concept were assigned a code. Codes were compared both within and across interviews and overarching themes were identified. All transcripts were independently reviewed and coded by two investigators (RVR, ADV) who met regularly to discuss the coding structure.

Ethics

Ethics approval was granted by the Medical Ethics Commission of the Brussels University Hospital (B.U.N. 143201732034) and the Institutional Review Board of Hospital Network Antwerp (ZNA, EC approval no 4939). Every participant gave written informed consent prior to participation. All obtained data were anonymized.

RESULTS

Participant characteristics

We performed 15 interviews with family caregivers of 15 different persons with young-onset dementia: patients' wives, husbands, daughters and brother. For details on patients' and caregivers' characteristics, see Table 1.

Table 1. SOCIO-DEMOGRAPHIC CHARACTERISTICS SAMPLE (N=15 family caregivers)

Respondents are family caregivers (right column) of persons diagnosed with young-onset dementia (left column)

	Patient characteristics		**Caregiver characteristics**	
	Frequency	Percentage	Frequency	Percentage
YEARS OF AGE	*age at diagnosis*		*age at interview*	
35 - 39	/	/	1	7
40 – 44	/	/	/	/
45 - 49	1	7	1	7
50 - 54	1	7	1	7
55 - 59	3	20	4	27
60 - 64	10	66	2	13
65 - 69	/	/	4	27
70 - 74	/	/	2	13
GENDER				
Female	8	53	9	60
Male	7	47	6	40
NATIONALITY				
Belgian	15	100	15	100
EDUCATION				
Primary	1	7	/	/
Secondary	4	27	2	13
Post-secondary non-tertiary	3	20	3	20
Tertiary	7	47	10	66
Undergraduate/ bachelor	*3*	*20*	*5*	*33*
Graduate/ master	*4*	*27*	*5*	*33*
RELATIONSHIP CAREGIVER TO PATIENT				
Wife			7	47
Husband			5	33
Brother			1	7
Daughter			2	13
LIVING SITUATION PATIENT				
Home	10	67		
With day-care	*4*	*27*		
Residential care	3	20		
Deceased	2	13		

Themes

The results that emerged from our analysis were categorized under two overarching themes: A) the extent to which respondents had engaged in planning for the future and the accompanying reasons, B) respondents' preferences for how to engage in advance care planning.

A. Extent of engagement in planning for the future and accompanying reasons

A1) The current conception of advance care planning

Plans perceived by caregivers as elements of advance care planning

During the interviews, caregivers initially focused on non-medical elements of planning for the future. For instance, they described engaging in financial planning, organizing the living situation and planning for after death, sometimes with the patients. Nevertheless, respondents also paid a lot of attention to specific end-of-life decisions during the interviews.

Euthanasia

One major topic that was spontaneously addressed by respondents during interviews concerns euthanasia. Of the eight patients who had ever expressed consideration of euthanasia, most mentioned this right after receiving a formal diagnosis of young-onset dementia as ‘the only way out’. Some patients mentioned the possibility of euthanasia during periods of distress, but after a while, when patients’ mood improved this idea faded away. Caregivers indicated reluctance to readdress the topic themselves for fear of distressing the patient. Most patients discussed the topic with a physician. Sometimes, the issue was addressed only (briefly) once, either because the patient did not want to discuss it any further or because physicians were, according to respondents, not inclined to discuss it in-depth.

Interestingly, several caregivers shared their thoughts on euthanasia during interviews even if patients never mentioned the topic. *“Thinking ahead like ‘now I* [patient] *have this disease so this means I will no longer be able to do this and that’ . . . According to me, he was actually never occupied with this. That’s why, in my opinion, he also never thought of things like euthanasia.”* (wife, 65y).

The completion of advance directives

According to their family caregivers, four patients had completed some advance directive. Six patients did not document any wishes and three caregivers stated not knowing. Remarkably, some caregivers who were unfamiliar with the term advance care planning, nevertheless indicated to have completed an advance directive with the patient. Patients completed these documents either before or after their diagnosis. When advance directives were completed before, re-evaluation after diagnosis did not take place because of patients’ (perceived) cognitive deficiencies or was deemed unnecessary since preferences were already known. In only half of the cases completing an advance directive resulted from dialogue: two patients discussed the content of the advance directive with their family caregivers of whom one also with the treating neurologist.

A2) Reasons for not discussing advance care planning

All respondents who had not undertaken advance care planning, the majority, gave multiple reasons for not doing so.

Not considering advance care planning significant or useful

Several caregivers felt that advance care planning would not change the situation at hand. Some elaborated on the unchangeable, irreversible nature of dementia and thus didn't consider engaging in conversations about future care as an added value, among others, as one simply needed to accept the situation. *"As yet, there's little planning for the future. And yes, we're… We're really still in an adjustment phase for now… also still a little of an acceptance phase. I'm not completely done with that yet. Although I have to accept it, there's nothing to do about it. I can't change it, I can't improve it"* (husband, 70y).

Patient behaviour hindering discussions

Some behavioural signs by the patient hindered advance care planning: e.g. handing over decision-making responsibility to caregivers, denying the situation or "superficially" reacting to discussions regarding future care. *"He* [patient] *brushed it off very easily when you would start about that* [future care]*. He didn't want to, he actually didn't want to discuss it. Yes, so, then it's difficult"* (wife, 65y). Multiple caregivers, however, described exhibiting similar behaviours*:* "*actually I'm very good at acting as if nothing's going on and just continuing living life*" (wife, 57y).

Adopting a day-to-day attitude

A first element to adopt a day-to-day attitude and focus strongly on the present, was the unfavourable nature of the future. *"Of course, it's also something* [progression of disease] *that you… I think, keep at a distance because… at that time, you're not that sick and… and you don't really want to think about that time, maybe"* (wife, 65y). Some caregivers mentioned fear of distressing patients when initiating advance care planning: *"You want them* [patients] *to enjoy some things. . . To give them a bunch of additional worries, you don't do that"* (wife, 54y). Similarly, several caregivers discussed difficulties of communicating freely to professionals about the (future) problems they face when patients are present.

One caregiver explained that adopting a day-to-day attitude was also based on preventing anxiety for herself: "*I have to stay calm, I have to keep my composure. A little more time. . . And then step by step.*" (wife, 59y).

Another factor contributing to focussing on the present, was shifting boundaries throughout the disease trajectory. *"You* [family caregiver] *think 'I'll never be able to deal with that'. But you're in it, you grow along and all of a sudden you're there* [stage of disease previously feared] *and it goes smoothly. . . That's also the case for people with young-onset dementia. You shift your boundaries"* (wife, 54y).

Also influencing such attitude, was the timing and acute manner in which problems were addressed. *"We'll only deal with the problem when the problem occurs. Whilst often, that will be too late"* (wife, 59y). Respondents felt problems were usually so unpredictable that it was difficult – or not useful- to prepare for them. "*That's* [unpredictability] *why we* [caregiver and patient] *deliberately chose to live day by day, do what we can and we'll see*" (wife, 54y).

Caregivers' need for self-protection

Some respondents feared that initiating or participating in advance care planning, and particularly euthanasia discussions, would cause them to feel guilty. One respondent explained how he felt like an "accomplice" after his sister received euthanasia. ". . . *You start to realise, to wonder, 'why have I done this? Why have I not tried to convince her* [patient] *to not do it* [euthanasia] *in the end*?" (brother, 65y). Another was asked by the patient to remind him of his euthanasia request in case his memory would lapse. Nevertheless, she mentioned: *"afterwards, you still have to be able to live with yourself, so I don't know if I would"* (wife, 57y).

On the other hand, advance care planning was perceived as a means for self-protection by some caregivers as knowing the patient's preferences could diminish emotional burden. As such, some expressed regret for not having discussed advance care planning. *"It would provide more peace of mind if you* [caregiver] *knew that this is his* [patient's] *wish*" (wife, 65y).

Questioning patients' cognitive capacity for advance care planning

Some caregivers feared or had experienced the patient making wrong assumptions about discussions: "*And what did mom* [patient] *eventually take from this entire conversation* [about future care] *'you're going to have me locked up'* " (daughter, 47y). On the other hand, the advantage of not having insight to one's own condition was mentioned: difficult decisions become easier for caregivers when patients no longer understand them. *". . . if he has to go somewhere* [residential care]*, I will find it devastating. I hope by then he doesn't realize it any longer. That would make it a lot easier for me."* (wife, 65y).

B. Preferences for advance care planning in young-onset dementia

B1) Who, when and what?

Who?

All respondents who shared thoughts on the ideal person to initiate advance care planning, named a third party and mainly physicians, because for relatives themselves, it could be hard to assume this role. "*It* [caregivers mentioning ADs] *looks like you're* [caregivers] *giving up on them* [patients] *a little*" (wife, 65y). However, physicians' attitudes also played a role: respondents distinguished between honest and empathic physicians versus those who didn't seem to see the patient as a person. One person felt a peer, or a "buddy", would be best suited to introduce advance care planning to persons with young-onset dementia. ". . . *because if a physician says it* [what to possibly plan for in the future], *it's immediately so definitive*" (wife, 54y).

When?

The ideal timing for initiating advance care planning was perceived slightly differently by several caregivers. The majority felt that initiation should take place timely: *"The sooner* [after diagnosis] *these things happen, the better for him* [patient] *because well, you don't know to* *what extent, how fast it* [patient's condition] *deteriorates"* (wife, 69y). *"If you wait too long… then it might be too late"* (husband, 70y). However, some also noted the necessity of allowing the patient time to process the diagnosis. "*I don't think that that* [discussing the future] *is possible in the beginning. Because one, one first has to…. well, be able to give it* [diagnosis] *a place"* (wife, 65y). Other recommendations entailed establishing a trusting relationship between patient and physician prior to advance care planning and repeatedly discussing preferences instead of just once.

What?

Some caregivers indicated that advance directives could be a means to start a broader conversation about future care to emphasize patients' remaining abilities, rather than emphasize their acquired limitations. Lastly, several mentioned an "individual approach" is of great importance.

B2) The need for information as a first step towards advance care planning

About disease trajectory

Five respondents explicitly stated they -including the patient- were not or barely given any information about what to expect regarding disease progression and its implications, sometimes despite their explicit request. For the few that did receive this type of information, the majority added they did so only after specifically asking for it themselves. Most caregivers argued that uncertainty and unpredictability of an individual disease trajectory makes it very difficult for physicians to provide concrete information. Additionally, *"you can't just scatter all of people's hope at once"* (husband, 59y). Indeed, one caregiver stated too much information "would only make her depressed".

About advance care planning

Many respondents were unfamiliar with the term advance care planning or its purpose, with only a few being informed about it by their physician. Several respondents expressed the need to let people know "advance care planning exists" and the need to "explain advance care planning in a simple manner".

About services

Unravelling information about services for persons with young-onset dementia was a major challenge for caregivers (and patients). Almost all respondents reported there is no clear overview of available services: they recommended that care options in young-onset dementia are clarified, centralized and made easily accessible. *"I mean, it's a fact, sometimes I'm completely tired of having to chase after everything* [in terms of care]" (wife, 55y).

B3) The need for high-quality care

The emphasis placed on a person-centred approach became evident from the recommended "individual approach" for advance care planning but was also expressed regarding the need for appropriate care services for people with young-onset dementia, accustomed to their age, activity level and clinical presentation. However, this was rarely the case. *"I had already visited several centres* [residential care facilities]. *But there are some that are truly horrible, I have to say. I was thinking 'you can't just put someone out like old trash this way?' Sitting in front of the television all day, laying in a sofa, playing bingo"* (wife, 70y). *"It* [service provision/ help] *is actually a hopeless situation"* (wife, 62y). More generally, caregivers stated that the options for care for people with young-onset dementia were far too limited. One respondent explained that the fewer options available in terms of care, the easier it was to make the "right" decision (and thus the less need for discussions). Another caregiver, rather cynically, stated that making the best choice was simplified by barely having options. The slow pace of service provision was also problematic. Additionally, (painful) dissimilarities in dementia vs. cancer care services were pointed out. One respondent spontaneously stated that considering persons with dementia as palliative patients ought to be more common practice, since this would ensure more and earlier care.

DISCUSSION AND CONCLUSION

Discussion

This is –to our knowledge- the first exploratory study to address engagement in and preferences for advance care planning in young-onset dementia from the perspective of family caregivers. Respondents in our study had limited engagement in advance care planning. Various barriers and needs clarified why advance care planning was not or could not be undertaken by persons with young-onset dementia and their family caregivers.
Throughout the interviews, the goal of protecting their loved ones surfaced as a profound factor in shaping the behaviour and decisions of family caregivers of people with young-onset dementia. Regarding advance care planning, the aim of safeguarding patients' state-of-mind prevented caregivers from initiating the process: they were afraid to upset the patient. Similarly, caregivers experienced significant difficulties in communicating openly to professionals in patients' presence. The protective role that caregivers of people with dementia take on towards patients, has been highlighted in prior research [19]. It is plausible that this protective role is even stronger in caregivers of people with young- as compared to caregivers of people with late-onset dementia, due to the interference of a dementia diagnosis with the life phase activities of this younger population, such as raising children, working and maintaining an active social life. Additionally, the stigma associated with (young-onset) dementia [13] might enhance the need to offer protection.

A surprising result was the need for family caregivers to not only protect patients but to also protect themselves from possibly long-lasting burdensome emotions. Interestingly, this concept was indicated both by a reluctance and desire to engage in advance care planning. Caregivers' efforts to avoid feelings of guilt led them to not initiate advance care planning. Conversely, caregivers explained a wish for advance care planning in that it would give them 'peace of mind' knowing the patient's preferences. As such, the needs to protect both their loved-ones and themselves seem intertwined and sometimes oppositional. In this context, it appears evident why caregivers name third parties, especially physicians, as ideally placed to initiate advance care planning, which is consistent with prior recommendations [16]. Our respondents also supported the recommendation [16] of timely advance care planning, wishing to ensure patients' ability to participate. Cognitive incompetence was indeed identified by respondents as a reason for not communicating about advance care planning: their recommendation of timely advance care planning might reflect caregivers' awareness of this 'missed opportunity'. Caregivers' protective role shows once again through another recommendation for advance care planning timing: a grieving period after diagnosis should be respected, as also shown in previous research [20].

A topic identified by respondents as specifically difficult to discuss with patients was euthanasia. In all but one interview euthanasia was raised spontaneously by caregivers, even when this was never discussed with patients. Specifically, patients' diagnosis made their caregivers form an opinion about euthanasia in young-onset dementia, regardless of whether they found this a desirable option. The possibility and even likelihood that specific medico-legal contexts influence existential decisions was recently suggested for people with amyotrophic lateral sclerosis [21]. Our study confirms that legislation might partially determine the framework in which people think about end-of-life decisions. As such, one could suggest that the legislation of euthanasia makes it an alternative that is taken into account almost 'naturally' or 'spontaneously' following a young-onset dementia diagnosis. However, this result might also be associated with the younger age of respondents and the patients discussed in this study. Prior research has indicated that persons in Belgium and the Netherlands who request euthanasia, tend to be younger (i.e. younger than 80 years old) [22]. It has been suggested that persons with young-onset dementia, in comparison to persons with late-onset dementia, place stronger emphasis on autonomy, possibly impacting end-of-life decisions [15]. In this regard, it is important to explicitly distinguish between persons' thoughts or considerations and persons' decisions or acts: research shows that in 2013 less than one percent of all reported euthanasia cases in Belgium involved persons diagnosed with dementia [23]. Future studies comparing different legal contexts and their impact on thought processes regarding end of life in distinct patient groups and across various generations are needed.

Our results from interviewing family caregivers underscore the central role of information in enabling people with young-onset dementia and their caregivers to engage in advance care planning: we found lack of information regarding the trajectory of young-onset dementia, advance care planning and available services to be particularly challenging. The often un-met needs for information [20,24,25,26,27] and for accustomed services to people with young-onset dementia and their caregivers [19,28,29,30] as highlighted in other research, was confirmed by our respondents and can be linked to other results in our study. For instance, not being explained which milestones one can expect throughout the disease trajectory, and as such coping with ‘uncertainty’, contributed to a day-to-day attitude in which (sometimes foreseeable) problems were dealt with only at the moment they actually occurred. Not addressing likely future problems (such as cognitive incompetence) might lead to the significance and benefits of advance care planning being unclear or down-played [31]. This suggests that it is paramount to adequately inform patients and caregivers about (general) expectations so they can prepare themselves if they wish so, as recommended by the EAPC taskforce on advance care planning for people with dementia [32]. On the other hand, prior research showed that focusing on the present is an effective coping strategy as it is associated with lower unmet needs, as opposed to worrying about the future [33]. Therefore, as also captured within an existing general guideline on advance care planning for people with dementia [16], we would recommend striving towards a balance between attention for both the present, and the future in the context of advance care planning specifically with younger persons with dementia and their family.

Such gap of information could potentially lead to a gap of care. This link between information and care is also suggested by the WHO in its recent global action plan: quality of care at the end of life should be improved by, among others, promoting awareness about advance care planning for all people living with dementia. Information provision is, more generally, regarded as a form of support to people with dementia and their carers [34]. However, as was both explained by our respondents and shown in previous research [35], the service provision that people with young-onset dementia receive is in itself poor. Despite information on advance care planning, these poor services might cause or amplify the perception of advance care planning as pointless [20]: engaging in the process would not meaningfully contribute to the care received since this care is flawed anyway. Our findings shed further light on other research showing that people with young-onset dementia and their caregivers barely use formal services [29], as these are scarce, difficult to access and insufficiently meet the needs specific to people who have dementia at a young age, and further support the plea [28] for age-appropriate specialist care for people with young-onset dementia and their caregivers. As the interviewed family caregivers in our study pointed out, care for persons with dementia at a younger age should take into account their distinct age, higher activity level and the specific clinical presentation of their condition. Part of this recommendation could be interpreted as yet another example of the strong protective role that family caregivers of persons with young-onset dementia take on: appropriate care tailored to younger patients with dementia should focus on their remaining abilities, and these might be found in other domains, for instance in physical possibilities, as compared to older persons with dementia. Our study highlights the importance of availability of high-quality care in the context of advance care planning in young-onset dementia, as quality of care might form a strong incentive to (meaningfully) plan for it and might substantially counter the perception of advance care planning as useless for people with young-onset dementia.

Strengths and limitations

This study holds several strengths. Interviewing 15 family caregivers with an average duration of 60 minutes, led to data saturation for our main themes. All interviews were conducted face-to-face, facilitating in-depth exploration. Additionally, a heterogeneous sample of respondents in terms of socio-demographic characteristics and in terms of their caregiving role (relationship to and disease stage of the patient) allowed us to grasp various perspectives. Nevertheless, this study also has several limitations. First, its homogeneity in terms of respondents' ethnicity, which calls for this study to be repeated against other backgrounds. Further, interviewing caregivers enabled us to learn about patients' views only indirectly. Future research on advance care planning for people with young-onset dementia should include patients themselves, valuing their unique experiential knowledge [36]. Since prior research showed that in older people with dementia decision trajectories differ for those with versus those without informal caregivers [17], it is important to include both younger patients who can and who can't rely on family caregivers. Additionally, future studies should inquire how our results from interviewing caregivers might be refined based on the subtype of young-onset dementia their loved-ones were diagnosed with, given that differing subtypes are characterized by differing clinical presentation.

Conclusion

By our family caregivers, the possibility for advance care planning in young-onset dementia is undoubtedly welcomed. Nevertheless, this endorsement -indicated throughout their reasons for not having undertaken advance care planning and their preferences if they were to engage in the process- can often not be put to practice.

We found that various advance care planning recommendations previously made for older people with dementia are equally applicable and desirable in young-onset dementia, as noted by family caregivers. However, some barriers and needs –such as lacking age-appropriate care- are specific to them and their caregivers. A gap of knowledge (what is advance care planning & what does dementia entail), a gap of information (how to engage in advance care planning & what to plan for), and a gap of care (low-quality young-onset dementia care services & a lack of advance care planning within usual young-onset dementia care) are intertwiningly hindering advance care planning engagement and maintain a cycle of deficiencies.

Practice Implications

This study shows that professional caregivers, institutions and policy-makers could reflect on the responsibility they hold in providing people with young-onset dementia and their caregivers a choice of whether or not to engage in advance care planning. At present, due to the personal and organisational barriers mentioned in our study, undertaking advance care planning is not an option equally accessible to them as not undertaking advance care planning.

KEY STATEMENTS

What is already known about the topic?

- The significance of advance care planning for people with dementia is widely acknowledged and highlighted
- The uptake of advance care planning in dementia is particularly low
- It has been hypothesized that people with young-onset dementia and their caregivers might have distinct needs and preferences for advance care planning, in comparison to their older peers

What does this paper add?

- The uptake of advance care planning is lacking or limited in young-onset dementia according to family caregivers
- Although family caregivers would welcome the possibility of advance care planning, the opportunity for them to actually engage in the process is complicated or hindered by several barriers
- A gap in knowledge about advance care planning and dementia, in information about the content and process of advance care planning, and a gap in the quality of available care are factors intertwiningly hindering such advance care planning undertaking for persons with young-onset dementia and their family caregivers

Implications for practice, theory or policy

- Adequate information provision (about disease trajectory, advance care planning & services) is a necessary first step for enabling advance care planning engagement in young-onset dementia
- High-quality care might form an incentive to engage in advance care planning in young-onset dementia
- Policy makers, institutions and professionals could reflect on the responsibility they hold in providing people with young-onset dementia and their family an actual opportunity or choice of undertaking advance care planning or not

Acknowledgements

Contributions of others who did not merit authorship but participated in the research:

The authors acknowledge the help of Mrs. Dian Thys (coordinator at day care centre ‘De Toren’ for people with YOUNG-ONSET DEMENTIA and their families, based in Antwerp, Belgium) and of Mrs. Gudrun Callewaert (founder of volunteer organization ‘Het Ventiel’ for people with YOUNG-ONSET DEMENTIA and their families, based in West-Flanders, Belgium). The authors also wish to genuinely thank the family caregivers for sharing their valuable insights.

Funding: This work was supported by the Research Foundation - Flanders (FWO, grant number 1S75919N (strategic basic pre-doctoral grant) and grant number 12V9718N).

Sponsor: No sponsor was involved in conducting this study.

Declarations of interest: none.

I confirm all patient/personal identifiers have been removed or disguised so the patient/ person(s) described are not identifiable and cannot be identified through the details of the story.

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CHAPTER 2

Comparing Advance Care Planning in Young-Onset Dementia in the USA vs Belgium: Challenges Partly Related to Societal Context

Van Rickstal, R., De Vleminck, A., Morrison, S. R., Koopmans, R. T., van der Steen, J. T., Engelborghs, S., Neugroschl, J., Aldridge, M. D., Sano, M., & Van den Block, L. (2020).

Published in: *Journal of the American Medical Directors Association*, *21*(6), 851–857.
https://doi.org/10.1016/j.jamda.2020.01.007

ABSTRACT

Objectives

Advance care planning in young-onset dementia largely remains a blind spot within current literature. This study aimed to explore the engagement in and the conceptualisation of advance care planning from the perspective of family caregivers of persons with young-onset dementia and to identify potential similarities and differences in this area between American and Belgian persons with young-onset dementia and their family caregivers.

Design

An exploratory qualitative study.

Setting and Participants

We purposively sampled adult family caregivers of persons with young-onset dementia: our respondents were 13 American and 15 Belgian caregivers with varying familial relationships to the patient.

Methods

We conducted 28 semi-structured interviews, using the same interview guide for American and Belgian respondents. Verbatim transcripts were analysed through the method of constant comparative analysis.

Results

Important similarities between American and Belgian respondents were: restricted knowledge of advance care planning, limited communication about advance directives, their recommendation for professionals to timely initiate advance care planning. Major differences were: attention paid to those end-of-life decisions depicted in legislature of their respective countries, American caregivers placed higher emphasis on financial planning than their Belgian peers, and in case of consulting professionals for advance directives American caregivers turned to lawyers, whereas Belgian caregivers relied on physicians.

Conclusions and Implications

Specific nuances and challenges in terms of advance care planning in young-onset dementia, arise from a particular societal and legal context on the one hand, and from patients' and caregivers' younger age on the other. Professionals' awareness of and responsiveness to these specificities could facilitate the advance care planning process. Based on our interpretation of results, several recommendations for practice and policy are made.

INTRODUCTION

Advance care planning (ACP) is a process of communication between patients, family and professional caregivers to explore patients' preferences for future (medical) care, including care at the end of life[1]. The overall aim of such communication process is to increase the chance that patients receive care that is consistent with their wishes and values, even at times when they have lost decisional capacity and are unable to make or express their own choices[2].

At the end of the 1960s, the concept of ACP began in the United States of America (USA) as a movement for establishing advance directives as legal guiding mechanisms to refuse treatment, also known as living wills[3]. At that time, documentation of an agreement between patient and physician in an advance directive document was the main focus of ACP[4]. At present, ACP has broadened to a process of communication which can result in, but no longer merely consists of documentation in advance directives[1,5].

The way in which ACP is conceptualized and implemented in practice differs between countries, partly because of different legal frameworks across nations and/or a nation's states. The USA currently knows a changing landscape in several states where physician assisted dying has been legalized or in which this option is politically and publicly debated[6,7]. Belgium has a specific medico-legal context for ACP. In 2002, three laws were passed in Belgium, all revolving around patient autonomy[8]: the laws on patient rights (including the right to refuse treatment)[9], on palliative care[10] and on euthanasia[11]. Both in Belgium[9] and in all 50 states of the USA and the District of Colombia[3] several types of health care directives are legalized: an instructional advance directive focussed on refusing treatment, known as a living will in the USA, and a directive to appoint a durable power of attorney to be patients' surrogate in decision-making. Additionally, Belgian law enables people to request euthanasia through an advance directive in the event they would end up in an irreversible persistent vegetative state[11].

Although relevant for all persons with a chronic illness, ACP is particularly important for persons with dementia, given that Alzheimer's disease and related disorders are characterized by progressive cognitive decline, ultimately resulting in decisional incapacity[12].

Dementia is typically associated with older age. However, in up to 9 % of all dementia cases the condition develops before the age of 65 years old, referred to by the term young-onset dementia[13]. The age limit of 65 is based on social, rather than on biological factors[14]; people with young-onset dementia often have young children and spouses, are often still active in the labour market and might have significant financial commitments[14,15]. Additionally, it is suggested that this phase of life leads to more preserved disease awareness for persons with young-onset dementia because their environment is more demanding which makes the loss of roles and status related to the disease clearer in comparison to older patients[16]. Based on their phase of life, it has been previously hypothesized that these younger patients and their family caregivers have distinct needs and preferences in terms of ACP, wanting more autonomy and requesting more involvement in the decision-making process[17]. It is generally recommended to involve family caregivers of persons with dementia as early as possible in the process of ACP[18], given they are likely to be faced with difficult care decisions for the patient at the end of life[19].

Nonetheless, the role, needs and preferences of family caregivers concerning ACP in young-onset dementia is currently unclear as, more generally, the topic of ACP in young-onset dementia remains poorly represented within existing literature[20,21]. Previous research showed that although Flemish family caregivers of persons with young-onset dementia would endorse engaging in ACP, several reasons keep them from actually doing so[22]. The substantial need to gain insights into how to tailor end-of-life care communication to the needs of its recipients[23], is consequently very tangible in this patient population. Additionally, it is unknown if the ideas, topics and preferences that arise during ACP communication in young-onset dementia vary in different societal contexts. Both the USA and Belgium are Western countries in which considerable attention has been given to ACP, yet both nations show significant differences in terms of the tradition of and the legal framework for ACP[3,6,7,8]. As such, comparison of those two nations potentially allows for detecting subtle nuances. As such, this study aimed to explore the engagement in and the conceptualisation of ACP from the perspective of family caregivers of persons with young-onset dementia and to inquire which potential similarities and differences exist in this area between American and Belgian persons with young-onset dementia and their family caregivers.

METHODS

Design

Given the exploratory, but also sensitive nature of our topic, a qualitative approach was chosen. Semi-structured interviews were most suitable since they standardize the interviews yet allow participants to touch on topics that were not anticipated by the researchers. Interviews with Belgian[22] and American caregivers were, respectively, all conducted face-to-face and over the phone. Data from the Belgian respondents led to a previous separate publication[22]. Data from the American caregivers were not the subject of any prior article.

Participants

Four eligibility criteria were put forward: respondents needed to be 1) the primary family caregiver of a person formally diagnosed with young-onset dementia, 2) at least 18 years old, 3) fluent in English or Dutch language (respectively for American and Belgian caregivers), and 4) needed to provide written informed consent prior to participation.

Data collection

Eligible respondents were approached by a physician or an involved health care professional working at specialist care settings that patients were enrolled in and were contacted by the researchers, after their consent. Interviews were scheduled at a mutually convenient time and, additionally in Flanders, at a location of respondents' choosing. Over an 18-month period, two researchers (RVR and ADV) conducted 15 interviews with Belgian and 13 interviews with American (New York) family caregivers which respectively lasted on average 60 and 46 minutes. The two researchers interviewed both Belgian and American caregivers, using the same interview guide for both groups (*see Figure 1*), which was developed and reviewed within the research team.

Figure 1. Summary of Interview Guide used for both Belgian and American caregivers

Knowledge of and information received from professionals or gathered themselves about: - Diagnosis and disease trajectory of young-onset dementia - ACP* (for Belgian respondents we used both the term 'advance care planning' and its Dutch translations *'voorafgaande of vroegtijdige zorgplanning'*)
Previous engagement in planning for the future (extent, manner, reason): - Plans for the future (in general)

- ACP and planning for (medical) care
    - ° Documentation of ACP plans?

Preferences for how to 'ideally' engage in ACP

- Person to initiate ACP
- Timing for ACP
- Content of ACP

*If respondents were not familiar with the concept, ACP was explained to them at the beginning of the interview as: 'a process of communication that supports adults at any age or stage of health in understanding and sharing their personal values, life goals, and preferences regarding future care, including end-of-life care[24]. The goal of ACP is to help ensure people receive care that is consistent with their values, goals and preferences, including at times when they lose decisional capacity and are no longer able to decide for themselves[2]'

Data analysis

All interviews were audio-recorded, transcribed verbatim and subsequently analysed through the method of constant comparative analysis[24]. This inductive approach allows for comparison within and between groups: through an iterative process codes are assigned to a discrete part of the transcript representing a certain idea or concept. Such codes are then grouped in concepts of a higher hierarchy based on their similarities or differences. In the last phase, these concepts are gathered in a limited number of overarching themes. For an example of how this method is applied, see Table 1. Coding and analysing were done independently by two researchers (RVR and ADV), who determined data saturation by consensus for the overarching themes described in the results. With regard to our Belgian data[22], the codes and concepts obtained through previous analysis were re-interpreted in the light of our comparative research question. Our American data were subjected to original analysis for this study.

Ethics

Ethics approval was granted by the Medical Ethics Commission of the Brussels University Hospital (B.U.N. 143201732034), the Institutional Review Board of Hospital Network Antwerp (ZNA, EC approval no 4939) and the Ethics Committee of GastHuisZusters Antwerpen (GZA, EC approval no. 170407ACADEM) for the study in Flanders and by the Institutional Review Board of Icahn School of Medicine at Mount Sinai, New York (GCO# 16-1046) for the study with American family caregivers.

Table 1. *Table to serve as demonstration of method of constant comparative analysis: Examples of instances of transcripts and accommodating codes to show how themes emerge.*

1. Ad verbatim transcript in which pieces of text are demarcated

Be Transcript C: That [general practitioner] is the most suited person, to talk about it [ACP] (1) and to say 'look, now that you are still able to, you know, judge objectively' (2)
Be Transcript E: I think for him [patient], the quicker these things happen [discussing ACP], the better for him (1), because you don't know to what extent, how fast it will decline (2)

US Transcript B: I think in the beginning of the disease [good moment to talk about ACP] (1). You know, while she could still be, you know involved in the decision making. (2)
US Transcript J: I have to decide all that [advance directives & DNR] for him (1) because he doesn't have the capability mentally to decide those things. (2)
US Transcript H: I think that when he got diagnosed, I think it [being informed about possible future decisions] probably would have been very helpful. (1)

2. Codes assigned to discrete sections of text representing a certain idea

Code C1: GP is most suited to talk about ACP
Code C2: GP should discuss ACP when patient is still able to "judge objectively"
Code E1: The quicker ACP takes place, the better for the patient
Code E2: Reason for E1= unpredictable speed of decline

Belgium

Code B1: "Beginning of disease" is good moment to talk about ACP
Code B2: Reason B1: patient can still be involved in decision making
Code J1: Caregiver has to decide on AD's and DNR
Code J2: Patient no longer has ability to decide on AD's and DNR (=> brother decides)
Code H1: information on future decisions after diagnosis would have been "helpful"

USA

3. Codes are grouped under concepts of a higher hierarchy

Preferences for the initiation of ACP as broader concept

Ideal person: Be code C1

Timing: Be code C2, US code B1, US code H1
Reason for preferred timing: Be code C2, Be code E2, US code B2, US code J2

Belgium & USA

4. Concepts are gathered under limited number of overarching themes

One similarity between American and Belgian caregivers in terms of ACP is:

They want to initiate ACP in time to enable the person with young-onset dementia to be involved in decision making

RESULTS

We had a sample of 28 participants with various familial relationships to the patient: 13 wives, 9 husbands, 2 siblings and 4 adult children. *Table 2* shows further details about our respondents. None of the persons who were contacted by the researchers declined participation or dropped out of the study. `

Table 2. Socio-demographic characteristics of sample (N=28 family caregivers)
Respondents are family caregivers (lower rows) of persons diagnosed with YOD (upper rows)

	American		**Belgian[22]**	
	Frequency	Percentage	Frequency	Percentage
PATIENT CHARACTERISTICS				
NATIONALITY				
American	13	100	/	/
Belgian	/	/	15	100
AGE AT DIAGNOSIS				
45 - 49	/	/	1	7
50 - 54	/	/	1	7
55 - 59	/	/	3	20
60 - 64	13	100	10	67
LIVING SITUATION PATIENT				
Home	4	31	10	67
With help/ day-care	*2*	*15*	*4*	*27*
Residential care	3	23	3	20
Deceased	6	46	2	13
GENDER				
Female	5	38	8	53
Male	8	62	7	47
CAREGIVER CHARACTERISTICS				
NATIONALITY				
American	13	100	/	/
Belgian	/	/	15	100
RELATIONSHIP CAREGIVER TO PATIENT				
Wife	6	46	7	47
Husband	4	31	5	33
Brother	1	8	1	7
Daughter	1	8	2	13
Son	1	8	/	/
GENDER				
Female	7	54	9	60
Male	6	46	6	40

When analysing the similarities and differences between American and Belgian family caregivers with regard to their engagement in and conceptualisation of ACP, we identified several subthemes. Three similarities were most striking: 1) their restricted knowledge of ACP, 2) the limited communication that took place with regard to advance directives, and 3) their recommendation for professionals timely initiating ACP. Three major differences also emerged: 1) attention to those end-of-life decisions encompassed by the different laws, 2) American caregivers placed higher emphasis on financial planning than Belgian respondents, 3) and Belgian caregivers relied on physicians, whilst American respondents relied on lawyers in case they communicated about advance directives with professionals.

Similarities:

1. **Restricted knowledge of the term ACP and its meaning**

Generally, we found that the familiarity of Belgian and American family caregivers with the term “advance care planning”, and its Dutch translations, was either limited or lacking. The majority of respondents were unaware of what ACP means or entails.

“Advance care planning? I don’t know, maybe in a different way. What do you mean by that?” (US caregiver, wife)

“I could imagine [what ACP is]*, but no... If you’re not working in that field...”* (Belgian caregiver, brother)

2. **Little communication about advance directives, even when confronted with dementia**

Unlike their Belgian peers, the majority of American caregivers indicated that they and the patients had completed living wills. However, neither groups seemed to have engaged in in-depth discussions regarding the patient’s future care when completing these advance directives. The lacking or limited nature of these ACP discussions goes for both the mutual communication between patients and family caregivers, as for their communication with healthcare professionals. Additionally, and mostly relating to American caregivers, a majority of these advance directives were in place prior to the patient’s diagnosis and were not re-evaluated after.

"We talked about the present, the present. We actually never talked about the future. Uhm… we had talked, before he had any impairment. We had discussed our end-of-life decisions and filed all the necessary papers." (US caregiver, wife)

". . . and we thought, we just better finish up all the details. Let's get the living will, let's get the do not resuscitate. You know all the things, you know, we're healthy and well. Let's do it now and let's just get organized." (US caregiver, wife)

3. Health care professionals timely initiating ACP is wanted

When asked about their opinion on how ACP should ideally take place in young-onset dementia, the vast majority of American and Belgian caregivers expressed that these discussions should preferably be initiated in a timely manner, meaning soon after diagnosis, and by a health care professional.

"That's an advice I'd give everyone "Do that [ACP] *as soon as possible"* (Belgian caregiver, wife).

According to most caregivers a physician would be best suited. One Belgian caregiver thought a peer or "buddy" would be ideal. By timely initiation caregivers wish to ensure their loved-ones are enabled to participate in discussions.

"In the beginning. . . I think that she might have been able to do some planning back then as opposed to now" (US caregiver, son).

Differences

1. Attention paid to specific end-of-life decisions that are encompassed in the law

None of the American caregivers, yet all but one Belgian caregiver who was interviewed raised the topic of euthanasia without any question or prompt from the researchers. Most of those Belgian caregivers spontaneously gave their opinion about the 'applicability' or 'desirability' of that option in their specific situation.

"The fact that it [euthanasia] *is a possibility, is already a relief for a lot of people. But that doesn't mean they will do it"* (Belgian caregiver, wife)

The Belgian caregivers touched upon the subject of euthanasia, regardless of whether or not they considered it a desirable option.

"Once you've accepted that you've got dementia, and that it is no longer reversible, well, then there's only one way out [refers to euthanasia]" (Belgian caregiver, brother)

"Because it's something completely different, to commit euthanasia on someone who, as a figure of speech, has an aggressive cancer of who you know is nearing the end of his life, than on someone who has neurological problems." (Belgian caregiver, husband)

Whether they were considered as options or not, the potential end-of-life decisions mostly discussed by American caregivers involved refusal or withdrawal of treatment and more rarely, decisions involving suicide or those that are not within the scope of the law.

"I was a very strong advocate for, whatever they call it these days, they always use euphemism, for self-deliverance. That's the euphemism now, for suicide. If anybody wants to kill themselves, that's fine with me, including myself. But when it comes to late life suicide in face of incurable disease I'm still very strongly in favor of that should the person want to." (US caregivers, wife)

"If I would get Alzheimer's disease, I would ask somebody to help me out of this life. I would not go through it. Having seen it, you know, I would try to find a way for someone to help me out." (US caregiver, husband)

"Listen, sometimes I'm thinking to make him [patient] *sleep for good. We* [patient's family] *were at that moment when we thought that way."* (US caregiver, wife)

Questioning and at times actively researching what is possible within the (state) legislation, was also more of an issue for some American than Belgian caregivers.

"I would like to know more about the details about end-of-life decisions and . . . about some legalities in the various states in the United States of the end-of-life decisions. And questioning the legality of many, many, . . . And what you can do, and what's legal, and what's not legal, and I… in my mind it's very confusing…" (US caregiver, husband)

2. American caregivers put more emphasis on financial planning than Belgian respondents

The patient receiving a young-onset dementia diagnosis prompted financial arrangements for many Belgian and American caregivers, and sometimes also patients themselves.

"When he got sick, I did go to the notary and then we [patient and herself] *arranged our* [financial] *will and made sure that our granddaughter would receive a part of what my daughter would receive."* (Belgian caregiver, wife)

However, financial planning was considerably more of an extensive and also alarming matter to American, as compared to Belgian, caregivers. Specifically, the costs of health care for the American patient, who this would be borne by, and health and long-term care insurance all arose as concerns.

"And then, now you wanna talk to a physician, which physician, who knows, are they gonna charge you to discuss it [disease trajectory]*?"* (US caregiver, husband)

"You've got to set it up so you can afford to take care of that person so that means getting them the insurance they need, because they can't do without insurance, and making sure that you're financially stable. Because if you're not financially stable and you're taking care of somebody else, that's not gonna work. So you've got to deal with their finance and your finance." (US caregiver, brother)

Providing financial security for patients was a substantial part of their caregiving task and was perceived by them as an important element of planning ahead in the context of care.

"Well yeah, he's on Medicare… So he has health coverage, so I have that in place." (US caregiver, brother)

"Yes, I had to buy health care insurance, long term health care insurance at the beginning because if anything happened to me, our money would go to me and not to her [patient] *. And I needed to make sure that there was money for her. So, I'm spending like a little under 12,000 dollars a year on long term care insurance myself which I may or may not need. But I have to. I have to have it."* (US caregiver, husband)

For Belgian caregivers, financial planning was also of importance, but usually concerned arrangements for after the patient's death (drawing a financial will) or for when patients would no longer have cognitive capacity to arrange their own finances.

"We've had to arrange all those things that we [patient's children] *have authorization of all bank accounts, because yeah, at a certain moment, if he's no longer cognitively competent, you're faced with that. So that's all taken care of."* (Belgian caregiver, daughter)

3. US caregivers consulted lawyers; Belgian caregivers consulted physicians for documents

We found that American –as opposed to Belgian- caregivers rely on professionals from non-medical domains for guidance in correctly completing living wills. Every American respondent had completed or discussed their advance directives with a legal professional (lawyer).

> *"That lawyer had us sign advance directives, but he was an estate lawyer."* (US caregiver, wife)

None of these caregivers reported that there had been any contact or co-operation between the consulted legal and the treating medical professionals.

> *"I don't know if a physician would really help with the end-of-life stuff because they're treating him* [patient] *now . . . So I discuss his health with him* [physician] *and I bring up issues with them* [physicians]*, but in terms of planning for the future* [refers to advance directives]*, that's more of a legal issue at this point."* (US caregiver, brother)

In the exceptional cases where Belgian patients and caregivers had discussed advance directives with a third party, they turned to healthcare professionals and specifically physicians.

DISCUSSION

Summary of the results

Throughout the interviews several similarities and differences between American and Belgian caregivers of persons with young-onset dementia emerged regarding engagement in and conceptualisation of ACP. Similarities were their limited knowledge of the term and the content of ACP, the lack of communication in case of completing an advance directive, and their desire for a professional to initiate ACP in a timely manner. Differences were respondents spontaneously paying attention to those end-of-life decisions that are encoded in the laws of their respective countries, Belgian caregivers emphasizing financial planning less than American respondents, and reliance on physicians by Belgian caregivers and on lawyers by American caregivers if professionals were consulted for advance directives.

Strengths and limitations of this study

An important strength of this study is that it provides novel insights into an ethically charged topic in an under-researched group, in varying societal and legal climates. Besides gathering narratives of people from both Belgium and the USA, we recruited respondents with varying familial relationships to the patient, striving for a range of perspectives. When possible, interviews were conducted face-to-face with the opportunity for researchers to be prompted by respondents' non-verbal behaviours. Although it could be considered a limitation that

American caregivers were interviewed over the phone, this could conversely also be deemed a strength since social desirability might decrease without face-to-face interaction. It seems possible that telephone interviews helped them feel comfortable enough to express certain thoughts, for instance regarding end-of-life decisions that are not legalized. Interviewing caregivers, however, leads to the limitation that patients' perspectives are only portrayed indirectly. Future research should therefore include persons with young-onset dementia themselves to inquire their unique experiential knowledge. Also, subsequent studies could provide additional meaningful insight through in-depth inquiry of the potential influence of legislation on ACP.

Interpretation of the results

The finding that both our respondent groups were rather unacquainted with ACP seems surprising when considering the societal and political attention that has been paid to end-of-life decisions in both countries[4,6,8]. As is put forward in the action plan 2017 – 2022 issued by the World Health Organization[13], information and quality of care could be linked in the sense that information might function as a form of support to persons with dementia and their caregivers. Lacking knowledge most likely attributable to lacking information, could partly explain our finding of low ACP engagement in both respondent groups. Specifically, despite the presence of legal statutes outlining ACP in each country, public education of these laws and their implication appears to be needed as policy and legal changes and debates in this area do not seem to co-occur with an enhanced understanding of the concept of ACP. Providing people with an adequate explanation about ACP, its possibilities and value is a necessary first step for enabling ACP in clinical practice. Although information on the topic might seem as an obvious prerequisite to engaging in ACP, a lack of knowledge of ACP was already reported as a barrier over 20 years ago[25]. This suggests that durably and repetitively identifying the need for information provision is not sufficient for establishing actual change. Like any other health care behaviour, ACP should be promoted as a public health message, targeted at the general public on the one hand and at professionals and health care students on the other[25,26].

Nonetheless our American respondents had more documents in place regarding future care, they too did not complete those within a process of discussion and exploration. As such, it can be concluded that ACP is still largely conceptualised as merely documentation, rather than as an ongoing process of communication. This in itself might impede patients' and caregivers' engagement in ACP, as a recent systematic review showed that people with dementia and their carers demonstrate higher willingness and readiness to undertake ACP if they perceived it as a flexible and ongoing discussion[27]. Professionals highlighting the dynamic characteristics of ACP communication is therefore recommended. Similar to our finding of restricted knowledge on ACP, the focus on documents can be seen as a discrepancy or at least delay between 'theoretical' societal changes (legislation and 'expert'/academic conceptualisation of ACP) on the one hand, and the implementation and perception of ACP in practice and in broader society on the other.

Importantly, our results show that respondents appear to think about end-of-life decisions within the framework that is depicted by the country's legal framework for these decisions. It seems that when a certain decision is made possible by legislature, this option is taken into account in the personal thought process on future (end-of-life) care. However, this does not equate to actual requests for these options. Distinguishing between our respondents' ideas and their acts is therefore of importance when interpreting this result. Conversely, several previous studies have shown that the medico-legal context does influence the actual behaviour of physicians[28,29]. More research is needed to inquire if and how the law might influence various stakeholders' thoughts, decisions and/ or behavior in the context of ACP and end-of-life care.

Our study showed that financial security, in the form of care insurance for the patient, might be perceived as an import element of ACP in young-onset dementia. Recent articles highlight the problems of dementia costs for families and underscore the current mismatch between dementia care needs, specifically at the end of life, and Medicare benefits[13,30,31]. Those who contribute most to dementia costs are family caregivers, both through 'out of pocket payments' and through unpaid hours spent caring for their loved-ones[31]. It seems plausible that these unpaid caregiving hours impact families facing young-onset dementia even more than those facing late-onset dementia, since younger caregivers more commonly have to cut back on paid work in order to provide care to their loved-ones. Moreover, it has been shown that the average time between diagnosis and enrolment in residential care is longer for people with young-onset dementia as opposed to people with late-onset dementia (respectively 9 vs. 4 years)[32] and that persons with young-onset dementia barely use formal services[33]. As such, one could assume the total time and therefore cost of unpaid caregiving provided by family members is higher when patients get diagnosed at a younger age. Additionally, finances and health care costs have been explicitly linked to (life-limiting) end-of-life decisions by both European and American organisations, respectively, the European Association for Palliative Care[34] and the American College of Physicians[7]. However, in this regard we should clarify that finances were a concern, rather than a basis for decision-making for our respondents. Additionally, it is plausible that the financial implications of young-onset dementia were clearer to American respondents as they more often cared for a person who had already gone through the disease trajectory and had passed away by the time of the interview.

Lastly, previous research in cancer has shown that completing, but not discussing advance directives with physicians is a longstanding problem[35]. Similarly, we found a disconnect for our American respondents who documented and discussed decisions regarding future medical care with legal professionals, yet did not discuss these wishes with medical professionals. This might indicate a sense of ‘form-over-substance’: making sure advance directives are filled out in the correct format is prioritized over discussing their actual care content, which might in turn jeopardize their meaning in clinical care. The ‘juridification of medical care’, signalling that legal discourse becomes more pronounced in medical practice, has been reported previously[28]. Our study suggests that this does not only influence medical professionals[28,29], but also affects caregivers and patients when handling their medical care. Remarkably, there was no continuity in care provided by the legal and the medical professional when our respondents completed living wills, which seems rather worrisome when considering the nature of these decisions. Pleas for co-operation between these professionals have been made in the past[36,37]. In the context of dementia, it seems all the more important that a physician is involved in the process given the progressive impact of the condition on cognitive functioning. Cognitive capacity is required in order to complete an advance directive and assessment of such capacity is a medical act.

Figure 2. Summary of recommendations for health care professionals and policy to facilitate ACP in YOD, according to our interpretation of their caregivers' perspectives

Based on the need for information and on caregivers' preference for professionals timely initiating the process, physicians should fulfil an active role in explaining and starting ACP conversations in YOD.

Health care professionals should explicitly ask people about potential worries regarding health care costs, as this topic might open up a broader conversation about (long-term) care.

Health care professionals should actively ask patients and caregivers about what they might have planned for already, with who and should offer further discussion if desired. This will facilitate follow-up between professionals from different domains (e.g. health care, law) and might increase the chance that completed documents have actual meaning in clinical care.

Health care professionals should be aware of the potential influence of legislation on the framework that people use to think about end-of-life decisions.

Thorough and accessible top-down information transfer about the concept and the legalities of ACP from governments to both professionals and practitioners and to the general public is necessary.

CONCLUSIONS AND IMPLICATIONS

Based on our interpretation of the results we were able to formulate several recommendations for practice and policy to facilitate ACP in young-onset dementia *(see Figure 2)*. Family caregivers of people with young-onset dementia are faced with multiple challenges. Comparing caregivers in the USA and in Belgium, we found that some aspects of their conceptualisation of and their engagement in ACP are shared and others are distinct. Differing emphasis placed by respondents themselves, can be traced back to their respective societal and legal context. Generic ACP guidelines for professionals are necessary and useful, nevertheless, and by no means wanting to culturally stereotype, it is important to take nuances between countries into account. Conclusively, this study shows that specific distinctions and challenges in terms of ACP in young-onset dementia, arise from a particular societal and legal context on the one hand, and appear to flow from patients' and caregivers' younger age on the other. Professionals' awareness of and responsiveness to this specificity could facilitate the ACP process in this population.

Funding:

This work was supported by the Research Foundation Flanders (Fonds voor Wetenschappelijk Onderzoek) through a pre-doctoral Strategic Basic Research Fellowship granted to Romy Van Rickstal (1S75919N), and through a post-doctoral fellowship granted to Aline De Vleminck (12V9718N). This study was also supported by a Fulbright Scholarship granted to De Vleminck (2015). The authors have no conflicts of interest to declare.

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CHAPTER 3

Experiences with and perspectives on advance care planning in young- and late- onset dementia: a focus group study with physicians from various disciplines

Van Rickstal, R., De Vleminck, A., Engelborghs, S. & Van den Block, L. (2023).

Published in: Frontiers in Aging Neuroscience, 15, 1130642.

https://doi.org/10.3389/fnagi.2023.1130642

ABSTRACT

Introduction

Despite the relevance of advance care planning for people with dementia, its uptake in this population is particularly low. Several challenges for advance care planning in dementia have been identified from physicians' perspectives. However, the literature available mainly includes general practitioners and focuses exclusively on the context of late-onset dementia. This is the first study to inquire physicians from four highly relevant specialisms in dementia care, with a focus towards potential specificities based on patients' age. The research question of this study is: "What are physicians' experiences with and perspectives on discussing advance care planning with people with young- and/or late-onset dementia?".

Method

Five online focus groups were conducted with 21 physicians (general practitioners, psychiatrists, neurologists and geriatricians) in Flanders, Belgium. Verbatim transcripts were analyzed through the qualitative method of constant comparative analysis.

Results

Physicians believed that the societal stigma related to dementia influences people's reaction to their diagnosis, at times characterized by catastrophic expectations for the future. In this regard, they explained that the topic of euthanasia is sometimes addressed by patients very early in the disease trajectory. Respondents paid ample attention to actual end-of-life decisions, including DNR directives, when discussing ACP in dementia. Physicians felt responsible for providing accurate information on both dementia as a condition, and the legal framework of end-of-life decisions. Most participants felt that patients' and caregivers' wish for ACP was more driven by who their personality than by their age. Nonetheless, physicians identified specificities for a younger dementia population in terms of ACP: they believed that ACP covered more domains of life than for older persons. A high consistency regarding the viewpoints of physicians from differing specialisms was noted.

Discussion

Physicians acknowledge the added value of advance care planning for people with dementia and especially their caregivers. However, they face several challenges for engaging in the process. Attending to specific needs in young-onset, in comparison to late-onset dementia, requires advance care planning to entail more than solely medical domains. However, a medicalized view on advance care planning still appears to be dominant in practice as opposed to its broader conceptualization in academia.

INTRODUCTION

Advance care planning (ACP) is defined as a process of communication between patients, family caregivers and professionals to explore patients' preferences for future (medical) care, including at the end of life (Sudore et al., 2017). The concept has evolved considerably over time, now focussing on an ongoing process that also helps prepare people for "in the moment decision making" when necessary, rather than focussing on the completion of advance directives (Van den Block, 2019 & Tishelman et al., 2021). In general, dementia leaves people with a relatively long timeframe of loss of ability to self-manage care and diminishing cognitive function (Gaster, Larson and Curtis, 2017). Despite ongoing discussions about the value of ACP (Tishelman et al., 2021), it is argued that ACP can be particularly relevant for people with dementia and their caregivers as the condition eventually precludes patients from taking part in their own treatment decisions (Alam et al., 2022). In case of Alzheimer's Disease, diagnosis can be made during stages of mild cognitive impairment. The larger timeframe for planning care, due to earlier diagnosis, increases the opportunity for and importance of ACP (Porsteinsson et al., 2021). Nonetheless, the uptake of ACP in dementia is low with less than 40% of patients worldwide undertaking ACP (Sellars et al., 2019). Research showed that having dementia, in comparison to other conditions, is negatively associated with discussing treatment preferences, indicating that there are certain specific challenges related to engaging in ACP in dementia (Evans et al., 2014).

Particularly in dementia, discussing future care is considered difficult due to uncertainties regarding the future and due to the jeopardized decisional capacity of people with dementia (Tilburgs et al., 2018a & Sellars et al., 2019). More specifically, a recent meta-review of systematic reviews and primary studies (Keijzer-van Laarhoven et al., 2020) showed that physicians feel responsible for providing high-quality end-of-life care to people with dementia but face moral dilemmas that may cause them to behave avoidantly towards initiating ACP. Among others, these dilemmas arise from not wanting to emotionally burden patients, trying to maintain hope, dealing with uncertainties in patients' prognoses and having ethical concern regarding patients' declining capacity (Keijzer-van Laarhoven et al., 2020). Fearing a shift in patients' preferences as the condition progresses was also identified as causing reluctance for physicians to make advance decisions with people with dementia (De Vleminck et al., 2014). Conversely, a qualitative study also found that beliefs about the perceived benefits of ACP can motivate physicians to engage people with dementia in the process, such as the belief that ACP would align patients', family caregivers' and clinicians' care goals (Alam et al., 2022).

For a more inclusive understanding of physicians' attitudes and challenges in terms of ACP in dementia, several physician specialties that are essential in dementia diagnosis and care should be inquired. Although there is literature available, these studies mainly include general practitioners and focus exclusively on the context of late-onset dementia (Tilburgs et al., 2018b & De Vleminck et al., 2014 & Alam et al., 2022). There is a dearth of studies that inquire physicians from various specialisms. Moreover, research in which physicians are questioned about their perspectives not only regarding late-onset, but also young-onset dementia (YOD) is absent. Globally, it is estimated that 370 000 people younger than 65 develop dementia symptoms before the age of 65 annually, defined by the term YOD (Hendriks et al., 2021). The very limited number of studies focussing on people with YOD and their family caregivers, showed that they barely engage in ACP, yet have clear preferences for how to do so (Van Rickstal et al., 2019). Among others, these include their wish for physicians to timely initiate and flexibly approach the process, provide accurate information and pay attention to more than only the medical aspects of care (Van Rickstal et al., 2019 & Van Rickstal et al., 2022).

To the best of our knowledge, this is the first inquiry of physicians from four highly relevant specialisms in dementia care (GP's, psychiatrists, neurologists and geriatricians) regarding ACP, with a specific interest towards the potential specificities depending on patients' age at diagnosis. The research question of this study is: "What are physicians' experiences with and perspectives on discussing ACP with people with young- and/or late-onset dementia?".

METHODS

Design

This exploratory study used the qualitative research method of focus groups, as this approach allows for open discussion and interaction between participants. Conducting focus groups online was necessary due to the Covid-regulations at the time yet was also an attempt to minimize participation burden for already challenged health care providers. In adherence with a recent guideline for virtual qualitative data-collection (Dos Santos Marques et al., 2021), the maximum participants per focus group was lowered (n=5) to facilitate in-depth discussion. This paper follows the COREQ-criteria for reporting qualitative research.

1. Introduction

Description of ACP provided by researchers:

> *"Advance care planning is a process of communication between patients, their family caregivers and professionals in which patients' views, values and preferences for future (medical) care are explored. This process should enable patients to help guide future decisions (also at those times when they are no longer able to make or express choices). ACP can, but does not necessarily, result in the documentation of wishes in advance directives"*

To what extent is this description similar to how you conceptualize ACP/ your understanding of the concept?

Throughout the following questions, respondents were systematically asked if there were any specificities in case of young- vs. late-onset dementia.

2. Experience with ACP

To what extent do you engage in ACP in your clinical practice?

If you engage in ACP with patients/family caregivers:
    Who usually initiates the communication?
    If at physician's initiative: How do you usually initiate ACP?

Is there, in your experience, **a right time** to initiate ACP?

Who is usually involved in ACP? (patients, family caregivers, other care professionals,…)

What are important **topics** to discuss within ACP?

Are there specific **hindering factors** when it comes to engaging in ACP in case of dementia?

3. Wish to engage in ACP from patients/caregivers

In your experience, to what extent do you feel there is a **need/ wish for ACP** from patients and their family caregivers?

What is the **added value** of engaging in ACP in dementia? Is there a difference in this value, in your perspective, for patients vs. for family caregivers?

Box 1. Focus Group Topic Guide.

Participants

To answer our research question, we aimed for a heterogeneous sample in terms of physicians' specialism within focus groups, to allow for in-depth insights. We included general practitioners, neurologists, psychiatrists and geriatricians as these specialties are crucial in the care for people with dementia. Physicians were purposively sampled through a personal email of the main researcher (RVR) or through a general recruitment mail spread within several organizations (Belgian Dementia Council, and the Flemish Associations for Psychiatry, Geriatrics and Neurology). After physicians expressed their willingness to participate, they were sent a doodle in which they could indicate suitable moments for the focus group to take place.

Data-collection

For these focus groups, an interview guide consisting of open-ended questions was developed within the research team (see Box1.). Participants were informed about some important 'ground rules' at the start of each focus group, such as no talking across each other, respecting confidentiality regarding others' participation, the content of discussions, etc. Each focus group was moderated and observed by two researchers (four by RVR & ADV, one by RVR & LVdB). The focus groups took place online through secured Zoom-meetings in November and December 2021. The focus groups were conducted in Dutch, were video- and audiotaped with participants' consent and were transcribed verbatim. After the fifth focus group, researchers reached consensus that data-saturation had been reached and no additional focus groups needed to be organized.

Data-analysis

Verbatim transcripts of the focus groups were analysed through the qualitative method of constant comparative analysis (Dierckx De Casterlé et al., 2012 & Hewitt-Taylor, 2001). In this inductive approach, a code is assigned to a certain idea or concept (usually one or two sentences). These codes are subsequently compared within and between transcripts, identifying broader themes or concepts. Two transcripts were read and coded in full independently by two researchers. After discussion and agreement on a coding structure, the remaining three transcripts were coded and analysed by RVR. Once coding was completed and codes were added to the coding framework, RVR and ADV together revised the transcripts and the obtained coding structure.

Ethics

The study was approved by the Ethics Committee of the University Hospital Brussels (B.U.N. 143201939497) as the central commission and by Hospital Network Antwerp (ZNA, approval n°5208) and GasthuisZusters Antwerp (GZA, 190304ACADEM) as local commissions. A signed informed consent was obtained by all participants prior to the start of the focus group.

RESULTS

The average duration of a focus group was 95 minutes. A total of 21 physicians took part in one of five focus groups (two n=5, two n=4, one n=3). Of these 21 physicians, five were general practitioners, three were specialised in psychiatry, six in neurology, and five in geriatrics. Except for one last-year neurology resident, all were board-certified specialists. Five women and 16 men participated.

Six major themes were identified from our data: 1) stigmatic image related to dementia as a specificity for ACP in this population, 2) physicians' focus on specific end-of-life decisions when discussing ACP in dementia, 3) physicians feeling responsible for providing information on dementia and on the law regarding end-of-life decisions, 4) the age of patients and caregivers as an influence on the content of ACP, 5) physicians seeing more benefits of ACP for family carers, and 6) congruency between medical professions. Several of our findings are generally related to dementia as a condition and can therefore be interpreted as applicable to both the young- and late-onset variant.

Stigmatic image related to the condition as a specificity for ACP in dementia

A factor that physicians believed to negatively influence patients' fears and concerns about the future, was the stigma related to dementia. In this regard they discussed how the popular media is at times responsible for diminishing nuances in people's image of dementia: the last phase of disease progression is portrayed as representative for the entire disease trajectory.

> *"That one quickly thinks that it's only about that last vegetative stage and that one would also end up there very soon etcetera. In the beginning, that's something*

> *that strongly traverses those conversations. . . One doesn't know that there are many years preceding that. . ." (FG 24, 138-140)*

Despite patients' initial expectations regarding their disease progression, physicians referred to people with dementia who, along the way, sometimes find their trajectory more manageable than initially expected. From their perspective, this posed a difficulty for engaging in ACP, since the evolution of patients' wishes was felt to be too unpredictable to offer guidance for future care decisions.

> *"If they say "I don't ever want to be in a wheelchair", or "I always want to be able to feed myself", or something like that, then eventually, when push comes to shove, they don't mind being wheeled around or they don't mind that they're being cooked for. So, it changes so much that it's not fully predictable." (FG23, 170)*

Some physicians explained that the "catastrophic" image of dementia at times caused patients to drastically react to receiving their diagnosis and that they, and especially younger patients, quite impulsively expressed a wish for euthanasia the moment of or soon after hearing their diagnosis.

> *"When disclosing the probable diagnosis or the results, people very often or at least several times show a catastrophic reaction and then they immediately start thinking about that last stage." (FG24, 146)*
>
> *"Yes, and with people with young-onset dementia. . . There are a few patients who at the moment of diagnosis nearly immediately say "okay, I have said that I want euthanasia in that case". (FG65, 89)*

This moment was said to be grasped by physicians as an opportunity for further exploration, explanation and broader discussion of preferences.

> *"If you then assess 'what motivates that (euthanasia) question?' or 'what is truly behind it?'… Then you actually arrive at a much broader framework of care planning that basically no longer entails what the initial question for euthanasia was, but more about care and planning and those things…" (FG24, 75/76)*

Physicians' focus on specific end-of-life decisions when discussing ACP in dementia

All participating physicians were familiar with the description of ACP provided at the beginning of the focus groups. However, it became apparent that physicians mostly elaborated on or re-directed the conversation to a specific aspect of ACP, namely to anticipatory end-of-life decision-making, such as DNR-orders (do not resuscitate) and euthanasia.

Physicians' perceived motives behind euthanasia requests

According to our participants, the request for euthanasia was usually a request for something else in terms of future care. In most cases, it turned out to be the patient's expression of a concern for which they sought guidance rather than an actual wish for euthanasia.

> *"In many cases it turns out that it (euthanasia request) is about other concerns that can easily be addressed in a different way and then the question disappears." (FG24,84)*
>
> *"Actually they are not asking for euthanasia, they are asking the question 'if I end up in circumstances that I don't find dignified, are you still going to help me?'" (FG43, 94)*

This was also explained by physicians through the motives on which they thought these patients' comments on euthanasia or euthanasia requests were based. Participants mentioned that these could stem from agitation about what the future will bring, unwillingness to move to a residential care facility and fear of the unknown.

> *"What is said frequently, is 'Yes, if I would have to go to a nursing home, then I'm done. I don't want to live like a vegetable. I've seen it with my mother or my father. Then, I would actually prefer euthanasia and I want you to write that down in my file like that'." (FR44,171)*

Physicians felt responsible for providing information on dementia and on the legal framework of end-of-life decisions

Many physicians also felt that media had contributed to both the public's awareness about euthanasia as an end-of-life option and had contributed to confusion about what is possible or impossible under Belgian law. Explaining patients about the legal framework was said to be an important task in clinical practice in terms of ACP.

> *"So, a big part of the time or a big part of the energy goes out to just explaining what's possible and what isn't possible." (FG44, 186)*

Additionally, it was mentioned that providing information (in terms of for instance law or prognosis) could function as a care intervention itself.

> *"I often notice that by discussing and explaining it (the legal framework) and by defining it, they sometimes find some peace already. That that request (euthanasia) sometimes stems from fear of the unknown and that informing them is at times already sufficient to find peace. That the questions then sometimes also fade away to the background." (FG 24, 72-74)*

According to our participants, patients tended to hold a "catastrophic" view of (young-onset) dementia, characterized by drastically declining functional and cognitive abilities. Driven by this alarming image, patients at times initiate ACP or euthanasia discussions according to physicians. In this regard, participants underscore a clear need for education in the sense of prognostic information.

> *"If we get the question (euthanasia), it's usually indeed a question for, yes… that has a whole lot to do with the stigma around dementia, I think. Many people regard someone with Alzheimer's disease as someone who sits in a wheelchair, drooling, in a nursing home, as a figure of speech. But of course that's not always the stage that everyone progresses to. So, I think that it's important to educate a bit in terms of what the possible patterns and expectations can be." (FG43, 101-104)*

In terms of discussing prognosis, physicians explained they typically use 'vague' terms and 'averages' when describing a patient's medical future. This manner of communication was based on both clinical uncertainty about the dementia trajectory according to participants, and physicians' wish to safeguard patient's hope and positive emotions.

> *"General terms are averages: but I try to avoid making individual predictions." (FG44, 109)*

> *"One of the biggest problems from my experience is that, often, we are also not honest towards our patients with dementia." (FG43, 171-172)*

Although patients' image of dementia might be "catastrophic" at times and in need of nuanced information, some physicians emphasized that one cannot deny the inevitable negative aspects when going through the entirety of a dementia trajectory. Participants felt that these aspects are difficult to disclose openly to patients.

> *"It doesn't always have to be as bad as dying drooling in a nursing home, but well, the cases in which the older man, the grumpy old man becomes the endearing father, those are less frequent than the other story" (FG43, 202)*
>
> *"If we take good care of them and place them in a decent nursing home, then they die of, well, what do they actually die of? Do they starve? Do they have a spontaneous fracture because they have been lying in bed for years?" (FG43, 176-177)*

They expressed that a longitudinal and trusting relationship between patient and physician increased their 'openness and honesty' in terms of disclosing prognostic information, for instance about the speed of disease progression or expected difficulties ahead.

> *"The way in which you get more concrete in terms of prognosis, that's also an advancing insight. After the diagnosis, the progression, the first two years. . . that always gives an indication of how quickly it could evolve." (FG65, 166-167)*
>
> *"And you don't name it with, yeah, terms that are hurtful, but yeah… sometimes we have known these people for years. Yes, then I dare to be honest about it (prognosis). I'm quite straightforward and the people who continue to come into consultations with me, are the ones who can tolerate that and even expect it." (FG43, 180-181)*

The age of patients and caregivers influences the content of ACP discussions

It was noted that both people with young-, as well as people with late-onset dementia are heterogeneous groups. The extent to which people wish to engage in ACP was generally regarded as connected to who the patient was as a person, rather than associated to the patient's age.

> *"There are people, both among younger patients, but also among older patients, who are very set on their autonomy and from that perspective can also be very verbal and have a clear request for ACP or other things. Just as well, there are younger patients who would rather avoid that type of conversation." (FG24, 98-99)*

Although some physicians said that younger vs. older people with dementia are usually more 'articulate', 'assertive' and 'have a higher need for control', the majority of physicians saw an equal amount of younger and older patients wanting to discuss ACP. However, they noted that the life context of younger people, with younger children and spouses, might make their questions about the end of life more salient.

> *"I can imagine that under those circumstances the questions about wishes for the end of life are much more prominently present and that one contemplates it much more at that age compared to at an older age. With these younger people, they (wishes for the end of life) will be brought up sooner or later." (FG65, 109-110)*

Several physicians talked about how YOD, in comparison to late-onset dementia, might lead to diminished acceptance of the diagnosis, higher grief and to more conflict within families, among others about financial matters.

> *"Older people already let go of life a bit more and accept that there they are at a high age at which things will end sooner or later." (FG65, 81)*
>
> *"There is also much more sadness of people with young-onset dementia, for so many good years lost." (FG65, 139)*

Respondents explained that caregivers at times had a higher wish to engage in ACP than patients. Examples provided were when patients did not grasp the implications of their condition, were no longer cognitively competent or when patients had expressed a death-wish to their caregiver, who wanted to discuss this further with the physician. Several physicians explained that during their consultations, caregivers of younger as opposed to older, people showed more tendency to bring up ACP.

> *"Of course I have people who have no illness awareness, and especially in that case that question will arise through the caregiver. Especially if there is no awareness of illness, then it all appears very 'far off' for the patient, and that can be difficult at times." (FG65, 57-58)*

> *“Yes, it regularly occurs that some type of death wish was expressed by the patient and that that actually is the impetus for the partner or the children to initiate that conversation. They often refer back to it like ‘you remember that you’ve said that, what do you actually mean by that?’.” (FG65, 64-65)*

Additionally, physicians explained that ACP discussions usually cover more domains in YOD due to the challenges the diagnosis brings along in multiple areas of patients’ and caregivers’ lives.

> *“Evidently, with younger people there is often the difficulty of the partner still working, that the children are still young, still studying, at times still living at home, which actually complicates it even more. . . Then that is a broader conversation, because it becomes even more difficult with caregivers themselves, that conversation.” (FG65, 135)*
>
> *“If there are children who are still young and who, just to give an example, become scared of their father or mother, or where their relationship changes entirely. . . Or a professional situation, people who are still working. You simply come across many more problems, which obliges one to consider at least a mid-long timeframe.” (FG44, 133-134)*

ACP was believed to especially benefit family caregivers

Several advantages of ACP engagement were discussed by physicians, for the majority relating to family caregivers. ACP was told to lead to an ‘emotional relief’, less conflict and less suffering since family caregivers were enabled to fulfil their need to provide care to their loved-one.

> *“And I think that for family it’s also very important to have that feeling like ‘we are doing well, we have done well’.” (FG24,33)*

Physicians believed that both patients and caregivers would assess the care provided as more positive, due to ACP.

> *“The bottom line is of course that people, the caregiver as well as the patient, will evaluate the care received more positively in the sense that they feel it is more closely aligned to what they wish.” (FG24, 23)*

From patients' perspective, physicians hypothesized that not wanting to be a burden to others might be a motivating factor ACP, aside from keeping their own best interest for the future in mind.

> *"By some (patients) it is indeed addressed that they somehow do it (ACP) for the caregivers, but it's not an 'or-or story', it's a combination of how they themselves feel about it." (FG65, 188)*

Patients' need and desire to take care of their family and ACP as a means to fulfil that need, was noted as well.

> *"That's also partly taking care of my children. That's drafting a care plan, so that my children know that it's okay what they do or not do with me." (FG43, 249)*

Congruency between medical professions

There were no divergent themes when comparing between physicians from differing specialties. Moreover, there appeared to be a consensus amongst respondents that general practitioners are usually able to play a key role in ACP, due to their usually longstanding relationship with the patient and his/ her family, and their professional context in which they are more likely to have frequent consultations with patients, possibly including home visits. It was noted that systematic sharing of ACP information between the various physicians involved in a patient's care was desirable, yet that such information flow was not sufficiently common.

DISCUSSION

Summary of results

This study shows that physicians believe that the societal stigma related to dementia impacts how people react to their diagnosis, including catastrophic expectations for their future. In this regard, they mentioned that the topic of euthanasia is at times addressed early in the disease trajectory by patients. Physicians themselves paid ample attention to actual end-of-life decisions, including DNR directives, when discussing ACP in dementia. As part of ACP, physicians felt it was their responsibility to provide accurate information on both dementia as a condition, and the legal framework of end-of-life decisions. Most participants felt that patients' and caregivers' wish for ACP was more driven by who they are as people than by their age. Physicians did identify specificities for a younger dementia population in terms of ACP: they believed that ACP covered more domains of life than for older persons. A high consistency regarding the viewpoints of physicians from differing specialisms was noted.

Strengths and limitations

The main strength of this study is that it assembled focus groups heterogeneously in terms of specialisms crucial in dementia care, allowing for in-depth insights from and for various medical disciplines. Our research question focused on people with late-onset, as well as with young-onset dementia. This led to findings that are insightful for clinicians, when caring for this underexposed group. A limitation of this study is that we did not observe actual practices, but analysed what respondents shared about these practices. Also, certain results might be less or not generalizable to other legal contexts besides those with physician- assisted dying laws. In this regard, however, we deem our results to be informative within the current internationally evolving landscape of physician-assisted dying legislation. Future comparative research in countries with varying legislative frameworks would be insightful for understanding the possible impact of the law on ACP and on ACP communication.

Interpretation of findings

Physicians explained that, at times, they struggled with disclosing prognostic information due to clinical uncertainty characteristic to dementia. The difficulty or even inability to provide accurate prognostic information experienced by our participants, has also been reported by patients and family caregivers in different countries (Sellars et al., 2019). It has been shown that patients and family caregivers felt a distrust towards clinician's mastery and knowledge of dementia (Groen-van de Ven et al., 2017). Physicians communicating openly to their patients about their uncertainty, might counter such feeling of distrust and contribute to a relationship of mutual confidence and trust. This could in turn facilitate ACP, as a sense of rapport was previously identified as a prerequisite for ACP in dementia by patients, their caregivers and general practitioners (Tilburgs et al., 2018b & Van Rickstal et al., 2019). Attending to not only patients' and caregivers' uncertainty in decision-making (Sellars et al., 2019), but also to that of physicians, might empower all parties when it comes to initiating ACP. Comparing our findings with existing literature, showed that there is an important commonality between barriers identified by professional caregivers on the one hand, and barriers identified by family caregivers and patients on the other.

Physicians explained that disclosing prognostic information might also be hampered by constraints they experience in openly and honestly communicating about disease progression. Although participants acknowledged that a dementia trajectory undeniably has negative elements, they simultaneously pleated for a more nuanced image of dementia, with a sometimes more steadily progression than expected or feared. Qualitative studies showed that people with dementia and their caregivers tend to oscillate between ‘wanting to know’ and ‘not wanting to know’ (Wawrziczny et al., 2016) and prefer to take it ‘one day at a time’ (Keijzer-van Laarhoven et al., 2020 & Van Rickstal et al., 2019). Additionally, people with late- and young-onset dementia and their caregivers have previously highlighted that, regardless of being diagnosed with dementia, there is still room for enjoyment (Dening et al., 2017 & Van Rickstal et al., 2019). Moreover, previous research showed that focussing on the present as opposed to worrying about the future, is associated with experiencing fewer unmet needs and therefore is an effective coping strategy (Millenaar et al., 2018). Having a realistic and truthful view on the future, yet also allowing hope and positivity to co-exist with this, appears a useful balancing act to be undertaken by all those involved when engaging in ACP. When placing our finding in the context of findings with patients and caregivers, it appears that physicians’ moral threshold to engaging in ACP, also described in previous research (Keijzer-van Laarhoven et al., 2020), is at times justified. According to participants, the societal negative image that is related to dementia increases the need for realistic information provision. Physicians in our study described how patients at times demonstrate catastrophic reactions to receiving a diagnosis, also based on the common, stigmatic image of dementia. If grasped by physicians, these reactions might function as a steppingstone to discuss ACP more broadly, according to them.

In the current study, ACP was considered by physicians to be a means or an opportunity for people with dementia of fulfilling a caretaking role towards their family. It has been previously stated by patients and caregivers that if people with dementia undertake ACP, one of their main purposes is to take care of their loved-ones (Van Rickstal et al., 2022). The relational, as opposed to purely individual, nature of ACP appears evident from the viewpoint of all parties involved. As such, particularly in the context of dementia, a family- rather than a solely patient-centred approached to ACP could be desirable. As physicians also expressed that their wish to safeguard patients' emotional wellbeing shapes their own behaviour in terms of ACP, the previous idea of a mutual protective role between people with dementia and their family caregiver (Van Rickstal et al., 2022) could be extended from a dyad to a protective triad which also includes the professional caregiver.

Physicians identified specificities for the content of ACP in YOD. The desire for ACP was put forward by our respondents as person- rather than age- and/or generation- related, depending on someone's personality. Nonetheless, several specificities in terms of age were mentioned. Young-onset dementia usually affects people in the prime of life, with possibly children still living at home, financial commitments, work, and at times caring for older relatives themselves (Withall, 2013 & Draper and Withall, 2016). According to our respondents, ACP in YOD was indeed considered to cover a broader range of domains due to the plurality of life-areas affected by the condition. Additionally, if the topic arose, it was told to be more prominently present in consultations with younger as opposed to older patients and caregivers. The general hypothesis that younger people with dementia and their caregivers have a higher need for ACP due to a higher wish for autonomy (Koopmans, van der Steen, Bakker, 2015), appears not to correspond with patients', family caregivers' (Van Rickstal et al., 2019 & Van Rickstal et al.,2022) or professionals' narratives. However, our former and current research shows that all parties do acknowledge that the content of ACP shows distinctions based on patients' younger vs. older age, mainly due to stage of life. Through insights of patients with YOD and their carers it was previously recommended to conceptualize ACP as holistic (Van Rickstal et al., 2022), consistent with respondents of the current study who explain that ACP in YOD can entail a broader range of topics. Overall consensus seems to exist that clinicians need to dedicate heightened attention to non-medical domains to adequately address ACP in this younger population. However, it was formerly shown that Flemish people with YOD and their carers spontaneously incorporate euthanasia in their thought framework on end of life (Van Rickstal et al., 2020), and as such, it can also be regarded as a sensitivity from our participating physicians towards their patients that they pay adequate attention to end-of-life decisions. It appears recommended to find a balance between broadening ACP to medical, social and relational domains (Tilburgs et al., 2018b & Van Rickstal et al., 2022), yet simultaneously elaborating on specific concerns patients have, such as euthanasia, if this were the case.

CONCLUSION

Overall, physicians acknowledge the benefits of ACP for people living with dementia and particularly for their family yet describe several challenges for actually engaging in the process. Some of these difficulties are related to dementia as a condition, others are associated with constraints for engaging in such conversations. Attending to specificities in terms of ACP for people with young-onset, compared to late-onset, requires physicians to pay attention towards non-medical domains. The finding that participants elaborated on actual end-of-life decisions, such as euthanasia and Do Not Resuscitate- directives, shows that the medicalized concept of ACP is still dominant in practice.

Statement of conflict

None of the authors have any conflicts to report.

Acknowledgements

The authors wish to sincerely thank all physicians who were willing to share their insights and experiences.

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PART III

IDENTIFYING THE EXPERIENCES WITH, PERSPECTIVES ON AND PREFERENCES FOR ACP OF PEOPLE LIVING WITH YOUNG-ONSET ALZHEIMER'S DISEASE AND THEIR FAMILY CAREGIVERS

Chapter 4

A qualitative study with people with young-onset dementia and their family caregivers on advance care planning: A holistic, flexible, and relational approach is recommended

Van Rickstal, R., Vleminck, A., Engelborghs, S., Versijpt, J., & Van den Block, L. (2022).

Published in: *Palliative medicine*, *36*(6), 964–975.
https://doi.org/10.1177/02692163221090385

ABSTRACT

Background

Broad consensus exists on the relevance of advance care planning in dementia. Although people with young-onset dementia and their family are hypothesized to have distinct needs and preferences in this area, they are hardly ever included in studies.

Aim

We aim to explore the experiences with and views on advance care planning of people with young-onset dementia and their family caregivers.

Design

A qualitative study was conducted, analyzing semi-structured interviews through the method of constant comparative analysis.

Setting/ participants

We included 10 people with young-onset dementia and 10 of their respective family caregivers in Flanders.

Results

Participants lacked awareness about the concept of advance care planning, especially as a communication process. They had not or barely engaged in planning future care yet pointed out possible benefits of doing so. Initially, people with young-onset dementia and their caregivers directly associated advance care planning with planning for the actual end of life. When discussing advance care planning as a communication process, they paid ample attention to non-medical aspects and did not distinguish between medical, mental and social health. Rather, respondents thought in the overarching framework of what is important to them now and in the future.

Conclusions

Engagement in advance care planning might be hindered if it is too medicalized and exclusively patient-centered. To accommodate advance care planning to people with young-onset dementia's and caregivers' needs, it should be presented and implemented as a holistic, flexible and relational communication process. Policy and practice recommendations are provided to do so.

INTRODUCTION

A precondition for appropriate care for people with dementia is efficient and timely communication between patients and their caregivers[1]. Providing an opportunity for advance care planning is regarded as an essential element of sound palliative care[2] and of ethically responsible long-term care after a diagnosis of dementia[3]. Advance care planning has been conceptualized as a process of communication between patients, family and professionals to explore patients' preferences for future (medical) care, including care at the end of life[4]. It has been 50 years since the concept of advance care planning arose as a movement for establishing advance directives as legal guiding mechanisms to refuse treatment[5]. Back then, documenting an agreement between patient and physician in an advance directive was the primary focus of advance care planning[6], centrally aimed to respect patient autonomy[7]. Advance care planning has conceptually evolved over time[8], and especially in recent years has undergone a shift in focus from documentation to a process of ongoing support in communication and shared decision making[9,10]. However, a recent review on advance care planning intervention studies showed the lack of unity within the scientific and clinical field on what constitutes advance care planning, as studies use varying definitions, target different aspects of advance care planning and put forward a large variety of outcome types[8]. Nonetheless, a review of 2021 showed that the evolved concept of advance care planning as a process has been adopted in recent, as opposed to older, studies with only 4% equating advance care planning to the completion of an advance directive[8].

The overall aim of advance care planning is to increase the chance that the care received is in accordance to the care desired, even at times when people are no longer able to make or express their own choices[11]. As such, advance care planning can be particularly important for people with dementia, given that Alzheimer's disease and related neurodegenerative disorders are characterized by progressive cognitive decline, which may ultimately result in decisional incapacity[3]. Since family caregivers are highly likely to be faced with difficult care decisions for their loved one, it is recommended to involve them in advance care planning as early as possible[12,13]. Despite consensus of its possible significance in dementia, the average uptake of advance care planning is particularly low in this patient population[14,15].

Although dementia is typically associated to older age, it is estimated that globally 3.9 million people live with young-onset dementia, implying their condition developed before the age of

65[16]. These younger patients and caregivers have been presumed to have distinct needs and preferences in terms of palliative care, and specifically in terms of advance care planning[17]. This hypothesis is based on their active phase of life, with significant social, financial and professional commitments[18, 19]. Nonetheless, this particular patient population and their family caregivers remain an underrepresented group in research. Moreover, studies that include people with young-onset dementia themselves are nearly non-existent. Yet, gathering a broader understanding of patients' insights and views is indispensable for conceptualizing advance care planning in a manner that is adjusted and accommodating to them.

This study's research question is: *"What are the experiences with and the views on advance care planning of people with young-onset dementia and their family caregivers?".* Through this study we wish to gather insights and formulate recommendations for policy and practice on what to take into consideration when initiating or engaging in advance care planning in young-onset dementia.

METHODS

Design

Given both the exploratory nature of our research question and the sensitivity of the topic, a qualitative study design was deemed most appropriate. Qualitative data were collected through face-to-face in-depth interviews, which were semi-structured for allowing respondents to elaborate on topics not prompted by the researcher. For reporting, COREQ guidelines were followed.

Setting

Respondents were recruited from two provinces in Flanders (Antwerp and West Flanders) and from Brussels Capital Regions by two neurologists, the coordinator of a day care center and the founder of a non-profit organization for people with young-onset dementia.

Population

Participants were recruited as dyads of a person formally diagnosed with young-onset dementia of the Alzheimer Type (symptom onset before the age of 65) and his/ her primary caregiver.

Recruitment

Inclusion criteria and phases within recruitment are described in detail in Table 1.

Inclusion Criteria for respondents (dyads of person with dementia & caregiver)
For **persons with dementia** (6): 1. Being formally diagnosed with young- or late-onset probable Alzheimer's disease (based on strictly applied standard diagnostic criteria; e.g. NIA-AA criteria) 2. Having a score of minimum 16 on the MMSE 3. Being diagnosed for at least 6 months (for reasons of sensitivity to grieving stage after diagnosis) 4. Being 18 years of age or older 5. Speaking Dutch 6. Signing written informed consent (themselves + caregivers' consent as witnesses is required)
For **family caregivers** of persons with dementia (4): 1. Being the primary caregiver of a person formally diagnosed with young- or late- onset probable Alzheimer's disease 2. Being 18 years of age or older 3. Speaking Dutch 4. Signing written informed consent (+ providing consent for person with dementia, as witness)
Steps within recruitment, with ethical safeguards throughout:
1. Potential respondents are informed about the study by intermediate person (neurologists/ coordinator day care centre/ founder of volunteering organization) 2. After giving consent to the intermediate person for sharing contact information, potential respondents were contacted by the first author (RVR) 3. If they expressed interest in participation, RVR sent potential respondents a copy of the informed consent form by mail in order to timely provide people with all relevant information 4. If initially recruited by someone other than a physician, respondents were asked permission for the first author to contact the patient's treating physician (ascertaining inclusion criteria 1/2/3 for patients) 5. If respondents decided to participate, a date, time and place of their choice was agreed upon with RVR for conducting the interviews

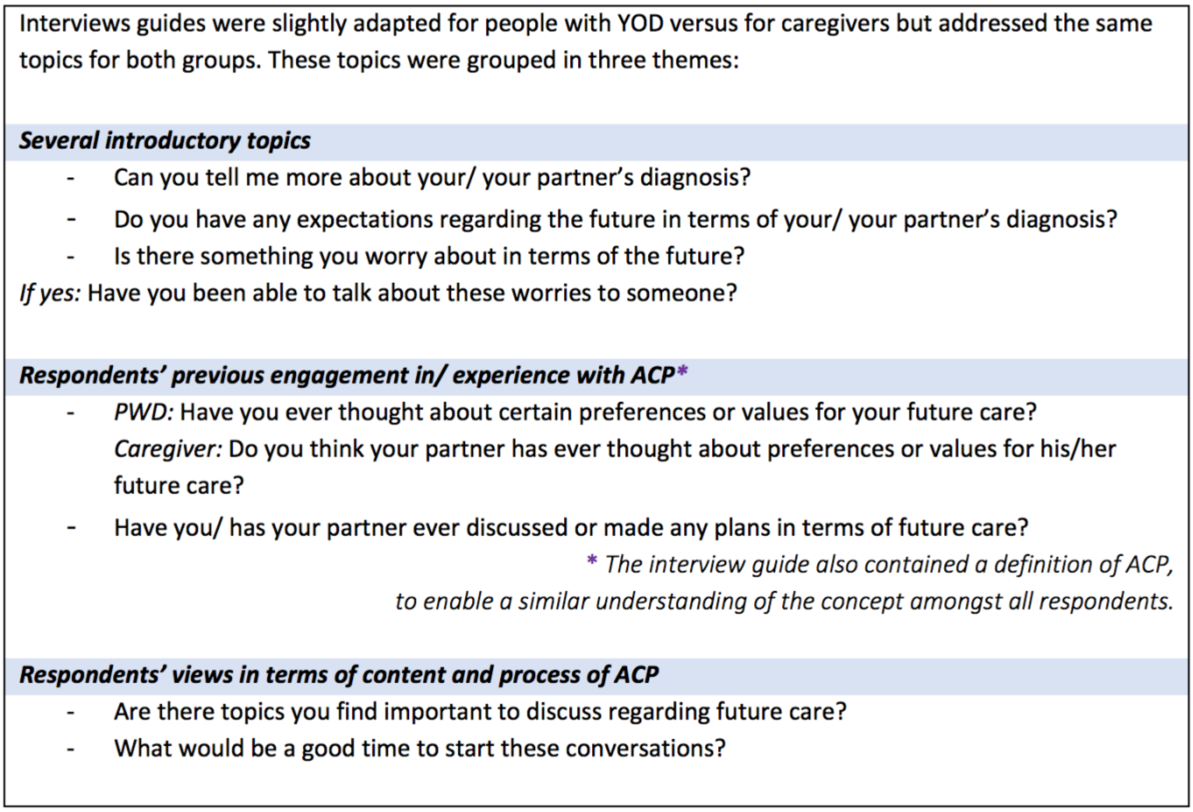
6. Prior to the actual interview, patients were asked a short yes-or-no survey* to guarantee their familiarity with (their rights within) the study
7. Before starting the interview, informed consent forms needed to be signed. For people with young-onset dementia, we installed a double consent procedure: caregivers were asked to also sign the informed consent form of their loved-one with dementia, as a 'witness' that patients were well informed about the study and their rights throughout the interview, and that they voluntarily and consciously chose to participate.

Table 1. Detailed overview of recruitment procedure.

*The survey contained 9 questions regarding the informed consent form. A cut-off score of 6 out of 9 correct answers was established by consensus in the research team as a condition for conducting an interview with a patient at that specific time. This step allowed for taking into consideration the fluctuating nature of cognitive capacity. A copy of the survey can be found in Appendix.

Data collection

Interviews were standardly conducted individually, yet as an ethical safeguard, persons with young-onset dementia were given the option of having their caregiver present if it made them feel more comfortable. Interviews were based on a topic guide, as shown in figure 1, and took place at a location of respondents' choosing, which was their home or the day care center. They took place from August until December 2019.



Interviews guides were slightly adapted for people with YOD versus for caregivers but addressed the same topics for both groups. These topics were grouped in three themes:

Several introductory topics

- Can you tell me more about your/ your partner's diagnosis?
- Do you have any expectations regarding the future in terms of your/ your partner's diagnosis?
- Is there something you worry about in terms of the future?

If yes: Have you been able to talk about these worries to someone?

Respondents' previous engagement in/ experience with ACP*

- *PWD:* Have you ever thought about certain preferences or values for your future care?
  Caregiver: Do you think your partner has ever thought about preferences or values for his/her future care?
- Have you/ has your partner ever discussed or made any plans in terms of future care?

** The interview guide also contained a definition of ACP, to enable a similar understanding of the concept amongst all respondents.*

Respondents' views in terms of content and process of ACP

- Are there topics you find important to discuss regarding future care?
- What would be a good time to start these conversations?

Figure 1. Summary of interview guide with exemplary questions for each theme.

Data analysis

All interviews were conducted and transcribed verbatim by the first author and were subsequently analyzed through constant comparative analysis[20], a qualitative method that entails consistent and comprehensive coding of open-ended data[21]. Transcripts were reviewed line-by-line and discrete text fragments representing a certain idea or concept were assigned a descriptive code. The first five transcripts were coded independently by two researchers (RVR & ADV), the others were coded solely by the first author. No software was used. The next phase in analysis, constructing a 'coding tree' of overarching themes that emerge from comparing codes both within and between interviews, was again done collaboratively by two researchers (RVR & ADV).

Ethics

This study was approved by the Ethics Committee of the University Hospital Brussels (B.U.N. 143201939497) as the central commission and by GZA Antwerp (190304ACADEM) and ZNA Antwerp (approval n° 5208) as local commissions.

RESULTS

Sample characteristics

Characteristics of people with young-onset dementia (n=10)	
Sex	
Male	8
Female	2
Mean age in years	
At time of diagnosis	60
At time of interview	63
Living situation	
Patients still living at home	10
Characteristics of family caregivers (n=10)	
Sex	
Male	2
Female	8
Mean age in years	
At time of interview	60
Characteristics of Dyads (person with young-onset dementia & caregiver)	
Spousal relationship	all dyads
With teenage or adult children	all dyads

Table 2. Characteristics of respondents (N=20)

Overview of results

Two overarching themes, with respective subthemes, emerged from our data. The first theme is centered around the finding that our participating people with young-onset dementia and their caregivers barely engaged in medical care planning as part of advance care planning. The second overarching theme that emerged shows respondents' thought framework of "what matters now & in the future" when they conceptualize advance care planning as a communication process.

1. Respondents barely engage in planning medical care as part of advance care planning

Most of the responding people with young-onset dementia and their caregivers were unaware of what the term advance care planning means or entails. If they had a notion of advance care planning, they discussed it as strictly related to medical end-of-life decisions. It emerged that only one couple had completed an advance directive (living will). A few caregivers and patients stated they rather impulsively and in panic had mentioned or had very briefly discussed the possibility of ending life, mostly right after being diagnosed. Some couples had documented that the caregiver would be the legal representative for the patient. People with young-onset dementia and caregivers elaborated on matters they had planned for the future. However, due to respondents' very limited engagement in planning care as part of advance care planning, our results cannot fully cover the original research question of people's experiences with the process. Our respondents provided several reasons for not discussing/ planning future (medical) care. Moreover, they describe potential benefits of doing so.

PLANNING FOR NON-MEDICAL DOMAINS

Regardless of their self-described lacking knowledge about young-onset dementia, most people with young-onset dementia and their caregivers were aware of the progressive nature of Alzheimer's disease, the inevitable decline in functioning and ultimately the end of life.

> *"The disease trajectory is not important in my case because they don't know anyway how it's going to progress, does it go fast, does it go slow. But the result is all the same huh. We are very well aware of that. (person with young-onset dementia, woman)*

As such, many patients and caregivers made plans for non-medical domains as they arranged financial wills and documented their spouse's right to control finances once the person with dementia would no longer be able to. Several patients also commented on their preferences following death such as funeral arrangements or donating their body to science.

> *"It seemed like a normality to me to arrange that [finances]" (person with young-onset dementia, man)*
>
> *"That I joke about that [funeral] and say like 'no fuss around that, as simple as possible'. . . Yeah that's for later, but of course it can be there quicker than you might think". (person with young-onset dementia, man)*

REASONS FOR NOT DISCUSSING CARE

Patients' attitudes hindering advance care planning communication

Although the majority of caregivers endorsed the idea of engaging in advance care planning, barely any had done so with the patient. Certain attitudes of the person with young-onset dementia, described by themselves and their caregivers, appeared to be hindering for actually discussing future care. More specifically, not worrying about their (future) situation, not experiencing a sense of urgency to arrange matters, and patients resigning in and accepting their situation seemed counter-productive for advance care planning.

> *"Maybe in the far future, it might still be too early. I'm still way too young. . . Plus, I'm still having fun, so at this point that [discussing care] is not necessary". (person with young-onset dementia, man)*
>
> *"I actually don't think about that yet, what the future will bring. It's not going to keep me up at night, let me put it that way. Not at all." (person with young-onset dementia, man)*
>
> *"That's how these things go. I imagine if things get more serious, then I will get more serious too" (person with young-onset dementia, man)*
>
> *"To me it's more like, it has to be done one day. . . But now I don't have the feeling like, yes, I have to [discuss care] as soon as possible". (person with young-onset dementia, man)*

Not knowing what the future holds impedes planning for medical decisions

A topic that emerged frequently among both persons with young-onset dementia and their caregivers was the uncertainty and the unpredictability of the disease trajectory. The majority of both patients and caregivers commented on how difficult it is to know what's coming throughout the progression of Alzheimer's disease and at what speed.

> *"It progresses differently for everyone, so you can't focus on that, right. So yes, we know something is coming, but we don't know what". (caregiver, woman)*

The 'individuality' of each person's disease trajectory complicated advance care planning, in the sense that respondents were doubtful regarding which decisions to prepare for. Additionally, some caregivers suggested advance care planning to be a flexible and vivid process; both as a response to the disease's erratic progression, and as a way to meet patients' potential fear for advance care planning's binding nature.

> *"Yes, it can change… I always have difficulty with ACP [advance care planning] on paper because it changes. Right, not wanting a few things any longer, yeah, until the time is there. . . To me it has to remain vivid. I am under the impression that if I write it down, then it's somewhere outside of me [my control]." (caregiver, woman)*
>
> *"You can discuss certain scenarios like 'I think of it like this', but at the moment it happens, you can never say like 'this is exactly identical to what we have discussed'". (caregiver, woman)*

Uncertainty about what the future holds, appeared associated to living with a 'day-to-day' attitude. Focusing attention to the present, trying to enjoy and "learning to live with it" on a daily basis were examples of such mindset, manifested by both patients and caregivers.

> *'Day by day, that is actually my motto. And there have been relatively many good days". (person with young-onset dementia, man)*

Needing to know more

Several caregivers and patients also emphasized their need for information both regarding young-onset dementia as a diagnosis and prognosis, and regarding advance care planning. Such information provision was deemed lacking and inaccessible by some.

> *"It's only by knowing what exists, that you know how you can be helped. . . The more you know, the better the trajectory will be. I am convinced of that." (person with young-onset dementia, man)*
>
> *"There is information, but you always have to go look for it yourself. . . Information is the most important step [within advance care planning". (caregiver, woman)*

On the other hand, some patients expressed not experiencing a desire for more information.

> Answering question on wish for information:
>
> *"Not too much at once. It's not going to get better, right." (person with young-onset dementia, man)*
>
> *"No, I don't think so. It's more the situation of, yeah, I didn't have anything left to say or do, it didn't matter." (person with young-onset dementia, man)*

PATIENTS' AND CAREGIVERS' HYPOTHESIZED BENEFITS OF ADVANCE CARE PLANNING

Several motives, expressed by both patients and caregivers, emerged that would encourage engagement in advance care planning. One reason for engaging in advance care planning was found in the fact that persons with young-onset dementia would be enabled to participate in decision making. A more frequently emerging motivation was found in the relief that planning in advance would bring for the caregivers.

> *"I don't have any problem with that [advance care planning]. Of course it is something that needs to be arranged, and now I still have a say in it. I mean, now I can still say myself how I would like things". (person with young-onset dementia, woman)*
>
> *"To me it's like, actually we have to do that now, because imagine he is suddenly not able to any longer, then we are too late and that's especially what I want to prevent. While I think that X (patient) is more like, yeah okay, I've still got some time for that, it's not necessary for now. To me it would actually be a relieve like, okay, you've arranged that too, you've got one less worry". (caregiver, woman)*
>
> *"Actually, if you do that [advance care planning, document a will], at this point I think that you do it more so for your relatives than for yourself. Because I am not yet convinced that those people [people living with more advanced stages of dementia]*

actually suffer pain or that those people experience an unbearable feeling". (person with young-onset dementia, man)

"I have to do that for myself as well, not just for him." (caregiver, woman)

2. Patients and caregivers broaden their thought framework to "what matters now & in the future" when thinking of advance care planning as a communication process

When respondents were informed about the concept of advance care planning as a communication process, they broadened their narratives to 'what their future might hold' and reflected in depth about what matters to them throughout their/ their loved-one's disease trajectory. Doing so, they provided meaningful insights into several broader care preferences, needs in young-onset dementia care, and into the inter-relational approach towards people with young-onset dementia.

Values expressed by patients and caregivers as important to them within care

Many recommendations and comments were formulated regarding care provision by both persons with young-onset dementia and their caregivers. These revolved around three major themes, which were mostly mentioned by respondents in relation to patients' younger age. Firstly, the need to enable people with young-onset dementia to remain socially and physically active according to their (remaining) possibilities.

"Every day I fight a battle with boredom" (reason for going do day care) (person with young-onset dementia, man)

"That I still have contact with the outside world, that I can still have conversations with people or a small talk, that is important to me. That I am not completely isolated, that is what I fear." (person with young-onset dementia, woman)

Secondly, a wish for family-centered care emerged. It was emphasized by both patients and caregivers that the condition does not solely affect the patient, and that support should be extended towards children and spouses.

"Especially with young-dementia I do miss a bit the information [about care availability] and the support toward a young family with children, with adolescents". (caregiver, woman)

Lastly, several patients and caregivers referred to the indispensability of autonomy and dignity within care provision.

> *"I was once allowed to join [in support group for patients and caregivers] and then the first time I could be with the group, uum… it was a meeting. And then the second time I wasn't allowed to participate anymore, only those who were not sick were allowed in that room. And I had to make drawings, do another thing. And then I was a bit angry… I was very angry actually" (person with young-onset dementia, man)*
>
> *"It's about being occupied with these people and leaving them in their dignity. And about actively doing things with them, go outside, I mean…, don't [leave them] in a wheelchair for entire days" (caregiver, woman)*

The importance of receiving high quality care

Our respondents generally perceived residential care provision as lacking quality. All, except one patient, were unwilling to address the topic of residential care, according to their caregiver, or had expressed their wish to stay home for as long as possible. Only one person with young-onset dementia stated to be at peace with the idea of someday moving to a facility. The majority of caregivers expressed fear for one day having to make the decision of transferring their loved-one to residential care.

> *"I hope something happens, so I never have to do that. Because knowing that he has always said "I do not want to end up there" and then for me, I also find it very difficult to have to admit to myself that this is where it ends at home now" (caregiver, woman)*
>
> *"They get you out of bed and then they put you back in at night. And for the rest it's done, I mean, as a figure of speech. . . That you are not treated like small children or anything like that. Because that's what you see at times, right, in certain hospitals, that's what you still see. And that is not how I want to end up, no I don't want that". (person with young-onset dementia, woman)*

The public perception of (young-onset) dementia

The stigma and taboo that is still associated to dementia and their impact on patients' self-esteem emerged throughout several patients' narratives. These usually revolved about people assuming that the person with young-onset dementia was unable to do or understand certain things.

> *"It's frustrating for somebody else that I'm not capable of doing some things, and it's a little difficult to deal with… be patient with me and don't make fun of me in some sense. . . then I don't feel very comfortable in life anymore". (person with young-onset dementia, man)*
>
> *"That they [people in general] do not have to immediately think or say to others 'oh, but he doesn't know it anyway'. Okay, that will be the impression these people have, you can't blame them, that a group of people still have that taboo and that they don't know…" (person with young-onset dementia, man)*

DISCUSSION

Summary of findings

It emerged that our respondents lacked awareness about advance care planning, particularly with its concept as a communication process. They had not or barely engaged in future (medical) planning, for which they provided several reasons. Nonetheless, they did also mention the hypothetical advantage of planning future care, and this mostly in relation to the caregiver. Throughout interviews, participants initially associated advance care planning with medically planning for the actual end of life. When discussing advance care planning as a communication process, they paid a lot of attention to non-medical aspects of care and did not compartmentalize medical, social and mental health.

To attune advance care planning to our respondents' views, we suggest the process to be approached holistically, flexibly and relationally as shown in Figure 2. We provide several recommendations for policy and practice on how to do so in Table 3.

Strengths and limitations

The main strengths of this study are the inclusion of people with young-onset dementia themselves and our method of in-depth interviews which allowed for rich and innovative data. Our insights shed renewed light on how to provide adequate long-term care for this specific population, yet likely sparks further questions regarding care needs in other patient groups as well. This paper is, to our knowledge, the first interview study on advance care planning to include people with young-onset dementia themselves. Meeting the at times challenging issue of ethically sound inclusion of people with young-onset dementia, we have combined multiple (novel) methodological ways for ensuring so. We believe this might serve other researchers in designing their study. However, as we recruited dyads, we were unable to gather insights of people with young-onset dementia without close informal caregivers, which would be a recommended topic for future research. Our sample was rather homogeneous, which can be regarded as both a strength and a weakness, since it has been shown that the caregiving experience for people with dementia is intertwiningly shaped by the relationship to the patient and by gender of the caregiver[22]. Our oversampling of female caregivers and male patients is a limitation. Solely interviewing spousal caregivers is a strength, as it enables future studies to compare our results with those of caregivers of a younger generation and, as such, identify whether our results are due to a cohort effect (i.e. related to our spousal age characteristic) or are shaped by the specificity of caring for someone with young-onset dementia.

Advance care planning as a holistic, as opposed to purely medicalized, process would match respondents' views

Our respondents themselves shared their views on care in a manner that did not compartmentalize its elements, but that entailed medical, social and mental health intertwiningly. The narrow and 'academically outdated' conceptualization of advance care planning as mainly documenting end-of-life care preferences and focusing on proximity to death, emerged from our interviews as still being dominant. Since its broadened concept is more attuned to respondents' views and needs, the understanding and awareness of advance care planning as the more holistic communication process it has evolved to needs to be increased. Our results underscore the importance of reciprocity between theory and practice: what is by now a consensus, process-oriented view on advance care planning in academia[8,9,10], does not yet appear to be embraced by practice or the larger community.

The interconnectedness of physical, social and mental wellbeing became apparent from our results regarding patients' reluctance to discuss residential care, their clear wish not to be admitted there and caregivers' concern of ever having to make that decision. The more sedentary services that are developed for older people's care are not adequate for younger persons with more physical possibilities, as also reported by our respondents[23]. Additionally, the need for dignified care and the fear of being socially isolated emerged. Unavailability of age-appropriate care for people with young-onset dementia and their caregivers is a durable issue: a review showed that findings about its limited availability remain largely unchanged during the past 26 years[24]. In line with previous research, the lack of high-quality care might cause people to not feel incentivized for engaging in advance care planning, since the care they will receive would be flawed anyway[25]. The current study shows that this finding might also apply to patients themselves, as our respondents with young-onset dementia also expressed concerns regarding inadequate residential care or simply stated wishing to never be admitted there.

Flexible advance care planning would meet multiple current challenges

Patients' and caregivers' uncertainty regarding disease progression, is a well-established research result in the field of dementia[15,26,27,28]. The current study highlighted yet again that a lack of knowledge regarding what the future holds, at what pace and which planning can be undertaken, is an important complicating factor. However, the field of tension between needing to and not needing to know more about disease progression became apparent in former young-onset dementia research[27,28], and was corroborated by our findings, specifically for people with young-onset dementia themselves. Consistent with previous research[26,27,31], our respondents acknowledged the terminal nature of dementia, regardless of their uncertainty about the trajectory. This was apparent by their arrangements for non-medical and mainly post-mortem domains, like finances and funeral arrangements. As such, there seems to be a 'different logic' when it comes to thinking about the future: planning for after death seems to be done more easily than planning for the period between the present and the end of life. Research by Sussman et al. indeed showed that contemplating about decline is more challenging and threatening than thinking about death[31]. Sensitive and step-by-step information provision on young-onset dementia and on advance care planning might to some extent incorporate thinking about the future into the above-mentioned attitude of living life on a day-to-day basis, and as such increase the readiness for the process in itself. This recommendation partly corresponds to the preference for advance care planning as a dynamic and flexible process, emerging from our current and from previous research[15,32], as it allows to synchronize the process with the individuality of each trajectory. Additionally, a recent study showed that physicians themselves struggle with 'premature' decisions for medical events that may present themselves many years later[33]. The sum of these studies underscores the significance for all stakeholders of ongoing dialogue about what constitutes current and future adequate care. As such, we support the idea[10] that the meaning of advance care planning might be found in its process, rather than in the plans it produces.

The added value of a relational, instead of an individualized, approach to advance care planning emerged

Our respondents saw the advantages of advance care planning more clearly for caregivers than patients themselves. Concordant with other recent research[31,34], our study highlighted the potential emotional benefits of advance care planning for family caregivers. Despite a wish for dialogue by most of the caregivers, advance care planning communication did not or barely take place within our sample. Caregivers appear confronted with conflicting demands in terms of not meeting their own and protecting their loved-one's emotional needs. To meet this complex dilemma of possibly divergent wishes, it was previously suggested[31] to frame advance care planning as an act of care of the person with dementia towards his/her caregiver. Our results indeed confirm that people with young-onset dementia might regard advance care planning as a mechanism to help safeguard their caregiver's emotional state. Additionally, our respondents with young-onset dementia who planned their finances and made funeral arrangements, clearly showed to prioritize their loved-ones' wellbeing. Moreover, our respondents highlighted the need for care provision to target a family, rather than the individual with young-onset dementia. The former suggests that people with young-onset dementia do not think or act in a void centralized exclusively towards them as patients. This is aligned to the concept of relational autonomy, which acknowledges relations as an essential part of decision making and pays attention to how relationships might enhance the proper exercise of autonomy[35]. Our findings indeed underscore a relational, rather than an individual, benefit of and need for advance care planning.

The importance of social embeddedness also shows from our results that people with young-onset dementia experience stereotyping and discrimination: they reported feeling excluded from activities, underestimated by others in terms of remaining capabilities and feared becoming socially isolated. How dementia is portrayed within mainstream culture leads to an increased affective and social distance towards people with dementia[36]. Experiencing stigma negatively effects people with dementia's self-identity and can lead to their social withdrawal[37]. Importantly, patients' hesitation to reflect on future deterioration might be associated to the stigma that comes with dementia[31,38]. Its impact might even be more tangible in young-onset dementia, as a recent study[39] showed that stigmatic attributions, such as stereotypes and negative emotions, were consistently higher when confronted with younger, as compared to older, people with dementia.

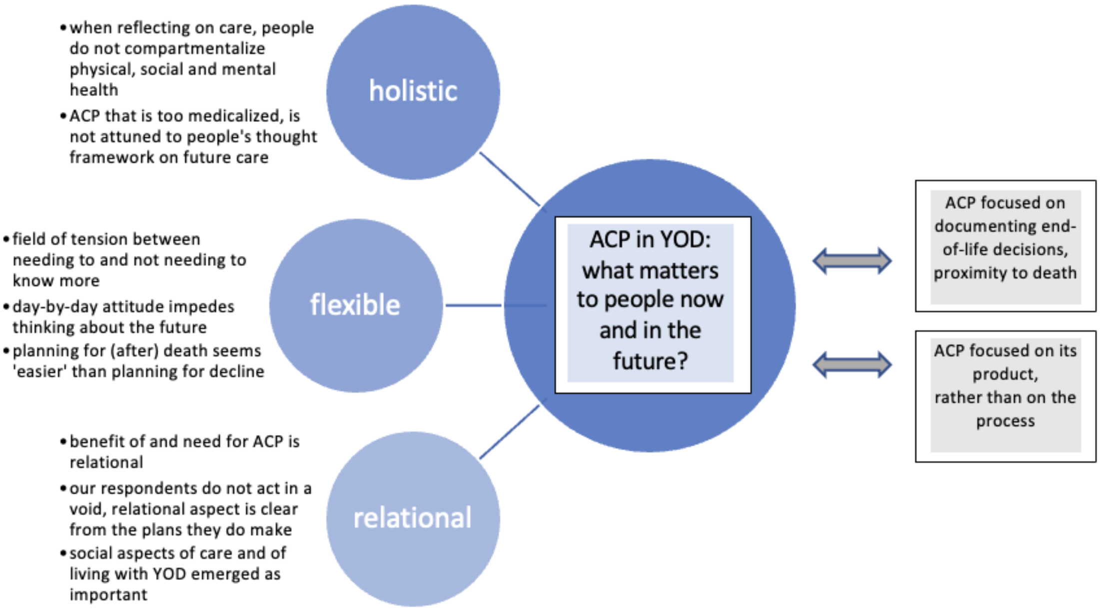


Figure 2. Conceptualization of advance care planning as holistic, flexible and relational.

Implications of our study

Several recommendations are formulated based on our findings. These are aimed at policy makers and practice/ professionals, yet also target care provision in general. Given that the current study is, to our knowledge, the first one to specifically ask people with young-onset dementia and their caregivers about their views on advance care planning, its insights form a solid basis for internationally extending the evidence base on this particular element of palliative care.

As the majority of our results were not explicitly associated to our respondents' age, we do not rule out the possibility that our findings and recommendations are generalizable to people with late-onset dementia and their caregivers. Results regarding age-appropriate care and support aimed at a young family, might on the other hand not be dementia specific and, as such, applicable for younger people living with another life-changing diagnosis than young-onset dementia.

Recommendations for holistic advance care planning

- Policy makers hold a responsibility in contributing to a more consistent public discourse and organizing larger information interventions to raise both professionals' and the general public's awareness of advance care planning as a communication process
- In medical practice, the overarching framework of 'what is important to people in terms of the present and the future' could serve as a starting point for advance care planning, as this might make the process more attuned to patients' and caregivers' own ideas of it
- We support the idea of advance care planning to entail conversations about harm reduction by avoiding unwanted treatments, but as a means to achieve what matters most to people[40].
- Care should be improved, with attention to social, physical and mental domains, to form an incentive for advance care planning

Recommendations for flexible advance care planning

- Adequate information provision about both dementia and advance care planning can be regarded as a necessary first step for enabling people with dementia and their caregivers in the process
- In accordance with a previous suggestion[29], we support the idea that disclosure of information on prognosis and on advance care planning to people with young-onset dementia and their caregivers should be embedded within the care pathway
- The process of advance care planning should be the focus, rather than its product

Recommendations for relational advance care planning

- Professionals might raise a dialogue about patients' and caregivers' mutually protective roles, as this might create broader communicative space for advance care planning
- Maximizing people with dementia's opportunity to participate in advance care planning could be regarded in itself as a means to counter stigmatic beliefs since it allows them to be active agents, to have their opinion heard and to have their capabilities, rather than their possible disabilities, highlighted. To do so, advance care planning should be a holistic, flexible and relational process.

Table 3. Recommendations for policy and practice on how to implement advance care planning as a holistic, flexible and relational process.

CONCLUSION

Our participating people with young-onset dementia and their caregivers still equated the term advance care planning to medical decisions for the actual end of life. A view on advance care planning that is too medicalized and solely centered towards the patient might impede engagement in the process, since our respondents think in an overarching concept of what is important to them now and in the future. To attune advance care planning to this thought framework, the communication process should be presented and implemented as holistic, flexible and relational in nature.

KEY STATEMENTS

What is already known:

- Despite consensus on its relevance, the uptake of advance care planning in dementia is low
- People with young-onset dementia and their caregivers are hypothesized to have distinct needs and preferences for palliative care and advance care planning
- There is a dearth of studies which include people living with young-onset dementia and their family

What this paper adds:

- People with young-onset dementia and their caregivers strictly associated advance care planning with medical planning for the end of life and reported to have not or barely engaged in this
- Respondents were unacquainted with the concept of advance care planning as a communication process, yet when discussing advance care planning as such, they broadened their narratives to 'what matters to them now and in the future'; in doing so they did not make a division between their medical, social or mental health
- An overly medicalized approach to advance care planning, restricted to planning death, might impede people's engagement in the process

Implications for practice, theory and policy:

- Within our sample, advance care planning was solely known as a narrow medical term, namely as documentation of end-of-life decisions. However, conceptualizing advance care planning as a communication process about what matters most might resonate better with their actual needs. Advance care planning can best be implemented in practice as a holistic, flexible and relational communication process
- As the majority of our respondents' preferences were not directly linked to their younger age, our findings and recommendations could possibly help guide research regarding the implementation of advance care planning in other patient populations
- Policy makers and clinicians jointly hold responsibility for raising awareness and for uniform messaging about the evolved concept of advance care planning as a broad communication process.

Acknowledgements

The authors wish to genuinely thank our respondents for sharing their valuable experiences and insights. They also wish to thank misses D. Thys and misses G. Callewaert for their help with recruitment.

Author contributions

Study concept and design: Van Rickstal, De Vleminck, Van den Block. Recruitment of respondents: Van Rickstal, Engelborghs, Versijpt. Acquisition of data: Van Rickstal. Analysis and interpretation of data: Van Rickstal, De Vleminck, Van den Block. Drafting of the manuscript: Van Rickstal. Critical revision of the manuscript for important intellectual content: De Vleminck, Engelborghs, Versijpt, Van den Block.

Declaration of conflicting interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship and/or publication of this article.

Funding

The author(s) disclosed receipt of the following financial support for the research, authorship and/or publication of this article: This work was supported by the Research Foundation – Flanders (FWO), grant number 1S75919N (Strategic Basic research mandate by Van Rickstal) and grant number 12ZY222N (post- doctoral mandate by De Vleminck). For publishing this article, we would like the acknowledge the support of the University Foundation of Belgium.

Appendix 1.

Short survey of 9 yes/no questions regarding Informed Consent to ascertain that the person living with YOD understands the study and the rights he/she has throughout.

1. For this study, will I be asked to take part in an interview that will last about one hour?
2. If I choose not to participate in this study, will this impact the care I usually receive?
3. If I take part in the interview, can I refuse to answer certain questions?
4. Even though I said I wanted to participate, can I still say at any time that I rather not take part in the interview?
5. If I would choose to stop my participation in this study, would this affect the care I usually receive?
6. Was I explained what the researchers' goal is with this study?
7. Am I voluntarily participating in this study?
8. Was I explained that I can be interviewed in the presence of my caregiver if I prefer so over an individual interview?
9. If something is written about this study in magazines, will people who read it know that I participated?

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Chapter 5

People with young-onset dementia and their family caregivers discussing euthanasia: a qualitative analysis of their considerations

Van Rickstal, R., De Vleminck, A. Chambaere, K. & Van den Block, L. (2022).

Submitted

ABSTRACT

Objectives

Research showed that people with young-onset dementia and their family caregivers raised the topic of euthanasia when talking about the broader topic of advance care planning. A better understanding of what people address and why may inform the evolving landscape of physician assisted dying. This study aimed to explore the considerations on euthanasia that people with young-onset dementia and their family caregivers expressed.

Methods

A secondary qualitative analysis on interviews with 10 Belgian people with young-onset dementia and 25 family caregivers, using constant comparative analysis.

Results

Respondents described similar contexts in which euthanasia had been discussed: the topic arose at ‘key’ moments, mostly with family caregivers, and was motivated by patients considering the impact of disease progression for themselves and their loved-ones. Caregivers shared opinions on the euthanasia law and discussed the emotional impact of discussing euthanasia.

Conclusions

Considerations of people with young-onset dementia towards euthanasia appear rooted in personal, as well as in anticipated interpersonal and societal suffering. The negative image associated with dementia and dementia care seemed to influence people’s expectations for and thoughts on the future.

Practice implications

Patient-physician communication should include detangling motives for euthanasia requests, openly discussing fears and reflecting on prognosis.

INTRODUCTION

It is estimated that up to 4 million people globally develop dementia symptoms before the age of 65, captured by the term young-onset dementia (YOD)[1]. A recent systematic review and meta-analysis on population-based studies, points to an incidence rate of 370,000 new YOD cases annually worldwide[2]. Given that Alzheimer's disease and related neurodegenerative disorders are characterized by progressive cognitive decline, the significance of advance care planning (ACP) for this specific patient population is widely recognized[3,4,5]. ACP has been conceptualized as a communication process between patients, family and professionals to explore patients' preferences for future (medical) care, including at the end of life[6].

Despite the hypothesis that people with YOD and their caregivers want more involvement and autonomy in palliative care and specifically ACP[7], previous research showed[4,8] that they barely take part in ACP and that they intuitively equate the term ACP with making medical decisions for the actual end of life[9]. Moreover, throughout interview studies, Belgian patients and caregivers spontaneously touched on the topic of euthanasia when discussing their views on ACP, as opposed to American family caregivers[8,9]. The interpretation of this discrepancy was that people with YOD and their caregivers think about end-of-life decisions within their countries' legal framework[8]. This valuable finding was not analyzed in depth in our previous main ACP analysis[4,9] as these unsolicited statements did not address the aim of our previous research questions. Given that the specific topic of euthanasia in YOD as seen from the perspective of people with YOD and family caregivers themselves has never been addressed within existing literature, we conducted a secondary qualitative analysis on the actual content of what respondents expressed about euthanasia.

At present, more and more nations across the world establish legal grounds for physician-assisted dying, an umbrella term usually referring to euthanasia and physician assisted suicide[10]. Euthanasia is the act of where a health care practitioner intentionally ends a patient's life by active drug administration at this patient's explicit request[11]. Countries such as Belgium and the Netherlands currently have the most progressive physician assisted dying laws[12]. The euthanasia law[11] that was passed in Belgium in 2002, puts forward several central substantive criteria; one must suffer unbearably from an incurable condition, without any prospect of improvement and must express a well-considered, voluntary and repeated request for euthanasia. This request has to be ‘current’, entailing that an advance directive for euthanasia is relevant solely in the specific case of an irreversible persistent vegetative state. As such, in Belgium people with dementia are eligible for euthanasia only in those stages of their condition where they are still competent to voice a current request. At present, public debate focuses on the legal status of an advance euthanasia directive for patients in late-stage dementia, where it might be challenging to cognitively substantiate their euthanasia request[13].

As access to assisted dying may be legalized or expanded in varying jurisdictions, research has to follow to assess the effects and impact on patients and other stakeholders[10]. However, a systematic review on the attitudes of patients and caregivers, health professionals and the general public on assisted dying in dementia showed that the perspectives of people with dementia and depicted in only a very few studies, which are mostly of low quality[14]. Within the internationally evolving landscape, it is useful to shed light on the views of people with (young-onset) dementia and their caregivers regarding euthanasia. As such, we wish to increase the understanding of people’s thoughts on euthanasia in the context of young-onset dementia.

The research question for this study is: “what do people with YOD and their family caregivers who raise the topic of euthanasia, during an interview on the broader topic of ACP, express regarding euthanasia itself and regarding communication about euthanasia?”.

METHOD

Research design

This explorative qualitative study is based on data collected through semi-structured face-to-face interviews from two previous qualitative studies (see Table 1 for the studies’ characteristics). Qualitative study designs were chosen because of both the exploratory topic and the sensitive research questions. We performed secondary data analysis, as the topic of euthanasia played a prominent role in remarks that respondents spontaneously added without any prompts by the interviewing researcher. Whenever respondents addressed the topic further discussion was enabled through one or more follow-up questions (such as inquiring how communication about euthanasia had taken place, with who etc.) and through 'silent probes' by the interviewer, before associating respondents' comments to a predetermined topic leading back to the interview guide.

Characteristics	**Study 1**	**Study 2**
Respondents	N = 15 Family caregivers of people with YOD	N = 20 Dyads of people with YOD (n=10) and their respective family caregivers (n=10)
Data-collection	Semi-structured in-depth interviews, face-to-face	Semi-structured in-depth interviews, face-to-face
Data-analysis	Constant comparative analysis	Constant comparative analysis
Timeframe	July - September 2017	August - December 2019
Publication	*Reference 4*	*Reference 9*
Inclusion criteria	No data from people with young-onset dementia	For **persons with dementia** (6): 1. Being formally diagnosed with young-onset probable Alzheimer's disease (based on strictly applied standard diagnostic criteria; e.g. NIA-AA criteria) 2. Having a score of minimum 16 on the MMSE 3. Being diagnosed for at least 6 months (for reasons of sensitivity to grieving stage after diagnosis) 4. Being 18 years of age or older 5. Speaking Dutch 6. Signing written informed consent (themselves + caregivers' consent as witnesses is required)
	For **family caregivers** of persons with dementia (4) 1. The main family caregiver of a person formally diagnosed with young-onset dementia (regardless of dementia subtype), 2. Aged 18 or older, 3. Dutch-speaking and 4. Needed to have given written informed consent to participate.	For **family caregivers** of persons with dementia (4): 1. Being the primary caregiver of a person formally diagnosed with young-onset probable Alzheimer's disease 2. Being 18 years of age or older 3. Speaking Dutch 4. Signing written informed consent (+ providing consent for person with dementia, as witness)

Ethics	- Ethics Committee of Brussels University Hospital (B.U.N. 143201732034) - Institutional Review Board of Hospital Network Antwerp (ZNA, approval n° 4939) and of Hospital GasthuisZusters Antwerp (GZA, 170407ACADEM)	- Ethics Committee of Brussels University Hospital Brussels (B.U.N. 143201939497) - Institutional Review Board of Hospital Network Antwerp (ZNA, approval n° 5208) and Hospital GasthuisZusters Antwerp (GZA, 190304ACADEM)

Table 1. Comparison of study characteristics

Participants

This study is based on data generated during 2 previous qualitative studies[4,9] in which we included family caregivers of patients with YOD and persons living with YOD of the Alzheimer Type themselves. See Table 1 for inclusion criteria. The interview study with family caregivers of people with YOD had 15 participants, with an average age of 61 years old. Nine respondents were female, six were male. The study in which dyads of people with YOD and their respective caregivers were recruited, had 20 respondents. Participating patients (two women and eight men) had an average age of 60 years old at the time of diagnosis and 63 years old at the time of the interview. Their respective caregivers (two men and eight women) were averagely 60 years old at the time of the interview.

Recruitment

Respondents in both studies were recruited through four intermediate people: two neurologists, a coordinator of a day care centre for people with YOD, and the founder of a non-profit organization that organizes activities for people with YOD and their families. Coordinators at each site approached eligible respondents, briefly explained the study and asked for permission to give their contact information to the researchers. Participants were contacted by phone or email by R.V.R. or A.D.V., addressing any questions and ascertaining willingness to take part in the study. After agreement, interviews were scheduled at a mutually convenient time, at a place of respondents' choice (usually their home or the day care centre).

Further details on recruitment and ethical safeguards when recruiting participants can be found elsewhere[9].

Data-collection

Data were collected through face-to-face in-depth semi-structured interviews by two researchers (RVR and ADV). One researcher (RVR) works as a doctoral researcher and obtained a master's degree in clinical psychology. The other researcher (ADV) is a professor with a background in sociology and is the daily supervisor of the former (RVR). For both studies two very similar interview guides were developed within the research team. In neither guide was a question about euthanasia. Data-collection of one included study dates from 5 years ago: we deemed that the time that has passed since these data were originally collected has not impacted the relevance of our findings, as our countries' legal framework for euthanasia has not changed during this time.

Data-analysis

All audio-recordings of the interviews were transcribed verbatim by the first author and subsequently analyzed through a way of thematic analysis. As stated previously by Braun and Clarke[15,16], thematic analysis can be best thought of as a family of or an umbrella term for a set of approaches for the analysis of qualitative data that have a shared focus on developing themes (patterns of meaning) from qualitative data. More specifically, we opted for the strategy of the constant comparative method[15]. Transcripts were reviewed line-by-line and codes were assigned to discrete fragments of text, usually one or two sentences representing a certain idea or concept. For the secondary data-analysis, all text-fragments relating to the topic of euthanasia were assembled throughout the 35 interview transcripts. Of those 35 transcripts, 24 had explicit references/quotes regarding euthanasia. These were collected within a new dataset to subsequently be compared within and between interviews by two researchers (RVR and ADV), in order to identify broader themes. The goal of our analysis was to identify and describe patterns in data, and to interpret their relationships[15]. Based on the personal preference of the first author (RVR), no analytic software was used.

Ethics

This secondary analysis was in line with the aims of the original research studies and their initial ethics approvals obtained *(see Table 1)*.

RESULTS

Over two third of all respondents spontaneously raised the topic of euthanasia and their previous communication about euthanasia when discussing ACP.

When respondents raised euthanasia during interviews, they addressed four major subtopics: (1) the context in which patients and family caregivers had already talked about euthanasia (when, with whom and why), (2) their views on the legality of euthanasia in dementia, (3) the ability to still experience joy after being diagnosed with young-onset dementia, and (4) the impact of euthanasia and euthanasia discussions as experienced by family caregivers.

The context in which patients and caregivers had already talked about euthanasia

When: *Euthanasia brought up at several 'key moments'*

Various key moments or events gave rise to euthanasia becoming a topic between people with YOD and their family caregiver. It emerged that the initial shock of receiving a YOD diagnosis could lead to patients, sometimes impulsively, discussing the option of euthanasia very early on in the disease trajectory. Other highly emotional moments, such as during arguments, were also identified as moments where euthanasia was explicitly brought into the conversation. Experiencing more difficulties in day-to-day activities and foreseeing or noticing deterioration could make the topic resurface later on.

> *"If it worsens, then I will take the necessary decisions myself, huh, for euthanasia or such… No, at that point I am done with it." (man with YOD, 2codeE17-19)*

Being confronted with other patients that actually received euthanasia, was also said to be a moment when the topic of euthanasia arose. One caregiver explained that her husband had discussed his own request for euthanasia with another person with YOD whose euthanasia request was granted shortly after. This caregiver believed that being confronted with another patient's euthanasia, had strengthened her husband's idea at the time.

> *"It reinforced him (patient) in the fact that it (euthanasia) was possible and that it was a good alternative".*

With whom: *euthanasia was discussed with family, rather than professional, caregivers*

For the vast majority, conversations regarding euthanasia initially arose between the person with YOD and his/her family caregiver(s). More specifically, they were typically initiated by the patient, were directed towards family caregivers such as spouses or siblings, and sometimes simply 'faded away' over time.

> *"But then I saw, the next day he (patient) was no longer occupied with that (euthanasia) at all." (caregiver, code D124)*

At other times, they were continued with medical or legal professional caregivers, mostly through the initiative of the patients' family caregiver.

Why: *Personal as well as interpersonal considerations when discussing euthanasia*

People with YOD's main motivation for thinking about or considering euthanasia, appeared to stem from their wish to spare themselves inevitable decline and suffering and to safeguard their sense of dignity.

> *"It's going to have to happen one day, dying, right… I would prefer (to make) the best choice… Little pain and such…" (man with YOD, 2, Code 0122).*
>
> *"To become incontinent for instance, I would have a lot of difficulties with that. And then it might be that I say that it needs to be over for me." (woman with YOD, 2, code E46)*

In this context, family caregivers discussed patients' attempt to avoid institutionalization and having the choice between living in a care facility (nursing home) or ending life through euthanasia.

> *(Quote 1, code L142) A woman with YOD wanted euthanasia "from the moment she needed to be locked up".*
>
> *"In fact, you (caregiver) are always pushing him (patient) towards the exit… And then he has still got the choice 'I am going to the institution' or 'I choose the other way' (euthanasia)" (caregiver, 1, M70-71)*

Both people with YOD and caregivers elaborated on the meaning and importance of social relations in the context of (wanting) euthanasia. For example, patients wishing euthanasia when they no longer recognize their loved-ones.

> *"I would not like to be… when the memory is completely gone, then it's not necessary anymore…" (man with YOD, 2, codes 0122-124).*
>
> *"He (patient) says 'if I am no longer able to speak, then it can stop for me." (caregiver, 2, code A34)*

Other considerations were not wanting to be a burden to their family and wishing to maintain the capability of having conversations with others.

> *"But the phenomenon of euthanasia when you are doing really bad or right on the edge… then I don't hesitate for a moment. And then of course that comes back to the story of X (wife) and X (daughter) who have to be all right." (man with YOD, 2, code Q109)*
>
> *"He (patient) has always said 'I do not want you (caregiver) to suffer, and if it is not attainable any longer, then I choose to stop." (caregiver, 2, CodeP51)*

Opinions on the euthanasia law and the criterium for cognitive capacity

During the interviews several caregivers shared their, and according to them also their loved-ones', viewpoints on the validity of euthanasia through an advance directive for people who have reached a stage of dementia in which they (might) lack the cognitive capacity to confirm their previous wish. The opinions expressed revolved around whether or not the current Belgian law should be expanded to allow people in advanced stages of dementia to receive euthanasia based on their previously stated request.

> *"Everyone knows that you have it (dementia), that you do not want to continue life like a plant… yeah guys… then, why not?" (caregiver, 1, code G168)*
>
> *"There are so many discussions about euthanasia for people with dementia, but actually the people who make the laws, should be in that situation themselves. They themselves should have a partner or a mother or a dear friend, and experience -for entire days- what that means. If X (patient) now, today, says 'if I am no longer able to do this, and that and that, then I want euthanasia', then I believe he has got the right to that". (caregiver, 2, codes N67/68/68)*

Both persons with YOD and their caregivers discussed the legal requirement of having cognitive capacity at the time of formulating a current euthanasia request and/or being granted euthanasia.

> *"One of her (patient who was granted euthanasia) statements was that 'euthanasia is something you need to do with a sound mind'" (caregiver, 1, code G157)*
>
> *"At some point, if you wait too long you can no longer (request euthanasia). I know that's an issue and I'm kind of aware of it but it's not in the fore-figure for me now, because I figure I still have six months or a year where I still have the capability to make decisions." (man with YOD, 2, code C28-29).*

In this sense, the need to address euthanasia recurrently, was discussed.

> *"And you need to bring it (euthanasia request) up regularly and see if they… And clarify to them (patients) that it is now or never in their case." (caregiver, 1codes M, p.15)*

Dementia as condition that still allows room for joy

Caregivers also put forward that euthanasia for people with dementia was different from euthanasia in more somatically "aggressive" diseases.

> *"I mean there are people here… many people… for whom that (euthanasia) probably isn't the best solution because they are still… physically relatively okay. Because they can still enjoy many things. And… and that will continue to be the case until they are too far to make a decision." (caregiver, 1, code165-166)*

This specificity was usually focused on the ability of people with dementia to still experience joy.

> *"Should you perform euthanasia on someone who can still eat with a tasty appetite every day? Who can still enjoy?" (caregiver, 1, codeK87)*
>
> *"Yes, but with a sound mind… Then you can also still enjoy life fully capable. Right. So the moment that you can still consciously enjoy all kinds of things with a sound mind, the beautiful weather, then you have to be able to say at the same time 'I want to die, because tomorrow it might be that I'm gone." (caregiver, 1, Gp10,A3)*

Also, people living with YOD themselves talked about experiencing feelings of well-being despite their diagnosis.

> *"There have been relatively many good days (since diagnosis)." (man with YOD, 2, codeO130)*

> *"Today I feel well again and then I say to myself 'yes, I don't need anything else'." (man with YOD, 2, codeQ49)*

Emotional impact of euthanasia and discussions about euthanasia for family caregivers

Caregivers elaborated on how emotionally challenging it felt to discuss euthanasia one-on-one with their loved-one.

> *"I have to bring that (euthanasia) up and that is not pleasant for me because it seems like you'd wish she (patient) were dead." (caregiver, 2E56)*

In this regard, several caregivers preferred to have a professional (an "outsider") involved.

> *"I find it difficult to do that, as a wife, to check how he feels towards euthanasia. I'm almost too involved. So I always find it very difficult to say something about that myself. I think it is best if he can talk to someone else about that (euthanasia)." (caregiver, 2D79-82)*

One caregiver also explained the difficulty when her loved-one with YOD no longer remembered his own wish for euthanasia.

> *"So, I thought, I don't have to hold on to that (patient saying he wanted euthanasia). I might have someone else inform with him how he feels about that, if there's a chance, but for the rest I've got to let that go. Because in the end he will get the impressions that I wish he were dead." (caregiver, code D124-125)*

Family caregivers of persons with dementia who were actually granted euthanasia, elaborated on the sense of guilt they experienced afterwards.

> *"Why have I not tried to convince her (patient) to not do it (euthanasia) after all." (caregiver 1, code G122)*
>
> *"You always feel guilty. . . the impression that you have not done enough." (caregiver 1, code M117 & M118)*

One caregiver explained feeling that his family regarded him as *"a bit of a murderer because he had encouraged it (sister's euthanasia)". (caregiver, 1codeG40)*

DISCUSSION AND CONCLUSION

Discussion

Over two third of our respondents spontaneously addressed the topic of euthanasia and how discussions in this regard took place. If euthanasia was discussed, four main results stood out: **(1)** People with YOD and their family caregivers described similar contexts in which these discussions about euthanasia arose. The topic was brought up at several key moments (such as the moment of diagnosis), usually with an informal caregiver, and was motivated by both personal patient considerations (the impact for the person with YOD), as well as by patients considering the impact of disease progression for their loved-ones. **(2)** Family caregivers paid a lot of attention to the criteria under which euthanasia is allowed in Belgium for people with YOD, specifically in the context of declining cognitive capacity. **(3)** Dementia is characterized as a condition that still allows room for enjoyment, making it different from other conditions in the context of euthanasia according to our participants. **(4)** Family caregivers elaborated on the emotional challenges they experienced when discussing euthanasia and, some, on the feelings of guilt after a loved-one with dementia was actually granted euthanasia.

The main strength of this study is its ability to shed light on an understudied topic through patients' and caregivers' own narratives. This is, to our knowledge, the first research to enter the new territory of euthanasia as discussed by people with YOD and their family caregivers. Since euthanasia was not included in the interview guides as a specific topic, these instances of communication were not consistently structured throughout interviews, and hence the topic was not discussed as 'in-depth' as it would have been in a study focused solely on euthanasia. As such, respondents' unsolicited remarks cover a broad range of ideas, for some of which we might not have reached data-saturation. However, given that the various viewpoints expressed are instances of unique experience-based perceptions on an undiscovered topic, they hold innovative value to both theory and practice. Nonetheless this study's findings form a steppingstone for further developing the international evidence base on this specific topic, it is necessary to put patients' and caregivers' experiences of and perspectives on euthanasia in YOD at the forefront of future research questions. As this study was homogeneous in terms of respondents' Belgian social and legal background, the way in which other legislative and social contexts possibly impact patients' and caregivers' considerations, should be subject of further qualitative research in various countries. Whilst some results seem to point to age-specific factors (for instance trying to avoid residential care), others appear rather dementia- than age-specific (for instance the issue of declining cognitive capacity). Future research should address this study's remaining question of to which extent our findings are cohort- or disease-specific, or both.

Interpersonal relationships are an aspect of euthanasia considerations

Our study shows the emotional impact of receiving a diagnosis of dementia at a young age, with patients who at times rather impulsively express their 'interest' in euthanasia. A large-scale interview study with German family caregivers of people in advanced stages of dementia showed that over one out of four patients in early stages of YOD experience feelings of suicidal ideation that fade over time[17]. Similar to the euthanasia considerations found in the current study, these ideations are possibly caused by the wish to spare oneself from suffering, as well as a wish to not to be a burden to family, rather than representing an actual wish to die[17]. Our respondents paid attention to social, and especially familial, relationships within their views on euthanasia. Not wanting to become a burden to loved-ones was an important consideration when thinking about the future. Yet again, this reiterates the previously reported protective role that people with YOD take on towards their family caregivers when contemplating their future care[9]. Our study further shows that people with YOD might wish to avoid not solely their personal, but also their families', suffering. Given the finding that the consequences of YOD for patients' loved-ones might be part of their euthanasia considerations, it seems important within practice to clarify and to the extent possible detangle this reason from the motive of unbearable personal suffering.

Societal image of dementia and dementia care are present in euthanasia considerations

The finding that considerations on euthanasia entailed an attempt to avoid decline and maintain dignity can be linked to several previous findings on how society as a whole deals with dementia. Already a decade ago it was reported that popular media tend to focus on portraying the terminal stage of dementia which becomes representative of the entire trajectory[18]. More recently, it was reiterated that stigma and stereotypes shape our image of (young-onset) dementia[19]. Moreover, the strong relationship between the general public's views about the quality of life in advanced dementia and their views about legalizing euthanasia for these patients through advance directives has been established through an experimental survey[20]. In a Dutch qualitative study, physicians expressed that society tends to consider dementia as a condition with hardly any quality of life and regards euthanasia as a more dignified alternative[21]. Also Belgian physicians, as shown from a recent focus group study, felt that society's negative portrayal of dementia impacted patients and was associated to catastrophic reactions to being diagnosed[22]. Importantly, the societal image in which a life with dementia is at times reduced to 'a life not worth living'[19,23,] , which can be a driving force behind euthanasia requests[24], is contradicted by the majority of patients with both young- and late-onset dementia[17,25]. The current study corroborates the finding that caregivers and patients themselves also underscore the remaining capacity of enjoyment within a dementia trajectory[4] and that they might consider the burden of dementia differently than 'healthy' individuals do[17]. Through their own involvement in research, patients have been providing a more nuanced and at times brighter outlook on dementia[25]. So did the people living with YOD and their caregivers who took part in our studies, when explaining that there is still room for 'enjoying life' regardless of the diagnosis and the burden it causes. Topics regarding the 'negative' image of dementia might deserve a place within consultations since they appear to influence patients' and caregivers' expectations of the future.

Stigmatizing beliefs, particularly regarding inadequate residential care for people with YOD, have been described previously[26]. The current study found that euthanasia is at times regarded as a mean to avoid institutionalization. This highlights the urgency to address both the needs in and the misconceptions about residential care for people with YOD. Moreover, this result might shed further light on previous findings that contemplating decline in dementia is more threatening than thinking about death itself[27]. It seems evident that when the law on euthanasia states that all reasonable alternatives need to be tried before granting a euthanasia request, these alternatives ought to be deemed reasonable and adequate from the patient's perspective as well. Public and political debates about the legal criteria for euthanasia, such as capacity, should co-exist with initiatives on care-improvement (e.g. residential care).

Legal context influences respondents' thought framework

Previous studies suggested that the framework people use to think about end-of-life decisions is likely to be influenced by the medicolegal context in which people find themselves[8,28].

Former research with American caregivers of people with YOD showed that the actual life-ending decisions discussed mainly revolved around suicide. It seems plausible that due to Belgian law, the end-of-life decision of suicide is replaced -at least partly- by its legalized alternative with the same end-result, namely euthanasia[8]. In both our studies with Belgian participants, the majority of respondents elaborated on euthanasia when discussing ACP in YOD. Moreover, this study's finding that some caregivers, from their and their loved-one's perspective, specifically elaborated on the requirements of capacity and therefore the timing of a euthanasia request within Belgian euthanasia law, echoes the current public and political debate and, as such, its relevance. Our study reiterates that professionals being open to conversations about concerns and fears, possibly including those about ideas of (hastening) death, can be regarded as a service to people with YOD and their family caregivers[29]. Our study shows that patients and caregivers themselves are an indispensable voice to be heard at the table as their lived experience most likely leads to unique perspectives on the practice of euthanasia.

Practice implications

It appears recommended for physicians to pay sufficient attention to familial relationships and interpersonal considerations when discussing euthanasia with people living with young-onset dementia and their caregivers. Also, our study suggests that patients and caregivers should be provided with the opportunity to openly discuss concerns and fears, including those about hastening death (euthanasia), and with the opportunity to reflect on their prognosis, in a timely and realistic manner. When outlining policy, it is important to reflect as a society on our portrayal of (young-onset) dementia and its ethical implications for people and families living with the condition. Moreover, it seems important for policy to re-evaluate the current care initiatives that are available in young-onset dementia and make improvements where necessary.

CONCLUSION

Our findings suggest that the considerations of people with YOD towards euthanasia are not solely rooted in expected personal unbearable suffering, but also appear to stem from the anticipated impact of their condition on significant others. The negative manner in which (young-onset) dementia is socially framed and the embedding of euthanasia as a legal end-of-life option, might influence the considerations of euthanasia.

Conflicts of interest

There are no conflicts of interest to be declared.

Acknowledgements

The authors wish to genuinely thank all respondents for their willingness to share their insights.

Funding

This work was supported by the Research Foundation—Flanders (FWO), grant number 1S75919N (Strategic Basic research mandate by Van Rickstal), and grant number 12ZY222N (post-doctoral mandate by De Vleminck).

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PART IV

GENERAL DISCUSSION

INTRODUCTION

The overall research objective of the current dissertation is to identify the experiences with, the preferences for and the perspectives on ACP in YOD of the main participants involved in the process. Therefore, we inquired the viewpoints of family and professional caregivers of people with YOD (Chapter 1,2,3), as well as people with YOD themselves, together with their respective family caregiver (Chapter 4,5).

In this part, the main findings of the studies undertaken to meet our objective will be summarized. This will be followed by a discussion of the strengths and the limitations of the methodologies chosen and by a general discussion of some of our most important findings. Lastly, several implications for practice, policy and future research are proposed.

SUMMARY OF MAIN FINDINGS

PART II: Identifying the experiences with, perspectives on and preferences for ACP in YOD of family caregivers and physicians

CHAPTER 1 consists of an interview study with 15 Flemish family caregivers of people with YOD[1]. This study showed that planning for the future typically revolved around non-medical domains such as financial planning, organizing the living situation or making funeral arrangements. Several factors appeared to intertwiningly hinder engagement in ACP. The limited or lacking engagement in the process, was clarified through various reasons: not regarding ACP as useful, patient behaviour that hindered ACP (such as ‘denying the situation’), adopting a day-to-day attitude, family caregivers emotionally protecting themselves and the uncertainty about patients’ cognitive competence to engage in ACP. The opportunity to engage in ACP was welcomed, as shown through respondents’ preferences for timely initiating ACP, preferably done by a third party (physicians were usually mentioned) and with a focus on patients’ remaining possibilities. Nonetheless, a potential wish for ACP can often not be put to practice, resulting in lacking or limited uptake of ACP in YOD.

CHAPTER 2 entails a comparative study between Belgian and American family caregivers of people with YOD in terms of their experiences with and preferences for ACP[2]. Throughout the interviews, several similarities and differences between American and Belgian caregivers emerged regarding both the engagement in, as well as the conceptualization of ACP. The main similarities in both respondent groups were a restricted knowledge of the term ACP and its content, lacking or limited communication both with professional and family caregivers when an advance directive is completed, and a desire for professionals to timely initiate ACP. Important differences were the attention paid to those end-of-life decisions depicted in the legislature of their respective countries, American caregivers who placed higher emphasis on financial planning than Belgian caregivers, and, in the case of consulting professionals for advance directives, American caregivers who usually consulted lawyers, whereas their Belgian peers relied on physicians. We concluded that specific nuances and challenges in terms of ACP in YOD arise from a particular societal and legal context on the one hand (e.g. which end-of-life decisions are discussed), and from patients' and caregivers' younger age on the other (e.g. issue of financial planning).

In CHAPTER 3 we inquired physicians from four different specialisms about their experiences with and their perspectives on discussing ACP with persons with young- and/ or late-onset dementia[3]. Throughout our results we noted a high consistency regarding the viewpoints of respondents from different medical specialties. Physicians acknowledged that ACP can be of added value to people with dementia and particularly to their family caregivers. However, engaging in the process was considered to be challenging due to different factors (such as clinical uncertainty and moral constraints). It emerged that physicians believe that the societal stigma related to dementia impacts how persons react to their diagnosis, at times holding catastrophic expectations for the future. In this regard, they explained that the topic of euthanasia is sometimes brought up very early in the disease trajectory by patients. Although physicians endorsed a broad conceptualization of ACP, they placed ample attention on actual end-of-life decisions, including 'do-not-resuscitate' codes, when discussing ACP in dementia. As such, in clinical practice a medicalized view on ACP appeared to still be dominant. As part of ACP physicians felt a responsibility to provide accurate information on both dementia as a condition, and the legal framework of end-of-life decisions. Most of our respondents believed that a wish for ACP was more driven by the personality of patients and caregivers, than by how old they are. Nonetheless, physicians did identify a specificity for a younger dementia population when they mentioned that ACP encompasses more domains of life.

Part III: Identifying the experiences with, perspectives on and preferences for ACP of people living with young-onset Alzheimer's disease and their family caregivers

CHAPTER 4 focuses on the experiences with and preferences for ACP of persons living with YOD and their respective family caregivers[4]. Throughout interviews, it was shown that people with YOD and their caregivers strictly associated ACP with medical planning for the end of life and were not familiar with its concept as a communication process. However, when considering ACP as a communication process, they paid a lot of attention to non-medical aspects of care and did not compartmentalize medical, social, and mental health. Rather, they broadened their narratives to "what matters to them now and in the future". Respondents had not or barely engaged in future (medical) planning, for which they provided several reasons (e.g. patients not worrying about the future and respondents' uncertainty about what the future holds). Nonetheless, both patients and family caregivers also reflected about the advantages of planning future care, and this mostly in relation to the family caregiver for whom this might be an emotional relief. An overly medicalized approach to ACP, restricted to planning the end of life, might impede people's engagement in the process. To attune ACP to our respondents' views, it is suggested to approach the process holistically, flexibly, and relationally.

CHAPTER 5 describes a secondary analysis of the interviews with people with YOD and their family caregivers of Chapter 4 and more precisely on their spontaneous expressions about euthanasia[5]. When they were asked about the broader topic of ACP, over two-third of our respondents spontaneously addressed the issue during interviews. They elaborated on the topic of euthanasia as such and, if they had ever discussed euthanasia, they also described how that communication had taken place. Our study showed that people with YOD and their family caregivers described similar contexts in which these discussions about euthanasia arose. The topic was brought up at several key moments (such as the moment of diagnosis), usually with a family caregiver, and was motivated by both personal patient considerations (the impact for the person with YOD), as well as by patients considering the consequences of disease progression for their loved-ones. Considerations of people with YOD about euthanasia appeared rooted in personal, as well as in expected interpersonal and societal suffering. People's anticipations for and ideas about the future seemed to be influenced by the negative image associated with both dementia as well as dementia care. Family caregivers also paid a lot of attention to the Belgian legal criteria under which euthanasia is allowed for people with YOD, specifically in the context of declining cognitive capacity. According to both patients and family caregivers, dementia is different from other conditions when it comes to euthanasia given that dementia still allows room for enjoyment. Family caregivers also elaborated on the emotional challenges they experienced when discussing euthanasia and, some, on the feelings of guilt after a loved-one with dementia was actually granted euthanasia.

METHODOLOGIC CONSIDERATIONS: STRENGTHS AND LIMITATIONS

Overarching strengths and limitations

Some of our strengths and limitations are applicable to several of the studies that we conducted to meet our research objective. As a qualitative approach was chosen throughout this dissertation, the most important strength is that we were able to present rich, 'thick' data regarding a sensitive and complex issue. More importantly, we have identified and described the experiences, perspectives and preferences regarding/of a patient population who has been largely overlooked (durably and globally) in research on end-of-life care. As this is the first doctoral dissertation to enter the new ground of ACP in YOD, it presents highly innovative results. Given that we included people with YOD themselves, as well as their formal, and informal caregivers, this thesis presents a complex issue from divergent angles (triangulation). We hope that through this work other researchers' curiosity will be sparked to conduct further studies in the domain of end-of-life care for people with YOD and their caregivers. In this regard, our studies might also offer guidance in terms of methodology (e.g. ethically sound inclusion of people living with YOD). Most importantly, we believe that the findings of this dissertation, and their applicability in practice, might improve the care that is provided to people with YOD and their relatives.

Due to reasons of 'convenience' when recruiting participants who were deemed not to be easily accessible, we have opted for purposive sampling in which only a limited number of inclusion criteria were put forward. An overall limitation, leading to necessary caution in interpreting our results, is the homogeneity of our samples. For instance, throughout our study, respondents had the same Caucasian ethnicity, which requires caution when generalizing to other ethnic groups. Additionally, aside from our second Chapter which also included American participants, we have solely recruited Belgian respondents. Given that Belgium has a specific medico-legal context for ACP, some of our findings might not be generalizable to other countries without or with another legislative framework for physician assisted dying. This is also suggested by the differences and similarities emerging from our Belgian vs. US comparative study. Particularly the findings regarding patients' and family caregivers' expressions on euthanasia (Chapter 5) and physicians' emphasis on end-of-life decisions such as euthanasia throughout focus groups (Chapter 3), might be subject to diminished generalizability. Considering the limitation due to purposive sampling, we believe our study insights would not only be expanded, but also refined if we would have been able to proceed with theoretical sampling, to ensure adequate representation of our results. Theoretical sampling means that participants are selected who can best contribute to the emerging findings and that, as such, data-collection and data-analysis concurrently take place[6,7]. However, our respondents were recruited independently from developments in analysis, hence this implies that our evolving analytic insights did not inform ongoing selection of participants. Although throughout chapters there was transparency regarding results for which data-saturation was (or was not) reached, theoretic sampling might have revealed necessary gaps to fill: gathering additional data from respondents with varying characteristics (e.g. in terms of educational level, social activity level, family composition etc.) would possibly stimulate nuance and revision of our findings. Nonetheless, the findings from this first exploratory study form an important steppingstone for further developing the international evidence base on the topic of ACP in young-onset dementia.

Interview studies

For our interview studies (Chapter 1, 2, 4), the design of semi-structured interviews was chosen[1,2,4]. This led to the general strength that respondents were enabled to touch upon issues that were not foreseen by the researcher. The methodologic value of allowing interviewees to spontaneously elaborate on topics, was seen from our secondary analysis (Chapter 5), which was purely based on such 'unsolicited expressions' regarding euthanasia. Given that the average duration of the individual interviews, across our studies, was about one hour, we were able to gather rich, in-depth data. For our Belgian caregivers (Chapter 1, 2, 4) and people with YOD (Chapter 4), interviews were conducted face-to-face, which facilitated in-depth exploration and enabled researchers to be prompted by respondents' nonverbal behavior. It might be deemed a limitation that interviews with American caregivers took place over the phone. However, this could conversely also be considered as a strength given that social desirability might have decreased without face-to-face interaction. Possibly, telephone interviews helped them feel comfortable enough to express certain delicate perspectives, for instance, regarding end-of-life decisions that fall outside the scope of the law[2]. A limitation regarding our interviews with family caregivers (Chapter 1, 2) is that patients' perspectives were only portrayed indirectly. This clarifies the major strength of Chapter 4 and 5, in which we included people with YOD themselves, and as such gained insight into their unique experiential knowledge[8]. A limitation stemming from recruiting dyads of people with YOD and their respective family caregivers, was that we did not inquire any patients without informal caregivers. As such, we did not gain any understanding about if or how the absence of close family caregivers impacts ACP. Additionally, we solely recruited people with the subtype of Alzheimer's disease. Therefore, we could minimize the confound in our results due to differences in clinical presentation (e.g. behavior in Alzheimer's vs. frontotemporal dementia). Conversely, this also meant that we were not able to simply assume our findings to be equally valid for people with other subtypes of YOD than Alzheimer's disease. Moreover, even within the population of people with young-onset dementia of the Alzheimer type, we should be careful to assume transferability of our findings. It is possible that a selection bias took place in the sense that we were referred to patients who were considered to have adequate awareness of changes and acquired deficits. Given that lacking awareness of such deficits (anosognosia) is common in Alzheimer's dementia and that the role played by such clinical entity in end-of-life decision-making is currently unclear[9,10], caution in terms of generalizing is necessary.

Focus group study

The main strength of our focus group (Chapter 3) study is that it assembled heterogeneous focus groups in terms of specialisms crucial in dementia care, allowing in-depth insights from and for various medical disciplines (general practitioners, geriatricians, neurologists and psychiatrists)[3]. Making the focus of our research question two-fold and inquiring about people with late-onset dementia as well as with YOD, led to findings that are insightful for clinicians, when caring for this underexposed group of younger patients with dementia. A limitation of this study, as is also inherently the case for our interview studies, is that we did not observe the actual practices of ACP, but analysed what participants shared about them.

Secondary qualitative analysis on patients' and caregivers' expressions on euthanasia

For our fifth chapter, we conducted a secondary analysis on a subset of interviews with family caregivers (Chapter 1, 4) and persons with YOD (Chapter 4) in which respondents elaborated on euthanasia without any prompts from the interviewer[5]. Given that euthanasia was not included as a topic in our interview guide, it was not discussed as ‘in-depth’ as it would have been if it were at the forefront of our research questions. As such, these unsolicited expressions of respondents covered a broad range of ideas, for some of which data-saturation might not have been reached. Nonetheless, the various viewpoints expressed portrayed unique experience-based perceptions on an undiscovered subject and held innovative value to both theory and practice.

GENERAL DISCUSSION OF THE MAIN FINDINGS

The current part is divided into six overarching themes, with their respective subthemes. An overview can be found below. The themes summarize and interpret the main findings from our research.

1. *Unmet prerequisites for ACP engagement in YOD*
    - Lacking awareness of what ACP is
    - Lack of information to meaningfully engage in ACP
    - Limited engagement in ACP, regardless of respondents' endorsement
2. *ACP and the role of family in YOD*
    - ACP is impacted by protective roles
    - Relational autonomy as a cornerstone of ACP in YOD
3. *A 'process over product' mindset could facilitate ACP*
    - Advance directives without communication seem incomplete for ACP
    - Process-approach could facilitate all participants' readiness for ACP
    - The impact of uncertainty on the process of ACP
    - Balancing act between preparing for the future and enjoying life day by day
4. *How society regards and cares for people with YOD impacts ACP*
    - Society's stigmatic portrayal of dementia might impact patients' perspectives
    - Quality of care and its potential link to ACP
5. *How a country's legal context impacts ACP in practice*
    - Legal framework impacts a person's thought framework
    - Respondents discuss current policy debate
6. *The specifics of ACP for people with YOD and their family*
    - Broader content and family-centered approach to ACP in YOD
    - Difficult to differentiate dementia-specific vs. age-specific findings

1) ***Unmet prerequisites for ACP engagement in YOD***

Lacking awareness of what ACP is

Amongst people living with YOD and family caregivers, we found that awareness of the concept of ACP, especially as a communication process, was lacking. If respondents had a notion of the concept, they discussed ACP as if it was mainly focused on decision making for or at the actual end of life (i.e. last days or hours of life). This was in line with our findings among professionals. Although physicians were familiar with the broad description of ACP as a communication process provided at the start of focus groups, they often re-directed the conversation to end-of-life decisions such as Do Not Resuscitate-orders or euthanasia. Somewhat contrary, we found that many participating family caregivers, physicians and some patients preferred not to wait too long after a diagnosis of dementia to start talking about ACP, a finding in line with other studies[11,12]. Preferring ACP to start 'in time' yet discussing ACP as in such close proximity to death, seems to point to a discrepancy between theory and practice.

Additionally, although physicians agreed that ACP should not limit itself to conversations about purely medical decisions, they did place great emphasis on medical end-of-life decision making throughout all focus groups. This rather narrow view on ACP as focused on making medical decisions for the actual end of life, emerging throughout this research, does not correspond to the broader communication process that ACP evolved to in academia[13,14]. Interestingly, when patients and caregivers were informed about this evolved concept during interviews, they broadened their thought framework for discussing care to "what is important now and for the future?", with ample attention to non-medical domains (for instance financial planning or the importance of social activity). This frame of thought appears to match physicians' perspectives about ACP needing to be a broader topic in case of young-onset, as compared to late-onset dementia, given the multitude of life areas possibly affected due to a diagnosis earlier on in life. The conceptualization of ACP in research[15], i.e. an ongoing process which is not a purely medicalized matter, should trickle down more to practice as this would correspond better with patients' and caregivers' thought framework on what matters most. For patients and their families, care is not compartmentalized in distinct elements, but entails medical, social as well as mental health intertwiningly (Chapter 4). Such interconnectedness of physical, social and mental wellbeing became apparent from our results (Chapter 4) regarding, among others, patients' reluctance for residential care, their fear for social isolation and the wish for dignified care. As our results showed that the process in YOD should address domains of care beyond solely medicine, the need for a holistic view on ACP is underscored. This corroborates a recent scoping review showing that, although holism is one of the core principles of palliative care, challenges still exist in its implementation in palliative care practice[16]. One specific finding was that open conversations, as opposed to assessment tools, lead to a more comprehensive understanding of the complexity of issues that people may experience in more than one care domain[16]. Providing holistic care, therefore appears to require that health care professionals make a clear commitment to actively and openly communicate with patients and caregivers, including on psychological and social matters.

Lack of information to meaningfully engage in ACP

The unmet need for information in dementia has been a well-established finding in literature[17,18,19]. A mixed-method study with family caregivers of persons with YOD showed that 70% of respondents experienced unmet needs, and that those were mainly of a psycho-educational nature[19]. Besides a lack of information on what ACP is and how to engage in it if wanted, persons with YOD and their caregivers in our studies also explained to be confronted with a lack of information regarding dementia itself, the expected course of disease progression, and the service provision available. According to patients and caregivers in our study but also across countries as shown from a review[20], uncertainty and unpredictability about what the future holds is a reason for them not to engage in ACP. Physicians on their part also elaborated on the clinical uncertainty they dealt with, for instance regarding the speed of disease progression in an individual case, as also seen in previous research[20,21,22]. Nonetheless, physicians agreed that certain milestones in a dementia disease trajectory, such as declining cognitive capacity, will inevitably present themselves sooner or later and should therefore be discussed with patients and caregivers. As a part of ACP, physicians in our study considered it their responsibility to provide adequate information about dementia and its disease prognosis, yet they also described moral constraints they felt in doing so, as found also in previous reviews[22,23]. Taken together, adequate information provision (about a YOD disease trajectory, ACP, and available services and resources) is a necessary first step for enabling ACP engagement in YOD, according to all stakeholders involved.

Limited engagement in ACP, regardless of respondents' endorsement

Regardless of (mostly emotional) constraints that family caregivers held towards starting a conversation about ACP, they acknowledged the importance of the opportunity to engage in the process. This became clear through the preferences they shared in all studies: i.e., they wished for a professional caregiver to initiate the process, in a timely manner and with a focus towards patients' remaining capabilities. Our finding of such explicit preferences, combined with the finding that our respondents had not or had barely engaged in ACP, suggests that the lack of ACP engagement is the result of an inability to do so, rather than a result of an unwillingness to do so. A gap of knowledge (about ACP and about dementia) and a gap of information (how can one engage in ACP and what to plan for in dementia*)* seem to hinder ACP engagement in YOD (Chapter 1, 4). Physicians in our focus group study also acknowledged the added value of ACP in case of dementia yet described several challenges for actually engaging in the process. Similar to previous research[20,21,22,24,25], some of these difficulties were related to dementia as a condition (e.g. uncertain speed of progression); others were associated with moral constraints for engaging in ACP conversations (e.g. not wanting to distress patients) according to our respondents.

2) ***ACP and the role of family in YOD***

ACP is impacted by protective roles

Alongside the actual unpredictability of an individual disease progression, physicians explained that providing prognostic information is difficult due to a fear of taking away patients' hope, a finding in line with a recent systematic review[22]. Family caregivers also clearly identified their motive of trying to safeguard their loved-one's emotional wellbeing. In this regard they, for instance, at times deliberately chose to focus on the present and not discuss the future with its unfavorable nature, a coping mechanism also previously described[22]. In line with prior qualitative research, the overall experience of family caregivers is profoundly shaped by the aim to protect a loved-one with dementia[26]. However, by not discussing the future, family caregivers stated to also protect themselves; they attempted to avoid potential feelings of guilt for distressing the patient. Family caregivers appeared to find themselves in a field of tension as they simultaneously explained that engaging in ACP would be a means of protecting themselves. More precisely, it would give them 'peace of mind' and diminish emotional burden in the future if they were to know the patient's preferences (Chapter 1). Families trying to protect loved-ones whilst compromising their own need for dialogue was similarly reported by Sussman et al.[27].

A systematic review[28] on patients' experiences with ACP showed, however, that people with life-limiting illness assume that being open about their wishes for care would reduce burden for their loved-ones. In our study, patients on the one hand and physicians on the other, also regarded the emotional relief that ACP could bring for family caregivers as its main benefit. As such, all those involved appeared to identify engaging in ACP as a possible caregiving act of patients towards their family. Our respondents thus seemed to support the previous suggestion[27] of framing ACP as an act of care from a person with dementia towards his/her caregivers. In line with this reasoning, the non-medical plans that patients had made and specifically those for after death, such as financial or funeral arrangements, could equally be considered as a caring act towards their family. Our study therefore suggested that patients' intentions as well as family caregivers' intentions for ACP engagement are shaped by a wish to offer protection to their loved ones.

Familial relations as a cornerstone of ACP in YOD

Patients, family caregivers and physicians all highlighted the importance of addressing families when providing care, rather than exclusively focusing on the person with YOD. Solely a person-centered approach, usually recommended in end-of-life care as well as in dementia care[11,12,29,30] might therefore not be sufficient in the case of YOD. According to our respondents, adequately meeting the needs of young people who face dementia (when possibly having young, working spouses and children or adolescents at home, and/or potentially carrying significant financial commitments[31,32]), might require a care approach that targets a family, rather than just the affected individual. Physicians who explained that ACP in younger people with dementia should cover a wider range of topics and address more domains of life, appeared to acknowledge the scope of living with YOD for an entire family. Moreover, patients', family caregivers' and physicians' views on the benefits of and desire for ACP, mainly revolved around considerations and consequences of the YOD diagnosis in the interplay within a family (Chapter 1, 3 & 5).

Equally, an important consideration for people with YOD when thinking about the future was not wanting to become a burden to loved ones, yet again showing the abovementioned protective role patients take on towards their family (Chapter 4 & Chapter 5). Both previous[33] as well as our own studies showed that patients with young-onset dementia wish to spare not only themselves, but also their relatives from suffering. Additionally, within respondents' perspectives on euthanasia, we found that they paid a lot of attention to familial relationships (Chapter 5). An article describing the clinical considerations in physician-assisted death for probable Alzheimer's disease, point to the attending physician having to consider physical, psychological and social factors that influence a patient's health status and in particular, vulnerability represented in, among others, a loss of autonomy[10]. More generally, acknowledging relations as part of decision making appears necessary for families to engage in ACP. Adopting a framework of relational autonomy[34], in which interactions of stakeholders and the influence of a sociocultural context is considered, could make the process more accustomed to those confronted with YOD. This suggestion can be best interpreted as an addition to, and by no means a detraction from, the central principle of patient autonomy.

3) ***<u>A 'process over product' mindset could facilitate ACP</u>***

Advance directives without communication seem incomplete for ACP

Family caregivers mentioned that advance directives might be a good starting point for initiating a broader conversation on ACP, as also suggested by general practitioners in previous research[35]. Nonetheless, we also found that many of the American and Belgian patients who had completed an advance directive, did so without engaging in in-depth discussions with family, nor with professional caregivers. Legally, advance directives generally do not need to be discussed with, completed in the presence of, or signed by a physician[36]. However, filling out advance directives without any accompanying discussion with physicians is a longstanding observation as shown also from previous research in cancer[37]. As discussed in Chapter 2, and particularly regarding American respondents, this might indicate a sense of 'form-over-substance' in which making sure documents are filled in correctly is regarded as more important than discussing their actual content in terms of preferred care. Such approach could jeopardize the meaning of documents in clinical practice. Moreover, it was previously shown that people with dementia and their caregivers are at times hesitant about engaging in ACP because previous care decisions could ‘lock them in a certain pathway’[20] and that patients with life-limiting illness might fear the consequences of written documents when they feel they are not able to change them later[28]. Considering advance directives as “living” instruments, as opposed to end-goals, that can guide dynamic and ongoing communication, could therefore increase patients’ and caregivers’ willingness and readiness for the process (Chapter 2, 4).

Process-approach could facilitate all participants’ readiness for ACP

Several of our insights suggested that a focus on process, rather than on product, could facilitate the initiation of and engagement in ACP for all those who participate in the process. Aside from many patients’ and family caregivers’ preference to take it one day at a time, physicians’ responsiveness towards individuals’ changing information needs and step-by-step information provision on ACP and on dementia, might enable people to gradually incorporate thinking about the future. This might, as such, increase people’s readiness for the process itself (Chapter 4). For physicians, a more process-oriented view on ACP might diminish an ethical issue that they experience: they explained, as did patients and family caregivers in our study, that it is very difficult to predict how people’s preferences for care will evolve throughout a disease trajectory. In this sense they said to struggle with making ‘premature’ decisions for medical situations that might only take place several years later, as found also in other research[38]. Throughout our focus groups, physicians also referred to people with dementia who, as it goes, find their trajectory more manageable than initially expected, which makes current preferences insufficient to guide future care decisions (Chapter 3). Differences in ACP definitions are seen in whether it focuses just on decision making for future medical care or also incorporates decision making for current care[28]. Our findings, from all respondent groups, point to the desirability to discuss both the present, as well as the future. Gradually and continuously engaging in discussions about preferences might attune ACP to the principle of a patient’s evolving wishes and boundaries throughout a disease trajectory. In anticipation of such a ‘response shift’ in dementia, ACP (documentation) should be evaluated regularly, as previously suggested[12]. Moreover, we agree that tailoring ACP to people with dementia and their caregivers should include a recognition that ACP is likely to be going a repeated process over time[29]. Additionally, a previous qualitative study showed that caregivers of people with dementia usually find ‘day to day’ decision making so burdensome that it prevents them from planning in the longer term[17]. In the light of these previous and our own findings, the importance of ACP as an ongoing, dynamic dialogue about what constitutes adequate present and future care for people with YOD is yet again underscored.

The impact of uncertainty on the process of ACP

Our studies showed that uncertainty is a stumbling block for engaging in ACP for all those involved. As also emerged from a recent review[20], patients and family caregivers in our study explained that uncertainty regarding dementia, its trajectory, and ACP are impeding factors for engaging in the process. Throughout the focus groups, it emerged that physicians consider information provision on dementia, on ACP and on its legal framework, as an important responsibility in clinical practice. As such, it is clear they wish to counter the issue of lacking knowledge and lacking information. However, physicians also explained that they struggle with clinical uncertainty about for instance the speed of progression, which they said can refrain them from discussing prognosis with patients and family caregivers. In order to start bridging the gap in care and establishing an opportunity for ACP as an integral part of care for people with YOD, it might be recommended that physicians are given room within ACP conversations to also attend to and be open about their own uncertainty. In a recent review on end-of-life-communication strategies[30], it was similarly suggested that health care professionals openly offer an explanation about their uncertainties as a part of information provision. A specific suggestion of how to do so was illustrating several scenarios to patients: the worst case, the best case and the most likely case[30]. Additionally, the realization that uncertainty as a barrier to ACP is a commonality between professional caregivers on the one hand, and family caregivers and patients on the other, might lower the threshold to address these sensitive topics and might empower all parties in the initiation of ACP (Chapter 3). Given that a level of uncertainty is an inevitable characteristic of dementia, it seems necessary to counter the potential prejudice that uncertainty is a ground for excluding ACP from care. One of the aims of the process is precisely to prepare people for ‘in-the-moment decision making’[13,14] through knowing the values, preferences and goals of care of people with dementia.

Balancing act between preparing for the future and enjoying life day by day

When discussing ACP, respondents paid a lot of attention to anticipatory decision making for the actual end of life, such as DNR-orders, advance directives and euthanasia (Chapter 3,4,5). However, preparing people for the abovementioned 'in-the-moment decision making' might be a way to attune ACP to their own preference of focusing on the present. A lack of knowledge on what the future holds, at what pace the disease might progress and what plans can be made (Chapter 4), was explained to contribute to an attitude of 'living life day by day'. The preference of taking it one day at a time, also shown in other research[12,20], was demonstrated by both people living with YOD and their family caregivers (Chapter 1, 4). In previous research it was suggested that living in the moment does not preclude ACP in dementia but does make discussions more difficult[12]. Family caregivers explained that problems are so unpredictable, that they were typically addressed in an acute manner, namely when they actually occur, even though these problems were at times foreseeable (e.g. related to cognitive incompetence). Physicians not addressing the problems that are likely to present themselves in the future, might lead to the potential significance and added value of ACP being unclear or minimized[39] (Chapter 1). Although physicians acknowledge the importance of providing adequate prognostic information, they explained that disclosing such information might be hampered by constraints in honestly communicating about disease progression (Chapter 3). Such moral threshold appears justified to a certain degree, as qualitative research shows that people with dementia and their family oscillate between 'wanting to know' and 'not wanting to know'[40], and that focusing on the present is associated with experiencing fewer unmet needs than worrying about the future[41]. However, a recent review[23] also showed that particularly family caregivers have a need for both realistic and hopeful communication about the future implications of their loved one's diagnosis. Having a truthful view on what the future might hold, yet allowing for hope and positivity to simultaneously exist, seems to be a helpful balancing act to be undertaken by everyone who is involved in ACP in YOD[3].

4) How society regards and cares for people with YOD impacts ACP

Society's stigmatic portrayal of dementia might impact patients' perspectives

A recurrent theme throughout this doctoral study was the way society looks at and cares for persons with YOD (and dementia in general). Through their own narrative, it appeared that patients with YOD experienced events where they feel underestimated and discriminated: people easily (and wrongfully) assume that they are no longer able to do or to understand certain things (Chapter 4). A systematic review of depictions of dementia in popular culture[42] showed that popular media often focusses on portraying the terminal stage of dementia which then becomes representative of the entire trajectory, a topic on which physicians in our study also elaborated (Chapter 3). Physicians believed that the stigmatic portrayal of dementia within our society and through media leaves little room for nuance in people's image of the condition, and as such negatively impact patients' fears and concerns about the future. Since it was shown that younger, in comparison to older people with dementia are subject to consistently higher stigmatic attributions[43], this might be the case even more for people living with YOD. Moreover, recent qualitative research showed that family and professional caregivers of younger people with dementia also encounter stigmatic experiences, mainly due to people's lacking knowledge about the condition[44].

Importantly, a previous qualitative study with people with dementia[45] showed that patients themselves create a more nuanced, and at times more positive outlook on dementia, when speaking about how they experience their life as meaningful and valuable. So did both people with YOD and family caregivers, as well as the physicians who took part in our studies: to a certain extent they have contradicted a 'solely catastrophic' view of dementia, when explaining that room for enjoyment and wellbeing still exists after receiving a diagnosis (Chapter 1, 5). Corroborating another recent finding[33], these results suggest that the burden of dementia might be assessed differently by patients and caregivers than by 'healthy' individuals or society in general. To obtain a more balanced and subtle outlook on YOD (and by extension on dementia), it appears necessary to prioritize the inclusion of perspectives of people with dementia themselves, in media, but also in public and policy debates, as well as in research. Moreover, our findings support the previous idea that ACP in dementia[12] should include health care professionals to explore possible misconceptions about the disease trajectory and provide a balanced view of what life can be like living with dementia. In the context of people with dementia seeking physician-assisted death, it is emphasized that they must understand the range of prognostic outcomes and experiences among patients[10,46]: adequate and effective education on the realities of life with dementia is needed for people in the early stages of the condition[46]. For such communication to take place, a good and trusting patient-physician relationship appears paramount[10,47] (Chapter 1, 3).

Family caregivers' recommendation that ACP should focus on a patient's remaining capabilities, instead of on acquired difficulties, could be regarded as a reply to the stereotype -described by patients themselves- of people with dementia as 'no longer competent'. Maximizing people with dementia's opportunity to take part in ACP, might in itself be a means to counter such stigmatic beliefs, as it enables them to be active agents in self-managing care, and have their capabilities, as opposed to their potential disabilities, addressed and utilized (Chapter 4). A systematic review of ACP experiences of patients with life-threatening or life-limiting illness indeed showed that participating in ACP made patients feel heard and respected[28].

Quality of care and its potential link to ACP

A discrepancy found between patients' and family caregivers' perspectives on the one hand, and physicians' perspectives on the other, was whether they shared views on the quality of care provided to persons living with YOD. Whilst physicians did not speak on this topic in the context of ACP, patients and family caregivers addressed it quite extensively. They explained that care in general is too fragmented to be easily accessible, that they are required to 'unravel' information about care provision and that especially residential care is not adjusted to younger patients. The lack of accustomed care provision for people with YOD is a durable problem as shown from experiences portrayed in previous research[48,49,50]. Patients' and family caregivers' perspective on the lack of appropriate care might cause them to not feel incentivized for engaging in ACP (Chapter 1, 4): even though one would plan for care, the actual care received would still be expected to be substandard. The finding from other research[27] that thinking about decline in dementia is possibly more threatening than thinking about death, might be related to both negative beliefs about dementia, as well as to negative beliefs about dementia care.

Moreover, our studies also revealed that patients' concerns about care might fuel discussions on end-of-life decisions. Both people with YOD and family caregivers, as well as physicians commented on patients' specific reluctance to ever be enrolled in residential care. Physicians further explained that patients' unwillingness to move to a care facility, was at times even a motive for them to express a euthanasia request. More generally, the sometimes 'impulsive' requests for euthanasia from patients (as perceived by the physician), usually not long after receiving their diagnosis, were seen as motivated by a concern or a fear that could be addressed differently. As with advance directives, these exclamations about euthanasia could be grasped as a steppingstone towards wider communication about concerns and needs, according to physicians. Previous research indeed showed that ACP is initiated earlier if patients express a wish for euthanasia[47]. However, our Belgian family caregivers (Chapter 1) also stated that euthanasia was at times only briefly discussed once, because physicians were, according to respondents, not inclined to discuss the subject in-depth. If physicians omit to further explore a patient's reasons for bringing up the topic of euthanasia, an opportunity to timely initiate ACP might be missed.

5) ***<u>How a country's legal context impacts ACP in practice</u>***

Legal framework impacts a person's thought framework

Corresponding to a previous finding in amyotrophic lateral sclerosis research[51], several of our results suggested that persons think about end-of-life decisions within the framework that is depicted by a country's legal framework for such decisions (Chapter 2, 5). Moreover, we found this to be the case within each respondent group: whilst most of our Belgian respondents (patients, family caregivers and physicians) elaborated on the topic of euthanasia, our American respondents (family caregivers) did not. Rather, they discussed the end-of-life decisions encoded in their nation's law (refusal or withdrawal of treatment), and at times suicide or decisions that fall without the scope of the law. If within a country certain end-of-life decisions were legalized, these options appeared to be 'naturally' considered in a person's thought process on end of life. It was recently stated that -amongst other- legal differences across countries may affect communication strategies for the end-of-life and that these differences should therefore be recognized in clinical settings[30].

Our studies show that Belgian persons with YOD and their family caregivers incorporated the topic of euthanasia when thinking about end of life (Chapter 1, 2, 4, 5) and that patients at times bring up the topic very early on in their disease trajectory. Aside from our recommendation for holistic ACP, it could therefore also be seen as a sensitivity from physicians towards their patients that they pay sufficient attention to medical decisions for the actual end of life (as they did throughout focus groups, Chapter 3). It is advisable to combine discussing specific concerns patients might raise (such as euthanasia), while simultaneously being open to discussing rather social and relational domains of ACP and care[47]. Specifically in the context of end-of-life requests by people with Alzheimer's dementia a similar recommendation was previously made, namely that psychosocial and existential suffering are to be regarded as part of the medical domain[10,52].

Respondents discuss the current policy debate

The current political and public debate in Belgium on the legality of advance euthanasia directives and, as such, the timeframe for euthanasia requests for people with dementia, was also reflected in our findings. Particularly family caregivers and also some patients paid attention to the legal criteria for euthanasia in Belgium, particularly in the context of declining cognitive capacity, and showed varied opinions on the matter (Chapter 1, 4). Following a prior suggestion[53], the responsible use of law as a tool for improving public health, requires a commitment to the pursuit and to the consideration of scientific evidence. However, a systematic review on the attitudes of patients and caregivers, health professionals, and the public showed that the perspectives of people with dementia and their caregivers are portrayed in only a very limited number of studies, which are mostly of low quality[54]. The spontaneous remarks of some of our respondents minimally show that the matter is of importance to them, that they are willing to discuss their perspective and that they are a voice to be heard in debates regarding the euthanasia law. Physicians also discussed the issue, yet mainly in the sense that they felt responsible for providing adequate information on what is possible or impossible for people with dementia under Belgian laws (Chapter 3).

6) The specifics of ACP for people with YOD and their family

Broader content and family-centered approach to ACP in YOD

Koopmans et al.[55] hypothesized that people with YOD and their family caregivers might have distinct needs for palliative care and specifically ACP when compared to their older peers, due to desiring higher involvement and autonomy in medical decision-making. Through the narratives of patients (Chapter 4, 5), family caregivers (Chapter 1, 2, 4, 5), or physicians (Chapter 3), we did not find evidence to support the suggestion[55] that people with YOD wish to play a more active role in ACP. All respondent groups did point to specificities in terms of the content of ACP when people with dementia and their family caregivers are younger. More concretely, according to respondents, ACP in YOD covered a broader range of domains when compared to late-onset dementia, mainly due to younger people's phase of life. One specific topic that was addressed by family caregivers and by patients themselves (Chapter 2, 4), was financial planning undertaken after the diagnosis. Amongst our caregivers, finances posed more of a concern to American than to Belgian caregivers: providing financial security was regarded by them as an important element of their caregiving task. However, it seems plausible that generally unpaid caregiving hours have a higher impact on families facing young-onset, as opposed to late-onset, dementia since younger caregivers more commonly have got to cut back on paid work in order to take care of their loved one (Chapter 3). It was recently reiterated that the management of YOD requires the consideration of age-specific issues, such as improved access to financial assistance[56]. Also, the need for patients to stay socially and physically active was underscored throughout our interview studies. Allowing room in clinical practice for those type of subjects, might open up communicative space for discussing other aspects of health, care and specific concerns that might be more salient in younger patients and relatives (e.g. providing for a family with children at home or at university when having to diminish or stop work). Agreement among our respondents indeed existed that professional caregivers need to pay heightened attention to non-medical domains to address ACP in this population (Chapter 3, 4). In terms of the process of ACP in YOD, a specificity mentioned by all respondents is the importance of targeting an entire family, as opposed to focusing merely on the affected individual. As with other recommendations, this could be equally applicable to older people living with dementia as it corresponds to existing guidelines for ACP in dementia; namely professionals' need to involve family or significant others in ACP as soon as possible[12].

As concluded in Chapter 3, generic ACP guidelines for professionals are necessary and useful. In addition to more general guidelines, it is important to also include nuances which might exist in a specific patient population like YOD. In summary, these nuances and/ or challenges appeared related to the particular societal and legal context in which people find themselves on the one hand, and to also flow from patients' and family caregivers' younger age on the other.

Difficult to differentiate between dementia-specific vs. age-specific findings

Many results from our studies, however, were not explicitly linked to the younger age of patients and family caregivers who face YOD. As discussed in Chapter 4, we do not rule out that our findings, as well as our recommendations, might be generalizable to older people with dementia and their respective family caregivers. Conversely, our results regarding needs for care initiatives aimed at a young family and the difficulty of finding age-appropriate care for younger patients might not be specific to dementia[4]. A recent scoping[57] review indeed pointed to the lack of options that younger people with disability, for instance due to acquired brain injury, have in terms of residential care facilities. As such, we believe that some of our insights might be generalizable towards younger persons who live with a different life-changing condition than YOD.

We are also aware of Belgium's specific medico-legal context for conducting research in the field of end-of-life care. As shown from our comparative study (Chapter 2), not all findings can be simply transferred to another societal and legal context. However, in our study on euthanasia considerations (Chapter 5), we equally found age- vs. dementia- specific results that could apply to patients in other contexts as well. For instance, trying to avoid residential care as an age-specific factor and the issue of declining cognitive capacity as a dementia-specific factor.

Although we are not able to provide certainty based on our current studies and therefore future research is needed to gain evidence, we hypothesize that our findings are a combination of cohort-specific and disease-specific results. Topics like financial considerations are highly likely to be related to the cohort of our respondents and can therefore also be expected in younger people with other (chronic) conditions. Conversely, issues such as the impact of a ‘day-by-day’ attitude on ACP, are highly likely to be an extension of a dementia diagnosis, rather than rooted in patients’ and caregivers’ age.

RECOMMENDATIONS FOR PRACTICE, POLICY AND RESEARCH

Based on the different findings throughout the studies in this dissertation, several recommendations for practice, policy and future research can be made.

FOR PRACTICE

Through our interviews with patients, family caregivers and physicians several recommendations for practice emerged. Although solely physicians whose specialisms makes them essential in dementia-care were recruited for our focus groups, many of these recommendations may apply to or be of benefit for other professional caregivers.

Dynamic, family-, and process-oriented ACP

Based on the clear need for information and family caregivers' preference for professionals to timely initiate the process, physicians should fulfil an active role in explaining ACP and initiating those discussions in case of YOD. Adequate information provision about both dementia and ACP can be regarded as a necessary first step for enabling people with dementia and their caregivers in the process. In accordance with a previous suggestion[58] we support the idea that disclosure on prognosis and familiarizing people with ACP should be in itself embedded as a process within a YOD trajectory. Overall, the process of ACP should be the focus, rather than the plans it produces. To counter several difficulties that we identified for engaging in the process (such as patients' and caregivers' day-to-day attitude and physicians' clinical uncertainty), ACP could be best considered as an ongoing dialogue about what constitutes adequate care. Also, based on our findings that both patients' and caregivers' intentions for ACP are shaped by a wish to protect each other, professionals might raise a dialogue about the impact of mutually protective roles.

ACP as a conversation with a holistic focus

As we found that patients and caregivers do not divide medical, mental and social health when thinking about care and expressed needs in these areas, we recommend that physicians pay heightened attention to non-medical domains when engaging in ACP in YOD. In medical practice, the overarching framework of "what is important to people in terms of the present and the future" could serve as a starting point for ACP, as this matches patients' and caregivers' own ideas of it. For instance, health care professionals could explicitly ask people about potential worries regarding health care costs or social connectedness, as these topics might facilitate a wider conversation about (long-term) care. As also suggested by others[59], ACP should be advocated as a means to achieve what matters most to people.

Advance directives seem to be most useful in clinical practice, when they are instruments for creating communicative space and are implemented as living documents to be re-discussed.

To facilitate care congruence between professionals -even from distinct domains (e.g. health care, law)- and to increase the probability that completed documents will be of meaning in clinical practice, health care professionals should actively ask patients and caregivers about what they might have planned for already, with who and should offer further discussion if desired.

Addressing the societal and legal context in which ACP takes place

Topics regarding our society's 'negative' image of dementia might deserve a place within consultations since they appear to influence patients' and family caregivers' expectations of the future. Maximizing people with dementia's opportunity to take part in ACP could be regarded by itself as a way for countering stigmatic beliefs about living with dementia since it allows patients to actively use their capabilities, rather than have potential disabilities emphasized.

It appears recommended to offer people with YOD the opportunity to discuss medical, social, and relational domains as part of ACP, yet simultaneously elaborate on particular concerns or questions patients might have, such as regarding euthanasia. Professionals being aware of and responsive to the legal specificities of ACP in their respective country, could facilitate the process of ACP. Our study on patients' and caregivers' expressions about euthanasia reiterated that professionals being open to conversations about concerns and fears, possibly including those about (hastening) death, can be regarded as a service to people with YOD and their family caregivers[3,60]. Patient-physician communication should include openly discussing potential fears, a (step-by-step) reflection on prognosis and detangling motives for possible euthanasia requests. Based on our finding that the consequences of YOD for their close relatives were part of patients' euthanasia considerations, it seems important within practice to clarify and detangle to the extent possible this reason from the motive of unbearable personal suffering.

FOR POLICY

Uniform messaging about ACP as a process of communication

Policy makers have the task of contributing to a more consistent public discourse about what ACP is. This can be done by organizing larger information interventions to raise both professionals' and the general public's awareness of ACP as a communication process. Policymakers, as well as clinicians, have a joint task of creating a uniform message towards patients and their family about the evolved concept of ACP nowadays[4]. Equally regarding the legalities of ACP, thorough and accessible top-down information transfer from governments to both health care professionals, as well as to the general public is needed.

Re-evaluating and improving YOD care where necessary

Based on our findings that many people with YOD and their family caregivers have an image of YOD care that is inadequate, it appears necessary to thoroughly re-evaluate and improve the current care and services provided to people with YOD as needed. Since lacking care was identified as a possible stumbling block to see an added value in ACP, this might secondarily incentivize people with YOD and their relatives to engage in the process. Within care initiatives, attention should be paid to social, physical and mental domains as they are not distinct elements in people with YOD's and their caregivers' view of care. Moreover, the Belgian law on euthanasia states that all reasonable alternatives need to be tried before granting a person's euthanasia request. It seems evident that these alternatives ought to be deemed reasonable and adequate from the patient's perspective as well[5]. As we found, for instance, that wanting to avoid residential care was at times a motive for people with YOD to express a euthanasia request, it seems evident that public and political debates about the legal criteria for euthanasia, such as capacity, should co-exist with initiatives on care-improvement (Chapter 5).

Including patients' and caregivers' perspectives to inform policy debate

Policymakers should make it a priority to include the perspectives and the opinions of patients and caregivers in the current political and policy debate, as their lived experience leads to unique insights (on the practice of) end-of-life decision-making in dementia.

FOR RESEARCH

Repeating and extending research against other social and legal contexts

Certain results in this doctoral thesis might be less or not generalizable to other legal contexts, which have no or different laws on physician-assisted dying. Our second Chapter (the Belgian - US comparative study) shows that differences for thinking about end-of-life decisions and ACP are highly likely to flow from differences in the legal framework for those decisions. Subsequent studies should provide further insight through in-depth inquiry of the potential influence of legislation on ACP and ACP communication.

Our fifth Chapter dealt with the spontaneous expressions of people with YOD and their caregivers on the topic of euthanasia. However, as this was rather an 'accidental finding', the issue of euthanasia (physician-assisted dying) should be put at the forefront of studies to deepen our understanding of patients' and caregivers' opinions[54,61]. Moreover, the way in which other legislative and social contexts possibly impact patients' and caregivers' considerations regarding care at the end of life, should be the subject of further qualitative research that compares findings from various countries. These insights should be regarded as highly valuable, or even indispensable, to inform patient-centered policy developments.

Broadening respondent groups

Throughout our studies, our biggest respondent group were family caregivers of people with YOD of the Alzheimer type, of which the vast majority were female, spousal caregivers. As previous research shows[62], the experience of taking care of a person with dementia is shaped by both the relationship to the patient, as well as by the gender of the caregiver. For broadening our understanding, it would be interesting to repeat our studies with family caregivers with different characteristics (for instance children, male). Including patients' children as respondents, would additionally allow to compare our results with those from a younger generation. As such it would be possible to identify whether our findings are due to a cohort effect (namely related to the age characteristic of our spousal respondents) or whether our findings reflect an experience specific to taking care of a person with YOD (or both).

Future studies should continue including people with YOD themselves. Two specific recommendations can be made in this regard. First, it would be insightful to also include patients with a different dementia diagnosis than Alzheimer's disease, as the possible impact of clinical presentation on the process and content of ACP might be identified. Second, it is important to also include people with YOD without any close family caregivers, as their decision trajectories might differ[63]. To gain an inclusive understanding, dyads of patients and their respective family caregivers, as well as patients without any relatives or friends who provide help should be involved in research. More generally, as the cut-off score for YOD is somehow arbitrary given that it is based on social, rather than biological factors, it would be advisable to deepen our understanding of such impacting 'social' factors rather than regarding age as the main (or sole) defining feature in YOD. Therefore, it would be insightful to pay sufficient attention to an individual's circumstances such as work life, social activity, familial context, physical health and so on, when trying to understand their specific context in terms of ACP or end-of-life care.

Associated to a limitation of the current dissertation, namely that of homogenous respondent groups, it is strongly advised to strive for a deepening of our present understanding. Continuing this research, more attention should be paid to characteristics of people living with young-onset dementia beyond the factor of age (in terms of, for instance, subtype of dementia, educational level, familial composition, ethnicity, etc.). When we do so in future studies, our current results (coding tree) could serve as a starting point for rather deductive thematic analysis and could be followed by inductive ways of thematic analysis in order to add dimensions to our present knowledgebase.

Given that we have solely included physicians as formal caregivers, it is recommended to widen this respondent group in future research. Bearing multi-disciplinarity in mind, also other professionals' perspectives and experiences (among others those of nurses and psychologists) need to be included for better understanding the clinical practice of this complex issue.

Conducting studies from real-life clinical practice

This doctoral thesis consists entirely of stakeholders' experiences and perspectives of ACP in clinical practice. As our findings are based on respondents' recollections, future research might focus on complementing those with evidence gathered in actual clinical practice. Observing ACP consultations with people with YOD through audio- or video-recordings and analyzing the communication therein (e.g. through conversation analysis[64]) might offer valuable insights into how specific communicative acts possibly impact the course of ACP discussions.

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PART V

SUMMARY

NEDERLANDSTALIGE SAMENVATTING

INTRODUCTIE

Voorafgaande zorgplanning (VZP) wordt beschouwd als een essentieel element van kwaliteitsvolle en van ethisch verantwoorde lange termijn zorg na een diagnose van dementie. VZP is een proces van communicatie tussen patiënten, mantelzorgers en professionele zorgverleners waarbij de voorkeuren van de patiënt inzake (toekomstige) zorg verkend worden, eveneens de zorg verleend aan het levenseinde. Het overkoepelende doel van VZP bestaat eruit om de kans te vergroten dat personen de zorg krijgen die ze zelf wensen, zelfs op momenten dat ze niet langer in staat zijn hun eigen keuzen te maken of uit te drukken. Dit opzet verklaart al meteen waarom VZP betekenisvol kan zijn bij Alzheimer dementie en soortgelijke neurodegeneratieve aandoeningen. Dementie betreft immers een progressieve aandoening, zonder huidige behandelmogelijkheden tot het verhelpen of stopzetten van ziekteprogressie. Ingevolge Alzheimer dementie zullen personen in toenemende mate moeilijkheden ontwikkelen op vlak van cognitie, geheugen en het dagdagelijks functioneren. Ziekteprogressie bij dementie zal vroeg of laat tot gevolg hebben dat patiënten niet langer over wilsbekwaamheid beschikken. Bij uitstek aan het levenseinde zullen mantelzorgers van personen met dementie geconfronteerd worden met mogelijk moeilijke medische keuzen voor hun geliefde.

Hoewel eensgezindheid heerst over de potentiële meerwaarde van VZP bij dementie, is de mate waarin aan VZP gedaan wordt bij uitstek laag in deze populatie. Er zijn aanwijzingen dat VZP bij dementie gepaard gaat met specifieke uitdagingen. Onder meer werd aangetoond dat artsen het belang van VZP erkennen, doch klinische, alsook morele struikenblokken ervaren om het proces te initiëren. Zo geven artsen aan dat ze, bijvoorbeeld, onzeker zijn over de prognose, hoe hieromtrent te communiceren en dat ze de patiënt geen hoop willen ontnemen of angst willen inboezemen.

Niettegenstaande dementie vaak geassocieerd wordt met oudere leeftijd, ontwikkelt tot 9% van de personen met dementie symptomen vóór de leeftijd van 65 jaar en is er bijgevolg sprake van jongdementie (young-onset dementia, YOD). Op heden wordt geschat dat er wereldwijd bijna vier miljoen mensen aan de aandoening lijden, hetgeen de nood aan adequate zorgverlening nogmaals onderstreept.

Jongdementie treft personen mogelijk ‘in de fleur van hun leven’. Niet zelden zijn ze nog actief op de arbeidsmarkt, hebben ze jonge kinderen, of dragen ze nog zware financiële verplichtingen. Voortvloeiend uit de actieve levensfase waarin mensen met jongdementie zich bevinden, wordt geopperd dat zij andere noden hebben inzake palliatieve zorg en specifiek inzake VZP. Concreet wordt verondersteld dat zij meer autonomie en een hogere betrokkenheid wensen in de besluitvorming omtrent zorgbeslissingen.

Desondanks bovenstaande hypothese, blijven de zienswijzen van personen met jongdementie aangaande VZP onontgonnen terrein binnen wetenschappelijk onderzoek. Bovendien bestaat er slechts gering inzicht in de perspectieven van hun mantelzorgers, ondanks de algemene aanbeveling om hen zo vroeg mogelijk te betrekken in het proces van VZP. Personen met jongdementie en hun mantelzorgers betreffen een langdurig en systematisch onderbelichte populatie in de literatuur. Daar wil dit onderzoek graag verandering in brengen.

Binnen dit doctoraat wensen we inzicht te verkrijgen via zowel zorgverleners van, alsook personen met jongdementie zelf omtrent hoe VZP kan worden afgestemd op hun noden en voorkeuren. Hiervoor zullen we ook peilen naar de zienswijzen van artsen omtrent VZP in zowel dementie ontstaan op jongere, als op oudere leeftijd.

METHODE

Op basis van onze verkennende, alsook gevoelige onderzoeksvragen werd voor dit volledige doctoraat geopteerd voor een kwalitatief onderzoeksopzet. Vier studies zijn gebaseerd op individuele semi-gestructureerde interviews met mantelzorgers en personen met jongdementie, een vijfde studie is gebaseerd op focus groepen met artsen.

Deze studies werden ongesplitst in twee delen, en meer bepaald op basis van hun respondenten. Deel 1 betreft onderzoek met zorgverleners van personen met jongdementie; zowel mantelzorgers, als artsen. Deel 2 bevraagt personen met jongdementie zelf, samen met hun respectievelijke mantelzorgers.

In totaal werden 48 individuele interviews afgenomen, waarvan 38 met mantelzorgers en 10 met personen met jongdementie. Voor de focus groepen includeerden we 21 artsen van verscheidene disciplines die essentieel zijn binnen dementiezorg (neurologen, psychiaters, geriaters en huisartsen). Al deze interviews en focus groepen werden letterlijk getranscribeerd om vervolgens te worden geanalyseerd volgens de 'constant vergelijkende methode', dewelke het mogelijk maakt om verschillen en gelijkenissen, zowel binnen als tussen groepen, te analyseren.

BELANGRIJKSTE BEVINDINGEN

Uit onze interviews met Vlaamse mantelzorgers (Hoofdstuk 1) van personen met jongdementie bleek dat zij -samen met de patiënt- voornamelijk plannen hadden ondernomen op andere dan medische vlakken (bv. financiën). De beperkte mate waarin slechts sprake was van ondernomen VZP, werd verduidelijkt op basis van verscheidene redenen. Zo bleek onder meer dat VZP soms simpelweg niet als nuttig wordt ingeschat, dat de persoon met jongdementie soms gedrag stelt dat VZP bemoeilijkt (bv. een ontkennende houding aannemen), en dat een dag-tot-dag levenswijze soms expliciet verkozen wordt boven zich zorgen maken om de toekomst. Daarenboven verklaarden mantelzorgers hun vermijdende houding om VZP aan te kaarten door hun wens om zichzelf van mogelijk schuldgevoel te ontzien (m.n. vanwege de patiënt emotioneel te belasten door over de toekomst te praten). Ten slotte kwam aan bod dat de vraag heerstte in hoeverre de persoon met jongdementie het gesprek mogelijk anders zou interpreteren dan bedoeld door de mantelzorger. Afgezien van deze redenen, bestond er evenwel een draagvlak voor VZP zoals bleek uit de voorkeuren van mantelzorgers indien ze zouden deelnemen aan het proces. Volgens de mantelzorgers werd VZP best 'tijdig' geïnitieerd, door een derde (velen verkozen een arts) en moest het proces zich toespitsen op de resterende mogelijkheden van de persoon met jongdementie. Ten slotte kwam uit deze interviews ook aan bod dat er een nood heerst aan informatie (over dementie, VZP en beschikbare zorgverlening) en dat er behoefte is aan kwaliteitsvolle, afgestemde zorg voor jongere mensen met dementie en hun familie.

De interviews met Vlaamse zorgverleners werden nadien opnieuw geanalyseerd, tegen het licht van interviews met Amerikaanse mantelzorgers van personen met jongdementie. Uit deze vergelijkende studie (Hoofdstuk 2) kwamen overkoepelend drie verschillen en drie gelijkenissen tussen beide respondentengroepen naar voren. De belangrijkste gelijkenissen waren dat (1) beiden een beperkte notie hadden van het begrip VZP, (2) het vervolledigen van schriftelijke wilsverklaringen slechts gepaard ging met gering overleg omtrent hun inhoud, en (3) dat zowel Belgische als Amerikaanse mantelzorgers de voorkeur te kennen gaven om VZP tijdig te initiëren. De voornaamste verschillen betroffen dat (1) de levenseindebeslissingen die mantelzorgers bespraken doorgaans degene waren die ingebed zijn in het wettelijk kader van hun respectievelijk land; waar Belgische mantelzorgers euthanasie vermeldden, spraken hun Amerikaanse lotgenoten namelijk over het weigeren van behandelingen, dan wel zelfdoding, (2) dat Amerikaanse mantelzorgers een grotere nadruk legden op financiële aangelegenheden, en (3) dat wanneer professionele hulp werd ingeroepen voor het vervolledigen van wilsverklaringen, Amerikaanse mantelzorgers zich tot juristen wendden, daar waar Belgische mantelzorgers een beroep deden op artsen.

Op basis van vijf focus groepen verkenden we de ervaringen met en zienswijzen op VZP van artsen bij dementie ontstaan op zowel jongere, als oudere leeftijd (Hoofdstuk 3). Hieruit bleek dat zij veronderstelden dat het maatschappelijk stigma dat aan dementie kleeft, mede bepaalde hoe personen reageren op hun diagnose. Deze reactie werd, volgens de artsen, soms gekenmerkt door catastrofale toekomstverwachtingen van de patiënt. In deze optiek verduidelijkten de artsen ook hoe patiënten soms aan het werkelijke begin van hun ziektetraject het onderwerp van euthanasie aankaartten. Hoewel artsen aangaven een beeld te hebben van VZP als zijnde een breed communicatieproces, besteedden ze doorheen de focus groepen bijzondere aandacht aan bepaalde levenseindebeslissingen, zoals de zorgcode 'niet reanimeren' of euthanasie. Artsen gaven aan zich verantwoordelijk te voelen om personen van adequate informatie te voorzien en dit zowel op vlak van dementie als aandoening, als omtrent het wettelijke kader voor VZP. Niettegenstaande artsen de potentiële meerwaarde van VZP onderkenden, bleek gelijktijdig dat bepaalde uitdagingen, zoals een vrees om patiënten hoop te ontnemen, de initiatie van het proces bemoeilijkten. De meeste artsen waren van oordeel dat de mate waarin patiënten met dementie en hun mantelzorgers een behoefte ervaren aan VZP eerder voortkomt uit wie ze zijn als personen, dan voortvloeit uit hun leeftijd. Ze haalden evenwel aan dat VZP communicatie bij personen met jongdementie doorgaans meer levensdomeinen behelst, ingevolge de levensfase van jonge patiënten. Ten slotte werd duidelijk dat er een hoge eensgezindheid bestond tussen artsen van verschillende disciplines.

Hoofdstuk 4 betreft een interviewstudie waarin we 10 personen met jongdementie en 10 van hun respectievelijke mantelzorgers vroegen naar hun ervaringen met en hun zienswijzen op VZP in jongdementie. Opnieuw bleek dat er slechts gering bewustzijn bestond onder respondenten omtrent VZP, en bij uitstek als een proces van communicatie. Hoewel ze niet of amper aan VZP gedaan hadden, noemden ze bepaalde voordelen die volgens hen gepaard zouden gaan met VZP. Hierbij werd voornamelijk geopperd dat mantelzorgers mogelijk minder emotionele draaglast ervaren wanneer ze de wensen van de patiënt kennen. Aanvankelijk stelden personen met jongdementie en hun mantelzorgers VZP gelijk aan het maken van medische beslissingen voor het eigenlijke levenseinde. Wanneer ze ingelicht werden over de huidige opvatting van VZP als communicatieproces en VZP als dusdanig bespraken, verbreedden ze hun denkkader naar "wat is belangrijk, nu en in de toekomst?". Binnen dit denkkader besteedden ze ruime aandacht aan niet-medische aspecten van zorg en bespraken ze medische, psychologische en sociale gezondheid als onderling verweven.

Het laatste hoofdstuk van dit doctoraat betreft een secundaire analyse van de interviews uit hoofdstuk 4, toegespitst op uitspraken aangaande euthanasie (Hoofdstuk 5). Niettegenstaande de onderzoeker geen vragen stelde omtrent euthanasie, werd dit onderwerp spontaan aangehaald door de meerderheid van respondenten. Personen met jongdementie en mantelzorgers die het onderwerp eerder (onderling) bespraken, gaven een gelijkaardige context weer waarin deze communicatie had plaatsgevonden. Meer bepaald kwam euthanasie aan bod tijdens bepaalde 'sleutelmomenten', doorgaans met mantelzorgers en kwam dit voort uit overwegingen van patiënten omtrent de gevolgen van hun ziekte(progressie) zowel voor zichzelf als hun naasten. Bovendien bleek dat het negatieve beeld dat heerst rond dementie, alsook rond dementiezorg, personen hun verwachtingen en gedachten omtrent de toekomst beïnvloedde.

ALGEMENE DISCUSSIE

VZP werd door de deelnemers aan ons onderzoek nog vaak opgevat als plannen voor het eigenlijke levenseinde, met nadruk op beslissingen als euthanasie. Dergelijke zienswijze stemt evenwel niet overeen met het denkkader dat patiënten en mantelzorgers naar voor schoven eens ze ingelicht werden over VZP als communicatieproces, m.n. ‘wat is er belangrijk, nu en in de toekomst?’ waarin grote aandacht uitging naar niet-medische domeinen. Teneinde tegemoet te komen aan de zienswijze van patiënten en mantelzorgers, zou VZP als breder communicatieproces (zoals reeds omarmd in wetenschappelijke studies) verder moeten doorsijpelen in de klinische praktijk. De onwetendheid van patiënten en naasten omtrent de term VZP en wat deze net inhoudt, blijkt slechts één aspect van een ruimere informatienood. Evenzeer omtrent dementie, het bijbehorende ziekteverloop en de mogelijke zorgvoorzieningen werd volgens de respondenten onvoldoende informatie verschaft. Een degelijke informatievoorziening omtrent VZP, dementie en het zorglandschap leek een onontbeerlijke voorwaarde om personen in staat te stellen om te plannen voor de toekomst. Ondanks de bevinding dat personen in onze studie niet of amper aan VZP hadden gedaan, bleek er wel een draagvlak voor te bestaan, en dit bij alle respondentengroepen. We concludeerden dan ook dat de randvoorwaarden om aan VZP te kunnen doen, op heden onvoldoende vervuld lijken in geval van jongdementie.

Een tweede thema was de belangrijke rol die uitgaat naar familie bij VZP in jongdementie. Meer bepaald kwam aan het licht dat het willen beschermen van geliefden een overweging was die mensen maakten binnen hun intentie om wel of net niet aan VZP te doen. Mantelzorgers, alsook artsen gaven aan dat ze gesprekken omtrent de toekomst soms vermeden om de patiënt van emotionele draaglast te ontzien. Anderzijds gaven zowel patiënten, artsen, alsook mantelzorgers zelf aan dat VZP mogelijk leidt tot enige gemoedsrust voor mantelzorgers daar waar ze op de hoogte zijn van de wensen van de patiënt. Op basis van deze bevindingen zou VZP bijgevolg beschouwd kunnen worden als een zorgdaad van de persoon met jongdementie ten aanzien van zijn of haar geliefden. Verder werd, door alle respondentengroepen, gewezen op hoe een diagnose van jongdementie impact had op een voltallig gezin en op een veelheid aan levensdomeinen. Zorg afgestemd op een familie, eerder dan op louter de persoon met jongdementie, bleek dan ook aangewezen.

Vervolgens werd gesuggereerd dat een opvatting van VZP waarbij het proces primeert over het product (bijvoorbeeld een wilsverklaring), VZP mogelijk faciliteert. Dergelijke zienswijze en implementatie komt mogelijk tegemoet aan verscheidene struikelblokken die op heden bestaan om deel te nemen aan VZP. Zo zou, onder meer, een stapsgewijze informatievoorziening omtrent VZP en dementie het mogelijk vergemakkelijken voor patiënten en mantelzorgers om na te denken over de toekomst, ondanks hun keuze om 'van dag tot dag' te leven. Daarenboven zou een voortschrijdende en dynamische dialoog omtrent VZP mogelijk tegemoet komen aan de moeilijkheden van veranderende wensen en ervaringen van de patiënt doorheen zijn of haar ziektetraject. Zowel zorgvoorkeuren voor het heden, als voor de toekomst zouden bij voorkeur een uitgangspunt zijn binnen VZP bij jongdementie. Hierbij bleek het bovendien wenselijk om patiënten en mantelzorgers voor te bereiden op het nemen van 'acute' beslissingen, daar waar onzekerheid werd voorgesteld als een onvermijdelijk kenmerk van dementie als aandoening. Niettegenstaande enerzijds personen met jongdementie en hun naasten een voorkeur uitten om zich te focussen op het heden, en anderzijds artsen aangaven dat het geven van prognostische informatie uitdagend is (bv door morele overwegingen), bleek evenwel dat voornamelijk mantelzorgers nood ervaren aan eerlijke en open communicatie omtrent toekomstverwachtingen. VZP bij jongdementie vereist daarom een evenwichtsoefening tussen het behouden/ bieden van hoop enerzijds, en het hebben/ weergeven van een realistisch toekomstbeeld anderzijds.

Bovendien bleek de maatschappelijke, vaak stereotyperende weergave van dementie mogelijk impact te hebben op VZP. Onze studies belichtten dat het stigma rond (jong)dementie mogelijk een invloed uitoefende op hoe patiënten naar hun toekomst keken. De negatieve manier waarop dementie vaak wordt voorgesteld in de media, werd door artsen beschouwd als een voedingsbodem voor catastrofale reacties op de diagnose. Anderzijds gaven patiënten en mantelzorgers binnen ons onderzoek ook een genuanceerdere weergave van leven met jongdementie, wanneer ze aangaven dat er ondanks de ziekte nog ruimte heerste voor genot. Daar waar personen met jongdementie in interviews aangaven zich vaak onderschat te voelen door anderen, kan VZP mogelijk beschouwd worden als een middel om stereotyperende beelden over jongdementie tegen te gaan: VZP zou personen met jongdementie immers het gevoel kunnen geven dat hun mogelijkheden aangesproken en ingezet worden. Een andere bevinding met betrekking tot de maatschappelijke context was dat de kwaliteit van zorg voor jongdementie door patiënten en naasten als laag werd ingeschat en dat dit hun drijfveer voor VZP-deelname mogelijk verminderde.

Verder toonden onze studies aan dat het wettelijk kader voor VZP mede het kader schetste waarin personen over levenseinde(beslissingen) nadachten. Daar waar vele Belgische respondenten spontaan over euthanasie spraken doorheen interviews, deden geen van de Amerikaanse deelnemers dit. Wanneer binnen een land een bepaalde levenseindebeslissing gelegaliseerd was, leek deze natuurlijkerwijs te worden opgenomen in overwegingen omtrent het levenseinde. We opperden bijgevolg dat de verschillen in wetgevende kaders tussen landen mogelijk de communicatie(strategieën) binnen VZP beïnvloeden. Naast de aangewezen holistische opvatting van VZP (zijnde met medische, sociale en psychologische aspecten), lijkt het gelijktijdig aangeraden dat artsen aandacht besteden aan deze verschillen en ingaan op specifieke zorgen of vragen die patiënten mogelijk uiten (zoals euthanasie). Het debat dat op heden in België heerst, met name over euthanasie middels een voorafgaande wilsverklaring bij personen die niet langer wilsbekwaam zijn, vond ook weerklank binnen de interviews, met soms uiteenlopende meningen. Ons onderzoek toont minimaal aan dat de perspectieven van personen met (jong)dementie en hun naasten eventuele beleidsontwikkelingen omtrent de euthanasiewet mee vorm kunnen geven.

Ten slotte werd ingegaan op de specifieke aspecten van VZP bij jongdementie die we over onze studies heen vonden. Deze bleken vooral betrekking te hebben op de inhoud van VZP, eerder dan op de mate waarin personen wensen deel te nemen aan VZP. Zo zou VZP bij jongdementie een ruimere reeks van onderwerpen bestrijken vanwege de veelheid aan levensdomeinen die mogelijk aangedaan zijn door de diagnose. Zo werd bijvoorbeeld meermaals gewezen op de financiële gevolgen voor een gezin (bv mantelzorgers die hun eigen werkzaamheden dienen te verminderen om zorg te dragen voor hun geliefde) en op het belang van financiële planning. Verder werd ook vaak gewezen op de behoefte dat zorgverlening zich richtte tot een familie, en niet louter tot de patiënt. Ook het belang van sociale en fysieke activiteit voor de patiënt met jongdementie werd meermaals belicht.

Algemeen gesteld is het op basis van het huidige doctoraat niet mogelijk om met zekerheid te besluiten of onze bevindingen voortkomen uit de jongere leeftijd van personen met jongdementie en hun mantelzorgers, dan wel een gevolg zijn van dementie als aandoening (ongeacht leeftijd). Hoewel sommige resultaten leeftijdsspecifiek lijken (bv gebrekkige residentiële zorg), lijken andere eerder dementie-specifiek (bv een dag-tot-dag levenswijze en de impact daarvan op VZP).

AANBEVELINGEN VOOR PRAKTIJK, BELEID EN ONDERZOEK

Voor praktijk:

- VZP als dynamisch en relationeel proces
- VZP als gesprek met een holistische focus
- VZP met aandacht voor de maatschappelijke en wettelijke achtergrond waartegen die plaatsvindt

Voor beleid:

- Een eenduidige boodschap verschaffen omtrent VZP als communicatieproces
- Het herevalueren van zorg voor jongdementie en verbeteren waar nodig
- Het includeren van perspectieven van personen met (jong)dementie en hun mantelzorgers binnen beleidsontwikkelingen

Voor onderzoek:

- Het herhalen en uitbreiden van studies uit dit doctoraat binnen een andere maatschappelijke en wettelijke context
- Verschillende groepen van mensen met jongdementie includeren in onderzoek (bv diegenen zonder mantelzorgers)
- De daadwerkelijke klinische praktijk van VZP bestuderen als aanvulling op personen hun narratieven hieromtrent

ENGLISH SUMMARY

INTRODUCTION

Advance care planning (ACP) is considered as an essential element of high quality and ethically sound long-term care after a diagnosis of dementia. ACP is a process of communication between patients, family caregivers and professional caregivers in which the patient's preferences regarding (future) care are explored, including end-of-life care. The overarching goal of ACP is to increase the chance that individuals will receive the care they desire, even at times when they are no longer able to make or to express their own choices. This aim immediately clarifies why ACP can be meaningful in Alzheimer's dementia and related neurodegenerative disorders. After all, dementia is a progressive condition, with no current options to halt or reverse the disease. As a result of Alzheimer's dementia, individuals will increasingly develop difficulties in cognition, memory and day-to-day functioning. Disease progression in dementia will sooner or later result in patients no longer having cognitive capacity. Especially at the end of life, family caregivers of persons with dementia will face potentially difficult medical choices for their loved-one.

Although there is agreement on the potential added value of ACP in dementia, the rate of ACP uptake is particularly low in this population. Evidence suggests that ACP in dementia is associated with specific challenges. Among others, it was shown that physicians recognize the importance of ACP, yet experience clinical, as well as moral stumbling blocks to initiate the process. For example, physicians express uncertainty about prognosis, how to communicate this, and not wanting to deprive patients of hope or instill fear.

Nonetheless dementia is often associated with old age, up to 9% of people with dementia develop symptoms before the age of 65, signaled by the term young-onset dementia (YOD). It is estimated that nearly four million people worldwide suffer from the condition, once again underscoring the need for adequate care. Dementia at a younger age potentially affects individuals ‘in the prime of their lives’. Not only are they possibly still active on the labor market, but they might also have young children and significant financial obligations. Due to the active phase of life in which people with YOD find themselves, it is suggested that they have other needs regarding palliative care and specifically ACP: they are hypothesized to want more autonomy and greater involvement in decision-making regarding care.

Despite the hypothesis above, the views of persons with YOD regarding ACP remain unexplored in scientific research. Moreover, the views of their caregivers are poorly understood, despite the general recommendation to involve them as early as possible in the process of ACP. Persons with YOD and their caregivers are a longstanding and systematically under-researched population in (end-of-life) care. This study would like to change that.

Within this doctoral dissertation, we wish to gain insight from both caregivers of, as well as individuals with YOD themselves on how ACP can be tailored to their needs and preferences. To this end, we will also explore physicians' views on ACP in both younger and older dementia.

METHOD

Based on our exploratory, as well as sensitive research questions, a qualitative research design was chosen for this entire doctoral study. Four studies are based on individual semi-structured interviews with family caregivers and persons with YOD, a fifth study is based on focus groups with physicians. These 5 studies were divided into two parts, and more specifically based on their respondents. Part 1 involves research with caregivers of persons with YOD, both family caregivers and physicians. Part 2 includes persons with YOD themselves, along with their respective caregivers.

A total of 48 individual interviews were conducted, including 38 with family caregivers and 10 with persons with YOD. For the focus groups, we included 21 physicians from several disciplines essential within dementia care (neurologists, psychiatrists, geriatricians and general practitioners).

All interviews and focus groups were transcribed verbatim for subsequently using the method of ‘constant comparative analysis’, which allows for identifying differences and similarities, both within and between groups.

MAIN FINDINGS

Our interviews with Flemish family caregivers (Chapter 1) of persons with YOD revealed that -together with the patient- they had mainly undertaken plans in areas other than medical care (e.g., finances). The limited extent of undertaken ACP was clarified through several reasons. These included that sometimes ACP is simply not perceived as helpful, that the person with YOD sometimes exhibits behaviors that hinder ACP (e.g., a denying attitude), and that a day-to-day lifestyle is sometimes explicitly preferred over worrying about the future. In addition, family caregivers explained their avoidance of bringing up ACP by their desire to spare themselves from possible guilt (e.g., from emotionally burdening the patient by talking about the future). Finally, the question arose as to the extent to which the person with YOD might interpret the conversation differently than intended by the family caregiver. Apart from these reasons, however, there was support for ACP as shown by the preferences expressed by the family caregivers for how to participate in the process. According to them, ACP was best initiated ‘timely’, by a third party (many preferred a physician) and should focus on the remaining capabilities of the person with YOD. Finally, these interviews also revealed a need for information (about dementia, ACP and available care services) and a need for high-quality, tailored care for younger people with dementia and their families.

The interviews with Flemish caregivers were subsequently reanalyzed against interviews with American informal caregivers of persons with YOD. Overall, three differences and three similarities between the two respondent groups emerged from this comparative study (Chapter 2). The main similarities were that (1) both respondent groups had a limited notion of the concept of ACP, (2) the completion of written advance directives was accompanied by only limited discussion regarding their content, and (3) that both Belgian and American family caregivers expressed a preference for the timely initiation of ACP. The main differences were that (1) the end-of-life decisions that family caregivers discussed were those embedded in the legal framework of their respective countries; namely, where Belgian caregivers mentioned euthanasia, their American peers talked about refusal of treatment, or suicide, (2) that American family caregivers placed greater emphasis on financial matters, and (3) that when professional help was sought to complete advance directives, American caregivers turned to lawyers, whereas Belgian caregivers turned to physicians.

Based on five focus groups, we explored physicians' experiences with and views of ACP in dementia at both younger, and older ages (Chapter 3). This revealed that physicians hypothesized that the social stigma attached to dementia partly determined how individuals responded to their diagnosis. According to them, this reaction was sometimes characterized by a patient’s catastrophic expectations for the future. In this view, physicians also clarified how patients sometimes raised the subject of euthanasia at the very beginning of their disease trajectory. Although physicians agreed with a perception of ACP as being a broad communication process, they paid particular attention to certain end-of-life decisions throughout focus groups, such as ‘do not resuscitate’ orders or euthanasia. Physicians expressed a sense of responsibility to provide individuals with adequate information, both in terms of dementia as a condition and regarding the legal framework for ACP. Notwithstanding physicians recognized the potential value of ACP, it was simultaneously apparent that certain challenges (such as a fear of depriving patients of hope) hindered initiation of the process. Most physicians believed that the extent to which patients with dementia and their caregivers experience a need for ACP stems from who they are as individuals rather than from their age. However, they expressed that ACP communication in persons with YOD tends to encompass more life domains, due to patients’ stage of life. Finally, it became clear that there was high congruency among physicians of different disciplines.

Chapter 4 concerns an interview study in which we asked 10 persons living with YOD and 10 of their respective caregivers about their experiences with and their views on ACP. Again, we found that there was only low awareness among respondents regarding ACP, and especially as a process of communication. Although they had not or barely engaged in ACP, they mentioned certain benefits they felt would be associated with it. It was mainly suggested that family caregivers may experience less emotional burden when they know the patient's wishes. Initially, persons with YOD and their family caregivers equated ACP with making end-of-life medical decisions. When they were informed about the current understanding of ACP as a communication process and discussed ACP as such, they broadened their thought framework to "what is important, now and in the future?". Within this frame of thinking, they paid ample attention to non-medical aspects of care and discussed medical, psychological and social health as intertwined.

The final chapter of this dissertation is a secondary analysis of the interviews from Chapter 4, focusing on statements regarding euthanasia (Chapter 5). Although the researcher did not ask questions about euthanasia, the topic was spontaneously raised by the majority of respondents. Individuals with YOD and family caregivers who had previously discussed the topic (among themselves) reflected a similar context in which this communication had taken place. Specifically, euthanasia came up during certain ‘key moments’, usually with family caregivers and stemmed from patients' considerations regarding the impact of their disease (progression) both on themselves and their loved-ones. Moreover, the negative image surrounding dementia, as well as dementia care, appeared to influence individuals' expectations and thoughts regarding the future.

GENERAL DISCUSSION

ACP was often still perceived by participants in our study as medical planning for the actual end of life. Such a view, however, does not correspond to the thought framework that patients and caregivers put forward once they were informed about ACP as a communication process, i.e. "what is important now and in the future?" and with ample attention to non-medical domains. In order to meet the views of patients and family caregivers, ACP as a broader communication process (as already embraced in academia) should trickle down further into clinical practice. Patients' and caregivers' unawareness of the term ACP and what it entails appeared to be only one part of a more extensive information need. Similarly, according to respondents, insufficient information was provided regarding dementia, the associated course of the disease and possible care services. Adequate information about ACP, dementia and the care landscape seemed an indispensable prerequisite to enable individuals to plan for the future. Despite the finding that individuals in our study had little or no experience with ACP, there appeared to be support for it among all respondents. Thus, we concluded that currently the prerequisites for actually engaging in ACP appear to be insufficiently met in case of YOD.

A second theme was the important role of family in ACP in YOD. Specifically, it emerged that wanting to protect loved-ones was a consideration that people make within their intention to engage or not to engage in ACP. Family caregivers, as well as physicians, indicated that they sometimes avoid conversations regarding the future in order to spare the patient of burdensome emotions. On the other hand, patients, physicians, as well as family caregivers themselves indicated that ACP and knowing the patient's preferences might lead to some peace of mind for family caregivers. Consequently, based on these findings, ACP could be considered an act of care by the person with YOD toward his or her loved-ones. Furthermore, it was pointed out, by all respondent groups, how a diagnosis of YOD impacted an entire family and a multitude of life domains. Care tailored to a family, rather than exclusively to the person with YOD, therefore appeared appropriate.

Further, it was suggested that a view of ACP in which the process is prioritized over the product (e.g., an advance directive) might facilitate ACP. Such a view and implementation potentially address several stumbling blocks that currently exist to participating in ACP. For example, among other things, a step-by-step information provision regarding ACP and dementia would potentially facilitate patients and caregivers to think about the future, despite their choice to live 'from day-to-day'. In addition, an ongoing and dynamic dialogue regarding ACP would potentially address the challenge of patients' changing wishes and experiences throughout their disease trajectory. Both care preferences for the present and for the future would preferably be a starting point within ACP in YOD. In doing so, it also appeared desirable to prepare patients and family caregivers to make 'acute' decisions, since uncertainty was presented as an inevitable feature of dementia. Notwithstanding the fact that persons with YOD and their relatives expressed a preference to focus on the present, and that physicians indicate that giving prognostic information is challenging (e.g. due to moral considerations), it appeared that mainly family caregivers experienced a need for honest and open communication regarding future expectations. ACP in YOD therefore requires a balancing act between maintaining/providing hope on the one hand and having/representing a realistic image of the future on the other.

Moreover, the societal, often stereotypical portrayal of dementia was found to potentially impact ACP. Our studies highlighted that the stigma surrounding (young-onset) dementia possibly impacted how patients viewed their future. The negative way in which dementia is often presented in the media was also seen by clinicians as fueling catastrophic reactions to the diagnosis. On the other hand, patients and family caregivers within our study also portrayed a more nuanced image of living with YOD when they indicated that, despite the condition, there was still room for enjoyment. As persons with YOD indicated in interviews that they often felt underestimated by others, ACP could possibly be considered as a way to counter stereotypical images about dementia: ACP could make individuals with YOD feel that their capabilities are being addressed and utilized. It also appeared that the low quality of care for YOD, as perceived by patients and their loved-ones, possibly reduced the incentive for ACP.

Additionally, our studies showed that the legal framework for ACP partly shaped the framework in which individuals thought about end-of-life decisions. Whereas many Belgian respondents spontaneously discussed euthanasia throughout interviews, none of the American participants did so. When a particular end-of-life decision was legalized within a country, it naturally seemed to be incorporated in end-of-life considerations. Consequently, we suggested that differences in legislative frameworks between countries may influence communication (strategies) within ACP. Besides the recommended holistic view of ACP (namely including medical, social and psychological aspects), it seems simultaneously recommended that physicians pay attention to these differences and address specific concerns or questions that patients may express (such as euthanasia). The debate that is currently going on in Belgium, particularly regarding the legality of an advance euthanasia directive for persons who have lost cognitive capacity, also resonated within the interviews, with sometimes divergent opinions. At a minimum, our research shows that the perspectives of persons with YOD and their loved ones can help shape possible policy developments regarding the euthanasia law.

Finally, we addressed the specific aspects of ACP in YOD that were found across our studies. These appeared to relate primarily to the content of ACP, rather than the extent to which individuals wish to participate in ACP: namely, ACP in YOD would cover a wide range of topics given the plurality of life domains potentially affected by the diagnosis. For example, the financial implications for a family (e.g., informal caregivers who need to reduce their own work to care for their loved-one) and the importance of financial planning were mentioned several times. Furthermore, the need for care provision to be directed to a family, and not merely to the patient, was also frequently mentioned. Lastly, the importance of social and physical activity for the patient with YOD was also highlighted.

In general, it is not possible to determine with certainty whether our findings stem from the younger age of persons with YOD and their caregivers or whether they are a consequence of dementia as a condition (regardless of age). While some results seem age-specific (e.g., regarding inadequate residential care), others seem rather dementia-specific (e.g., day-to-day lifestyle and its impact on ACP).

RECOMMENDATIONS FOR PRACTICE, POLICY AND RESEARCH

For practice:

- ACP as a dynamic and relational process
- ACP as a conversation with a holistic focus
- ACP with attention to the societal and legal background against which it takes place

For policy:

- Provide a uniform message regarding ACP as a communication process
- Re-evaluating care for YOD and improving as needed
- Including people with (young-onset) dementia’s and their family caregivers’ perspectives within policy developments

For research:

- Repeating and expanding studies of this dissertation within a different societal and legal context
- Including different groups of people with YOD in research (e.g., those without informal caregivers)
- Studying the actual clinical practice of ACP to complement individuals' narratives about how this practice is experienced

PART VI

CURRICULUM VITAE

CURRICULUM VITAE

Romy Van Rickstal was born September 28th 1992 in Antwerp, Belgium. At the Vrije Universiteit Brussel (VUB), she obtained her bachelor's degree in Psychology and her master's degree in Clinical Psychology. In her second master year, she was selected for the VUB Honours Program. Shortly after her studies, she joined the End-of-Life Care Research Group as a doctoral researcher. After one year, Romy was granted a PhD Fellowship from the Research Foundation Flanders (FWO), supervised by Prof. Dr. Lieve Van den Block and Prof. Dr. Aline De Vleminck, and -further in the project- also by Prof. Dr. Sebastiaan Engelborghs. Romy's research focusses on advance care planning in young-onset dementia and has been disseminated through publications in high-impact peer-reviewed journals and (inter)national conferences.

EDUCATION

Master of Science in Clinical Psychology, Vrije Universiteit Brussel (July 2017)

Summa cum laude (ranking: 1st/171)

Master dissertation: Advance care planning for people with young-onset dementia: an exploratory interview study with primary family caregivers

Bachelor in Psychology, Vrije Universiteit Brussel (July 2015)

Magna cum laude (ranking: 1st/141)

PROFESSIONAL CAREER

2019 - 2023

Fellow at the Research Foundation – Flanders (FWO)

Pre-doctoral mandate for Strategic Basic Research, from January 1st 2019 onwards

Doctoral Researcher at the End-of-Life Care Research Group

2017 - 2018

Appointed by Vrije Universiteit Brussel (VUB)

Doctoral Researcher at the End-of-Life Care Research Group

ACADEMIC PUBLICATIONS

Van Rickstal, R., De Vleminck, A., Engelborghs, S., & Van den Block, L. (2023). Experiences with and perspectives on advance care planning in young- and late- onset dementia: A focus group study with physicians from various disciplines. *Frontiers in aging neuroscience*, *15*, 1130642. https://doi.org/10.3389/fnagi.2023.1130642 [2021 IF 5.702; Ranking Q1; ranking 16/109 neurosciences in SCIE edition]

Van Rickstal, R., De Vleminck, A., Chambaere, K., & Van den Block, L. (2022). People with young-onset dementia and their caregivers discussing euthanasia: a qualitative analysis of their consideration. Submitted to Patient Education and Counseling (December 23rd, 2022).

Van Rickstal, R., De Vleminck, A., Engelborghs, S., Versijpt, J., & Van den Block, L. (2022). A qualitative study with people with young-onset dementia and their family caregivers on advance care planning: A holistic, flexible, and relational approach is recommended. *Palliative medicine*, *36*(6), 964–975. doi: 10.1177/02692163221090385. [2020 IF 4.762; Ranking Q1; ranking 18/108 in health care sciences & services in SCIE edition]

Van Rickstal, R., De Vleminck, A., Morrison, S. R., Koopmans, R. T., van der Steen, J. T., Engelborghs, S., Neugroschl, J., Aldridge, M. D., Sano, M., & Van den Block, L. (2020). Comparing Advance Care Planning in Young-Onset Dementia in the USA vs Belgium: Challenges Partly Related to Societal Context. *Journal of the American Medical Directors Association*, *21*(6), 851–857. doi: 10.1016/j.jamda.2020.01.007. [2018 SCI impact factor 4.899; Ranking Q1; ranking n° 6 of 53 in geriatrics & gerontology]

Van Rickstal, R., De Vleminck, A., Aldridge, M. D., Morrison, S. R., Koopmans, R. T., van der Steen, J. T., Engelborghs, S., & Van den Block, L. (2019). Limited engagement in, yet clear preferences for advance care planning in young-onset dementia: An exploratory interview-study with family caregivers. *Palliative medicine*, *33*(9), 1166–1175. doi:10.1177/0269216319864777. [2018 SCI impact factor 4.956; Ranking D1; ranking n° 6 of 98 in health care sciences & services]

ACADEMIC BOOK CHAPTER

Koopmans, R., van der Steen, J., Perry, M., & **Van Rickstal, R.** (2021). Palliative care in people with young onset dementia. In de Vugt, M., & Carter, J. (Ed.), Understanding young unset dementia: evaluation, needs and care. Routledge. ISBN 9780367568160

ACADEMIC PRESENTATIONS

2022

Oral presentation (teaching activity) at Radboud Universiteit Nijmegen (UMC Radboud, NL)
Course: ‘Interprofessional Education for physicians and professionals: working with young people with dementia’

2021

Oral presentation for the Advisory Board of the Dutch National Expertise Centre of dementia at a young age

Oral presentation at the International Conference of Alzheimer Europe (online event)
Presentation Title: “A qualitative study with people with young-onset dementia and their family caregivers on advance care planning”

Oral presentation at the Alzheimer Disease International Conference (online event)
Presentation Title: “Exploring similarities and differences in ACP in YOD: a qualitative study with American and Belgian family Caregivers”

Poster Presentation at the 11th World Research Congress of the EAPC (European Association for Palliative Care)
Poster Title: “Comparing advance care planning in young-onset dementia in the USA vs Belgium: challenges relate to societal context”

2019
Poster presentation for the Flemish – Dutch Science Days on end-of-life care
Poster Title: “Experiences with and preferences for advance care planning in young-onset dementia”

Oral presentation at the International Conference of Alzheimer Europe, The Hague (NL)
Presentation Title: “Comparing advance care planning in Young-Onset Dementia: US vs. Belgium”

Oral presentation (teaching activity) at Radboud Universiteit Nijmegen (UMC Radboud, NL)
Course: ‘Interprofessional Education for physicians and professionals: working with young people with dementia’

2018
Oral Presentation (plenary), Nursing Home Research International Working Group Conference; 6th edition, Rome (Italy)
Presentation title: “Experiences with and preferences for advance care planning in Young-Onset Dementia”

Oral presentation for the Flemish non-profit organization Right to a Dignified Death, regarding master dissertation, “Advance care planning in young-onset dementia: an interview study with family caregivers”

GRANTS OR AWARDS RECEIVED

Personal **PhD Fellowship ‘Strategic Basic Research’**, granted by the Research Foundation Flanders (FWO)
- Two terms of two years:
  - 2019-2020 (initial mandate),
  - 2021-2022 (renewal after successful evaluation of first two-year term)
- Granted an extension based on application for Covid-19 delay (Jan-Feb-March ‘23)

Prize Léon Favyts – Hugo Van den Enden, 2018
Awarded by Recht op Waardig Sterven (non-profit Right to Die organization), on November 2nd 2018 for ‘a Master dissertation with societal relevance regarding the subject of end of life and end-of-life decisions, contributing to patient autonomy’