Young onset dementia

Care needs & Service provision

Christian Bakker

Colofon
The research presented in this thesis was performed by a researcher of the care organization 'Florence' in the Hague, the Netherlands, in collaboration with the department of Primary and Community Care, Centre for Family Medicine, Geriatric Care and Public Health, Radboud University Nijmegen, Medical Centre, the Netherlands, The Department of Psychiatry and Neuropsychology, School for Mental Health and Neuroscience, Maastricht University, the Netherlands and VU University Amsterdam, the Netherlands. This study was funded by the Florence Care Group in the Hague, the Dutch Alzheimer's Society and Stichting Wetenschapsbevordering Verpleeghuizen (SWBV).
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Care Needs & Service Provision

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Promotoren:	Prof. dr. R.T.C.M. Koopmans
	Prof. dr. F.R.J. Verhey (Universiteit Maastricht)
Copromotor:	Mw. dr. M.E. de Vugt (Universiteit Maastricht)

Manuscriptcommissie:	Prof. dr. W.J.H.M. van den Bosch
	Prof. dr. M.G.M. Olde Rikkert
	Prof. dr. R.M. Droës (VU Medisch Centrum)

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Introduction

Introduction

In our ageing society dementia is a growing public health concern profoundly affecting the health related quality of life (HRQoL) of those affected by the disease and their families and at the same time increasing demands on health care services and care expenditures.[2] Because dementia is typically regarded as a disease of old age, the importance and implications of the occurrence of dementia at a younger age may be overlooked. Young onset dementia (YOD) is defined as dementia with the onset of disease symptoms before the age of 65. This conceptualisation is somewhat arbitrary and probably more closely related to the fact that in most developed countries the age of 65 is considered to mark the beginning of a new life phase rather than that it refers to patient characteristics such as vitality, life expectancy or social engagement. Currently, there is also some debate about the terminology. The term 'presenile dementia' used until about 10 years ago, is no longer considered appropriate.[3] In current research the term 'early onset dementia' is widely used. However, some state that the term 'early onset dementia' refers to the early stages of the dementia process or should be reserved for people with a disease onset before the age of 45 years. Others propose that from a person-centred point of view, people affected by the dementia at an early age consider themselves to be 'young' suggesting that the term 'young onset dementia' (YOD), 'younger onset dementia' or 'younger people with dementia' should be preferred. Because of the person centred focus of our research, the term that these younger persons and their families prefer themselves, 'young onset dementia' will be used throughout this thesis.

Alzheimer's disease

It is difficult to understand for the people that surround us.
They observe the changes, and you, you feel more and more alone.

Not knowing or understanding, feels like not being there at all.
Living in uncertainty, a life that causes so much pain.

Not being able to communicate with those I love angers me so much. Misunderstood, not
knowing, all the arguing that follows. It hurts to realize the future, gradually losing who I am.

I must confess: the fear of not knowing my husband, children and those I love torments me.
Losing one another, unreachable. This disease soon will become unbearable.

Losing yourself, trapped in the past. Losing control over yourself and your decisions. One of
many symptoms of Alzheimer's disease, that's a fact.

A reality to all of us.

Gerda Arkesteijn[1]

Epidemiology

Studies on the epidemiology of YOD are scarce. Harvey and colleagues[4] estimated that the prevalence of YOD, in two London boroughs in the UK, was 54.0 per 100,000 in the 30-64 age group and 98.1 per 100,000 to 118.0 per 100,000 in the 45-64 year age group. A Japanese study estimated a similar prevalence of 42.3 per 100,000 in the 18-65 age group.[5] While a recent study from Australia estimated a prevalence of 68.2 per 100 000 [95% CI 53.9-82.4] in the 30-64 age group, with approximately 1/750 of the population at risk of those aged 45-64 and 1/1500 of those aged 30-64 (Withall et al, 2012). An important issue regarding epidemiological studies on YOD concerns a likely underestimation of population prevalence of YOD, because cases were identified through service contact only. The recent WHO report estimates that the proportion of YOD of all people with dementia may be as high as 6-9%.[2]

In the UK study, Alzheimer's disease (AD) (34%) was the most common type of dementia, followed by vascular dementia (18%) and frontotemporal dementia (FTD) (12%).[4] They also found a relative high proportion (19%) of other less common causes of dementia suggesting that the aetiology of dementia in younger patients is much more heterogenic than in late onset dementia (LOD). In contrast, Ikejima and colleagues[5] found that in Japan vascular disease was the most common cause of dementia, followed by AD. While Withall and colleagues (Withall et al. 2012) found that Alcohol related dementia was the most common type (21%) followed by Alzheimer's disease (17%), unspecified dementia (13%), dementia secondary to other medical illness (including Huntington disease, multiple sclerosis, acquired immune deficiency syndrome (AIDS), epilepsy and Creutzfeldt-Jakob disease; 17%); and frontotemporal dementia (including Pick's disease, semantic dementia and progressive non-fluent aphasia; 12%).

There have been series of convenience samples which all lend support to the view that Alzheimer dementia, dementia with Lewy bodies, and vascular cognitive impairment compromise a smaller portion of cases in younger patients than in the older populations, with a relatively higher prevalence of frontotemporal dementia (FTD) and alcohol related dementia in patients with YOD.[3] However, the younger the onset of the dementia, the more likely it is that the patient has a genetic or metabolic disease. Kelley et al. published a retrospective medical chart observational study of patients with cognitive decline between the ages of 17 to 45 years of the Mayo Clinic Rochester over a 10-year inclusion period.[6] Of the 235 cases 31.1% presented with neurodegenerative aetiologies' while Alzheimer disease was uncommon. In their sample autoimmune or inflammatory causes accounted for 21.3% while at last follow-up, for 18.7% of patients no cause for the dementia could be established, despite exhaustive evaluation.

Psychosocial implications

A diagnosis of dementia has serious consequences for those who are affected by the disease and results in an important loss of health related quality of life (HRQoL). Within the concept of HRQoL, the individual response to the physical, mental and social effects of the disease on daily living influence the extent to which personal satisfaction with life circumstances can be achieved.[7] When the onset of the disease is in a mid-life age, people may face specific consequences and problems, further compromising HRQoL.

A mayor concern for YOD patients and their families is related to difficulties in obtaining a dementia diagnosis. This is probably related to a lack of awareness of the occurrence of dementia in younger people both among the general public as among general practitioners to who people usually first present. Symptoms are often misunderstood for other more common causes in younger people, such as depression, anxiety and other illnesses. Also the fact that some people present with predominant behavioural changes often contributes to a complex and lengthy nature of the diagnostic process in YOD.[3, 8, 9] Consequently, there is a significant delay in the time to diagnosis prolonging the period of uncertainty and delaying the initiation of appropriate support and health care services.[10]

YOD patients are much more likely to suffer from the loss of different roles within the family like for instance being a parent, financial provider or spouse, resulting in the loss of sense of self and identity.[11] Because of the occurrence of cognitive, functional and behavioural changes in a more demanding environment, YOD patients often have to renounce their work in an early stage of the disease resulting in financial and legal issues[12] and a loss of self-esteem. With the loss of a job, YOD patients may also lose an important part of their social contacts as well as the ability to give meaning to their life. The more profound loss of these roles in younger dementia patients is likely to add to the loss of HRQoL.
Caregivers of YOD patients are known to experience high levels of caregiver burden, even more so than caregivers of older dementia patients.[13] This has been suggested to be related to the considerable number of psychosocial problems these younger caregivers encounter, including relational difficulties, family conflict, employment and financial issues, and the aforementioned negative experiences regarding the diagnostic process.[14] Furthermore, neuropsychiatric symptoms in YOD are highly prevalent because of the high prevalence of frontotemporal dementia, in which neuropsychiatric symptoms are a key symptom. Because younger people are physically fit and stronger, the impact of neuropsychiatric symptoms might also be more profound. This will add to the burden felt by these younger caregivers and likely further complicate the caregiving process, with serious implications for caregiver HRQoL.

Care needs and service use in YOD

The psychosocial implications of YOD might also result in different care needs in these younger patients and their caregivers. However, research on care needs and service use in YOD is scarce and the literature that exists is largely of an experiential nature. Although the experiences of patients and health care professionals are an important resource for clinical practice and evaluation, also scientific evidence concerning the factors underlying the use or non-use of health services in YOD during the caregiving process is vital for further improving the quality of care for these patients and their families.

Most dementia patients are cared for at home for a considerable proportion of the caregiving trajectory, with the time until institutionalization ranging from 2.5 to 3.5 years in late onset dementia (LOD).[15] It is unclear whether this is also true for YOD. Persons with dementia and their families want to prevent institutionalization as long as possible due to concerns about the quality of care in nursing homes and the potential negative impact of institutionalization on patients' HRQoL. Additionally, institutional care is costly and far exceeds the cost of informal care in high income countries.[16] Therefore, in Western Europe, policy is aimed at delaying institutionalization in persons with dementia through the provision of (community) support programs for patients and caregivers.[17] The desire to prevent or postpone institutionalization may particularly apply for younger people with dementia and their families because they perceive main stream dementia services as inappropriate for their loved ones.[18] This stresses the importance of a better understanding of their needs and which factors underlie the initiation and continued use of services and support in this specific patient group.

The behavioural model,[19] which was originally introduced in 1968 and since then has been revised until the late 90s, can be used to highlight different aspects that might be important in the caregiving trajectory in YOD. The behavioural model suggests that people's use of health care services, such as psychosocial support, home care, day care and residential services is a function of their predisposition to use services, factors that enable or impede use, and their need for care. Predisposing factors constitute demographic characteristics (i.e. age, gender), genetic factors, psychological characteristics (i.e. mental dysfunction, cognitive impairment and autonomy), social structures (i.e. social networks, social interactions and culture) and health beliefs. The enabling variables include personal/familial/social and community enabling resources as well as organizational factors, e.g. how health care services are organized and whether or not health insurance benefits are available. People must have the means and knowledge to get to appropriate services to be able to make use of them. Also health care professionals and facilities must be available for individuals.

The latter being an important issue in YOD because in most countries YOD specialized services are scarce and patients and their families therefore have to resort to regular dementia services that are often designed for the elderly. Because, for instance, younger patients are more likely to be physically fit and sexually active, have different interests, and are likely to identify more closely with staff than LOD patients, regular dementia services might have difficulties addressing the needs of these younger patients.[20] Several researchers have suggested that in YOD comprehensive dedicated multidisciplinary services should be commissioned in order to be able to meet the specific needs of these younger patients and their caregivers during the caregiving trajectory.[18, 20, 21] Implicating that this might prevent YOD patients and their caregivers from being pushed 'from pillar to post' in their search for services that can meet their needs.[21]

In the behavioural model the need component is specified as the most immediate cause of health service use and therefore of special interest in YOD. The need component consists of both perceived and evaluated health status. Perceived need refers to how people view their own general health and functional state, as well as how they experience symptoms of illness, pain and worries about their health. This perception of their own HRQoL will determine whether or not they perceive their problems to be pressing enough to seek care and support. Complimentary in the model, evaluated need is related to the professional judgement about people's health status and their need for care by health care professionals, such as the general practitioner. It is likely that in YOD the delay in the time to diagnosis, experiences with existing healthcare services as well how patients and caregivers perceive the illness and are able to address the issues at hand influence the initiation and use of health care services. However, little is known about the course of care needs in YOD, the experiences of YOD patients and their caregivers with existing health care services and whether or not the same factors influence the decision to use health care services as in LOD.

Aims and outline of this thesis

The objective of this thesis is to study different aspects of the caregiving process in young onset dementia. To our knowledge, the NeedYD-study is the first prospective study investigating the course of young onset dementia and exploring care needs of both patients and caregivers. The aims, design and methods of the NeedYD-study on which the studies presented in this thesis were based are described in chapter 2. This thesis focuses on three main research questions:

(1) *What are the care needs of young onset dementia patients?*

As stated earlier in this introduction needs can be considered one of the major components explaining the use and non-use of health care services, such as psychosocial support, home care, day care and residential care. No research regarding the care needs of young onset dementia patients and their caregivers is available, while this information is essential for the management of YOD. Therefore, in chapter 3 we explored the experiences of a spouse of a young person with dementia, and studied how the needs of both the patient and his wife developed and changed over time. Furthermore, we explored experiences and views concerning provided health care services. In chapter 4 a detailed overview of the care needs of YOD patients is provided, examining the personal views of YOD patients and their caregivers on care needs and the extent to which these needs are met.

(2) *What are the consequences of unmet care needs on the course of neuropsychiatric symptoms and health related quality of life in YOD?*

Clinical experience as well as the limited research that is available on the issue of care needs in YOD suggests that the needs of YOD patients and their caregivers differ from older patients and their caregivers. It is suggested that mainstream dementia services as a result will have difficulties in addressing the care needs of these younger patients and their caregivers, leaving care needs unmet.[8] We expect that in YOD patients an increase in unmet needs will be associated with an increase in neuropsychiatric symptoms, considering the association that has been found previously between unmet needs and several neuropsychiatric symptoms, such as anxiety and depression [22], agitation [23, 24] and resistiveness to care.[25] In chapter 4 the relationship between changes in care needs during a two-year period with changes in the severity of neuropsychiatric symptoms is explored.

We also hypothesize that when needs cannot be met, this will have implications for patient and caregiver functioning and wellbeing, negatively influencing their HRQoL. Therefore, in chapter 5 the relationship between unmet needs and health related quality of life of both patients and caregivers is investigated.

(3) *What factors are related to the use of (in)formal care and institutionalization in community dwelling young onset dementia patients?*

Little is known about the caregiving trajectory in YOD, the provision of informal and formal care and which factors are related to the initiation of care. Health care services are much more likely to be beneficial to YOD patients and their caregivers when they can be more properly attuned to the care family members can provide themselves. Gaining insight into the beliefs and experiences about health care services as well as factors determining the use of these services and institutionalization might accommodate health care professionals in designing services that can meet the needs and personal preferences of these younger patients and their caregivers.

In chapter 6 the amount of formal and informal care use in YOD patients and their primary caregivers is explored in the period prior to institutionalization. Also factors associated with the use of formal and informal care in YOD are investigated.

In chapter 7 the time from symptom onset to institutionalization in YOD as well as factors predicting the time to institutionalization in YOD are investigated. These findings are compared with a convenience sample of LOD patients.

Finally, in chapter 8 a summary of the main findings and conclusions is given and these are discussed within the context of clinical practice. Also methodological limitations of the research as well as the implications for people with YOD, their caregivers as well as health care professionals and the relevance of this study for further research are discussed.

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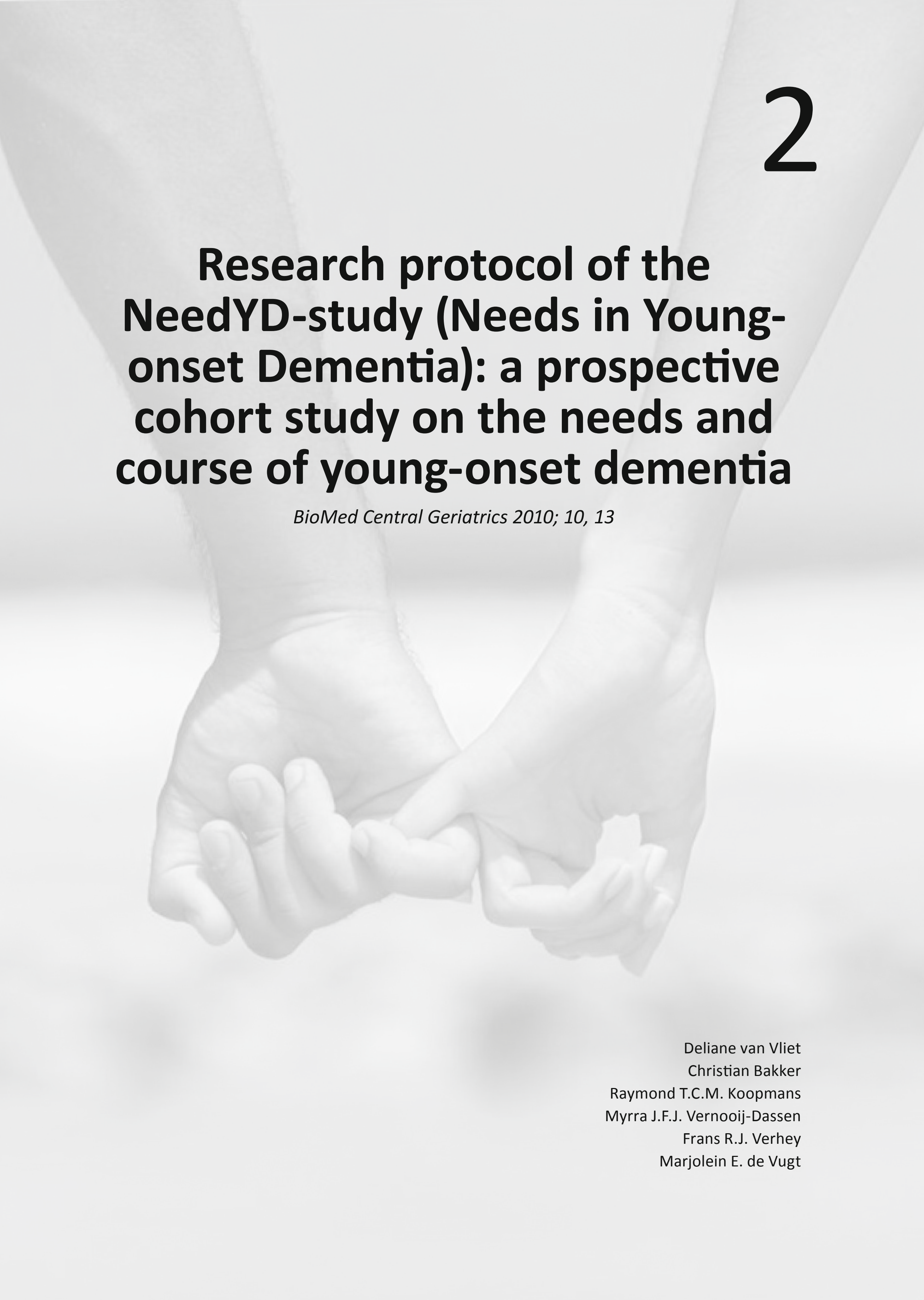

2

Research protocol of the NeedYD-study (Needs in Young-onset Dementia): a prospective cohort study on the needs and course of young-onset dementia

BioMed Central Geriatrics 2010; 10, 13

Deliane van Vliet
Christian Bakker
Raymond T.C.M. Koopmans
Myrra J.F.J. Vernooij-Dassen
Frans R.J. Verhey
Marjolein E. de Vugt

Abstract

Background: Young-onset dementia has serious consequences for patients and their family members. Although there has been growing attention for this patient group, health care services are still mainly targeted at the elderly. Specific knowledge of the needs of young-onset dementia patients and their families is limited but necessary for the development of adequate health care services and specific guidelines. This research project is mainly targeted at delineating the course of young-onset dementia, the functional characteristics and needs of young-onset dementia patients and their caregivers, the risk factors for institutionalization and the interaction with the caring environment.

Methods/Design: The NeedYD-study (Needs in Young-onset Dementia) is a longitudinal observational study investigating young-onset dementia patients and their caregivers (n=215). Assessments were performed every six months over two years and consisted of interviews and questionnaires with patients and caregivers. The main outcomes were (1) the needs of patients and caregivers, as measured by the Camberwell Assessment of Needs for the Elderly (CANE) and (2) neuropsychiatric symptoms, as measured by the NeuroPsychiatric Inventory (NPI). Qualitative analyses were performed in order to obtain more in-depth information on the experiences of YOD patients and their family members. The results of this study were compared with comparable data on late onset dementia from a historical cohort.

Discussion: The study protocol of the NeedYD-study is presented here. To our knowledge, this study is the first prospective cohort study in this research area. Although some limitations exist, these do not outweigh the strengths of this study design.

Background

Dementia is often regarded as a disease of old age. However, there is also a group in which the symptoms of the disease develop before the age of 65. Prevalence rates of young-onset dementia (YOD) have been reported to range between 54 and 260 cases per 100,000 in the 30-64 age group.[1-3]

YOD is recognized as an important psychosocial and medical health problem with serious consequences for patients and their families.[4, 5] YOD is more difficult to recognize than late onset dementia (LOD) in the early stages of the disease because of the lower prevalence rate, the wider range of etiologies [6, 7] and the use of other mental health services (e.g., community mental health teams). These factors cause an important delay before an accurate diagnosis can be established, commonly resulting in feelings of insecurity and frustration for both patients and their families.[8] A proper diagnosis is an important prerequisite for receiving adequate (in)formal support and health care services.

YOD also may have a different clinical manifestation than LOD due to the relatively high prevalence rate of frontotemporal dementia (FTD), in which problem behavior is more prevalent as the presenting sign of probable dementia.[6, 7] Recent research on the impact of problem behavior on caregivers and vice versa shows that these specific aspects of the dementia, more so than cognitive and functional changes, have severe consequences for patients and their family members.[9] Behavioral problems are the most important risk factor for caregiver burden and a strong predictor of institutionalization [10-12] but also an important starting point for interventions.[13] Recent studies have shown that psychological factors, such as disease awareness,[14] and environmental factors, such as caregiver management strategies,[15] influence the development and persistence of behavioral problems in LOD. Similar studies on YOD have not yet been conducted.

Furthermore, YOD patients are in a life phase in which they often have an active role in society and often have young children. The loss of roles and responsibilities is, therefore, greater than in older people. They also have to deal with specific issues such as marital problems, family conflict, (un)employment and financial issues.[5] Furthermore, many YOD patients of the post-war baby boom generation grew up in a society that is very different from that of the older generation. The needs of YOD patients may, therefore, be different and demand a different approach than in LOD.

Despite these differences, the availability of specialized healthcare services is still limited in most countries, forcing YOD patients and their family members to use services designed for the elderly. In the Netherlands, specialized services are available, but their geographic distribution is limited, as is the range of services offered. Furthermore, specific knowledge on the characteristics and needs of YOD patients and their families is lacking but prerequisite for the development of suitable health care services. Adequate diagnostics, (in)formal support and services like support groups, day care facilities or respite care may help patients and their families cope with the situation and may even postpone institutionalization. This multidisciplinary research project focuses on the course of YOD, the functional characteristics of YOD patients, the needs of YOD patients and their caregivers, the risk factors for institutionalization and the interaction with the caring environment. We expect that the study will yield important data that can be used to design specific guidelines and improve the development of health care services for YOD patients and their families.

Aim and research questions

NeedYD is a prospective cohort study with the following primary objectives: (1) to investigate the (un)met needs of YOD patients and their family members during different phases in the course of the disease (e.g., the diagnostic phase and the phase in which (specialized) day care is provided) and (2) to investigate the course of neuropsychiatric symptoms and possible risk factors (e.g., comorbidity, age, communication problems, biological factors, disease awareness, interaction with environment).
The secondary objectives are:

- To gain insight into the course of other functional domains in YOD (cognition, activities of daily living);
- To explore the experiences and feelings of patients and their caregivers during the diagnostic period;
- To investigate the impact of the diagnosis of dementia on YOD patients and their family members;
- To study the course of functioning of the caregivers of YOD patients and the problems they (and possibly other family members) experience;
- To identify factors that influence the use of respite care and determine institutionalization;
- To explore to what extent stigma and taboo concerning dementia interfere with adequate communication within the family;
- To compare these findings with findings of studies on LOD.

Methods

Design

The NeedYD-study is a prospective cohort study with a follow-up of two years in which a group of YOD patients and their families were assessed at six month intervals. The study design is similar to that of the MAAstricht Study of BEhavior in Dementia (MAASBED) and the WAAL BEhavior in Dementia (WAALBED) study,[16-19] conducted in the Netherlands.

Subjects

The study population consisted of dyads of patients with YOD and their caregivers. Patients with onset of disease symptoms before the age of 65 are included in the study (age at inclusion could be later than 65). Diagnoses of dementia subtype were made according to regular criteria.[20-25] Patients were recruited through the memory clinics of the three Alzheimer's centers in the Netherlands located in Amsterdam, Nijmegen and Maastricht, the memory clinics of general hospitals and through other mental health services in the south of the Netherlands as well as through specialized day care facilities that are affiliated with the Dutch National YOD Taskforce. Thus, a group of patients without day care or receiving non-specialized day care, as well as a group of patients receiving specialized day care, were included in the study. For some of the research questions, these groups will be compared. The exclusion criteria were: (1) dementia caused by HIV, traumatic brain injury, Down's syndrome, Huntington's chorea or alcohol-related dementia, (2) lack of a reliable informant or (3) lack of informed consent of the participant. Furthermore, children of YOD patients who were living at home and were older than 14 years of age at the time of the baseline assessments were recruited through their parents.

Measures

Primary outcome measures

(Un)met needs were assessed with the Dutch version of the Camberwell Assessment of Needs in the Elderly (CANE).[26, 27] This assessment is a semi-structured interview consisting of 24 domains that cover social, physical, psychological and environmental needs. The interview started with an open question concerning a specific domain, followed by questions regarding help and (in)formal support the patient receives in that particular domain, as well as the amount of help and support that was needed. These items were scored on a three point scale ranging from little (1) to a lot of help (3). Satisfaction with the amount and quality of the help and support received was also assessed. The answers were used to determine whether or not the participant experienced a need and whether or not this need was met. The experienced needs of patients were based on patient and proxy (primary caregiver) interviews. The need for information and the psychological needs of the caregiver were also assessed by means of the CANE. Reliability and validity were found to be adequate. [26, 27]

Neuropsychiatric symptoms in the patient and related caregiver burden were assessed with the Dutch version of the Neuropsychiatric Inventory (NPI).[28] The NPI is a structured interview with the primary caregiver and, when available, a health care professional. After institutionalization, the nursing home version of the NPI (NPI-NH) was used.[29] Ten neuropsychiatric and two neuro-vegetative symptoms were assessed: delusions, hallucinations, agitation/aggression, dysphoria, anxiety, euphoria, apathy, disinhibition, irritability/lability, aberrant motor behavior, night-time behavior disturbances and appetite/eating abnormalities. Screening questions were asked to determine whether behavioral changes were present. In the case of a positive answer, further questions were asked and the severity and frequency of the behavioral disturbances were determined. The Dutch version of the NPI was found to have high inter-rater agreement and to be a valid rating scale for measuring a wide range of behavioral and psychological symptoms of dementia.[30] Furthermore, the experience of caregiver distress due to these neuropsychiatric symptoms was determined according to the six point NPI caregiver distress scale (NPI-D) ranging from no distress (0) to extreme distress (5).[31] The NPI-D provides a reliable and valid measure of subjective caregiver distress in relation to the neuropsychiatric symptoms measured by the NPI.

For an overview of all measurements see Table 1.

Secondary outcome measures for the patient

The Global Deterioration Scale (GDS) was administered to assess the severity of the dementia. The GDS is a widely used instrument which has been validated against behavioral, neuro-anatomic and neurophysiologic measures, for which significant correlations have been found.[32] The Interview for Deterioration in Daily Living in Dementia (IDDD) was used to assess the activities of daily living. The internal consistency of this scale is high (Cronbach's alpha 0.94).[33] Cognitive functioning was measured using the Mini Mental State Examination (MMSE), which is a reliable and valid test of cognitive function.[34] When the MMSE score was below 15, the Short Severe Impairment Battery (s-SIB) was administered, which has been found to be a reliable and valid test of cognitive function in moderate to severe dementia patients.[35, 36] Furthermore, executive functioning was assessed using the Frontal Assessment Battery (FAB). The FAB has good inter-rater reliability, internal consistency and discriminant validity.[37] The Guidelines for the Rating of Awareness of Deficits (GRAD) [38, 39] were administered in order to investigate disease awareness. This instrument has substantial inter-rater reliability.[40] The Quality of Life-Alzheimer's Disease scale (QoL-AD), which has good content, criterion and construct validity and excellent inter-rater reliability and internal consistency,[41] was used to assess the quality of life of the patient, as perceived by the patient and his caregiver. The Dutch version of the Cornell Scale for Depression in Dementia (CSDD) [42, 43] was administered to identify depressive symptoms in the patient. This scale has adequate inter-rater reliability, internal consistency and sensitivity. The amount of formal care the patient receives and the time the caregiver spends caring for the patient were obtained using the Resource Utilization Scale (RUD-Lite), which covers 95% of the resource use, the complete RUD covers.[44] Therefore, it is a good alternative for the complete RUD when the assessment battery is large.

Table 1: Flowchart of measures used during the assessments

Outcome measure	**Operationalization (Type of instrument)**	**Time of assessment**					
Patient		S	B	F1	F2	F3	F4
Primary outcomes							
Needs	CANE [26, 27] (SSI)		p/c	p/c	p/c	p/c	p/c
Frequency and severity neuropsychiatric symptoms	NPI [28]		c/n	c/n	c/n	c/n	c/n
	NPI-NH [29] (SI)						
Secondary outcomes							
Severity of dementia	GDS [32] (RS)		p/c	p/c	p/c	p/c	p/c
Depressive symptoms	CSDD [42, 43] (SI)		c	c	c	c	c
Cognitive functioning	MMSE [34] (CT)		p	p	p	p	p
Cognitive functioning	SIB [35] If MMSE <15 (CT)		p	p	p	p	p
Executive functioning	FAB [37] (CT)		p		p		p
ADL disabilities	IDDD [33] (Q)		c		c		c
Disease awareness	GRAD [38, 39] (SSI)		p/c	p/c	p/c	p/c	p/c
Amount of formal and informal care	RUD-lite [44] (SI)		c	c	c	c	c
Quality of life	QoL-AD [41] (SI/Q)		p/c	p/c	p/c	p/c	p/c
Additional variables							
Inclusion/exclusion criteria		r/p/c					
Informed Consent		p/c					
Demographic data	Age, gender, education level, marital status, employment		p/c				
Diagnosis	First complaints, date of diagnosis, physician that gave diagnosis		p/c				
Life events	Disease, institutionalization, conflict, divorce, other		p/c	p/c	p/c	p/c	p/c
Medical record investigation	Current diagnosis, possible prior diagnoses, examinations that lead to diagnosis, medical history		r				
Treatment and other information	Physical complaints, current treatment/ use of formal care, medical history, substance use, dementia/ genetic diseases in family		p/c	p/c	p/c	p/c	p/c

Continuation table 1 on next page

Continuation table 1: Flowchart of measures used during the assessments

Outcome measure	**Operationalization (Type of instrument)**	**Time of assessment**					
Caregiver							
Primary outcomes							
Needs	CANE [26, 27] (SSI)		c	c	c	c	c
Experienced burden as a result of behavioral disturbances	NPI [28] (SI)		c/n	c/n	c/n	c/n	c/n
Needs and experiences	(SSI)		c		c		c
Sense of competence	SSCQ [45] (SI)		c	c	c	c	c
Secondary outcomes							
Depressive symptoms	MADRS [46]. (SI)		c		c		c
Psychological and somatic complaints	SCL-90 [47] (Q)		c	c	c	c	c
Coping strategies	UCL (Schreurs, Willige et al. 1988) (Q)		c				
Quality of life	RAND-36 [48] (Q)		c		c		c
Quality of the marital relationship	Four items of the University of Southern California Longitudinal Study of Three-Generation Families measures of positive affect [49]		c	c	c	c	c
Emotional instability	Subscale neuroticism of NEO-FF-I [50] (Q)		c				
Caregiver management strategy	Caregiver management strategy [15] (SI)		c	c	c	c	c
Additional variables							
In/exclusion criteria		c					
Informed consent		c					
Demographic data	Age, gender, education level, marital status, employment		c				
Information on informal care	hours giving care, contact hours, other informal caregivers		c	c	c	c	c
Information on employment	Hours working, date stopped working		c	c	c	c	c
Children							
Needs and experiences	(SSI)		ch				
Demographic data	Age, gender, education level, employment, living situation		ch				
Data on informal care	Hours spent care giving, contact hours with demented parent		ch				

S= Screening, B= Baseline, F1= Follow up measurement 1, F2= Follow up measurement 2, F3= Follow up measurement 3, F4= Follow up measurement, SSI= Semi Structured Interview, SI= Structured Interview, CT= Cognitive Test, Q= Questionnaire, RS= Rating Scale, C= informant is caregiver, P= Informant is patient, N= Informant is health care professional from nursing home, Ch= informant is child

Secondary outcome measures for the caregiver

The Short Sense of Competence Questionnaire (SSCQ) [45] was administered to assess caregiver's feelings of being capable to care for a demented individual. The SSCQ was reported to have satisfactory reliability and validity.[45] Depressive symptoms were measured by the Montgomery Asberg Depression Rating Scale (MADRS), which has adequate inter-rater reliability and exhibits construct and concurrent validity.[46] Psychological and physical complaints were measured with the Symptom Checklist 90 (SCL-90). Reliability and construct validity of the SCL-90 are satisfactory.[47] Emotional instability was assessed with the neuroticism subscale of the Dutch version of the NEO- Five Factor Inventory (NEO-FF-I). Internal consistency and test-retest reliability are high for this scale, as is the construct validity.[51] Coping strategies were assessed by means of the Utrechtse Copinglijst (UCL). The reliability of this scale is reasonable and the validity has been found to be sufficient despite some inconsistencies in the literature.[52] General health was measured with the Dutch translation of the RAND-36.[48] The Dutch version of the RAND-36 appears to be a reliable, valid and sensitive measure for general health.[53] In addition, the quality of the marital relationship and the changes that have occurred since the onset of the disease were measured by four items of the University of Southern California Longitudinal Study of Three-Generation Families measures of positive affect. Cronbach's alpha for this scale is 0.85.[49]The caregiver management strategy was assessed by means of questions reflecting three caregiver strategies: a caring, supporting or non-adapting strategy.[15] This scale has not yet been validated. Furthermore, a semi-structured interview was administered to the caregiver and, when applicable, to children living at home. The interview included topics concerning the period prior to diagnosis, the impact of the diagnosis, changes in the interpersonal relationships within the family, the communication within the family about the disease, the problems experienced by the patient and family members, experiences and beliefs concerning (in)formal support and health care services, transitions in care (e.g., day care, institutionalization) and future perspectives.

Additional data

By means of a structured interview and examining the patients file information, medical and demographical information of the patient were obtained. For a full description of these data, see Table 1.

Procedures

YOD patients and their caregivers received five assessments at six-month intervals (B, F1, F2, F3, F4; Table 1). Before inclusion in the study, (S) information on in/exclusion criteria was collected and informed consent was obtained. Patients who were not able to sign informed consent were asked to give oral consent and their legal representative had to give written consent that the patient was able to participate. Children living at home who were older than age 14 were asked at baseline to participate in a semi-structured interview. Children aged between 15 and 18 years, as well as their legal representatives, both had to sign informed consent. When individuals did not agree to participate, the reason plus age, gender and diagnosis of the patients were registered.

When participants, after inclusion in the study, did not wish to participate in one of the assessments, caregivers were asked to participate in an interview by telephone, so the CANE, NPI and Sense of Competence questionnaire could still be administered and to fill out all of the questionnaires required for that assessment. If this was not possible, the researcher asked them to answer several questions about their own and the patients' functioning and about the use of formal care. If caregivers refused this as well, the reason for refusal was asked. When a patient had died, data on the use of (in)formal care and the needs of caregivers before and after the patient died was collected from the caregiver as well as the date and cause of death.

Ethical considerations

The study protocol was approved by the Medical Ethics Committee of the University Medical Center Maastricht. The local ethics committees of the participating institutions have also given consent. The research project was performed according to the principles of the Declaration of Helsinki (version January 2004; www.wma.net) and in agreement with the law regarding medical-scientific research in humans (WMO). An independent physician was assigned to the study. Participants were informed about the possibility of contacting him for further questions about the study.

Sample size

Based on a power calculation (two groups: diagnostic phase and the phase of specialized day care; ANOVA) with an alpha of 0.05, a power of 0.85 and an expected effect size of 0.25, 128 YOD patients were required to participate in the study. With an expected loss to follow-up of 37% in a two year follow-up period based on data of the MAAstricht Study of BEhavior in Dementia (MAASBED) study,[15] 200 patients needed to be included.

Data analysis

Data entry was performed twice to safeguard data integrity. Statistical analyses were performed using the Statistical Package for Social Sciences. Descriptive statistics were used to describe characteristics of patients and caregivers, i.e., age, sex, distribution of diagnoses, etc. Both quantitative and qualitative data were used in the analyses.

Diagnosis matched patients with LOD from a historical cohort (MAASBED study) were used to make a comparison with YOD for some of the research questions. Baseline differences between groups were analyzed to investigate the comparability of the groups. Depending on the research question and which variables were analyzed, parametric or non-parametric analyses were performed. Comparisons between groups were made with independent samples t-tests or AN(C)OVAs for continuous and normally distributed variables and Pearson's Chi square test, Fisher's exact or Mann-Whitney U tests for categorical and non-normally distributed variables. Comparisons between the memory clinic and day care groups, the YOD group and the LOD group and within groups across measurements will be performed using linear mixed models analyses. A survival analysis will be performed to study predictors of institutionalization. If participants withdrew from the study, they were not excluded. The data collected could still be analyzed, because of the use of linear mixed models. However, the characteristics of the dropouts and losses to follow-up were described and taken into account.

Qualitative data were analyzed using the method of constant comparative analysis.[54] These qualitative analyses were performed in order to obtain a more in-depth understanding of the experiences of YOD patients and their family members. The interviews that were held with the caregivers were transcribed and first read by one researcher. They were then read a second time to develop codes that were grouped into categories. Categories were grouped into themes. Another researcher independently applied the same procedure. The analyses were performed using Atlas.ti.

Discussion

The current paper presents the study protocol of a prospective cohort study: the NeedYD-study. This project mainly focuses on the course of YOD, the functional characteristics of YOD patients, the needs of YOD patients and their caregivers, the risk factors for institutionalization and the interaction with the caring environment.
To our knowledge, this is the first study that addresses these issues longitudinally in a large cohort. It will contribute widely to our knowledge about the course of YOD, the caring process and the needs of the patient and caregiver as they develop during the course of the disease. A longitudinal design is necessary to examine the predictive value of study variables in observational data. Despite the many positive aspects of this design, there are some limitations.

Sample bias could be a factor in our study. Although patients are recruited through a large range of different institutions, which is likely to be representative of the Dutch population, the group that gives consent may be different from the group that refuses to participate. Furthermore, selective attrition due to early death is inherently associated with the current study.

In addition, the data from the present study and the historical cohort of the LOD sample (MaasBED study) are different in several ways, as the participants are not matched. The LOD patients were mostly seen right after receiving the diagnosis, whereas in the YOD group patients in different stages are included. The YOD group is, therefore, probably more heterogeneous in terms of disease severity, cognitive functioning and ADL disability. Furthermore, the dementia of the YOD patients is possibly more severe because establishing a diagnosis in YOD often takes longer than in LOD. However, these factors as well as other possible confounders are collected in order to take these into account during the statistical analyses.

Furthermore, one may argue that the proxy ratings we use for several patient characteristics are not as reliable as patient ratings. However, in this patient group such ratings are inevitable since dementia patients gradually become cognitively impaired and may suffer from a lack of awareness. Therefore, proxy ratings are preferred to keep the informant during follow-up reliable and constant.

In conclusion, the strengths of this study outweigh its few limitations as long as they are dealt with properly.

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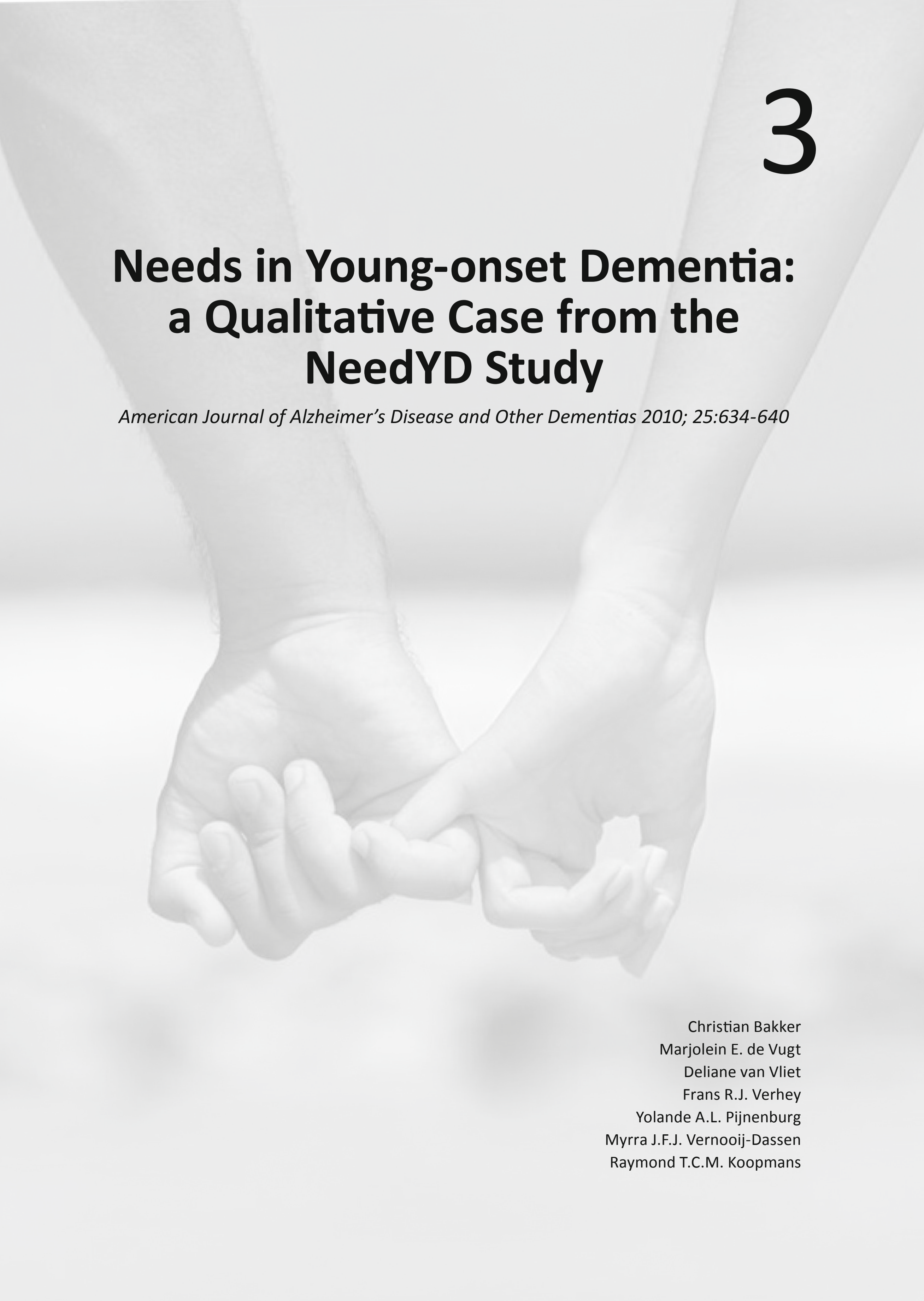

3

Needs in Young-onset Dementia: a Qualitative Case from the NeedYD Study

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Christian Bakker
Marjolein E. de Vugt
Deliane van Vliet
Frans R.J. Verhey
Yolande A.L. Pijnenburg
Myrra J.F.J. Vernooij-Dassen
Raymond T.C.M. Koopmans

Abstract

Objectives: The aim was to explore the experiences of a caregiver of a young-onset dementia (YOD) patient and the needs of patient and caregiver.

Methods: A single case study design was used to explore (1) (un)met needs of patient and caregiver (2) caregiver's experiences of transitions in care and health care services. A qualitative analysis was used to examine the data.

Results: This study shows that a YOD patient and caregiver were confronted with specific issues during the care process, namely (1) prolonged time to diagnosis, (2) a lack of fit between needs and services, (3) the strain of dedication to care versus the caregiver's own future perspective (4) the need for response of health care services to changing individual preferences.

Conclusion: This study illustrates specific issues related to YOD that require specialized YOD care such as the differential and changing needs of patients and caregivers.

Introduction

Dementia is typically regarded as a disease of old age, but in 3-5% of the patients, onset of the disease occurs before the age of 65.[1-4] When dementia occurs at an early age, the psychosocial implications for both the patient and family are numerous. Young-onset dementia (YOD) patients are in a life phase in which they often play an active role in society and may have young children. The loss of roles and responsibilities is, therefore, greater than that of older people. They must also deal with specific issues such as difficulties in obtaining a proper diagnosis, marital problems, family conflict, (un)employment and financial issues.[5-7] Furthermore, many YOD patients of the post-war generation grew up in a society that is quite different from that of the older generation. The needs of YOD patients may as a result differ from the needs of late onset dementia patients and demand a different approach. A need is defined here as a situation in which an individual experiences a significant problem that impedes functioning

Despite these differences, the availability of specialized healthcare services remains limited in most countries. This forces YOD patients and their family members to use services that are designed for the elderly and are less likely to meet their needs. At present, there is no agreement among researchers regarding whether specific guidelines and services should be developed to meet the needs of these younger individuals and their families or whether existing healthcare services must be more responsive to these specific needs.[8] This stresses the importance of gaining insight into all possible needs of YOD patients and their families.

From research on needs of older people with dementia in residential care, it is known that unmet needs are associated with an increase in neuropsychiatric symptoms and that younger age is associated with a higher number of unmet needs.[9] Roest et al. (2007) found that the needs of older dementia patients and their caregivers are less related to instrumental needs, but more associated with how they cope with the disease and wellbeing.[10] It is, to our knowledge, unclear whether this is also true for YOD patients and their caregivers. Furthermore, domains that influence the quality of life of the dementia patient and his/her primary caregiver are subject to change as the disease progresses and, with that, needs are also likely to change. Transitions in care, for example when daycare begins or the patient is admitted into a nursing home, especially lead to a shift in the hierarchy of the domains that influence the perception of quality of life.[11]

Most of the current research on needs and services for dementia patients and their caregivers focuses on elderly sufferers. Specific knowledge on the characteristics and needs of YOD patients and their families is lacking, but this knowledge is an important prerequisite for the development of suitable health care services. Adequate diagnosis, (in)formal support and services such as support groups, daycare facilities and respite care may help patients and their families cope with the situation and may even postpone institutionalization.[12] Especially in YOD, there is a need to understand the effect of the disease on both patients' and family members' functioning and family roles with regard to their specific life phase.

In the current case report, we explored the experiences of a spouse of a young person with dementia, and studied how the needs of both the patient and his wife developed and changed over time from the perspective of the spouse. Furthermore, we explored experiences and views concerning provided health care services.

Methods

We used a single case study design. Cognitive functioning of the patient, disease state and neuropsychiatric symptoms were assessed with validated measures. Qualitative interviews with the caregiver were used to explore how the caregiver perceived met and unmet needs of both the patient and caregiver and how she experienced transitions in care and health care services provided. We used qualitative methods because they are especially suitable to study complex topics and allowed us to thoroughly explore experiences and needs.[13] A patient who underwent transitions in care was randomly selected from a sample of 217 cases of a two-year follow-up study on Needs in Young-onset Dementia (NEEDYD).[12] Data were collected at time of inclusion and after six and twelve months. Written informed consent was obtained for both the patient and caregiver.

Cognitive functioning of the patient at baseline was assessed with the Mini Mental State Examination.[14] This is a widely used screening instrument in dementia. In addition, the Short Severe Impairment Battery (SIB) was used because it is especially suitable for assessing cognitive functioning in severely impaired dementia patients.[15,16]

Severity of the dementia was assessed with the Global Deterioration Scale using a seven point scale (1-7), ranging from 'no global impairment' (1) to 'very severe global impairment' (7).[17] Neuropsychiatric symptoms and caregiver burden were assessed at baseline, six and twelve months using the Neuropsychiatric Inventory (NPI).[18] Depressive symptoms of the caregiver were measured at baseline and after twelve months using the Montgomery Asberg Depression Rating Scale (MADRS), which has adequate inter-rater reliability and exhibits construct and concurrent validity.[19]

A trained researcher (CB) held semi-structured interviews at baseline and after twelve months according to an interview guide (see topics listed in Table 1) with topics derived from clinical practice and the relevant literature. In the NeedYD study the Camberwell Assessment of Needs in the Elderly (CANE) is used to assess needs of patient and caregiver.[20, 21] Because of the explorative character of this case study to gain new insights into the complex experiences and evolving needs of the caregiver, we used the narratives as a result of the administration of the CANE, for the qualitative analysis in addition to the data of the semi-structured interviews.

The audiotapes of the semi-structured interviews were fully transcribed. An inductive content analysis[22] was used by the first author (CB) to compare similarities and differences in order to understand relationships in the data concerning problems met, perceived needs of both the patient and his wife and experiences with health services. Quotes of the caregiver were coded. Codes referring to the same phenomenon were grouped into categories and the categories were grouped into themes. A second researcher (MdV) independently applied the same procedure. After thorough discussion, consensus was reached about categories, subcategories and the major themes.

Table 1. Interview guide

Period prior to diagnosis
Diagnosis
Period after diagnosis
Caring
Transitions in care
Future perspective

Results

Case description

The patient was a 59-year-old male with young-onset Alzheimer's disease. The first symptoms retrospectively occurred when the patient was 50 years old. At that time, he often forgot appointments, experienced difficulties in managing finances and also lost his job as a salesman because he was not able to function properly. He attempted to obtain other jobs, but without success. At the time of the first symptoms of the disease, his wife was 46 and she still worked full-time. She noticed that her husband showed less initiative, experienced difficulties executing simple household tasks and began drinking excessively. She also noticed that her husband became irritated when she confronted him with these things. During the five years that followed, both these functional problems and neuropsychiatric symptoms became more severe and she sent her husband to the general practitioner (GP). The GP and a psychologist both thought that her husband was not suffering from a serious problem and suggested a minor concentration problem as well as marital difficulties. Nearly one year passed and symptoms worsened. When the patient's wife forced a referral to a memory clinic, a diagnosis of young-onset Alzheimer's disease was established within three weeks. The patient was included in the NeedYD study at the age of 59 (patient's baseline characteristics are summarized in table 2). At that time, he was still living at home and visited a specialized daycare centre five days a week.

At the time of inclusion, the cognitive functioning of the patient was severely impaired (table 2). Furthermore, the caregiver reported delusions, hallucinations, agitation, depression, anxiety, euphoria, disinhibition, irritability, aberrant motor behavior and nighttime disturbances on the NPI (NPI frequency x severity total score: 39). The caregiver found these neuropsychiatric symptoms to be quite distressing (NPI-D total score: 33). At that time, she also reported minor feelings of tension and lassitude, but no other depressive symptoms (MADRS total score: 4/60). In the first month after inclusion, the severity of neuropsychiatric symptoms and especially anxiety and aggressive behavior worsened according to the caregiver and the patient was admitted into a nursing home at a dementia special care unit for the elderly. In spite of the prescription of psychotropic medication, neuropsychiatric symptoms continued, and apathy/indifference was also observed as a new behavioral problem at that time.

Two months after admission, the patient was transferred to a special care unit for YOD patients. We visited the patient a second time and it appeared that the frequency and severity of neuropsychiatric symptoms had decreased (NPI total score: 15). However, at time of the third assessment, agitation/aggression and irritability reappeared, especially when the nursing staff offered help with activities of daily living. Furthermore, apathy and aberrant motor behavior continued (NPI total score: 20). At that time the neuropsychiatric symptoms were moderately distressing for the caregiver (NPI-D total score: 13). The caregiver experienced no depressive symptoms at the third assessment (MADRS total score: 1/60).

Table 2. Baseline characteristics of the patient

Gender	Male
Diagnosis	Alzheimer’s disease
Age at onset	50
Age at diagnosis	56
Age at inclusion in the NeedYD study	59
Disease state	
Global Deterioration Scale – score	5/7
Cognitive functioning	
Mini Mental State Examination – score	4/30
Short Impairment Battery – score	24/37

Note: NeedYD = Needs in Young-onset Dementia

Qualitative results

Analysis of the qualitative data resulted in three major themes in the caregiver's experiences of the care process: (1) Making choices in the care situation, (2) Conditions for the use of care, and (3) Involvement in care. An overview of codes, categories and themes is provided in table 3.

Making choices in the care situation

Throughout the course of the disease, the caregiver encountered a range of emotional and moral dilemmas in which the decision making *process* was an important issue, leading to an increase in experienced caregiver burden. The *reciprocity* in the relationship gradually diminished and, in the end, the caregiver was forced to make all of the decisions concerning work, finances, the household and the care of her husband.

> Cg: '*I really felt abandoned. At that time, I really thought: I am going to take care of things myself because managing all these things together with my husband is not going to work.'*
> (…) *'I had all these things on my mind, and my husband just sat there all day doing nothing.'*

A proper and timely diagnosis was perceived as an important prerequisite for (1) being able to perceive the (behavioral) problems that occurred such as aggression, irritability and excessive drinking of her husband as symptoms caused by the disease and (2) obtaining appropriate care in time. The patient denied that he had a problem.

> Cg: *'At that time, I did not know that he was ill. He did not want to see a doctor. I thought: If you are not ill, I will leave you….it could have ended in a divorce.'*

Furthermore, the GP and the psychologist that she and her husband visited did not recognize the disease. This resulted in a delay of the caregiver's search for appropriate support and in feelings of guilt.

> Cg: *'My husband went three times and the psychologist was more or less angry with me. I had to give my husband more space; there was no dementia, no brain tumor, only a minor concentration problem. I left and thought: maybe I am interfering with his life too much. I blamed myself.'*

Only after the diagnosis was established the caregiver was able to *change her perspective* on the situation and to seek appropriate support such as daycare and a support group. However difficult, the diagnosis was an important marking point, and led the caregiver to decide to stay in the marriage and care for her husband. It also allowed her to begin *grieving*.

> Cg: *'Finally, I had the answer to what had gone wrong in the past four, five years.'*
> Cg: *'When my husband was asleep, I was in the living room, crying and thinking, not knowing what the future might bring.' (…) 'I was only just 50 years old.' (…) 'How devastating for him…'*

Furthermore, the *duality* in the caring situation, combining continuing care with maintaining a life of her own, was an important issue from the perspective of the caregiver. The caregiver gradually realized (with the support she received from the coordinator at the daycare centre) that in order to remain able to care for her husband, she also had to meet her own needs.

> Cg: '*I would like to be able to enjoy myself again, but I am not sure that I really can.' (…) 'But I will have to try. Imagine that he will be at home for two more years. No vacation for two or three years with continuing care. When he's at home I constantly have to be on my guard. If I do something wrong, he gets angry, thus I do everything to keep him happy.'*

In terms of respite care, she expressed:

> Cg: *'It is very difficult, but I do know, and I approach this rationally, he has to be there for a few nights. As a result, I will be able to care for him at home much longer, he has got to get used to being here. This will become his future home eventually.'* (…)*'The fact that he has daycare and sleeps at the nursing home two days a week, makes that I have been given back a piece of my life.'*

Conditions for the use of care

As the disease progressed and especially during transitions in care, such as the start of daycare or the admission of the patient into the nursing home, needs *changed*. At times, there was a need for specific information about the course of the disease, dealing with neuropsychiatric symptoms, psychosocial problems of the patient or practical information about addressing the personal or instrumental needs of the patient. This information, for example, was provided by the coordinator of the daycare centre and partially by a caregiver support group that she infrequently visited:

> Cg: *'When my husband gets in a different stage, I just want to know how others deal with that. I don't have to go every month, which would be too much.'* (…) *'I do not want to listen to their stories; I have enough on my mind as is.'*

There was also a need for c*onfirmation* from health care professionals of the decisions that the caregiver made, such as planning a vacation and admitting her husband into the nursing home.

> Cg: *'I thought I will not take that vacation, but then I talked to the coordinator of the daycare centre and she said: "If you never take a few days off, you will not be able to manage." And that is true.'* (…) *'I am glad that she said that.'*
>
> Cg: *'Maybe I can talk to a psychologist who can alter my way of thinking. Maybe he can take away the guilt.'*

As needs changed, the *availability* of supportive professional caregivers was mentioned as another important prerequisite throughout the caring process.

> Cg: '*You have to grow into each stage of the disease. But I do need the support of others, who you can call, whenever you need to. I have that now here at the daycare centre, I can call them.'* (…) *'We have a fine GP and neurologist, I can always call them.'*

Furthermore, there was also a need for *continuity* in care, meaning the availability of support throughout the course of the disease for both the patient and caregiver from the same health care professionals**.** This was important, for example, when talking about the fact that she had met another man and that this fact troubled her:

> Cg: '*Friends do understand. I do know friends I can talk to about these things, but they are not professionals.'* (…) *'What I do need now is a professional I can talk to about the changes in my life.'* (…) *'You would think that would be possible at the nursing home where my husband is staying. Especially for partners of young dementia patients who want to pick up their own lives again. They should be much more able to understand these kinds of issues.'*

Involvement in care

The great commitment to the care of her husband resulted in little room for other things in the caregiver's life, such as a personal job, social contacts and relaxing.

> Cg: *'The most important issue for me is that he is comfortable. I hope, in spite of everything, that he is happy.'* (…) *'I spent so much time caring for him. My life revolves around Alzheimer's disease.'*

Despite the caregiver's dedication during the caring process, it became clear that (1) the caregiver and her husband needed additional help from others, such as friends and professionals, and (2) the caregiver *had to let go* and allow others to gradually take over.

> Cg: '*I just want to know how we can get through this together. You have to accept practical help. I saw people who did not want help for their partner, wanted to do everything themselves. But then, you are not going to cope. I do not want to listen to them. I want to do it my way. That is better for me.'*

Regarding the decision to admit her husband into the nursing home, she stated:

> Cg: '*My biggest concern is that I will not be able to let go. I don't want to let go. I hope I will not have the urge to go see him every day. I hope that I can let go, let go of caring. I suppose I have to grow into that as well.'*

In this respect, *trust* was also an important issue that influenced the caregiver's ability to allow others to tend to the needs of her husband. Especially when there was not a good *fit* between support, services and the needs of her husband, this was particularly distressing for both the patient and caregiver. When talking about the first weeks of admission into a dementia special care unit for the elderly in the nursing home, the caregiver expressed:

> Cg: *'At first, he was very rebellious; he did not want people helping him.'* (…) *'He was among older people with dementia with fewer activities and he just walked around there, lost.'* (…) *'It was very difficult for him.'* (…) *'He was in distress during that period. He said: "I'll jump of that roof".*

The importance of a good fit between needs and services is illustrated by the transfer of the patient to a special care unit for YOD patients. A few months after the transfer, the patient's neuropsychiatric symptoms decreased and wellbeing improved. This also enabled the caregiver to reduce her share in the care for her husband.

> Cg: *'This unit is right for him.'* (…) *'Now I let them take care of him. I also have confidence in them now. Before, I constantly had the feeling I had to visit him. I can somewhat let go now because I can see there is more personal attention from people working at this unit.'* (…) *'I think the way they approach people in this unit is different. I think it is less patronizing, more direct, more personal.'*

Although the care environment seemed to be an improvement for the patient, the caregiver noticed that the personal needs of her husband were not fully met by the nurses. This influenced her perception of her role in the caring process while her husband was residing at the nursing home. When talking about his incontinence, she stated:

> Cg: *'I already told them before; you have to change his incontinence material (diapers) regularly. I already noticed several times that he is wet, just because they don't replace the incontinence material in time.'* (…) *'I wonder what happens when I am not there.'*

Table 3. Overview of codes, categories and major themes

Themes	Categories	Codes
Making choices in the care situation	Decision making process	Making decisions without the patient, ambivalence, combining (new) roles, fear of diagnosis, financial and legal issues, patients' approval, social consequences.
	Diminishing reciprocity	Loss of roles, feelings of abandonment, loneliness, giving and taking, quality of the relationship, communication.
	Changing perspectives	Diagnostic issues, changing symptoms of the disease, grief, burden of care.
	The duality in the caring situation	Entanglement, maintaining a life of your own.
	Adjusting to change	Dealing with loss, dependency, coping with behavioral changes.
	Need for information	Questions about symptoms of the disease, the course of the disease, practical issues, availability of services, legal/financial issues.
Conditions for the use of care	Changing needs	Practical needs, functional needs, activity, being among peers.
	Confirmation	need for reassurance, guidance, comprehension.
	Availability of (in)formal support	Safety net, practical aid, someone to talk to, social support.
	Continuity in care	Understanding YOD, burden of transitions in care, access to the same professionals.
Involvement in care	Commitment and dedication to care	Feelings of guilt, concern for the patients' future, fear of harm, comfort patient, changing relationship.
	The process of letting go	Retain control, adjusting to change, personal growth, ambivalence, emotional problems, practical boundaries.
	Trust	Experiences with (in)formal care, referral to appropriate services, image of health care services, medical care.
	The fit between needs and services	Ability of services to meet the patients needs, patients' personal needs, selfhood and self esteem, resisting care.

Note: YOD = Young-onset Dementia

Discussion

This case study revealed three major themes in the care process of a YOD patient. First, the caregiver perceived the decision making process as complicated and stressful. The decision making process was complicated by problems related to the specific life phase of the patient and his caregiver. Second, the fit between the needs of both the patient and caregiver and the characteristics of the available support and services was a main concern for the caregiver. As the disease progressed, and especially when there were transitions in care, the expectations towards healthcare services and (in)formal support changed. Third, the involvement of the caregiver during the care giving process was influenced by the caregiver's perception of the changing needs of her husband, the quality and availability of support and health care services as well as the dilemma between the dedication to caring for her husband by herself and her own changing future perspectives.

In the process of caring for a YOD patient, it appears as though the caregiver is confronted with difficulties and issues that are specifically related to their younger age and life phase. Besides the care for her husband, the caregiver was confronted with issues concerning work, financial difficulties, and the household, while her husband became increasingly unable to fulfill his roles as husband and financial provider. These conflicting roles are much more likely to occur in a younger, active life phase than in late life. Furthermore, the caregiver had to deal with severe neuropsychiatric symptoms that remained distressing throughout the caring process. Neuropsychiatric symptoms are known to have important adverse effects on the psychological and physical health of informal caregivers[23] and are suggested to be an especially important caregiver issue in YOD.[24] Moreover, this case strikingly showed the dilemma of a younger caregiver dedicated to the care of her family member versus her own future perspective. This complicated the decision making process while caring and led to an increase in perceived caregiver burden.

Additionally, obtaining a diagnosis is especially problematic for people with YOD, prolonging the period of uncertainty and adversely affecting the relationship between the patient and his family members. As illustrated by this case, YOD is often not recognized as such by health care professionals. This is likely due to the fact that YOD is not as prevalent as late onset dementia, has a different clinical manifestation (i.e., predominant behavioral changes such as those of our patient), and different cognitive symptoms compared to late onset dementia. This is consistent with the available literature that addresses this issue in YOD.[25-28] The timely diagnosis of young-onset Alzheimer's disease was an important prerequisite for the caregiver in our case to change her perspective on their situation, start grieving and to seek the appropriate support and health care services, such as specialized daycare, a support group for caregivers of YOD patients, individual support and assistance with neuropsychiatric symptoms and intermittent stays at the nursing home. In this case, the availability of these services and support enabled the caregiver to prolong the period that she could care for her husband at home.

As the disease progressed, the needs of both the patient and caregiver were subject to change. This case suggests that this is a dynamic process that is influenced by the cognitive and psychological state of the patient as well as by the presence of neuropsychiatric symptoms and the ability of both the patient and caregiver to cope with the consequences of the disease. From this case, it is also clear that a good fit between the developing needs and the support and healthcare services available is important. When these services were responsive to the individual needs of the patient and his caregiver, the patient's neuropsychiatric symptoms decreased. This also enabled the caregiver to entrust others with the care of her husband and eventually led to an increase in both the patient's and caregiver's wellbeing.

Services designed for the elderly, like the unit where the patient first resided after admission to the nursing home did not adequately address the needs of the patient. It is likely that services designed for the elderly have more difficulty recognizing the specific needs of these younger people and have less resources to meet those needs.[29] In our case, the overall combination of services specifically designed for YOD patients seemed beneficial for both patient and caregiver. The caregiver perceived that these YOD services did better fit her and her husband's needs, but still had difficulty addressing the functional and ADL needs of the patient and her need for individual support. Thus, also YOD services can be improved by further integrating care throughout the different stages of the dementia process and by offering the possibility for both patient and caregiver to receive care and support from the same health care professionals, like for instance a case manager during the course of the disease. It is likely that healthcare professionals are able to be more responsive to the individual wishes and preferences when they are involved throughout the course of the disease and are aware of the issues of both the patient and caregiver.

Conclusions

This study shows that a YOD patient and caregiver were confronted with specific issues during the caring process, namely (1) the prolonged time to diagnosis, (2) the fit between the patient and caregiver needs and the available health services, (3) the strain of dedication to care versus the caregiver's own future perspective and (4) the need for response of health care services to the changing individual wishes and preferences during the course of the disease. This study illustrates that there is a need to develop specific care and care programs for younger people with dementia and their families. Based on this study we suggest that these care programs should include the monitoring of changes in patient and caregivers' needs and address the issue of continuity in care. We believe that these younger patients and their families would benefit from the support of a casemanager that has specific knowledge on the issues in YOD and has insight in which YOD services are available.

This explorative study also has provided important insights into the domains that should be addressed in future (longitudinal) research. This research is needed to further explore the themes found in this case study in a larger group and to more closely examine the effect of a good fit with healthcare services on the functioning and wellbeing of both YOD patients and their caregivers.

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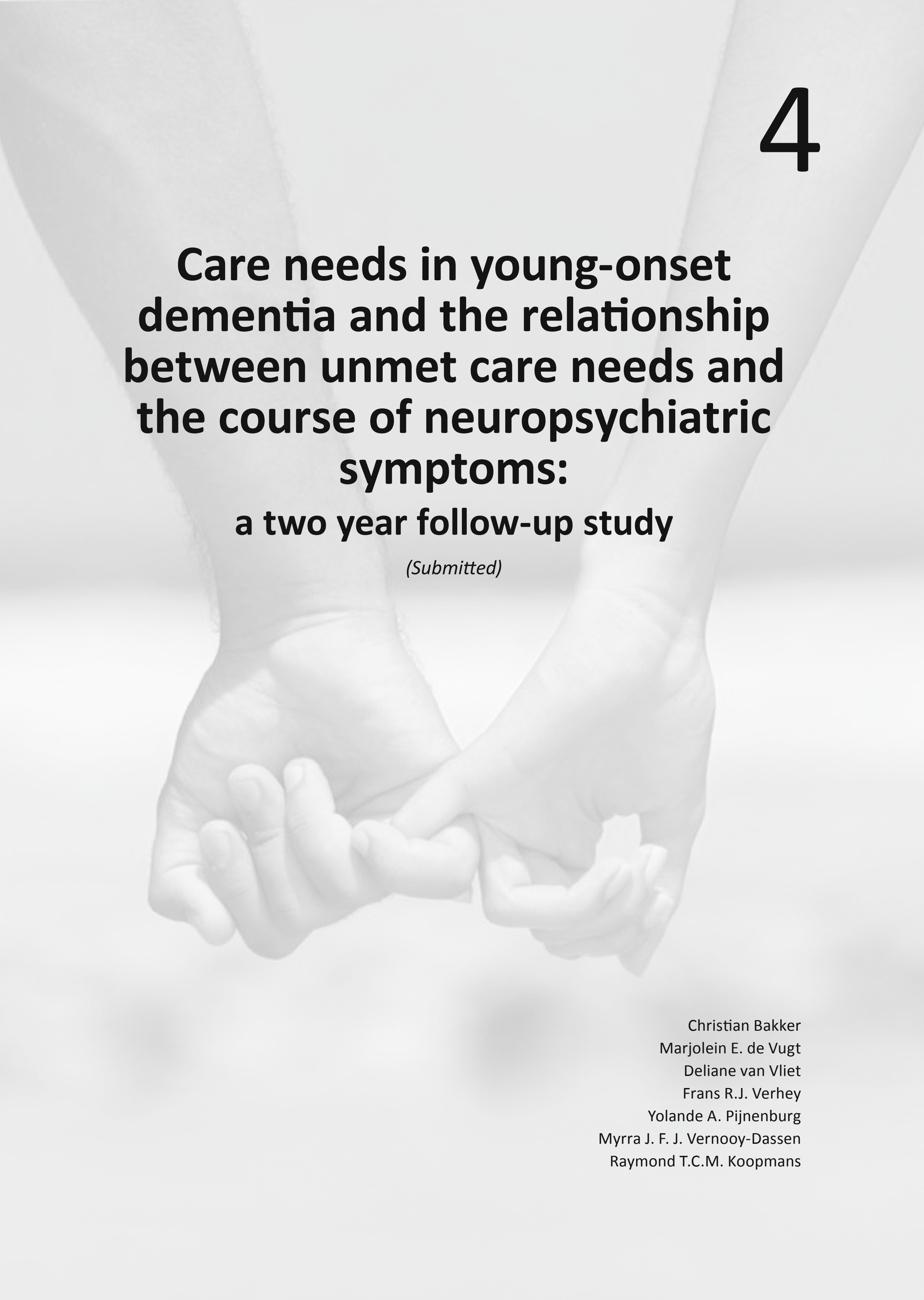


4

Care needs in young-onset dementia and the relationship between unmet care needs and the course of neuropsychiatric symptoms:

a two year follow-up study

(Submitted)

Christian Bakker
Marjolein E. de Vugt
Deliane van Vliet
Frans R.J. Verhey
Yolande A. Pijnenburg
Myrra J. F. J. Vernooy-Dassen
Raymond T.C.M. Koopmans

Abstract

Background: Little is known about care needs in young-onset dementia (YOD) while this information is essential for service provision and future care planning.

Objective: We explored (1) care needs of people with YOD, (2) the level of agreement within patient-caregiver dyads on (un)met care needs, and (3) the relationship between care needs and neuropsychiatric symptom levels over time.

Research design: A community-based prospective study of 215 YOD patients-caregiver dyads.

Measures: The level of agreement between patient and caregivers' report on the Camberwell Assessment of Need for the Elderly (CANE) was calculated using kappa (κ) coefficients. The relationship between (un)met needs and neuropsychiatric symptoms over time, assessed with the Neuropsychiatric Inventory (NPI), was explored using linear mixed models.

Results: Patients and caregivers generally agreed on the areas in which needs occurred. Only modest agreement existed within patient-caregiver dyads regarding whether needs could be met. Patients experienced high levels of unmet needs in areas such as daytime activities, social company, intimate relationships and information. A strong relationship existed between both met and unmet needs and neuropsychiatric symptoms over time.

Conclusions: Our findings indicate that in YOD there are specific areas of life in which unmet needs are more likely to occur. The high proportions of unmet needs and the relationship between (un)met needs and neuropsychiatric symptoms we found warrant interventions that target neuropsychiatric symptoms as well as the prevention of unmet needs. This underlines the importance of the periodic investigation of care needs, in which patient and caregiver perspectives are considered complementary.

Introduction

A diagnosis of dementia presents with serious consequences and care concerns at any age, but for younger dementia patients and their families, it is known to cause additional challenges.[1, 2] These challenges are associated with diagnostic issues,[3, 4] work-related and financial issues, loss of self-esteem and identity, and a sense of a disrupted future perspective.[1, 5] Young-onset dementia (YOD) caregivers must balance care with other responsibilities, such as work and parenting, and are often forced to prioritize caregiving responsibilities above their own future plans, such as career opportunities or further education.[6]

The care giving process in YOD is likely also hampered by the high levels of neuropsychiatric symptoms found in these patients.[7, 8] Neuropsychiatric symptoms are known to cause high levels of caregiver distress[9] and are considered a main predictor for institutionalization,[10] at least in late onset dementia (LOD). Neuropsychiatric symptoms often increase the patients reluctance to use services,[11] leading to a postponement of service use. YOD caregivers frequently postpone the use of formal care services until advanced stages of the disease.[12] In addition to the other personal and contextual factors known to delay service use in YOD, there is a lack of specialized dementia services, and mainstream dementia services have difficulty meeting the care needs of these younger patients and their families.[6, 13] At the same time, research regarding the care needs in YOD is largely lacking. This research is essential for providing appropriate care. Care needs that are not fulfilled are likely to have an adverse effect on patient and caregiver wellbeing and may negatively influence the course of neuropsychiatric symptoms.

Research on care needs in late onset dementia indicates a difference in the perspectives of patients and caregivers regarding the levels and areas of care needs.[14] In more advanced disease stages it will become increasingly difficult to establish care needs of persons with dementia, likely increasing the risk that their care needs will remain unfulfilled. This dilemma is in line with the need-driven, dementia-compromised behavior (NDB) model, which suggests that because of the inability of a person with dementia to make his or her needs known and the resulting difficulties of (in)formal caregivers in understanding these needs, neuropsychiatric symptoms may occur. Behavior in the NDB model is viewed as an attempt by the patient to communicate psychological or physical distress when unmet needs occur.[15] The literature suggests that reducing patients' unmet care needs leads to a decrease in the occurrence and severity of neuropsychiatric symptoms.[16] Kovach et al. argue that providing anticipatory care may help to prevent a cascading effect of unmet care needs on behavior, thereby enhancing the quality of life of both patients and caregivers.[17] Therefore, it is important to study the relationship between neuropsychiatric symptoms and care needs in YOD over time as well as the level of agreement between patients and caregivers regarding these care needs.

The aim of this study is to (1) explore the care needs of people with YOD, (2) explore patient and caregiver agreement on care needs and the extent to which these needs can be met and (3) explore the relationship between changes in care needs and changes in the severity of neuropsychiatric symptoms. We expect that the number of unmet needs in YOD increases over time, and these unmet needs are related to the severity of neuropsychiatric symptoms.

Methods

Subjects

This study is part of a Dutch prospective cohort study, the NeedYD study (Needs in Young-onset Dementia), which has been described in detail elsewhere.[18] This study recruited 215 participants with YOD with an age at onset before 65 years. Assessments with patients and their primary caregiver were performed every six months for two years. Patients were sampled from specialized day care facilities (n = 115) or were community dwelling (referred by University Medical Centers in Maastricht and Amsterdam (n = 56), regional hospitals (n = 10), or regional community mental health services (n = 20), or the patients applied to participate (n = 14)). Inclusion criteria were a diagnosis of dementia beginning ≤ 65 years and availability of an informant who had contact with the patients at least once a week. A dementia diagnosis was made on the basis of clinical, neuropsychological and brain imaging findings according to the criteria from the 4th edition of the Diagnostic and Statistical Manual of Mental Disorders, text revision (2000)[19], and the Dutch consensus guidelines,[20] which use internationally accepted criteria for diagnosing dementia subtypes[21-25]. Clinical diagnoses were checked against each patient's clinical patient file. Exclusion criteria were (1) lack of informed consent of the participant, (2) living in a nursing home or (3) dementia caused by HIV, traumatic brain injury, Down's syndrome, Huntington's disease or alcohol-related dementia.

Data collection and assessment instruments

Well-trained researchers and research assistants collected data using structured interviews and questionnaires.

Assessment of neuropsychiatric symptoms

Neuropsychiatric symptoms were assessed with the Dutch version of the *Neuropsychiatric Inventory (NPI)*.[26] This instrument has high inter-observer reliability and is a valid rating scale for a wide range of neuropsychiatric symptoms of dementia.[27] The frequency (F) and severity (S) of each symptom is rated on a five- (0-4) and four-point (0-3) Likert scale, respectively. Scores are summed to calculate total scores for four neuropsychiatric sub-syndromes, hyperactivity (agitation, euphoria, disinhibition, irritability and aberrant motor behavior), mood (depression and anxiety), psychosis (delusions, hallucinations and night-time behavioral disturbances) and apathy (apathy and appetite or eating abnormalities), based on the results of the European Alzheimer Disease Consortium.[28]

Assessment of care needs

(Un)met needs were assessed with the Dutch version of the *Camberwell Assessment of Need for the*

Elderly (CANE).[29, 30] The CANE is a semi-structured interview consisting of 24 domains that cover the social, physical, psychological, and environmental needs of the patient. The patient self-report and caregiver proxy were used to determine whether the patient experienced a need and whether this need was met. The reliability and validity of the CANE were found to be adequate.[29, 30]

Analyses

The analyses were performed using the Statistical Package for Social Sciences (SPSS) version 18.0.2 (SPSS Inc., Chicago, USA). Proportions or means were calculated to describe the characteristics of the patients and caregivers. Group comparisons were performed by Chi-square tests for categorical variables and t-tests for continuous variables. Frequency distributions were calculated by identifying met and unmet needs in the different areas of the CANE. The level of agreement between the patient's self-report and the caregiver's proxy was calculated using kappa (κ) coefficients, which were rated as having poor (0.00-0.20), fair (0.21–0.40), moderate (0.41–0.60), good (0.61–0.80) or very good agreement (81–1.00).[31] A Kruskal-Wallis test was performed to compare means across the five assessments of the total amount of reported needs. To examine the relationship between (un)met needs and neuropsychiatric symptoms over time, linear mixed models were used. Mixed models analysis allows for an estimation of an intercept and slope for each participant based on all available data for that individual, augmented by the data from the entire sample. Because missing data at one or more time points are common in prospective studies, the use of mixed models allows individual slopes to be estimated using the available data for that participant. In the model, neuropsychiatric symptoms (NPI total score) were included as the dependent variable, and the number of met and unmet needs were included as the key time-varying covariates. The model also included another time-varying covariate, disease severity (GDS; mild, moderate or severe), and the following time-invariant covariates: age of both patient and caregiver at baseline, patient's gender, patient's and caregiver's educational levels (low, average or high), type of dementia (Alzheimer's dementia (AD), frontotemporal dementia (FTD), mixed or vascular dementia (mixed/vasc) and other type of dementia), relationship of the caregiver to the patient (spouse, child or other) and the duration of the disease at time of inclusion. Sensitivity analyses were performed to explore whether any effects of the key time-varying covariates applied to all four neuropsychiatric sub-syndromes (hyperactivity, mood, psychosis and apathy). Because a relatively large proportion of patients were institutionalized during our study, a sensitivity analysis was performed to determine whether the effects remained after this group was removed from the analysis. All tests were two-tailed unless otherwise specified. For all analyses, $p < 0.05$ was used as the threshold for statistical significance.

Results

Dropouts and missing data

From the original sample, 209 (out of 215) patient-caregiver dyads were included in the study. Six caregivers refused to participate due to high levels of subjective burden. For the baseline assessment, self-reports of 57 (out of 209) patients regarding their own needs were unavailable because these patients were unable to complete the CANE interview and were therefore not included in the analyses. Non-completers had more severe dementia (U = 5,529, $p < 0.001$) and a higher score on the apathy sub-syndrome (U = 5,088.5, $p = 0.21$).

Fifty-four dyads (25.8%) were lost to the 2-year follow-up assessment: 16 (7.7%) were lost due to the death of the patient, and 38 (18%) dyads discontinued participation mainly due to high levels of burden or lack of time as a result of the caregiving situation. There were no significant differences between those who dropped out the study and those who did not regarding age, gender, education, the number of (un)met needs or the level of neuropsychiatric symptoms. However, there was a difference between the groups regarding the severity of dementia; patients who dropped out had more severe dementia compared to patients who completed all assessments (U = 3,969, $p < 0.001$).

Patient and caregiver characteristics

More than half of the patients were male, and most patients had moderate disease severity (Table 1). In our group, Alzheimer's disease (AD) was the most common cause of dementia, followed by frontotemporal dementia and vascular dementia. Neuropsychiatric symptoms were highly prevalent in our sample, with 90.4% of the patients having at least one clinically relevant behavioral symptom. The patients had the highest severity scores for the hyperactivity and apathy sub-syndromes.

Table 1. Baseline characteristics of patients and caregivers

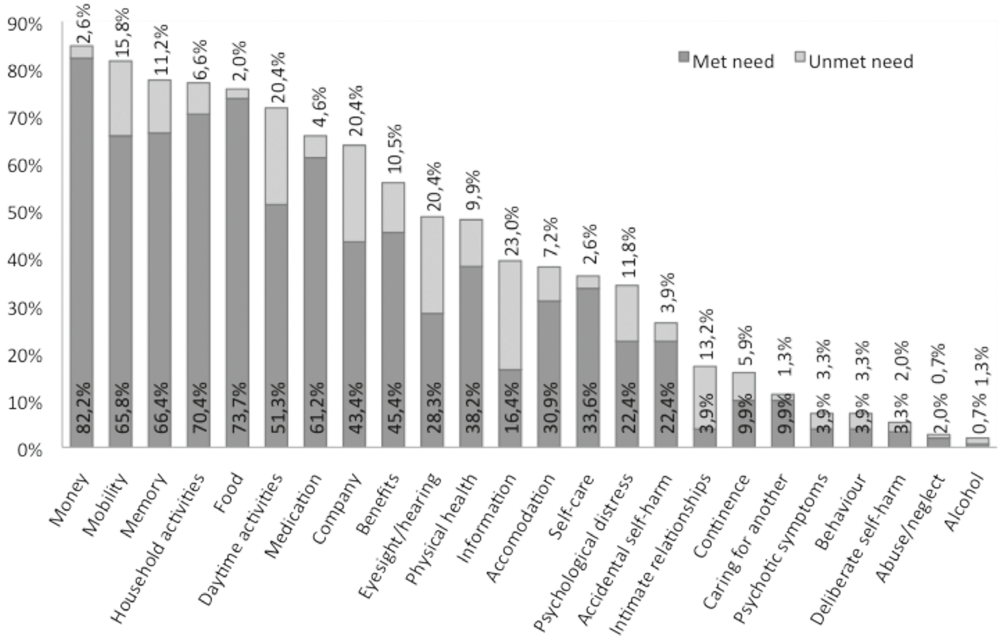
Patient gender	Male (%)	114	54.5%
Patient age at inclusion	Mean (SD)	61.1	5.5
	Range		[43-74]
Patient education			
Low		90	43.1%
Average		72	34.4%
High		17	22.5%
Dementia type			
Alzheimer's dementia (AD)		119	56.9%
Frontotemporal dementia (FTD)			
	Behavioral variant	28	13.4%
	Primary progressive aphasia (PPA)	13	6.2%
Vascular dementia (VD)		24	11.5%
Mixed dementia		8	3.8%
Lewy body dementia		7	3.3%
Dementia due to another cause		10	4.8%
Disease severity (GDS)			
Mild (2, 3)		34	16.3%
Moderate (4, 5)		130	62.2%
Severe (6, 7)		35	16.7%
Neuropsychiatric symptoms	(NPI) Mean (SD)	26.0	19.1
Hyperactivity sub-syndrome	Mean (SD)	10.7	10.8
Affective sub-syndrome	Mean (SD)	3.9	5.0
Psychosis sub-syndrome	Mean (SD)	2.8	5.1
Apathy sub-syndrome	Mean (SD)	8.1	7.2
Disease duration	Mean (SD) in months	90	46.4
Patients institutionalized during the study		71	34.0%
Caregiver gender	Female (%)	125	59.8%
Caregiver age at inclusion	Mean (SD)	58.4	9.3
	Range		[20-78]
Caregiver education			
Low		64	30.6%
Average		89	42.6%
High		56	26.8%
Caregiver relationship			
Spouse		191	91.4%
Child		13	6.2%
Other		5	2.4%

Notes: GDS = Global Deterioration Scale; NPI = Neuropsychiatric Inventory.

Patients' self-reports of met and unmet needs at baseline

At baseline, patients reported a mean of 7.9 (SD 3.3) areas with met needs and a mean of 2.0 (SD 2.7) areas in which an unmet need existed. The majority of patients reported that they needed or received some kind of formal or informal support in managing their finances, mobility (e.g., walking, climbing stairs and travelling), memory (e.g., remembering appointments and coping with memory loss), or household activities and food (e.g., shopping and preparing meals) (Figure 1). The highest proportion of unmet needs were in the areas of information, eyesight and hearing, company, daytime activities, mobility, intimate relationships and psychological distress.

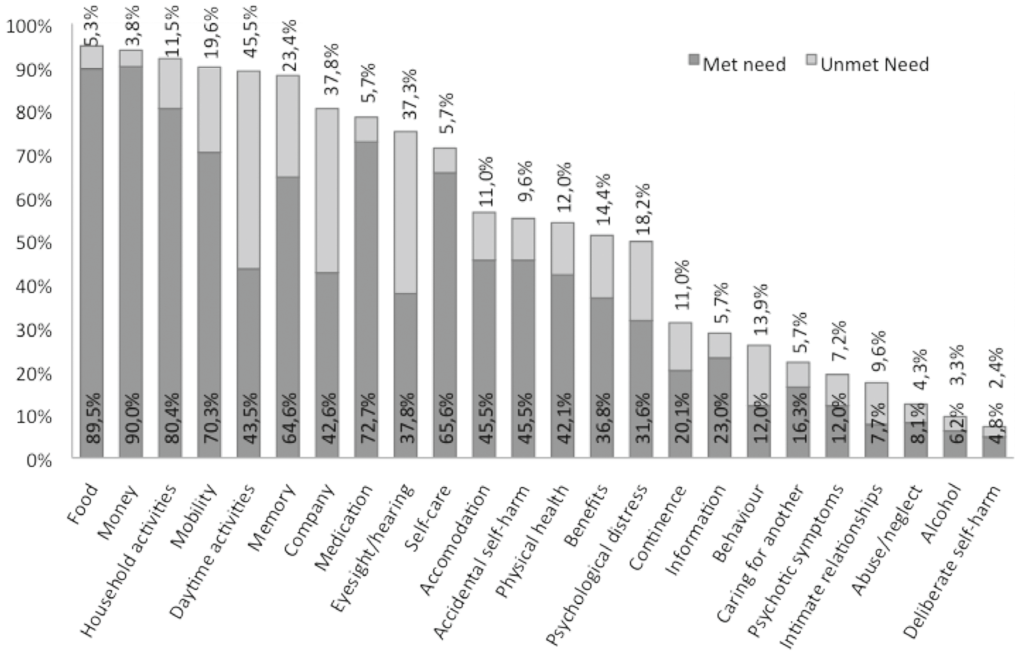
Figure 1. Patients' self-reports of met and unmet needs by percentages (N=152)



Caregivers' reports of met and unmet needs of patients at baseline

On average, patients had 9.6 (SD 2.9) areas in which a need existed that could be met and 3.1 (SD 2.4) areas in which an unmet need remained. The majority of caregivers reported that the patient needed or received (in)formal support with food, money, household activities, mobility, daytime activities, memory, company, medication, eyesight and hearing and self-care (Figure 2). The proportion of unmet needs varied between 2.4% for the domain of deliberate self-harm and 45.5% for daytime activities. The highest proportions of unmet needs included daytime activities, the company of others, eyesight and hearing (i.e., difficulties in communicating, reading and writing), memory, mobility and psychological distress.

Figure 2. Caregivers' proxy reports on met and unmet needs by percentages (N=209)



Agreement between patients and caregivers on care needs at baseline

There was large variation in the degree of agreement (κ) between the patients' self-reports and the proxy of the caregivers, with agreement varying between 0.06 (poor agreement) and 0.51 (moderate agreement) (Table 2.). The highest agreement was found in the areas of medication, continence and caring for another (e.g., caring for a child). Poor agreement was found in the areas of food, abuse/neglect, information, accidental self-harm, intimate relationships, memory and deliberate self-harm.

Table 2. Agreement between patients and caregivers on care needs

	κ coefficient	n
Medication	0.51	150
Continence	0.46	150
Caring for another	0.41	152
Physical health	0.39	151
Accommodation	0.37	152
Household activities	0.34	152
Self-care	0.33	152
Psychotic symptoms	0.32	152
Money	0.31	152
Company	0.29	150
Psychological distress	0.28	148
Mobility	0.27	151
Benefits	0.27	126
Alcohol	0.25	151
Behavior	0.23	150
Eyesight and hearing	0.23	151
Daytime activities	0.22	149
Food	0.20	151
Abuse or neglect	0.20	150
Information	0.18	141
Accidental self-harm	0.17	152
Intimate relationships	0.14	147
Memory	0.11	146
Deliberate self-harm	0.06	151

Note: Cases with an unknown need in a specific area were not included in the analyses to obtain equal cross tables; therefore, the n varies for the different κ values.

The two-year course of needs

According to the caregiver report, the total number of areas in which a need existed did not change significantly during the two-year study period H (4) = 4.21, p = .378. However, according to the patient report, there was a significant decrease in the number of areas in which a need occurred over time H (4) = 17.25, p = .002). Both caregivers and patients reported that the percentage of unmet needs decreased from approximately 24% (caregiver) and 18% (patient) at baseline to 14% and 10%, respectively, at 2 years follow up, with an obvious increase in the percentage of needs that were met.

The relationship between (un)met needs and neuropsychiatric symptoms over time

A linear mixed model analysis showed that the time-varying number of met needs and the number of unmet needs covaried with the level of total neuropsychiatric symptoms over time (table 3). Patients with higher levels of met or unmet needs over time exhibited higher levels of neuropsychiatric symptoms over time, with the largest effect for unmet needs. Sensitivity analyses (not shown) for the four separate behavioral sub-syndromes (hyperactivity, psychosis, mood and apathy) revealed that the effect for unmet needs remained significant. However, the effect for met needs remained significant for all sub-syndromes except for the hyperactivity cluster, for which there was only a trend (t=1.837; df=624,99, p=.067). When the patients who where institutionalized during the study were removed from the analysis, the effects for met and unmet needs on neuropsychiatric symptoms remained significant (not shown).

FTD patients showed significantly higher levels of neuropsychiatric symptoms over time compared with AD patients. A similar effect was found for the ‘other causes’ group in our sample. Patients exhibiting severe dementia showed higher levels of neuropsychiatric symptoms over time compared with patients with mild dementia. Furthermore, patients with higher education showed higher levels of neuropsychiatric symptoms over time. However, although this effect was significant for a moderate level of education, the effect was only near significant for a high level of education.

Table 3. Linear mixed model of the relationship between changes in unmet needs and neuropsychiatric symptoms adjusted for covariates

	Estimate	SE	df	P	95% CI	
					Lo	Up
Intercept	-11.0	14.23	204.10	0.44	-39.10	17.10
Key predictors:						
Number of met needs	1.41	0.24	813.85	<0.001	0.94	1.88
Number of unmet needs	2.98	0.32	815.60	<0.001	2.36	3.60
Covariates:						
Patient’s age	-0.06	0.27	183.94	0.837	0.59	0.48
Male gender patient	4.01	2.33	188.32	0.087	-0.58	8.61
Patient’s level of education						
Low	0a					
Moderate	5.34	2.22	171.62	0.017	0.96	9.72
High	5.01	2.67	170.46	0.062	-0.26	10.27
Diagnosis						
Alzheimer’s disease	0a					
Vascular/mixed dementia	4.56	2.77	172.86	0.102	-0.91	10.03
Frontotemporal dementia	9.35	2.55	182.44	<0.001	4.31	14.39
Other causes	7.10	3.55	175.57.	0.047	0.09	14.11
Disease severity						
Mild	0a					
Moderate	-1.92	2.08	813.15	0.358	-6.01	2.17
Severe	7.10	2.54	793.81	0.005	2.11	12.07
Caregiver’s age	0.20	0.22	186.31	0.900	0.37	-0.24
Caregiver education						
Low	0a					
Moderate	0.23	2.32	171.12	0.921	-4.35	4.81
High	-4.35	2.77	170.24	0.119	-9.82	1.12
Caregiver relationship						
Spouse	0a					
Child	0.89	7.38	187.27	0.904	-13.67	15.45
Other	-10.77	6.14	188.33	0.081	-22.88	1.34
Disease duration	0.03	0.02	189.59	0.231	-0.02	0.07
Assessment						
Baseline	0a					
Six months	-1.69	1.36	660.87	0.216	-4.36	0.99
Twelve months	-2.81	1.39	670.77	0.043	-5.54	-0.09
Eighteen months	-2.83	1.48	701.01	0.055	-5.73	0.66
Twenty-four months	-1.15	1.54	715.66	0.456	-4.17	1.87

[a] Reference category

Discussion

This is the first prospective study to explore the course of care needs and the relationship between changes in these needs and changes in the severity of neuropsychiatric symptoms in people with YOD. Patients and caregivers reported similar areas in which they perceived needs. However, only modest agreement existed within patient-caregiver dyads regarding whether needs could be met. This study also reveals that YOD patients experience high levels of unmet needs in important areas of life, including daytime activities, social company, intimate relationships, eyesight and hearing/communication, memory, mobility, psychological distress and unmet information needs. Contrary to our expectations, the number of unmet needs decreased over time. Furthermore, as we hypothesized, we found a strong relationship between unmet needs and the level of neuropsychiatric symptoms over time. We also found a similar relationship between neuropsychiatric symptoms and the amount of met needs.

There was fair agreement within patient-caregiver dyads on the areas in which a need occurred. Nonetheless, patients reported fewer (un)met needs than did their caregivers. Patients also reported a significant decrease in the number of areas in which a need existed over time, but their caregivers did not. Non-completers of the needs interview in our study had more severe dementia, suggesting that as the disease progresses, patients find it more difficult to reflect on their needs and how these needs can be met. This is in line with earlier research on the subjective needs of LOD patients.[14, 32, 33] The agreement within patient-caregiver dyads in our sample on whether or not needs could be met was generally modest, which is consistent with earlier research in LOD.[14, 34] We found the highest agreement in relatively unambiguous areas, such as medication use, incontinence or caring for another person, and the lowest agreement in areas in which patients' judgment might rely on patients' disease awareness, such as abuse/neglect, accidental or deliberate self-harm, food and memory. There was little agreement on information needs and needs regarding intimate relationships, as caregivers contrary to the patient reported no unmet needs existed in these areas. The difference between patients' and caregivers' ratings of (un)met needs might not be unproblematic because caregivers base care decisions partially on their assessment and prioritization of the patient's needs,[35] increasing the risk that care needs remain unmet.

Patients and caregivers reported high proportions (18% and 24%, respectively) of unmet needs of patients at baseline. Both patients and caregivers reported that the proportion of unmet needs diminished with time by 8% and 10%, respectively. This result may indicate that during the course of the disease, appropriate support could be initiated. This underlines the importance of early introduction of professional services in the caregiving trajectory in YOD, especially given the known delay in the initiation of professional help and support in these young patients.[12] Caregivers reported the highest proportions of unmet needs in the areas of social company, daytime activities, eyesight and hearing/communication, memory, mobility and psychological distress. Patients additionally mentioned the areas of information and intimate relationships among the areas with the highest proportion of unmet needs. Van de Roest et al. conducted a study that explored the needs of community-dwelling older people with dementia and found similar results.[14] However, in their study, unmet mobility needs and unmet needs regarding intimate relationships were not among the areas with the highest proportions of unmet needs. Furthermore, overall proportions of unmet needs for the various areas of life in our study appear much higher. These differences may be due to study differences despite the use of the same assessment instrument, but it may also indicate age-related differences in the perception and weighting of unmet needs. For instance, losing the ability to work, perform social roles or drive/travel may have a more profound impact on younger patients.

We found that patients who reported more unmet needs during the course of our study also showed significantly higher levels of neuropsychiatric symptoms over time. This finding indicates that an increase in the number of unmet needs leads to the occurrence or worsening of neuropsychiatric symptoms, which is in line with the NDB model. In contrast, it is possible that high levels of neuropsychiatric symptoms hamper care provision in these patients, decreasing the likelihood that care needs will be fulfilled. Sensitivity analyses revealed that the effect of unmet needs applied to all four behavioral sub-syndromes. This finding is consistent with previous research on LOD patients, which found that unmet needs were associated with neuropsychiatric symptoms, such as anxiety and depression,[36] agitation[37, 38] and resistiveness to care.[39] Contrary to our expectations, we also found an effect of met needs on the occurrence of neuropsychiatric symptoms over time. This finding suggests that the relationship between (un)met needs and neuropsychiatric symptoms is indeed bi-directional in nature. It may also indicate that appropriate help and support with care needs do not necessarily counter the overall adverse effect of dementia on patients with YOD. As in YOD patients, the loss of functional abilities, increasing dependency on others and the consequential loss of various roles within the family and society lead to an important loss of sense of self, self-esteem and identity.[5] This may be especially true for patients with active careers and many social responsibilities because of the relationship we identified between higher levels of education and higher levels of neuropsychiatric symptoms over time. However, our findings indicate that it is worthwhile to target unmet needs to reduce neuropsychiatric symptoms and the related caregiver distress in YOD. Some of the unmet needs identified in our study, such as unmet needs related to social interaction, daytime activities, information about the disease and psychological distress, may be amendable to intervention.[15] Our previous research on awareness in YOD suggests that young-onset AD patients are more aware of the disease and related issues compared to late onset AD patients. Thus, these patients may be open to or may benefit from day care, volunteers' help with daytime activities or psychological counseling.[40]

As expected, patients with a diagnosis of frontotemporal dementia as well as patients with more severe dementia showed significantly higher levels of neuropsychiatric symptoms over time. Thus, the caregivers of these patients may be particularly at risk of being adversely affected because neuropsychiatric symptoms are known to adversely affect the quality of marital relationships[41] and to result in high levels of caregiver distress.[9, 42]

Several limitations of this study should be discussed. First, our findings may not be valid for the entire YOD population because we did not include patients with alcohol-related dementia, HIV/AIDS-related dementia complex, Huntington's disease, traumatic brain injury or Down's syndrome. We enrolled patients and caregivers who sought care and provided consent for participation. Patient-caregiver dyads we subsequently missed may have differed in terms of care needs, neuropsychiatric symptoms or dementia severity. Additionally, a large proportion of patients were unable to complete the needs assessment. We found that this inability was related to more severe dementia. Given the explorative nature of our study and our interest in studying the needs of all patients (including those with severe dementia), we used the caregiver proxy for the analyses. Consequently, our findings may have been biased because of the known differences between patient and caregiver perspectives. A strength of our study is that we were able to explore patients' care needs and their relationship with neuropsychiatric symptoms in a unique and relatively large sample of YOD patients and caregivers despite low prevalence rates and difficulties in diagnosing YOD.

This study provides important insights into a relatively unexplored area of YOD. Our findings suggest that health care professionals should be aware that in YOD there are specific areas of life in which unmet needs are more likely to occur. The high proportion of unmet needs in some areas, such as daytime activities, social company and information, in these younger patients should raise some concerns. Our study also suggests an important bi-directional relationship between (un)met needs and the course of neuropsychiatric symptoms, stressing the need for multi-component interventions that target neuropsychiatric symptoms directly and provide anticipatory care as well to prevent unmet needs in YOD. This warrants the periodic assessment of care needs in which patient and caregiver views on patients' needs are considered complementary.

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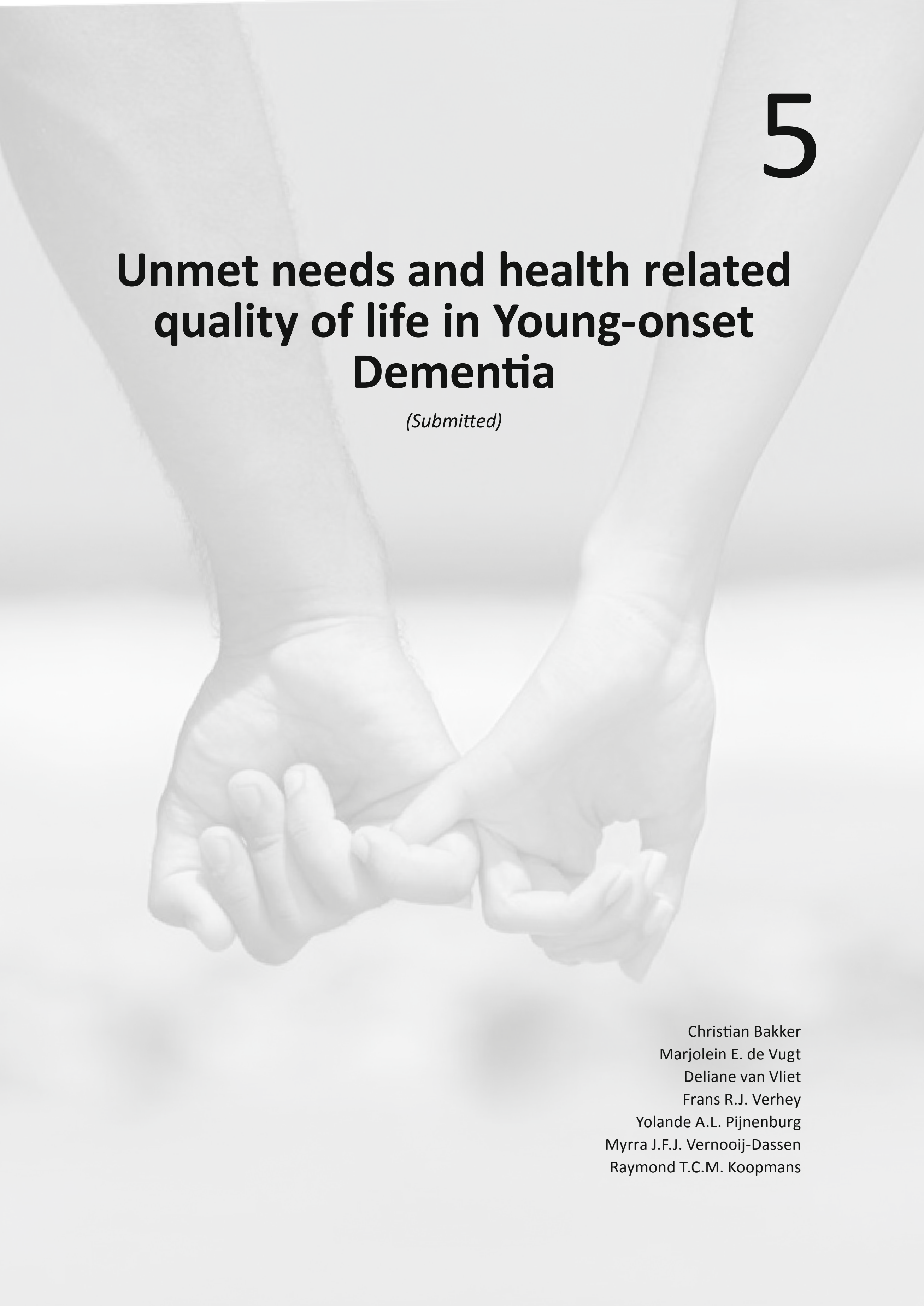

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Unmet needs and health related quality of life in Young-onset Dementia

(Submitted)

Christian Bakker
Marjolein E. de Vugt
Deliane van Vliet
Frans R.J. Verhey
Yolande A.L. Pijnenburg
Myrra J.F.J. Vernooij-Dassen
Raymond T.C.M. Koopmans

Abstract

Objective: Young-onset dementia (YOD) causes specific challenges and issues that are likely to affect health-related quality of life (HRQoL). This study explores patient and caregiver HRQoL and its association with unmet needs in YOD.

Design/Setting: A cross-sectional design was used.

Participants: 215 community dwelling YOD patients and their primary caregivers.

Measurements: Multiple linear regression analyses were performed to determine the relationship between unmet needs assessed with the Camberwell Assessment of Need in the Elderly (CANE) and patient and caregiver HRQoL, controlling for other variables such as demographic characteristics, patient functional status, neuropsychiatric symptoms and caregiver sense of competence.

Results: Patient HRQoL was not associated with unmet needs. However, we found that the unmet needs of both patient and caregiver related to several domains of caregiver HRQoL.

Conclusions: This study shows that patient and caregiver unmet needs are related to caregiver HRQoL in YOD. However, the relationship between HRQoL and unmet needs is complex. The assessment of unmet needs within the context of HRQoL appears to be an important prerequisite for personalizing care in YOD. Adjusting supportive services to match the individual needs and preferences of these young patients and their caregivers is likely to enhance their quality of life.

Introduction

Health-related quality of life (HRQoL) is an increasingly important outcome measure in dementia research. In young-onset dementia (YOD), defined as dementia occurring before the age of 65, specific issues and challenges may add to the negative impact of dementia on the HRQoL of both patients and caregivers.

Younger patients are in a more active life phase, in which cognitive and functional impairments and changes in personality may adversely affect HRQoL through the loss of social roles, responsibilities and identities.(1) In dementia patients over the age of 65, younger age has been associated with worse HRQoL.(2) Also, behavioral problems, depression, insight into the disease and impaired cognition have been associated with lower HRQoL.(3, 4) It is unclear whether the aforementioned associations exist within the YOD population. Obtaining a proper diagnosis is essential for patients and caregivers to understand and adjust to the changes that occur and to seek appropriate support. However, in YOD the diagnosis is often substantially delayed,(5) delaying the initiation of appropriate support and health care services. Research suggests that unmet needs lead to more behavioral problems and increased caregiver burden, both of which adversely affect HRQoL(6, 7) and increase the risk for institutionalization.(8, 9) Unmet needs may also directly be negatively associated with HRQoL in YOD because it may be especially difficult to meet the needs of YOD patients and their families.(10) An unmet need can be defined as a need in a particular area of life of the individual for which insufficient or inadequate support is provided or appropriate support is unavailable.(11) A YOD patient might for example experience a need for meaningful daytime activities while attending a day care centre that arranges for activities that are mainly focused on the elderly. The lack of meaningful activities could consequently lead to a loss of HRQoL. It is necessary to better understand this relationship as HRQoL may be improved by addressing the unmet needs of YOD patients and their families.

Caregivers of younger patients experience high levels of caregiver burden(12) that may negatively impact their own HRQoL. Work-related and financial issues, conflicting roles, severe behavioral problems, and inadequate health care services may add to the burden and negatively influence caregiver HRQoL.(5) Also, the patient's level of cognitive impairment, the time committed to care, lack of respite services, the patients' younger age, and the presence of behavioral problems are related to decreased caregiver HRQoL, at least in late onset dementia (LOD).(13, 14) We found one small study in YOD-AD caregivers that addressed caregiver HRQoL in YOD. This study found that increased caregiver age and greater patient insight were significantly associated with better HRQoL in caregivers.(15)

In this study, we explored (1) HRQoL in YOD patients and possible differences with caregiver perspectives on patient HRQoL, (2) HRQoL in YOD caregivers compared to the general population, and (3) the relationship between unmet needs and both patient and caregiver HRQoL in YOD. We hypothesized that the number of unmet needs in YOD is related to both patient and caregiver HRQoL.

Methods

Study design and selection of participants

This cross-sectional study is part of a larger longitudinal study, the Needs in Young-onset Dementia (NeedYD) study. The design of the NeedYD study has been described previously.(16) Baseline data from 215 patient-caregiver dyads were used. All patients were living at home at time of inclusion. Patients were consecutively referred from university medical centers (n=56), regional hospitals (n=10), community mental health services (n=20), self-referred (n=14), or recruited through YOD-specialized day care facilities (n=115). People with symptom onset before age 65 were included in the study. Therefore, age at inclusion could be over 65. Dementia diagnosis was made by the clinician on the basis of clinical, neuropsychological and brain imaging findings according to the criteria from the 4th edition of the Diagnostic and Statistical Manual of Mental Disorders, text revision (2000) (17), and the Dutch consensus guidelines (18) that uses internationally accepted criteria for diagnosing dementia subtypes (19-23). Clinical diagnoses were checked by the researcher against clinical patient files for each patient. Exclusion criteria were (1) lack of informed consent of the participant, (2) living in a nursing home or (3) dementia caused by HIV, traumatic brain injury, Down's syndrome, Huntington's disease or alcohol related dementia.

The Medical Ethics Committee of the Maastricht University Medical Center and the local ethics committees of the participating institutions approved the study protocol. The research project was performed according to the principles of the Declaration of Helsinki (version January 2004; www.wma.net) and is in agreement with the law regarding medical-scientific research in humans (WMO).

Data collection and assessments

Written informed consent from patients or their legal representatives was obtained prior to the study. Patients who were not able to sign informed consent were asked to give oral consent and their legal representative had to give written consent that the patient was able to participate. Trained researchers and research assistants used structured interviews and questionnaires to collect the data. A one-day training session and regular consensus meetings safeguarded the integrity of the data collection.

Assessment of health-related quality of life

For the assessment of patient HRQoL, the *Quality of Life-Alzheimer's Disease Scale* (QOL-AD) was used. The QOL-AD is a dementia-specific self-report scale covering 13 domains of HRQoL: physical health, energy, mood, living situation, memory, family, marriage, friends, chores, fun, money, self, and life as a whole. These domains are scored on a four-point Likert scale.(24) The QOL-AD can be used as a patient self-reporting instrument or as a caregiver report of the patient's HRQoL. Summing the domain scores results in total scores ranging from 13 to 52, with a higher score representing a higher level of HRQoL. Participant self-reported HRQoL and the caregiver's report of the participant's HRQoL were obtained. The QOL-AD is applicable to mild-moderate dementia stages and has good inter-observer reliability, content and criterion-concurrent validity;(25) there is also evidence for its reliability in severe dementia.(25, 26)

HRQoL in the caregiver was assessed with the Dutch version of the *RAND-36*.(27) The RAND-36 consists of 36 questions that evaluate HRQoL scores in five mental and three physical domains: role limitations caused by emotional problems, social functioning, emotional well-being, vitality, general health perception, physical functioning, and role limitations caused by physical health problems and pain. These domains can be expressed in two summary scores (range 0–100), reflecting the mental (MCS) and physical (PCS) component scores. The Dutch version of the RAND-36 is a reliable, valid and sensitive measure for general health.(28)

Assessment of (un)met needs

(Un)met needs were assessed with the Dutch version of the *Camberwell Assessment of Need for the Elderly* (CANE).(29) The CANE is a semi-structured interview consisting of 24 domains that cover social, physical, psychological, and environmental needs of the patient. The answers are used to determine whether or not the patient experiences a need and whether or not this need is met. Also, psychological needs of the caregiver were inventoried, such as the need for psychosocial support, psychoeducation, or time off from the care giving situation. The needs of patients and caregivers were assessed by interviewing the caregivers. Reliability and validity were found to be adequate.(29)

Patient and caregiver factors

The severity of dementia was assessed with the *Global Deterioration Scale* (GDS), which is a seven-point scale (1-7) ranging from ‘no impairment’ (1) to ‘very severe cognitive impairment’ (7). The GDS is a widely used instrument that has been validated against behavioral, neuro-anatomic and neurophysiologic measures, where significant correlations have been found.(30) Neuropsychiatric symptoms were assessed with the Dutch version of the *Neuropsychiatric Inventory* (NPI).(31) The frequency (F) and severity (S) of each symptom are rated on a five- (0-4) and four-point (0-3) Likert scale, respectively. These scores are summed to calculate a total score ranging from 0-144. The NPI has high inter-observer reliability and is a valid rating scale for assessing a wide range of neuropsychiatric symptoms of dementia.(32) The *Cornell Scale for Depression in Dementia* (CSDD) was administered to the caregiver to identify depressive symptoms in the patient. The CSDD has adequate inter-observer reliability, internal consistency and sensitivity.(33) The amount of informal care used by the patient and the use of supportive services by the caregiver were assessed with the *Resource Utilization in Dementia-Lite* (RUD-Lite) questionnaire; this assessment covers 95% of the resource use covered by the complete RUD.(34) The *Short Sense of Competence Questionnaire* (SSCQ) was administered to assess caregiver capabilities in dealing with the burden of caring for a dementia patient. Three domains are evaluated: (1) satisfaction with the demented person as a care recipient; (2) satisfaction with one’s own performance as a caregiver; and (3) consequences of involvement in care for the personal life of the caregiver. The SSCQ consists of seven items rated on a 5-point Likert scale ranging from 1 (agree very strongly) to 5 (disagree very strongly). The SSCQ is reported to have satisfactory reliability and validity.(35) Additionally, the duration of the care giving process and demographic characteristics of both patients and caregivers were assessed using a structured interview.

Statistical analysis

Analyses were performed using the Statistical Package for Social Sciences (SPSS) version 18.0.2 (SPSS Inc., Chicago, USA). Proportions or means were calculated to describe the characteristics of patients and caregivers. Group comparisons were performed (to determine whether the characteristics of patients and caregivers whose data were incomplete differed from the rest of the sample) using the chi-squared test for categorical variables and the t-test for continuous variables.
Paired-sample t-tests were performed to compare the patient-reported QOL-AD scores with the caregiver proxy score for the different types of dementia. Domain and summary scores of the RAND-36 were calculated and compared to the norms from the general Dutch population, with scores derived from the original reference population of the Dutch version of the RAND-36, using a one-sample t-test. (36) The reference population for the RAND-36 was aged between 18 and 89 years (mean 44.1 years). Multiple regression analyses were used to study the association between patient HRQoL and unmet needs. Thus, the QOL-AD patient self-report score was analyzed as the dependent variable, with the number of unmet needs as correlate and patient age, sex, disease severity (GDS), depressive symptoms (CSDD, total score) and behavioral problems (NPI Frequency x Severity total score) as covariates. Additional multiple regression analyses were used to explore the association between caregiver HRQoL and unmet needs. In the analysis the RAND-36 domains (role limitations caused by emotional problems, social functioning, emotional well-being, vitality, general health perception, physical functioning, and role limitations caused by physical health problems and pain) were analyzed as dependent variables, with the number of unmet needs of the patient and unmet needs of the caregiver as correlates of HRQoL. Caregiver age, sex, sense of competence (SSCQ), the amount of informal care provided by the caregiver (RUD-lite), the duration of the care giving process and the use of supportive services by the caregiver (RUD-lite) were entered into the analyses as covariates. All tests were performed two-tailed and the significance level was set at <alpha> = .05.

Results

Patient and caregiver characteristics

Two hundred and nine patient-caregiver dyads were included from the original sample (N=215). Six caregivers refused to participate because of high levels of subjective burden, and four caregivers did not fill out the RAND-36 assessment. These caregivers did not differ from the group that was included. One hundred and sixty-one patients (78.5%) completed the QOL-AD questionnaire. Non-completers were more likely to be female (Chi-squared test, χ^2=4.168, df=1, p<0.05) and had a higher disease severity (Mann-Whitney U test, Z = 7.033, p < 0.001). Most of the caregivers (96.8%) completed the QOL-AD proxy report.

Mean patient age was 61.1 years (SD ± 5.4), and the male to female ratio was approximately equal (Table 1). The majority (62%) had a moderate disease severity, and AD was the most frequent cause (57.1%) of dementia. Caregivers reported that patients experienced different amounts of unmet needs on the CANE, ranging from zero to 13 domains in which a need existed. Almost half of the patients (45.5%) experienced an unmet need regarding daytime activities. Also a large proportion of patients reported they lacked companionship (37.8%), experienced difficulties in performing tasks that depend on eyesight or hearing (37.3), or depend on memory (23%).

Caregiver mean age was 58.4 years (SD ± 9.3), and the majority were patient spouses. Forty-three percent of caregivers reported that they had unmet psychological needs.

Table 1. Baseline demographic characteristics of patients and caregivers in the study (N=205)

Sex (Patient)	Male (%)	113	(55.1%)
Patient age	Mean (SD)	61.1	(5.4)
	Range		[43 – 74]
Dementia type:			
Alzheimer's Disease (AD)		117	(57.1%)
Frontotemporal dementia (FTD)			
Behavioural variant		27	(13.2%)
Primary progressive aphasia (PPA)		13	(6.3%)
Vascular dementia (VD)		24	(11.7%)
Mixed dementia		8	(3.9%)
Dementia due to another cause		16	(7.8%)
Disease severity (GDS):			
Mild (2, 3)		34	(16.6%)
Moderate (4, 5)		127	(62.0%)
Severe (6, 7)		34	(16.6%)
Patients with unmet needs (CANE):		183	(87.6%)
	Mean (SD)	3.10	2.45
	Range		[0-13]
Sex (Caregiver)	Female (%)	124	(60.5%)
Caregiver age	Mean (SD)	58.4	(9.3)
	Range		[20 – 78]
Relationship of caregiver to the patient:			
Spouse		187	(91.2%)
Child		13	(6.3%)
Other		5	(2.5%)
Caregivers with unmet needs (CANE):		89	(43.4%)

Notes: GDS = Global Deterioration Scale, 4.9% of patient scores were not available; CANE indicates the Camberwell Assessment of Need in the Elderly; the mean for the patients represents the amount of domains of the CANE in which a need exists that cannot be met.

Health-related quality of life

Patient HRQoL

The mean patient self-reported score on the QOL-AD was 36.2 (SD 5.7), significantly higher than the QOL-AD proxy score as assessed by the caregivers, which was 29.1 (SD 5.6) (Table 2.). This difference remained statistically significant for all diagnostic subgroups , with the largest absolute difference in the group with frontotemporal lobar degeneration (FTLD), in which patients reported the highest level of HRQoL.

Table 2. Paired sample t-test of QOL-AD patient self-report compared with caregiver proxy report

	Self-report Mean (SD)	Proxy report Mean (SD)	*p*	*df*
Patient				
QOL-AD score entire group	36.2 (5.7)	29.1 (5.6)	<0.001	151
AD	36.8 (5.4)	29.8 (5.7)	<0.001	87
Vascular/mixed	33.9 (4.8)	28.1 (4.8)	0.001	22
FTD	37.9 (6.5)	28.3 (5.1)	<0.001	27
Other	33.7 (5.3)	27.1 (6.8)	0.038	12

Note: QOL-AD indicates Quality of Life-Alzheimer Disease Scale; AD, Alzheimer's disease; FTD, frontotemporal dementia. Only cases in which both the patient and caregiver report were available were entered in the analyses.

Although the multiple linear regression model was significant overall, only depression predicted patient HRQoL (Table 3.).

Table 3. Multiple linear regression model of unmet needs and patient HRQOL adjusted for covariates

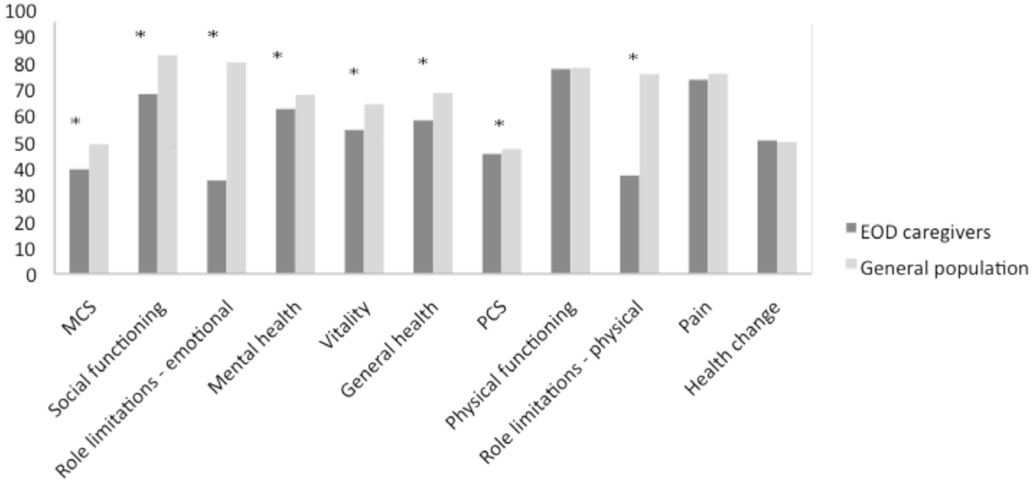
				95% CI	
	B	SE	p	Lower bound	Upper bound
Number of unmet needs (CANE)	-0.25	0.226	0.275	-0.694	0.199
Depression (CSDD-score)	-0.35	0.099	0.001*	-0.545	-0.153
Disease severity (GDS-score)	0.81	0.588	0.170	-0.351	1.974
Behavioural problems (NPI FxE score)	0.03	0.032	0.364	-0.034	0.093
Patient age	-0.06	0.087	0.500	-0.231	0.113
Female gender	0.63	1.027	0.538	-1,397	2.667

Note: Model summary patient HRQOL: adjusted r^2 = 0.13; *df* = 7,137; *F* = 4.476; p ≤ 0.001. 95% CI indicates 95% confidence interval; CANE, Camberwell Assessment of Need in the Elderly; GDS, Global Deterioration Scale; CSDD, Cornell scale for depression in dementia; NPI, Neuropsychiatric Inventory.

Caregiver HRQoL

The caregivers' mean score (SD) on the mental component score (MCS) of the RAND-36 was 41.6 (SD 6.9) and was 47.7 (SD 7.3) on the physical component score (PCS). These scores were 10 and 2 points lower than the MCS and PCS of the general Dutch population, respectively (Figure 1). One-sample t-tests revealed that on the MCS, caregiver scores were significantly lower than the general population in terms of social functioning, vitality, role limitations due to emotional problems, and general health. Also on the PCS, caregiver scores were significantly lower for role limitations due to physical problems. The absolute difference was largest for role limitations that were due to emotional and physical problems.

Figure 1. HRQoL caregivers as measured by RAND-36 compared to the general population



Note: MCS = Mental Component total Score, PCS = Physical Component total Score; MCS and PCS score for the general population were derived from the original article on the RAND-36 psychometric properties [38]; *Group comparisons were done using one-sample t-tests, *df* = 204 (for both significant and non-significant results), except for role limitations physical (*df* = 203) and general health (*df* = 201), p < 0.001.

Caregivers' HRQoL and unmet needs

Multiple linear regression analysis showed that caregivers taking care of patients with a higher number of unmet needs reported feeling less vital and had lower levels of general health (Table 4). Furthermore, we found that caregivers taking care of patients with a higher number of unmet needs reported more difficulties in performing daily tasks due to physical problems and higher levels of physical pain (Table 5).

Additionally, unmet needs among caregivers were correlated with increased limitations in performing social activities and higher levels of mental health. However, caregivers with an increased number of unmet needs experienced fewer limitations in performing work and other roles due to emotional problems.

Table 4. Association of unmet needs and RAND-36 Mental Component domains (Caregiver HRQoL) adjusted for covariates

	Soc. Functioning					Role limitations - emotional					Mental health				
				95% CI					95% CI					95% CI	
	B	SE	p^a	lo	up	B	SE	p^a	lo	up	B	SE	p^a	lo	up
Unmet needs, caregiver	-8.29	3.89	0.034*	-15.96	-0.62	17.35	6.01	0.004*	5.49	29.21	-7.26	2.70	0.008*	-12.95	-1.93
Unmet needs, patient	-1.23	0.82	0.135	-2.84	0.38	0.42	1.26	0.739	-2.07	2.91	-0.73	0.57	0.201	-1.85	0.39
Age of caregiver	-0.05	0.21	0.804	-0.47	0.36	-0.38	0.33	0.242	-1.02	0.26	-0.09	0.15	0.560	-0.37	0.20
Sex of caregiver	-4.97	3.98	0.213	-12.82	2.88	10.66	6.15	0.085	-1.48	22.80	-8.05	2.77	0.004*	-13.51	-2.59
Duration of care	0.41	0.44	0.360	-0.47	1.28	-1.00	0.68	0.147	-2.35	0.35	0.55	0.31	0.076	-0.06	1.16
Sense of competence	0.40	0.37	0.280	-0.33	1.12	-0.64	0.57	0.262	-1.76	0.48	0.54	0.26	0.037*	0.03	1.04
Supportive services	-3.94	1.14	0.001*	-6.19	-1.69	8.42	1.76	<0.001*	4.94	11.90	-2.41	0.79	0.003*	-3.97	-0.84
Amount of informal care	-0.01	0.01	0.320	-0.02	0.01	0.02	0.01	0.154	-0.01	0.04	-0.01	0.01	0.322	-0.02	0.01

Notes: Model summary social functioning: adjusted $R^2 = 0.14$; df = 8,180; F = 4.891; $p \leq 0.001$.
Model summary role limitations: adjusted $R^2 = 0.22$; df = 8,180; F = 7.732; $p \leq 0.001$.
Model summary mental health: adjusted $R^2 = 0.21$; df = 8,180; F = 7.416; $p \leq 0.001$.
95% CI indicates 95% confidence interval;
*$p < 0.05$, ^{a}T-test

Table 4. (Continued) Association of unmet needs and RAND-36 Mental Component domains (Caregiver HRQoL) adjusted for covariates

	Vitality					General health				
				95% CI					95% CI	
	B	SE	p^a	lo	up	B	SE	p^c	lo	up
Unmet needs, caregiver	-4.70	2.84	0.100	-10.30	0.90	1.69	3.17	0.594	-4.56	7.94
Unmet needs, patient	-1.53	0.60	0.011*	-2.71	-0.35	-2.42	0.66	<0.001*	-3.72	-1.13
Age of caregiver	0.03	0.15	0.826	-0.27	0.34	-0.08	0.17	0.653	-0.41	0.26
Sex of caregiver	-4.27	2.90	0.144	-10.00	1.47	1.48	3.22	0.646	-4.88	7.84
Duration of care	0.25	0.32	0.434	-0.38	0.89	-0.33	0.36	0.352	-1.04	0.37
Sense of competence	0.19	0.27	0.471	-0.34	0.72	0.64	0.30	0.033*	0.05	1.24
Supportive services	-3.28	0.83	<0.001*	-4.93	-1.64	-3.41	0.92	<0.001*	-5.23	-1.60
Amount of informal care	-0.01	0.01	0.036*	-0.02	-0.001	-0.001	0.01	0.817	-0.1	0.1

Model summary vitality: adjusted $R^2 = 0.20$; df = 8,180; F = 6.926; $p \leq 0.001$.
Model summary general health: adjusted $R^2 = 0.17$; df = 8,178; F = 5.614; $p \leq 0.001$.
95% CI indicates 95% confidence interval;
*$p < 0.05$, ^{a}T-test, df = 188, ^{c}T-test, df =186

Table 5 Association of unmet needs and RAND-36 Physical Component domains (Caregiver HRQoL) adjusted for covariates

	Phys. functioning					Role limitations - physical					Pain				
				95% CI					95% CI					95% CI	
	B	SE	p[b]	lo	up	B	SE	p[b]	lo	up	B	SE	p[a]	lo	up
Unmet needs, caregiver	-1.36	3.37	0.687	-8.00	5.29	5.67	6.19	0.361	-6.54	17.89	-1.14	3.76	0.762	-8.56	6.27
Unmet needs, patient	-2.24	0.71	0.002*	-3.63	-0.85	1.18	1.30	0.365	-1.38	3.74	-1.83	0.79	0.022*	-3.39	-0.27
Age of caregiver	-0.45	0.18	0.014*	-0.81	-0.09	-0.01	0.33	0.974	-0.67	0.65	-0.26	0.20	0.207	-0.66	0.14
Sex of caregiver	-3.12	3.45	0.368	-9.93	3.69	8.87	6.33	0.163	-3.63	21.37	-3.09	3.85	0.423	-10.68	4.50
Duration of care	-0.60	0.38	0.119	-1.35	0.16	0.64	0.70	0.367	-0.75	2.03	-0.51	0.43	0.238	-1.35	0.34
Sense of competence	0.06	0.32	0.859	-0.57	0.68	-0.81	0.58	0.169	-1.96	0.35	0.01	0.36	0.973	-0.69	0.71
Supportive services	-2.48	0.99	0.013*	-4.42	-0.53	6.83	1.82	<0.001*	3.24	10.42	-3.10	1.10	0.005*	-5.28	-0.93
Amount of informal care	-0.01	0.01	0.267	-0.02	0.01	0.02	0.01	0.072	-0.002	0.05	-0.01	0.01	0.147	-0.03	0.004

Notes: Model summary physical functioning: adjusted $R^2 = 0.14$; df = 8,179; F = 4.823; $p \leq 0.001$.
Model summary role limitations: adjusted $R^2 = 0.16$; df = 8,179; F = 5.488; $p \leq 0.001$.
Model summary pain: adjusted $R^2 = 0.14$; df = 8,180; F = 3.678; $p \leq 0.001$.
95% CI indicates 95% confidence interval;
*$p < 0.05$, [a] T-test, df = 188, [b] T-test, df = 187

Discussion

To our knowledge, this is the first study that focuses on patient and caregiver HRQoL in YOD and the relationship of HRQoL with unmet needs. Patient HRQoL was not associated with unmet needs. However, patient HRQoL was associated with depression. Caregivers rated their own HRQoL lower than that of the general population. Unmet needs of both patients and caregivers were related to several domains of caregiver HRQoL.

Consistent with findings from other studies (37), patients in all diagnostic subgroups of our sample rated their HRQoL significantly higher than did their primary caregivers. We did not find an association between unmet needs and patient HRQoL, which is surprising because the domains in which our patients experienced unmet needs also refer to important domains in HRQoL, such as performing meaningful daytime activities or enjoying the company of others. However, there can be many different reasons why we did not find a relationship between unmet needs and HRQoL in our sample. For instance we investigated patients' unmet needs using the caregiver proxy report. The caregiver's perspective might not fully represent a patient's perspective on his or her needs,(38) explaining why we did not find a relationship with patient self-reported HRQoL. We did not use the patients' self-reports of unmet needs for the analysis, because such reports were not available for 30% of our patient sample. We found during the interviews that patients had difficulties in reflecting on different aspects of all 24 domains of the needs interview. They were unable to recall the help they actually received, and experienced difficulties in reflecting on the kind of help they wanted to receive. This is a known issue in the assessment of needs especially in patients with higher levels of cognitive impairment and higher levels of dependency. (11, 39) As in our sample, we included patients with more advanced disease stages, we chose to use the caregiver view to get a complete picture of all domains in which unmet needs might exist. As depression is related to unmet needs,(40) and high levels of depression were related to worse patient HRQoL in our sample, a suppressive effect of depression on unmet needs would also explain our findings. We indeed found a weak, but significant correlation between unmet needs and depression (r =0.490, p <0.001). We found no significant effect for age nor any of the other covariates on patient HRQoL.

Compared to the general Dutch population, caregivers reported significantly lower HRQoL scores on the different HRQoL sub-domains. The largest difference was observed in the area of experienced role limitations due to both emotional and physical problems. We believe this illustrates the double burden in YOD of caring for a young dementia patient while simultaneously performing other roles, such as parenting, working and running the household. We found that unmet needs of both patients and caregivers were negatively associated with several domains of caregiver HRQoL. Unmet needs of the patient were associated with a loss of vitality and perceived general health in the caregiver as well as higher levels of physical pain and difficulties in performing daily tasks due to physical problems. We believe that young caregivers find it difficult to accept unmet needs in the patient as they watch the patient struggle with the loss of cognitive and functional abilities, daily activities and social contact.

The loss of these abilities experienced by YOD patients and their changing roles in society have been shown to adversely affect patients' sense of self and identity,(1) thereby causing more stress in caregivers.(41)

In our study, caregivers with more unmet psychological needs, such as the need for psychosocial support or respite care, seem especially at risk for experiencing limitations in social functioning and feelings of depression and anxiety. This may relate to the delay in the use of supportive services in these families, leading to a significant increase in unmet needs as the disease progresses. In general, dementia caregivers are known to postpone the use of supportive services until advanced stages of the disease, when it is less likely that these services can be beneficial to them.(42) This is related to caregivers' unawareness of services available to them, a perceived lack of need of support and the patients' reluctance to accept help.(43) This may also be true for these young caregivers. Additionally, in YOD, several factors may further hamper the use of supportive services, such as the delay in diagnosis, professionals' lack of knowledge about YOD and the limited availability of services that can address the individual needs of these families.(10, 44) In our analysis we included the use of supportive services as a covariate and found a strong significant relationship between the use of supportive services and several caregiver HRQoL domains. Although some caution should be used when interpreting these findings this may suggest that when caregivers do initiate supportive services, caregivers may experience fewer limitations due to emotional and physical problems, given the positive association we found with these domains. However, also strong negative relationships emerged with other HRQoL domains, illustrating that the use of supportive services may not counter the negative impact of the double strain of providing high levels of informal care while performing other necessary roles.(45) Furthermore, more unmet psychological needs were associated with fewer limitations experienced by caregivers in performing work and other roles due to emotional problems. This may relate to the fact that YOD caregivers often have to renounce their social roles, diminish their working hours or stop working altogether due to the patient's increasing needs.(5) We believe that caregivers who may be too burdened to initiate or benefit from supportive services and who become more and more isolated in the community might especially be at risk of being adversely affected by the care-giving situation. Since dyads of varying ages were included, differences between the youngest-young and older dyads could be expected. However we did not find an effect for age on domains of HRQoL, except in the area of physical functioning of the caregiver. This suggests that older caregivers may be less resilient and, as a result, are at risk for developing physical complaints due to the care-giving situation.

This study has some limitations. First, because a large proportion of participants was recruited through existing healthcare services, it is likely that patients with an uncommon cause of dementia or phenotype (i.e. predominant behavioural disturbances) were not included because these patients often do not present at a memory clinic or dementia health care facility. As we excluded patients with HIV/AIDS related dementia complex, Huntington's disease, traumatic brain injury and down's syndrome, all of which are important contributors to YOD and may provide unique challenges to caregivers, generalizability of our study is limited. Additionally, a large proportion of patients were unable to complete the

HRQoL assessment. Perhaps if all patients had completed the QOL-AD scale, an association between unmet needs and HRQoL would have been found, considering the higher disease severity among non-completers. However, we were able to explore HRQoL in a unique and large sample of YOD patients and caregivers in spite of low prevalence rates and difficulties in diagnosing YOD. Also, some caution should be used when interpreting the findings of the multiple multiple regression analyses with the RAND-36 as there may be a risk of an inflated Type I error rate, considering the large number of tests in the analysis. However, most relationships we found and discuss in our study appear to be relatively strong (p <0.01 or less) and correspond with clinical practice. To confirm these results and the corresponding hypotheses this has to be tested in future studies in YOD. Furthermore, no LOD comparison group was available, making it impossible to directly compare both groups of patients. Despite these limitations, we believe that this study provides us with important insights in a relatively unexplored area in YOD.

This study shows that unmet needs are related to caregiver HRQoL in YOD. However, the relationship between HRQoL and the concept of unmet needs is complex. The assessment of unmet needs within the context of HRQoL appears to be an important method of identifying those at risk of being most affected by the disease; this assessment is important for enhancing service provision in YOD. When assessing HRQoL, the exploration of HRQoL domain scores should be preferred to generate an individual HRQoL profile as a guideline for more personalized service provision. It is essential that health care workers focus on non-pharmacological interventions aimed at improving the quality of life of both patient and caregiver, especially because pharmacological treatment options for dementia symptoms are limited.(46) Adjusting supportive services to match the individual needs and preferences of these young patients and their caregivers is likely to enhance quality of life.

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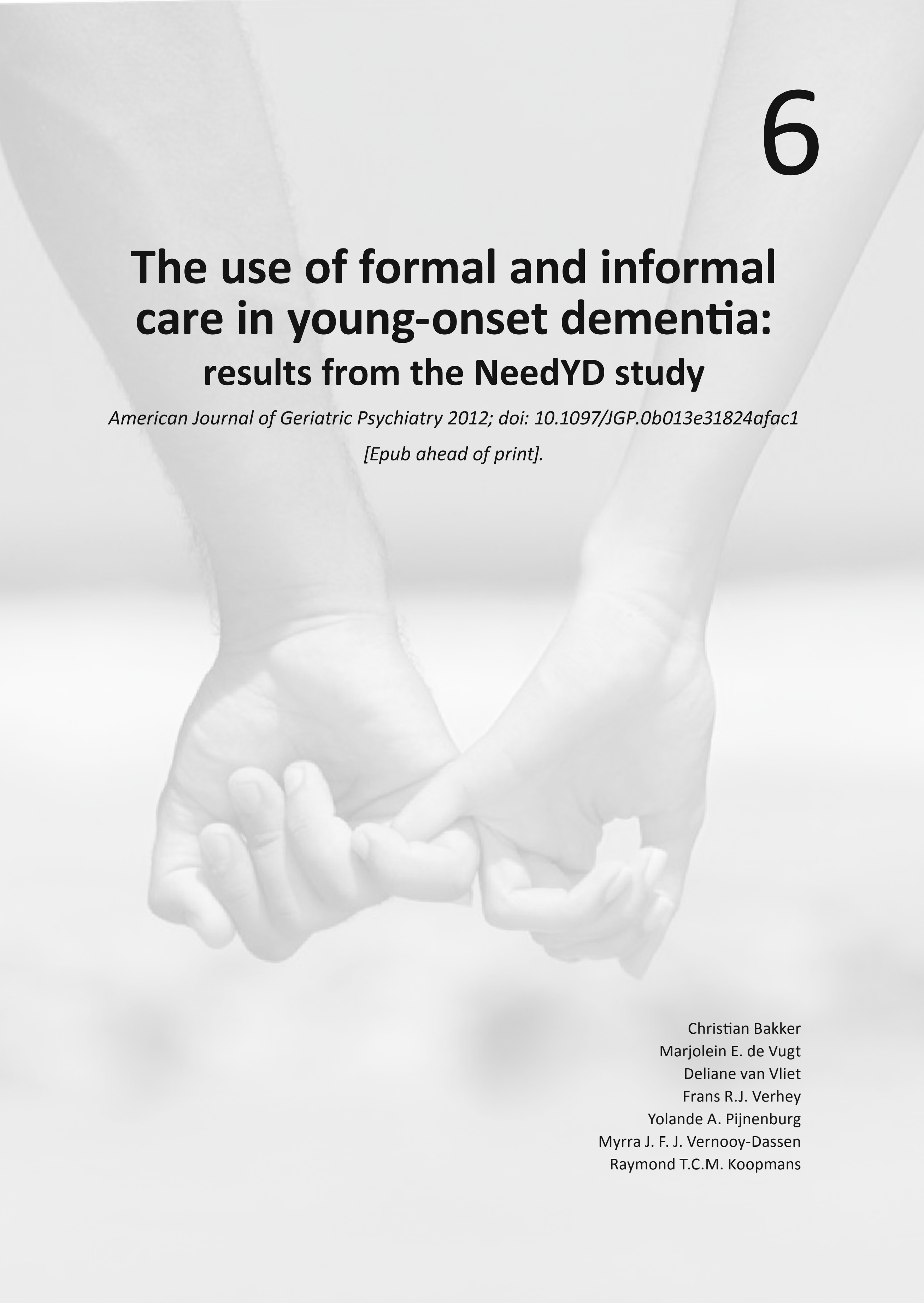

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The use of formal and informal care in young-onset dementia:

results from the NeedYD study

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Christian Bakker
Marjolein E. de Vugt
Deliane van Vliet
Frans R.J. Verhey
Yolande A. Pijnenburg
Myrra J. F. J. Vernooy-Dassen
Raymond T.C.M. Koopmans

Abstract

Objective: Young-onset dementia (YOD) poses specific challenges and issues for both the patient and (in)formal care. This study explores the use of (in)formal care prior to institutionalization, and its association with patient and caregiver characteristics.
Design/Setting: Participants were part of a community-based prospective longitudinal study of 215 patients and their informal caregivers.
Participants: Baseline data of a subsample of 215 patient-caregiver dyads were analyzed.
Measurements: Analyses of covariance were performed to determine correlates of (in)formal care use assessed with the Resource Utilization in Dementia questionnaire (RUD) Lite.
Results: Informal care had a 3:1 ratio with formal care. Supervision/surveillance constituted the largest part of informal care. In more than half of cases patients had only one informal caregiver. The amount of informal care was associated with disease severity, showing more informal care hours in advanced disease stages. Fewer informal care hours were related to more caregiver working hours, especially in younger patients. The amount of formal care was related to disease severity, behavioral problems and initiative for ADL.
Conclusions In YOD it appears that family members provide most of the care. However, other social roles still have to be fulfilled. Especially in spousal caregivers of younger patients in advanced disease stages there is a double burden of work and care responsibilities. This finding also indicates that even within the YOD group there might be important age related differences. The relatively higher amount of formal care use during advanced disease stages suggests a postponement in the use of formal care.

Introduction

Young-onset dementia (YOD), defined as dementia with an onset before the age of 65, is increasingly recognized to have important medical and social consequences. Prevalence rates of YOD have been estimated to range between 54 and 260 cases per 100,000 individuals in the 30-64-year-old age group.[1] YOD, poses specific challenges and problems such as life stage issues, diagnostic difficulties, work related and financial issues, coping with behavioral problems, and relationship problems which adversely affect patient and caregiver wellbeing.[2, 3] It is likely that these issues influence YOD caregivers in the decision-making process regarding their own involvement in care and the use of formal care services.

Literature shows that the presence of informal care reduces but not replaces the use of formal care and delays institutionalization.[4, 5] Thus suggesting that the relationship between informal and formal care is that of a complementary nature, rather than informal or formal care being merely a substitute for one another. The use of formal care services, such as home or day care can be beneficial for both patient and caregiver. But formal care use is generally low in dementia, with family members often providing high levels of care themselves.[6] The behavioral model on service use by Andersen suggests that the use of formal care is a function of predisposing factors (i.e. demographic factors, social characteristics and health beliefs) factors that impede or enable service use (i.e. familial/personal factors and contextual factors) and the families need for care. Within the model the need for care is perceived as the most immediate determinant on the decision to use services.[7]

Indeed, Brodaty et al. found in an extensive literature review that service non-use is related to the perceived lack of need by caregivers.[6] These caregivers often feel low levels of burden, are caring for a patient with diverse levels of impairment, have high care giving skills and adequate amounts of social support. In YOD, caregivers will be reluctant to accept formal help because of the stigma attached to dementia.[8] Reassuring themselves that they are still managing and that their young spouse or relative is not ready for dementia services such as day care yet. However, when the patient also has physical disabilities, services are more likely considered, at least in late onset dementia (LOD) caregivers. This suggests physical disability in dementia is more acceptable as a reason for service use than cognitive decline.[6] Furthermore, also willingness of the patient to accept care is an important factor.[6, 9] Patients who refuse help are characterized by high levels of cognitive impairment[6] and behavioral problems, especially agitation and aggression.[9] This might especially be the case in YOD because of the high prevalence of behavioral problems.[10] When the patient refuses support from others this is likely to isolate the caregiver and add to the high levels of caregiver burden in these younger caregivers[11] and with that also further impede the initiation of formal care.[6]

Other factors associated with service use relate to service characteristics, such as availability of appropriate services, their operating hours or service costs. These issues are believed to be especially relevant in YOD because age appropriate services for these patients and their caregivers in most countries are limited and when these services exist caregivers are not likely to be aware of their existence. This forces caregivers to resort to services designed for the elderly which often have difficulties in meeting the specific care needs of these younger patients and their caregivers.[12] It is plausible that caregivers will renounce services that they perceive as inappropriate, especially when needs of the patient are concerned. Younger caregivers are also likely to look for services accommodating to their own needs and individual preferences. For instance when considering day care the operating hours of the day care centre will be preferred to coincide with their working hours to offer them relief, while most day care centers do not provide flexible operating hours. An important issue in YOD is that younger people often lose their income, while at the same time do not qualify for receiving the same benefits as those available for the elderly because YOD often is not categorized as a disability or terminal illness.[3] This will further dissuade caregivers to initiate formal care services.

There is a lack of studies that examine the use of informal and formal care in YOD and which factors are related to the initiation of (in)formal care. However, this knowledge is important for the development of appropriate interventions to facilitate the use of formal care in YOD. Therefore, the objectives of the current study are (1) to explore the amount of formal and informal care use in YOD patients and their primary caregivers in the period prior to institutionalization, and (2) to identify patient and caregiver factors that are related to the use of formal and informal care in YOD. To our knowledge this is the first explorative study regarding the use of (in)formal care in YOD. We hypothesize that, as in LOD, the amount of informal care will be considerably higher than the amount of formal care. Furthermore, we expect patient factors such as behavioral problems, disease severity and caregiver factors such as age and the ability of the caregiver to deal with the burden of caring for the patient to be associated with (in)formal care use.

Methods

Study design, selection of patients, ethical considerations

This study is part of the Needs in Young-onset Dementia (NeedYD)-study. The aims and design of this two-year follow-up study are described elsewhere.[13] Baseline data from a total of 215 patient-caregiver dyads were used for this study. All patients were living at home at time of inclusion. Patients were consecutive referrals from University Medical Centers (n=56), regional hospitals (n=10), or regional community mental health services (n=20). Additionally, dyads applied for participation (n=14) or were recruited through specialized day care facilities (n=115). People with onset of disease symptoms before the age of 65 were included in the study thus age at inclusion could be over 65. Dementia subtypes were established according to regular criteria.[7, 14-18]

The Medical Ethics Committee of the Maastricht University Medical Center approved the study protocol. The local ethics committees of the participating institutions also gave consent.

Data collection and assessments

Trained researchers and research assistants collected data using structured interviews and questionnaires. A one-day training session and regular consensus meetings safeguarded the integrity of the data collection.

Assessment of formal care

Patient and caregiver formal care use was assessed with the Resource Utilization in Dementia (RUD)-lite questionnaire. The RUD-lite is a shortened version of the RUD instrument which has high intra-rater reliability and has been found a valid instrument.[19, 20] It covers 95% of the resource-use questions examined in the complete RUD questionnaire.[21] In interviews with primary caregivers, the number of service visits and the duration of these visits over a period of three months were assessed. In the RUD-lite, formal services are defined as support from a district nurse, home care, meals-on-wheels, day care, help with transport, temporary admissions at a nursing home and 'other services' mentioned by the caregiver. Formal services used by the caregiver were defined as the use of information, psycho-education, individual support, support group and 'other services'.

Assessment of informal care

The RUD-lite questionnaire was also used to assess informal care provided by the primary caregiver, addressing three components: (1) support in activities of daily living (ADL); (2) support of instrumental activities of daily living (IADL); and (3) supervision/surveillance, defined as the prevention of dangerous events and managing behavioral and psychological symptoms in dementia (BPSD). Informal care provided by others, such as children, neighbors and friends, was assessed using a semi-structured interview assessing who provided which type of support (ADL, IADL and/or supervision).

Patient and caregiver factors

Dementia severity was assessed with the Global Deterioration Scale (GDS), which is a seven point scale (1-7) ranging from 'no global impairment' (1) to 'very severe global impairment' (7). The GDS is a widely used instrument which has been validated against behavioral, neuro-anatomic and neurophysiologic measures, for which significant correlations have been found.[22] ADL disabilities were assessed with the Interview for Deterioration in Daily living activities in Dementia (IDDD). A higher IDDD score means that more assistance is needed. This scale has good construct validity, test-retest reliability, and responsiveness to changes over a six month period.[23] Neuropsychiatric symptoms were assessed with the Dutch version of the Neuropsychiatric Inventory (NPI).[24] This instrument has high inter-rater agreement and is found to be a valid rating scale.[25] The frequency (F) and severity (S) of each symptom is rated on a five (0-4) and four-point (0-3) likert-scale, respectively. These scores are summed to calculate a total score. The Short Sense of Competence Questionnaire (SSCQ) was administered to assess caregiver's competence in dealing with the burden of caring for a dementia patient. The SSCQ is based on the family-crisis model and includes issues from the Zarit's Burden Scale. Three domains are distinguished: (1) satisfaction with the demented person as a care recipient; (2) satisfaction with one's own performance as a caregiver; and (3) consequences of involvement in care for the personal life of the caregiver. The SSCQ consists of 7 items rated on a 5-point likert scale from 1 (agree very strongly) to 5 (disagree very strongly). The SSCQ was reported to have satisfactory reliability and validity.[26] Additionally, characteristics such as physical health problems of the patient, care duration and demographic characteristics of both patients and caregivers were assessed through a semi-structured interview.

Statistical analysis

Analyses were performed using the Statistical Package for Social Sciences (SPSS) version 18.0. Proportions or means were calculated to describe the characteristics of patients and caregivers. Group comparisons were done by Chi-square tests for categorical variables and t-tests for continuous variables. Analyses of co-variance (ANCOVA) were used to analyze factors associated with the use of both informal and formal care. First, associations between separate characteristics and the two outcome variables were determined. Factors likely associated with the outcome variable on the basis of literature studies and factors with associations with $p \leq 0.25$ were included in the model. These factors included age of the patient, disease severity, physical health problems of the patient, behavioral problems, ADL initiative, gender of the caregiver, age of the caregiver, sense of competence, number of caregiver working hours and whether the patient and the caregiver lived together. These factors were then entered in a model with respectively the number of hours of informal care and the number of hours of formal care as the dependent variables.

Because resource use data are often skewed, as in our data, parametric estimates with 95% confidence intervals were estimated with non-parametric bootstrapping with 5000 replications and bias-corrected, and accelerated (BCa) confidence intervals in order to determine whether the skewness of the data affected the outcomes of the model.[27] Because the results of the bootstrap did not differ substantially from the ANCOVA analysis, we present the results of the ANCOVA analysis.

Results

Patient and caregiver characteristics

Out of the original sample (N = 215), 209 patient-caregiver dyads were included in this study. Six caregivers did not want to participate due to high levels of perceived burden. The mean age of the patients was 61.8 years (SD ± 5.5), and the male to female ratio was approximately equal (Table 1). The majority of patients had a moderately severe stage of dementia (GDS stage 4). The majority of patients suffered from Alzheimer’s disease. The mean age of the caregivers was 58.4 years (SD ± 9.3) and approximately half of the caregivers still worked at time of inclusion. All patients lived at home at time of inclusion.

Table 1. Characteristics patients and caregivers participating in this study (N=209)

	Frequency (n)	Percentage (%)[b]	Mean age ± SD (Range)
Patient gender			
Male	114	(54.5)	61.8 ± 5.0 (47 – 74)
Female	95	(45.5)	60.2 ± 5.9 (43 – 73)
Disease severity (GDS) [a]			
Mild (3)	34	(16.3)	
Moderate (4)	76	(36.4)	
Moderately severe (5)	53	(25.4)	
Severe (6)	29	(13.9)	
Very severe (7)	6	(2.9)	
Dementia type			
Alzheimer's Disease	119	(56.9)	
FTD, behavioral variant	28	(13.4)	
Primary Progressive Aphasia (PPA)	13	(6.2)	
Vascular dementia	24	(11.5)	
Mixed dementia	8	(3.8)	
Other causes	17	(8.1)	
Living arrangements			
Alone	14	(6.7)	
With spouse	153	(73.2)	
Spouse and children	38	(18.2)	
With children	2	(0.95)	
Other	2	(0.95)	
Caregiver relationship			
Spouse	191	(91.4)	59.9 ± 6.7 (42 – 78)
Son/Daughter	13	(6.2)	33.6 ± 7.7 (20 – 42)
Other	5	(2.5)	66.0 ± 5.6 (58 – 73)
Caregiver gender			
Male	84	(40.2)	61.9 ± 6.5 (45 – 78)
Female	125	(59.8)	56.2 ± 10.2 (20 – 74)
Still working			
Patient	9	(4.3)	
Caregiver	102	(48.8)	

Notes: FTD indicates frontotemporal dementia;[a] Disease severity scores of 11 cases (5.3%) were not available because those patients were not seen by the researcher
[b] All percentage values are presented as percentages of the total group

The use of formal and informal care

Formal care was provided in 79.9% of the cases with an average amount of 79.6 h/month (Table 2). In most cases multiple services were used. Formal care consisted of day care (66%), home care (29.7%) or the use of a district nurse (7.2%). Of the patients receiving day care, 114 patients attended a specialized day care center for YOD patients, and 24 patients visited a regular day care center. Approximately half of all cases used transportation services, mostly provided by the day care center. The Meals-on-wheels service was used in only a few cases (n =5).

Other services mentioned included individual support (9.1%), individual support with daytime activities (7.7%), temporary admission into a nursing home (7.7%), and other kinds of support, such as support groups, information meetings or courses (7.2%).

Most caregivers used multiple types of support. Approximately 48% of the caregivers used information from different resources like self-help books, brochures, the Internet and information meetings held by the Dutch Alzheimer's Society. Caregivers also frequently used support from a social worker, psychiatric nurse or case manager (38.7%) or participated in a support group (33.0%). A separate psycho-education service was used in a small proportion of cases (13.9%).

Informal care was provided in all cases. The amount of informal care was more than three times the amount of formal care at 259.6 h/month. Although supervision/surveillance was only reported by approximately half of the caregivers, this constituted the largest proportion in time (230 h/month), which was larger than support for ADL (62.9 h/month) or IADL (82.4 h/month).

Almost half of the patients (46%) also received informal support from someone other than the primary caregiver. This support primarily consisted of support with IADL activities (71.7%) or supervision (17.4%). Additional informal support was mainly provided by children (53.3%) or other family members (24%).

Table 2. Mean hours spent during a period of 4 weeks on formal and informal care, (N=209)

	Frequency (n)	Percentage (%)[a]	Mean h/ month	(± SD)[b]
Formal care patient				
Specialized day care	114	(54.5)	79.7	(31.3)
Day care	24	(11.5)	59.1	(33.5)
Home care	62	(29.7)	24.5	(47.3)
District nurse	15	(7.2)	10.7	(11.7)
Total formal care patient	172 [c]	(79.9)	79.6	(48.2)
Other psychosocial support	65	(31.0)	n.a.	
Informal care patient				
ADL	129	(61.7)	62.9	(61.3)
IADL	195	(93.3)	82.4	(59.8)
Supervision/surveillance	118	(56.4)	230.0	(239.3)
Total informal care	215	(100.0)[c]	259.6	(274.8)
Formal care Caregiver				
Information	101	(48.3)	n.a.	
Individual support	81	(38.7)	1.0	(1.4)
Support group	69	(33.0)	1.7	(1.4)
Psychoeducation	29	(13.9)	n.a.	

Notes: ADL indicates activities of daily living; IADL = instrumental activities of daily living; [a] All percentage values are presented as percentages of the total group, most subjects used different types of care_; [b] Excluding non-users; [c] n and percentage represent the total number of patients who received any kind of formal or informal care; n.a.= not available

The association between patient and caregiver characteristics and (in)formal care

Informal care

The general linear model (analysis of covariances) showed that disease severity was statistically significantly associated with the number of hours of informal care, F (4,169) = 2.45, p < .05, *partial* η^2 = .05 (Table 3, univariate analyses are not shown, but are available on request). Jonckheere's test revealed a significant trend (p < 0.001) in the data with a higher disease severity resulting in a higher amount of informal care hours, *J* = 8.84, *z* = 4.50, r = 0.32. Additionally, there was an interaction effect between patient age and the number of caregiver work hours and informal care hours. This suggested that, in younger patients, more caregiver work hours were related to less informal care hours, F(1,169) = 6.04, p < .05, r = .03.

Formal care

The general linear model (analysis of covariances) showed that disease severity was significantly associated with the amount of formal care hours F(4,168) = 9.41, p < .001, *partial* η^2 = .18. Jonckheere's test revealed a significant trend (p < 0.001) in the data with a higher disease severity leading to more formal care hours, *J* = 8.67, *z* = 4.39, r = 0.32. Additionally, behavioral problems were found to be significantly related with formal care, with higher levels of behavioral problems associated with more formal care hours, F(1,168) = 4.77, p < .05, r = .03. Furthermore, the initiative of the patient in ADL functioning was significantly associated with formal care hours, with higher levels of patient initiative relating to fewer formal care hours, F(1,168) = 5.66, p < .05, r = .03.

Table 3. General linear model (ANCOVA) of factors associated with the use of formal and informal care

	Informal care					Formal care				
	B^b	SE	*P*-value	95% CI		B^b	SE	*P*-value	95% CI	
				Lower	Upper				Lower	Upper
Intercept	148.62	152.12	.330	-151.69	448.92	176.76	28.67	< .001	120.17	233.36
Independent variables										
Patient factors										
Age of the patient	2.36	4.85	.627	-7.21	11.93	1.34	.95	.159	-.53	3.22
Disease severity			.048					< .001		
GDS 3	0^a					0^a				
GDS 4	56.45	56.23	.317	-54.57	167.44	-17.31	10.57	.103	-38.19	3.56
GDS 5	104.64	58.65	.076	-11.14	220.42	5.65	11.21	.615	-16.48	27.78
GDS 6	201.96	69.89	.004	63.99	339.93	18.19	13.13	.168	-7.74	44.11
GDS 7	165.34	118.19	.164	-67.97	398.66	100.89	22.27	< .001	56.93	144.85
Physical problems	-14.06	42.38	.740	-97.72	69.60	-13.06	8.00	.104	-28.85	2.73
Behavioral problems	2.24	1.17	.057	-.07	4.55	.49	.23	.030	.05	.93
Initiative in ADL functioning	-.48	.43	.261	-1.32	.36	-.21	.09	.018	-.38	-.04
Caregiver factors										
Gender of the caregiver	-1.58	44.81	.972	-90.04	86.88	-8.21	8.54	.338	-25.08	8.65
Age of the caregiver	-.95	3.30	.775	-7.47	5.58	-1.13	.65	.085	-2.41	.16
Sense of competence (SSCQ)	-4.66	3.99	.245	-12.53	3.22	-.32	.77	.677	-1.84	1.20
Number of caregiver work hours	-3.72	1.45	.011	-6.57	-.87	.21	.27	.432	-.32	.74
Living together	159.18	85.64	.066	-10.47	328.82	7.22	16.34	.659	-25.03	39.48
Interaction between patient's age and the number of caregiver work hours	-.53	.22	.015	-.96	-.11	-.05	.04	.250	-.13	.03

Notes: ANCOVA = analysis of covariance; ^{a} reference category ^{b} all continuous variables were centered at mean value

Model summary informal care: adjusted r2 = 0.18; *df* = 13, 184; *F* = 3.906; p < .001. Model summary formal care: adjusted r2 = 0.30; *df* = 13,183; *F* = 6.504; p <.001.

Discussion

To our knowledge, this is the first study examining formal and informal care in YOD. In YOD informal and formal care had a 3:1 ratio. In more than half of the cases, patients had only one informal caregiver. The amount of informal care was associated with disease severity, showing more informal care hours with advanced disease severity. Fewer informal care hours were associated with more caregiver work hours, in particularly when the patient was younger. The amount of formal care was not related to caregiver characteristics but to patient factors such as disease severity, behavioral problems and ADL initiative.

All patients in our sample received informal care, and in 80% of cases, formal care was also provided. In the latter case, family and relatives provide the greater part of the care. This is in line with figures from other countries on care provision of patients with LOD. Previous studies reported a 3:1 ratio in the USA[28] and a 7:1 ratio in an Italian study.[29] In almost half of our cases, the caregiver received additional help from friends, relatives or neighbors. A similar study on LOD care found that 57% of patients were receiving additional informal support from someone other than the primary caregiver.[30] Because we did not assess the amount of support provided by others, it is difficult to compare these findings. However, this may indicate that the social support system is limited for younger caregivers, which might be related to the caregivers' dedication in caring for the patient themselves, the patient's reluctance to accept support from others[12] or a lack of people able or willing to offer support.
Supervision/surveillance, which we defined as the prevention of dangerous episodes and management of behavioral and psychological symptoms in dementia (BPSD), constituted the largest part of informal care, larger than hours spent for ADL and IADL combined. This ratio is consistent with findings from previous research in this area on LOD care.[31, 32] BPSD is highly prevalent, even in earlier stages of the dementia. The caregiver has to cope with these problems and often supervise the patient to prevent accidents or issues in interaction with other people, adding to the time that the caregiver has to spend with the patient. In YOD, this is an especially important issue because of the higher prevalence of FTLD, which is characterized by high rates of BPSD.[33] This will further complicate caregiving and add to the burden felt by the caregiver.[34]

The amount of informal care was associated with disease severity with a higher amount of informal care in more advanced stages of the disease. This finding was expected because the dependency of the patient increases with progression of the disease. An increase in informal care hours is likely to adversely affect the health and well-being of the caregiver, especially when the care duration is lengthy.[35, 36] The timely provision of formal care might counter these adverse effects. Rosness et al. found that in YOD, domiciliary nursing care protected caregivers against high levels of depression, and this protection was also suggested to occur with the use of day care.[37] Additionally, we found that less informal care was provided to younger patients by caregivers who worked more hours. This may seem obvious because working caregivers will have less time available to care for the patient. However, it demonstrates that especially in YOD caregivers might experience difficulties in finding a balance between caregiving and other social roles, such as breadwinning and being a parent.[12, 38] In our study, we demonstrated that this might especially be true for caregivers of the youngest patients.

Disease severity was also associated with the amount of formal care. When looking at the different stages of disease severity, we found a different pattern in the amount of formal care compared to the pattern in informal care. Especially in advanced stages of the disease, a relatively higher increase in formal care was found compared to earlier stages of the disease. This might be explained by the fact that caregivers postpone the use of formal care until it becomes inevitable. This delay in the use of formal services is probably related to difficulties in accepting help and the stigma attached to dementia.[8, 9] Moreover, the lack of specialized dementia services for these younger patients and their caregivers probably increases the barriers against using formal care. Additionally, the patient's lack of insight regarding cognitive and behavioral changes hampers their acceptance of help or support.[9, 12] This will especially be true for FTLD, which is characterized by unawareness of deficits.

Furthermore, the severity of behavioral problems was associated with higher levels of formal care hours, but not with informal care hours. Behavioral problems are an important source of stress for informal caregivers[39] and are a main predictor of institutionalization.[40] The results of our study suggest that also in the period prior to institutionalization, more formal care is provided when behavioral problems are present. This emphasizes the need to target these behavioral symptoms, not only to increase patient and caregiver well-being, but also to reduce the costs of dementia care.

There are some limitations of this study. First, this is a cross sectional study with no LOD comparison group that recruited participants through memory clinics as well as through existing health care services. This will result in overestimation of the use of formal services as well as limit the generalizability of our findings, especially when considering the heterogeneity of the YOD population. However, in this way, we were able to recruit a unique and large sample of YOD patients and caregivers in spite of low prevalence rates and difficulties in diagnosing YOD. Second, we did not take contextual factors into account in our analyses, such as economic data of the caregiver, availability of appropriate services, their operating hours or service costs. It is likely that these factors also are related to the use of (in)formal care. These issues should be addressed in further research.

Conclusions

In YOD, it appears that the spouse and other family members provide most of the care, while other social roles still have to be fulfilled. Especially in spousal caregivers of younger patients in advanced disease stages, there is a double burden of work and care responsibilities. This double strain needs to be addressed in caregiver support programs and has implications for service provision in YOD. This finding also indicates that, even within the YOD group, there might be important age-related differences that need more exploration in future research. The relatively higher amount of formal care use in advanced disease stages suggests a postponement of service use. Development or improvement of tailored care in YOD patients and support for the acceptance of care is needed because earlier service utilization can have important implications for patient and caregiver well-being, such as delaying institutionalization.[41] Finally, longitudinal research can provide more insight on the interaction between formal and informal care over time and the decision-making process of initiating formal care.

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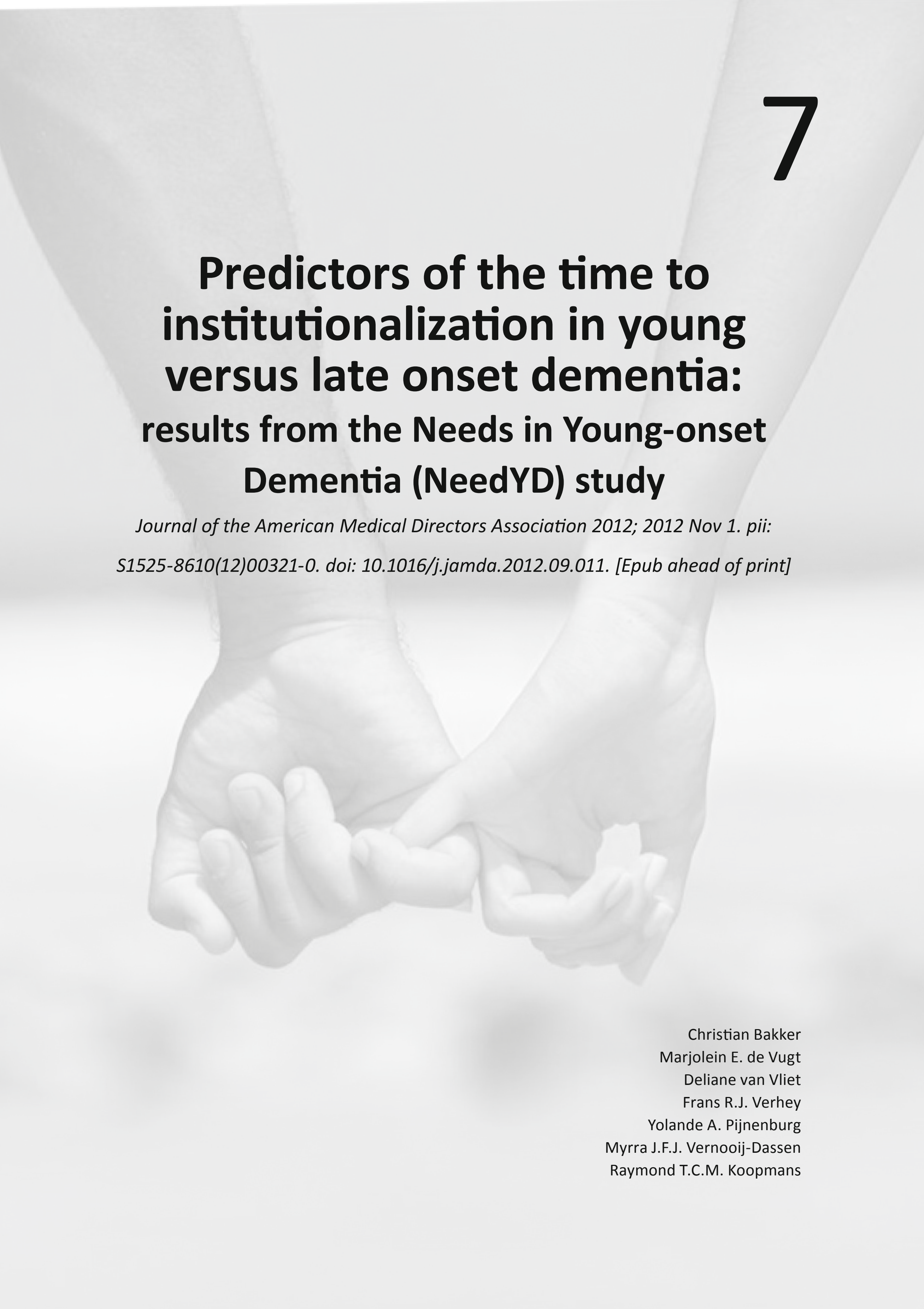

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Predictors of the time to institutionalization in young versus late onset dementia: results from the Needs in Young-onset Dementia (NeedYD) study

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Christian Bakker
Marjolein E. de Vugt
Deliane van Vliet
Frans R.J. Verhey
Yolande A. Pijnenburg
Myrra J.F.J. Vernooij-Dassen
Raymond T.C.M. Koopmans

Abstract

Objective: The aim of this study is to (1) investigate the time from symptom onset to institutionalization in persons with young-onset dementia (YOD) and compare these findings with late onset dementia (LOD) and (2) to determine which factors predict institutionalization in persons with YOD compared with LOD.

Design/Setting: Longitudinal study of community-dwelling YOD and LOD patients and their caregivers.

Participants: A total of 226 YOD and 102 LOD patients and their informal caregivers were recruited through memory clinics and health care facilities.

Measurements: Cox proportional hazard (CPH) models were used to relate covariates with time to institutionalization. The main outcome was time from symptom onset to institutionalization. Key predictors were cohort (YOD or LOD), neuropsychiatric symptoms, caregiver distress due to neuropsychiatric symptoms and caregivers' sense of competence (SSCQ total score).

Results: The time from symptom onset to institutionalization was nearly nine years for YOD patients compared to approximately four years for LOD patients. In the YOD group, apathy significantly predicted time of institutionalization. Furthermore, the caregiver's competence in caring for the person with dementia significantly predicted institutionalization in both groups.

Conclusion: YOD patients are cared for at home for a longer period of time than LOD patients. The results of the present study underline the importance of a timely diagnosis for these patients and their families to facilitate the initiation of appropriate care and support. Support programs aimed at enhancing the caregivers' sense of competence and ability to deal with neuropsychiatric symptoms, especially apathy, may postpone the institutionalization of the patient.

Introduction

Dementia is the primary reason for institutionalization in the elderly, with 20% of persons with dementia being institutionalized in the first year after diagnosis, 50% after five years and up to 90% after eight years.[1] In Western Europe, policy is aimed at delaying institutionalization in persons with dementia through the provision of support programs for patients and caregivers.[2] Institutional care is costly and far exceeds the cost of informal care in high income countries.[3] Furthermore, persons with dementia and their families have a particular desire to prevent institutionalization due to concerns about the quality of care in nursing homes and the potential negative effects of institutionalization on patients' quality of life.

For young-onset dementia (YOD), defined as dementia with symptom onset before the age of 65, the decision to place a relative in a long-term care facility is likely to be complicated by the specific issues and problems that these families face.[4] Caring for a dementia patient is known to adversely affect caregiver mental and physical health, especially when the patient is younger.[5] YOD caregivers must often deal with severe neuropsychiatric symptoms, causing high levels of distress.[6] Neuropsychiatric symptoms, and especially the related caregiver distress, increase the risk for institutionalization in late onset dementia (LOD) [7, 8] and may also be main predictors of institutionalization in YOD, considering the high prevalence of neuropsychiatric symptoms in these patients.[4] The literature suggests that the course of dementia in persons with YOD is more aggressive compared to that in LOD, likely shortening the time to institutionalization.[9] Some evidence for a more rapid progression of dementia has been found in AD patients without the ApoEε4 genotype,[10] which is a common cause of dementia in younger patients.[11]

Furthermore, YOD caregivers are faced with the double burden of providing care while performing other roles, such as parenting, working and managing the household.[12] Due to this double strain, these caregivers may feel hampered in caring for the patient at home even in less advanced stages of the dementia. This stresses the importance of timely support programs aimed at providing respite care and enhancing the caregivers' feelings of ability to care for the patient. The caregivers' sense of competence has been identified as a strong determinant of delaying institutionalization.[13] However, in most countries, support programs specifically designed for younger patients and their caregivers are largely lacking, limiting respite possibilities. This lack of support may compel caregivers to place their relative in a long-term care facility despite their wish to care for the patient at home as long as possible.

The aim of the current study is to (1) investigate the time from symptom onset to institutionalization among people with YOD and compare this with findings from those with late onset dementia (LOD) and (2) to determine which factors predict the time to institutionalization among people with YOD compared with those with LOD. We hypothesize that YOD produces greater caregiver burden and, therefore, that the time from symptom onset to institutionalization is shorter than that for LOD. Furthermore, we expect that neuropsychiatric symptoms and the related caregiver distress are stronger predictors of the time to institutionalization in the case of YOD than in LOD. We also hypothesize that caregiver sense of competence is a stronger predictor of the time to institutionalization in YOD than in LOD, considering the aforementioned double burden of care in YOD.

Methods

Study design and selection of participants

The present study is part of the Needs in Young-onset Dementia (NeedYD) study. The design of this two-year follow-up study has been described previously.[14] Baseline data from 215 patient-caregiver dyads were used. Patients were consecutively referred from university medical centers (n=56), regional hospitals (n=10), or regional community mental health services (n=20). Additionally, dyads applied for participation (n=14) or were recruited through YOD-specialized day care facilities (n=115). People with symptom onset before age 65 were included; therefore, age at inclusion could be over 65. The diagnoses were made by the clinician on the basis of clinical, neuropsychological and neuroimaging data, according to the criteria from the 4th edition of the Diagnostic and Statistical Manual of Mental Disorders, text revision (2000) for dementia,[15] and the Dutch consensus guidelines,[16] that include internationally accepted criteria for the subtype diagnoses.[17-21] Clinical diagnoses were confirmed by the researcher through clinical files for every patient and medical hospital records, when available. The exclusion criteria were as follows: (1) lack of a reliable informant (contact at least once per week), (2) lack of participant informed consent, (3) participant residence in a nursing home and (4) dementia caused by HIV, alcohol, traumatic brain injury, Down's syndrome or Huntington's chorea.

The comparison group consisted of 119 LOD patient-caregiver dyads. These participants were selected from a previously performed two-year follow-up cohort study, the MAAstricht Study of BEhaviour in Dementia (MAASBED) study.[22] The NeedYD study protocol was based on the methods of the MAASBED study, using the same design, assessment instruments and diagnostic criteria to enable the comparison of outcomes. In the MAASBED study, patients with dementia were included irrespective of the diagnostic subtype, and they were consecutively enrolled from the Memory Clinic of the MUMC+ or from the Regional Institute for Community Mental Health Care (RIAGG) in Maastricht.[17] The MAASBED cohort included YOD patients, who were added to the YOD group in the present study.

Data collection and assessments

The Medical Ethics Committee of the Maastricht University Medical Center approved the study protocol. The local ethics committees of the participating institutions also gave consent. Written informed consent was obtained from the patients or their legal representatives prior to the study. Trained researchers and research assistants utilized structured interviews and questionnaires to collect the data in six-month intervals during a period of two years. A one-day training session and regular consensus meetings safeguarded the validity of the data collection.

Time to institutionalization

Using a semi-structured open-ended interview, the primary caregivers were asked for the year of onset and the nature of the first signs or symptoms. Caregivers were then asked to elaborate on their answers and identify any earlier signs or symptoms. The year of the earliest sign or symptom, in the cognitive, behavioral or functional domain, was recorded as the year of symptom onset. For patients who were admitted to a nursing home, the duration (in months) between symptom onset and institutionalization was calculated by subtracting the year of symptom onset from the year of institutionalization. As the only data available at symptom onset were demographic characteristics, the duration from study inclusion to institutionalization was calculated as well to be able to perform analyses with the data from the baseline assessment.

Neuropsychiatric symptoms, related caregiver distress and caregivers' sense of competence

Neuropsychiatric symptoms were assessed with the Dutch version of the *Neuropsychiatric Inventory (NPI)*.[23] This instrument has high inter-observer reliability and is a valid rating scale for a wide range of neuropsychiatric symptoms of dementia.[24] The frequency (F) and severity (S) of each symptom are rated on five- (0-4) and four-point (0-3) Likert scales, respectively. These scores are summed to calculate total scores for the following four neuropsychiatric syndromes: hyperactivity (agitation, euphoria, disinhibition, irritability and aberrant motor behavior), mood (depression and anxiety), psychosis (delusions, hallucinations and night-time behavioral disturbances) and apathy (apathy and appetite and eating abnormalities), based on the results from the European Alzheimer Disease Consortium.[25] Caregivers also rated the level of distress they experienced as a result of the various behavioral symptoms on a scale from 0 (none) to 5 (extreme). The NPI-Distress (NPI-D) score is the sum of these ratings (range 0–60). The *Short Sense of Competence Questionnaire (SSCQ)* was administered to assess caregivers' capabilities in dealing with the burden of caring for a dementia patient. Three domains are evaluated, as follows: (1) satisfaction with the demented person as a care recipient; (2) satisfaction with one's own performance as a caregiver; and (3) consequences of involvement in care for the personal life of the caregiver. The SSCQ consists of 7 items rated on a 5-point Likert scale from 1 (agree very strongly) to 5 (disagree very strongly). The SSCQ is reported to have satisfactory reliability and validity.[26]

Other covariates

Dementia severity was assessed with the *Global Deterioration Scale (GDS)*, a seven-point scale (1-7) ranging from 'no impairment' (1) to 'very severe cognitive impairment' (7). The GDS is a widely used instrument that has been validated against behavioral, neuro-anatomic and neurophysiologic measures, for which significant correlations have been found.[27] Furthermore, demographic data of both the patient and caregiver, such as age, gender, education, patient's living arrangement and patient-caregiver relationship, were collected through a structured interview with the primary caregivers.

Statistical analysis

Analyses were performed using the Statistical Package for Social Sciences (SPSS) version 18.0.2 (SPSS Inc., Chicago, USA). Proportions and means were calculated to describe the characteristics of the patients and caregivers. Group comparisons were conducted using Chi-squared tests for categorical variables and t-tests for continuous variables. Cox proportional hazards (CPH) models were used in two steps to relate covariates with time to institutionalization, treating deaths and drop-outs as censored. In the first step, a CPH model was fit for the time from *symptom onset* to institutionalization (in months) and the patient and caregiver characteristics that were known at disease onset. These covariates were as follows: group (YOD or LOD), gender of patient and caregiver, patient's relationship with the caregiver (spouse or child), diagnosis (Alzheimer dementia (AD), frontotemporal dementia (FTD), mixed or vascular dementia (mixed/vasc) and other type of dementia), and patient's and caregiver's educational levels (low, average or high). In the second step, another CPH model was fit for the time from *inclusion* in the study to institutionalization (in months) and baseline predictors. The key predictors in the model were group (YOD or LOD), behavioral problems (NPI hyperactivity, mood, apathy and psychosis total score), caregiver distress due to neuropsychiatric symptoms (NPI-Distress total score) and caregiver's sense of competence (SSCQ total score). Additional covariates that were entered in the model were age, patient's gender, caregiver's gender, severity of the disease (the variable was dichotomized, with a score of ≥ 5 representing advanced dementia) and diagnosis (Alzheimer dementia (AD), frontotemporal dementia (FTD), mixed or vascular dementia (mixed/vasc) and other type of dementia). Because we included patients from different phases of the caregiving trajectory, there are differences in the length of time from disease onset to inclusion in our study. Therefore the time from disease onset until inclusion was also added as a covariate in this analysis to adjust for these differences. $P < 0.05$ was used as the threshold for statistical significance. Missing values for baseline covariates were not common in the YOD or LOD group. In the YOD group, there were missing values for disease severity (4.5%), neuropsychiatric symptoms (3.2%), and caregivers' sense of competence (1.4%). In the LOD group, there were missing values for caregivers' sense of competence scores (3.7%). Missing values on the sense of competence questionnaire were substituted by the mean of the item's sub-scale, provided that only one item of the sub-scale was missing.

Results

Patient and caregiver characteristics

From the original samples, 226 patient-caregiver dyads were included in the YOD group and 102 dyads were included in the LOD group. In the YOD group, six caregivers refused to participate due to high levels of subjective burden. The demographic characteristics of these caregivers did not differ from those of the included caregivers. Information about symptom onset was not available for four YOD and two LOD patients.

There were significant differences between the YOD and LOD group on demographic variables (Table 1). Patients in the YOD group exhibited more severe dementia than patients in the LOD group. In the YOD group, frontotemporal dementia was the second most common cause of dementia, whereas in the LOD group, this was vascular dementia. Mean disease duration differed between the two groups, with 88.4 months (SD 45.5) for the YOD and 39.0 months (SD 29.3) for the LOD group. Furthermore, there were significant differences in behavioral symptoms between the groups. YOD patients had significantly higher levels of both hyperactive symptoms and apathy and significantly lower levels of mood symptoms. There was no difference in caregiver distress related to the neuropsychiatric symptoms.

Table 1. Baseline characteristics of patients and caregivers

Characteristics		YOD (N=226)		LOD (N=102)		Test value	P value
Patient gender	Male (%)	125	(55.3%)	38	(37.3%)	$\chi^2 = 9.2$	.002
Patient age at inclusion	Mean (SD)	61.3	(5.4)	80.9	(5.8)		
	Range		[43 – 74]		[69-99]		
Age at symptom onset	Mean (SD)	53.8	(6.0)	77.8	(6.2)		
	Range		[34-64]		[66-97]		
Patient education						$\chi^2 = 57.5$	<.001
Low		99	(43.8%)	65	(63.7%)		
Average		77	(34.1%)	28	(27.5%)		
High		50	(22.1%)	9	(8.8%)		
Dementia type						$\chi^2 = 18.4$*	<.001
Alzheimer’s dementia (AD)		135	(59.7%)	74	(72.5%)		
Frontotemporal dementia (FTD)							
Behavioral variant		28	(12.4%)	2	(2.0%)		
Primary progressive aphasia (PPA)		13	(5.8%)	1	(1.0%)		
Vascular dementia (VD)		25	(11.1%)	19	(18.6%)		
Mixed dementia		8	(3.5%)	3	(2.9%)		
Lewy body dementia		7	(3.1%)				
Parkinson’s dementia				3	(2.9%)		
Dementia due to another cause		10	(4.4%)				
Disease severity (GDS):						U = 9125.5	.009
Mild (2, 3)		36	(16.0%)	20	(19.6%)		
Moderate (4, 5)		145	(64.1%)	86	(79.5%)		
Severe (6, 7)		35	(15.5%)	1	(1.0%)		
Neuropsychiatric symptoms (NPI)	Mean (SD)	24.6	(19.3)	23.1	(22.3)	U = 10269.0	.245
Hyperactivity sub syndrome	Mean (SD)	10.1	(10.7)	7.4	(9.9)	U = 9494.0	.016
Affective sub syndrome	Mean (SD)	3.8	(4.9)	5.7	(6.3)	U = 13603.0	.008
Psychosis sub syndrome	Mean (SD)	2.7	(5.0)	4.2	(7.2)	U = 12578.0	.099
Apathy sub syndrome	Mean (SD)	7.8	(7.2)	5.7	(6.3)	U = 9491.0	.013
Disease duration	Mean (SD) in months	88.4	(45.8)	39.0	(29.3)	U = 3441.0	<.001
Caregiver gender	Female (%)	136	(60.2%)	66	(64.7%)	$\chi^2 = .609$	.435
Caregiver age at inclusion	Mean (SD)	58.7	(9.2)	64.3	(12.7)		
	Range		[20 – 78]		[37-90]		
Caregiver education						$\chi^2 = 15.6$	.049
Low		70	(31.0%)	35	(34.3%)		
Average		98	(43.4%)	43	(42.2%)		
High		58	(25.7%)	24	(23.5%)		

Continuation table 1 on next page

Continuation table 1: Baseline characteristics of patients and caregivers

Characteristics		YOD (N=226)		LOD (N=102)		Test value	P value
Caregiver relationship:							
Spouse		207	(91.6%)	48	(47.1%)		
Child		14	(6.2%)	46	(45.1%)		
Other		5	(2.2%)	8	(7.8%)		
Caregiver competence (SD)	SSCQ total score	22.5	(5.4)	22.7	(6.7)	U = 11806.0	.581
Caregiver distress (SD)	NPI-D total score	12.7	(9.2)	11.7	(10.2)	U = 10099.0	.167

Notes: GDS = Global Deterioration Scale; NPI = Neuropsychiatric Inventory; SSCQ = Short Sense of Competence Questionnaire; *based on the categories: (1) AD, (2) FTD, (3) VaD and mixed dementia and (4) Other

Time from symptom onset to institutionalization in YOD versus LOD

In the YOD group, 33% of the patients (n=74) were institutionalized within the study period, compared to 44% (n=45) of the LOD group. The duration between symptom onset and institutionalization was more than twice as long for YOD patients (107 months) than for LOD patients (50 months) (U = 3441.0, p<0.001). Being in the YOD group was associated with an approximate 70% decrease in the likelihood of institutionalization compared with the LOD group (hazard ratio [HR]= 0.286; 95% CI:0.17-0.48) (p < 0.001; Table 2). This difference remained statistically significant after correcting for caregivers' gender, caregivers' relationship, diagnosis and patients' and caregivers' levels of education.

Table 2. Cox proportional hazard model of the time to institutionalization from symptom onset (YOD and LOD cohort)

	Hazard ratio	95% Confidence interval		Sign.
		Lower	Upper	
YOD group	0.286	0.171	0.480	*< 0.001*
Male caregiver	2.089	1.366	3.193	*0.001*
Spousal caregiver	0.447	0.261	0.766	*0.003*
Diagnosis				
Alzheimer's dementia (reference)				
Frontotemporal dementia	0.591	0.325	1.075	0.085
Mixed/Vascular dementia	0.508	0.261	0.987	*0.046*
Other dementias	1.517	0.732	3.144	0.262
Patient education				
Low (reference)				0.489
Average	1.139	0.588	2.206	0.700
High	1.387	0.750	2.563	0.297
Caregiver education				
Low (reference)				0.529
Average	0.750	0.420	1.341	0.332
High	0.959	0.578	1.590	0.870

Notes: entire model: -2 Log Likelihood: 1009.865 (p <0.001); YOD = Young-onset dementia

Predictors of time to institutionalization in YOD versus LOD

Cox proportional hazards showed that in the YOD group, the likelihood of institutionalization increased 8% with every one-point increase on the apathy score of the NPI (hazard ratio [HR]=1.081; CI: 1.008-1.159) (p < 0.05) (Table 3). No such relationship was found in the LOD group. No relationship was found with caregiver distress related to neuropsychiatric symptoms. However, the ability of the caregiver to deal with the caregiving role was a nearly significant predictor in the model (p= 0.061). Although the correlation between caregiver distress and the caregiver's sense of competence was low (0.313), caregivers sense of competence emerged as a significant predictor of institutionalization (hazard ratio [HR]= 0.955; CI: 0.917-0.995) (p = 0.029) when caregiver distress related to neuropsychiatric symptoms was omitted from the model. As no interaction effect between group and caregiver's sense of competence was found this relationship applied for both groups. Furthermore, institutionalization was nearly twice as likely for patients with a male caregiver (hazard ratio [HR]=1.88; 95% CI: 1.21-2.94) (p < 0.01). The risk for institutionalization decreased when the primary caregiver was a spouse (hazard ratio [HR]=0.348; CI: 0.144-0.843)(p<0.05). In the YOD group, patients with mild dementia had a decreased risk for institutionalization compared with patients with advanced dementia (hazard ratio [HR]=0.355; CI: 0.137-0.919) (p<0.05). Furthermore, in both groups, patients with frontotemporal dementia were less likely to be institutionalized compared with AD patients (hazard ratio [HR]=0.453) (p<0.05).

Table 3. Cox proportional hazard model of the time from inclusion to institutionalization (YOD and LOD cohort)

	Hazard ratio	95% Confidence interval		Sign.
		Lower	Upper	
YOD group	0.985	0.332	2.292	0.978
Caregiver age	1.007	0.976	1.039	0.675
Male caregiver	1.881	1.214	2.941	*0.005*
Spousal caregiver	0.348	0.144	0.843	*0.019*
Advanced dementia	0.786	0.368	1.678	0.786
Diagnosis				
Alzheimer's dementia (reference)				
Frontotemporal dementia	0.453	0.233	0.882	*0.020*
Mixed/Vascular dementia	0.602	0.280	1.295	0.194
Other dementias	1.282	0.601	2.737	0.520
Neuropsychiatric symptoms (NPI)				
Hyperactivity	1.005	0.977	1.034	0.722
Psychosis	0.911	0.950	1.033	0.665
Mood	0.998	0.956	1.042	0.937
Apathy	0.595	0.901	1.019	0.178
Caregiver distress (NPI-D)	1.011	0.975	1.048	0.559
Caregiver sense of competence (SSCQ)	0.959	0.918	1.002	0.061
Time from symptom onset until inclusion	1.000	0.995	1.005	0.951
Interaction Group with dementia severity	0.355	0.137	0.919	*0.033*
Interaction Group with Apathy score	1.081	1.008	1.159	*0.029*

Notes: entire model: -2 Log Likelihood: 610.388 ($p < 0.001$); YOD = Young-onset dementia

Discussion

Contrary to our expectations, we found that the time from symptom onset to institutionalization was more than twice as long for YOD patients (nine years) compared with LOD patients (four years). Our hypothesis that both neuropsychiatric symptoms and the related distress would predict institutionalization in YOD was not fully confirmed. Only apathy emerged as a significant predictor of the time from inclusion to institutionalization. Contrary to our expectations the distress related to neuropsychiatric symptoms did not significantly predict institutionalization.

We did not expect such a large delay in institutionalization in YOD. Luppa et al. reviewed studies on institutionalization in LOD and found a duration of 2.5 to approximately 3.5 years.[1] Differences in the results of these studies are related to varying methodological approaches, as most of the studies reported the median time from study entry to institutionalization without information on symptom onset. Most studies used disease severity to correct for this problem, with only a few studies including the time from symptom onset to diagnosis in the analyses. A study that explored only time to institutionalization in incident dementia cases reported a duration of 33.5 months,[28] which is similar to the present findings regarding the LOD group. The apparent delay in YOD institutionalization in the current study might be due to methodological differences or national differences in the way community services are organized.

The present results suggest that in YOD, the caregiving trajectory is significantly extended compared with LOD. This might be related to the fact that YOD patients and their caregivers have a better physical health compared to LOD patients and their caregivers. However, to our knowledge currently there are no studies that included medical comorbidity in their comparison of YOD and LOD patients. Furthermore, research regarding the relationship between the physical health of dementia patients and their caregivers and institutionalization in dementia is inconclusive.[1, 29] Our findings might also be explained by the fact that functional problems might be detected at an earlier stage of the disease in YOD patients compared with LOD patients, as younger patients experience greater challenges in daily life. This is supported by earlier research that demonstrated that YOD patients were less severely impaired at the time of diagnosis than LOD patients,[30] even though there is a significant delay in the time to diagnosis in YOD.[31] The first subtle signs and symptoms of disease do not lead to a diagnosis at that time. However, caregivers will likely retrospectively recall these changes as the time of disease onset. Another explanation for the delay in institutionalization may be the lack of age-appropriate residential facilities, differences in the progression of YOD or the stigma attached to dementia.[12] One may argue that a large proportion of YOD patients in the current study attended a specialized day care center, which could have delayed institutionalization.[29] However, this is unlikely given the delay in diagnosis and subsequent delay in service use in the sample, as reported previously.[12, 32] A delay in service use has been suggested to increase the risk of institutionalization, as caregivers are likely to become overburdened and nursing home placement becomes inevitable.[33]

Neuropsychiatric symptoms, especially aggression, depression and hallucinations, are known predictors of institutionalization in LOD.[1, 29] However, in the YOD group, only apathy emerged as a significant predictor of time to institutionalization. We found significantly higher levels of both apathy and hyperactivity symptoms and significantly lower levels of mood symptoms in the YOD group compared to the LOD group. This result may be due to the higher proportion of frontotemporal dementia in the YOD group, as apathy and hyperactivity are often key symptoms in FTD.[34] An explanation for apathy predicting institutionalization may be that in YOD patients' apathy results in an increase in the informal care demands on the primary caregiver as a result of the patients' lack of initiative and increasing dependency. The caregiver may not be able to meet these demands due to the double burden of caring while maintaining other responsibilities such as work or parenting. Moreover, previous research suggests that apathy is associated with a deterioration of the marital relationship,[35] and higher levels of caregiver distress.[34] Increasing the risk for institutionalization, as caregivers may find it difficult to care for a patient in the absence of reciprocity.[36, 37] However, the current study did not find that caregiver distress per se predicted the time to institutionalization.

Although neuropsychiatric symptoms are common in YOD,[4] as supported by the present findings, Gaugler et al. (2010) suggest that a persistent occurrence of neuropsychiatric symptoms is less likely to lead to an increase in caregiver distress than newly occurring problems.[8] YOD caregivers are exposed to neuropsychiatric symptoms for a long period of time,[11] which might lead to a decrease in behavior-related distress. The current findings suggest that the caregivers' feelings of competence to care may be a more reliable determinant of YOD patients' risk for institutionalization. Thus, the further development of interventions aimed at enhancing caregivers' sense of competence is warranted.

The current study also showed that in the YOD group, the probability of institutionalization was lower for FTD compared to AD patients. AD is a strong predictor of institutionalization in LOD.[1, 29] Furthermore, FTD patients may show less functional dependency during earlier and moderate stages of the disease compared to AD patients and also resist the initiation of services including institutionalization, which will likely delay the decision to institutionalize these patients. In the YOD group, advanced dementia emerged as a predictor for institutionalization, which is in line with previous research.[1, 29] We also found that the caregiver's relationship to the patient and the caregiver's gender predicted institutionalization in both samples. Spousal caregivers were less likely to institutionalize the care recipient in our sample, which is inconsistently supported by previous studies.[29] In addition, the risk of institutionalization more than doubled for patients with a male caregiver, which is in line with earlier research.[1]

Strength and limitations

The current study is the first to comprehensively assess the time to institutionalization in a relatively large sample of YOD patients in comparison with LOD patients. The study provides important insights in the predictors that increase the risk for institutionalization in YOD. However, several limitations should be considered. Because dementia commonly has an insidious onset, it is difficult to accurately ascertain symptom onset.[38] An important and inevitable limitation of the study is the retrospective estimation of the time of symptom onset, which could bias study outcomes. In the YOD group, the time from symptom onset to institutionalization may have been overestimated. In this group, the first month of the year of symptom onset was used in the analyses, whereas symptom onset was estimated by subtracting disease duration until diagnosis from the date of diagnosis in the LOD group. However, this difference in estimation cannot explain the large difference found between the groups. We also included patients from different phases of the caregiving trajectory. This inevitably leads to differences in the length of time from symptom onset of the dementia to inclusion in our study, likely biasing our analysis. We have controlled for this issue by including the time from symptom onset to inclusion in our analysis. Given the fact that this appeared not significant in our model we can assume that this has not influenced our model. Furthermore, the recruitment strategy for the study groups was slightly different, which may have also biased the results. The LOD group was recruited through memory clinics and psychiatric services, whereas the YOD patients were recruited through memory clinics and specialized day care centers. In the YOD group 115 patients attended a specialized day care centre while for the LOD group information about how many patients were involved in day care and for how long was not available. Furthermore, the number of patients in some of the diagnostic categories was small, limiting the generalizability of our findings for the entire YOD population. A selection bias is also likely because we enrolled patients who sought care and provided consent for participation. The patient – caregiver dyads that were not included in the study or did not consent may exhibit greater severity of neuropsychiatric symptoms and caregiver distress. Finally, in addition to the factors we included in our analyses also the patients' and caregivers' physical and mental health as well as contextual factors (e.g., waiting lists, financial status and facility characteristics) are also likely to influence the decision to institutionalize; however, these were not included in the analyses, but should be considered in future research.

Conclusions

The findings of this study have important implications for service provision given the prolonged period that YOD patients are cared for at home. The results stress the importance of a timely diagnosis for these patients and their families to facilitate the initiation of appropriate care and support.[32] Early introduction of, for example, case management within the context of a multidisciplinary YOD care program may ameliorate transitions in care and enhance the caregivers' sense of competence in dealing with the caregiving situation.[39, 40] In YOD, providing the caregiver with support in dealing with neuropsychiatric symptoms, especially apathy, may increase caregiver competence and postpone the institutionalization of the patient.[41]

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8

General discussion

Summary

Clinical implications

Future directives

Introduction

The studies described in this thesis are part of a large prospective, multi-centre study, the Needs in Young-onset Dementia (NeedYD)-study (Chapter 2). In this final chapter, the main results of the studies presented in this thesis (Chapter 3-7) will be summarized and discussed. Also the methodological strengths and limitations will be considered. Finally, implications and recommendations for clinical practice and future research are provided.

Summary of findings

(1) *What are the care needs of young-onset dementia patients?*

We found that when dementia occurs at a young age it causes additional concerns and challenges in the caregiving situation, hampering the extent to which needs of both patient and caregiver are met. These concerns result from the young and active life phase in which patients and their caregivers are confronted with an invalidating degenerative disease. Furthermore, due to the low prevalence of YOD health care professionals do not often encounter these patients, hampering the diagnostic process as well as causing specific issues during the caregiving trajectory. Our case study (chapter 3) showed that a delay in diagnosis was perceived as an important obstacle to initiate appropriate help and support, leaving care needs of patient and caregiver unmet. Unfortunately, this delay in diagnosis is common in YOD, [1, 2] suggesting that a delay in service use and sub-sequential high prevalence of unmet needs in the entire YOD population is likely to occur. In YOD, caregivers are confronted with a double strain of providing care while at the same time fulfilling other active and necessary roles like being a parent, financial provider and managing the household (chapter 3). This shift in responsibilities likely adds to the burden felt by young caregivers and puts an important strain on their own future perspective (chapter 3).

Needs occur in many different areas in YOD and these needs are highly heterogeneous (chapter 4). There was fair agreement between patients and caregivers on the areas in which a need occurred, however agreement on the total number of (un)met needs was only modest. These findings suggest that the difference between the patient and caregiver perspective on patient care needs found in LOD[3, 4] also applies for YOD. Both patients and caregivers reported high proportions of unmet needs at baseline, 18% and 24% respectively. The highest proportions of unmet needs were reported by YOD caregivers in the areas of daytime activities, social company, eyesight and hearing, memory, mobility and psychological distress. Patients additionally mentioned unmet information needs and unmet needs regarding intimate relationships among the most common unmet needs. During the two-year follow-up the proportion of unmet needs decreased with 8-10% respectively, with an obvious similar increase in the proportion of met needs, suggesting that during the course of the disease appropriate help and support was initiated. However, a relatively high proportion (10-14% respectively) of care needs remained unmet at the end of our study.

(2) *What are the consequences of unmet care needs on the course of neuropsychiatric symptoms and health related quality of life in YOD?*

We did not find a relationship between unmet needs and patient HRQoL (Chapter 5). However, we found that the occurrence of unmet needs in the patient adversely affected caregiver HRQoL. Patient unmet needs were associated with caregivers reporting a loss of vitality and perceived general health, next to higher levels of physical pain and difficulties in performing daily tasks due to physical problems. Also, the unmet psychological needs of the caregivers themselves were found related to caregivers experiencing more limitations in social functioning as well as higher levels of depression and anxiety. The use of supportive services by the caregiver was associated with caregivers experiencing fewer limitations due to emotional and physical problems but at the same time associated with caregivers reporting lower scores on other HRQoL domains (chapter 5). These findings suggest that the use of supportive services, such as psychosocial support or respite care may be beneficial to caregivers. Nonetheless, indicating that supportive services might not counter the negative impact of the double strain of providing high levels of informal care while performing other necessary roles in YOD (chapter 3 and chapter 6). Research in LOD patients shows that when health care services are initiated early in the disease process it is most likely these services can be of benefit to them.[5, 6] Implicating that this might prove an important challenge in YOD, considering the delay in services use in these patients (chapter 6).

Our findings also indicate that an increase in the number of unmet needs may lead to the occurrence or worsening of neuropsychiatric symptoms. In contrast, it is also possible that high levels of neuropsychiatric symptoms hamper care provision in these patients, decreasing the likelihood that care needs will be fulfilled. Sensitivity analyses revealed that the effect of unmet needs applied to all four behavioural sub-syndromes (psychosis, hyperactivity, mood and apathy). Our findings are consistent with previous research in LOD patients, showing that unmet needs were associated with neuropsychiatric symptoms, such as anxiety and depression,[7] agitation[8, 9] and resistiveness to care.[10] Contrary to our expectations, we also found an effect of met needs on the occurrence of neuropsychiatric symptoms over time. This finding suggests that the relationship between (un)met needs and neuropsychiatric symptoms is indeed bi-directional in nature. It may also indicate that appropriate help and support with care needs do not necessarily counter the overall adverse effect of dementia on patients with YOD. As in YOD patients, the loss of functional abilities, increasing dependency and the consequential loss of various roles within the family and society lead to an important loss of the sense of self, self-esteem and identity.[11] The relationship between (un)met needs and neuropsychiatric symptoms we found has important clinical implications as neuropsychiatric symptoms have been found to predict cognitive and functional decline in LOD patients,[12] as well as high levels of caregiver distress,[13, 14] and institutionalization.[12, 15]

(3) *What factors are related to the use of (in)formal care and the time to institutionalization in community dwelling young-onset dementia patients?*

In chapter 6 we found that caregivers provide most of the care in YOD, with an informal and formal care ratio of 3:1. In more than half of all cases all informal care was provided by only one caregiver, most often the spouse or a child and in a minority of cases by another caregiver. This might, at least to some extent, explain the high levels of caregiver burden found in caregivers of YOD patients.[16, 17] We found that higher levels of informal care use were associated with advanced disease severity. Caregivers of the youngest patients provided fewer amounts of informal care, which was related to a higher number of working hours. No relationship was found between caregiver factors and the use of formal health care services. Increased formal care use however, was found to be related to patient factors such as advanced dementia severity, higher levels of neuropsychiatric symptoms as well as lower levels of patient ADL initiative. These findings suggest that YOD caregivers delay the initiation of formal services until late in the course of the caregiving trajectory, when informal care options have run out. The case study (chapter 3) showed that the caregiver felt that the use of dedicated health care services, such as specialized day care, intermittent stays at the nursing home and psychosocial support were beneficial to her and her husband and contributed to a delay in institutionalization.

In YOD the caregiving trajectory is significantly longer compared with LOD, with the time from symptom onset to institutionalization being more than twice as long in YOD (nine years) compared to LOD (four years) (chapter 7). In the YOD group apathy appeared an important predictor of the time to institutionalization. The probability of institutionalization was significantly higher for those patients exhibiting higher levels of apathy symptoms. Also patient diagnosis predicted institutionalization in both groups, with frontotemporal dementia associated with a decreased likelihood of institutionalization. In both the YOD and LOD group lower levels of caregiver's sense of competence in dealing with the caregiving situation, male gender of the caregiver and the caregiver being a child were found associated with an increase in the likelihood of institutionalization.

Methodological considerations

Sample selection

Because of our sampling strategy (through existing health care services, such as memory clinics and YOD day care centres) our sample may not be representative of the entire YOD population. Only patients and caregivers who sought care and provided consent for participation were enrolled. Patient-caregiver dyads that were consequently not included might very well differ in terms of care needs, neuropsychiatric symptoms or caregiver distress. Also no patients with alcohol related dementia, HIV/Aids related dementia complex, Huntington's disease, traumatic brain injury or Down's syndrome were included. Therefore the results of this study may not be valid for all community dwelling YOD patients and their caregivers. However, our findings are very relevant for clinical practice because we were able to recruit a relatively large sample of YOD patients and caregivers across various settings in spite of the low prevalence of YOD and difficulties in diagnosing these patients.

To investigate possible differences in the time to institutionalization between YOD and LOD patients we compared our data with data of the MAAstricht Study of BEhaviour in Dementia (MAASBED) study. The NeedYD study was based on the methods of the MAASBED study, using a similar design, assessment instruments and diagnostic criteria to enable the comparison of the various outcomes. However, the recruitment strategy for the study populations was slightly different, which may have biased results. Although both studies sampled from memory clinics and community mental health care facilities after a diagnosis had been established, the NeedYD study also included patients who attended day care centres that provided specialized care for YOD patients. This difference may have influenced the distributions of for example neuropsychiatric symptoms and disease severity, overestimating the differences between groups.

An inevitable issue in longitudinal research concerns a loss to follow-up. At 2-year follow-up 25.8% of patient-caregiver dyads were lost to follow up. Although no differences were found on important outcome variables and patient and caregiver characteristics, patients who were lost to follow-up had more severe dementia at baseline than those who remained in the study. This suggests a selective attrition, with an overrepresentation at the later assessments in our study of less severely impaired YOD patients. In our longitudinal analyses we used linear mixed models. Mixed models analysis allows for an estimation of an intercept and slope for each participant based on all available data for that individual, augmented by the data from the entire sample. Because missing data at one or more time points are common in prospective studies, the use of mixed models allows individual slopes to be estimated using the available data for that participant. It is unlikely that the attrition bias affects our main conclusions, especially considering the strong relationship we found between (un)met needs and neuropsychiatric symptoms after controlling for dementia severity. Despite this limitation the strength of this longitudinal design is that we were able to study changes in various outcomes over time allowing us to also make inferences about cause and effect.

Assessment instruments

We used the Global Deterioration Scale (GDS) to establish dementia severity because it assesses both cognitive and functional impairment in patients. However, because the GDS also addresses behavioural changes (delusions, anxiety and aggression) in the more advanced disease stages this could have led to an over-inclusion in our analyses of the course of neuropsychiatric symptoms of people with these behavioural symptoms in the later stages. Furthermore, the GDS, as most staging scales, has originally been designed to establish dementia severity in patients with Alzheimer`s disease, indicating that it might be less valid for the assessment of dementia severity in other diagnostic categories. However, deterioration in dementia is often reflected by changes in patient functioning, cognitive abilities and behaviour. This suggests that the assessment of dementia severity should comprise an instrument that covers all of these domains. As the GDS is a global assessment, with broad descriptions and criteria we preferred to use this particular assessment instrument.

We assessed neuropsychiatric symptoms using the Dutch version of the NPI,[18] which is a broad instrument for the assessment of most, but not all possible neuropsychiatric symptoms. The NPI for instance does not assess sexual problems,[19] nor does it assess self-injurious behaviour,[20] loneliness or boredom. From our clinical experience we know that these problems are more likely to occur in young, vital persons with dementia. Therefore, the use of the NPI may have resulted in an underestimation of the prevalence of neuropsychiatric symptoms in our sample. Also, the NPI only globally assesses aggression and agitation symptoms, suggesting that given the high prevalence of these symptoms in our sample adding the Cohen Mansfield Agitation Inventory (CMAI) would have allowed for a more differentiated insight in these symptoms in YOD. Furthermore, in our study we used the scores on behavioural sub-syndromes instead of the total score of symptoms. Although this results in a data-reduction of the twelve symptoms of the NPI it also allowed for a more thorough investigation of for example the differential effect of unmet needs on behaviour. There is evidence that the sub-syndromes we used for our analyses are consistent for the different dementia diagnoses in LOD patients.[21] This would suggest that this also applies for YOD, however also stressing that some caution should be used when interpreting our findings.

An important consideration concerns the use of patients' self-report versus the caregiver report in the assessment of quality of life. In more advanced stages of the dementia it will become increasingly difficult for dementia patients to reflect on issues that affect their quality of life. There are known differences between caregiver proxies and subjective experiences of dementia patients regarding their health related quality of life when using the various assessment instruments that are currently available.[22] We chose to use the patient report of the quality of life scale because research suggests that in dementia and even in more advanced stages of the dementia the patient self-report of the QOL-AD is reliable and the self-report should therefore be considered the golden standard.[22] Of our sample 78.5% of patients were able to complete the quality of life scale. For the QOL-AD non-completers were more likely to be female or to have a higher disease severity. This might have biased our results, because these patients may very well experience lower levels of HRQoL, resulting in an overestimation of HRQoL in our group. This might also suggest that there are gender differences that should be addressed when assessing HRQoL in YOD. We believe that despite these limitations we were able to explore a relatively unknown area of YOD outweighing the potential bias that may have occurred.

A similar issue relates to the decision whether or not to use the patient self-report of the Camberwell Assessment of Need in the Elderly (CANE) in our analyses. One could argue that in the assessment of care needs the patient self-report should be preferred and not the caregiver proxy like we did, because caregivers might overestimate unmet care needs, for instance when experiencing high levels of caregiver burden. However, almost 30% of our patients were unable to complete the needs interview. The CANE is a rather long and demanding interview in which patients spontaneously have to reflect on the difficulties they experience in different areas of their lives and recall whether they receive help in that area, the quantity and kind of help they receive, whether or not this is enough and if additional or other help is necessary. We experienced during the interviews that patients had difficulties in reflecting on different aspects of the 24 domains of the CANE, that they were unable to recall the help they actually received, and that they experienced difficulties in reflecting on the kind of help they wanted to receive. Our experiences are supported by a review of van de Roest et. al. (2007), and papers from Orrel et. al. (2008) and van de Roest et. al. (2009) that found that 30% - 40% of the patients in their samples were not able to complete all items of the CANE and that this was amongst other things related to increased cognitive impairment and levels of dependency.[3, 4, 23] Considering the explorative nature of our study in which we included patients from various phases of the caregiving trajectory, we chose to use the caregiver view to get a complete picture of all domains in which unmet needs might exist.

Implications

Implications for clinical practice

Currently an estimated 35.6 million people are affected by dementia and this number will double by 2030 and even triple by 2050.[24] This means that in the forthcoming years not only the disease will affect an increasing number of people but also the societal and economic burden of the disease will increase accordingly. There is an increasing awareness among health care professionals that when the dementia occurs at a younger age this causes specific challenges and issues,[25] with important consequences for service provision. However, considering the low prevalence of YOD, with an estimated 6-9% of patients affected by the disease before the age of 65,[24] the needs and wants of these patients and their families might be easily overlooked. The issue of YOD should therefore be included in national dementia programs, similar to the French National Dementia Program. The French program contains two specific research objectives regarding the improvement of both the diagnostic process and the management of YOD. When the specific concerns regarding YOD and related research objectives are included in national dementia programs this might reduce the risk that governments and policy makers will underestimate the economic and societal burden of YOD.

Our findings underline the importance of providing general practitioners and occupational physicians with guidelines for the early detection of probable YOD as they are most likely to encounter these patients in the early stages of the disease. Earlier findings of the NeedYD study showed that the diagnostic phase in YOD is lengthy and problematic, resulting in a mean of 4.4 years until a diagnosis can be established compared to a mean of 2.8 years in LOD.[1] The education of general practitioners and occupational physicians about the specific early signs and symptoms of the dementia in YOD and how these should be addressed[2] could significantly raise awareness of YOD and probably shorten the diagnostic phase in YOD patients. Findings of this thesis suggest that the problematic diagnostic phase in YOD results in an important delay in the use of formal health care services in YOD, stressing the importance of a timely diagnosis. However, our findings also indicate that even when a proper diagnosis has been established, YOD caregivers postpone service use until this becomes inevitable. The problem with delayed service use is that issues related to the dementia, such as functional problems, neuropsychiatric symptoms and caregiver burden will eventually become more severe and complex to solve.[5, 6] The diagnostic phase in YOD should therefore also include the introduction of a specialized YOD casemanager who can provide psychosocial support as well as support the family in advance care planning. The latter can be established through regular discussions with patients and their caregivers about which factors influence their quality of life, what (in)formal care options are available within the context of the current disease phase as well as their uncertain future perspective. This underlines the importance that the casemanager remains involved with the family throughout the entire caregiving trajectory to allow for continuity in care as well as a timely recognition of issues that may adversely affect the caregiving situation such as relationship problems, financial difficulties or difficulties in managing neuropsychiatric symptoms. The early introduction of casemanagement in the caregiving trajectory might even result in a reduction of service use in later stages of the caregiving trajectory and a sub sequential reduction in care expenditures in YOD.

Our findings also underline the importance of educating general practitioners and occupational physicians how to support and properly refer YOD patients and their family members who turn to them with mental or physical complaints likely related to the caregiving situation. There is an evident loss of productive years in these young patients as they are affected at a time they are still working. Our findings indicate that this might also apply for YOD caregivers given the lengthy caregiving trajectory, in which high levels of informal care are provided by caregivers themselves and the sub-sequential double burden these caregivers are faced with. High levels of informal caregiving have been found associated with decreasing health and well-being of dementia caregivers, especially when the care duration is lengthy.[26, 27] The double burden of providing high levels of informal care for an extended period of time while performing other responsibilities such as working, parenting and running the household may force YOD caregivers to eventually reduce their working hours, take sick leave or stop working altogether. This will add to the financial difficulties these families already have to face,[25] but will also cause an important loss in productive years in these caregivers. Sustaining a job will allow for the caregiver to somewhat distance themselves from the caregiving situation, regain their strength and prevent them from becoming more and more isolated in the community.

In the Netherlands there is an increasing tendency to integrate hospital based diagnostic services with community services for people with dementia. However, due to the low prevalence of YOD these diagnostic and community services are unlikely to be familiar with the needs and wants of these younger patients and their families and have less resources to meet their needs.[28] Also when diagnostic services do specialize in YOD this does not necessarily coincide with an appropriate follow up of care and support. Some researchers have proposed that in YOD providing dedicated services is the way to go.[29-32] We agree that dedicated services, integrating diagnostic and community services might more easily allow for more personalized service provision in YOD. It will prove important to offer a range of multi-component interventions, targeting both the patient and the caregiver, which can be used throughout the entire caregiving trajectory, considering the extensive period of time these patients are cared for at home. Despite similarities in the areas of life in which needs occur in YOD and LOD, we found high proportions of unmet needs in various areas of life as well as unmet needs in which the high proportion appears specifically related to YOD, such as the need for information about the disease and care options and the need for intimate relationships. The care needs we found are at least to some extent amendable to interventions. For example the need for meaningful daytime activities and social contact could be addressed through specialized day care. Meeting other peers who encounter the same issues and who have similar needs and wants might improve patient wellbeing. Given the nature of the other unmet needs that our study revealed (i.e. difficulties in communicating, memory problems, mobility issues and psychological distress) YOD services should also comprise a multi-disciplinary team of for example occupational therapists, physiotherapists, speech therapists and psychologists to be able to address these needs as they occur. Our findings also suggest that health care professionals should aim at enhancing the caregivers' sense of competence in dealing with the caregiving situation and neuropsychiatric symptoms, in particular apathy, as this may significantly delay institutionalization. From our clinical experience we know that support groups, in which these issues are discussed with other caregivers in the presence of the casemanager and a psychologist, are beneficial. The experiences during these sessions can afterwards be used to support the caregiver in the decision making process regarding the caregiving trajectory. This illustrates the importance of an integrative approach to the management of YOD, in which interventions can be combined to fit the individual needs of these families, and with that, enhance the effect of these interventions. Health care professionals should focus on empowering YOD patients and caregivers in retaining control over the caregiving situation and designing services that can complement informal care. When dedicated services are difficult to establish, for example in low populated areas, policymakers and health care professionals should at least aim at integrating existing health care services to ensure better continuity in care for these younger families.

Considering that (unmet) needs are diverse in the YOD population and change during the caregiving trajectory the periodical assessment of (un)met needs, for instance using the Camberwell Assessment of Need in the Elderly (CANE) should be an integral part of caregiving in YOD. Within the context of providing anticipatory care a thorough assessment of (un)met patient and caregiver needs is a prerequisite for a more personalized approach in the management of YOD. In the assessment of (un) met needs the subjective experiences of patients and the experiences of their caregivers should be regarded complementary, because of differences in their views on which needs should be prioritized and whether a need is met. Given the adverse relationship between unmet needs and the course of neuropsychiatric symptoms and caregiver HRQoL it is important that the number of unmet needs in YOD is reduced. Educating YOD caregivers about the risk of high levels of unmet needs and discussing with them at what time professional support might complement informal care and be most beneficial will enable them to retain control over the caregiving situation. Special attention should be paid to the issue of patient acceptance of services. This can be achieved by the exploration, together with the patient, how services and support could be beneficial to them. It is important that this exploration takes place within the context of the patients own needs and wants as early as possible in the course of the disease as patients are more likely to be able to reflect on their situation and concerns for the future. One should also discuss which services or support could be initiated in order to avoid things the patient might be afraid of, such as the loss of autonomy and institutionalization. This will also likely prove important to raise patients' acceptance of services.

Implications for research

Although the studies described in this thesis provide important first insights into care needs and the caregiving trajectory in YOD there are still many different aspects that deserve more attention in future research.

Data regarding the prevalence of YOD and the different causes of dementia in younger patients for most countries is still scarce and inconsistent. This underlines the need for a large community based, Dutch prevalence study because this data is essential for assessing the impact of YOD on society and future care planning. Moreover, there is concern among health care professionals about a possible increase in the incidence of YOD with the increase of known risk factors for dementia such as obesity, diabetes and cardiovascular disease in developed countries. Midlife obesity, high (systolic) blood pressure and high total cholesterol level have been associated with increased risk for developing dementia, at least in older persons.[33-35] This warrants further epidemiologic research on the possible causal pathways of these risk factors and the occurrence of dementia at a younger age.

When dementia occurs at a younger age this touches the lives of all members of the family. In this thesis we investigated characteristics and care needs of YOD patients and the primary caregiver. Clinical experience shows that also other family members of YOD patients, such as children, parents or siblings experience difficulties in for example dealing with feelings of grief or managing functional and behavioural changes. This may even raise the boundaries for these family members to become more actively involved in the caregiving process, which is undesirable considering the limited social support system in YOD. Data regarding the course of needs and wants of other family members in YOD is largely lacking, suggesting this should deserve more attention in future research.

Our findings indicate that neuropsychiatric symptoms are common in YOD, although our earlier research shows that there are differences in the prevalence of neuropsychiatric symptoms when comparing young-onset and late onset Alzheimer's disease. In our study we specifically looked at the behavioural sub-syndromes of the neuropsychiatric inventory instead of the total score of symptoms. Our findings indicate that there are differences between YOD and LOD patients in which sub-syndromes are most prevalent, as we found that apathy as well as hyperactivity symptoms appear to be significantly more prominent in YOD compared to LOD. There is strong evidence that the sub-syndromes we used for our analyses are consistent for the different dementia diagnoses in LOD.[21] However, more research is needed to investigate whether the sub-syndromes are also consistent across dementia diagnoses and the separate dementia stages in YOD, especially considering the heterogeneity of dementia causes in YOD.[2]

As we have seen our findings suggest that policy makers and health care professionals should aim at the establishment of dedicated services in YOD, targeting both patient and caregiver. These services should focus on empowering YOD patients and caregivers in retaining control over the caregiving situation and designing services that can compliment informal care. Although the studies presented in this thesis provide important first insights into what services could be beneficial to YOD patients and their caregivers, little is known about proper timing of available services and which services are most beneficial at different stages in the caregiving trajectory. Also the efficacy of interventions specifically designed for YOD patients and their caregivers, such as specialized day care, YOD casemanagement or YOD support groups should be evaluated in further research. Longitudinal research can provide more insight in these issues and advance care planning in YOD. Our research shows that in the evaluation of services in YOD (un)met needs and HRQoL should be considered important outcomes.

Conclusions

Young dementia patients and their caregivers should not be overlooked in dementia care. The studies presented in this thesis suggest that in the management of YOD an integrative approach to caregiving is essential. This can be established through the initiation of dedicated YOD services or a better integration of existing mental health care services. A thorough understanding of (un)met needs of these patients and their caregivers by health care professionals should be considered a prerequisite for establishing appropriate help and support. A better understanding of the needs and wants of these families will allow health care professionals aim at empowering these patients and caregivers in retaining control over the caregiving situation and designing services that can compliment informal care.

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9

Summary

Samenvatting

Summary

Dementia is typically regarded as a disease of old age and as a result dementia health care services are designed to address the needs of these older persons and their families. However, in approximately 6-9% of dementia cases the onset of the disease is before the age of 65 years. This has important implications for service provision, as mainstream dementia services are known to have difficulties in addressing the specific needs and wants of these younger individuals and their families. Little is known about care needs and service provision in young-onset dementia (YOD). Therefore the aim of this thesis is to address three particular aspects of caregiving in YOD: (1) care needs of persons with YOD and their caregivers (chapter 3 &4), (2) the impact of unmet care needs on the course of neuropsychiatric symptoms and health related quality of life (chapter 4 & 5), and (3) factors related to the use of (in) formal care and institutionalization (chapter 6 & 7).

The studies presented in this thesis are part of a prospective cohort study, the Needs in Young-onset Dementia (NeedYD) study. In the NeedYD-study 215 community-dwelling YOD patient-caregiver dyads are assessed every six months during a two-year period investigating the course of the dementia, clinical characteristics (i.e. cognitive functioning, neuropsychiatric symptoms and quality of life) and the course of care needs. The methodology of the NeedYD-study is discussed in chapter 2 of this thesis.

The study rationale and research questions are provided in the introduction of this thesis (Chapter 1). In chapter 3 we describe the course of care needs and experiences with health care services using a single case study. In this study we perform an inductive content analysis on the interviews held with the spouse of a young person with Alzheimer's disease. An inductive content analysis can be used to understand relationships within the narratives and generate mayor themes in the caregiving process concerning problems met, care needs and experiences with health services. Findings indicate that these relatively young individuals and their caregivers are confronted with specific issues during the care process, namely (1) a prolonged time to diagnosis, (2) a lack of fit between needs and services, (3) the strain of dedication to care versus the caregiver's own future perspective, and (4) the need for a response of health care services to changing individual preferences.

In chapter 4 we more closely explore (un)met care needs in the entire YOD sample as well as the level of agreement within care recipient-caregiver dyads on (un)met care needs. Additionally, we investigate the longitudinal relationship between care needs and neuropsychiatric symptom levels. Findings indicate that care recipients and caregivers generally agree on the areas in which needs occur. Only modest agreement exists within dyads regarding whether needs can be met. Our findings indicate that in YOD there are specific areas of life in which unmet needs are more likely to occur. Persons with YOD experience high levels of unmet needs in areas such as daytime activities, social company, intimate relationships and information. There is also a strong relationship between both met and unmet needs and neuropsychiatric symptoms over time. The high proportions of unmet needs and the relationship between (un)met needs and neuropsychiatric symptoms warrant interventions that target neuropsy-

chiatric symptoms as well as the prevention of unmet needs. This underlines the importance of the periodic investigation of care needs, in which care recipient and caregiver perspectives are considered complementary.

In chapter 5 we perform multiple linear regression analyses to determine the relationship between unmet needs and HRQoL, controlling for other known associations. We find no relationship between unmet needs and HRQoL of the person with the dementia. However, the occurrence of unmet needs in the care recipient adversely affects caregiver HRQoL. Unmet needs of the care recipient are associated with caregivers reporting a loss of vitality and perceived general health, next to higher levels of physical pain and difficulties in performing daily tasks due to physical problems. Also, the unmet psychological needs of the caregivers themselves are related to caregivers experiencing more limitations in social functioning as well as higher levels of depression and anxiety. The use of supportive services by the caregiver is associated with caregivers experiencing fewer limitations due to emotional and physical problems but at the same time associated with caregivers reporting lower scores on other HRQoL domains. These findings suggest that the use of supportive services, such as psychosocial support or respite care may be beneficial to caregivers. Therefore, the assessment of unmet needs within the context of HRQoL appears to be an important prerequisite for personalizing care in YOD. Adjusting supportive services to match the individual needs and preferences of these young individuals and their caregivers is likely to enhance their quality of life.

In chapter 6 the use of (in)formal care prior to institutionalization, and its association with care recipient and caregiver characteristics are explored. In YOD informal care has a 3:1 ratio with formal care. Supervision/surveillance constitutes the largest part of informal care. In more than half of cases the person with YOD has only one informal caregiver. Furthermore, our findings show that the amount of informal care is associated with disease severity, showing more informal care hours in advanced disease stages. Fewer informal care hours are related to more caregiver working hours, especially in younger care recipients. The amount of formal care is related to disease severity, behavioral problems and initiative in ADL functioning. In YOD it appears that family members provide most of the care. However, other social roles still have to be fulfilled. Especially in spousal caregivers of younger care recipients in advanced disease stages there is a double burden of work and care responsibilities. This finding also indicates that even within the YOD group there might be important age related differences. The relatively higher amount of formal care use during advanced disease stages suggests a postponement in the use of formal care.

In chapter 7 we investigate the time from symptom onset to institutionalization in persons with YOD compared to LOD. We also determine which factors predict institutionalization in persons with YOD compared with LOD. Our findings show that the time from symptom onset to institutionalization is nearly nine years for persons with YOD compared to approximately four years in LOD. In the YOD group, Cox proportional hazard models show that apathy significantly predicts the time to institutionalization. Furthermore, the caregiver’s competence in caring for the person with dementia significantly predicts the time to institutionalization in both groups. These findings underline the importance of a timely diagnosis in YOD to facilitate the initiation of appropriate care and support, especially given the long period these younger individuals are cared for at home. Support programs aimed at enhancing the caregivers' sense of competence and ability to deal with neuropsychiatric symptoms, especially apathy, may reduce the risk for institutionalization in YOD.

In the final chapter, the main findings of this thesis as well as methodological considerations and implications for clinical practice and future research are discussed.

Samenvatting

Dementie wordt algemeen beschouwd als een ouderdomsziekte. De dementiezorg is derhalve voornamelijk gericht op het voorzien in de (zorg)behoeften van ouderen met dementie en hun familieleden. Er is echter een belangrijke groep mensen waarbij de eerste verschijnselen van de dementie voor het 65^{e} levensjaar aanvangen. Geschat wordt dat het hier gaat om 6-9% van het totaal aantal mensen met dementie. Dit heeft belangrijke consequenties voor de klinische praktijk aangezien de ervaring leert dat reguliere dementievoorzieningen onvoldoende lijken toegerust om in de specifieke (zorg)behoeften van deze jonge mensen met dementie en hun gezinsleden te voorzien. Er is echter weinig bekend over zorggebruik en -behoeften bij deze doelgroep. De centrale doelstelling van dit proefschrift is dan ook om drie specifieke aspecten in de zorg voor jonge mensen met een dementie en hun mantelzorgers nader in kaart te brengen en wel: (1) zorgbehoeften van jonge mensen met een dementie en hun mantelzorgers (hoofdstuk 3 & 4), (2) de impact van behoeften waarin niet kan worden voorzien op het beloop van neuropsychiatrische symptomen en ervaren kwaliteit van leven (hoofdstuk 4 & 5), en (3) voorspellers van het gebruik van (in)formele zorg en opname in het verpleeghuis (hoofdstuk 6 & 7).

De afzonderlijke studies in dit proefschrift maken deel uit van een uitgebreidere prospectieve studie, de Needs in Young-onset Dementia (NeedYD) studie. In de NeedYD-studie worden 215 thuiswonende jonge mensen met een dementie en hun mantelzorger elke zes maanden onderzocht gedurende twee jaar. Onder meer het beloop van de dementie, specifieke kenmerken (zoals cognitief functioneren, neuropsychiatrische symptomen en kwaliteit van leven) naast het beloop van zorgbehoeften worden hierbij in kaart gebracht. In hoofdstuk 2 wordt in detail ingegaan op de opzet en methodologie van de NeedYD-studie.

In het inleidende hoofdstuk (hoofdstuk 1) worden de rationale en vraagstellingen van dit proefschrift beschreven. Om het beloop van zorgbehoeften en ervaringen met zorgbehoeften nader in kaart te brengen werd een kwalitatieve case studie verricht, waarvan de resultaten in hoofdstuk 3 beschreven staan. Op basis van de interviews gehouden met de mantelzorger van een jonge man met de ziekte van Alzheimer werd met behulp van een inductieve inhoudsanalyse gezocht naar mogelijk centrale thema's in de zorgsituatie van jonge mensen met een dementie. De resultaten laten zien dat jonge mensen met een dementie en mantelzorgers zich geconfronteerd zien met specifieke problemen gedurende het zorgtraject, namelijk (1) een langere duur totdat een juiste diagnose kan worden gesteld, (2) onvoldoende aansluiting tussen zorgbehoeften en beschikbare zorg, (3) de belasting van het spanningsveld tussen het bieden van zorg enerzijds en het eigen toekomstperspectief voor de mantelzorger anderzijds, en (4) het belang van aandacht vanuit zorgprofessionals voor veranderende zorgbehoeften van patiënt en mantelzorger.

In hoofdstuk 4 zijn vervolgens de zorgbehoeften en de mate waarin hierin werd voorzien bij de gehele studiegroep in kaart gebracht. Daarbij is ook de mate van overeenstemming in de beoordeling van zorgbehoeften tussen degene die de zorg ontving en de mantelzorger alsmede de relatie tussen zorgbehoeften en neuropsychiatrische symptomen over de hele studieperiode onderzocht. Resultaten laten een redelijke overeenstemming zien tussen jonge mensen met een dementie en hun mantelzorgers wat betreft de gebieden waarin zorgbehoeften werden gesignaleerd. Echter, de overeenstemming over de mate waarin in deze zorgbehoeften werd voorzien was beperkt. De resultaten laten verder zien dat bij jonge mensen met een dementie het waarschijnlijker is dat in specifieke levensgebieden zorgbehoeften optreden waarin niet of moeilijk kan worden voorzien. Patiënten gaven hoge proporties niet tegemoet gekomen zorgbehoeften aan op verschillende terreinen, bijvoorbeeld wat betreft het vinden van een zinvolle dagbesteding, het vinden van gezelschap, het onderhouden van intieme relaties en op het gebied van informatie over de dementie en zorgmogelijkheden. Daarnaast bleek er een belangrijke relatie te bestaan tussen tegemoetgekomen én niet tegemoetgekomen zorgbehoeften enerzijds en het beloop van neuropsychiatrische symptomen anderzijds. In de behandeling van neuropsychiatrische symptomen is het dan ook belangrijk dat professionals in de zorg zich direct richten op het verminderen van deze symptomen maar ook aandacht hebben voor het terugdringen van het aantal niet tegemoet gekomen zorgbehoeften. Dit zal alleen mogelijk blijken wanneer periodiek zorgbehoeften en de mate waarin hierin wordt voorzien in kaart worden gebracht. Het in kaart brengen van het oordeel van zowel degene die de zorg ontvangt als dat van de mantelzorger zou daarbij uitgangspunt moeten zijn.

Omdat ook een relatie kon worden vermoed tussen niet tegemoet gekomen zorgbehoeften en ervaren kwaliteit van leven van zowel degene met de dementie als diens mantelzorger is dit in hoofdstuk 5 in kaart gebracht. Er werd geen relatie gevonden tussen niet tegemoet gekomen zorgbehoeften en kwaliteit van leven van jonge mensen met een dementie zelf. Er werd echter wel gevonden dat niet tegemoet gekomen zorgbehoeften van degene die de zorg ontving verschillende aspecten van de ervaren kwaliteit van leven van de mantelzorger negatief beïnvloedden. Mantelzorgers van jonge mensen met een dementie met een groter aantal zorgbehoeften waaraan niet tegemoet werd gekomen gaven aan een verlies van vitaliteit en algehele gezondheid, meer pijn en meer belemmeringen in het dagelijks functioneren ten gevolge van fysieke problemen te ervaren. Tegelijkertijd bleken niet tegemoet gekomen zorgbehoeften van de mantelzorger zelf te resulteren in meer depressieve gevoelens en angstklachten naast beperkingen in sociaal functioneren bij de mantelzorger. Wanneer mantelzorgers gebruik maakten van professionele hulp en ondersteuning bleek dit gerelateerd aan een vermindering van ervaren beperkingen ten gevolge van zowel emotionele als fysieke problemen. Tegelijkertijd bleek dit gerelateerd aan een ervaren verslechtering op andere domeinen van kwaliteit van leven van de mantelzorger. Een meer persoonsgerichte benadering van de zorg voor deze doelgroep vraagt daarmee om het meer toegespitsen van voorzieningen op persoonlijke behoeften en wensen, wat een belangrijke bijdrage kan leveren aan de ervaren kwaliteit van leven.

In hoofdstuk 6 wordt het gebruik van formele en informele zorg en de relatie met kenmerken van de ontvanger van de zorg en die van de mantelzorger nader in kaart gebracht. Het blijkt dat tweederde van de totale zorg uit mantelzorg bestaat, waarbij het grootste deel van deze informele zorg bestaat uit het toezicht houden op degene met de dementie. In meer dan de helft van de gevallen was er slechts één mantelzorger beschikbaar. De resultaten laten verder zien dat in meer gevorderde stadia van de ziekte het aantal uren informele zorg toeneemt. Omgekeerd zagen we dat bij werkende mantelzorgers, zeker naar mate degene met de dementie jonger was minder informele zorg geboden werd. De hoeveelheid formele zorg bleek in onze studie in belangrijke mate te worden bepaald door een toename in de ernst van de dementie, hogere niveaus van neuropsychiatrische symptomen en verminderde zelfredzaamheid van degene met de dementie ten aanzien van de zelfzorg. Bij dementie op jonge leeftijd wordt dus het grootste deel van de zorg geboden door familieleden van degene met de dementie, waarbij tegelijkertijd allerlei andere rollen vervuld moeten worden. Met name voor mantelzorgers van de jongste individuen in de meer gevorderde stadia van de dementie blijkt er een groot spanningsveld te bestaan tussen de noodzaak om te werken enerzijds en de verantwoordelijkheid voor de zorg anderzijds. Dit laat zien dat het waarschijnlijk is dat er ook binnen de groep van jonge mensen met een dementie en hun mantelzorgers belangrijke leeftijdsgerelateerde verschillen kunnen bestaan. De relatie tussen een meer gevorderd stadium van de dementie en het gebruik van formele zorg suggereert daarbij dat dit gebruik zo lang mogelijk wordt uitgesteld bij deze doelgroep.

In hoofdstuk 7 is de duur van eerste ziekteverschijnselen tot aan opname tussen jonge en oudere mensen met een dementie vergeleken. Daarbij is ook gekeken naar factoren die samenhangen met deze duur voor beide groepen. De resultaten laten zien dat bij jonge mensen met een dementie het 9 jaar duurt vanaf de eerste ziekteverschijnselen tot aan opname, terwijl dit voor de ouderen ongeveer 4 jaar betrof. In de groep jonge mensen met een dementie blijkt apathie een belangrijke voorspeller van de duur tot opname. In beide groepen was de mate waarin de mantelzorger zich toegerust voelt om zorg te bieden aan iemand met een dementie eveneens een belangrijke voorspeller van de duur tot opname. Waarbij een hoger gevoel van competentie was gerelateerd aan een verlenging van de periode tot aan opname. Dit onderstreept het belang van een tijdige, juiste diagnose zodat adequate hulp en ondersteuning kan worden ingezet, zeker in het licht van de lange periode dat deze jonge mensen thuis worden verzorgd. Het risico op opname zal mogelijk kunnen worden verkleind wanneer interventies voor de mantelzorger worden ontwikkeld die meer zijn gericht op het vergroten van het ervaren gevoel van competentie en het omgaan met neuropsychiatrische symptomen, in het bijzonder apathie.

In het laatste, achtste hoofdstuk worden de belangrijkste bevindingen van dit proefschrift besproken, evenals methodologische overwegingen en implicaties voor zowel de klinische praktijk als toekomstig onderzoek.

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Curriculum Vitae

Christian Bakker werd op 23 juni 1974 geboren te ’s Gravenhage, Nederland. In 1993 behaalde hij zijn VWO diploma aan het Fioretti college in Lisse. Van 1993 tot 1998 studeerde hij klinische en gezondheidspsychologie aan de Leidse Universiteit. Na zijn afstuderen bleef hij in dienst van de Universiteit, vakgroep Psychiatrie en was betrokken bij het onderzoek naar dynamische persoonlijkheidsdiagnostiek met behulp van het “Ontwikkelingsprofiel”. In 2000 maakte hij zijn overstap vanuit de psychiatrie naar de ouderenzorg en kwam in dienst bij zijn huidige werkgever, Stichting KVV, het huidige Florence Zorggroep. Vanuit de gemeenschappelijke interesse van Florence en de Radboud Universiteit naar dementie op jonge leeftijd werd hij aangesteld als junior onderzoeker bij het NeedYD-project, het onderzoek naar het beloop van dementie en zorgbehoeften op jonge leeftijd. Dit onderzoek heeft geresulteerd in dit proefschrift. Momenteel is hij werkzaam als psycholoog en zorgprogrammaleider bij het Expertisecentrum voor Jonge Mensen met een Dementie van Florence en als onderzoeker bij de afdeling Eerstelijnsgeneeskunde, Centrum voor Huisartsgeneeskunde, Ouderengeneeskunde en Public Health van de Radboud Universiteit.