Maastricht University

Family caregivers of persons with young-onset dementia

Citation for published version (APA):

Bruinsma, J. (2022). *Family caregivers of persons with young-onset dementia: tailoring web-based psychosocial support*. [Doctoral Thesis, Maastricht University]. Ridderprint. https://doi.org/10.26481/dis.20220420jb

Document status and date:
Published: 01/01/2022

DOI:
10.26481/dis.20220420jb

Document Version:
Publisher's PDF, also known as Version of record

Please check the document version of this publication:

• A submitted manuscript is the version of the article upon submission and before peer-review. There can be important differences between the submitted version and the official published version of record. People interested in the research are advised to contact the author for the final version of the publication, or visit the DOI to the publisher's website.
• The final author version and the galley proof are versions of the publication after peer review.
• The final published version features the final layout of the paper including the volume, issue and page numbers.

Link to publication

General rights
Copyright and moral rights for the publications made accessible in the public portal are retained by the authors and/or other copyright owners and it is a condition of accessing publications that users recognise and abide by the legal requirements associated with these rights.

• Users may download and print one copy of any publication from the public portal for the purpose of private study or research.
• You may not further distribute the material or use it for any profit-making activity or commercial gain
• You may freely distribute the URL identifying the publication in the public portal.

If the publication is distributed under the terms of Article 25fa of the Dutch Copyright Act, indicated by the “Taverne” license above, please follow below link for the End User Agreement:

www.umlib.nl/taverne-license

Take down policy
If you believe that this document breaches copyright please contact us at:

repository@maastrichtuniversity.nl

providing details and we will investigate your claim.

Download date: 05 okt.. 2022

Family caregivers of persons with young-onset dementia

Tailoring web-based psychosocial support

Jeroen Bruinsma

Colophon

Family caregivers of persons with young-onset dementia: Tailoring web-based psychosocial support

ISBN/EAN: 978-94-6416-983-6

Cover: Jeroen Bruinsma

Printing: Ridderprint, the Netherlands

ISBN: 978-94-6416-983-6

Financial support for printing this dissertation was kindly provided by Alzheimer Netherlands.

Family caregivers of persons with young-onset dementia

Tailoring web-based psychosocial support

PROEFSCHRIFT

ter verkrijging van de graad van doctor aan de Universiteit Maastricht,
op gezag van de Rector Magnificus, Prof.dr. Pamela Habibović
volgens het besluit van het College van Decanen,
in het openbaar te verdedigen
op woensdag 20 april 2022 om 16:00 uur

door

Jeroen Bruinsma
Geboren op 20 januari 1989 te Delft

Promotoren

Prof. dr. M.E. de Vugt

Prof. dr. F.R.J. Verhey

Copromotoren

Dr. K.K.B. Peetoom

Dr. C. Bakker (Radboud Universiteit Nijmegen)

Beoordelingscommissie

Prof. dr. C.M. van Heugten (voorzitter)

Prof. dr. R. Crutzen

Prof. dr. D.L. Gerritsen (Radboud Universiteit Nijmegen)

Prof. dr. J.P.H. Hamers

Dr. J.M. Papma (Erasmus Universiteit Rotterdam)

Contents

Part two: Tailoring and evaluating web-based support for caregivers of persons with young-onset dementia and frontotemporal dementia

Chapter 5

Evaluating the Dutch RHAPSODY program: a web-based information and support program for caregivers of persons with young-onset dementia

Submitted

Chapter 6

Tailoring and evaluating the web-based 'Partner in Balance' intervention for family caregivers of persons with young-onset dementia

In Internet Interventions, 2021

Chapter 7

Tailoring the web-based 'Partner in Balance' intervention to support spouses of persons with frontotemporal dementia

In Internet Interventions, 2021

Chapter 8

General discussion

Addendum

- Summary | Nederlandse samenvatting
- Impact paragraph
- Author information and list of publications
- Dankwoord (acknowledgements)
- Thesis defenses from MHeNs

Chapter 1

General Introduction

Chapter 1

The prevalence, etiology, and presentation of young-onset dementia

Around 47 million persons have dementia worldwide (World Health Organization, 2017). Although most persons with dementia are older than 65 years, dementia can also occur at a younger age (Rossor et al., 2010). The term young-onset dementia (YOD) refers to dementia with a symptom onset before the age of 65 years (van de Veen et al., 2021). In the Netherlands, between 14.000 and 17.000 persons have YOD (Hendriks et al., 2021).

Alzheimer's disease is the most prevalent cause of dementia in general, but also among persons with YOD (Harvey et al., 2003; Hendriks et al., 2021). The underlying pathology in YOD is more diverse than dementia occurring at older age. One-third of the persons with YOD have Alzheimer's dementia, whereas in older peers this is around two-thirds (Jefferies and Agrawal, 2018). Frontotemporal dementia (FTD) is a specific pathology that is relatively more prevalent in persons under the age of 65 years (Bang et al., 2015). Approximately 10% of the persons with YOD have FTD, compared to 3% in older persons with dementia (Hogan et al., 2016). Other common pathologies causing YOD are vascular dementia, dementia with Lewy bodies, Parkinson's disease, and Huntington's disease (Draper and Withall, 2016; Rossor et al., 2010). More rare diseases causing YOD are corticobasal degeneration, Creutzfeldt-Jacob disease, or progressive supranuclear palsy (Alzheimers Society, 2021).

The clinical presentation of YOD is more diverse than dementia occurring in older persons (Draper and Withall, 2016). In general, early symptoms of YOD more often include difficulty organizing and planning, selfishness, apathy, agitation, social withdrawal, low self-awareness, compulsiveness, language problems, social awkwardness, or altered eating behaviors (Jefferies and Agrawal, 2018). Compared to Alzheimer's dementia at older age, the early presentation of YOD Alzheimer's dementia is less likely to include memory deficits, and is more often characterized by apraxia, language difficulty, and problems in executive functioning (Koedam et al., 2010). Specific characteristics of FTD are language problems, loss of semantic knowledge, or profound changes in personality and behavior, such as apathy, impulsiveness, loss of empathy, uninhibited behavior, lacking social insight, and absence of disease awareness (Rabinovici and Miller, 2010; Ducharme et al., 2020). This diverse clinical presentation of symptoms at young age can delay the diagnosis (Draper et al., 2016). Establishing a dementia diagnosis generally takes around 2.8 years, while in YOD this is 4.4 years. The delay can even last up to 6.4 years in the case of FTD at a young age (van Vliet et al., 2013). The diagnostic delay is problematic because it impedes the access to post-diagnostic care and support (van Vliet et al., 2011; Rasmussen et al., 2019).

The impact of young-onset dementia on family caregivers

Providing caregiving to a relative with dementia at home can cause high levels of burden and distress. This is particularly evident for YOD and FTD caregivers. Compared with older peers, YOD caregivers report lower quality of life (Hvidsten et al., 2020; Millenaar et al., 2016). Persons with YOD are more likely to be in good physical health than their older peers and tend to live at home longer before institutionalization (Gerritsen et al., 2016). More specifically, persons with YOD live at home for nine years before institutionalization, in older persons with dementia this is four years (Bakker et al., 2013a). Before institutionalization, around two-thirds of the support for persons with YOD and FTD is provided by family, friends, or neighbors (Bakker et al., 2013b). It is often the spouse who becomes primarily responsible for organizing care at home (Cartwright et al., 2021). Spouses can experience context-specific problems with combining caregiving and employment or with raising their children. They are also at risk for having financial problems because the loss of income of their partner with YOD or FTD (van Vliet et al., 2010). Additionally, spouses have to cope with the specific symptoms of YOD that elicit high levels of burden and distress, particularly if they have a relative with the behavioral variant of FTD (Cabote et al., 2015; Caceres et al., 2016). Coping with profound behavioral symptoms can be challenging for caregivers, for example because symptoms often result in heated arguments and are accompanied by stigma (Diehl-Schmid et al., 2013; Nunnemann et al., 2012). This may adversely affect the sense of couplehood experienced by spouses as they may perceive to lose the emotional connection to their loved one with YOD or FTD (Holdsworth and McCabe, 2018). However, caregiving may also have positive effects on the relationship of spouses that provide caregiving, as they may feel closer to their partner (de Vugt et al., 2003).

Providing caregiving to a person with YOD or FTD at home affects roles and relationships within families. Given the young age, children may still live at home and gradually become involved in caring for their parent who has YOD or FTD. Children are often in puberty, adolescence, or (young) adulthood but can also be younger. When they become involved in caregiving they can experience a reversed child-parent role as care responsibility increases (Grundberg et al., 2021; Cartwright et al., 2021). This can be stressful and may result in social isolation (Grundberg et al., 2021). Additionally, (adult) children can have financial concerns, worry about the burden on their healthy parent, and may postpone plans, such as moving out or studying (Millenaar et al., 2014; Grundberg et al., 2021). Despite these adverse effects, caring for a parent with YOD or FTD can have positive effects on children who can feel to mature faster and may experience a stronger bond with other relatives (Grundberg et al., 2021).

Support for caregivers of persons with young-onset dementia

Caregivers of persons with YOD or FTD often perceive a mismatch between service availability and their need for specific and age-appropriate support (Mayrhofer et al., 2018). To illustrate, they may feel it is difficult to relate to support designed for older caregivers (Cations et al., 2017). Particularly caregivers of persons with FTD may experience this because most information and support is designed with Alzheimer's dementia in mind. Given the low prevalence of YOD and particularly FTD, healthcare professionals may have limited experience in coping with specific symptoms. This can make it difficult for professionals to provide practical advice to YOD and FTD caregivers (Diehl-Schmid et al., 2013). Generally, only limited support is available that is specifically designed to support (adult) children and other relatives of persons with YOD (Cations et al., 2017; Cartwright et al., 2021). The limited availability of appropriate support may be problematic as it could undermine the confidence that YOD and FTD caregivers have in professional healthcare. In turn, they may postpone the initiation of professional support services (Cations et al., 2017). Accepting professional support in an early phase is often difficult for caregivers, for example because they feel they can still manage themselves (Boots et al., 2015). Further delay of initiating professional support may impede the ability of YOD and FTD caregivers to adapt to the caregiving role. This may have negative effects on their well-being (Tookey et al., 2021).

Even though there is a consensus that support for YOD caregivers should be age-appropriate, there is little knowledge about what works (Mayrhofer et al., 2018). In the Netherlands, family members caring for a relative with YOD at home can use supportive services such as daycare, support from a nurse, information services, psychoeducation, and caregiver support groups (Bakker et al., 2013). These professional services aim to provide psychosocial support by helping caregivers in coping with the caregiving role (de Vugt and Verhey, 2013; American Psychological Association, 2021). Psychosocial support is known to increase the resilience of caregivers by helping them to think positively, increase self-efficacy, and be resourceful (Kobiske and Bekhet, 2018). Promoting the use of adaptive coping strategies in caregivers can also positively contribute to their quality of life (Tookey et al., 2021). This may help YOD and FTD caregivers to continue organizing adequate care for their relative at home (Mayrhofer et al., 2018).

Ideally, support for YOD and FTD caregivers is accessible, affordable, flexible, and should offer unique and tailored content (Cations et al., 2017). Given the low prevalence of these conditions, eHealth may provide a promising direction to offer support for YOD and FTD caregivers at home. An eHealth approach is in line with the current digital trend and may allow service access at a convenient time and place, even in rural areas (Diehl-Schmid et al., 2013). Although there is no clear definition of eHealth, the term often refers to technology used to promote web-based healthcare services delivered through the internet (Ho et al., 2015).

That eHealth can be an acceptable support tool for YOD caregivers has been demonstrated by the RHAPSODY program (Kurz et al., 2016; Metcalfe, 2019). This is a web-based educational program for caregivers of persons with YOD including psychoeducation and practical advice. The program provides information on the medical background of YOD and subtypes, on managing behavioral symptoms, dealing with role change, and balancing caregiving with daily life (Kurz et al., 2016). A pilot in Germany, France, and the United Kingdom demonstrated language tailored versions of the RHAPSODY program were usable and acceptable. The RHAPSODY program also positively contributed to caregiver well-being (Metcalfe, 2019). These effects are promising but there was no Dutch version of the RHAPSODY program available in the Netherlands.

Another promising web-based intervention for caregivers is the Partner in Balance intervention. Partner in Balance is an eight-week web-based intervention including video vignettes, psychoeducation, self-reflection, and goal-setting. Currently, the intervention is being implemented in the Netherlands (Christie et al., 2020; Christie et al., 2021). During the intervention caregivers complete four self-chosen modules and receive online coaching from a trained healthcare professional, often a dementia casemanager or psychologist. Examples of Partner in Balance module themes include acceptance of the caregiving role, coping with communication difficulties, and positive thinking. Previously, the Partner in Balance intervention has shown to be usable, feasible, and acceptable (Boots et al., 2016). Following the Partner in Balance intervention, caregivers improved on measures on self-efficacy, sense of mastery, and quality of life (Boots et al., 2018; Boots et al., 2016). Although these results are promising, YOD caregivers felt most of the program content was too generic. More specifically, they felt intervention materials portrayed the experiences of older peers and they preferred specific content on YOD and FTD (Boots et al., 2017).

Objectives and outline of this dissertation

This dissertation aimed to explore the lived experiences of YOD and FTD caregivers regarding the disease trajectory to identify directions for support (part one). Subsequently, the RHAPSODY program and the Partner in Balance intervention were tailored for this specific group. Tailored versions included YOD-specific content for spouse caregivers, children in their puberty or older, and other relatives involved in caregiving. This dissertation aimed to evaluate these tailored web-based interventions (part two). More specifically, the tailored version of the RHAPSODY program was evaluated in terms of user acceptability, usability, satisfaction, and user behavior. Different tailored versions of the Partner in Balance intervention were evaluated in terms of usability, feasibility, acceptability, and explorative effects on self-efficacy, sense of mastery, distress, anxiety, and depression.

Part one **aims to obtain more understanding of the lived experiences of YOD caregivers throughout the disease trajectory to identify directions for support (chapters 2, 3, and 4).**

Chapter 2 describes a quantitative exploration using data from the Needs in Young-onset Dementia (NeedYD) study. Our study addressed the following research question:

‘Which factors can predict changes in the quality of the relationship perceived by spouses caring for a partner with young-onset dementia living at home?’.

Chapter 3 reports a qualitative study by means of focus group discussions with family caregivers of persons with FTD. The research question was:

‘What are lived experiences of family members caring for a relative with FTD regarding recognizing early symptoms, the diagnostic trajectory, and post-diagnostic support?’

Chapter 4 documents a case study about one family who has a relative with the behavioral variant of FTD living at home. The following research question was addressed:

‘How do symptoms of the behavioral variant of frontotemporal dementia interfere with family dynamics?’

Part two documents evaluations of existing web-based interventions that were tailored to support caregivers who have a relative with YOD or FTD living at home (chapters 5, 6, and 7).

Chapter 5 describes a cross-sectional study using quantitative and qualitative measures to evaluate a recently developed Dutch version of the web-based RHAPSODY program for YOD caregivers. The program was publicly available via the website of the Dutch Alzheimer's Society [*Alzheimer Nederland*]. The following research question was addressed:

'How do YOD caregivers perceive a Dutch version of the RHAPSODY intervention in terms of user acceptability, usability, satisfaction, and user behavior?'

Chapter 6 documents a prospective study to evaluate tailored versions of the Partner in Balance intervention for spouses and other family members who have a relative with YOD living at home. Qualitative and quantitative measures were used to address the following research question:

'How do family caregivers of persons with YOD perceive the usability, feasibility, acceptability, and effects of tailored versions of the Partner in Balance intervention?'

Chapter 7 reports a prospective study evaluating Partner in Balance content tailored specifically for spouses caring for a partner with FTD living at home. Using qualitative and quantitative measures the following research question was addressed:

'How do spouses of persons with FTD perceive a tailored version of Partner in Balance in terms of their expectations, aspects of usability, feasibility, acceptability, and effects?'

Finally, **Chapter 8** is a general discussion, and provides future directions.

References

Alzheimer's Society. (2021, August 20). Rarer types of dementia. Retrieved from: https://www.alzheimers.org.uk/about-dementia/types-dementia/rarer-types-dementia#content-start

American Psychological Association (2021, August 20). Dictionary of psychology. Retreived from: https://dictionary.apa.org/psychosocial-support

Bakker, C., de Vugt, M. E., van Vliet, D., Verhey, F. R., Pijnenburg, Y. A., Vernooij-Dassen, M. J., and Koopmans, R. T. (2013a). Predictors of the time to institutionalization in young- versus late-onset dementia: results from the needs in young onset dementia (NeedYD) study. *J Am Med Dir Assoc, 14*(4), 248-253. doi:10.1016/j.jamda.2012.09.011

Bakker, C., de Vugt, M. E., van Vliet, D., Verhey, F. R., Pijnenburg, Y. A., Vernooij-Dassen, M. J., and Koopmans, R. T. (2013b). The use of formal and informal care in early onset dementia: results from the NeedYD study. *Am J Geriatr Psychiatry, 21*(1), 37-45. doi:10.1016/j.jagp.2012.10.004

Bang, J., Spina, S., and Miller, B. L. (2015). Frontotemporal dementia. *Lancet, 386*(10004), 1672-1682. doi:10.1016/s0140-6736(15)00461-4

Boots, L. M., de Vugt, M. E., Kempen, G. I., and Verhey, F. R. (2018). Effectiveness of a blended care self-Management program for caregivers of people with early-stage dementia (Partner in Balance): randomized controlled trial. *J Med Internet Res, 20*(7), e10017. doi:10.2196/10017

Boots, L. M., de Vugt, M. E., Smeets, C. M., Kempen, G. I., and Verhey, F. R. (2017). Implementation of the blended care self-management program for caregivers of people with early-stage dementia (Partner in Balance): process evaluation of a randomized controlled trial. *J Med Internet Res, 19*(12), e423. doi:10.2196/jmir.7666

Boots, L. M., de Vugt, M. E., Withagen, H. E., Kempen, G. I., and Verhey, F. R. (2016). Development and initial evaluation of the web-based self-management program "Partner in Balance" for family caregivers of people with early stage dementia: an exploratory mixed-methods study. *JMIR Res Protoc, 5*(1), e33. doi:10.2196/resprot.5142

Boots, L. M., Wolfs, C. A., Verhey, F. R., Kempen, G. I., and de Vugt, M. E. (2015). Qualitative study on needs and wishes of early-stage dementia caregivers: the paradox between needing and accepting help. *Int Psychogeriatr, 27*(6), 927-936. doi:10.1017/S1041610214002804

Cabote, C. J., Bramble, M., and McCann, D. (2015). Family caregivers' experiences of caring for a relative with younger onset dementia: a qualitative systematic review. *J Fam Nurs, 21*(3), 443-468. doi:10.1177/1074840715573870

Caceres, B. A., Frank, M. O., Jun, J., Martelly, M. T., Sadarangani, T., and de Sales, P. C. (2016). Family caregivers of patients with frontotemporal dementia: an integrative review. *Int J Nurs Stud, 55*, 71-84. doi:10.1016/j.ijnurstu.2015.10.016

Cartwright, A. V., Stoner, C. R., Pione, R. D., and Spector, A. (2021). The experiences of those affected by parental young onset dementia: A qualitative systematic literature review. *Dementia,* online ahead of print. doi:10.1177/1471301220988231

Cations, M., Withall, A., Horsfall, R., Denham, N., White, F., Trollor, J., Loy, C., Brodaty, P. S., Gonski, P., Demirkol, A., Cumming, R. G., and Draper, B. (2017). Why aren't people with young onset dementia and their supporters using formal services? Results from the INSPIRED study. *PLoS One, 12*(7), e0180935. doi:10.1371/journal.pone.0180935

Christie, H. L., Boots, L. M. M., Peetoom, K., Tange, H. J., Verhey, F. R. J., and de Vugt, M. E. (2020). Developing a plan for the sustainable implementation of an electronic health intervention (partner in balance) to support caregivers of people with dementia: case study. *JMIR Aging 3*(1), e18624. doi:10.2196/18624

Christie, H. L., Boots, L. M. M., Tange, H. J., Verhey, F. R, J., and de Vugt, M. E. (2021). Implementations of edidence-based ehealth interventions for caregivers of people with dementia in municipality contexts (Myinlife and Partner in Balance): evaluation study. *JMIR Aging 4*(1), e21629. doi:10.2196/21629

de Vugt, M. E., and Verhey, F. R. (2013). The impact of early dementia diagnosis and intervention on informal caregivers. *Prog Neurobiol, 110*, 54-62. doi:10.1016/j.pneurobio.2013.04.005

Diehl-Schmid, J., Schmidt, E. M., Nunnemann, S., Riedl, L., Kurz, A., Forstl, H., Wagenpfeil, S., and Cramer, B. (2013). Caregiver burden and needs in frontotemporal dementia. *J Geriatr Psychiatry Neurol, 26*(4), 221-229. doi:10.1177/0891988713498467

Draper, B., Cations, M., White, F., Trollor, J., Loy, C., Brodaty, H., Sachdev, P., Gonski, P., Demirkol, A., Communig, R. G., and Withall, A. (2016). Time to diagnosis in young-onset dementia and its determinants: the INSPIRED study. *Int J Geriatr Psychiatry, 31*(11), 1217-1224. doi:10.1002/gps.4430

Draper, B., and Withall, A. (2016). Young onset dementia. *Intern Med J, 46*(7), 779-786. doi:10.1111/imj.13099

Ducharme, S., Dols, A., Laforce, R., Devenney, E., Kumfor, F., van den Stock, J., . . . Pijnenburg, Y. (2020). Recommendations to distinguish behavioural variant frontotemporal dementia from psychiatric disorders. *Brain.* doi:10.1093/brain/awaa018

Gerritsen, A. A. J., Bakker, C., Verhey, F. R. J., de Vugt, M. E., Melis, R. J. F., Koopmans, R. T. C. M.,and 4C study team. (2016). Prevelance of comorbidity in patients with young-onset Alzheimer disease compared with late-onset: a comparative cohort study. *J Am Med Dir Assoc, 17*(4), 318-323. doi:10.1016/j.jamda.2015.11.011

Grundberg, A., Sandberg, J., and Craftman, A. G. (2021). Childrens' and young adults' perspectives of having a parent with dementia diagnosis: A scoping review. *Dementia*, online ahead of print. doi:10.1177/14713012211023653

Eysenbach, G. (2001). What is e-health. *J Med Internet Res, 3*(2). doi:10.2196/jmir.3.2.e20

Harvey, R. J., Skelton-Robinson, M. S., and Rossor, M. N. (2003). The prevalence and causes of dementia in people under the age of 65 years. *J Neurol Neurosurg Psychiatry, 74*(9), 1206-1209. doi:10.1136/jnnp.74.9.1206

Hendriks, S., Peetoom, K., Bakker, C., van der Flier, W. M., Papma, J. M., Koopmans, R., Verhey, F. R., de Vugt, M., and Köhler, S. (2021). Global prevalence of young-onset dementia: a systematic review and meta-analysis. *JAMA Neurol.* doi:10.1001/jamaneurol.2021.2161

Hogan, D. B., Jette, N., Fiest, K. M., Roberts, J. I., Pearson, D., Smith, E. E., Roach, P., Krik, A., Pringsheim, T., and Maxwell, C. J. (2016). The prevalence and incidence of frontotemporal dementia: a systematic review. *Can J Neurol Sci, 43*(1), S96-S109. doi:10.1017/cjn.2016.25

Holdsworth, K., and McCabe, M. (2018). The impact of younger-onset dementia on relationships, intimacy, and sexuality in midlife couples: a systematic review. *Int Psychogeriatr, 30*(1), 15-29. doi:10.1017/S1041610217001806

Hvidsten, L., Engedal, K., Selbaek, G., Wyller, T. B., Benth, J. S., BruvikF., and Kersten, H. (2020). Quality of life of family carers of persons with young-onset compared to late-onset dementia. *Aging ment health, 24*(9), 1394-1401. doi:10.1080/13607863.2019.1617245

Jefferies, K., and Agrawal, N. (2018). Early-onset dementia. *Adv in Psychiatr Treat, 15*(5), 380-388. doi:10.1192/apt.bp.107.004572

Kobiske, K. R., and Bekhet, A. K. (2018). Resilience in caregivers of partners with young onset dementia: a concept analysis. *Issues Ment Health Nurs, 39*(5), 411-419. doi:10.1080/01612840.2017.1400625

Koedam, E. L., Lauffer, V., van der Vlies, A. E., van der Flier, W. M., Scheltens, P., and Pijnenburg, Y. A. (2010). Early-versus late-onset Alzheimer's disease: more than age alone. *J Alzheimers Dis, 19*(4), 1401-1408. doi:10.3233/JAD-2010-1337

Kurz, A., Bakker, C., Bohm, M., Diehl-Schmid, J., Dubois, B., Ferreira, C., Gage, H., Graff, C., Hergueta, T., Jansen, S., Jones, B., Komar, A., de Mendonça, A., Metcalfe, A., Milecka, K., Millenaar, J., Orrung Wallin, A., Oyebode, J., Schneider-Schelte, H., Saxl, S., and de Vugt, M. (2016). RHAPSODY - Internet-based support for caregivers of people with young onset dementia: program design and methods of a pilot study. *Int Psychogeriatr, 28*(12), 2091-2099. doi:10.1017/S1041610216001186

Mayrhofer, A., Mathie, E., McKeown, J., Bunn, F., and Goodman, C. (2018). Age-appropriate services for people diagnosed with young onset dementia (YOD): a systematic review. *Aging Ment Health, 22*(8), 927-935. doi:10.1080/13607863.2017.1334038

Mendez, M. F. (2019). Early-onset alzheimer disease and its variants. *Continuum (Minneap Minn), 25*(1), 34-51. doi:10.1212/CON.0000000000000687

Metcalfe, A., Jones, B., Mayer, J., Gage, H., Oyebode, J., Boucault, S., Aloui, S., Schwertel, U., Böhm, M., Tezenas du Montcel, S., Lebbah, S., De Mendonça, A., De Vugt, M., Graff, C., Jansen, S., Hergueta, T., Dubois, B., and Kurz, A. (2019). Online information and support for carers of people with young-onset dementia: A multi-site randomised controlled pilot study. *Int J Geriatr Psychiatry, 34*(10), 1455–1464. doi:10.1002/gps.5154

Millenaar, J. ., de Vugt, M. E., Bakker, C., van Vliet, D., Pijnenburg, Y. A. L., Koopmans, R. T. C. M., and Verhey, F. (2016). The impact of young onset dementie on informal caregivers compared with late onset dementia: results from the NeedYD study. *Am J Geriatr Psychiatry, 24*(6), 467-474. doi:10.1016/j.jagp.2015.07.005

Millenaar, J. K., van Vliet, D., Bakker, C., Vernooij-Dassen, M. J., Koopmans, R. T. C. M., Verhey, F. R., and de Vugt, M. E. (2014). The experiences and needs of children living with a parent with young onset dementia: results from the NeedYD study. *Int Psychogeriatr, 26*(12), 2001-2010. doi:10.1017/S1041610213001890

Nunnemann, S., Kurz, A., Leucht, S., and Diehl-Schmid, J. (2012). Caregivers of patients with frontotemporal lobar degeneration: a review of burden, problems, needs, and interventions. *Int Psychogeriatr, 24*(9), 1368-1386. doi:10.1017/S104161021200035X

World Health Organization. (2017). *Global action plan on the public health response to dementia 2017 - 2025*. Retreived from: https://www.who.int/publications/i/item/global-action-plan-on-the-public-health-response-to-dementia-2017---2025

Rabinovici, G. D., and Miller, B. L. (2010). Frontotemporal Lobar Degeneration Epidemiology, Pathophysiology, Diagnosis and Management. *CNS Drugs, 24*(5), 375-398. doi:10.2165/11533100-000000000-00000

Rasmussen, H., Hellzen, O., Stordal, E., and Enmarker, I. (2019). Family caregivers experiences of the pre-diagnostic stage in frontotemporal dementia. *Geriatr Nurs, 40*(3), 246-251. doi:10.1016/j.gerinurse.2018.10.006

Rossor, M. N., Fox, N. C., Mummery, C. J., Schott, J. M., and Warren, J. D. (2010). The diagnosis of young-onset dementia. *Lancet Neurol, 9*(8), 793-806. doi:10.1016/s1474-4422(10)70159-9

Tookey, S. A., Greaves, C. V., Rohrer, J. D., and Stott, J. (2021). Specific support needs and experiences of carers of people with frontotemporal dementia: A systematic review. *Dementia*, online ahead of print. doi:10.1177/14713012211022982

van de Veen, D., Bakker, C., Peetoom, K., Pijnenburg, Y., Papma, J. M., PRECODE study group., de Vugt, M., and Koopmans, R. (2021). An Integrative Literature Review on the Nomenclature and Definition of Dementia at a Young Age. *J Alzheimers Dis*. doi:10.3233/JAD-210458

van Vliet, D., de Vugt, M. E., Bakker, C., Koopmans, R. T., Pijnenburg, Y. A., Vernooij-Dassen, M. J., and Verhey, F. R. (2011). Caregivers' perspectives on the pre-diagnostic period in early onset dementia: a long and winding road. *Int Psychogeriatr, 23*(9), 1393-1404. doi:10.1017/S1041610211001013

van Vliet, D., de Vugt, M. E., Bakker, C., Koopmans, R. T., and Verhey, F. R. (2010). Impact of early onset dementia on caregivers: a review. *Int J Geriatr Psychiatry, 25*(11), 1091-1100. doi:10.1002/gps.2439

van Vliet, D., de Vugt, M. E., Bakker, C., Pijnenburg, Y. A., Vernooij-Dassen, M. J., Koopmans, R. T., and Verhey, F. R. (2013). Time to diagnosis in young-onset dementia as compared with late-onset dementia. *Psychol Med, 43*(2), 423-432. doi:10.1017/S0033291712001122

Chapter 2

The quality of the relationship perceived by spouses of people with young-onset dementia

Bruinsma, J., Peetoom, K., Millenaar, J., Köhler, S., Bakker, C., Koopmans, R.,

Pijnenburg, Y., Verhey, F., and de Vugt, M.

International psychogeriatrics, 2020

Abstract

Young-onset dementia (YOD) has a profound impact on spouses. However, little is known on how the quality of the relationship changes over time in YOD. This study aims to determine how the quality of the relationship changes over time and identify predictors of this change.

Methods: This study used data from the Needs in Young onset Dementia (NeedYD) study. The primary outcome measure was the quality of the relationship perceived by spouses measured throughout 24 months. Baseline characteristics of persons with YOD and spouses were also measured to assess their predictive value.

Results: Totally, 178 dyads were included. The perceived quality of the relationship deteriorated over time. A longer symptom duration, a diagnosis of frontotemporal dementia, lower levels of awareness of deficits, lower levels of initiative toward daily living activities, and higher levels of apathy, hyperactivity, depression, and anxiety in the person with YOD were associated with a lower perceived quality of the relationship by spouses. A coping style characterized by palliative and passive reacting patterns and higher levels of neuroticism in spouses was also associated with a lower quality of the relationship.

Conclusion: The quality of the relationship as perceived by spouses deteriorated over time and was influenced by characteristics of the person with YOD as well as their spouse. Helping spouses to come to terms with factors that threaten their sense of couplehood might help them to develop a more positive attitude toward their spousal relationship and improve the quality of the relationship and care.

Keywords: Young-onset dementia; Carers; Neuropsychiatric symptoms; Longitudinal studies; Frontotemporal dementia (FTD)

Introduction

Young-onset dementia (YOD) is defined as dementia with a symptom onset before the age of 65. The onset of symptoms at a young age can cause specific challenges for spousal caregivers that may impact their perception of the spousal relationship. Having a close and high-quality relationship seems to provide people with a sense of security and support but will also have a positive effect on well-being and quality of life (Vangelisti and Perlman, 2006). Specific challenges that occur due to YOD may affect the quality of the spousal relationship. For example, the prolonged time to diagnosis can be a challenge since early symptoms of YOD are frequently mislabeled as marital problems, burn-out, or depression (Draper and Withall, 2016; van Vliet et al., 2011). Spouses are often confronted with changes in personality and behavior in the person with YOD. Misunderstanding the cause of personality and behavioral change may affect the quality of the relationship (Holdsworth and McCabe, 2018). Spouses are also challenged by a longer period of care at home in YOD, and the mean time until institutionalization is reported to be 8.9 years in YOD compared to 4.2 years in late-onset dementia (LOD) (Bakker et al., 2013). This can result in a higher risk of overburden in spouses (van Vliet et al., 2010b).

Previous research has shown that spouses of people with YOD experience changes in the quality of the relationship due to a decline in reciprocity and changes in sexuality (Lockeridge and Simpson, 2013), shifting roles and responsibilities (Massimo et al., 2013), and social isolation (Kimura et al., 2015). Changes in mood, behavior, and daily functioning in the person with YOD may result in feelings of disappointment, anger, frustration, and sometimes result in conflict between spouses (van Vliet et al., 2011). While similar changes have been reported by spouses of people with LOD, the impact of these changes is different in a younger life phase. Spouses of people with YOD often experience increased financial pressure due to a loss of income together with financial responsibilities, such as a mortgage and the education of children (Holdsworth and McCabe, 2018). Although most of these challenges seem to impact the quality of the relationship, some spouses of people with dementia, but not specifically YOD, might still experience high levels of emotional closeness (de Vugt et al., 2003; Riedijk et al., 2008). Indeed, spouses of persons with YOD have reported that they are able to spend more time together and experience a stronger bond (Wawrziczny et al., 2016).

Although YOD has a profound impact on the quality of the relationship, there seems to be a lack of age-appropriate information and support services for spouses of people with YOD that help them cope with changes in their relationship (Holdsworth and McCabe, 2018). In LOD, behavioral changes in persons with LOD have been associated with a lower quality of the spousal relationship as perceived by the spouses (de Vugt et al., 2003), while literature on the factors that influence the quality of the spousal relationship in YOD is scarce. Previous studies often used small sample sizes or qualitative data, and most did not specifically target the quality of the relationship itself, but had a broader focus such as describing the general caregiver experiences (Holdsworth and McCabe, 2018). Hence, quantitative studies examining changes in the quality of the relationship over time are needed due to the progressive nature of YOD.

To improve information and support services for spouses of people with YOD, this study aims to determine how the quality of the relationship changes over time and to identify factors that influence the quality of the spousal relationship. Better understanding factors that impact the quality of the relationship may provide clinical possibilities to increase well-being and quality of life as perceived by spousal caregivers (Holdsworth and McCabe, 2018). This may reduce caregiver distress and may help caregivers to better fulfill caregiving tasks. In turn, this may lower the need for professional care and delay the time to institutionalization (Bakker et al., 2013; Miller et al., 2012). In line with previous studies, we hypothesize that spouses of people with YOD may experience deterioration in the perceived quality of the relationship over time due to characteristics of the person with YOD, such as neuropsychiatric symptoms, and characteristics of the spouse, such as personality and coping strategies.

Methods

Study design and setting

The current study used data from the longitudinal two-year cohort of the Needs in Young onset Dementia (NeedYD) study (van Vliet et al., 2010a). Persons with YOD and their primary caregiver were recruited between 2007 and 2011 through Dutch memory clinics at Amsterdam and Maastricht, regional hospitals, and mental health services. Additionally, a cohort of participants was recruited through specialized day care facilities affiliated with the Dutch National YOD taskforce. Persons with YOD completed clinical tests and participated in a semi-structured interview at baseline and after 6, 12, 18, and 24 months. Spouses also completed a questionnaire and participated in a semi-structured interview at the same time points. This study is reported following the guidelines of STROBE (von Elm et al., 2014).

Participants

The NeedYD study included 209 persons with YOD and their caregivers. Persons with symptom onset before the age of 65 were included, regardless of their age at which they received a dementia diagnosis. In accordance with the Dutch consensus guidelines (CBO, 2005) and the

Diagnostic and Statistical Manual of Mental Disorders (American Psychiatry Association, 2000), the dementia diagnosis was based on clinical and neuropsychological assessments, in combination with neuroimaging. Diagnosis of dementia subtypes was made in accordance with regular guidelines (Erkinjuntti, 1994; McKeith, 2006; McKhann et al., 1984; Mesulam et al., 2003; Neary et al., 1998). Persons with YOD caused by HIV, traumatic brain injury, Down syndrome, Huntington's disease, or alcohol abuse were excluded.

In the current study, persons with YOD and their spousal caregiver were included if they were in a spousal relationship (dyad), shared the same household, and completed items about the quality of the relationship at a minimum of two different time points.

Primary outcome: The perceived quality of the relationship by spouses over 24 months

The primary outcome, the perceived quality of the relationship by spouses over 24 months, was quantitatively assessed by four items from the University of Southern California Longitudinal Study of Three-Generation Families measures of positive affect (Lawrence et al., 1998). The four items assess the current level of (1) closeness ("how close do you feel in the relationship between you and your spouse with dementia?"), (2) communication ("how is the communication between you and your spouse with dementia?"), (3) sharing life views ("how similar are your views about life to those of your spouse with dementia?"), and (4) getting along ("how well do you and your spouse with dementia get along together?"). Each item was rated on an ordinal scale (1 = not close/well/similar/good at all, 4 = very close/well/similar/good). In line with Lawrence et al. (1998), we used a sum score as outcome variable (range: 4–16; Cronbach's α = .71). In addition to the sum score, spouses were also asked at all timepoints to assign a grade (1 = not good at all, 10 = very good) to their current perceived quality of the relationship ("how would you grade your current relationship with your spouse with dementia?"). Assessments were completed at baseline, 6, 12, 18, and 24 months (van Vliet et al., 2010a).

Baseline predictor characteristics of the person with YOD

In addition to demographic characteristics such as gender and age and dementia diagnosis, the dementia severity (1 = no cognitive decline, 7 = severe dementia) in the person with YOD were assessed at baseline during a semi-structured interview using the Global Deterioration Scale (GDS; Reisberg et al., 1982). In addition, the awareness of deficits (1 = absent awareness, 4 = intact awareness) in the person with YOD was rated using the Global Rating of Awareness in Dementia (GRAD; Verhey et al., 1993) at baseline.

During the baseline interview with the spouse, the presence (0 = no, 1 = yes), severity (1 = minor, 3 = severe), and frequency (1 = less than once a week, 4 = once or more than once a day) of neuropsychiatric symptoms in the person with YOD were assessed using the Neuropsychiatric Inventory (NPI; Cumming, 1997; Kat et al., 2002). A continuous score per neuropsychiatric symptom was obtained by multiplying severity and frequency scores (range = 0–12). A summed total NPI score (range: 0–144) and four NPI sub-syndrome scores were assessed (Aalten et al., 2008): apathy (sum of apathy, eating abnormalities, range: 0–24), hyperactivity (sum of agitation, euphoria, disinhibition, irritability, aberrant motor behavior, range: 0–60), depression and anxiety (sum of depression and anxiety, range: 0–24), and psychosis (sum of delusions, hallucinations, nighttime disturbances, range: 0–36). During the baseline interview, spouses also completed the Interview for Deterioration in Daily Living Activities (IDDD; Teunisse and Derix, 1997). Spouses rated (0 = never, 4 = always) whether the person with YOD showed initiative toward (sum of 9 items, range: 0–36) and needed assistance to perform (sum of 11 items, range: 0–44) daily living activities, such as washing oneself, preparing a meal, and handling finances.

Baseline predictor characteristics of the spouse

In addition to demographic characteristics such as gender and age, spouses completed 12 items (1 = not at all, 5 = totally agree) of the NEO Personality Inventory (NEO-FFI; Costa and McCrae, 1989) to measure neuroticism. In line with Chapman (2007) and Saucier (1998), two subscales of neuroticism were assessed: negative affect (items 1, 11, 16, 31, and 46) and self-reproach (items 6, 21, 26, 36, 41, 51, and 56). In addition, spouses completed 47 items (1 = never, 4 = very often) of the Utrechtse Coping List (UCL; Schreurs et al., 1993) to determine spousal coping strategies. Specifically, seven items measured active coping (e.g. clarifying and acting immediately), eight items measured palliative reaction patterns (e.g. diverting attention and engaging in other activities), eight items measured avoidance (e.g. avoiding certain situations), six items measured seeking social support (e.g. looking for comfort from others), five items measured reassuring thoughts (e.g. thinking that worse things can happen), seven items measured passive reaction patterns (e.g. feeling incapable), and three items measured the expression of emotions (e.g. expressing anger and frustration)

Data analysis

To test for change over time in the two primary outcomes (sum score and grade) of perceived quality of the relationship, separate linear mixed models (LMMs) were used. The fixed effect of the continuous time variables gives the average change in quality of the relationship per six months.

To evaluate associations between the baseline predictor variables and levels of the perceived quality of the relationship, averaged over 24 months, each baseline predictor characteristic was separately added to the LMMs. Separately for each baseline predictor characteristic, a fixed effect was included. Subsequently, the LMMs were extended with an interaction effect time*predictor to test whether change over time in relationship quality was influenced by the predictor. For each LMM, model fit was examined by likelihood-testing (X^2, $p < .05$), which compared the maximum loglikelihood (−2LL) and the degrees of freedom (df) of the nested models. Random intercept and random slope models with an unstructured correlation matrix were used as this gave best fit.

To gain insight into how the four individual items of the sum score changed over 24 months, General Estimation Equation (GEE) models were constructed using the individual items as dependent variables. GEE models were preferred over LMMs due to the skewed distribution and ordinal measurement level of the individual items on closeness, communication, sharing life views, and getting along. While both approaches are suitable for repeated measures and give largely similar estimates for continuous outcomes, the individual-specific LMM model can differ from the marginal GEE model when the dependent variable is categorical, with the latter being appropriate if one is interested in population-average change (the regression estimate) and less in the individual-specific (random effect) part (Heck et al., 2012; Twisk, 2007). In the GEE models, time was included as a predictor variable to indicate change over 24 months. An independent correlation matrix was specified for the GEE models.

All analyses were conducted in SPSS 25.0. An alpha level of .05 (in two-sided tests) was used, and a p-value of <.05 was considered to reflect statistical significance.

Ethical considerations

The NeedYD-study protocol was approved by the Medical Ethics Committee of Maastricht University Medical Center (MEC 07-2-38), the Netherlands.

Results

Participants

Of the initial 209 dyads, 178 were eligible for inclusion. Ten dyads were excluded because the spouse did not complete items about the quality of the relationship at two different time points. No differences were found regarding baseline predictor variables or outcome variables, except for the excluded spouses who reported more symptoms of psychosis in the person with YOD.

In total, 102 males and 76 females with YOD were included, with an average age of 61.6 years (SD = 5.3) at baseline (Table 1). On average, symptom-onset started 7.4 years (SD = 3.9) before inclusion. Alzheimer's dementia (59.6%) was the most prevalent diagnosis of YOD, followed by frontotemporal dementia (FTD) (19.1%) and vascular dementia (11.2%). Of the spouses, 75 were male and 103 female (including two female–female and one male–male dyad), with an average age of 60.5 years (SD = 6.8) at baseline.

The averaged sum score for the quality of the relationship was 10.7 (SD = 2.4) at baseline and 9.8 (SD = 2.3) after 24 months. Spouses graded the relationship quality as merely satisfactory with a 6.4 (SD = 2.14) at baseline and 6.0 (SD = 2.36) after 24 months.

Table 1. *Baseline characteristics of persons with YOD and their spouses (n = 178)*

	Persons with YOD	Spouses
Gender, N (Male / Female)	102 / 76	75 / 103
Age in years, Mean (SD)	61.6 (5.3)	60.5 (6.8)
Symptom duration before baseline in years, Mean (SD)	7.4 (3.9)	
Diagnosis, N (%)		
- Alzheimer's dementia	106 (59.6)	
- Frontotemporal dementia	34 (19.1)	
- Vascular dementia	20 (11.2)	
- Lewy Body dementia	6 (3.4)	
- Mixed Alzheimer/vascular dementia	6 (3.4)	
- Not otherwise specified	6 (3.4)	
GDS, Mean (SD)	4.5 (1.0)	
GRAD, Mean (SD)	2.6 (.96)	
IDDD, Mean (SD)		
- Initiative towards daily living activities	15.0 (10.1)	
- Performance of daily living activities	20.2 (13.6)	
NPI[1], Mean (SD)		
- Total	25.8 (18.9)	
- Hyperactivity (n = 138)	13.3 (10.6)	
- Apathy (n = 139)	10.3 (6.6)	
- Psychosis (n = 68)	6.4 (6.3)	
- Depression and anxiety (n = 120)	5.5 (5.1)	

Table 1. *(Continued)*

	Persons with YOD	Spouses
Neuroticism, Mean (SD)		
- Self-reproach		15.5 (5.0)
- Negative affect		14.0 (4.1)
UCL, Mean (SD)		18.4 (3.7)
- Active coping		16.7 (3.2)
- Palliative reaction pattern		15.3 (3.2)
- Avoidance		12.6 (3.1)
- Seeking social support		11.7 (2.5)
- Reassuring thoughts		11.2 (3.3)
- Passive reaction pattern		5.4 (1.5)
- Expressing emotions		

[1] *Only scores ≥1 were used to compute average NPI subsyndrome scores. GDS = Global Deterioration Scale. GRAD = Global Rating of Awareness in Dementia.. IDDD = Interview for Deterioration in Daily Living Activities using domain scores for initiative. NPI = Neuropsychiatric Inventory. Neuroticism = NEO-FFI. UCL = Utrechtse Coping List, using active coping.*

The perceived quality of the relationship by spouses over time

To indicate change in the sum score and the grade for the perceived quality of the relationship over 24 months (Table 2), two LMMs were constructed (Table 3). A linear decline in both the sum score for the quality of the relationship (slope = -.038, $p < .001$) and the grade for the quality of the relationship (slope = -.020, $p < .001$) was observed over 24 months. In-depth analyses at item-level using GEE indicated that experienced feelings of emotional closeness, satisfaction with communication, and satisfaction with sharing views deteriorated over 24 months. The item regarding getting along well did not significantly increase or decrease over 24 months (Table 4).

Table 2. *Descriptive data of the items measuring perceived quality of the relationship over 24 months*

	Baseline	6 months	12 months	18 months	24 months
Relationship grade, Mean (SD)	6.4 (2.14)	6.1 (2.33)	6.1 (2.36)	6.0 (2.5)	6.0 (2.36)
Relationship sum score, Mean (SD)	10.7 (2.4)	10.2 (2.4)	9.9 (2.4)	9.8 (2.3)	9.8 (2.3)
Closeness, %					
- not at all	1.1	1.7	1.8	3.6	1.9
- somewhat	11.9	13.3	13.1	15.7	15.5
- fairly	21.6	30.6	35.7	36.1	37.9
- very	65.3	54.3	49.4	44.6	44.7
Communication, %					
- not at all	37.0	54.0	51.8	56.0	59.0
- somewhat	41.0	31.4	32.1	32.5	28.0
- fairly	18.5	14.0	13.1	9.0	11.2
- very	3.5	4.7	3.0	2.4	1.9
Sharing views, %					
- not at all	28.2	43.2	46.1	48.5	46.5
- somewhat	26.0	30.8	29.7	30.3	31.4
- fairly	36.6	23.1	20.0	16.4	18.9
- very	5.2	3.0	4.2	4.8	3.1
Getting along, %					
- not at all	3.4	3.5	3.6	4.3	3.1
- somewhat	13.2	15.2	16.1	11.1	13.7
- fairly	36.2	44.4	42.7	43.8	44.1
- very well	47.1	36.8	38.7	40.7	39.1

This table represents valid percentage of scores on the four ordinal items of the University of Southern California Longitudinal Study of Three-Generation Families measures of positive affect.

Baseline characteristics as a predictor of perceived quality of the relationship by spouses

Baseline characteristics of the person with YOD were separately added as predictors to the LMMs to examine their predictive value regarding the perceived quality of the relationship over 24 months. A lower sum score for quality of the relationship was associated with a higher total NPI score, higher levels of apathy, higher levels of hyperactivity, a presence of FTD, symptom duration, and lower levels of awareness in the person with YOD. All these baseline variables were also associated with a lower grade for the quality of the relationship over 24 months (Table 5).

Table 3. *Change in the perceived quality of the relationship over 24 months*

	Relationship sum score			Relationship grade		
	Estimate	Sig.	95% CI	Estimate	Sig.	95% CI
Linear time effect	**-.128**	**.003**	**-.213 to -.043**	**-.09**	**.047**	**-.185 to -.001**
Quadratic time effect	.007	.125	-.002 to .016	.007	.162	-.003 to .017
Cubic time effect	-.001	.226	-.001 to .001	-.001	.224	-.001 to .001

Significant time effects are displayed in **bold**. *Linear time = 0, 6, 12, 18 and 24 months. Quadratic time = time* time. Cubic time = time*time* time.*

Table 4. *General Estimation Equation: change on individual items of the sum score for perceived quality of the relationship by spouses over 24 months*

	Estimate	Sig.	95% CI
Closeness			
Linear time	**-.026**	**<.001**	**-.038 to -.014**
Communication			
Linear time	**-.035**	**<.001**	**-.049 to -.021**
Sharing views			
Linear time	**-.020**	**.001**	**-.032 to -.008**
Getting along			
Linear time	-.009	.150	-.020 to .003

The four ordinal items of the University of Southern California Longitudinal Study of Three-Generation Families measures of positive affect are presented. Significant time effects are displayed in **bold**.

In addition, the sum score for the perceived quality of the relationship by spouses was associated to a lower level of initiative toward performing daily living activities in people with YOD. A lower grade for the quality of the relationship over 24 months was associated with higher levels of depression and anxiety in persons with YOD.

When adding interaction effects between time and the baseline predictor variables of the person with YOD to the LMMs, the association between the total NPI score in the person with YOD and the sum score for quality of the relationship significantly diminished over 24 months (slope = .005, $p = .007$). The associations between apathy and the sum score for the quality of the relationship (slope = .017, $p < .001$) and hyperactivity and the sum score for the quality of the relationship (slope = .016, $p <.001$) also significantly diminished over 24 months. No significant interaction effects between time and other baseline predictor variables of the person with YOD were found.

Next, in separate LMMs, the baseline characteristics of the spouses were added as predictors to the LMMs. The sum score for the quality of the relationship over 24 months was associated with the spouses' palliative and passive reaction patterns. This coping style was also associated to a lower grade for the quality of the relationship over 24 months. More expression of emotions and more seeking of social support by spouses were also associated to a lower grade for the quality of the relationship. In addition, a higher level of negative affect in spouses was associated to a lower sum score for the quality of the relationship. In addition, a lower grade was associated to higher levels of self-reproach in the spouse, but this effect diminished over 24 months due to a significant interaction effect with time (slope = .016, $p < .047$). No other significant interaction effects between time and baseline predictor variables of the spouse were found.

Table 5. *LMMs: Predictors of the perceived quality of the relationship by spouses over 24 months*

	Relationship sum score			Relationship grade		
Baseline predictor	Estimate	Sig.	95% CI	Estimate	Sig.	95% CI
Baseline predictor variables of the person with YOD						
Age	-.034	.233	-.091 to .022	-.038	.168	-.093 to .016
Presence of FTD	**-.871**	**.025**	**-1.629 to -.113**	**-.750**	**.044**	**-1.479 to -.021**
Symptom duration	**-.558**	**<.001**	**-.850 to -.265**	**-.430**	**.003**	**-.715 to -.145**
Dementia severity	-.255	.099	-.559 to .049	-.191	.193	-.480 to .098
Awareness of deficits	**.717**	**<.001**	**.430 to 1.004**	**.464**	**.001**	**.183 to .746**
Initiative towards ADL	**.310**	**.042**	**.012 to .608**	.133	.366	-.157 to .424
Performance in ADL	-.167	.276	-.469 to .134	-.008	.955	-.299 to .283
NPI total score	**-.053**	**<.001**	**-.069 to -.037**	**-.038**	**<.001**	**-.052 to -.025**
NPI total*time	**.005**	**.007**	**.002 to .010**			
Hyperactivity	**-.956**	**<.001**	**-1.274 to -.638**	**-.721**	**<.001**	**-.993 to -.450**
Hyperactivity*time	**.016**	**.016**	**003 to .030**			
Apathy	**-1.069**	**<.001**	**-1.376 to -.762**	**-.701**	**<.001**	**-.972 to -.430**
Apathy*time	**.017**	**.010**	**.004 to .030**			
Psychosis	-.015	.921	-.317 to .286	-.062	.674	-.353 to .228
Depression and anxiety	-.290	.057	-.590 to .009	**-.302**	**.040**	**-.589 to -.014**
Baseline predictor variables of the spouse						
Gender	-.548	.076	-1.154 to .059	-.566	.056	-1.148 to .015
Age	-.016	.488	-.060 to .029	-.020	.358	-.063 to .023
Self-reproach	**-.548**	**<.001**	**-.841 to -.255**	**-.567**	**<.001**	**-.876 to -.258**
Self-reproach*time				**.016**	**.047**	**.001 to .031**
Negative affect	**-.366**	**.017**	**-.667 to -.065**	-.231	.118	-.520 to .059
Active coping	-.141	.365	-.448 to .165	.088	.556	-.207 to .383
Palliative reacting	**-.537**	**<.001**	**-.836 to -.238**	**-.404**	**.006**	**-.692 to -.115**
Avoidance	-.190	.215	-.493 to .112	-.151	.307	-.442 to .140
Seeking social support	-.281	.064	-.577 to .016	**-.370**	**.012**	**-.658 to -.082**
Reassuring thoughts	-.025	.868	-.327 to .276	.203	.170	-.088 to .494
Passive reacting	**-.427**	**.005**	**-.725 to -.129**	**-.325**	**.027**	**-.614 to -.037**
Expression of emotions	-.286	.063	-.587 to .016	**-.529**	**<.001**	**-.881 to -.248**

Reported estimates are standardized effects. Significant effects are displayed in **bold**. *ADL = daily living activities.*

Discussion

This relatively large longitudinal study showed that on average, spouses of people with YOD grade their relationship quality as merely satisfactory as they grade the relationship with a six on a scale between one and ten. Over 24 months, the perceived quality of the relationship was found to deteriorate in general. Higher levels of apathy, higher levels of hyperactivity, a presence of FTD, symptom duration, lower levels of awareness of deficits, lower levels of initiative toward performing daily living activities, and higher levels of depression and anxiety in the person with YOD were associated with a lower perceived quality of the relationship by the spouse. In addition, a coping style characterized by palliative and passive reaction patterns, seeking for social support, and the expression of emotions in the spouse was associated to a lower perceived quality of the relationship. Higher levels of neuroticism (both self-reproach and negative affect) were also associated to a lower quality of the relationship.

Interpretation

Our quantitative results correspond with previous qualitative findings (Kimura et al., 2015; Lockeridge and Simpson, 2013; Massimo et al., 2013; Riedijk et al., 2008) and the review of Holdsworth and McCabe (2018) that spouses of people with YOD perceive a statistically significant deterioration in the quality of the relationship over time. However, in our study, the absolute average deterioration was relatively small and relationship quality was still graded as merely satisfactory over the course of the disease. Indeed, previous research indicates that spouses of persons with YOD often report to still love their husband or wife (Massimo et al., 2013), were able to get along with the person with YOD, and experience relatively high levels of emotional closeness, similar to spouses of persons with LOD (de Vugt et al., 2003; Riedijk et al., 2008). However, several characteristics of persons with YOD and spouses were associated to a lower quality of the relationship. For example, neuropsychiatric symptoms such as apathy and hyperactivity can be difficult to understand for spouses (Ducharme et al., 2013; Massimo et al., 2013; Paton et al., 2004). In previous qualitative research, caregivers acknowledged to miss their comrade due to apathy and experienced a decline in reciprocity in the person with YOD (de Vugt et al., 2003; Millenaar et al., 2017). Hyperactivity is characterized by agitation, euphoria, disinhibition, irritability, and aberrant motor behavior. These symptoms may cause inappropriate behavior of the person with YOD and result in anger in the spouse that diminishes the quality of the relationship (Massimo et al., 2013).

In contrast with previous research on LOD (de Vugt et al., 2003), in our study, hyperactivity in persons with YOD was associated to a lower quality of the relationship by the spouses. The higher prevalence of FTD in our younger sample might explain why hyperactivity was associated to a lower quality of the relationship because FTD is characterized by stereotypical behaviors, disinhibition, and more distress related to neuropsychiatric symptoms (Bozeat et al., 2000; de Vugt et al., 2006). Next, a diagnosis of FTD and a lower level of deficit awareness by the person with YOD were also associated to a lower quality of the relationship perceived by the spouse. This confirms earlier findings that a lack of disease awareness in the person with YOD is linked to a lower quality of life in caregivers (Baptista et al., 2016) and more unmet needs and difficulties to arrange care and support by caregivers (Millenaar et al., 2017). A lower quality of the relationship was also associated to lower levels of initiative toward daily living activities in the person with YOD. In contrast, the assistance a person with YOD required in order to perform daily living activities was not associated to the quality of the relationship. It is conceivable that spouses have more understanding for the inability to perform daily living activities in a person with YOD and are less understanding of the lack of initiative toward daily living activities. A lack of initiative in persons with YOD can be difficult to understand for spouses because it is not often attributed to dementia (Paton et al., 2004). As a result, spouses might blame the person with YOD for not showing enough initiative.

Subsequently, a higher level of self-reproach in the spouse was associated to a lower quality of the relationship by the spouse. Self-reproach, a subdomain of neuroticism, includes feelings of inferiority, worthlessness, helplessness, and being ashamed (Chapman, 2007). Previous research has shown that dementia caregivers with higher levels of neuroticism more often apply nonadaptive coping strategies (de Vugt et al., 2004). The use of these nonadaptive coping strategies might result in a higher burden, more frustration, and perhaps more relational conflict. This could explain why spouses who experience higher levels of self-reproach report a lower quality of the relationship. In addition, palliative and passive reacting patterns used by spouses were associated to a lower quality of the relationship. Surprisingly, seeking social support and the expression of emotions by spouses were also associated to a lower quality of the relationship. This might be due to reversed causality. Spouses who perceive the quality of their relationship as low might seek more social support from friends and relatives to express their emotions and cope with the situation (Knippenberg et al., 2018; Lockeridge and Simpson, 2013)

System theory might provide a framework to better understand the complex interactions and interrelationships between aspects of the spousal relationship and YOD. Dementia is known to influence the emotional connection between the person with dementia and caregivers (Chapman et al., 2019). As social relationships are continuously redefined, a loss in the emotional connection with the person with YOD may deteriorate the quality of the relationship perceived by spousal caregivers. Over time, spouses might learn to come to terms with the situation and develop a positive attitude toward their caregiving role (Lloyd et al., 2016). A positive attitude toward the caregiving role and the development of an adaptive coping strategy might help spouses to cope with changes in their relationship (Lockeridge and Simpson, 2013). This might explain why spouses of people with YOD were found to feel relatively satisfied with regard to their emotional closeness and getting along.

The development of a more positive attitude toward the caregiving role or an adaptive coping strategy might explain why the influence of apathy and hyperactivity in the person with YOD diminished over time. Another explanation for the diminishing influence of apathy and hyperactivity might be that we only used baseline measurements of these predictor variables. The progressive nature of YOD and the fluctuating course of neuropsychiatric symptoms might have weakened the association between the baseline predictors and perceived quality of the relationship over time.

Strengths and limitations

A diverse population of people with YOD and their relatives was followed over time and the longitudinal design with repeated assessments allowed us to investigate changes in the quality of the relationship over time and identify factors that are associated to the quality of the relationship. Our study findings correspond with previous and international literature, which increases the likelihood of external validity and generalizability.

Although we intended to examine predictors of perceived quality of the relationship over time, the associations we found could be bidirectional. In terms of causality, this means that a lower relationship quality can also influence, for example, the behavior of the person with YOD or coping mechanisms of the spouse. Also, some of the effects might have occurred prior to inclusion while other effects might still occur after the 24-month follow-up.

In addition, there seems to be a lack of validated measurement tools to examine the quality of the relationship in a person with dementia and their spouse. In this study, we could not explore the perceived quality of the relationship as it was experienced by the person with YOD, as in the NeedYD study only the spouse provided information about the quality of the relationship. The quality of the relationship experienced by the person with YOD might influence their behavior and thus have an effect on the perceived quality of the relationship by the spouse. Future studies that examine the quality of the relationship should take the view of the person with YOD into account.

The use of multiple statistical tests in our study increased the risk of a type 1 error. However, the results of our explorative study correspond with previous qualitative literature results (Holdsworth and McCabe, 2018), which creates a certain degree of confidence in the conclusions presented here.

Practical implications

Our findings can tailor interventions aimed at maintaining or improving the quality of the relationship in couples with YOD. Departing from a social perspective on health, persons with YOD and their spouses can still experience well-being and a high quality of life (Huber et al., 2011; de Vugt and Dröes, 2016). Helping spouses to accept or come to terms with factors that threaten their sense of couplehood may help them to develop a more positive attitude toward their spousal relationship and improve the quality of the relationship and care.

Conclusion

This study revealed that spouses of people with YOD experience a deterioration in their perceived quality of the relationship over 24 months. This deterioration was found to occur in various relationship domains and was associated with a broad set of characteristics of both the person with YOD and the spouse. Our findings can tailor interventions aimed at maintaining or improving the quality of the relationship in couples with YOD.

Conflict of interest

None.

Description of authors' roles

J. Bruinsma conducted the analyses and wrote the paper in close collaboration with K. Peetoom, S. Köhler, J. Millenaar, F. Verhey, and M. de Vugt. Other authors had a substantial contribution to the design and data collection of the NeedYD study and critically revised this manuscript.

References

Aalten, P., Verhey, F. R., Boziki, M., Bullock, R., Byrne, E. J., Camus, V., ... and Robert, P. H. (2008). Consistency of neuropsychiatric syndromes across dementias: results from the European Alzheimer Disease Consortium. Part II. *Dement Geriatr Cogn Disord, 25*, 1-8. doi:10.1159/000111082

American Psychiatry Association. (2000). Diagnostic and statistical manual of mental disorders DSM-IV-TR (Tekst revision). Washington: *Americal Psychiatry Association.*

Bakker, C., de Vugt, M. E., van Vliet, D., Verhey, F. R., Pijnenburg, Y. A., Vernooij-Dassen, M. J., and Koopmans, R. T. (2013a). Predictors of the time to institutionalization in young- versus late-onset dementia: results from the needs in young onset dementia (NeedYD) study. *J Am Med Dir Assoc, 14*(4), 248-253. doi:10.1016/j.jamda.2012.09.011

Baptista, M. A. Baptista, M. A. T., Santos, R. L., Kimura, N., Lacerda, I. B., Johannenssen, A., Barca, M. L., ... and Dourado, M. C. N. (2016). Quality of life in young onset dementia: an updated systematic review. *Trends Psychiatry Psychother, 38,* 6-13. doi:10.1590/2237-6089-2015-0049

Bozeat, S., Gregory, C. A., Lambon Ralph, M. A., and Hodges J. R. (2000). Which neuropsychiatric and behavioural features distinguish frontal an dtemporal variants of frontotemporal dementia from Alzheimer’s disease? *J Neurol Neurosurg Psychiatry, 69*, 178-186. doi:10.1136/jnnp.69.2.178

CBO. (2005). Guideline diagnosis and pharmacological treatment of dementia. Alphen aan den Rijn: *Van Zuiden Communications B.V.*

Chapman, B. P. (2007). Bandwidth and fidelity on the NEO-Five Factor Inventory: replicability and reliability of Saucier's (1998) item cluster subcomponents. *J Pers Assess, 88*(2), 220-234. doi:10.1080/00223890701268082

Chapman, M., Philip, J., and Komesaroff, P. (2019). Towards an Ecology of Dementia: A Manifesto. *J Bioeth Inq, 16*(2), 209-216. doi:10.1007/s11673-019-09910-8

Costa, P. T. and McCrae, R. R. (1989). The NEO-PI/NEO-FFI Manual Supplement. Odessa: *Psychological Assessment Resources.*

Cummings, J. L. (1997). The Neuropsychiatric Inventory- Assessing psychopathology in dementia patients. *Neurology*, *48.*

de Vugt, M. E., and Droes, R. M. (2017). Social health in dementia. Towards a positive dementia discourse. *Aging Ment Health, 21*(1), 1–3. doi:10.1080/13607863.2016.1262822

de Vugt, M. E., Stevens, F., Aalten, P., Lousberg, R., Jaspers, N., Winkens, I., ... and Verhey, F. R. (2003). Behavioural disturbances in dementia patients and quality of the marital relationship. *Int J Geriatr Psychiatry, 1*(2) 149-154. doi:10.1002/gps.807

de Vugt, M. E., Stevens, F., Aalten, P., Lousberg, R., Jaspers, N., Winkens, I., Jolles, J., and Verhey, F. R. (2004). Do caregiver management strategies influence patient behaviour in dementia? *Int J Geriatr Psychiatry, 19*(1), 85-92. doi:10.1002/gps.1044

de Vugt, M. E., Riedijk, S. R., Aalten, P., Tibben, A., van Swieten, J. C., and Verhey, F. R. (2006). Impact of behavioural problems on spousal caregivers: a comparison between Alzheimer's disease and frontotemporal dementia. *Dement Geriatr Cogn Disord*, *22*(1), 35-41. doi:10.1159/000093102

Draper, B. and Withall, A. (2016). Young onset dementia. *Int Medical J, 46*(7), 779-786. doi:10.1111/imj.13099

Ducharme, F., Kergoat, M.-J., Antoine, P., Pasquier, F. and Coulombe, R. (2013). The Unique Experience of Spouses in Early-Onset Dementia. *Am J Alzheimer's Dis Other Dement, 28*(6), 634-641. doi:10.1177/1533317513494443

Erkinjuntti, T. (1994). Clinical criteria for vascular dementia: The NINDS-AIREN criteria. *Dementia*, *5*(3-4), 189-192. doi:10.1159/000106721

Holdsworth, K., and McCabe, M. (2018). The impact of younger-onset dementia on relationships, intimacy, and sexuality in midlife couples: a systematic review. *Int Psychogeriatr, 30*(1), 15-29. doi:10.1017/S1041610217001806

Heck, R. H., Thomas, S. L. and Tabata, L. N. (2012). Multilevel Modeling of Categorical Outcomes Using IBM SPSS. New York: *Taylor & Francis Group.*

Huber, M., Knottnerus, J. A., Green, L., Van Der Horst, H., Jadad, A. R., Kromhout, D., ... and Smid, H. (2011). Health how should we define it? *BMJ, 343*, 235-237 doi:10.1136/bmj.d4163

Kimura, N. R., Maffioletti, V. L., Santos, R. L., Baptista, M. A. and Dourado, M. C. (2015). Psychosocial impact of early onset dementia among caregivers. *Trends Psychiatry Psychother*, *37*(4), 213-219. doi:10.1590/2237-6089-2015-0038

Kat, M. G., de Jonghe, J. F. M., Aalten, P., Kalisvaart, C. J., Dres, R. M. and Verhey, F. R. J. (2002). Neuropsychiatrische symptomen bij dementie: Psychometrische aspecten van de Nederlandse Neuropsychiatric Inventory (NPI). Tijdschrift voor Gerontologie en Geriatrie, 33, 150–155.

Knippenberg, R. J. M., de Vugt, M. E., Ponds, R. W., Verhey, F. R. J. and Myin-Germeys, I. (2018). Emotional reactivity to daily life stress in spousal caregivers of people with dementia: An experience sampling study. PLos One, 13(4). doi:10.1371/journal.pone.0194118

Lawrence, R. H., Tennstedt, S. L. and Assmann, S. F. (1998). Quality of the caregiver–care recipient relationship: Does it offset negative consequences of caregiving for family caregivers? *Psychol Aging, 13*(1), 150-158. doi:10.1037//0882-7974.13.1.150

Lloyd, J., Patterson, T., and Muers, J. (2016) The positive aspects of caregiving in dementia: A critical review of qualitative literature. *Dementia, 15*(6), 1534-1561. doi:10.1177/1471301214564792

Lockeridge, S. and Simpson, J. (2013). The experience of caring for a partner with young onset dementia: how younger carers cope. *Dementia, 12*(5), 635-651. doi:10.1177/1471301212440873

Massimo, L., Evans, L. K. and Benner, P. (2013). Caring for loved ones with frontotemporal degeneration: the lived experiences of spouses. *Geriatr Nurs, 34(*4), 302-306. doi:10.1016/j.gerinurse.2013.05.001

McKeith, I. G. (2006). Consensus guidelines for the clinical and pathologic diagnosis of dementia with Lewy bodies (DLB): report of the Consortium on DLB International Workshop. *J Alzheimer's Dis, 9*(s3), 417-423. doi:10.3233/jad-2006-9s347

McKhann, G., Drachman, D., Folstein, M., Katzman, R., Price, D., and Stadlan, E.M. (1984). Clinical diagnosis of Alzheimer's disease: report of the NINCDS-ADRDA Work Group under the auspices of Department of Health and Human Services Task Force on Alzheimer's Disease. *Neurology, 34*(7), 939-944. doi:10.1212/WNL.34.7.939

Mesulam, M. M., Grossman, M., Hillis, A., Kertesz, A., and Weintraub, S. (2003) The core and halo of primary progressive aphasia and semantic dementia. *Ann Neurol*, *54*(s5), S11-14. doi:10.1002/ana.10569

Millenaar, J. K., Bakker, C., van Vliet, D., Koopmans, R. T., Kurz, A., Verhey, F. R., and de Vugt, M. E. (2017). Exploring perspectives of young onset dementia caregivers with high versus low unmet needs. *International Journal of Geriatric Psychiatry*, *33*, 340-347. doi:10.1002/gps.4749

Miller, E. A., Rosenheck, R. A., and Schneider L. S. (2012). Caregiver burden, health utilities, and institutional service use in Alzheimer's disease. *Int J Geriatr Psychiatry, 27*(4), 382-393. doi:10.1002/gps.2730

Neary, D., Snowden, J. S., Gustafson, L., Passant, U., Stuss, D., Black, S. A. S. A., ... and Benson, D. F. (1998). Frontotemporal lobar degeneration: a consensus on clinical diagnostic criteria. *Neurology, 51*(6), 1546-1554. doi:10.1212/wnl.51.6.1546

Paton, J., Johnston, K., Katona, C., and Livingston, G. (2004). What causes problems in Alzheimer's disease: attributions by caregivers. A qualitative study. *Int J geriatr psychiatry, 19*(6), *527-532.* doi:10.1002/gps.1118

Reisberg, B., Ferris, S. H., de Leon, M. J. and Crook, T. (1982). The global deterioration scale for assessment of primary degenerative dementia. *Am J Psychiatry*, 139, 1136-1139. doi:10.1176/ajp.139.9.1136

Riedijk, S., Duivenvoorden, H., Rosso, S., Van Swieten, J., Niermeijer, M. and Tibben, A. (2008). Frontotemporal Dementia: Change of Familial Caregiver Burden and Partner Relation in a Dutch Cohort of 63 Patients. *Dement Geriatr Cogn Disord*, *26*(5), 398-406. doi:10.1159/000164276

Saucier, G. (1998). Replicable item-cluster subcomponents in the NEO Five-Factor Inventory. *J Pers Assess*, *70*(2), 263-276. doi:10.1207/s15327752jpa7002_6

Schreurs, P. J. G., van de Willige, G., Brosschot, J. F., Tellegen, B. and Graus, G. M. H. (1993). De Utrechtse Coping Lijst: UCL. Lisse: *Swets en Zeitlinger*.

Teunisse, S. and Derix, M. M. A. (1997). The Interview for Deterioration in Daily Living Activities in Dementia: Agreement Between Primary and Secondary Caregivers. *Int Psychogeriatr*, *9*(4), 155-162. doi:10.1017/S1041610297004845

Twisk, J. W. R. (2007). Applied Logitudinal Data Analysis for Epidemiology. Cambridge: Cambridge University Press.

Vangelisti, A., and Perlman, D. (2006). Cambridge handbook of personal relationships. Cambridge: *Cambridge University Press.*

van Vliet, D., Bakker, C., Koopmans, R. T., Vernooij-Dassen, M. J., Verhey, F. R., and de Vugt, M. E. (2010). Research protocol of the NeedYD-study (Needs in Young onset Dementia): a prospective cohort study on the needs and course of early onset dementia. *BMC Geriatr, 10*, 1-8. doi:10.1186/1471-2318-10-13

van Vliet, D., de Vugt, M. E., Bakker, C., Koopmans, R. T., Pijnenburg, Y. A., Vernooij-Dassen, M. J., and Verhey, F. R. (2011). Caregivers' perspectives on the pre-diagnostic period in early onset dementia: a long and winding road. *Int Psychogeriatr, 23*(9), 1393-1404. doi:10.1017/S1041610211001013

Verhey, F. R. J., Rozendaal, N., Ponds, R. W. H. M. and Jolles, J. (1993). Dementia, awareness and depression. *Int J Geriatr Psychiatry*, *8*(10), 851-856. doi:10.1002/gps.930081008

von Elm, E., Altman, D. G., Egger, M., Pocock, S. J., Gøtzsche, P. C., Vandenbroucke, J. P., and Strobe Initiative. (2014). The Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) Statement: guidelines for reporting observational studies. *Int J Surg*, *12*(12), 1495-1499. doi:10.1016/j.jclinepi.2007.11.008

Wawrziczny, E., Antoine, P., Ducharme, F., Kergoat M. J. and Pasquier, F. (2016). Couples' experiences with early-onset dementie: An interpretative phenomenological analysis of dyadic dynamics. *Dementia*, *15*(5), 1082-1099. doi:10.1177/1471301214554720.

Chapter 3

“They simply do not understand”: a focus group study exploring the lived experiences of family caregivers of people with frontotemporal dementia

Bruinsma, J., Peetoom, K., Bakker, C., L. Boots., Verhey, F., and de Vugt, M.

Aging & Mental Health, 2020

Abstract

Frontotemporal dementia (FTD) has a profound impact on the spouse and other family caregivers involved. While caregivers have a need for support, it is difficult for healthcare providers to respond to their specific needs. This qualitative study explores the lived experiences and needs of caregivers of persons with FTD to facilitate the development of support.

Methods: Three focus group discussions were organized to explore the lived experiences of Dutch FTD caregivers. The included caregivers (n = 24) were aged 16 years or older and were involved in the care of a relative with FTD. Two researchers independently performed an inductive content analysis using open and axial coding.

Results: The main category emerging from the data was a lack of recognition, acknowledgment, and understanding experienced by caregivers. This was linked to caregivers' experiences with (1) complex emotional and behavioral symptoms in the person with FTD, (2) the trivializing responses of family and friends, (3) a perceived lack of knowledge and support from healthcare professionals, and (4) the bureaucratic procedures that accompany caregiving. As a result, caregivers felt lonely and solely responsible for the caregiving role.

Conclusion: Caregivers of persons with FTD experience a lack of understanding in caring for their relative with FTD, which contributes to feelings of loneliness. A specialized support approach is needed to address the specific needs of caregivers of persons with FTD. Support should address strategies that caregivers can use to inform and involve family and friends in the caregiving situation to prevent loneliness in FTD caregivers.

Keywords: Frontotemporal dementia; Young-onset dementia; Caregivers; Lived experiences; Loneliness

Introduction

Frontotemporal dementia (FTD) has a profound impact not only on the persons with FTD themselves but also on their spouses and other family caregivers involved (Kaizik et al., 2017; Nunnemann, Kurz, Leucht, & Diehl-Schmid, 2012). Diagnostic disclosure is important for caregivers as it helps them to understand and cope with symptoms in their relative with FTD (Caceres et al., 2016; Nunnemann et al., 2012). In retrospect, caregivers often describe the prediagnostic phase as a period full of uncertainty and frustration (Caceres et al., 2016; Nunnemann et al., 2012). This is probably attributed to the prolonged time to diagnosis, as it is difficult for caregivers and healthcare professionals to recognize FTD due to the insidious onset of symptoms at young age (van Vliet et al., 2013). It is challenging to recognize FTD due to its heterogeneous clinical presentation including symptoms such as disinhibition, apathy, compulsive behavior, loss of insight, loss of empathy, aphasia, impaired semantic knowledge, loss of speech production, agrammatism, and apraxia (Bang, Spina, & Miller, 2015; Hogan et al., 2016). Behavioral and personality change and a lack of disease insight in the person with FTD may especially delay help-seeking behavior, thereby further prolonging the diagnosis (Draper et al., 2016; Rosness, Haugen, Passant, & Engedal, 2008b; van Vliet et al., 2013).

Although caregivers often already perform caregiving tasks prior to diagnosis, diagnostic disclosure marks the start of the caregiving trajectory (de Vugt & Verhey, 2013; Ducharme, Lévesque, Lachance, Kergoat, & Coulombe, 2011). Especially in the behavioral variant of FTD, caregivers often experience the loss of an emotional connection with the person with FTD and feel forced to sacrifice parts of their social life to fulfil their caregiving role. As a result, caregivers can experience social isolation and feelings of loneliness (Caceres et al., 2016; Massimo, Evans, & Benner, 2013; Nunnemann et al., 2012). In particular, coping with behavioral changes in the person with FTD may impose high levels of burden and distress in FTD caregivers (Nunnemann et al., 2012).

While FTD caregivers express a high need for support after the diagnosis, it is difficult for healthcare providers to recognize and respond to their specific needs (Barca, Thorsen, Engedal, Haugen, & Johannessen, 2014; Caceres et al., 2016; Rosness, Haugen, & Engedal, 2008a). This may be related to the low prevalence and heterogeneous clinical presentation of FTD (Onyike & Diehl-Schmid, 2013). Caregivers of persons with the behavioral variant of FTD, for example, often have to cope with challenging behavior from their relative with FTD, such as social awkwardness, a loss of manners, and egoistic behavior (Gossink et al., 2018b; Ibañez & Manes, 2012; Mendez et al., 2014). Caregivers of persons with a language variant of FTD, either non-fluent primary progressive aphasia or semantic dementia, are confronted with a profound decline in the linguistic abilities of their relative, such as difficulties with language production or loss of word meaning (Bang et al., 2015). Both changes in behavior and difficulties with communicating are known to be challenging and burdensome for caregivers (Caceres et al., 2016; Diehl-Schmid et al., 2013). While the wide variety of symptoms poses unique challenges for caregivers, there seems to be a knowledge gap regarding effective support for caregivers of persons with FTD (Gossink et al., 2018a; Karnatz et al., 2019). Therefore, this study aims to explore the lived experiences and needs of spouses and other family caregivers of persons with FTD to facilitate the development of support strategies and interventions.

Methods

In this qualitative study, focus group discussions were organized to explore lived experiences and needs of spouses and other family caregivers of persons with FTD in the Netherlands. The results are reported using the consolidated criteria for reporting qualitative research (COREQ) (Tong, Sainsbury, & Craig, 2007).

Recruitment and participants

In spring 2019, participants were recruited by (1) distributing information leaflets in waiting rooms in the memory clinic of the Maastricht University Medical Center (MUMC+), (2) addressing the focus groups during bimonthly meetings with healthcare organizations affiliated with Dutch Young-onset Dementia Knowledge Center [*Kenniscentrum Dementie op Jonge Leeftijd*], and (3) providing information via peer-support meetings, social media and the website of the Dutch FTD peer-support organization [*FTD lotgenoten*]. An ambassador of the Dutch FTD peer-support organization also actively informed caregivers about the study.

All potential participants registered via email and were then informed about the study objectives in a telephone conversation. To obtain a comprehensive understanding on how caregivers perceived caring for a relative with FTD throughout the caregiving trajectory we used purposeful sampling to include participants that varied in their level of experience, their relationship to the person with FTD (spouses, children, and other relatives), FTD subtype (behavioral and language variants), and gender. This allowed us to gain insight into experiences from caregivers in different stages of the caregiving trajectory. The focus group discussions were conducted with caregivers from two urban areas and one rural area in the Netherlands.

Caregivers were eligible for participation if they were involved in the care of their spouse, parent, or other relative with either the behavioral or a language variant of FTD. In accordance with guidelines of the medical ethical committee, caregivers younger than 16 years were excluded from participation.

All eligible participants received written information about the study by post before inclusion and were phoned to check whether they had additional questions. All participants provided informed consent before participating in the focus group discussions.

Procedures and data collection

A context mapping approach with generative techniques was used to sensitize caregivers prior to the focus groups to obtain insight into their lived experiences and to initiate discussion during the focus groups (Visser, Stappers, van der Lugt, & Sanders, 2005). In line with grounded-theory multiple techniques were used to collect data (Charmaz, 2014; Glaser & Strauss, 1967). For example, prior to their participation, caregivers completed a booklet containing questions about their everyday life such as ‘What is important for you when caring for your relative with FTD?’ and ‘What gives you positive energy in your daily life?’ In addition, caregivers reflected on their experiences by writing a letter containing advice on caregiving to peers (Boots, Wolfs, Verhey, Kemplen, & de Vugt, 2015).

Each focus group discussion was moderated by a (neuro)psychologist (MdV/CB) with clinical and research expertise regarding FTD and with experience in moderating focus group discussions. During each focus group, the process was monitored and field notes were made by the first author (JB), who has a background in nursing, psychology and health sciences. In addition, two assistants with a background in nursing or health sciences made fieldnotes as well.

To guide the focus group discussions, a topic list was developed based on the literature, qualitative interviews from the Needs in Young-onset Dementia study (NeedYD) (van Vliet et al., 2010), and expert discussions. The topic list covered (1) the prediagnostic period, (2) the postdiagnostic period, and (3) caregiver coping strategies. After each focus group discussion, a consensus meeting was organized with the research team to evaluate the data collection process and refine the topic list if needed. Each focus group discussion was video recorded to gain insights into the interactions between participants. All focus groups were transcribed verbatim using F5 transcript software.

Data analysis

To derive categories from the data, two researchers (JB/KP) independently performed an inductive content analysis by openly coding the data in Atlas.ti version 8.3.1 (Elo & Kyngäs, 2008; Evers, 2015; Savin-Baden & Major, 2013). They compared texts that were assigned to codes by constant comparison to reach consensus on the code definitions and code structure (Bradley, Curry, & Devers, 2007). Then, related codes within the code structure were merged using axial coding and both researchers independently developed a mind-map to elicit categories and subcategories from the codes. In line with grounded theory, an iterative process was used to interpret the findings and derive theory from the data using selective coding (Charmaz, 2014). First, the main category and subcategories were derived by combining the mind-maps through discussion with the last author (MdV). Next, the main category and subcategories were discussed with the wider research team to deepen the findings. Then, the results were verified by discussing the findings with the chairman of the Dutch FTD peer-support organization. Throughout the process, the first and second authors continuously checked whether new information emerged from the data to establish data saturation (Patton, 2002).

Trustworthiness of data

To ensure the trustworthiness of the data, multiple techniques to achieve method and data source triangulation were used, such as the use of fieldnotes, video recordings of the group discussions, and the use of the booklet containing questions about everyday life (Creswell, 2013). During meetings, researchers with different backgrounds reflected on the general research process, the data collection, and the results during team meetings to ensure investigator triangulation (Carter, Bryant-Lukosius, DiCenso, Blythe, & Neville, 2014).

Ethical considerations

The study protocol was approved by the Medical Ethics Committee of Maastricht University (#2018-0675).

Results

A total of 48 FTD caregivers registered for participation. Using purposeful sampling, 24 caregivers were invited for three focus group discussions (Table 1). Eight caregivers were included in the first focus group in a rural area in the Netherlands. Subsequently, two additional focus group discussions, each with eight participants, were organized in urban areas. The remaining 24 caregivers were added to a waiting list for an additional fourth focus group. Data saturation was established after the third focus group discussion as no new information emerged. Therefore, no fourth focus group discussion was organized. All focus groups were organized in the evenings or on weekends to facilitate the participation of caregivers who were working or studying. The focus groups lasted between 2 h and 2.5 h and were held in meeting rooms of one hospital and two healthcare organizations.

Participants

In the three focus groups, 17 females and seven males between the ages of 31 and 79 participated. The majority of the participants were spouses. Other family caregivers, such as children, and siblings of the person with FTD, also participated. As some were part of the same family, the 24 included caregivers were involved in the care for 20 persons with FTD; 15 with the behavioral variant and five with a language variant of FTD. On average, the first symptoms in the persons with FTD started about 9 years before the focus group discussion (range between 4 and 12 years), and the diagnosis was on average established 4 years ago (range 0.5–9 years)

Table 1. Characteristics of the family members (n = 24) of persons with FTD (n = 20)

Variable	N
Gender (n = 24)	
Male (%)	7 (29.2)
Female (%)	17 (70.8)
Age (n = 24)	
Mean (min-max)	55.7 (31-79)
Relationship to the person with FTD (n = 24)	
Spouse	16
Child	5
Child-in-law	1
Divorced from spouse with FTD	1
Sibling	1
Gender person with FTD (n = 20)	
Male (%)	13 (65.0)
Female (%)	7 (35.0)
Clinical syndrome in person with FTD (n = 20)	
Behavioral variant	15
Language variant	5
Living situation of the person with FTD (n = 20)	
Living at home	5
Living at home and using daytime facilities	3
Institutionalized	10
Person with FTD passed away	2

In total, 24 relatives of 20 persons with FTD participated in the focus group interviews.

***FTD** = Frontotemporal dementia.*

Main category: the experienced lack of recognition, acknowledgment, and understanding throughout the caregiving trajectory

After an iterative open and axial coding process, consensus was reached about the used codes and their definitions. Using selective coding iteratively theory was derived from the data by yielding one main category from the data and four subcategories (Figure 1). The data revealed that throughout the caregiving trajectory, caregivers perceived a lack of recognition, acknowledgment, and understanding not only from the person with FTD but also from family, friends, and healthcare professionals. Caregivers, for example, experienced that FTD symptoms often did not fit within the image that people have of dementia making it difficult for family, friends, and healthcare professionals to recognize symptoms of FTD. In turn, caregivers perceived a lack of understanding and felt not acknowledged in their caregiving role. This made caregivers feel solely responsible for the caregiving role and this induced feelings of loneliness. According to caregivers, this made caring for a relative with FTD particularly exhausting and stressful.

"People simply do not understand the impact of FTD on the caregivers involved. This is very difficult to cope with."
– Son-in-law, behavioral variant, FG3 –

"It is a very lonely process. I have a lot of people around me, but you can't really share it."
– Spouse, behavioral variant, FG3 –

Although caregivers stressed that there were profound differences between the behavioral and language variants of FTD, a common perception among caregivers was that they perceived a lack of recognition, acknowledgment, and understanding. This was linked to four subcategories related to caregivers' experiences with (1) complex emotional and behavioral symptoms in the person with FTD, (2) the trivializing responses of family and friends, (3) a perceived lack of knowledge and support from healthcare professionals, and (4) the bureaucratic procedures that accompanied the caregiving role. The extent to which these subcategories influenced the experienced lack of recognition, acknowledgment, and understanding varied during the three phases of the caregiving trajectory namely, recognizing early symptoms of FTD, during the diagnostic process, and while gaining access to social and professional support.

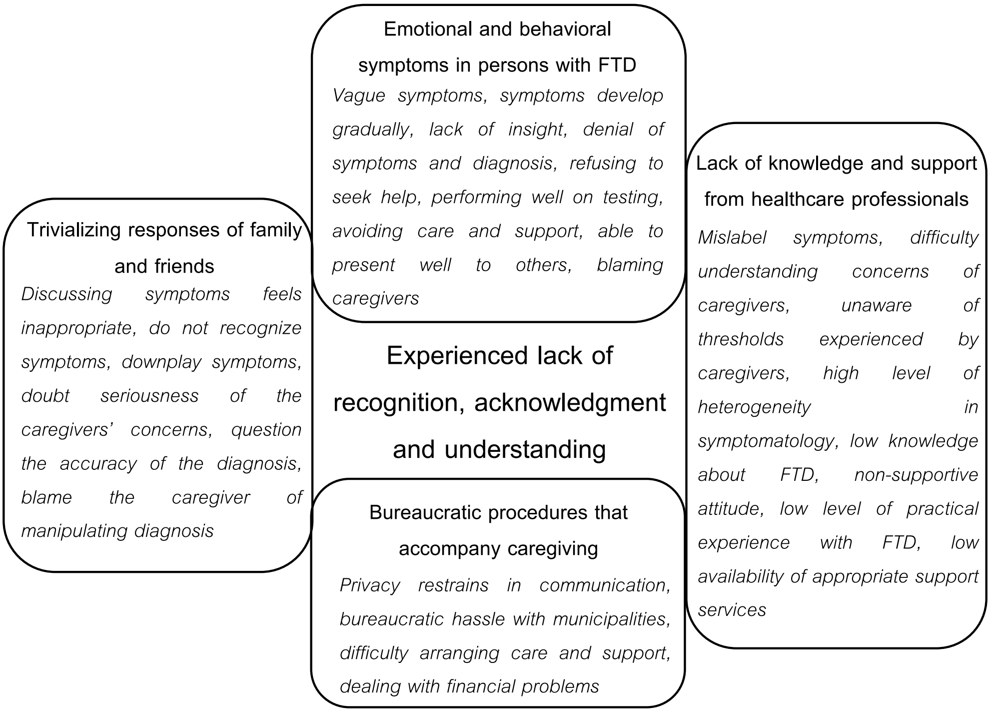

Figure 1. *Caregiver experiences throughout the caregiving trajectory*

Caregivers' experiences with the recognition of the early symptoms of FTD

In the first phase of the caregiver trajectory, caregivers experienced egoistic, paranoid, jealous, impulsive, stereotypic, and apathetic behavior as early symptoms in their relative with FTD. Caregivers of persons with a language variant of FTD described apathy, a loss of semantic knowledge, mutism, and difficulty speaking fluently. This made caregivers feel that their relationship with the person with FTD deteriorated because there was less reciprocity and interaction.

Most caregivers experienced difficulties with recognizing and understanding these symptoms as they developed gradually over time. According to caregivers of persons with the behavioral variant, recognizing early symptoms was complicated due to a lack of disease insight in the person with FTD, who often denied that something was wrong. This made caregivers feel insecure about the seriousness of the symptoms and sometimes caregivers felt that they themselves were to blame for the emotional and behavioral changes in their relative with FTD.

“The behavioral change was going on for years. You have no idea what is going on. You think you are the one to blame.”

– Spouse, behavioral variant, FG1 –

Caregivers also expressed that it was difficult to discuss their concerns about early symptoms with their family and friends. They suggested this might be due to feelings of guilt and shame, as they perceived it as complaining or bad-mouthing their relative with FTD.

“My father lost the ability to speak. (…) In our cultural community, they thought he did something bad. They thought the symptoms were supernatural or that it was voodoo. This was very stressful for our family. So, you simply do not talk about it. Perhaps, this is due to shame.”

– Daughter, language variant, FG1 –

“I always felt that something was wrong. (…) I was worried, but I could not go anywhere with my worries.”

– Spouse, behavioral variant, FG3 –

In addition, trivializing responses from family and friends downplaying the severity of the caregivers’ concerns made caregivers feel as if they had overreacted to the situation. This made caregivers doubt the seriousness of their concerns.

“When people came to visit us, they said ‘He seems to be doing very well’. This made me doubt.”

– Spouse, behavioral variant, FG2 –

Caregivers perceived that it was difficult to discuss their concerns with family and friends who often did not recognize symptoms in the person with FTD, such as apathy, agitation, or compulsive behavior. As a result, caregivers missed social support and this made them feel unrecognized in coping with early symptoms. As a result, many caregivers expressed to feel lonely.

Caregivers also experienced that early symptoms of FTD being regularly mislabeled as work-related distress or marital problems by employers and occupational physicians. Looking back, caregivers perceived a lack of knowledge in healthcare professionals regarding FTD and felt this had contributed to a delayed diagnosis. In addition, caregivers felt frustrated by privacy restraints that hindered communication with the employer and occupational physician of their relative with FTD.

“When I asked the occupational physician what was going on, he said, ‘Privacy. I can only tell you that stress does strange things to people’.”

– Spouse, behavioral variant, FG2 –

Caregivers perceived that the unwillingness of the occupational physician to disclose any information about problems at work felt as an unnecessary bureaucratic barrier, and it significantly delayed the start of the diagnostic process and therefore obstructed gaining access to professional help and support.

Caregivers' experiences with the diagnostic process and diagnostic disclosure

In the diagnostic process, caregivers perceived several barriers that delayed help seeking. Caregivers explained they postponed visiting the general practitioner (GP), as the person with FTD denied symptoms and refused to go to the GP. Some caregivers visited the GP alone, while others eventually succeeded in persuading the person with FTD to visit the GP by, for example, asking their children to accompany the person with FTD. Caregivers also postponed visiting the GP because of the trivializing responses of family and friends who often doubted the seriousness of symptoms. However, some caregivers indicated that on some occasion's family or friends also noticed early symptoms and motivated them to reach out for professional help.

> *"Everybody in our environment had an opinion about what is going on. (…) This is why you do not discuss the situation."*
> – Daughter, language variant, FG1 –

> *"My daughter took a year off to travel. When she came home, she felt like, 'What happened here?' This is why we visited the general practitioner."*
> – Spouse, behavioral variant, FG3 –

According to caregivers, GPs often had difficulty understanding the caregivers' concerns because the persons with FTD often presented themselves well to GPs and performed well on neuropsychological testing. Caregivers acknowledged that it was difficult for GPs to recognize FTD because symptoms are difficult to interpret and developed gradually over time. They also felt that early recognition was complicated by work-related stress, marital problems, or comorbid disease, such as a neurological pain disorder, low-back, problems, or multiple sclerosis. Additionally, caregivers felt that GPs were often unaware of the threshold perceived by caregivers in talking openly about emotional and behavioral changes in their relative with FTD, especially when caregivers and persons with FTD both consulted the GP. Some GPs, for example, confronted the person with FTD with the caregivers' concerns during a consult, which made caregivers feel misunderstood and unsupported. Caregivers also felt that GPs should have had more knowledge about the early symptoms of FTD, as this would have fostered the early recognition of symptoms.

"It feels bad to talk about your spouse like this with the GP. Especially when he kept asking my husband, 'How do you feel if she [caregiver] says all these things about you?'."
– Spouse, behavioral variant, FG3 –

Looking back at the diagnostic process, some caregivers also expressed highly positive experiences with regard to the role of their GP. Some caregivers, for example, felt that the GP took their concerns seriously, acted very supportively, and seemed highly motivated to determine what was going on. This made caregivers feel heard and recognized.

"Our GP was very supportive from the beginning but lacked knowledge about FTD. GPs should have more knowledge about the early symptoms, but this is difficult because GPs already need to know so much."
– Spouse, language variant, FG2 –

According to caregivers, the FTD diagnosis helped them to better understand the emotional and behavioral changes in their relative with FTD. Diagnostic disclosure led to a sense of recognition among caregivers. Some felt relieved that pieces of the puzzle had finally fallen into place. Receiving the diagnosis also helped caregivers address and explain the situation to family and friends.

"I thought 'If this continues, then I will quit'. In that sense, the diagnosis was a blessing. You finally get it and think, 'Okay, for better and for worse'. This is why I continue."
– Spouse, behavioral variant, FG2 –

"The diagnosis enabled me to tell my children why their father acted like this. That it was his illness and not because he did not love them anymore."
– Spouse, behavioral variant, FG2 –

Some caregivers explained that family members said they had doubts about whether the diagnosis was correct. Some caregivers of persons with the behavioral variant of FTD, explained that family sometimes accused them of manipulating the diagnosis. As a result of these trivializing responses, caregivers felt sad and abandoned by family and friends. This made caregivers feel solely responsible for the caregiving role.

"My family and my sisters think that I manipulated the diagnosis. They think that there is nothing going on with my husband. They even said that I should see a doctor. This makes me really sad."
– Spouse, behavioral variant, FG3 –

"You have to cope with all of this by yourself. You need to find some kind of best practice for all the shit you go through."

– Spouse, behavioral variant, FG2 –

Caregivers' experiences with gaining access to social and professional support

Caregivers stated that emotional, behavioral, and language symptoms progressed in the phase after the diagnosis. As a result, caregivers felt that they were losing the emotional connection with their relative. For example, due to increasing levels of apathy and a lack of empathy. Caregivers perceived the diagnostic process as an emotional burden and experienced feelings of sadness and grief as they felt they were gradually losing their relative with FTD.

"He is still here, but he no longer behaves as my husband. Losing your spouse really hurts."

– Spouse, behavioral variant, FG2 –

Caregivers explained this was frustrating for them to cope with the lack of awareness of the disease in their relative with FTD, who in turn, devalued, neglected, and underestimated the impact of their caregiving role. As a result, they felt unrecognized as caregivers. Most caregivers acknowledged that social support helped them to cope with stress and recharge their battery, but it was difficult to gain sufficient support from family and friends after diagnosis. Involving family and friends in the caregiving situation was experienced as exhausting as they were often unable to recognize symptoms and acknowledge the caregivers' needs, despite the diagnosis.

"When people say, 'He visited us but we did not notice anything'. I always think, 'What are you trying to say? That I am crazy?'."

– Spouse, behavioral variant, FG3 –

"Sometimes I feel completely upset. Then, my children tell me, 'It is not that bad.' (…) They simply do not understand. This is why being a caregiver is lonely."

– Spouse, behavioral variant, FG3 –

Caregivers perceived this as a lack of understanding from family and friends. This made caregivers feel unsupported and lonely. In turn, caregivers felt it was very difficult to achieve a sense of balance in their life and learn to adjust to combining caregiving with having a life of their own.

"I was very tired and stressed when my husband still lived at home. (…) I learned that it is very important to find a balance and think about yourself."

– Spouse, behavioral variant, FG2 –

Caregivers often experienced that it was difficult for GPs, psychologists, and specialized dementia nurses to provide practical advice about coping with emotional and behavioral change in the person with FTD. As a result, caregivers felt unrecognized and misunderstood by healthcare providers and postponed the use of care and support services. Due to the lack of appropriate support services for FTD caregivers, they stressed the importance of peer-support groups to elicit a sense of recognition and understanding. During the focus groups, some caregivers expressed that they only needed half a word to understand other FTD caregivers. When it came to professional support and peer-support, child caregivers expressed feeling left in the dark.

"As a daughter, there was no support for me at all. I visited a peer-support group once, but everyone there was a spouse. I was hard for me to connect with them."
– Daughter, language variant, FG1 –

Caregivers also expressed frustration with the bureaucratic procedures that accompanied the caregiving role, such as struggles with the municipality to receive the postal mail, and applying for personal care funding. Some caregivers explained that they had lost their specialized dementia nurse and peer-support group because their relative with FTD became institutionalized. Caregivers explained it was frustrating to feel unsupported, neglected, and misunderstood by healthcare providers.

"I do not understand why I am not allowed to visit peer-support meetings anymore now that my husband is hospitalized. All of a sudden, your support is gone."
–Spouse, language variant, FG1 –

Some caregivers of persons with the behavioral variant also explained that they ended up with serious financial problems as a result of their relative having FTD. After the diagnosis, some caregivers found that the person with FTD was gambling, made risky investments, or had refused to pay the bills. Coping with financial difficulties was often frustrating for caregivers, and arranging financial custody could only be achieved by following lengthy procedures. This generally resulted in increased financial debts.

"My parents are in deep financial debt and may lose their house. We are unable to put my dad [the person with FTD] under financial supervision due to the paperwork. The process is really frustrating."
– Daughter, behavioral variant, FG3 –

At the same time, caregivers experienced little compassion from financial institutions such as banks and debt collectors, who often did not understand the impact of FTD. As a result, caregivers felt unacknowledged and misunderstood.

> *"The frustrating part is that no one has any understanding. If he [the person with FTD] would stab and kill someone, they would probably declare him unaccountable. With regard to financial debts, they do not. You are all alone with those debt collectors breathing down your neck."*
>
> – Son-in-law, behavioral variant, FG1 –

Discussion

Our study demonstrates that FTD caregivers experience a lack of recognition, acknowledgment and understanding while caring for their relative with FTD. According to caregivers, FTD symptoms often do not fit within the image people have of dementia. In turn, they felt that emotional and behavioral symptoms were difficult to recognize for family, friends, and healthcare professionals. This made caregivers feel that no one understands them and this resulted in feelings of loneliness.

In the early stages of the caregiving trajectory, caregivers themselves also had difficulty recognizing and understanding the early symptoms of FTD, as these often occurred at a young age and the severity of symptoms gradually increased over time. This may be specific to FTD, as 70–80% of the persons with FTD develop symptoms well before the age of 65 (Knopman & Roberts, 2011; Rabinovici & Miller, 2010). While Alzheimer's dementia (AD) is still the most prevalent cause of young-onset dementia (YOD) (Rossor, Fox, Mummery, Schott, & Warren, 2010), only 5% of all persons with AD have a symptom onset at a young age (Zhu et al., 2015). In FTD, there is a longer delay in diagnosis compared to YOD caused by AD, ranging between 4.1 and 6.4 years (Rosness et al., 2008b; van Vliet et al., 2013). This has important negative consequences for caregivers as diagnostic delay is associated with high levels of uncertainty, caregiver distress, family or marital conflict, and even financial problems in the period prior to diagnosis (van Vliet et al., 2011). Caregivers may experience several internal and external barriers that delay the diagnosis (van Vliet et al., 2011; van Vliet et al., 2013). Specific internal factors for caregivers of persons with FTD are, for example, feelings of uncertainty whether or not the symptoms in their relative need to be taken seriously. In line with van Vliet et al. (2013), trivializing remarks from family, friends, and healthcare professionals seemed to enhance these feelings of uncertainty. In turn, caregivers felt they delayed their decision to seek professional help.

Prediagnostic uncertainty further increased by typical characteristics of the behavioral variant of FTD, such as changes in character and behavior and a lack of insight, as these symptoms are often not associated with dementia by healthcare professionals (Rosness et al., 2008b; Spreadbury & Kipps, 2018; van Vliet et al., 2011). In particular, the behavioral variant of FTD is difficult to recognize for healthcare professionals due to the overlapping symptomatology with psychiatric disorders (Gossink et al., 2016). Our findings also demonstrate that comorbid disease made it difficult for healthcare professionals to interpret early symptoms of FTD. Early recognition is further hampered by the low prevalence of FTD (Onyike & Diehl-Schmid, 2013; Rossor et al., 2010). Obtaining a timely diagnosis is important as it helps caregivers with adapting to the caregiving role (de Vugt & Verhey, 2013). In our study, caregivers perceived the diagnosis as an important milestone because it allowed them to understand the changes in their relative with FTD. Nevertheless, caregivers of persons with the behavior variant of FTD felt that family and friends often continued to struggle with recognizing symptoms. It is known that emotional and behavioral symptoms of dementia are often misattributed to environmental factors or to fall within personal control (Polenick et al., 2018). Although family, friends, and healthcare professionals misattributed symptoms in all variants of FTD, this might occur more often in the behavioral variant which is characterized by the inability to empathize with others, recognize emotions, and comply with social norms (Harciarek & Cosentino, 2013; Ibañez & Manes, 2012). These symptoms also disrupt the relational connection between the caregiver and the person with FTD and are difficult to recognize for family and friends (Massimo et al., 2013; Polenick et al., 2018). In line with Vasileiou et al. (2017), this resulted in feelings of loneliness in caregivers.

Caregivers in our study confirmed that there is a need for support in the postdiagnostic phase (Rosness et al., 2008a; Spreadbury & Kipps, 2018). According to caregivers there is only limited appropriate support available and healthcare professionals often have little knowledge about FTD and the challenges that caregivers face. These negative experiences with healthcare will lower the confidence that caregivers have in professional care and may increase the risk for delay in initiation of care and support services (Rabanal, Chatwin, Walker, O'Sullivan, & Williamson, 2018; Sikes & Hall, 2018). Particularly in the behavioral variant of FTD this is problematic given the high levels of burden and distress experienced by caregivers (Mioshi et al., 2013). Therefore, specialized support services and educational programs on FTD for healthcare professionals seem needed to improve postdiagnostic care in FTD (Rabanal et al., 2018; Sikes & Hall, 2018).

Strengths and limitations

A strength of our study is that we recruited a heterogeneous sample with spouses and other family caregivers who were in different phases of the caregiving trajectory. This provides a comprehensive picture of the lived experiences of FTD caregivers while they engage in the caregiving role. Focus group discussions require caregivers to talk openly about their experiences in front of others, therefore, some caregivers may have felt hesitant to discuss certain sensitive topics. As multiple caregivers from the same family were included, some participants may have provided socially desirable answers or some experiences are overrepresented. To address this, we also asked caregivers to individually reflect on the caregiving role prior to their participation in the group discussion by completing a booklet containing questions and writing a letter to a caregiver peer. We mainly recruited participants via a Dutch peer-support group. It could be that caregivers who experience difficulty in gaining support may experience more need for peer-support. Therefore, these caregivers may be overrepresented in our study.

Future directions and conclusions

A specialized support approach is needed to improve prediagnostic and postdiagnostic care in FTD (Rosness et al., 2008a). Our findings provide directions for developing adequate support for FTD caregivers by emphasizing their need for recognition, acknowledgement, and understanding. Care and support services that elicit a sense of recognition in FTD caregivers may lower the threshold to gain timely access to support. In turn, this will facilitate the role adaptation process, which may reduce caregiver burden and distress. Healthcare professionals could play an important role to reduce barriers that caregivers experience when gaining social support (Dam, Boots, Boxtel, Verhey, & de Vugt, 2018). Our study adds that healthcare professionals can achieve this by reducing feelings of shame and stigma and by supporting FTD caregivers with actively informing and involving family and friends in the caregiving situation. The medical training of healthcare professionals on FTD seems a prerequisite for facilitating adequate postdiagnostic support to FTD caregivers. Additionally, our findings underscore the importance of peer-support as recognition from peers made caregivers feel heard and reduced feelings of loneliness. As FTD does not fit within the general image people have of dementia, peer-support groups that specifically include caregivers of persons with FTD seem needed to elicit a sense of recognition. Specialized support programs may also be beneficial for FTD caregivers by increasing sense of competence and decreasing burden and distress (Gossink et al., 2018a). Given the active life phase of younger caregivers and the low prevalence of FTD, online peer-support and web-based psychosocial support may provide an opportunity to support FTD caregivers, even in remote areas (Diehl-Schmid et al., 2013). Our findings show that FTD caregivers often postpone the use of professional healthcare services. Several factors could lower barriers to access support, such as designing tailored support programs that meet the specific needs of FYD caregivers and offering support also outside the healthcare sector, for example, by online programs that can be accessed at a time and place that is convenient for FTD caregivers. Allowing timely access to support may help caregivers of persons with FTD not only to gain more understanding themselves but also experience it from others.

Acknowledgments

We would like to acknowledge the important role of the Dutch peer-support organization [*FTD lotgenoten*] during this study. In particular, we would like to express our appreciation to Margreet Mantel for her role as ambassador of the project while recruiting participants. We thank Hendrik-Jan van der Waal, chair of the Dutch peer-support organization, for his assistance with interpreting some of the results. In addition, we would like to express appreciation for Yvette Daniels, Esther Gerritzen, and Anne Marieke Doornweerd for their assistance during the focus group discussions.

Disclosure statement

No potential conflict of interest was reported by the authors.

Funding

This study was funded by the Dutch Brain Foundation [*Hersenstichting*], grant number BG-FTD-SBB.

References

Bang, J., Spina, S., and Miller, B. L. (2015). Frontotemporal dementia. *The Lancet, 386*(10004), 1672-1682. doi:10.1016/s0140-6736(15)00461-4

Barca, M. L., Thorsen, K., Engedal, K., Haugen, P. K., and Johannessen, A. (2014). Nobody asked me how I felt: experiences of adult children of persons with young-onset dementia. *Int Psychogeriatr*, *26*(12), 1935-1944. doi:10.1017/S1041610213002639

Boots, L. M. M., Wolfs, C. A. G., Verhey, F. R. J., Kemplen, G. I. J. M., and de Vugt, M. E. (2015). Qualitative study on needs and wishes of early-stage dementia caregivers: the paradox between needing and accepting help. *Int Psychogeriatr, 27*(6), 927-936. doi:10.1017/s1041610214002804

Bradley, E. H., Curry, L. A., and Devers, K. J. (2007). Qualitative data analysis for health services research: developing taxonomy, themes and theory. *Health Serv R, 42*(4), 1758-1772. doi:10.1111/j.1475-6773.2006.00684.x

Caceres, B. A., Frank M. O., Jun, J., Martelly, M. T., Sadarangani, T., and de Sales, P. C. (2016). Family caregivers of patients with frontotemporal dementia: An integrative review. *Int J Nurs Stud, 55*, 71-84. doi:10.1016/j.ijnurstu.2015.10.016

Carter, N., Bryant-Lukosius, D., DiCenso, A., Blythe, J., and Neville, A. J. (2014). The use of triangulation in qualitative research. *Oncol Nurs Forum, 41*(5), 545-547. doi:10.1188/14.ONF.545-547

Charmaz, K. (2014). Constructing grounded theory. London, United Kingdom: Sage.

Creswell, J. W. (2013). Qualitative inquiry and research design: Choosing among five approaches. Los Angeles, California: Sage.

Dam, A. E. H., Boots, L. M. M., Boxtel, M. P. J., Verhey., F. R. J., and de Vugt, M. E. (2018). A mismatch between supply and demand of social support in dementia care: a qualitative study on the perspectives of spousal caregivers and their social network members. *Int Psychogeriatr*, *30*(6), 881-892. doi:10.1017/S1041610217000898

de Vugt, M. E., and Verhey, F. R. J. (2013). The impact of early dementia diagnosis and intervention on informal caregivers. *Prog Neurobiol, 110*, 54-62. doi:10.1016/j.pneurobio.2013.04.00

Diehl-Schmid, J., Schmidt, E. M., Nunnemann, S., Riedl, L., Kurz, A., Förstl, H., Wagenpfeil, S., and Cramer, B. (2013). Caregiver burden and needs in frontotemporal dementia. *J Geriatr psychiatry neurol, 26*(4), 221–229. doi:10.1177/0891988713498467

Draper, B., Cations, M., White, F., Trollor, J., Loy, C., Brodaty, H., Sachdev, P., Gonski, P., Demirkol, A., Cumming, R. G., and Withall, A. (2016). Time to diagnosis in young-onset dementia and its determinants: the INSPIRED study. *Int J Geriatr Psychiatry, 31*(11), 1217–1224. doi:10.1002/gps.4430

Ducharme, F., Lvesque, L., Lachance, L., Kergoat, M.-J., and Coulombe, R. (2011). Challenges associated with transition to caregiver role following diagnostic disclosure of Alzheimer disease: a descriptive study. *Int J Nurs Stud, 48*(9), 1109-1119. doi:10.1016/j.ijnurstu.2011.02.011

Elo, S., and Kyngäs, H. (2008). The qualitative content analysis. *J Adv Nurs, 62*(1), 107-115. doi:10.1111/j.1365-2648.2007.04569.x

Evers, J. (2015). Kwalitatieve analyse: kunst én kunde. Amsterdam, Nederland: Boom Lemma uitgevers.

Glaser, B. G., and Strauss, A. L. (1967). The discovery of grounded theory: Strategies for qualitative research. Chicago, Illinois: Aldine.

Gossink, F., Pijnenburg, Y., Scheltens, P., Pera, A., Kleverwal, R., Korten, N., Stek, M., Dröes, R. M., and Dols, A. (2018a). An intervention programme for caregivers of dementia patients with frontal behavioural changes: an explorative study with controlled effect on sense of competence. *Psychogeriatrics, 18*(6), 451-459. doi:10.1111/psyg.12351

Gossink, F., Schouws, S., Krudop, W., Scheltens, P., Stek, M., Pijnenburg, Y., and Dols, A. (2018b). Social cognition differentiates behavioral variant frontotemporal dementia from other neurodegenerative diseases and psychiatric disorders. *Am J Geriatr Psychiatry, 26*(5), 569-579. doi:10.1016/j.jagp.2017.12.008

Gossink, F. T., Dols, A., Krudop, W. A., Sikkes, S. A., Kerssens, C. J., Prins, N. D., Scheltens, P., Stek, M. L., and Pijnenburg, Y. A. (2016). Formal psychiatric disorders are not overrepresented in behavioral variant frontotemporal dementia. *J Alzheimers Dis, 51*(4), 1249-1256. doi:10.3233/JAD-151198

Harciarek, M., and Cosentino, S. (2013). Language, executive function and social cognition in the diagnosis of frontotemporal dementia syndromes. *Int Rev Psychiatry, 25*(2), 178-196. doi:10.3109/09540261.2013.763340

Hogan, D. B., Jetté, N., Fiest, K. M., Roberts, J. I., Pearson, D., Smith, E. E., Roach, P., Kirk, A., Pringsheim, T., and Maxwell, C. J. (2016). The prevalence and incidence of frontotemporal dementia: a systematic review. *Can J Neurol Sci, 43*, S96-S109. doi:10.1017/cjn.2016.25

Ibañez, A., and Manes, F. (2012). Contextual social cognition and the behavioral variant of frontotemporal dementia. *Neurology, 78*(17), 1354-1362. doi:10.1212/WNL.0b013e3182518375

Kaizik, C., Caga, J., Camino, J., O'Connor, C. M., McKinnon, C., Oyebode, J. R., Piguet, O., Hodges, J. R., and Mioshi, E. (2017). Factors underpinning caregiver burden in frontotemporal dementia differ in spouses and their children. *J Alzheimer's Dis, 56*(3), 1109-1117. doi:10.3233/JAD-160852

Karnatz, T., Monsees, J., Wucherer, D., Michalowsky, B., Zwingmann, I., Halek, M., Hoffmann, W., and Thyrian, J. R. (2019). Burden of caregivers of patients with frontotemporal lobar degeneration - a scoping review. *Int Psychogeriatr*. 1-21. doi:10.1017/S1041610219000176

Knopman, D. S., and Roberts, R. O. (2011). Estimating the number of persons with frontotemporal lobar degeneration in the US population. *Journal of Molecular Neuroscience, 45*(3), 330-335. doi:10.1007/s12031-011-9538-y

Massimo, L., Evans, L. K., and Benner, P. (2013). Caring for loved ones with frontotemporal degeneration: the lived experiences of spouses. *Geriatr Nurs, 34*(4), 302-306. doi:10.1016/j.gerinurse.2013.05.001

Mendez, M. F., Fong, S. S., Shapira, J. S., Jimenez, E. E., Kaiser, N. C., Kremen, S. A., and Tsai, P. H. (2014). Observation of social behavior in frontotemporal dementia. *Am j Alzheimer's Dis Other Dem, 29*(3), 215-221. doi:10.1177/1533317513517035

Mioshi, E., Foxe, D., Leslie, F., Savage, S., Hsieh, S., Miller, L., Hodges, J. R., and Piguet, O. (2013). The impact of dementia severity on caregiver burden in frontotemporal dementia and Alzheimer disease. *Alzheimer Dis Assoc Disord, 27*(1), 68-73. doi:10.1097/WAD.0b013e318247a0bc

Nunnemann, S., Kurz, A., Leucht, S., and Diehl-Schmid, J. (2012). Caregivers of patients with frontotemporal lobar degeneration: a review of burden, problems, needs, and interventions. *Int Psychogeriatr, 24*(9), 1368-1386. doi:10.1017/s104161021200035x

Onyike, C. U., and Diehl-Schmid, J. (2013). The epidemiology of frontotemporal dementia. *Int Rev Psychiatry, 25*(2), 130-137. doi:10.3109/09540261.2013.776523

Patton, M. Q. (2002). Qualitative evaluation and research methods. Los Angeles, California: Sage.

Polenick, C. A., Struble, L. M., Stanislawski, B., Turnwald, M., Broderick, B., Gitlin, L. N., and Kales, H. C. (2018). "The filter is kind of broken": Family caregivers' attributions about behavioral and psychological symptoms of dementia. *Am J Geriatr Psychiatery, 26*(5), 548-556. doi:10.1016/j.jagp.2017.12.004

Rabanal, L. I., Chatwin, J., Walker, A., O'Sullivan, M., and Williamson, T. (2018). Understanding the needs and experiences of people with young onset dementia: a qualitative study. *BMJ Open, 8*, e021166. doi:10.1136/bmjopen-2017-021166

Rabinovici, G. D., and Miller, B. L. (2010). Frontotemporal lobar degeneration epidemiology, pathophysiology, diagnosis and management. *CNS Drugs, 24*(5), 375-398. doi:10.2165/11533100-000000000-00000

Rosness, T. A., Haugen, P. K., and Engedal, K. (2008a). Support to family carers of patients with frontotemporal dementia. *Aging Ment Health, 12*(4), 462-466. doi:10.1080/13607860802224334

Rosness, T. A., Haugen, P. K., Passant, U., and Engedal, K. (2008b). Frontotemporal dementia: a clinically complex diagnosis. *Int J Geriatr Psychiatry, 23*(8), 837-842. doi:10.1002/gps.1992

Rossor, M. N., Fox, N. C., Mummery, C. J., Schott, J. M., and Warren, J. D. (2010). The diagnosis of young-onset dementia. *Lancet Neurol. 9*(8), 793-806. doi:10.1016/S1474 4422(10)70159-9

Savin-Baden, M., and Major, C. H. (2013). Qualitative Research: The Essential Guide to Theory and Practice. Abingdon, England: Routledge.

Sikes, P., and Hall, M. (2018). The impact of parental young onset dementia on children and young people's educational careers. *Br Educ Res J, 44*(4), 593-607. doi:10.1002/berj.3448

Spreadbury, J. H., and Kipps, C. M. (2018). Understanding important issues in young-onset dementia care: the perspectives of healthcare professionals. *Neurodegener Dis Manag, 8*(1), 37-47. doi:10.2217.nmt-2017-0029

Tong, A., Sainsbury, P., and Craig, J. (2007). Consolidated criteria for reporting qualitative research (COREQ): a 32-item checklist for interviews and focus groups. *Int J Qual Health Care, 19*(6), 349-357. doi:10.1093/intqhc/mzm042

van Vliet, D., Bakker, C., Koopmans, R. T., Vernooij-Dassen, M. J., Verhey, F. R., and de Vugt, M. E. (2010). Research protocol of the NeedYD-study (Needs in Young onset Dementia): a prospective cohort study on the needs and course of early onset dementia. *BMC Geriatr, 10*, 1-8. doi:10.1186/1471-2318-10-13

Van Vliet, D., De Vugt, M., Bakker, C., Pijnenburg, Y., Vernooij-Dassen, M., Koopmans, R., and Verhey, F. (2013). Time to diagnosis in young-onset dementia as compared with late-onset dementia. *Psychol Med, 43*(2), 423-432. doi:10.1017/S0033291712001122

van Vliet, D., de Vugt, M. E., Bakker, C., Koopmans, R. T., Pijnenburg, Y. A., Vernooij-Dassen, M. J., and Verhey, F. R. (2011). Caregivers' perspectives on the pre-diagnostic period in early onset dementia: a long and winding road. *Int Psychogeriatr, 23*(9), 1393-1404. doi:10.1017/S1041610211001013

Vasileiou, K., Barnett, J., Barreto, M., Vines, J., Atkinson, M., Lawson, S., and Wilson, M. (2017). Experiences of loneliness associated with being an informal caregiver: A qualitative investigation. *Front Psychol, 8*, 1-11. doi:10.3389/fpsyg.2017.00585

Visser, F. S., Stappers, P. J., van der Lugt, R., and Sanders, E. B. N. (2005). Contextmapping: experiences from practice. *CoDesign, 1*(2), 119-149. doi:10.1080/15710880500135987

Zhu, X. C., Tan, L., Wang, H. F., Jiang, T., Cao, L., Wang, C., Wang, J., Tan, C. C., Meng, X. F., and Yu, J. T. (2015). Rate of early onset Alzheimer's disease: a systematic review and meta-analysis. *Ann Transl Med, 3*(3), 1-6. doi:10.3978/j.issn.2305-5839.2015.01.19

Chapter 4

Behind closed doors. A case study exploring the lived experiences of a family of a person with the behavioral variant of frontotemporal dementia

Bruinsma, J., Peetoom, K., Bakker, C., Verhey, F., and de Vugt, M.

Submitted.

Abstract

The behavioral variant of frontotemporal dementia (bvFTD) is characterized by profound changes in personality and behavior that often start before the age of 65 years. These symptoms interfere with family life, particularly if (adult) children live at home. In research on young-onset dementia or bvFTD, the family itself is hardly ever a unit of analysis. Insight in the perspectives of different family members from the same household allow for a deeper understanding of the complex interference of symptoms of bvFTD with family dynamics.

Methods: This case study explored the perspectives of one family having a relative with bvFTD living at home. Over the course of four months, different family members were individually interviewed twice. Two authors independently performed a directed content analysis using open coding.

Results: The family consisted of a father, mother, and three adult children. Around three years before the interviews the father was diagnosed with bvFTD.

One main category was identified from the analysis, namely a change in family dynamics over the disease trajectory. Three subcategories characterized the change in family dynamics, namely (a) the change in existing roles, relationships and interactions in the family due to early symptoms, (b) a redefinition of roles and responsibility in the family once the diagnosis was established, and (c) the formation of new roles, relationships and interactions in the family by organizing post-diagnostic support at home.

Conclusion: Symptoms of bvFTD have a complex and profound impact on family dynamics and change existing roles, relationships, and interaction patterns. Psychosocial support may help families by accounting for individual differences in involvement, coping, and bereavement. This may help to create a sense of mutual understanding between family members that could potentially strengthen the relationships and help families to deal with the difficult challenge of organizing care for a relative with bvFTD who lives at home.

Keywords: Frontotemporal dementia; Caregiving; Family; Case study

Introduction

The behavioral variant of frontotemporal dementia (bvFTD) is characterized by changes in personality and behavior, such as apathy, loss of empathy, uninhibited behavior, compulsiveness, lacking social insight, and absence of disease awareness (Rabinovici and Miller, 2010; Ducharme et al., 2020). In the majority of the persons with bvFTD, symptoms have an onset before the age of 65 years, making it a specific subvariant of young-onset dementia (Nunnemann et al., 2012; van de Veen et al., 2021). The interference of the symptoms of bvFTD with family life is profound. Given their young age, persons with bvFTD and their spouses are likely to be employed and may have children living at home (Caceres et al., 2016). Typically, the symptom onset of bvFTD is insidious and symptoms are initially misattributed for work-related stress, psychiatric symptoms, marital issues, or as a result of a midlife crisis (Ducharme et al., 2020; Bruinsma et al., 2020a). This complicates a timely diagnosis and results in high levels of pre-diagnostic uncertainty in family members (van Vliet et al., 2011). Obtaining a diagnosis can help family members to understand the changes in their relative who has bvFTD (Bruinsma et al., 2020a; Tookey et al., 2021). Additionally, for family members the diagnosis often confirms the irreversibility of symptoms (de Vugt and Verhey, 2013). This can lead to feelings of grief in anticipation of losing their relative to symptoms of bvFTD (Tookey et al., 2021).

For family members, disclosure of a bvFTD diagnosis often marks the beginning of identifying themselves as caregivers (de Vugt and Verhey, 2013; Bruinsma et al., 2020a). At a young age, it is often a spouse who becomes the primary caregiver (Tookey et al., 2021). This can cause shifts in the distribution of responsibility and changes in interpersonal relationships (Kaizik et al., 2017; Caceres et al., 2016). To illustrate, persons with bvFTD tend to show only little affection towards others. In turn, spouses may experience a diminished sense of couplehood (Holdsworth and McCabe, 2018; Bruinsma et al., 2020b). For (adult) children, the child-parent relationship may reverse as children gradually become more responsible for their parent who has FTD (Kaizik et al., 2017; Oyebode et al., 2013). Children in puberty, adolecence or (young) adulthood may worry about the burden on their healthy parent and may postpone plans such as moving out or studying because of their involvement in care (Millenaar et al., 2014; Cartwright et al., 2021). Although caring for a parent with bvFTD can be stressful, it can also have a positive effect on children who may feel to mature faster as they adopt more responsibility. Children can also experience stronger bonds with other relatives (Grundberg et al., 2021).

Although specific symptoms of bvFTD can interfere with family life, there is only little understanding of the experiences of children living at home (Caceres et al., 2016; Tookey et al., 2021). In most research, the family itself is hardly ever a unit of analysis, thereby ignoring the dynamic context in which caregiving experiences emerge (Kumamoto et al., 2004; Chapman et al., 2019). Exploring the perspectives of different family members from the same household allows for a deeper understanding of changes in roles, relationships, and interaction patterns (Kindell et al., 2014; Jokogbola et al., 2018). This may help to identify directions to support the family as a whole (Kilty et al., 2019; Roach et al., 2013). Currently, most support does not account for family dynamics and is often designed with a primary caregiver, Alzheimer's dementia, or old age in mind (Kobiske and Bekhet, 2018; Tookey et al., 2021; Mayrhofer et al., 2018). Therefore, this study explores the lived experiences of one family who has a relative with bvFTD who lives at home.

Methods

A case study design was used to examine the perspectives of different members from the same family. This allowed an in-dept understanding of the complex interaction between the symptoms and consequences of bvFTD for family life. Between April and August 2021, two rounds of individual interviews were conducted with individual family members. Follow-up interviews after four months enabled an insight in their experiences over time. The consolidated criteria for reporting qualitative research (COREQ) was used to report the findings (Tong, 2007).

Recruitment

Inclusion criteria were (a) the person with bvFTD was younger than 65 years at symptom onset and was living at home, (b) the spouse and at least one adult child were willing to participate, (c) at least one of the children was living at home, and (d) the diagnosis was established at least six months ago to be able to reflect on the adaptation process following the diagnosis. As a secondary selection criterion, the person with bvFTD was opted to participate to account for the patient perspective (Gove et al., 2018).

Recruitment took place via healthcare organizations affiliated with the Dutch Young-onset Dementia Knowledge Center [*Kenniscentrum Dementie op Jonge Leeftijd*] and the Dutch FTD peer-support organization [*FTD lotgenoten*]. Healthcare professionals and caregivers linked to these organizations were asked if they were involved with a family of a person with bvFTD, and to inquire if the family was willing to participate. One family was recruited, and the first author had a telephone call with potential participating family to inform them about the study. Before inclusion, all participating family members gave written consent for participation.

Data collection

Individual interviews were preferred over group interviews to ensure that family members voiced their own perspectives (Reczek, 2014). The initial semi-structured interviews lasted around 60 minutes and were conducted in separate rooms at the parental house of the family. To guide the initial interviews, a topic list was developed based on data from earlier conducted focus group discussions (Bruinsma et al., 2020a) and interviews with relatives of persons with bvFTD conducted in the Needs in Young onset Dementia (NeedYD) study (van Vliet et al., 2010). The topic list was discussed with caregivers of persons with bvFTD who were not included in this study to substantiate the interview questions. The final topic list asked family members to reflect on different phases of the disease trajectory, namely the pre-diagnostic phase, the diagnostic trajectory, the present, and the future. Questions aimed to openly explore perceptions, emotions, and experiences regarding role division, relationships, interaction patterns, personal goals, expectations, and distribution of responsibility.

Four months after the initial interviews, follow-up interviews were conducted to deepen the findings by exploring the lived experiences of the family over time. This enabled us to clarify ambiguities and allowed for member checking (Carter et al., 2014). The follow-up interviews lasted around 60 minutes and the children not living at the parental house preferred to be interviewed via a video call, using Zoom or Microsoft Teams.

Data analysis

Interviews were audio-recorded and transcribed verbatim. Fieldnotes were made about interactions in-between the interview sessions. After the initial interviews, the first and second authors independently performed a directed content analysis by means of open coding in Atlas.ti (Savin-Baden and Major, 2013; Hsieh and Shannon, 2005). In a directed content analysis theoretical concepts are used to collect data and direct the coding process (Hsieh and Shannon, 2005). Therefore, the topic list and coding process were structured according to phases of the disease trajectory (i.e. the pre-diagnostic phase, the diagnostic trajectory, the present, and the future) (Bruinsma et al., 2020a). The first and second author organized discussion sessions to reach consensus on the used codes (Bradley et al., 2007). Codes and (sub)categories following the initial interviews were visualized in a mind-map to support discussion among the first, second, and last authors to summarize the main findings (Suter, 2012). Then, findings were discussed in several meetings with the entire research team to substantiate the results and formulate directions for the follow-up interviews. In the follow-up interviews, participants reflected on the past four months. Subsequently, the main findings were briefly discussed, and questions were asked to deepen the findings and clear ambiguities. Again, the first and second authors independently coded the transcripts from the follow-up interviews and discussed how this substantiated the findings.

Trustworthiness

By interviewing family members twice, this study allowed for a more comprehensive picture of their experiences. This increased the credibility of findings as it allowed to ask additional questions, clear ambiguities, and shortly discuss results to deepen our findings (Suter, 2012). This allowed for member checking of the main findings and preliminary conclusions (Merriam, 2009; Tracy, 2010). To account for researcher bias, all analyses were performed independently by two researchers and the results were discussed extensively with the entire research team to establish investigator triangulation (Carter et al., 2014). More specifically, in monthly meetings the research team reflected on the process of the data collection, the analysis, findings, and conclusions. Members of the research team had varied backgrounds including nursing, health sciences, (neuro)psychology, and neuropsychiatry. All researchers were experienced in conducting qualitative research with caregivers of persons with young-onset dementia and bvFTD.

Ethical considerations

The study protocol was approved by the Research Ethics Committee of the Faculty of Health, Medicine and Life Sciences of Maastricht University, the Netherlands (FHML-REC/2020/102).

Results

Case description

One family was eligible for participation. The family lived in a rural area of the Netherlands, and consisted of a father, mother, and three children. The father of the family was diagnosed with bvFTD around three years before the initial interviews. At the time of the interviews, the family was supported by a dementia casemanager and the father attended daycare for four days a week. The mother of the family was employed parttime. The children (two sons, and one daughter) were aged in their late-20s and early-30s. The oldest son lived at his parents' house and took over the family business after the diagnosis. His siblings had left the parental house around ten years before the diagnosis. The father having bvFTD was unable to participate in our study given the intensity of symptoms.

Early symptoms

The family expressed that, in retrospect, the exact onset of the first bvFTD symptoms was difficult to pinpoint but likely started around eight years before a diagnosis of bvFTD was established. This was attributed to the insidious start, slow progression, and subtle changes in the character and behavior of their father. Looking back on the pre-diagnostic phase, the family noticed symptoms such as apathy, lack of empathy, hoarding, uninhibited eating, and changes in social behavior, which are characteristic for bvFTD.

Current symptoms

Symptoms of bvFTD had intensified in the years following the diagnosis. At time of the first interviews, the family coped with daily apathetic, stereotypic, and uninhibited behavior of the father. To illustrate, the father watched TV for hours, was fixated on food and eating, repeatedly sang one-liners, hoarded items, and performed rituals by closing doors and turning of lights repeatedly. According to the family key symptoms were the lack of disease insight and loss of emotional responsiveness.

During the follow-up interviews after four months, the family noticed that impairment in cognitive functioning had worsened. For instance, they mentioned disorientation in time, inability to understand basic everyday facts, and lacking ability to process information.

Main category: changing family dynamics over the disease trajectory

The directed content analysis resulted in one main category, namely a change in family dynamics over the disease trajectory. Three subcategories were identified that characterize the change in family dynamics, namely (a) the change in existing roles, relationships, and interactions in the family due to early symptoms, (b) a redefinition of roles and responsibilities in the family once the diagnosis was established, and (c) the formation of new roles, relationships, and interactions in the family by organizing post-diagnostic support at home.

Change in existing roles, relationships, and interactions in the family due to early symptoms

Pre-morbid characteristics of the father influenced coping with early symptoms. Before symptoms of bvFTD had onset, there was a certain level of stability in roles, responsibility, and interaction patterns. The father was the ‘pater familias’. He was the head of the family and ran the family business. The family lived up to his expectations and standards. When early symptoms of bvFTD insidiously started, his directive leadership made it difficult to ignore his strong opinion, and to question his behavior. This led to friction between the father and his daughter who had left the parental house on a young age. Later on, his role became more indifferent, indecisive, and less involved. The family noticed these changes but struggled to take over responsibility, given his authority. This led to uncertainty in the mother and adult children. To illustrate, the oldest son experienced an internal struggle regarding his position and his future within the family business.

“My father always knew what he was doing but gradually I got doubts like ‘Does it add up to what he does?’, ‘why am I still here?’, and ‘Should I leave?’. However, before I was able to leave, things had to be arranged but that was his responsibility. So basically, I was stuck.”
– Oldest son –

The apathetic and indifferent behavior of the father adversely influenced the relationships with his family members. Relationships with him became remote and emotionally distanced due to his lack of social responsiveness. To illustrate, he showed no empathy and was hardly interested in others. In turn, the mother experienced that the relationship with her husband lacked reciprocity. This made her feel neglected and she thought about filing for divorce.

“My husband never treated me badly, but he was authoritarian and laid down the law. Slowly this changed and he started to agree more. This was very unsatisfying because I had the impression he simply was not interested in me anymore.”
– Mother –

The apathetic behavior of the father changed the interaction patterns within the family as communication with him became superficial. Also, he denied and trivialized the apathetic behavior. In turn, the other family members increasingly started to discuss their concerns with each other. This was accelerated when they noticed changes in the social behavior of the father. More specifically, he became more selfish and started to make road trips without being able to explain his destination.

Based on the conversations about the father’s behavior, the family came to the joint decision to consult the general practitioner (GP). The father denied symptoms and refused to go to the GP, but the family was very concerned about his behavior and his driving abilities. Therefore, the oldest son took a leadership role and took his father’s car keys as leverage to visit the GP.

“I was shaking, wondering how he was going to react after I took his car keys. His reaction was ‘Just give me the keys back. Just give them back’. Non-stop. This continued for days. I stood my grounds to force him to visit the general practitioner.”
– Oldest son –

Upon consultation, the GP noticed social inappropriate behavior of the father in the waiting room leading and referred to a specialized neurologist. The diagnosis of bvFTD followed upon neuropsychological assessment and an MRI-scan.

A redefinition of roles and responsibilities in the family once the diagnosis was established

The diagnosis facilitated a better understanding of symptoms and clarified the apathetic behavior of the father having bvFTD. This allowed for a new perspective on relationships, roles, and responsibility.

The mother realized the deterioration in the quality of their relationship was not her husband's fault. The diagnosis made her aware her role had changed into being the primary caregiver. This insight strengthened her in taking the lead in the decision-making process about care options. This was an important insight because her husband had always been the one making the important decisions in the family.

"The diagnosis is devastating because there is no cure and symptoms progress. However, the diagnosis is also a relief. (…) I thought 'now it's me who is going to make the decisions.' More as caregiver than as his wife."

– Mother –

The diagnosis also helped the daughter to reevaluate the relationship with her father. She reflected on the love-hate relationship they previously had. This led to the insight she had distanced herself from her relatives in last years, and now had a more passive role within the decision-making process.

"The positive effect of the diagnosis was that it helped me realize our complicated relationship was not only my fault."

– Daughter –

After the bvFTD diagnosis the family reevaluated their expectations towards the father. This allowed the children to define new roles and take responsibility. For example, the sons felt that they had to take on a more supportive role to help their mother in organizing care. The oldest son took the lead in the family business and felt strengthened to make decisions without consulting his father.

"The diagnosis made me realize it was not my father who could make the decisions anymore. We were in charge. He kept saying 'No, no, no. You don't have to do this'. However, to get things done you have to ignore him. In this sense, taking his car keys was already a good practice."

– Oldest son –

Formation of new roles, relationships, and interactions in the family by organizing support

In the years after the diagnosis, the dynamics in the family further changed due to the fact that all care and support was organized at home. Although in the decision-making process regarding care the mother was primarily responsible, the sons were closely involved. This led to role reversal as the mother and children now shared authority over daily decisions, instead of the father. This had improved their communication and strengthened the relationship among the family members directly involved in organizing care. On the other hand, the relationship to the father had drastically changed by progressing symptoms of bvFTD.

"My father basically is part of the furniture. It is difficult to stay emotionally attached to him. We feed him, supervise him, and he pulls of a happy façade."

– Oldest son –

Organizing support together had created a sense of cohesion between the family members directly involved. The youngest son also helped his older brother with running the family business. This had strengthened the sense of companionship between the brothers.

"In the phase following the diagnosis everything changed for the positive. We started to talk about what we were going to do with the family business. If you talk about it, you go for it together. This gave us energy."

– Middle son –

The daughter felt more distant from organizing care. She studied and experienced it as confronting to see her father deteriorate. According to the daughter, she applied an emotional coping style and needed more space to grieve than her brothers. Therefore, she deliberately distanced herself from the care situation at home. Her mother had noticed this and tried to protect her daughter by not burdening her with discussing care related matters.

"I think everyone has different needs in coping with this. (…) I do not necessarily need to cope with this the same way my brothers do. I need more space to grieve."

– Daughter –

On the contrary, the mother and sons were able to emotionally detach from the situation. Their high level of involvement was grounded in using similar coping strategies. They were very creative in adapting their daily lives and formed an alliance in organizing care at home for as long as possible, even though they were uncertain about the future. This created cohesion between them.

> *"We already placed a lock on the refrigerator. If necessary, we will store the knifes behind a lock too. We would even get rid of the stove if he would turn on the gas. We are very adaptive. He [my father] taught us to be resourceful himself."*
>
> – Oldest son –

Discussion

Key results

This case study allowed a unique insight in the changing family dynamics that accompany having a relative with bvFTD. The findings reveal that early symptoms of bvFTD progressed and slowly altered the role and position of the father within his family. His apathetic behavior and low levels of emotional responsiveness adversely affected the relationships with his family. Obtaining the diagnosis allowed the family to understand the apathetic behavior and the emotionally distanced relationship with their father having bvFTD. This led the family to reevaluate their expectations, allowed to redistribute responsibility, and redefine new roles. The mother and sons formed a strong alliance and were practical in organizing daily care. This created cohesion, improved their communication, and strengthened their relationships. In contrast, the daughter was more distanced from organizing care. She had more need to cope emotionally and needed space for bereavement.

Interpretation of findings

Similar to prior research, in our study insufficient role fulfilment of the person having bvFTD led to pre-diagnostic role ambiguity in family members (Kilty et al., 2019). We found that early symptoms affected the role, level of involvement, and the position of the person having bvFTD within the family system. More specifically, family members experienced ambiguity about the cause of the apathetic behavior of their relative with undiagnosed bvFTD. Subsequently, this led to ambiguity about their own role, responsibility, and future. For example, the oldest son experienced the family business deteriorated due to his father's apathic behavior but struggled to take responsibility because it was difficult to transcend his father's authority. This pre-diagnostic role ambiguity is probably directly related to the characteristic symptoms of bvFTD, such as apathy and impaired social cognition (Ducharme et al., 2020; Russell et al., 2020). Similar to our findings, these symptoms affect relationships of persons with bvFTD as interactions become more remote and emotionally distanced (Tookey et al., 2021; Holdsworth and McCabe, 2018). Without the diagnosis these symptoms of apathy and emotional bluntness are easily mistaken for selfishness (Massimo et al., 2013). In turn, family members may blame their relative with bvFTD for early symptoms, which can adversely affect the experienced level of closeness (Holdsworth and McCabe, 2018; Paton et al., 2004).

In our study the family had postponed visiting the GP, this may be explained by that fact that the early symptoms of bvFTD are particularly difficult to label as signs of a pathology (Rasmussen et al., 2019; Bruinsma et al., 2020a). Postponing help-seeking behavior in the pre-diagnostic phase can contribute to the diagnostic delay of bvFTD (van Vliet et al., 2011; van Vliet et al., 2013). Typically, early symptoms of bvFTD are also difficult to recognize for GPs. This is attributed to the young age and the symptomatic overlap with psychiatric disorders (Beber and Chaves, 2013; Giamarelou et al., 2020). Our findings show the diagnosis was accelerated because the GP referred to a specialized neurologist after noticing impairments in social cognition. Confirming previous findings, obtaining the diagnosis was important as it facilitated understanding and acceptation of symptoms (de Vugt and Verhey, 2013; Bruinsma et al., 2020a). Prior work has documented obtaining the diagnosis may introduce new uncertainty, for example regarding the future (Grundberg et al., 2021; Tookey et al., 2021). In contrast, we found the diagnosis positively helped the family to plan for the future. More specifically, the diagnosis helped the family with reevaluating expectations, redistribute responsibility, and redefine new role patterns. This empowered the family to transcend the strong opinion and authority of their relative having bvFTD. This helped them to take responsibility in the decision-making process. Until now, only limited research has reported these positive aspects that accompany diagnostic disclosure (Tookey et al., 2021).

Although (adult) children may not perceive themselves as caregivers, they are often an essential part of the support structure for persons with young-onset dementia or bvFTD living at home (Grundberg et al., 2021; Cartwright et al., 2021). We found that organizing care together increased cohesion, strengthened interaction and communication, and created a sense of companionship between family members directly involved. The adult children in our study experience a reversed child-parent role and similar as documented in other studies they adopted increasingly more responsibility (Kaizik et al., 2017; Hall and Sikes, 2018). Although this can be stressful and have a negative impact on the future development of children (Johannessen et al., 2015; Millenaar et al., 2014), adopting more responsibility can also have positive effects as children may grow into a leadership role and perceive to mature (Cartwright et al., 2021). Coping strategies can influence how children cope with the changing role patterns, intensifying symptoms, and feelings of grief (Grundberg et al., 2021; Millenaar et al., 2014). Some children may become increasingly involved whereas others may distance themselves from the caregiving situation as a mechanism of self-protection. Both are normal mechanisms of coping with a parent having bvFTD (Cartwright et al., 2021). Our findings showed the level of engagement in organizing caregiving was related to the position of the children in the pre-diagnostic phase, the use of coping strategies, and having the ability to emotionally detach from the person having bvFTD. To illustrate, in the pre-diagnostic phase the daughter was more distanced from the situation at home, compared to the oldest son who lived at home and worked in his father's business. After the diagnosis, he was already closely involved and therefore able to take over responsibility and form an alliance with his mother and brother in organizing care. In contrast, after the diagnosis the daughter needed more space to emotionally cope with her feelings of bereavement and deliberately distanced herself from the care situation. In turn, she was less involved in the decision-making process. A direction for the future is to support families by accounting for these individual differences in involvement, coping, and bereavement caused by the dynamic interplay between symptoms and family life. Creating a sense of mutual understanding between family members could potentially strengthen the relationships and help families to deal with the difficult challenge of organizing care for a relative with bvFTD who lives at home.

Strengths and limitations

The case study design allowed a unique insight in the complex interplay between the symptoms and consequences of bvFTD and changes in family dynamics. The case study design helped to get a detailed understanding of the individual perspectives of different family members involved. For studying such complex phenomena a case study can be particularly useful (Flyvbjerg, 2006). Our study aimed to bridge a knowledge gap by using the family as a unit of analysis and adhere to a holistic and family-centered approach (Roach et al., 2013). It was not our aim to provide generalized claims about the impact of bvFTD on family life and we recognize the complexity and uniqueness of each family, person, and care situation.

Interviewing the family twice over the course of four months allowed an iterative process between data collection and analysis. This enabled member checking the findings and allowed for a unique insight in the experiences of caregivers over time. A study with an even longer interval would allow a more detailed understanding of the experience of caregivers, for example by exploring the transition towards institutionalization. A limitation of this study is it did not include the perspective of the person with bvFTD. Adhering to recommendations of Alzheimer Europe the person with bvFTD was opted to participate in our study (Gove et al., 2018). However, in our case the participation was complicated by the intensity of symptoms of bvFTD. Therefore, including a family in the phase directly after the diagnosis may allow for including persons with bvFTD. This remains a direction for future research.

Conclusion

Our findings confirm that symptoms of bvFTD have a profound impact on family life. Particularly, behavioural symptoms and deficits in social cognition changed existing roles, relationships, and interaction dynamics. This started already in the years before the diagnosis. Complicating is that the diagnosis of bvFTD is often delayed, resulting in a long pre-diagnostic phase. In this phase, relationships diminish as the role and position of the person with bvFTD changes. Disclosure of the diagnosis allowed for a sudden redefinition of roles and redistribution of responsibility within the family. Post-diagnostic support should focus on supporting caregivers to cope with these abrupt shifts. A system approach could focus on supporting all family members, for example by accounting for the individual differences in positions, involvement, coping, and need for bereavement. This may elicit a sense of mutual understanding between family members, and could potentially strengthen the relationships and improve organizing care together. Our findings showed that organizing care together had a range of positive effects for family members such as creating cohesion, improving communication, and strengthening relationships.

Acknowledgements

This study is the result of the family's willingness to share their feelings and experiences. Therefore, we acknowledge their high level of engagement in this study and thank them for their openness. Additionally, we appreciate the support during the recruitment provided by the Dutch FTD peer-support organization [*FTD lotgenoten*] and healthcare organizations affiliated with the Dutch Young-onset Dementia Knowledge Center [*Kenniscentrum Dementie op Jonge Leeftijd*]. We also want to acknowledge Roos Roberts for her help with transcribing the interview data.

Conflict of interest

None.

Funding

This study was funded by the Dutch brain foundation [*Hersenstichting*], grant number BG-FTD-SBB.

References

Beber, B. C., and Chaves, M. L. F. (2013). Evaluation of patients with behavioral and cognitive complaints: misdiagnosis in frontotemporal dementia and Alzheimer's disease. Dement Neuropsychol, 7(1), 60-65. doi:10.1590/S1980-57642013DN70100010

Bradley, E. H., Curry, L. A., and Devers, K. J. (2007). Qualitative data analysis for health services research: developing taxonomy, themes, and theory. Health Serv Res, 42(4), 1758-1772. doi:10.1111/j.1475-6773.2006.00684.x

Bruinsma, J., Peetoom, K., Bakker, C., Boots, L., Verhey, F., and de Vugt, M. (2020). “They simply do not understand”: a focus group study exploring the lived experiences of family caregivers of people with frontotemporal dementia. Aging Ment Health. doi:10.1080/13607863.2020.1857697

Bruinsma, J., Peetoom, K., Millenaar, J., Kohler, S., Bakker, C., Koopmans, R., . . . de Vugt, M. (2020). The quality of the relationship perceived by spouses of people with young-onset dementia. Int Psychogeriatr, 1-10. doi:10.1017/S1041610220000332

Caceres, B. A., Frank, M. O., Jun, J., Martelly, M. T., Sadarangani, T., and de Sales, P. C. (2016). Family caregivers of patients with frontotemporal dementia: an integrative review. Int J Nurs Stud, 55, 71-84. doi:10.1016/j.ijnurstu.2015.10.016

Carter, N., Bryant-Lukosius, D., DiCenso, A., Blythe, J., and Neville, A. J. (2014). The use of triangulation in qualitative research. Oncol Nurs Forum, 41(5), 545-547. doi:10.1188/14.ONF.545-547

Cartwright, A. V., Stoner, C. R., Pione, R. D., and Spector, A. (2021). The experiences of those affected by parental young onset dementia: A qualitative systematic literature review. Dementia, online ahead of print. doi:10.1177/1471301220988231

Chapman, M., Philip, J., and Komesaroff, P. (2019). Towards an Ecology of Dementia: A Manifesto. J Bioeth Inq, 16(2), 209-216. doi:10.1007/s11673-019-09910-8

de Vugt, M. E., and Verhey, F. R. J. (2013). The impact of early dementia diagnosis and intervention on informal caregivers. Prog Neurobiol, 110, 54-62. doi:10.1016/j.pneurobio.2013.04.00

Ducharme, S., Dols, A., Laforce, R., Devenney, E., Kumfor, F., van den Stock, J., . . . Pijnenburg, Y. (2020). Recommendations to distinguish behavioural variant frontotemporal dementia from psychiatric disorders. Brain 143(6), 1632-1650. doi:10.1093/brain/awaa018

Flyvbjerg, B. (2006). Five Misunderstandings About Case-Study Research. Qual Inq, 12(2), 219-245. doi:10.1177/1077800405284363

Giamarelou, A., Polychronopoulos, P., Skokou, M., Messinis, L., and Gourzis, P. (2020). Frontotemporal dementia misdiagnosed as schizophrenia or other psychotic disorder. Eur Psychiatry, 41(S1), s812-s812. doi:10.1016/j.eurpsy.2017.01.1575

Gove, D., Diaz-Ponce, A., Georges, J., Moniz-Cook, E., Mountain, G., Chattat, R., . . . European Working Group of People with Dementia. (2018). Alzheimer Europe's position on involving people with dementia in research through PPI (patient and public involvement). Aging Ment Health, 22(6), 723-729. doi:10.1080/13607863.2017.1317334

Grundberg, A., Sandberg, J., and Craftman, A. G. (2021). Childrens' and young adults' perspectives of having a parent with dementia diagnosis: A scoping review. Dementia, online ahead of print. doi:10.1177/14713012211023653

Hall, M., and Sikes, P. (2018). How do young people 'do' family where there is a diagnosis of dementia? Fam Relationsh Soc, 7(2), 207-225. doi:10.1332/204674316x14818999694306

Holdsworth, K., and McCabe, M. (2018). The impact of younger-onset dementia on relationships, intimacy, and sexuality in midlife couples: a systematic review. Int Psychogeriatr, 30(1), 15-29. doi:10.1017/S1041610217001806

Hsieh, H. F., and Shannon, S. E. (2005). Three approaches to qualitative content analysis. Qual Health Res, 15(9), 1277-1288. doi:10.1177/1049732305276687

Johannessen, A., Engedal, K., and Thorsen, K. (2015). Adult children of parents with young-onset dementia narrate the experiences of their youth through metaphors. J Multidiscip Healthc, 8, 245-254. doi:10.2147/JMDH.S84069

Jokogbola, O. R., Solomon, C., and Wilson, S. L. (2018). Family as Caregiver: Understanding Dementia and Family Relationship. Advances in Clin Trans Res, 2(2), 1-5. doi:10.31887/DCNS.2009.11.2/hbrodaty

Kaizik, C., Caga, J., Camino, J., O'Connor, C. M., McKinnon, C., Oyebode, J. R., Piguet, O., Hodges, J. R., and Mioshi, E. (2017). Factors underpinning caregiver burden in frontotemporal dementia differ in spouses and their children. J Alzheimer’s Dis, 56(3), 1109-1117. doi:10.3233/JAD-160852

Kilty, C., Boland, P., Goodwin, J., and de Roiste, A. (2019). Caring for people with young onset dementia: an interpretative phenomenological analysis of family caregivers’ experiences. J Psychosoc Nurs Men Health Serv, 57(11), 37–44. doi:10.3928/02793695-20190821-02

Kindell, J., Sage, K., Wilkinson, R., and Keady, J. (2014). Living with semantic dementia: a case study of one family's experience. Qual Health Res, 24(3), 401-411. doi:10.1177/1049732314521900

Kobiske, K. R., and Bekhet, A. K. (2018). Resilience in Caregivers of Partners With Young Onset Dementia: A Concept Analysis. Issues Ment Health Nurs, 39(5), 411-419. doi:10.1080/01612840.2017.1400625

Kumamoto, K., Arai, Y., Hashimoto, N., Ikeda, M., Mizuno, Y., and Washio, M. (2004). Problems family caregivers encounter in home care of patients with frontotemporal lobar degeneration. Int Psychogeriatr, 4, 33-39. doi:10.1111/ j.1479-8301.2004.00064.x

Massimo, L., Evans, L. K. and Benner, P. (2013). Caring for loved ones with frontotemporal degeneration: the lived experiences of spouses. Geriatr Nurs, 34(4), 302-306. doi:10.1016/j.gerinurse.2013.05.001

Mayrhofer, A., Mathie, E., McKeown, J., Bunn, F., and Goodman, C. (2018). Age-appropriate services for people diagnosed with young onset dementia (YOD): a systematic review. Aging Ment Health, 22(8), 927-935. doi:10.1080/13607863.2017.1334038

Merriam, S. B. (2009). Qualitative research: A guide to design and implementation. San Francisco: Jossey-Bass.

Millenaar, J. K., van Vliet, D., Bakker, C., Vernooij-Dassen, M. J., Koopmans, R. T. C. M., Verhey, F. R., and de Vugt, M. E. (2014). The experiences and needs of children living with a parent with young onset dementia: results from the NeedYD study. Int Psychogeriatr, 26(12), 2001-2010. doi:10.1017/S1041610213001890

Nunnemann, S., Kurz, A., Leucht, S., and Diehl-Schmid, J. (2012). Caregivers of patients with frontotemporal lobar degeneration: a review of burden, problems, needs, and interventions. Int Psychogeriatr, 24(9), 1368-1386. doi:10.1017/S104161021200035X

Oyebode, J. R., Bradley, P., and Allen, J. L. (2013). Relatives' experiences of frontal-variant frontotemporal dementia. Qual Health Res, 23(2), 156-166. doi:10.1177/1049732312466294

Paton, J., Johnston, K., Katona, C., and Livingston, G. (2004). What causes problems in Alzheimer’s disease: attributions by caregivers. A qualitative study. Int J geriatr psychiatry, 19(6), 527-532. doi:10.1002/gps.1118

Rabinovici, G. D., and Miller, B. L. (2010). Frontotemporal lobar degeneration epidemiology, pathophysiology, diagnosis and management. CNS Drugs, 24(5), 375-398. doi:10.2165/11533100-000000000-00000

Rasmussen, H., Hellzen, O., Stordal, E., and Enmarker, I. (2019). Family caregivers experiences of the pre-diagnostic stage in frontotemporal dementia. Geriatr Nurs, 40(3), 246-251. doi:10.1016/j.gerinurse.2018.10.006

Reczek, C. (2014). Conducting a multi family member interview study. Fam Process, 53(2), 318-335. doi10.1111/famp.12060

Roach, P., Keady, J., Bee, P., and Williams, S. (2013). ‘We can’t keep going on like this’: identifying family storylines in young onset dementia. Ageing Soc, 34(8), 1397–1426. doi:10.1017/s0144686x13000202

Russell, L. L., Greaves, C. V., Bocchetta, M., Nicholas, J., Convery, R. S., Moore, K., . . . Genetic Ftd Initiative, GENFI. (2020). Social cognition impairment in genetic frontotemporal dementia within the GENFI cohort. Cortex, 133, 384-398. doi:10.1016/j.cortex.2020.08.023

Savin-Baden, M., and Major, C. H. (2013). Qualitative Research: The Essential Guide to Theory and Practice. Abingdon, England: Routledge.

Suter, W. N. (2012). Qualitative data, analysis, and design. In Introduction to educational research: A critical thinking approach (pp. 342-386). (2nd ed.). Thousand Oaks, CA: SAGE Publications, Inc.

Tong, A., Sainsbury, P., and Craig, J. (2007). Consolidated criteria for reporting qualitative research (COREQ): a 32-item checklist for interviews and focus groups. Int J Qual Health Care, 19(6), 349-357. doi:10.1093/intqhc/mzm042

Tookey, S. A., Greaves, C. V., Rohrer, J. D., and Stott, J. (2021). Specific support needs and experiences of carers of people with frontotemporal dementia: A systematic review. Dementia, online ahead of print. doi:10.1177/14713012211022982

Tracy, S. J. (2010). Qualitative Quality: Eight “Big-Tent” Criteria for Excellent Qualitative Research. Qual Inq, 16(10), 837-851. doi:10.1177/1077800410383121

van de Veen, D., Bakker, C., Peetoom, K., Pijnenburg, Y., Papma, J. M., PRECODE study group., de Vugt, M., and Koopmans, R. (2021). An Integrative Literature Review on the Nomenclature and Definition of Dementia at a Young Age. J Alzheimers Dis. doi:10.3233/JAD-210458

van Vliet, D., Bakker, C., Koopmans, R. T., Vernooij-Dassen, M. J., Verhey, F. R., and de Vugt, M. E. (2010). Research protocol of the NeedYD-study (Needs in Young onset Dementia): a prospective cohort study on the needs and course of early onset dementia. BMC Geriatr, 10, 1-8. doi:10.1186/1471-2318-10-13

van Vliet, D., de Vugt, M. E., Bakker, C., Koopmans, R. T., Pijnenburg, Y. A., Vernooij-Dassen, M. J., and Verhey, F. R. (2011). Caregivers' perspectives on the pre-diagnostic period in early onset dementia: a long and winding road. Int Psychogeriatr, 23(9), 1393-1404. doi:10.1017/S1041610211001013

van Vliet, D., de Vugt, M. E., Bakker, C., Pijnenburg, Y. A., Vernooij-Dassen, M. J., Koopmans, R. T., and Verhey, F. R. (2013). Time to diagnosis in young-onset dementia as compared with late-onset dementia. Psychol Med, 43(2), 423-432. doi:10.1017/S0033291712001122

Chapter 5

Evaluating the Dutch RHAPSODY program: a web-based information and support program for caregivers of persons with young-onset dementia

Daemen, M., Bruinsma, J., Bakker, C., Groot Zwaaftink, R., Koopmans, R., Oostijen, A., Loose, B., Verhey, F., de Vugt, M., and Peetoom, K.

Submitted.

Abstract

Caregivers of persons with young-onset dementia (YOD) have an explicit need for tailored information and support about YOD. Therefore, during the European RHAPSODY project a web-based information and support program for YOD caregivers was developed. The program was recently tailored to the Dutch context. This study evaluates the Dutch version on user acceptability, usability, user satisfaction, and user behavior.

Methods: A cross-sectional study was conducted to evaluate the publicly available Dutch RHAPSODY program. Surveys and semi-structured interviews were used to evaluate how visitors perceived the program in terms of acceptability, usability, and their satisfaction. Web metrics registered user behavior. Quantitative data were analyzed using descriptive statistics and a deductive content analysis was used to analyze qualitative data.

Results: Most participants were caregivers and healthcare professionals. They perceived the program as acceptable, usable, and were satisfied about the quality of the content. The majority recommended the program to others. Participants emphasized the necessity and desirability of a central platform incorporating educational and practical information about YOD. The page providing a background explanation about YOD was the most viewed. The most time was spent on pages about the diagnostic process and difficulties in executing daily activities.

Conclusion: The Dutch RHAPSODY program showed good user acceptability, usability, and user satisfaction. The program adds value to existing available support for YOD caregivers and met the need for tailored information and support regarding YOD. Raising awareness about the programs existence among healthcare professionals may help caregivers to find appropriate information after obtaining the diagnosis. The program also provides educational opportunities for healthcare professionals.

Keywords: Young-onset dementia; Caregiver; Web-based; E-learning; Education

Introduction

The first symptoms of young-onset dementia (YOD) have an onset before the age of 65 years (Koopmans and Rosness, 2014; van de Veen et al., 2021). Unlike dementia symptoms in late-onset dementia (LOD), the presentation of symptoms in YOD is more varied due to the larger variety in underlying pathology and differences in clinical manifestation (Rossor et al., 2010). For example, behavioral symptoms occur more often in YOD, including changes in social abilities, apathy, impulsivity, or uninhibited behavior (Ducharme and Dickerson, 2015; Kuruppu and Matthews, 2013). Partly, this is due to the higher prevalence of frontotemporal dementia in YOD, compared to LOD (Onyike and Diehl-Schmid, 2013). Additionally, Alzheimer's dementia in individuals younger than 65 years is more likely to be characterized by non-memory symptoms, such as language deficits and executive dysfunctions (Barnes et al., 2015; Koedam et al., 2010).

Coping with these symptoms poses unique challenges for caregivers of persons with YOD and can result in high levels of burden, distress, and depressive symptoms (Ducharme et al., 2013; Lim et al., 2018; Millenaar et al., 2014). Spousal caregivers are often in their 50s, they may be employed and can have children living at home (Cartwright et al., 2021; Grundberg et al., 2021). This can result in difficulty in balancing the caregiving role with professional responsibilities (Ducharme et al., 2013) or financial problems due to reduced working hours or early retirement (Mayrhofer et al., 2021; van Vliet et al., 2010). Additionally, caring for a family member with YOD is known to result in profound shifts in family roles and relationships (Cabote et al., 2015; Svanberg et al., 2011).

Caregivers experience significant difficulty in coping with these circumstances and express an explicit need for age-appropriate professional support (Arai et al., 2007; Lockeridge and Simpson, 2013; Millenaar et al., 2014; Millenaar et al., 2016). Especially following the diagnosis, caregivers have an explicit need for specific information about what YOD entails, the prognosis, advice on informing others, questions regarding heredity, and available support and care. They also prefer to have practical information, for example on coping with symptoms and obtaining support for financial problems (Ducharme et al., 2014; Mayrhofer et al., 2018; Millenaar et al., 2016; Rosness et al., 2012). Currently, YOD caregivers often experience difficulty finding age-appropriate information as most information is too generic as it is focused on caregivers of persons with dementia in old age (Cations et al., 2017; Mayrhofer et al., 2018; Millenaar et al., 2016). Psychoeducation is an opportunity to adequately inform caregivers and may help them to acquire coping skills (Cations et al., 2017; Spreadbury and Kipps, 2018). Potentially, psychoeducation could also be used to inform healthcare professionals since they, in general, have limited knowledge about YOD (Bruinsma et al., 2020; Spreadbury and Kipps, 2018).

Previous research demonstrated that web-based services are flexible, easily accessible, and cost-effective, especially in areas hindered by geographical barriers (Godwin et al., 2013; Klimova et al., 2019; Waller et al., 2017). Previously, the European RHAPSODY consortium (http://www.rhapsody-project.eu) developed a web-based information and support program for YOD caregivers (Kurz et al., 2016). A pilot study in Germany, France, and the United Kingdom showed positive results regarding user acceptability, program satisfaction, and caregiver well-being (Metcalfe et al., 2019). Building upon these promising results, a Dutch version of the RHAPSODY program was developed and tailored to the Dutch context. The current study evaluates the Dutch RHAPSODY program in terms of user acceptability, usability, user satisfaction, and user behavior.

Methods

This cross-sectional study used quantitative and qualitative measures to evaluate user acceptability, usability, user satisfaction, and user behavior of the Dutch version of the RHAPSODY program. Between October and December 2020, online surveys and semi-structured telephone interviews were conducted. Additionally, web metrics were registered. The online program was freely and publicly available via the website of the Dutch Alzheimer's Society [*Alzheimer Nederland*], www.dementie.nl [*Online training dementie op jonge leetijd*]. The guidelines of the CONSORT-EHEALTH were used for reporting the results of this study (Eysenbach et al., 2011).

Development of the Dutch RHAPSODY program

The European RHAPSODY project is a European consortium, consisting of partners from six countries. The Dutch RHAPSODY program was built upon the previously developed versions designed and evaluated for caregivers of persons with YOD in Germany, France, and the United Kingdom (Kurz et al., 2016; Metcalfe et al., 2019). As in the previous versions, the Dutch program provides separate chapters describing different topics, such as medical information, management of challenging behavior, dealing with changes in role patterns within the family, access and availability of appropriate support, and (re)gaining a sense of balance during the caregiving trajectory. The content of the previous RHAPSODY versions was translated, revised, and tailored to the Dutch context by the research team. Caregivers, healthcare professionals, and field experts provided feedback and input during the development process. The language use in RHAPSODY aimed to address the general public and lay audience. The Dutch program (see Appendix A) contains textual information, explanatory pictures, animations, reflective questions, and video vignettes featuring three family caregivers and two YOD experts (CB and MdV). One chapter was completely rewritten to provide information on dedicated post-diagnostic care and support for YOD available in the Netherlands.

Recruitment

Both YOD caregivers and healthcare professionals were invited to use the online program and to participate in our study to evaluate user acceptability, usability, user satisfaction, and user behavior. Information about the program was spread via newsletters, social media, presentations, flyers, and by using personal mailing lists of Alzheimer Center Limburg, Radboudumc Alzheimer Center, the Dutch Alzheimer's Society, the Dutch YOD Knowledge Center [*Kenniscentrum Dementie op Jonge Leeftijd*], and the Academic Network of Nursing Homes Nijmegen [*UKON*]. All visitors who accessed the program were automatically invited to complete a short pop-up survey that appeared if participants visited a page longer than 30 seconds (Figure 1, see route 1). Subsequently, if they were willing to participate in an additional and more extensive survey, their email-address was requested to send them the link to the survey. Participants were also able to contact the research team directly to request participation (Figure 1, see route 2). All participants who completed the extensive survey were invited for an in-depth interview that was conducted via telephone.

Measurements

Participant characteristics

Participants were asked about their background in the pop-up survey. In the extensive survey, more detailed questions were asked about either their relationship to the person with YOD or about their healthcare profession. Participants were also asked about their gender and age.

Exploring user acceptability, usability, and user satisfaction

Quantitative and qualitative measures were used to examine user acceptability, usability, and user satisfaction. Acceptability was defined as the perceived ease of use and usefulness of the Dutch RHAPSODY program (Rahimi et al., 2018). Similar to the study evaluating previous versions of the RHAPSODY program, four subscales of the Technology Acceptance Model (TAM) were used (Metcalfe et al., 2019). The TAM predicts individual adoption of internet driven technology and can explain around 40% of the variance in the actual use and intention to use (Venkatesh and

Bala, 2008). The TAM examines technology in terms of perceived usefulness, ease of use, behavioral intention to use, and computer self-efficacy. The current study assessed these four domains in the extensive survey using a 7-point Likert scale, ranging from 1 ‘totally disagree’ to 7 ‘totally agree’.

Usability was defined as the extent to which the Dutch RHAPSODY program could be used in daily life and met the needs of the target group (Dix, 2003). To illustrate, usability was examined in the extensive survey by asking if participants found it difficult to allocate time to embed the program in daily life. Additionally, questions focused on the quantity of information provided. Usability items were rated on a 5-point Likert scale, ranging from 1 ‘totally disagree to 5 ‘totally agree’.

User satisfaction referred to the quality and relevancy of the program content and likelihood of recommending the Dutch RHAPSODY program to others. Like the study performed by Metcalfe et al., (2019), user satisfaction was assessed by asking about the perceived quality, relevancy, understandability, applicability of content in daily life, and layout of the program. Items in the extensive survey were rated on a 5-point Likert scale, ranging from 1 ‘very bad’ to 5 ‘very good’. One item in the pop-up survey asked about recommending the program to others and used a scale from 1 ‘very unlikely to 10 ‘very likely’.

The in-depth and semi-structured interviews were audio-recorded and conducted via telephone by two researchers (MD and KP). Interviews lasted around 40 minutes and were structured using an interview guide asking in-depth questions about user acceptability, usability, and user satisfaction (see Appendix B). For example, questions asked participants how they perceived the quality of the content, language use, usefulness of topics, the likelihood that they would recommend the program to others, and how they experienced navigating through the program content.

User behavior

Web metrics were collected anonymously to explore the behavior of visitors in terms of the number and duration of page visits. Due to the use of website cookies, a visitor would count as a unique page visitor once every 24 hours. Web metrics were also gathered three months after the evaluation study to monitor the implementation of the Dutch RHAPSODY program.

Data analysis

Quantitative data was analyzed using SPSS Statistics version 27. Descriptive statistics were calculated (mean, mode, and standard deviation) to explore participant characteristics, user acceptability, usability, user satisfaction, and to analyze the web metrics. The qualitative data was analyzed using a deductive content analysis, using transcripts of the interviews (Elo and Kyngäs, 2008). Qualitative data was examined from a pragmatic theoretical perspective, examining whether the program met the needs of the target group (Kaushik and Walsh, 2019). Therefore, transcripts were deductively coded by the first author (MD) using Atlas.Ti version 9.0.7. Subsequently, codes were summarized in a thematic mind-map and discussed with KP to verify the results.

Ethical considerations

Prior to participation in the extensive survey and in-depth interview, participants received an information letter by email and gave online informed consent for participation. The study protocol was approved by The Faculty of Health, Medicine and Life Sciences Research Ethics Committee of Maastricht University (FHML-REC/2020/090).

Results

A total of 26 participants completed the pop-up survey. Next, 19 participants completed the extensive survey, and ten participated in the in-depth interviews. The two recruitment routes are shown in Figure 1.

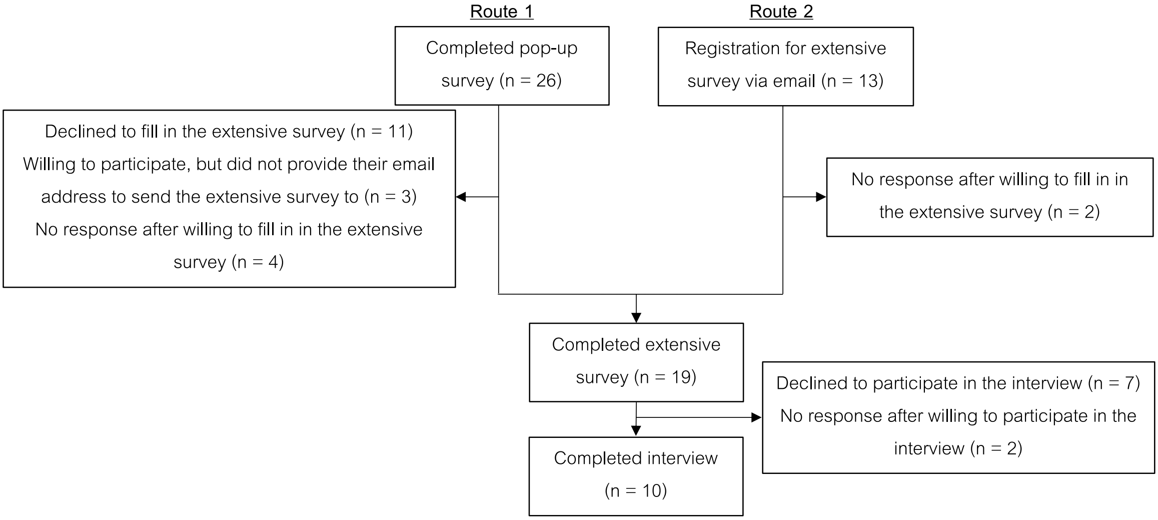

Figure 1. *Routes for participation flow diagram*

Participant characteristics

The majority of participants who completed the pop-up survey had a relative with dementia (53.8%) or were professionally involved with dementia (46.2%). A varied sample of participants completed the extensive survey and participated in the in-depth interviews, such as spouses, children, or other relatives of persons with different YOD-subtypes, and healthcare professionals with different backgrounds (Table 1).

Table 1. *Participant characteristics*

		Extensive survey (n = 19)	Interview (n = 10)
Caregivers		N (= 10)	N (= 7)
Gender	Male	5	4
	Female	5	3
Age	30-39 years	2	1
	40-49 years	0	0
	50-59 years	4	3
	60-65 years	2	1
	> 65 years	2	2
Relationship to person with dementia	Spouse	6	4
	Brother/sister	2	2
	Father/mother (in-law)	1	1
	Not specified	1	0
Diagnosis	Alzheimer's dementia	6	4
	Frontotemporal dementia	2	2
	Not specified	2	1
Years since diagnosis	< 3 years	2	1
	3-5 years	4	4
	> 5 years	2	1
	Not specified	2	1
Healthcare professionals		N (= 6)	N (= 2)
Gender	Male	0	0
	Female	5	2
	Not specified	1	0
Age	40-49 years	1	0
	50-59 years	2	1
	60-65 years	2	1
	> 65 years	1	0

Table 1. *(continued)*

		Extensive survey (n = 19)	Interview (n = 10)
Healthcare professionals		N (= 6)	N (= 2)
Background	Case manager	2	0
	Dementia care coordinator	1	1
	Dementia counselor	2	1
	Volunteer	1	0
Other		N (= 3)	N (= 1)
Gender	Male	0	0
	Female	3	1
Age	40-49 years	1	0
	50-59 years	2	1
Background	Caregiver and care professional	1	0
	Person with Alzheimer's dementia	1	1
	Not specified	1	0

Acceptability

Most participants who completed the extensive survey had high levels of computer self-efficacy (Table 2). The majority strongly agreed the program was useful, easy to use, and they would use it again.

Table 2. *Scores on user acceptability*

Technology Acceptance Model	Mean (SD)	Range
Perceived usefulness	6.42 (1.39)	1-7
Ease of use	5.95 (1.13)	1-7
Behavioral intention to use	5.63 (1.30)	1-7
Computer self-efficacy	6.11 (0.81)	1-7

In the in-depth interviews, participants expressed they had mixed feelings regarding the ease of use. Some experienced the navigation within the program as clear, whereas others experienced it as confusing. Participants suggested minor improvements to enhance the navigation. For example, a clear home-button redirecting to the entrance page, a table of contents at the start of each chapter, and providing clarification if a hyperlink led to an external web page.

"Sometimes it was hard to know where you were in the program. I wondered how to go back to the entrance page, especially if a link led me to an external web page."
– Dementia care coordinator –

Usability

Nine out of 19 participants (47.4%) who completed the extensive survey agreed it was challenging to find time to follow the program, five participants (26.3%) disagreed, and the remaining five (26.3%) neither agreed nor disagreed. Furthermore, two-thirds of the participants reported the level of detail, number of chapters, duration of chapters, and overall length of the program as exactly right, while the remaining one-third rated it as a lot.

Participants valued the possibility they could follow the program at any time and any location. Although most participants felt the program contained comprehensive information, it was highlighted that this is necessary for a topic like YOD. According to participants, this provided a reason to return to the program at a later stage when other topics are applicable.

"The program contains comprehensive information, but all the information is needed. YOD is a complex and diverse disease."
– Dementia counselor –

Furthermore, participants valued the language use in the program, and expressed it was understandable for those with and without a medical background. They felt the Dutch RHAPSODY program was relevant for YOD caregivers and healthcare professionals but also for persons not directly involved with YOD. For example, to create awareness and understanding about YOD among employers.

User satisfaction

Participants who completed the extensive survey were highly satisfied with the overall quality of the program, as 94.7% rated the quality good to excellent. Participants valued the relevance, understandability, and applicability of the content in daily life. Moreover, they rated each individual chapter and the reflective questions as useful. Approximately 80% of participants rated the overall layout and the layout of each individual chapter as clear. On the pop-up survey, 21 of 26 participants (80.8%) agreed or totally agreed the program helped in coping with YOD in daily life. The vast majority would recommend the program to others (Mean = 8.0, SD = 1.3, on a scale from 1 to 10). Qualitatively, participants highlighted the need for a program including educational and practical information about YOD bundled in one place.

"As a caregiver, there is always the question 'where do I find all the information?'. You can't see the forest for the trees. After looking into this program, this is the place I would recommend people visit."
– Daughter of a person with Alzheimer's dementia –

Participants experienced the alternation between texts and videos as pleasant. The great usefulness and applicability of practical tips were mentioned serval times during the interviews. Furthermore, participants recognized the information in their own daily experience and felt it retrospectively enhanced their understanding of symptoms of YOD. Participants were satisfied with the variety of topics covered in the program, especially regarding different subtypes of YOD, coping with behavioral symptoms, and availability of care and support services. However, several participants felt they had already obtained some of the information themselves in the past years. Thus, not all information was still relevant.

"At the beginning we searched for a lot of information. If only I had this training before, it would have been of great help, also to better understand my wife's behavior."
– Husband of a person with frontotemporal dementia –

"The tips provided in the program are good. Not only for caregivers but also for persons with dementia themselves."
– Person with Alzheimer's dementia –

User behavior

The entrance page of the Dutch RHAPSODY program registered 2,461 unique page views between October and December 2020 (see Appendix C). The page providing a background explanation about YOD (chapter 1) had the most visits and registered 360 unique page views. Visitors had spent most time reviewing content about the diagnostic trajectory (6.5 minutes) and difficulties in executing daily activities (4.4 minutes). The number of unique page views gradually decreased from the first to the last chapter. Overall, this same pattern was seen within the different pages of each individual chapter. In the three months after evaluating the program, the entrance page registered 2,218 page views, of which 1,855 were unique.

Discussion

Our quantitative and qualitative findings demonstrate that the Dutch RHAPSODY program had good user acceptability, usability, and user satisfaction. Participants indicated they would use the program again and recommended it to others. In-depth interviews emphasized the high need for education and tailored information about YOD, which was provided by the RHAPSODY program. Both caregivers and healthcare professionals were satisfied with the quality of the content and topics, such as information on different YOD-subtypes, coping with behavioral symptoms, and available care and support services. Minor suggestions for improvement were identified, such as enhancing the navigation within the program.

Participants valued the usefulness, relevance, and applicability of information in daily life. These results are consistent with the previous study on other versions of the RHAPSODY program (Metcalfe et al., 2019). For YOD caregivers, flexibility is an important aspect of using support programs (Cations et al., 2017). This also applied to the RHAPSODY program which allowed participants to visit it from home at a convenient time. Qualitative interviews imply that the Dutch RHAPSODY program is of added value to already existing support as the program met the information needs of YOD caregivers and healthcare professionals.

Approximately one-third of the participants reported that the amount of information within the program was a lot. This may explain why nine out of 19 persons agreed it was difficult to allocate time to follow the program. For caregivers it can be difficult to find time to follow the program as they already have to balance caregiving with other responsibilities, such as work, raising children, and maintaining social relationships (Brodaty and Donkin, 2009; Hvidsten et al., 2019). Interviewees revealed that a table of contents would enable more direct access to the desired information and would improve the navigation through the program content. Furthermore, participants highlighted it was pleasant to receive information tailored for different YOD-subtypes, because most available support is focused solely on Alzheimer's dementia or elderly peers. Previous research confirmed that caregivers have a need for YOD-subtype specific information (Bruinsma et al., 2021b; Rosness et al., 2008). The web metrics showed that most time was spent on the page about the diagnostic process. Diagnosing YOD is often complex and is associated with uncertainty for caregivers (van Vliet et al., 2011).

The Dutch RHAPSODY program was launched and evaluated during the Covid-19 pandemic. The need and usage of digital support programs for caregivers of persons with dementia accelerated during the pandemic (Cuffaro et al., 2020; Dourado et al., 2020). The pandemic may have facilitated the familiarization with digital services (Cuffaro et al., 2020). Due to the program's free and public availability through the Internet, persons could visit the program regardless of restrictions imposed by the Covid-19 pandemic. The program was co-designed and hosted by the Dutch Alzheimer's Society via their website. This partnership ensured its implementation, which helps to translate interventions into practice (Christie et al., 2018). Web metrics confirmed the program continued to have visitors even after actively recruiting participants for this study.

Although web-based programs are considered as easily accessible, they are often used in addition to face-to-face support (Klimova et al., 2019). Previous research indicates that caregivers feel to benefit most if online support is offered in addition to the support of a healthcare professional. Caregivers may for example perceive online support as impersonal and distant (Huis in het Veld et al., 2018). In the Netherlands there is tailored support available that blends a web-based approach with personal coaching, namely the Partner in Balance intervention (Bruinsma et al., 2021a; Bruinsma et al., 2021b). The RHAPSODY program may provide caregivers a steppingstone to participate in the other support options, for example Partner in Balance intervention.

To allow timely access to appropriate information regarding YOD, it is recommended that caregivers are aware of the program after obtaining the diagnosis. Our findings reveal that participating caregivers had spent a lot of time searching for reliable information online. Therefore, creating awareness about the Dutch RHAPSODY program is an important future direction. Healthcare professionals, such as dementia casemanagers, have an essential role in getting the right information at the right time to the right caregiver. Actively and personally informing caregivers about the program will likely stimulate caregivers to use it. This may also improve adherence, which is often limited in web-based programs (Kelders et al., 2012). Lastly, although the current study showed that the program is acceptable and usable, a direction for future research is to explore long-term effects on caregiver well-being or coping skills.

Our findings revealed that nearly half of the participants who completed the pop-up survey were healthcare professionals. This percentage is relatively high considering that caregivers are the primary target group of the program. This may indicate there is an educational need among healthcare professionals regarding YOD. Previous studies demonstrated that educational e-learning programs allow healthcare professionals to boost their knowledge (Delf, 2013). Therefore, e-learning plays an important role in (inter)professional education on YOD (Casimiro et al., 2009; Menard and Varpio, 2014). Involving care organizations affiliated with the YOD Knowledge Center in the Netherlands that support persons with YOD and their families could facilitate the use of the Dutch RHAPSODY program for the education of healthcare professionals. Ultimately, this improves the quality of care and support they provide to YOD caregivers. Embedding the Dutch RHAPSODY program in (inter)professional education programs on YOD offered by health academies and universities of applied science in the Netherlands would also enable healthcare professionals to get acquainted with the program.

Strengths and limitations

An important strength of the Dutch RHAPSODY program is that it was based on an international evidence-based program. This allowed us to cost-effectively dedicate our resources to tailor it to the Dutch context in close collaboration with caregivers, healthcare professionals, and field experts. Another strength is the current study allowed all visitors who accessed the program to participate in the evaluation study. No strict inclusion or exclusion criteria were defined as the program was freely and publicly accessible. This allowed for various ways of recruiting, for example via newsletters, social media, and flyers. For sustainable implementation, the program was embedded on the well-visited website (www.dementie.nl) of the Dutch Alzheimer's Society. In 2020, this website had over one million visits. This helped to raise awareness about the program's existence in a large audience of caregivers and healthcare professionals. Additionally, it ensures free, public, and structural access to the program.

The current study is accompanied by some limitations, such as the relatively small and heterogeneous sample, that may impede the generalizability of the results. Partly, the limited number of responses may be the result of visitors clicking away the pop-up survey. Participants who clicked away the pop-up survey did not receive a reminder. Moreover, the pop-up survey only appeared if participants visited a page longer than 30 seconds. Although the web metrics are a valuable contribution to the study, they should be interpreted with caution. For example, the duration of page visits is difficult to interpret as the amount and type of information per page differed, there is no insight into whether visitors viewed the program on different devices, or if they paused the online program while using it.

Conclusions

The Dutch RHAPSODY program provides an opportunity to sustainably educate and inform caregivers and healthcare professionals on YOD. Findings indicated good user acceptability, usability, and user satisfaction. The program met the information needs of caregivers of persons with YOD and was useful in daily life. Additionally, the RHAPSODY program had potential as an educational tool for healthcare professionals. Therefore, raising awareness about the RHAPSODY program among early stage YOD caregivers and their healthcare professionals is recommended. The partnership with the Dutch Alzheimer's Society was important as it facilitates the sustainable implementation of the program, this is essential to ensure free, public, and structural availability.

Conflict of interest

None.

Funding

The project was funded by The Netherlands Organization for Health Research and Development [*ZonMw*] and the Dutch Alzheimer's Society [*Alzheimer Nederland*]. RHAPSODY is an EU Joint Program Neurodegenerative Disease Research project.

Description of authors' roles

Development of the Dutch RHAPSODY program: M. Daemen and K. Peetoom developed the program in close collaboration with C. Bakker, M. de Vugt, A. Oostijen, R. Groot Zwaaftink, B. Loose, and J. Bruinsma. Program content was revised by J. Bruinsma, R. Koopmans, and F. Verhey.

Evaluating the Dutch RHAPSODY program: M. Daemen and K. Peetoom collected the data. R. Groot Zwaaftink monitored data collection, and together with other authors provided support in the recruitment of participants. M. de Vugt supervised the development and data collection.

Drafting the manuscript: M. Daemen performed the analyses and wrote this paper in close collaboration with J. Bruinsma and K. Peetoom. All other authors critically revised the paper.

Acknowledgments

The authors would like to thank the caregivers participating in the videos incorporated in the Dutch RHAPSODY program. Additionally, we express appreciation for caregivers and healthcare professionals who reviewed the program content and played an active role in recruiting the participants.

References

Arai, A., Matsumoto, T., Ikeda, M., and Arai, Y. (2007). Do family caregivers perceive more difficulty when they look after patients with early onset dementia compared with late onset dementia? Int J Geriatr Psychiatry, 22(12), 1255–1261. doi:10.1002/gps.1935

Barnes, J., Dickerson, B. C., Frost, C., Jiskoot, L. C., Wolk, D. and van der Flier, W. M. (2015). Alzheimer's disease first symptoms are age dependent: evidence from the NACC dataset. Alzheimers Dement, 11(11), 1349-1357. doi:10.1016/j.jalz.2014.12.007

Brodaty, H., and Donkin, M. (2009). Family caregivers of people with dementia. Dialogues Clin Neurosci, 11(2), 217. doi:10.31887/DCNS.2009.11.2/hbrodaty

Bruinsma, J., Peetoom, K., Bakker, C., Boots, L., Verhey, F., and de Vugt, M. (2021). "They simply do not understand": a focus group study exploring the lived experiences of family caregivers of people with frontotemporal dementia. Aging Ment Health. doi:10.1080/13607863.2020.1857697

Bruinsma, J., Peetoom, K., Bakker, C., Boots, L., Millenaar, J., Verhey, F., and de Vugt, M. E. (2021a). Tailoring and evaluating the web-based 'Partner in Balance' intervention for family caregivers of persons with young-onset dementia. Internet Interv, 25. doi:10.1016/j.invent.2021.100390

Bruinsma, J., Peetoom, K., Boots, L., Daemen, M., Verhey, F., Bakker, C., and de Vugt, M. E. (2021b). Tailoring the web-based 'Partner in Balance' intervention to support spouses of persons with frontotemporal dementia. Internet Interv, 26. doi:10.1016/j.invent.2021100442

Cartwright, A. V., Stoner, C. R., Pione, R. D., and Spector, A. (2021). The experiences of those affected by parental young onset dementia: A qualitative systematic literature review. Dementia, online ahead of print. doi:10.1177/1471301220988231

Casimiro, L., MacDonald, C. J., Thompson, T. L., and Stodel, E. J. (2009). Grounding theories of W(e)Learn: A framework for online interprofessional education. J Interprof Care, 23(4), 390-400. doi:10.1080/13561820902744098

Cations, M., Withall, A., Horsfall, R., Denham, N., White, F., Trollor, J., Loy, C., Brodaty, P. S., Gonski, P., Demirkol, A., Cumming, R. G., and Draper, B. (2017). Why aren't people with young onset dementia and their supporters using formal services? Results from the INSPIRED study. PLoS One, 12(7), e0180935. doi:10.1371/journal.pone.0180935

Cabote, C. J., Bramble, M., and McCann, D. (2015). Family caregivers' experiences of caring for a relative with younger onset dementia: a qualitative systematic review. J Fam Nurs, 21(3), 443-468. doi:10.1177/1074840715573870

Cuffaro, L., Di Lorenzo, F., Bonavita, S., Tedeschi, G., Leocani, L., and Lavorgna, L. (2020). Dementia care and COVID-19 pandemic: a necessary digital revolution. Neurol Sci, 41(8), 1977-1979. doi:10.1007/s10072-020-04512-4

Christie, H. L., Bartels, S. L., Boots, L. M. M., Tange, H. J., Verhey, F. R. J., and de Vugt, M. E. (2018). A systematic review on the implementation of eHealth interventions for informal caregivers of people with dementia. Internet Interv, 13, 51–59. doi:10.1016/j.invent.2018.07.002

Delf, P. (2013). Designing effective eLearning for healthcare professionals. Radiography, 19(4), 315-320. doi:10.1016/j.radi.2013.06.002

Dix, A., Finlay, J., Abowd, G. D., and Beale, R. (2003). Human-computer interaction. Pearson Education.

Dourado, M. C. N., Belfort, T., Monteiro, A., Lucena, A. T. D., Lacerda, I. B., Gaigher, J., ... and Marinho, V. (2020). COVID-19: challenges for dementia care and research. Dement neuropsychol, 14(4), 340-344. doi:10.1590/1980-57642020dn14-040002

Ducharme, F., Kergoat, M.-J., Antoine, P., Pasquier, F. and Coulombe, R. (2013). The Unique Experience of Spouses in Early-Onset Dementia. Am J Alzheimer's Dis Other Dement, 28(6), 634-641. doi:10.1177/1533317513494443

Ducharme, F., Kergoat, M. J., Coulombe, R., Lévesque, L., Antoine, P., and Pasquier, F. (2014). Unmet support needs of early-onset dementia family caregivers: a mixed-design study. BMC Nurs, 13(1), 1-10. doi:10.1186/s12912-014-0049-3

Ducharme, S. and Dickerson, B. C. (2015). The neuropsychiatric examination of the young-onset dementias. Psychiatric Clinics, 38(2), 249-264. doi:10.1016/j.psc.2015.01.002.

Elo, S., and Kyngäs, H. (2008). The qualitative content analysis process. J Adv Nurs, 66(11), 2460–2470. doi:10.1111/j.1365-2648.2007.04569.x

Eysenbach, G. (2011). CONSORT-EHEALTH: improving and standardizing evaluation reports of Web-based and mobile health interventions. J Med Internet Res, 13(4), e126. doi:10.2196/jmir.1923

Godwin, K. M., Mills, W. L., Anderson, J. A., and Kunik, M. E. (2013). Technology-driven interventions for caregivers of persons with dementia: a systematic review. Am J Alzheimer's Dis other Dement, 28(3), 216-222. doi:10.1177/1533317513481091

Grundberg, A., Sandberg, J., and Craftman, A. G. (2021). Childrens' and young adults' perspectives of having a parent with dementia diagnosis: A scoping review. Dementia, online ahead of print. doi:10.1177/14713012211023653

Huis in het Veld, J. G. Verkaik, R., van Meijel, B., Verkade, P-J., Werkman, W., Hertogh, C. M. P. M., and Francke, A. L. (2018). Self-management support and ehealth when managing changes in behavior and mood of a relative with dementia: an asynchronous online focus group study of family caregivers' needs. Res Gerontol Nurs, 11(3), 151-159. doi:10.3928/19404921-20180216-01

Hvidsten, L., Engedal, K., Selbaek, G., Wyller, T. B., Benth, J. S., BruvikF., and Kersten, H. (2020). Quality of life of family carers of persons with young-onset compared to late-onset dementia. Aging ment health, 24(9), 1394-1401. doi:10.1080/13607863.2019.1617245

Kaushik, V., and Walsh, C. A. (2019). Pragmatism as a research paradigm and its implications for social work research. Soc Sci, 8(9), 255. doi:10.3390/socsci8090255

Kelders, S. M., Kok, R. N., Ossebaard, H. C., and van Gemert-Pijnen, J. E. (2012). Persuasive system design does matter: a systematic review of adherence to web-based interventions. J Med Internet Res, 14(6), e152. doi:10.2196/jmir.2104

Klimova, B., Valis, M., Kuca, K., and Masopust, J. (2019). E-learning as valuable caregivers' support for people with dementia–A systematic review. BMC Health Serv Res, 19(1), 1-7. doi:10.1186/s12913-019-4641-9

Koedam, E. L., Lauffer, V., van der Vlies, A. E., van der Flier, W. M., Scheltens, P., and Pijnenburg, Y. A. (2010). Early-versus late-onset Alzheimer's disease: more than age alone. J Alzheimers Dis, 19(4), 1401-1408. doi:10.3233/JAD-2010-1337

Koopmans, R., and Rosness, T. (2014). Young onset dementia–what does the name imply?. Int Psychogeriatr, 26(12), 1931-1933. doi:10.1017/S1041610214001574

Kuruppu, D. K., and Matthews, B. R. (2013). Young-onset dementia. In Seminars in neurology (Vol. 33, No. 04, pp. 365-385). Thieme Medical Publishers. doi:10.1055/s-0033-1359320

Kurz, A., Bakker, C., Bohm, M., Diehl-Schmid, J., Dubois, B., Ferreira, C., Gage, H., Graff, C., Hergueta, T., Jansen, S., Jones, B., Komar, A., de Mendonça, A., Metcalfe, A., Milecka, K., Millenaar, J., Orrung Wallin, A., Oyebode, J., Schneider-Schelte, H., Saxl, S., and de Vugt, M. (2016). RHAPSODY - Internet-based support for caregivers of people with young onset dementia: program design and methods of a pilot study. Int Psychogeriatr, 28(12), 2091-2099. doi:10.1017/S1041610216001186

Lim, L., Zhang, A., Lim, L., Choong, T. M., Silva, E., Ng, A., and Kandiah, N. (2018). High caregiver burden in young onset dementia: what factors need attention?. J Alzheimer's Dis, 61(2), 537-543. doi:10.3233/JAD-170409

Lockeridge, S. and Simpson, J. (2013). The experience of caring for a partner with young onset dementia: how younger carers cope. Dementia, 12(5), 635-651. doi:10.1177/1471301212440873

Mayrhofer, A. M., Greenwood, N., Smeeton, N., Almack, K., Buckingham, L., Shora, S., and Goodman, C. (2021). Understanding the financial impact of a diagnosis of young onset dementia on individuals and families in the United Kingdom: Results of an online survey. Health Soc Care Community, 29(3), 664-671. doi:10.1111/hsc.13334

Mayrhofer, A., Mathie, E., McKeown, J., Bunn, F., and Goodman, C. (2018). Age-appropriate services for people diagnosed with young onset dementia (YOD): a systematic review. Aging Ment Health, 22(8), 927-935. doi:10.1080/13607863.2017.1334038

Menard, P., and Varpio, L. (2014). Selecting an interprofessional education model for a tertiary health care setting. J Interprof Care, 28(4), 311-316. doi:10.3109/13561820.2014.893419

Metcalfe, A., Jones, B., Mayer, J., Gage, H., Oyebode, J., Boucault, S., Aloui, S., Schwertel, U., Böhm, M., Tezenas du Montcel, S., Lebbah, S., De Mendonça, A., De Vugt, M., Graff, C., Jansen, S., Hergueta, T., Dubois, B., and Kurz, A. (2019). Online information and support for carers of people with young onset dementia: A multi-site randomised controlled pilot study. Int J Geriatr Psychiatry, 34(10), 1455–1464. doi:10.1002/gps.5154

Millenaar, J. K., van Vliet, D., Bakker, C., Vernooij-Dassen, M. J., Koopmans, R. T. C. M., Verhey, F. R., and de Vugt, M. E. (2014). The experiences and needs of children living with a parent with young onset dementia: results from the NeedYD study. Int Psychogeriatr, 26(12), 2001-2010. doi:10.1017/S1041610213001890

Millenaar, J. K., Bakker, C., Koopmans, R. T., Verhey, F. R., Kurz, A., and de Vugt, M. E. (2016a). The care needs and experiences with the use of services of people with young-onset dementia and their caregivers: a systematic review. Int J Geriatr Psychiatry 31(12), 1261–1276. doi:10.1002/gps.4502

Onyike, C. U., and Diehl-Schmid, J. (2013). The epidemiology of frontotemporal dementia. Int Rev Psychiatry, 25(2), 130-137. doi:10.3109/09540261.2013.776523

Rahimi, B., Nadri, H., Afshar, H. L., and Timpka, T. (2018). A systematic review of the technology acceptance model in health informatics. App Clic Inform, 9(03), 604-634. doi:10.1055/s-0038-1668091

Rosness, T. A., Haugen, P. K., and Engedal, K. (2008). Support to family carers of patients with frontotemporal dementia. Aging Ment Health, 12(4), 462-466. doi:10.1080/13607860802224334

Rosness, T. A., Haugen, P. K., Gausdal, M., Gjøra, L., and Engedal, K. (2012). Carers of patients with early-onset dementia, their burden and needs: a pilot study using a new questionnaire--care-EOD. Int J Geriatr Psychiatry, 27(10), 1095-1096. doi:10.1002/gps.2818

Rossor, M. N., Fox, N. C., Mummery, C. J., Schott, J. M., and Warren, J. D. (2010). The diagnosis of young-onset dementia. Lancet Neurol, 9(8), 793-806. doi:10.1016/s1474-4422(10)70159-9

Spreadbury, J. H., and Kipps, C. M. (2018). Understanding important issues in young-onset dementia care: the perspectives of healthcare professionals. Neurodegener Dis Manag, 8(1), 37-47. doi:10.2217.nmt-2017-0029

Svanberg, E., Spector, A., and Stott, J. (2011). The impact of young onset dementia on the family: a literature review. Int Psychogeriatric, 23(3), 356. doi:10.1017/S1041610210001353

van Vliet, D., de Vugt, M. E., Bakker, C., Koopmans, R. T., and Verhey, F. R. (2010b). Impact of early onset dementia on caregivers: a review. Int J Geriatr Psychiatry, 25(11), 1091–1100. doi:10.1002/gps.2439

van Vliet, D., de Vugt, M. E., Bakker, C., Koopmans, R. T., Pijnenburg, Y. A., Vernooij-Dassen, M. J., and Verhey, F. R. (2011). Caregivers' perspectives on the pre-diagnostic period in early onset dementia: a long and winding road. Int Psychogeriatr, 23(9), 1393-1404. doi:10.1017/S1041610211001013

van de Veen, D., Bakker, C., Peetoom, K., Pijnenburg, Y., Papma, J. M., PRECODE study group., de Vugt, M., and Koopmans, R. (2021). An Integrative Literature Review on the Nomenclature and Definition of Dementia at a Young Age. J Alzheimers Dis. doi:10.3233/JAD-210458

Venkatesh, V., and Bala, H. (2008). Technology acceptance model 3 and a research agenda on interventions. Decis Sci, 39(2), 273-315. doi:10.1111/j.1540-5915.2008.00192.x

Waller, A., Dilworth, S., Mansfield, E., and Sanson-Fisher, R. (2017). Computer and telephone delivered interventions to support caregivers of people with dementia: a systematic review of research output and quality. BMC Geriatr, 17(1), 1-20. doi:10.1186/s12877-017-0654-6

Appendix A

Table 3. *Six chapters incorporated in the Dutch RHAPSODY program*

Chapter	Topics
Introduction about YOD General information on YOD.	- What is YOD? How does it differ from late-onset dementia? - Medical background about the different brain functions and consequences of impairment - Diagnostic trajectory - Heredity of YOD - Most common subtypes of YOD: Alzheimer's dementia, frontotemporal dementia, Lewy body dementia, and vascular dementia - Medical and non-medical treatment options
Problems and solutions Tips on coping with daily challenges.	- Cognitive symptoms such as memory deficits and changes in social behavior - Difficulties in executing daily activities - Physical health and bodily functions
Behavioral changes What are behavioral changes? Tips on coping.	- Behavioral changes and causes - Recognizing emotional symptoms - Changes in social behavior - Lack of initiative and passive behavior - Physical behavior including repetitive behavior, agitation, and aggressive behavior - Perceptions on reality and hallucinations
Impact on family members Changing family roles and suggestions to improve communication.	- Changes in family roles and responsibilities - Consequences for children and suggestions on supporting them - Talking about YOD and accompanying changes with family members - Coping with emotions such as guilt, anger, and grief
Self-care Look after yourself, and balance caregiving with personal life.	- Experienced burden and preventing distress - Finding a balance between caregiving and personal needs - Having unrealistic expectations towards yourself - Focus on activities that are still possible to undertake - Searching and accepting support

Table 3. *(continued)*

Chapter	Topics
Available support in the Netherlands Where to find appropriate care?	- Specialized YOD healthcare and availability in the Netherlands - Explanation about the roles of healthcare professionals involved after obtaining the YOD diagnosis - Available types of care and support for persons with YOD - Available types of care and support for caregivers - An overview of the relevant laws and legislation - Care arrangements, including daycare, advanced care planning, and financial matters - Arranging nursing home admission - Arranging practical issues after passing away

Appendix B

Table 4. *Semi-structured interview guide*

Main questions	Subtopics
How did you experience the use of the online program?	
What do you think about the content of the online program?	- Which chapters did you follow? - Was the information relevant for your situation? - What do you think of the tips provided in the program? - What do you think of the amount of information provided?
What do you think of the understandability of the program?	- Which parts were easy or hard to understand?
What do you think of the layout of the program?	- What do you think about the chapter layout? The pictures? The videos?
What do you think of the usability of the program?	- Which parts were easy or hard to use? For example, the navigation, external weblinks, and hand-outs.
What do you think of the videos within the online program?	- Was the information provided in the videos clear and understandable? - Are the videos of added value to the program?
Do you think you would use the program again?	- If yes, any chapters in particular? - If no, why not?
Would you recommend the program to others?	- If yes, to whom? Why?
Do you have any points for improvement for the online program?	- Did you miss information?

Appendix C

Table 5. *User behavior from October to December 2020*

	Page views (n)	Unique page views (n)	Average time on page (sec)
Entrance page	3,027	2,461	129.8
Main page chapter 1	1,256	750	50.4
YOD explanation	439	360	137.5
Medical background	400	344	150.7
Diagnostic process	278	242	392.8
Heredity of YOD	190	165	186.1
YOD-subtypes	221	179	129.3
Treatment	120	109	204.8
Main page chapter 2	428	268	58.8
Cognitive problems	285	146	66.9
Daily activities	164	146	265.0
Physical health	92	51	34.6
Main page chapter 3	402	250	38.7
Behavioral changes	138	125	196.7
Mood changes	98	86	126.9
Social behavior	98	81	60.6
Lack of initiative	93	73	47.6
Physical behavior	61	54	88.1
Perceptions on reality	68	59	70.6
Main page chapter 4	196	121	36.9
Changes in relationships	53	45	126.9
Children	49	47	98.6
Communication	37	35	56.5
Emotions	57	51	62.4
Main page chapter 5	127	90	46.0
Burden	45	41	121.5
Balancing caregiving	45	44	169.9
Unrealistic expectations	48	43	45.3
Activities	39	37	51.3
Finding help	30	27	63.8

Table 5. *(continued)*

	Page views (n)	Unique page views (n)	Average time on page (sec)
Main page chapter 6	133	103	92.1
YOD healthcare	28	26	169.3
Healthcare professionals	50	43	82.4
Support for persons with dementia	41	24	75.7
Support for caregivers	40	25	54.4
Laws	57	50	216.9
Regulations	29	26	54.3
Nursing home admission	18	17	45.8
After passing away	14	14	34.6

Chapter 6

Tailoring and evaluating the web-based 'Partner in Balance' intervention for family caregivers of persons with young onset dementia

Bruinsma, J., Peetoom, K., Bakker, C., Boots, L., Millenaar, J., Verhey, F., and de Vugt, M.

Internet Interventions, 2021

Abstract

Young-onset dementia (YOD) poses specific challenges for caregivers involved. However, most available support does not address their specific needs. Previously, the web-based Partner in Balance intervention showed promising results and facilitated role adaptation in dementia caregivers. Although the web-based format proved a good fit for YOD caregivers, the evaluation showed a need for tailored content on YOD. Therefore, new content was iteratively developed respectively for spouses and other family caregivers of persons with YOD. This study evaluates how caregivers perceived the tailored content.

Methods: A pre-post design was used to prospectively evaluate how end-users perceived two tailored versions of the Partner in Balance intervention, one for spouses and one for other family members of people with YOD. After the intervention, participants were interviewed for approximately 60 minutes in-person or by telephone using the Program Participation Questionnaire. A qualitative deductive content analysis was used to evaluate (1) usability, (2) feasibility and acceptability, (3) perceptions on intervention content. To evaluate if the intervention facilitated role adaptation, preliminary effects were examined using pre-post questionnaires on self-efficacy, mastery, stress, anxiety, and depression.

Results: Spouses (n = 11) and other family members (n = 14) both positively evaluated the tailored content on YOD and valued that the web-based approach could easily be integrated in daily life. Participants perceived the intervention as usable, feasible and acceptable. Participants valued the recognizability of the content. Goal-setting helped participants to translate the intervention to daily life, although for some participants setting goals was difficult. Caregivers of persons with frontotemporal dementia suggested incorporating specific content to further increase recognizability. After participation, participants felt better equipped for the caregiving role. In line with previously demonstrated effects on generic modules of Partner in Balance, the tailored version increased levels of self-efficacy in the group of other family caregivers, $t(12) = 3.37$, $p = .006$.

Conclusion: The tailored Partner in Balance intervention was positively evaluated by YOD caregivers. Offering participants more guidance on goal-setting and adding content about frontotemporal dementia may facilitate implementation.

Keywords: Young-onset dementia; Caregiver; eHealth; Psychosocial support; Intervention; Tailoring

Introduction

Symptoms of young-onset dementia (YOD) start before the age of 65 and pose specific challenges, not only to the person with YOD but also to the family members involved (Cabote et al., 2015; Rossor et al., 2010). Due to the young age at onset, spouses often combine caregiving with employment (Caceres et al., 2016; van Vliet et al., 2010b), making it difficult for the caring partner to balance care related responsibilities and family life (Kilty et al., 2019; Roach et al., 2013). Children may still live at home and gradually become caregiver for their parent with YOD (Millenaar et al., 2014). This may have negative long-term effects on the future of children. For example, because they postpone plans regarding studying or moving out of the parental house (Millenaar et al., 2014). Family members often experience difficulty in coping with the profound changes in personality and behavior in their relative with YOD and feel uncertain about the future. This is known to cause high levels of burden and distress in family members (Cabote et al., 2015; van Vliet et al., 2010b). Additionally, family members are known to experience a shift in their social role as they become increasingly responsible for their relative with YOD (Hutchinson et al., 2016; Richardson et al., 2016). The ability to adapt to the role of caregiver is complicated by specific characteristics of YOD that prolong the time to diagnosis. The time to diagnosis is 4.4 years in YOD while in late onset dementia this is 2.8 years (van Vliet et al., 2013). This diagnostic delay impedes role adaptation because a diagnosis helps family members to understand the changes in their relative and to see the caregiving role in perspective (de Vugt and Verhey, 2013).

Psychosocial support may help caregivers to come to terms with their new role (de Vugt and Verhey, 2013; Kilty et al., 2019; Millenaar et al., 2018). However, most available support is targeted at spouses of persons with YOD or caregivers of older persons with dementia (Cations et al., 2017; Millenaar et al., 2016a). Given the impact of YOD on family life it is indicated that support should target the family as a whole (Chapman et al., 2019; Hutchinson et al., 2016; Kaizik et al., 2017; Kilty et al., 2019). Supporting the whole family may create a sense of togetherness and this may empower the caregiving system by making caregivers more resilient (Cabote et al., 2015; Roach et al., 2013). As all family members have their unique support needs (Millenaar et al., 2014), a tailored approach seems required to adequately support spouses and other family members of persons with YOD. Given the low prevalence of YOD and the active life phase of the family members involved, web-based support tools may provide an opportunity to facilitate accessible support (Diehl-Schmid et al., 2013; Harvey et al., 2003). However, web-based support for family members of persons with YOD is scarce or primarily aimed at psychoeducation (Kaizik et al., 2017; Kurz et al., 2016; Nichols et al., 2013). To facilitate role adaptation, more attention is needed for support that helps family members cope with daily challenges.

The web-based Partner in Balance intervention uses self-management principles to facilitate role adaptation in caregivers by increasing levels of self-efficacy, sense of mastery, and the quality of life in caregivers (Boots et al., 2018; Boots et al., 2016). In Partner in Balance, caregivers watch video vignettes, read background information, make self-reflection assignments, and set goals for the future together with a personal coach. Currently, efforts are made to implement the intervention (Christie et al., 2020). The web-based format showed to be a good fit for caregivers of persons with YOD as they are more likely to participate due to the online nature, compared to older caregivers (Boots et al., 2018). Despite this, a process evaluation revealed that caregivers of persons with YOD missed specific content on YOD in the Partner in Balance intervention, as most original content portrays elderly (Boots et al., 2017). Therefore, new tailored content was separately developed for spouses and other family caregivers of persons with YOD in order to be incorporated in the intervention. This study reports the results of two end-user tests regarding these tailored versions of the Partner in Balance intervention. Based on previous research (Boots et al., 2018; Boots et al., 2016), we hypothesized that the tailored Partner in Balance intervention would provide a good fit in terms of usability, feasibility, and acceptability. Additionally, we hypothesized that participants would report higher levels of self-efficacy and mastery, and lower levels of stress, anxiety, and depression after the intervention.

Methods

This feasibility study used a pre-post design to prospectively evaluate how end-users perceive newly incorporated content in the Partner in Balance intervention in terms of usability, feasibility, acceptability, and perceptions on content (Aggarwal and Ranganathan, 2019).

The Partner in Balance intervention

The web-based Partner in Balance intervention is an effective and feasible support tool for family caregivers of persons with dementia who still live at home (Boots et al., 2018). The intervention has been iteratively developed using a stepwise approach guided by the Medical Research Council (MRC) framework for developing and evaluating complex interventions (Figure 1) (Boots et al., 2016; Craig et al., 2008). The intervention incorporates self-management principles to help caregivers find a balance between caregiving and daily life (Boots et al., 2017). Caregivers follow the intervention individually and receive personal online coaching from a trained healthcare professional while they subsequently follow four self-chosen thematic modules online (Table 1).

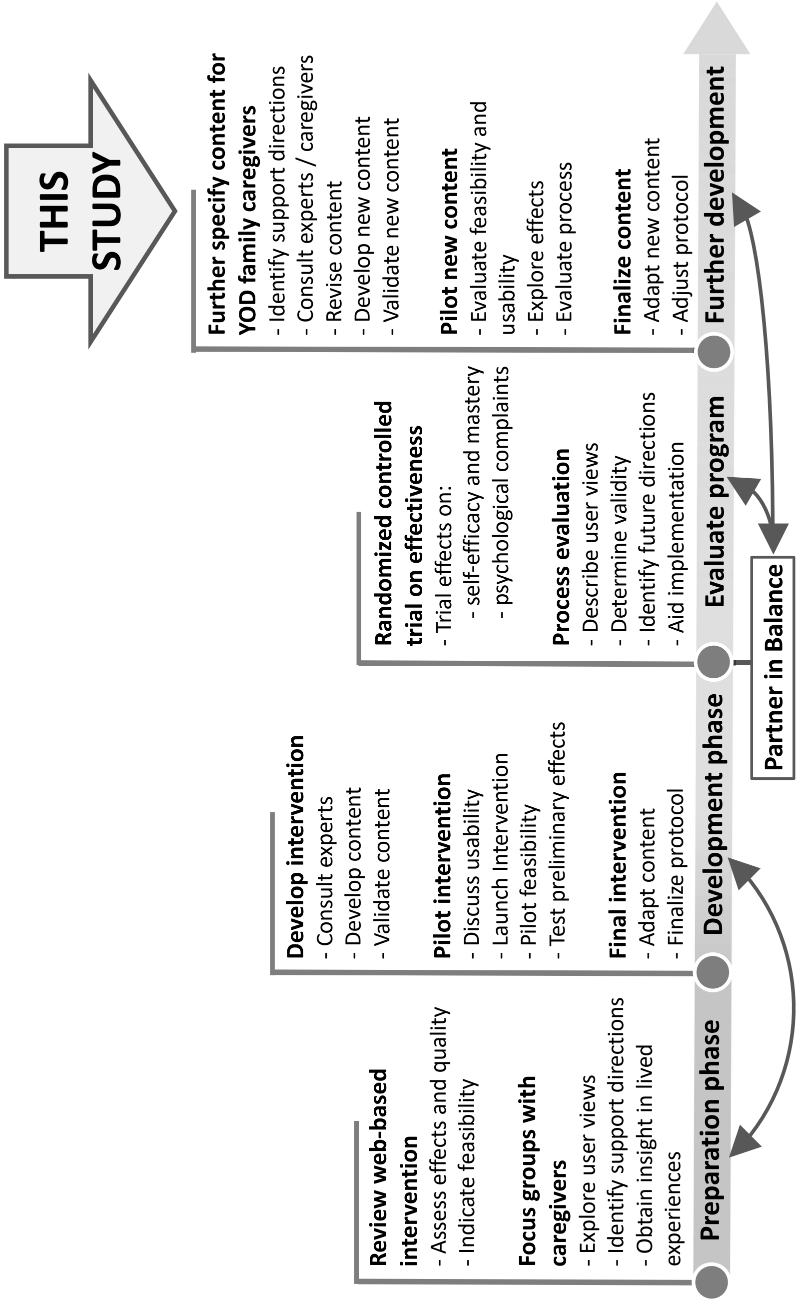

Figure 1. *Development of the Partner in Balance intervention*

The duration of Partner in Balance is approximately eight to ten weeks, but this is flexible (Boots et al., 2017). Each module includes (1) a video vignette in which caregivers share their experiences about a specific theme, (2) psychoeducation including a narrative story and practical tips, (3) a self-reflection assignment, and (4) a step-by-step change plan (Boots et al., 2016). Per module, the coach provides feedback to support caregivers with reflecting on their situation, and to formulate specific and attainable goals. Using a built-in chat, caregivers are also able ask questions directly to the coach. After completing four modules, caregivers reflect on their personal development together with the coach. On average, coaches spend around 6 hours during eight weeks to supervise caregivers using Partner in Balance (Boots et al., 2017).

Incorporating new content for YOD caregivers in the Partner in Balance intervention

After the effect evaluation, a process evaluation revealed that the Partner in Balance intervention needed specific tailoring for spouses and other family caregivers (e.g. children, brothers or sisters, and parents) of persons with YOD (Boots et al., 2018; Boots et al., 2017). New intervention content was iteratively developed using the MRC-framework. First, experiences and needs of YOD caregivers were identified from the literature and derived from the results of the Needs in Young onset Dementia (NeedYD) study (Bakker et al., 2013; van Vliet et al., 2010a). Second, directions for adaptation of the intervention were verified by consulting field experts (researchers, a neurologist, a clinical geneticist), healthcare professionals (dementia casemanagers, psychologists), and family members of persons with YOD. All original intervention content was critically revised to fit the specific needs of YOD caregivers. This resulted in the development of module content specific for either spouses or other family members of persons with YOD. Additionally, four new modules were developed covering (1) the impact of dementia on family life, (2) combining caregiving tasks with work or school, (3) sexuality and intimacy, and (4) worries about heredity (see Table 1). New video vignettes were developed by conducting in-depth interviews with four spouses and three other family members (i.e. two children and a sister) of a person with YOD. The interviews also allowed validating and supplementing the newly developed module content. Subsequently, a draft version was put online, and feedback was asked to field experts, healthcare professionals, spouses, and other family members of persons with YOD. This resulted in two separate versions of the Partner in Balance intervention; one for spouses and one for other family members. Two end-user tests were conducted to evaluate both versions. In the end-user tests participants followed four tailored modules in conjunction with receiving online coaching. All participants were interviewed afterwards to evaluate the newly developed modules and completed a questionnaire before and after the intervention to obtain insight in preliminary effects on role adaptation.

Table 1. Incorporated modules in the Partner in Balance intervention

Modules	**Original modules**	**New modules for YOD caregivers**	
	Boots et al., 2016	Spouses	Other family members
Combining care with work		x	x
Impact on family life		x	x
Sexuality and intimacy		x	
Worries about heredity			x
Coping with stress*	x		
Acceptance	x	x	x
Balance in activities	x	x	x
Changes in relative with YOD	x	x	x
Communication	x	x	x
Focusing on the positive	x	x	x
Insecurities and rumination	x	x	x
Self-understanding	x	x	x
Social relationships and support	x	x	x

** The module on coping with stress was merged with focusing on the positive and insecurities and rumination.*

Recruitment of participants

Spouses and other family members were recruited for the end-user tests by (1) spreading information within the network of Alzheimer Center Limburg/MUMC+, (2) providing information about the study in bimonthly meetings with healthcare providers affiliated with the Dutch Young-onset Dementia Knowledge Center [*Kenniscentrum Dementie op Jonge Leeftijd*], and by (3) distributing information via social media and meetings of the Dutch peer-support organization [*FTD lotgenoten*]. Spouses and other family caregivers contacted the first author if they wanted to participate. A previous study demonstrated that the inclusion of ten participants was sufficient to evaluate generic modules of Partner in Balance (Boots et al., 2016). Other feasibility studies on interventions for dementia caregivers included smaller sample sizes (Fick et al., 2011; Lai et al., 2012). Due to the heterogeneity of YOD we aimed to include ten spouses and 15 other family caregivers in our end-user test. Participants were eligible for participation if (1) they were aged 16 years or older, (2) they had a spouse or other relative with dementia with a symptom onset before the age of 65, and (3) their relative with dementia still lived at home.

During the end-user test, participants received online coaching (Boots et al., 2017). Therefore, psychologists and dementia casemanagers (specialized nurses or social workers) were recruited to become a coach. Preferably, these healthcare professionals would coach a caregiver from their own caseload. Healthcare professionals were eligible when they were employed as a dementia casemanager or psychologist and had practical experience with supporting YOD caregivers. At the start of the intervention, all healthcare professionals received a two-hour training to get acquainted with Partner in Balance, principles of self-management, and elements of web-based support (Boots et al., 2016). Following suggestions of Christie et al. (2021), a low threshold was pursued for technological support and intervision with more experienced coaches. Therefore, the first author contacted the coaches on a biweekly basis to monitor the process and evaluate if they encountered any difficulties. Also, intervision was organized between experienced and less experienced coaches. In addition to external healthcare professionals, coaching was also performed by five coaches of the Alzheimer Center Limburg with a background in psychology and extensive experience with coaching in Partner in Balance.

Measurements used during the end-user test

In line with previous studies on the Partner in Balance intervention (Boots et al., 2018; Boots et al., 2016), qualitative and quantitative measures were used to evaluate the newly developed content.

The Program Participation Questionnaire

To obtain insight in how participants perceived the intervention, semi-structured interviews were conducted after the intervention by means of the Program Participation Questionnaire (PPQ) (Boots et al., 2016). The PPQ consists of 33 items covering (1) how the intervention was used and implemented in daily life, (2) if the intervention was feasible, usable, and acceptable, and (3) how the quality and quantity of the content was perceived. Each item can be scored on a Likert scale ranging from 1 “strongly disagree” to 7 “strongly agree”. To obtain insight in participant reflections and experiences with the intervention, caregivers were encouraged to elaborate on their scores during the interview, while the researchers made fieldnotes. The PPQ also examines role adaptation by asking if the intervention positively influenced coping and if caregivers felt more confident towards their role as a caregiver.

Analysis of the Program Participation Questionnaire

The results of the PPQ were first analyzed quantitatively by calculating descriptive statistics such as mean, range, and percentiles. Items with a mean lower than 5 “slightly agree” were considered as a direction for improvement. To estimate the overall feasibility, usability and acceptability, a total PPQ-score was calculated ranging between 33 and 231 (Median = 132). In line with previous studies (Boots et al., 2016; Campbell et al., 2012), the median was used as a cut-off score to determine overall feasibility, usability, and acceptability. Subsequently, a deductive qualitative content analysis was performed using fieldnotes to interpret the quantitative scores on the PPQ (Elo and Kyngäs, 2008). A pragmatic theoretical stance was used to evaluate if the intervention matched the needs of the target group (Morgan, 2014). Therefore, the first author deductively coded and classified all fieldnotes in five categories focusing on the (1) use of the intervention in daily life, (2) feasibility, usability, and acceptability, (3) quantity and quality of the content, (4) role adaptation and well-being, and (5) suggestions for improvement and directions for the future. The coded and categories were discussed in a consensus meeting with the second author, after which the findings were also discussed with the wider research team to verify and substantiate the findings.

Preliminary effects on role adaptation

Additionally, we aimed to evaluate if the effects of the tailored versions were in line with those of the previously conducted feasibility study and randomized control trial (Boots et al., 2016; Boots et al., 2018). Therefore, a set of questionnaires was composed to examine preliminary effects and evaluate if the tailored versions of the Partner in Balance intervention facilitated role adaptation. Similar to earlier studies, questionnaires covered both self-efficacy and mastery. Self-efficacy was assessed with the Self-Efficacy Scale (CSES) (Fortinsky et al., 2002). Items covered care-management (six items) and service use (four items) and could be scored from 1 “not at all” to 10 “very”. Sense of mastery was assessed with the Pearlin Mastery Scale (PMS) (Pearlin and Schooler, 1978). Items covered sense of control and problem-solving ability, using seven items that could be scored between 1 “not at all” to 5 “totally agree”.

Role adaptation may have a positive effect on psychological well-being (Bandura, 1997; Boots et al., 2018; Boots et al., 2016). The questionnaire therefore also contained ten items of the Perceived Stress Scale (PSS) (Cohen, 1988). Items examined perceived levels of stress during the past seven days and were scored between 0 “never” to 4 “very often”. The Hospital Anxiety and Depression Scale (HADS) was also included (Zigmond and Snaith, 1983). Items assessed levels of anxiety (six items) and depression (seven items) and could be scores from 0 “not at all” to 3 “often”.

Analysis of the preliminary effects on role adaptation

To evaluate preliminary effects, the averaged questionnaire scores before and after the intervention were compared. Paired-sample t-testing was used to assess if any significant effects occurred. All analyses were conducted in SPSS (version 25.0) using an alpha level of 0.05 for two-sided tests.

Ethical considerations

The study protocol was approved by the Medical Ethics Committee of Maastricht University Medical Center, the Netherlands (METC: 2018-0443). Before inclusion, all participants received a letter including information about the study protocol, data-security, and privacy by email and were phoned to see if they had any additional questions. In advance to enrolling the end-user test, all participants completed a consent form.

Results

Eventually, 15 out of 19 (78.9%) spouses and 25 out of 38 (65.8%) other family members (i.e. 21 children, three siblings, and one parent) gave consent and participated in the end-user tests (Figure 2). Alzheimer's dementia (AD) (n = 27) was the most prevalent cause, followed by frontotemporal dementia (FTD) (n = 9), vascular dementia (n = 3), and Lewy Body dementia (n = 1). The majority of participants received coaching from a coach of the Alzheimer Center Limburg, while six spouses and six other family members received coaching from their casemanager or psychologist. The end-user test was completed by 11 of the 15 (73.3%) spouses, and 14 of the 25 (56.0%) other family members (Table 2). According to the non-completers their reason to withdraw from participation was not related to the intervention but to important life events such as the passing away of their relative with YOD (n = 1) or another relative (n = 1), moving abroad (n = 1), depression (n = 2), work or school related stress (n = 7), or unknown (n = 3). The non-completers did not significantly differ from the completers on demographic characteristics or outcome variables.

Table 2. Characteristics of included participants

Caregiver	N (%)	male / female	Age (min-max)
Completers			
Spouse	11 (45.8)	4 / 7	58.7 (50-70)
Child	9 (33.3)	3 / 6	33.5 (18-44)
Sibling	4 (12.5)	1 / 3	59.8 (56-64)
Parent	1 (4.2)	0 / 1	74.0
Non-completers (n = 15)			
Spouse	4 (26.6)	3 / 1	58.3 (53-65)
Child	11 (73.3)	4 / 7	30.9 (17-45)

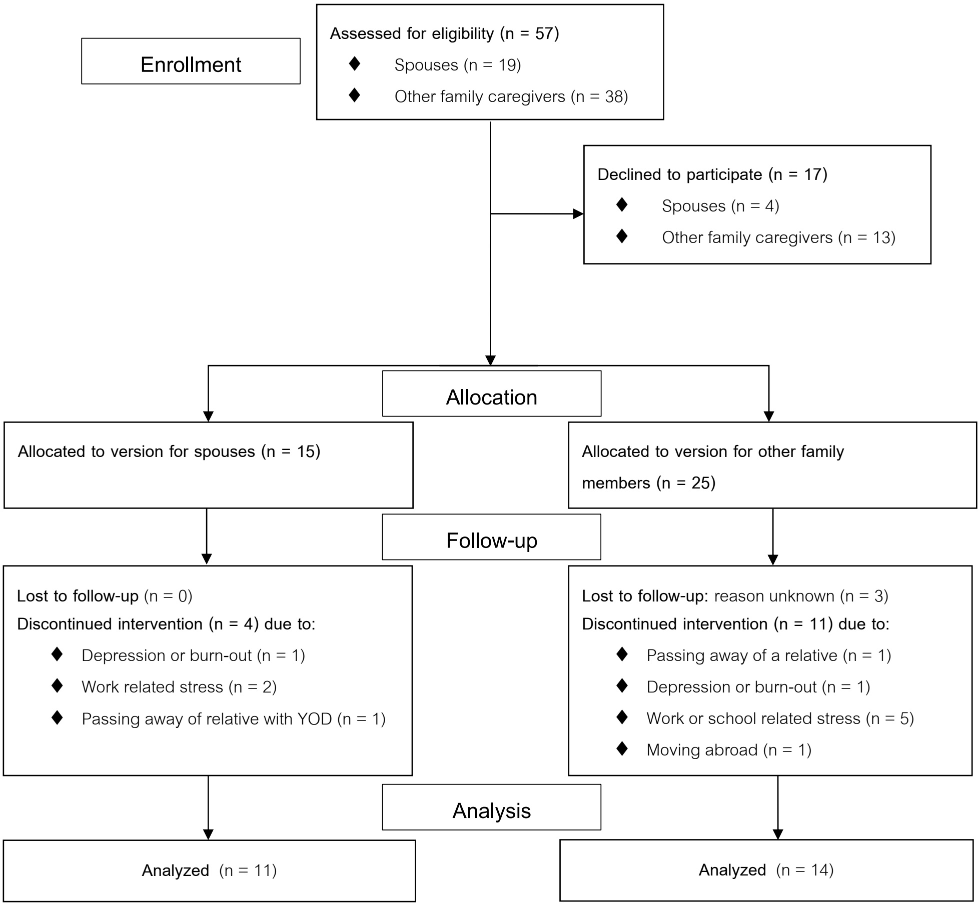

Figure 2. *CONSORT flow diagram on participation in the end-user test (Moher et al., 2001)*

The Program Participation Questionnaire

After completing the intervention, the average total score on the PPQ was 195.8 (SD = 15.8) for the 11 spouses. The total score for the 14 other family members was 211.3 (SD = 13.1). Both scores are higher than the cut-off score of 132, indicating a good overall usability, feasibility, and acceptability. On average, the spouses scored all items higher than 5 “slightly agree”, except for the item covering the use of the chat function (Figure 3). Percentiles revealed that 25% of the spouses scored the use of the chat and goal-setting lower than 5 “slightly agree”. The end-user test with other family members showed that all averaged scores and 25% percentiles were higher than 5 “slightly agree”, indicating good feasibility.

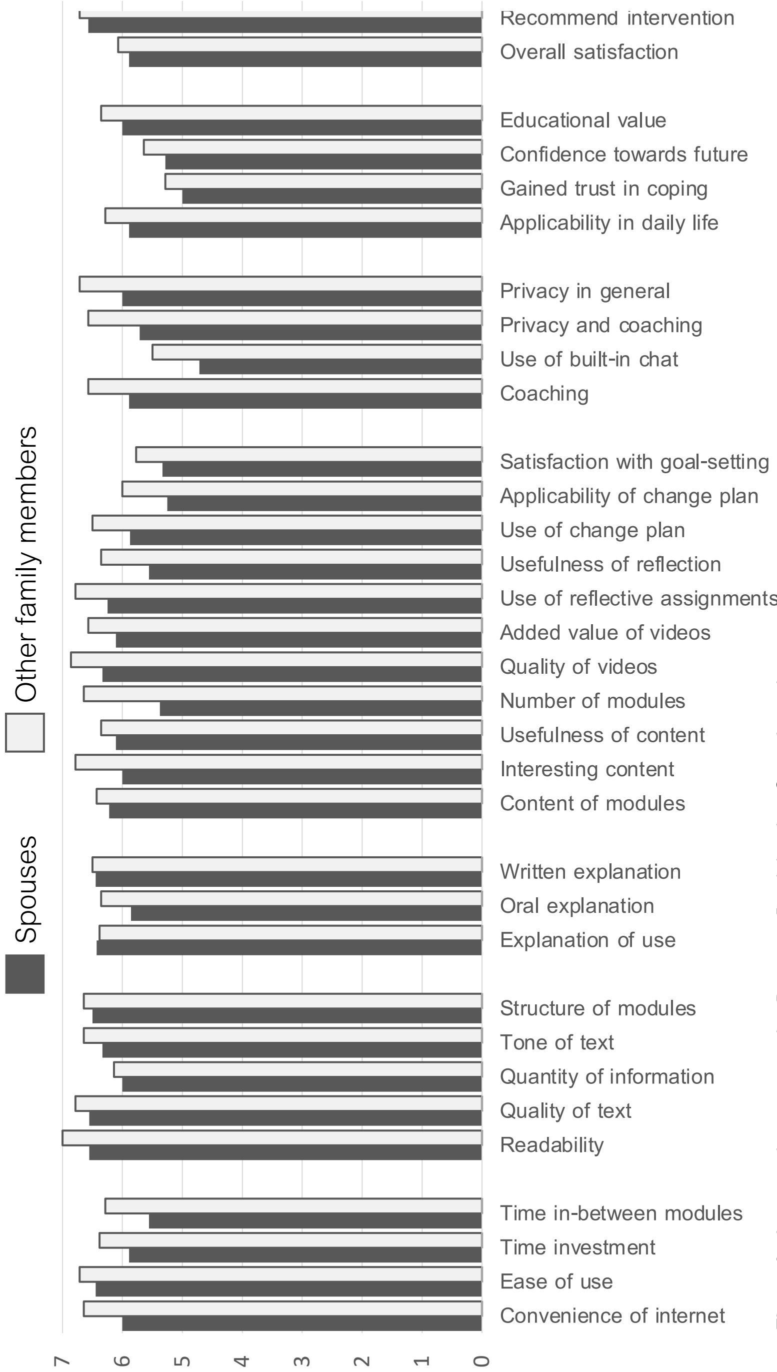

Figure 3. *Averaged scores on the Program Participation Questionnaire*

Qualitative findings

In line with the scores on the PPQ, a deductive qualitative content analysis of the fieldnotes showed that both spouses and other family members valued the intervention for its usability in daily life. They particularly appreciated the web-based format because it allowed them to follow the intervention in their own time at home. This facilitated the use of the intervention as most participants combined caregiving with work or studying, family life, and social activities. Spouses and other family members described the lay-out as self-explanatory, well structured, and user friendly. Particularly children (aged 18 or older), felt it was easier for them to express their feelings online, compared to traditional in-person meetings with healthcare professionals. Some children suggested to further improve the usability by making the intervention compatible with smartphone usage to allow them to use it in public transport while commuting to school or work.

“The internet component was nice because you can easily share your feelings. (…) I was able to share things outside the presence of my parents.”
– 17-year-old son of a person with frontotemporal dementia –

The quality of the tailored content was appreciated and considered highly acceptable by both spouses and other family members. They valued the recognizability of the video vignettes and narrative stories as this made them feel recognized. All participants felt that the written materials were well articulated, complete, and recognizable. However, some spouses and other family members of persons with FTD explained having difficulty recognizing themselves in the personal stories, which mainly focused on AD. They suggested to incorporate more specific content about FTD in the intervention to improve recognizability.

“The videos make you feel recognized and understood. They also tackle the taboo to talk about dementia.”
– 44-year-old daughter of a person with Alzheimer's dementia –

Spouses and other family members also valued the self-reflection assignments as it helped to translate the intervention to their personal situation. The reflection assignments helped to critically think about their needs and helped to prioritize activities. Most participants also perceived the step-by-step change plan as an important element because it structured their way of thinking by offering a stepwise approach to work towards a personal goal. However, some participants in both groups struggled with the step-by-step change plan experiencing difficulty formulating specific, measurable, and attainable goals. The feedback from the coach was experienced as an important source of inspiration while setting goals as it helped participants to gain new insights.

"The coach is the most important part of Partner in Balance. The personal contact is very nice, and the coach helps you to set goals by giving advice."

– 64-year-old brother of a person with Lewy Body dementia –

According to most spouses and other family members the coach also motivated them to apply the intervention in daily life. Both spouses and other family members expressed they felt better equipped as caregiver and more prepared for the future. They felt the Partner in Balance intervention helped them to prioritize and adjust their expectations regarding their relative with YOD. In turn, some felt more able to cope with behavioral symptoms in their relative with YOD.

"I am especially more self-aware, and Partner in Balance gave me the tools to tackle unwanted situations."

– 58-year-old spouse of a person with frontotemporal dementia –

Some participants expressed that the use of the chat function felt unnecessary, explaining why spouses provided on average a lower grade (Mean = 4.7, SD = 1.8) for the chat in the PPQ. An explanation stems from the fact that six of the 11 (54.5%) spouses received coaching from their own dementia casemanager or psychologist. In these cases, care as usual continued and most spouses also had in-person meetings with their coach.

Preliminary effects on role adaptation

Eleven spouses and 14 other family members completed the questionnaire before and after participating in the intervention. Although all average scores increased or decreased in line with our hypothesis (Figure 4), only the effect on self-efficacy regarding care-management was statistically significant in other family members, $t(12) = 3.37$, $p = .006$.

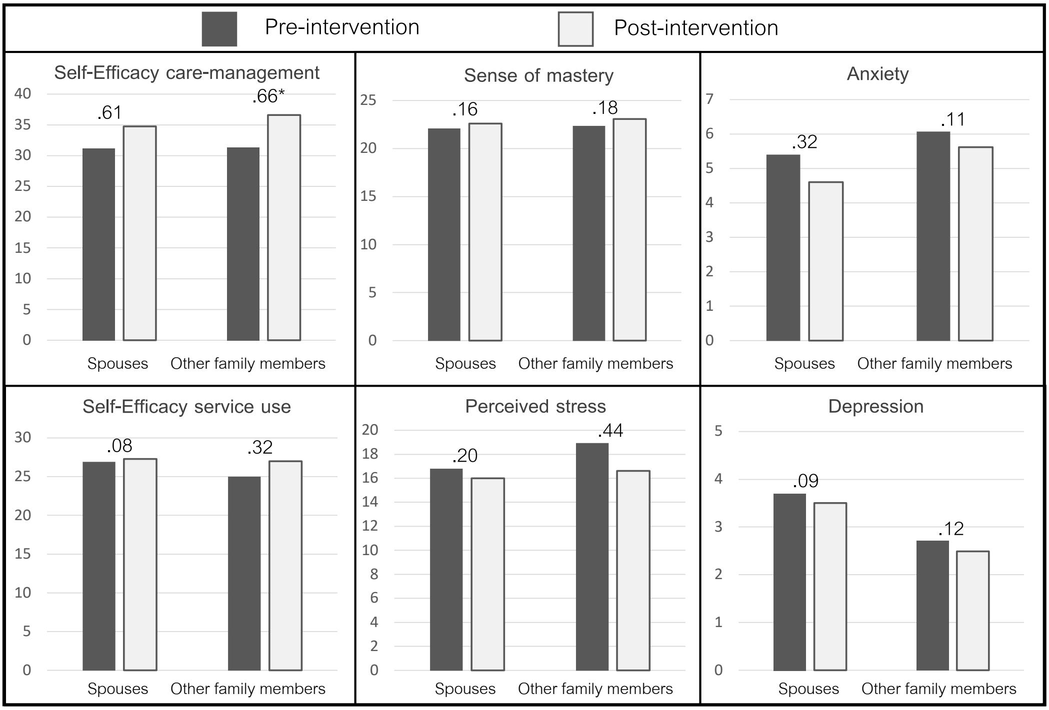

Figure 4. *Scores on the pre- and post-questionnaire*

Note. d = Cohen's d, * = statistically significant (p ≤ .05)

Discussion

The findings reveal that the tailored content for spouses and other family members of people with YOD was positively received. The participants valued the quality of the content provided and felt the intervention was easy to integrate in daily life. Similar to previous findings on generic modules included in Partner in Balance (Boots et al., 2018; Boots et al., 2016), the tailored content facilitated role adaption as participants felt better equipped for the caregiving role after the intervention. In both groups, caregivers expressed to feel more confident to cope with future challenges after participating in the intervention. In the end-user test with other family members, the effect on self-efficacy regarding care-management also increased statistically significant. This is in line with previously demonstrated effects of Partner in Balance in a randomized controlled trial (Boots et al., 2018). It is suggested that helping caregivers to become more resilient by increasing self-efficacy may also prevent long-term psychological distress and increase well-being (Bandura, 1997; Lazarus and Folkman, 1984).

Similar to previous studies, participants in our study felt the Partner in Balance intervention offered them a different perspective on their role as caregiver (Boots et al., 2018; Boots et al., 2016). Dementia is often defined in terms of loss and disability. As a result caregivers may perceive a lack of future perspective and experience feelings of hopelessness (Chapman et al., 2019; de Vugt and Dröes, 2017). Providing caregivers with a perspective that acknowledges their loss but also encourages them to think in terms of possibilities may empower them and may help caregivers regain a sense of balance in combining their caregiving role with other roles and responsibilities (de Vugt and Verhey, 2013). This may particularly apply to YOD caregivers given challenges they experience to balance caregiving with family life, employment, or studying (Cabote et al., 2015; van Vliet et al., 2010b). Therefore, it is important that interventions to support YOD caregivers can be easily integrated in daily life. Our findings reveal that Partner in Balance offered a flexible support tool that was easily integrated in daily life and allowed participants to follow the intervention at a convenient place and time. It is known that children of persons with YOD have specific support needs as they seem more reluctant to seek help from healthcare professionals (Millenaar et al., 2014). Additionally, children often feel that there is hardly any appropriate support available for them (Barca et al., 2014). Our findings indicate a web-based design is a particular good fit for children as some felt it was easier to share emotions online, compared to traditional face-to-face support. To further tailor Partner in Balance to the specific needs of children they suggested to improve the compatibility of the website with smartphone use. For example, to facilitate use in public transport.

Caregivers of persons with YOD often perceive mainstream dementia services and support as age-inappropriate because they primarily focused on elderly with dementia. In turn, YOD caregivers are known to delay the initiation of care and support services (Rabanal et al., 2018; Sikes and Hall, 2018). This is problematic because YOD can cause high levels of burden and distress in caregivers (Cabote et al., 2015; Millenaar et al., 2016b). Previously studies on Partner in Balance confirmed that YOD caregivers experienced difficulty recognizing their situation in the Partner in Balance intervention because most content focused on caregivers of elderly with dementia (Boots et al., 2017). Our findings show the newly incorporated content on YOD was positively received by YOD caregivers as they valued the high level of recognizable content provided. For example, in the video materials and narrative stories. However, our findings revealed that some caregivers of persons with FTD felt it was difficult to relate to the stories and content that mainly focused on YOD caused by AD. Caregivers of persons with FTD are known to experience a lack of understanding in caring for their relative, also from the low availability of recognizable support services (Bruinsma et al., 2020). Addressing their specific needs is important because FTD caregivers experience specific challenges that complicate adapting to the caregiving role. In particular, coping with profound changes in social behavior can impose high levels of burden and distress in FTD caregivers (Nunnemann et al., 2012).

Strengths and limitations

Our study aimed to evaluate the feasibility and usability of the proven effective Partner in Balance intervention in a new target group, and to identify directions for further improvement. Preliminary effects were also examined to evaluate if the intervention facilitated role adaptation, similar to previous findings (Boots et al., 2018; Boots et al., 2016). This end-user test with limited statistical power, shows a promising trend as absolute levels of self-efficacy and mastery increased, and levels of stress, anxiety and depression decreased but not statistically significant. Despite the small sample, self-efficacy in other family members did improve statistically significant. Increased self-efficacy may have a long-term beneficial effect on experienced stress, anxiety, and depression (Bandura, 1997; Lazarus and Folkman, 1984). An explanation for not finding significant improvement on stress, anxiety and depression may stem in the fact that our study used a short follow-up with a small sample. Therefore, studying long-term effects using a larger sample remains a direction for future research.

A strength of our study is that qualitative and quantitative data was collected to evaluate how caregivers perceived that the newly incorporated content. Additionally, a diverse sample was included that varied in age, gender, relationship to the person with YOD, and dementia subtype in their relative with YOD. A limitation is that experience with using web-based technology was not inventoried because this may influence how participants use Partner in Balance in daily life. In the interviews were no indicators found that participants perceived technological barriers. Given the young age of our sample (mean = 50.1, ranging from 17 to 74) most participants probably had experience with using web-based technologies in daily life, such as smartphones and computers.

During our study, usual care and support provided to informal caregivers by their casemanager or other healthcare professionals continued. To illustrate, some caregivers received coaching by their own healthcare professional. Previously, this showed to strengthen the bond between healthcare professionals and caregivers (Boots et al., 2017). Additionally, caregivers were allowed to receive other support such as peer-support. This may have biased our results, but it also resembles how Partner in Balance is used in the reality of daily practice.

Future directions and conclusion

In line with the results of the evaluation study of Partner in Balance for caregivers of people with late-onset dementia (Boots et al., 2018; Boots et al., 2016), the tailored intervention for YOD caregivers showed to be a good fit. Similar to earlier studies, the qualitative and quantitative measures indicate Partner in Balance has the potential to facilitate adaptation to the caregiving role. Our findings confirm that goal-setting is an important intervention element because it helped caregivers to translate the intervention to daily life. However, some caregivers experienced difficulty formulating specific, measurable, and attainable goals during our study. Therefore, incorporating strategies in the intervention that further facilitate goal-setting may support caregivers with applying the intervention in daily life. As the coach proved an important source of inspiration, it may be helpful to embed an introduction on goal-setting at the start of the intervention to help caregivers with making goals more specific. Additionally, goal-setting may be facilitated by asking caregivers when and where they want to attain certain goal-directed behavior in the step-by-step change plan (Vohs and Baumeister, 2016). Incorporating specific content for caregivers of FTD patients would improve the fit with this specific subgroup of YOD, and fulfil their need for tailored interventions (Bruinsma et al., 2020).

Furthermore, an implementation strategy is required to sustain the Partner in Balance intervention because less than 3% of the dementia care and support interventions are implemented in daily practice (Gitlin et al., 2015). Therefore, the development of a business-model including a license agreement is underway to ensure that healthcare professionals can structurally work with the intervention (Christie et al., 2020). To facilitate the development of the business-model, a cost-effectiveness study seems a direction for the future to obtain insight in long-term benefits of the Partner in Balance intervention such as prevention of psychological problems in caregivers or delayed institutionalization of persons with dementia. It would be interesting to evaluate effects on long-term stress, anxiety, and depression. Additionally, it would be interesting to investigate how the effects of Partner in Balance could be enhanced on the long-term, and how the effects relate to other psychoeducational interventions or psychosocial support, such as peer-support or case management.

To persuade healthcare organizations to implement the intervention, we aimed to use end-user tests as a steppingstone to facilitate implementation from the start (Christie et al., 2019). In a subsequent phase, healthcare professionals are asked to see if they are willing to sustain working with the Partner in Balance intervention. As organizational sponsorship seems to be a facilitator of implementation (Christie et al., 2018), we aim to involve healthcare organizations affiliated with the Dutch Young-onset Dementia Knowledge Center in the future implementation process.

Our findings demonstrate that tailoring support can help to make support services more appealing to caregivers of persons with YOD. Throughout the caregiving trajectory, YOD caregivers encounter a wide variety of supportive services, that often do not match their specific situation (Rabanal et al., 2018; Sikes and Hall, 2018). Partner in Balance is an addition to existing support services, by combining personal coaching and a web-based approach. However, this may not appeal to all caregivers of persons with YOD. Therefore, the development of other tailored supportive services remains an important direction for the future.

Funding

This work was supported by an undisclosed funder of Alzheimer Research Fund Limburg; and partly performed during the UNited for Implementation of a healthCare standard In The Young (UNICITY)-project funded by *ZonMw*, [grant number: 60-63900-98-001].

Declaration of competing interest

None.

Acknowledgements

We would like to acknowledge the important role of the Dutch Young-onset Dementia Knowledge Center [*Kenniscentrum Dementie op Jonge Leeftijd*] and the Dutch FTD peer-support organization [*FTD lotgenoten*] during the recruitment phase. We also like to express appreciation for all healthcare professionals and caregivers who helped with revising the materials of the Partner and Balance intervention and participated in the end-user tests. Additionally, we would like to express appreciation for Esther Gerritzen and Anne Marieke Doornweerd for their assistance during the data-collection.

References

Aggarwal, R., and Ranganathan, P. (2019). Study designs: part 4 – interventional studies. *Perspect Clin Res, 10*(3), 137–139. doi:10.4103/picr.PICR_91_19

Bakker, C., de Vugt, M. E., van Vliet, D., Verhey, F. R., Pijnenburg, Y. A., Vernooij-Dassen, M. J ., and Koopmans, R. T. (2013). The use of formal and informal care in early onset dementia: results from the NeedYD study. *Am J Geriatr Psychiatr, 21*(1), 37–45. doi:10.1016/j.jagp.2012.10.004

Bandura, A. (1997). Self-efficacy: The Exercise of Control. W. H. Freeman and Company, New York.

Barca, M. L., Thorsen, K., Engedal, K., Haugen, P. K., and Johannessen, A. (2014). Nobody asked me how I felt: experiences of adult children of persons with young-onsetdementia. *Int Psychogeriatr, 2*(12), 1935–1944. doi:10.1017/S1041610213002639

Boots, L. M., de Vugt, M. E., Withagen, H. E., Kempen, G. I., and Verhey, F. R. (2016). Development and initial evaluation of the web-based self-management program "partner in balance" for family caregivers of people with early stage dementia: an exploratory mixed-methods study. *JMIR Res Protoc, 5*(1), e33 doi:10.2196/resprot.5142

Boots, L. M., de Vugt, M. E., Smeets, C. M., Kempen, G. I., and Verhey, F. R. (2017). Implementation of the blended care self-management program for caregivers of people with early-stage dementia (partner in balance): process evaluation of a randomized controlled trial. *J Med Internet Res, 19*(12), e423 doi:10.2196/jmir.7666

Boots, L. M., de Vugt, M. E., Kempen, G. I., and Verhey, F. R. (2018). Effectiveness of a blended care self-management program for caregivers of people with early-stage dementia (partner in balance): randomized controlled trial. *J Med Internet Res, 20*(7), e10017 doi:10.2196/10017

Bruinsma, J., Peetoom, K., Bakker, C., Boots, L., Verhey, F., and de Vugt, M. (2020). "They simply do not understand": a focus group study exploring the lived experiences of family caregivers of people with frontotemporal dementia. *Aging Ment Health.* doi:10.1080/13607863.2020.1857697

Cabote, C. J., Bramble, M., and McCann, D. (2015). Family caregivers' experiences of caring for a relative with younger onset dementia: a qualitative systematic review. *J Fam Nurs, 21*(3), 443–468. doi:10.1177/1074840715573870

Caceres, B. A., Frank, M. O., Jun, J., Martelly, M. T., Sadarangani, T., and de Sales, P. C. (2016). Family caregivers of patients with frontotemporal dementia: an integrative review. *Int J Nur Stud, 55*, 71–84. doi:10.1016/j.ijnurstu.2015.10.016

Campbell, S., Azuara-Blanco, A., Campbell, M., Francis, J., Greene, A., Ramsay, C., and Burr, J. (2012). Developing the specifications of an Open Angle Glaucoma screening intervention in the United Kingdom: a Delphi approach. *BMC Health Serv Res, 12*(447), 1–10. doi:10.1186/1472-6963-12-447

Cations, M., Withall, A., Horsfall, R., Denham, N., White, F., Trollor, J., Loy, C., Bordaty, H., Sachdev, P., Gonski, P., Demirkol, A., Cumming, R. G., and Draper, B. (2017). Why aren't people with young onset dementia and their supporters using formal services? Results from the INSPIRED study. *PLoS One 12*(7), e0180935. doi:10.1371/journal.pone.0180935

Chapman, M., Philip, J., and Komesaroff, P. (2019). Towards an Ecology of Dementia: A Manifesto. *J Bioeth Inq, 16*(2), 209-216. doi:10.1007/s11673-019-09910-8

Christie, H. L., Bartels, S. L., Boots, L. M. M., Tange, H. J., Verhey, F. R. J., and de Vugt, M. E. (2018). A systematic review on the implementation of eHealth interventions for informal caregivers of people with dementia. *Internet Interv, 13*, 51–59. doi:10.1016/j.invent.2018.07.002

Christie, H. L., Martin, J. L., Connor, J., Tange, H. J., Verhey, F. R. J., de Vugt, M. E., and Orrell, M. (2019). eHealth interventions to support caregivers of people with dementia may be proven effective, but are they implementation-ready? *Internet Interv, 18*, 100260 doi:10.1016/j.invent.2019.100260

Christie, H. L., Boots, L. M. M., Peetoom, K., Tange, H. J., Verhey, F. R. J., and de Vugt, M. E. (2020). Developing a plan for the sustainable implementation of an electronic health intervention (partner in balance) to support caregivers of people with dementia: case study. *JMIR Aging 3*(1), e18624. doi:10.2196/18624

Christie, H. L., Boots, L. M. M., Tange, H. J., Verhey, F. R, J., and de Vugt, M. E. (2021). Implementations of edidence-based ehealth interventions for caregivers of people with dementia in municipality contexts (Myinlife and Partner in Balance): evaluation study. *JMIR Aging 4*(1), e21629. doi:10.2196/21629

Cohen, S. (1988). Perceived stress in a probability sample of the United States. In: Spacapan, In S., Oskamp, S. (Eds.), The Social Psychology of Health. Sage Publications, Newbury Park, pp. 31–67

Craig, P., Dieppe, P., Macintyre, S., Michie, S., Nazareth, I., and Petticrew, M. (2008). Developing an evaluating complex interventions: the new Medical Research Council Guidance. *BMJ 337*. doi:10.1136/bmj.a1655.

de Vugt, M. E., and Droes, R. M. (2017). Social health in dementia. Towards a positive dementia discourse. *Aging Ment Health, 21*(1), 1–3. doi:10.1080/13607863.2016.1262822

de Vugt, M. E., and Verhey, F. R. (2013). The impact of early dementia diagnosis and intervention on informal caregivers. *Prog Neurobiol, 110*, 54–62. doi:10.1016/j.pneurobio.2013.04.005

Diehl-Schmid, J., Schmidt, E. M., Nunnemann, S., Riedl, L., Kurz, A., Forstl, H., Wagenpfeil, S., and Cramer, B. (2013). Caregiver burden and needs in frontotemporal dementia. *J Geriatr Psychiatry Neurol, 26*(4), 221–229. doi:10.1177/0891988713498467

Elo, S., and Kyngäs, H. (2008). The qualitative content analysis process. *J Adv Nurs, 66*(11), 2460–2470. doi:10.1111/j.1365-2648.2007.04569.x

Fick, D. M., Steins, M. R., Mion, L. C., and Walls, J. L. (2011). Computerized decision support for delirium superimposed on dementia in older adults. *J Gerontol Nur, 37*(4), 39–47. doi10.3928/00989134-20100930-01

Fortinsky, R. H., Kercher, K., and Burant, C. J. (2002). Measurement and correlates of family caregiver self-efficacy for managing dementia. *Aging Ment Health, 6*(2), 153–160. doi:10.1080/13607860220126763

Gitlin, L. N., Marx, K., Stanley, I. H., and Hodgson, N. (2015). Translating evidence-based dementia caregiving interventions into practice: state-of-the-science and next steps. *Gerontologist 55*(2), 210–226. doi:10.1093/geront/gnu123

Harvey, R. J., Skelton-Robinson, M. S., and Rossor, M. N. (2003). The prevalence and causes of dementia in people under the age of 65 years. *J Neurol Neurosurg Psychiatry, 74*(9), 1206–1209. doi:10.1136/jnnp.74.9.1206

Hutchinson, K., Roberts, C., Kurrle, S., and Daly, M. (2016). The emotional well-being of young people having a parent with younger onset dementia. *Dementia 15*(4), 609–628. doi:10.1177/1471301214532111

Kaizik, C., Caga, J., Camino, J., O'Connor, C. M., McKinnon, C., Oyebode, J. R., Piguet, O., Hodges, J. R., and Mioshi, E. (2017). Factors underpinning caregiver burden in frontotemporal dementia differ in spouses and their children. *J Alzheimer's Dis, 56*(3), 1109-1117. doi:10.3233/JAD-160852

Kilty, C., Boland, P., Goodwin, J., and de Roiste, A. (2019). Caring for people with young onset dementia: an interpretative phenomenological analysis of family caregivers' experiences. *J Psychosoc Nurs Men Health Serv, 57*(11), 37–44. doi:10.3928/02793695-20190821-02

Kurz, A., Bakker, C., Bohm, M., Diehl-Schmid, J., Dubois, B., Ferreira, C., Gage, H., Graff, C., Hergueta, T., Jansen, S., Jones, B., Komar, A., de Mendonça, A., Metcalfe, A., Milecka, K., Millenaar, J., Orrung Wallin, A., Oyebode, J., Schneider-Schelte, H., Saxl, S., and de Vugt, M. (2016). RHAPSODY - internet-based support for caregivers of people with young onset dementia: program design and methods of a pilot study. *Int Psychogeriatr, 28*(12), 2091–2099. doi:10.1017/S1041610216001186

Lai, C. K. Y., Wong, L. F., Liu, K-H., Lui, W., Chan, M. F., and Yap, L. S. Y. (2012). Online and onsite training for family caregivers of people with dementia: results from a pilot study. *J Geriatr Psychiatry, 28*(1), 107–108. doi:10.1002/gps.3798

Lazarus, R., and Folkman, S. (1984). Stress, Appraisal, and Coping. Springer Publishing, New York.

Millenaar, J. K., van Vliet, D., Bakker, C., Vernooij-Dassen, M. J., Koopmans, R. T., Verhey, F. R., and de Vugt, M. E. (2014). The experiences and needs of children living with a parent with young onset dementia: results from the NeedYD study. *Int Psychogeriatr, 26*(12), 2001–2010. doi:10.1017/S1041610213001890

Millenaar, J. K., Bakker, C., Koopmans, R. T., Verhey, F. R., Kurz, A., and de Vugt, M. E. (2016a). The care needs and experiences with the use of services of people with young-onset dementia and their caregivers: a systematic review. *Int J Geriatr Psychiatry 31*(12), 1261–1276. doi:10.1002/gps.4502

Millenaar, J. K., de Vugt, M. E., Bakker, C., van Vliet, D., Pijnenburg, Y. A. L., Koopmans, R. T. C. M., and Verhey, F. R. J. (2016b). The impact of young onset dementia on informal caregivers compared with late onset dementia: results from the NeedYD study. *Am J Geriatr Psychiatr, 24*(6), 467–474. doi:10.1016/j.jagp.2015.07.005

Millenaar, J. K., Bakker, C., van Vliet, D., Koopmans, R., Kurz, A., Verhey, F. R. J., and de Vugt, M. E. (2018). Exploring perspectives of young onset dementia caregivers with high versus low unmet needs. *Int J Geriatr Psychiatry, 33*(2), 340–347. doi:10.1002/gps.4749

Moher, D., Schulz, K. F., and Altman, D. G. (2001). The CONSORT statement: revised recommendations for improving the quality of reports of parallel-group randomised trials. *BMC Med Res Methodol, 1*(2) doi:10.1186/1471-2288-1-2

Morgan, D. L. (2014). Pragmatism as a paradigm for social research. *Qual Inq, 20*(8), 1045–1053. doi:10.1177/1077800413513733

Nichols, K. R., Fam, D., Cook, C., Pearce, M., Elliot, G., Baago, S., Rockwood, K., and Chow, T. W. (2013). When dementia is in the house: needs assessment survey for young caregivers. *Can J Neurol Sci, 40*(1), 21–28. doi:10.1017/s0317167100012907

Nunnemann, S., Kurz, A., Leucht, S., and Diehl-Schmid, J. (2012). Caregivers of patients with frontotemporal lobar degeneration: a review of burden, problems, needs, and interventions. *Int Psychogeriatr, 24*(9), 1368–1386 doi:10.1017/S104161021200035X

Pearlin, L. I., and Schooler, C. (1978). The structure of coping. *J Health Soc Behav, 19*(1), 2–21. doi:10.2307/2136319

Rabanal, L. I., Chatwin, J., Walker, A., O'Sullivan, M., and Williamson, T. (2018). Understanding the needs and experiences of people with young onset dementia: a qualitative study. *BMJ Open 8*(10), e021166. doi:10.1136/bmjopen-2017-021166

Richardson, A., Pedley, G., Pelone, F., Akhtar, F., Chang, J., Muleya, W., and Greenwood, N. (2016). Psychosocial interventions for people with young onset dementia and their carers: a systematic review. *Int Psychogeriatr, 28*(9), 1441–1454. doi:10.1017/S1041610216000132

Roach, P., Keady, J., Bee, P., and Williams, S. (2013). 'We can't keep going on like this': identifying family storylines in young onset dementia. *Ageing Soc, 34*(8), 1397–1426. doi:10.1017/s0144686x13000202

Rossor, M. N., Fox, N.C., Mummery, C. J., Schott, J. M., and Warren, J. D. (2010). The diagnosis of young-onset dementia. *Lancet Neurol, 9*(8), 793–806. doi:10.1016/s1474-4422(10)70159-9

Sikes, P., Hall, M. (2018). The impact of parental young onset dementia on children and young people's educational careers. *Br Educ Res J, 44*(4), 593–607. doi:10.1002/berj.3448

van Vliet, D., Bakker, C., Koopmans, R. T., Vernooij-Dassen, M. J., Verhey, F. R., and de Vugt, M. E. (2010a). Research protocol of the NeedYD-study (Needs in Young onset Dementia): a prospective cohort study on the needs and course of early onset dementia. *BMC Geriatr, 10*(13) doi:10.1186/1471-2318-10-13

van Vliet, D., de Vugt, M. E., Bakker, C., Koopmans, R. T., and Verhey, F. R. (2010b). Impact of early onset dementia on caregivers: a review. *Int J Geriatr Psychiatry, 25*(11), 1091–1100. doi:10.1002/gps.2439

van Vliet, D., de Vugt, M. E., Bakker, C., Pijnenburg, Y. A., Vernooij-Dassen, M. J., Koopmans, R. T., and Verhey, F. R. (2013). Time to diagnosis in young-onset dementia as compared with late-onset dementia. *Psychol Med, 43*(2), 423–432. doi:10.1017/S0033291712001122

Vohs, K. D., and Baumeister, R. F. (2016). Handbook of Self-regulation, Second Edition: Research, Theory, and Applications, Second edition. Guilford Press, New York.

Zigmond, A. S., and Snaith, R. P. (1983). The Hospital Anxiety and Depression Scale. *Acta Psychiatr Scand, 67*(6), 361–370. doi:10.1111/j.1600-0447.1983.tb09716.x

Chapter 7

Tailoring the web-based 'Partner in Balance' intervention to support spouses of persons with frontotemporal dementia

Bruinsma, J., Peetoom, K., Boots, L., Daemen, M., Verhey, F., Bakker, C., and de Vugt, M.

Internet Interventions, 2021

Abstract

Frontotemporal dementia (FTD) typically starts before the age of 65 years, and symptoms differ from other dementias (e.g. Alzheimer's dementia). Spouses are often caregiver and experience difficulty coping with the profound changes in personality and behavior accompanying FTD. Most interventions available to these spouses do not match their need for tailored and flexible psychosocial support. Therefore, tailored content for spouses of persons with FTD was recently incorporated in the proven effective and web-based Partner in Balance intervention.

Methods: This feasibility study prospectively evaluated the tailored Partner in Balance content for spouses of persons with FTD. Spouses followed the 8-week intervention, and qualitative and quantitative measures were used to evaluate expectations and barriers prior to participation and aspects of usability, feasibility, and acceptability of content. Additionally, effects were explored regarding caregiver self-efficacy, sense of mastery, stress, depression, and anxiety.

Results: Twenty-seven spouses caring for a spouse with FTD at home started the intervention. Eventually, 20 completed the intervention (74.1%). Partner in Balance matched the expectations of participating spouses and helped them to find a better balance between caregiving and personal life, acquire more peace of mind, and facilitated coping with behavioral and communication difficulties. Before participation, time restraints were identified as a potential barrier, but afterwards spouses positively evaluated the flexibility of the web-based approach that allowed them to participate at a convenient time and place. They valued the recognizability of the videos and narrative stories on FTD. Post-intervention, spouses qualitatively felt more confident, more at ease, and strengthened as a caregiver. Quantitatively, levels of self-efficacy, anxiety, and depression significantly improved.

Conclusion: Partner in Balance is a usable, feasible, and acceptable intervention for spouses caring for a spouse with FTD at home. Healthcare organizations could consider adopting Partner in Balance in their daily practice to offer flexible and tailored support to spouses.

Keywords: Partner-in-Balance; Frontotemporal dementia; Web-based; Support; Caregivers

Introduction

In 70-80% of the persons with frontotemporal dementia (FTD) the symptoms start before the age of 65 years (Knopman and Roberts, 2011; Rabinovici and Miller, 2010). Given the young age of onset, spouses often perform the role of primary caregiver (Bakker et al., 2013). This is challenging for most spouses because they are likely to be employed (Caceres et al., 2016; Nunnemann et al., 2012), and children may still live at home (Kaizik et al., 2017). Another challenging factor is that persons with FTD are often in good physical condition and the symptoms differ from those of other dementias. Compared to persons with Alzheimer's dementia, the short-term memory often remains relatively intact in persons with FTD. Symptoms more often involve deficits in social cognition, disinhibition, and passive behavior (Shnall, 2009; Lindau et al., 2000). The presentation of FTD is diverse and generally three variants are distinguished. The behavioral variant of FTD is the most prevalent and characterized by personality and behavioral changes such as apathy, disinhibition, compulsive behavior, and a lack of social insight (Onyike and Diehl-Schmid, 2013; Bang et al., 2015). Non-Fluent progressive Aphasia is a FTD variant characterized by difficulty with language production and word comprehension. The semantic variant of FTD is accompanied by aphasia and loss of anomia of words, persons, places, and objects (Bang et al., 2015). In all variants, behavioral and emotional symptoms can occur such as apathy, repetitive behavior, and depression (Bang et al., 2015). Coping with these symptoms is challenging for spouses and linked to burden and distress (Caceres et al., 2016; Kaizik et al., 2017). The young age and symptomatic overlap with psychiatric disorders is known to complicate and delay the diagnosis of FTD (van Vliet et al., 2013; Ducharme et al., 2020). Establishing an FTD diagnosis may last up to 6.1 years, compared to 4.4 years in young-onset dementia in general (van Vliet et al., 2013; Draper et al., 2016). The delay in diagnosis is problematic as it impedes the ability of spouses to adapt to the caregiving role because a diagnosis helps with understanding the changes in their spouse with FTD (Bruinsma et al., 2020).

In the phase after obtaining the diagnosis, spouses of persons with FTD often feel socially and professionally unsupported (Bruinsma et al., 2020; Rosness et al., 2008). For example, they experience that family and friends trivialize the severity of symptoms. Additionally, they often feel that healthcare professionals struggle with providing advice on coping with symptoms of FTD (Bruinsma et al., 2020). The majority of available support for caregivers is designed with the elderly and in particular Alzheimer's dementia in mind, resulting in a mismatch between support and the needs of spouses of persons with FTD (Bruinsma et al., 2021a; Nunnemann et al., 2012). Therefore, spouses postpone the initiation of professional care and support services (Bruinsma et al., 2020). This is problematic because support can facilitate adaptation to the caregiving role. For example, by increasing levels of confidence, and decreasing levels of burden and distress in caregivers (Gossink et al., 2018; Boots et al., 2018). Tailored, accessible, and flexible support services for spouses of persons with FTD may facilitate timely access (Cations et al., 2017). However, due to the low prevalence of FTD it is difficult for spouses to find access to appropriate support close to home. Therefore, web-based support may allow for flexibility and accessibility, also to caregivers living in rural areas (Diehl-Schmid et al., 2013). Previously, web-based approaches already showed potential in improving caregiver well-being (Boots et al., 2014). Therefore, this study used the proven effective web-based Partner in Balance intervention as a starting point. Recent studies showed that Partner in Balance helped caregivers to prepare for the role of informal caregiver. Post-intervention, caregivers improved in self-efficacy, sense of mastery, and quality of life (Boots et al., 2018, Duits et al., 2020, Boots et al., 2016, Bruinsma et al., 2021). The web-based design also showed a good fit for family caregivers of persons with young-onset dementia (Boots et al., 2017). Although these results are promising, spouses of persons with FTD had difficulty recognizing their personal situation in the generic intervention content that primarily addressed spouses of persons with Alzheimer's dementia (Bruinsma et al., 2021). Therefore, tailored content was recently incorporated in Partner in Balance by developing tailored videos, personal stories, and psychoeducation for spouses of persons with FTD. This feasibility study evaluates how spouses perceive this tailored intervention content on FTD.

Methods

This pre-post design feasibility study evaluated tailored content for spouses of persons with FTD that was incorporated in the Partner in Balance intervention. Spouses caring for a spouse with FTD at home participated in the intervention, and qualitative and quantitative measures evaluated expectations and barriers before participation, and perceptions regarding usability, feasibility, and acceptability of the tailored content. Additionally, explorative effects were examined. The CONSORT-EHEALTH was used as a guideline for reporting (Eysenbach et al., 2011).

The Partner in Balance intervention

Partner in Balance is a web-based self-management intervention that aims to facilitate role adaptation by supporting caregivers with finding a balance between caregiving and daily life (Boots et al., 2017; Boots et al., 2018). During the intervention, caregivers receive online coaching from a trained healthcare professional and follow subsequently four self-chosen modules online (Table 1). Caregivers and healthcare professionals access the Partner in Balance intervention via a website and spent around 6 hours on following the intervention in a period of eight weeks. In Partner in Balance there are tailored modules for caregivers of elderly with dementia, young-onset dementia, and Parkinson's disease (Boots et al., 2016; Bruinsma et al., 2021; Duits et al., 2020). Each module includes (1) a video portraying the experiences of other caregivers, (2) narrative stories, psychoeducation, and practical advice, (3) a self-reflection assignment, and (4) a step-by-step change plan to help caregivers to set a personal goal (Boots et al., 2016; Bruinsma et al., 2021).

Table 1. *Available modules in Partner in Balance*

Collection	Generic	Young-onset dementia		Parkinson's disease	FTD
Study	*Boots et al. (2016)*	*Bruinsma et al. (2021)*		*Duits et al. (2020)*	*This study*
	Spouses	Spouses	Others	Spouses	Spouses
<u>Modules</u>					
Combining care with work		x	x		x
Impact on family life		x	x		x
Sexuality and intimacy		x			x
Worries about heredity			x		x
Coping with stress*	x			x	
Acceptance	x	x	x	x	x
Balance in activities	x	x	x	x	x
Changes accompanying dementia	x	x	x	x	x
Communication	x	x	x	x	x
Focusing on the positive	x	x	x	x	x
Insecurities and rumination	x	x	x	x	x
Self-understanding	x	x	x	x	x
Social relationships and support	x	x	x	x	x

**The module on coping with stress was merged with focusing on the positive and insecurities and rumination.*

Building upon previous development (Boots et al., 2018; Boots et al., 2017; Boots et al., 2016; Bruinsma et al., 2021), tailored content on FTD was recently incorporated in Partner in Balance. Figure 1 presents an overview of the iterative development process and incorporation of tailored content on FTD, using the Medical Research Council (MRC) framework (Craig et al., 2008). This feasibility study concerns the final step in the developmental process and evaluated the expectations and perceptions of spouses regarding tailored content on FTD. Tailored content was developed using data from focus group discussions (Bruinsma et al., 2020), and in close collaboration with an advisory committee comprising experts, healthcare professionals, and spouses of persons with FTD. To illustrate, the tailored content consisted of videos and personal stories reflecting the perspective of spouses caring for a person with FTD at home. Additionally, the advisory committee reviewed and supplemented psychoeducation and practical advice. For example, about heredity risks and coping with linguistic or behavioral symptoms of FTD.

Recruitment

Caregiving spouses were eligible for participation in the feasibility study when their spouse had FTD and lived at home. They were recruited via newsletters and social media of the Dutch FTD peer-support organization [*FTD lotgenoten*]. Additionally, dementia casemanagers and psychologists experienced in supporting persons with FTD and their caregivers were recruited to provide coaching during the study. Spouses were asked if their casemanager or psychologist was willing to facilitate coaching. Casemanagers and psychologists were also recruited to coach a spouse from their caseload by spreading information via newsletters and bimonthly meetings with healthcare providers affiliated with the Dutch Young-onset Dementia Knowledge Center [*Kenniscentrum Dementie op Jonge Leeftijd*]. Four experienced coaches with a background in psychology from Alzheimer Center Limburg were also available to coach spouses during the study when needed. Prior to coaching, all casemanagers and psychologists unfamiliar with Partner in Balance received a training comprising an introduction session, eLearning, and a consultation meeting with experienced coaches. On a biweekly basis the casemanagers and psychologists were contacted by the research team to monitor progress and verify protocol adherence. This also allowed to pursue a low threshold for support (Christie et al., 2021). Care as usual continued throughout the study.

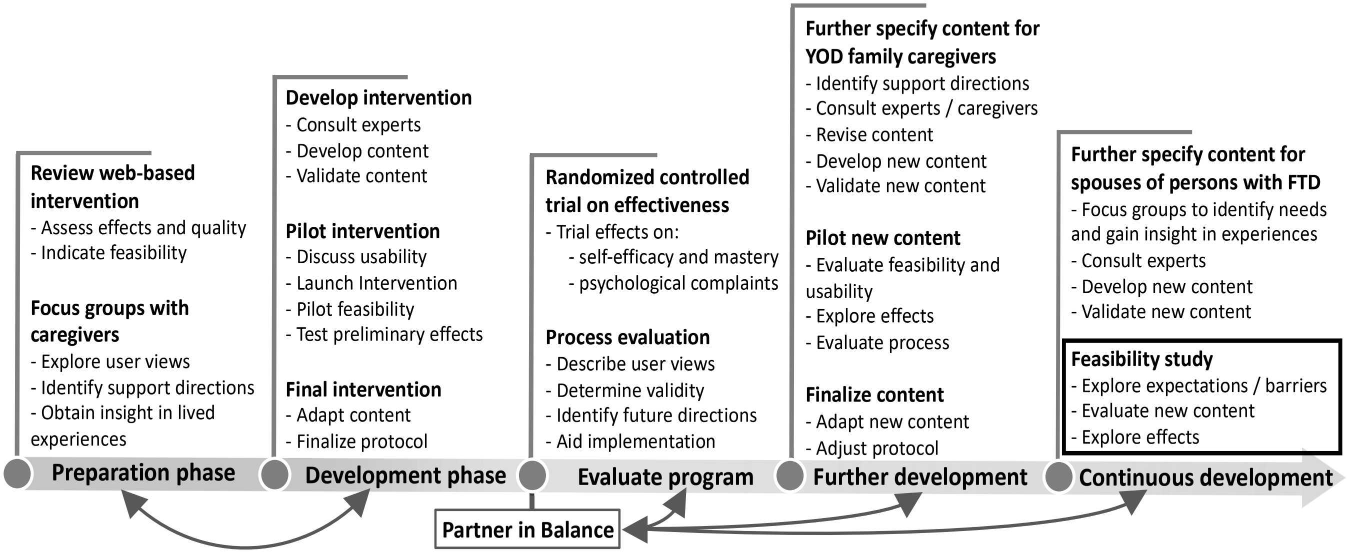

Figure 1. *The iterative development process of the Partner in Balance intervention*

Measurements

Qualitatively expectations and barriers perceived by spouses before participating in the intervention were explored. Then, quantitative and qualitative measures were used to explore perceptions regarding usability, feasibility, and acceptability of the tailored content on FTD. Additionally, it was explored if quantitative effects were in line with those of previous studies on Partner in Balance (Bruinsma et al., 2021; Duits et al., 2020; Boots et al., 2018; Boots et al., 2016).

Participant expectations and barriers before enrolling in the intervention

Pre-intervention, semi-structured interviews of 30 minutes were conducted to obtain insight in the motives of spouses to participate, their expectations, and potential barriers interfering with participation. To illustrate, questions involved “what persuaded you to participate in Partner in Balance?”, “what are your expectations regarding the intervention?”, and “what might interfere with participation?”. The study was conducted between April 2020 and May 2021, during the Covid-19 pandemic. Therefore, interviews were conducted via telephone and participants were asked about the impact of the pandemic on their role as caregiver, social life, and use of professional support.

Perceptions regarding usability, feasibility, and acceptability of the tailored content

Post-intervention, spouses were interviewed for 1 hour via telephone using the Program Participation Questionnaire (Boots et al., 2016; Bruinsma et al., 2021). This questionnaire was specifically developed to evaluate usability, feasibility, and acceptability aspects of Partner in Balance in the past (Boots et al., 2016). It contains 33 items scored from 1 “strongly disagree” to 7 “strongly agree”. Items covered (1) the use of the intervention in daily life, (2) feasibility of participation, (3) quality of the content provided, (4) experiences with coaching, and (5) perceptions on role adaptation and coping. Throughout the interview, participants were continuously encouraged to elaborate on their experiences. For example, by asking “how did you use the intervention is daily life?”, “what did you like or dislike about the narrative stories?”, and “how can we further improve the intervention materials?”. Additionally, participants were asked about the timing of the intervention and barriers encountered in using Partner in Balance.

Explorative effects

We substantiated our aim to evaluate feasibility by exploring if effects were in line with those of other versions of Partner in Balance (i.e. generic modules and content on young-onset dementia). Therefore, participants completed a pre-post questionnaire using identical scales as used in previous studies on feasibility and effectiveness (Boots et al., 2018; Boots et al., 2016; Bruinsma et al., 2021). The pre-post questionnaire explored self-efficacy regarding care-management (six items) and service use (four items) using the Self-Efficacy Scale (CSES). This scale has demonstrated good reliability and internal consistency (Fortinsky et al., 2002). Caregiver mastery was explored with seven items from the Pearlin Mastery Scale (PMS) (Pearlin and Schooler, 1978). A good validity and reliability of the PMS has been demonstrated in diverse populations (Edwards et al., 2000; Marshall and Lang, 1990; Walford-Kraemer and Light, 1984). Ten items measured the amount of stress experienced in the last week by using the Perceived Stress Scale (PSS) (Cohen, 1988). Previously, the PSS demonstrated good internal consistency and validity (Andreou et al., 2011). Anxiety (six items) and depression (seven items) were measured using the Hospital Anxiety and Depression Scale (HADS) (Zigmond and Snaith, 1983). Psychometric properties of the HADS indicate good reliability and validity (Spinhoven et al., 1997).

Analysis

An iterative process was used to analyze the qualitative and quantitative findings from a pragmatic theoretical stance (Morgan, 2014). Therefore, descriptive statistics (means, standard deviation, range) were calculated for items from the Program Participation Questionnaire. To interpret the scores, the interview transcripts were deductively coded by the first author, using Atlas.ti. Codes were summarized in a mind-map and discussed with the second author to derive categories from the data. Then, the findings were discussed with the other authors to substantiate the results. The research team included (neuro)psychologists, health scientists, and a neurologist. All researchers were experienced in conducting qualitative and quantitative research about caregivers of persons with FTD.

To explore if effects were in line with those of previously conducted studies (Bruinsma et al., 2021; Boots et al., 2018; Boots et al., 2016; Duits et al., 2020), the average scores on the pre-post questionnaire were compared for the CSES, PMS, PSS and HADS. This was done in SPSS using paired-sample t-testing to evaluate for statistical significance, using an alpha of 0.05 for two-sided tests (Boots et al., 2016; Bruinsma et al., 2021).

Ethical considerations

The study protocol was approved as a non-medical study by the Medical Ethics Committee of Maastricht University Medical Center, the Netherlands (METC2019-1286). Prior to participation, all spouses received information about the study, were phoned to see if they had questions, and completed the informed consent procedure.

Results

Between April 2020 and May 2021, 33 spouses contacted the research team about participating in Partner in Balance. Twenty-seven out of 33 spouses (81.8%) started the intervention (Figure 2). Two spouses were excluded prior to participation because their spouse was institutionalized or recently passed away. Four others decided not to participate for various reasons (e.g. currently too busy at work or experiencing psychological distress). Twenty out of 27 participating spouses received coaching from their own casemanager (n = 19) or psychologist (n = 1). The other seven spouses had a coach without a pre-existing therapeutic relationship from Alzheimer Center Limburg. The average age of the 27 participating spouses was 64.5 years, ranging from 52 to 81 years. Three of the 27 spouses were male. Eleven spouses were employed, of whom two in fulltime employment. On average, the diagnosis in the person with FTD was established around two years ago, ranging from 3 months to 8 years. Most of the caregiving spouses cared for a spouse with the behavioral variant of FTD (bvFTD; n = 15), followed by semantic dementia (SD; n = 6), and primary progressive aphasia (PPA; n = 4). Two caregiving spouses cared for a spouse with a combination of FTD and amyotrophic lateral sclerosis.

Eventually, 20 of the 27 participating spouses (74.1%) completed the intervention by completing all four self-chosen modules. Spouses who ceased participation attributed this to time constraints or experienced distress. For example, as the result of moving or stress at work. One spouse ceased participation on behalf of advice of her casemanager because she was suspected to have dementia herself.

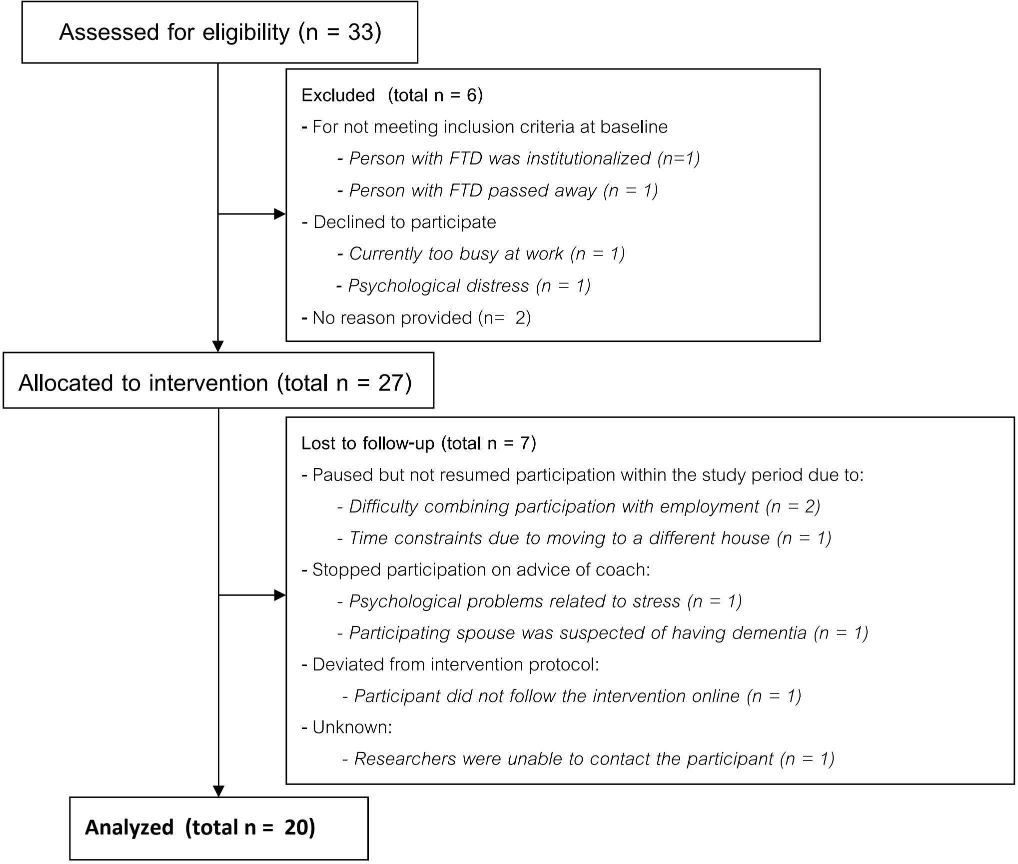

Figure 2. *Flow diagram on study participation*

Expectations and potential barriers

In the interview pre-intervention, most spouses explained they were persuaded to participate by reading about the newly incorporated Partner in Balance modules specifically on FTD. This elicited a sense of recognition regarding themes such as coping with behavioral symptoms, communication difficulties or worries about heredity. Most perceived Partner in Balance as an opportunity to learn more about themselves and acquire more peace of mind by learning more about setting boundaries, coping with challenging behaviors, improving communication skills, and achieving a balance between caregiving and personal life.

"I want to feel more at ease and achieve peace of mind. I hope this [Partner in Balance] helps me to grow as a caregiver. (…) I expect the coach to help me to think outside the box."

– Spouse of a 61-year-old person a bvFTD diagnosis for 4 months –

Prior to the intervention, most spouses felt confident towards completing Partner in Balance. Some perceived time constraints as a potential barrier, particularly when they combined caregiving with employment. As a result of the Covid-19 pandemic, some spouses worked from home while daycare facilities were closed. In turn, they expected that it would be additionally challenging to find time and privacy to follow Partner in Balance.

> *"As a result of the lockdown he is unable to go out and I have to work from home. He is very paranoid and constantly wants to know what I am doing, who I am calling. This is very exhausting."*
>
> **– Spouse of a 54-year-old person with a bvFTD diagnosis for 6 months –**

Usability, feasibility, and acceptability

Post-intervention, all spouses positively evaluated the web-based approach and valued the tailored content on FTD. The sum score on the Program Participation Questionnaire was 217.8, indicating good overall usability, feasibility, and acceptability because the score is higher than the cut-off score of 144. On average, all items were scored 6 or higher, on a scale from 1 "strongly disagree" to 7 "strongly agree" (Figure 3).

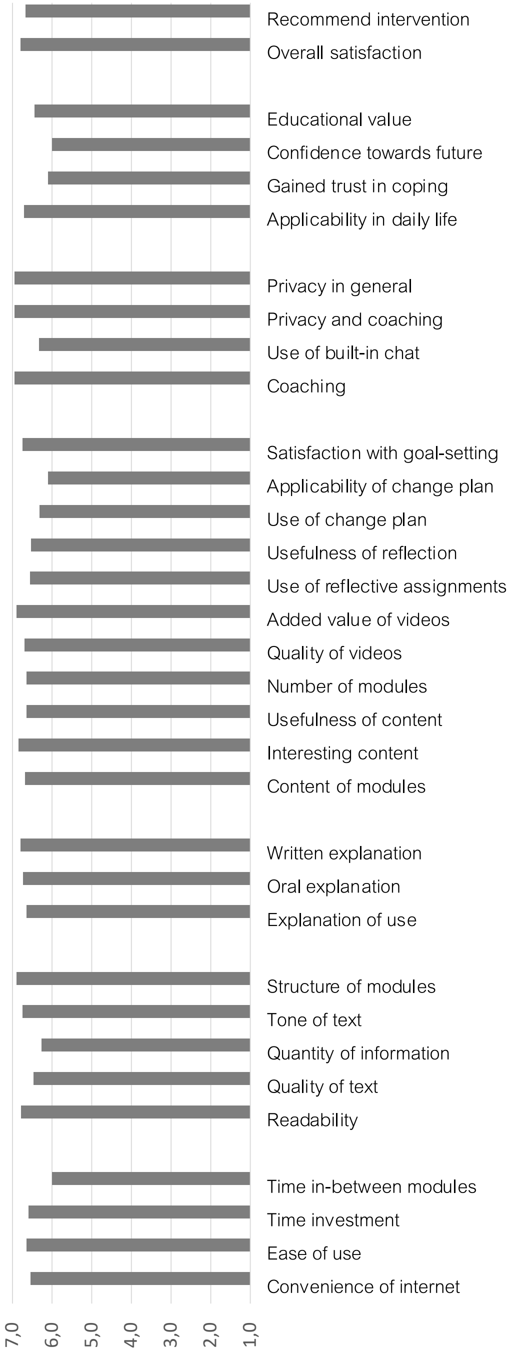

Figure 3. *Scoring on the Program Participation Questionnaire*

Spouses particularly valued the flexibility offered by the web-based design, enabling them to participate at a convenient time and place. According to spouses they had spent around one and a half hour per module, and it took them around 8 to 10 weeks to complete four modules. Most participants felt this was adequate, and stressed it was important to schedule time to engage in the intervention.

"The difficulty is you have to find the time and concentration for self-reflection and goal-setting. This can be difficult after a long day. You have to schedule time to embed it in your daily routine."

– Spouse of a 71-year-old person with a bvFTD diagnosis for one year –

All spouses perceived Partner in Balance as self-explanatory and well-structured. They valued the recognizability of the videos and textual information, and appreciated that real caregivers shared their experiences in the videos. Although they perceived each care situation as different, the videos and narrative stories gave them the feeling they were not alone. Some felt the videos could be further improved by adding experiences of a spouse caring for a person with SD, as the current videos only portray spouses of persons with the bvFTD or PPA.

"The videos give you the feeling you are not an exception. You see others openly talk about FTD. This encourages you to share your feelings and ask for support."

– Spouse of a 54-year-old person with a SD diagnosis for three years –

The participating spouses felt the self-reflection component helped to translate content of Partner in Balance to the personal context. Some felt this helped them to identify their own needs and helped them to prioritize. The goal-setting component in the change plan was often described as the most important element because it helped to set things in motion and apply Partner in Balance in daily life.

"Self-reflection reveals a part of yourself that is normally subconscious. This helps you to clarify things."

– Spouse of a 72-year-old person with a SD diagnosis for one year –

"Making a plan helped me to focus my attention and achieve my goals. (…) I sometimes I wonder why I didn't do this before."

– Spouse of a 65-year-old person with a bvFTD diagnosis for four years –

Spouses coached by a healthcare professional with a pre-existing therapeutic relationship felt their bond had strengthened. They attributed this to feeling more comfortable in addressing issues and felt their casemanager or psychologist had a better understanding of the caregiving situation. Some spouses preferred having a coach without a pre-existing relationship. For example, because they did not have a good relationship with their current healthcare professional or had a need for independent advice from a different angle. According to the participating spouses, the coach also made them feel heard and helped them to see their caregiver role from a different perspective by reevaluating expectations they had towards themselves. The feedback, questions, and advice from the coach also helped to set specific goals for the future.

"The added value is that I see things from a different angle. I feel more confident. More comfortable to ask for help when I am in need. (…) I feel more at ease and less frustrated about his behavior."

– Spouse of a 65-year-old person with a bvFTD diagnosis for six months –

Spouses felt the appropriate time to offer Partner in Balance would be in the phase directly after receiving the diagnosis because they had a high need for information in this phase. Some perceived Partner in Balance as a valuable tool throughout the caregiving trajectory because Partner in Balance boosted their confidence and helped them to come to terms with the role of informal caregiver. In retrospect, spouses reported that Partner in Balance matched their expectations as they felt better equipped as a caregiver following the intervention. They felt more at ease and strengthened in managing difficult behavior or communication difficulties. Although most felt more confident, they still experienced uncertainty towards the future due to the unpredictable nature of FTD.

"Eventually I will be unable to keep up with the progression of FTD. It may be a matter of weeks, months, or years. This makes the future unpredictable."

– Spouse of a 57-year-old person with a bvFTD diagnosis for four months –

"It [Partner in Balance] gives you the confidence you need because you come to the conclusion you are doing the best you can."

– Spouse of a 57-year-old person with a bvFTD diagnosis for four months –

Explorative effects

Explorative effects showed that post-intervention the levels of self-efficacy (CSES) regarding care-management (M = 37.3, SD = 8.86) were higher compared to pre-intervention (M = 35.0, SD = 8.96), t(19) = 2.33, p = .031. Additionally, post-intervention levels of depression (M = 4.8, SD = 2.28) were lower than pre-intervention (M = 5.9, SD = 2.21), t(19) = -2.926, p = .009. Post-intervention levels of anxiety (M = 4.8, SD = 2.67) were also lower than to pre-intervention (M = 6.5, SD = 3.15), t(19) = -3.157, p = .005 (Figure 4).

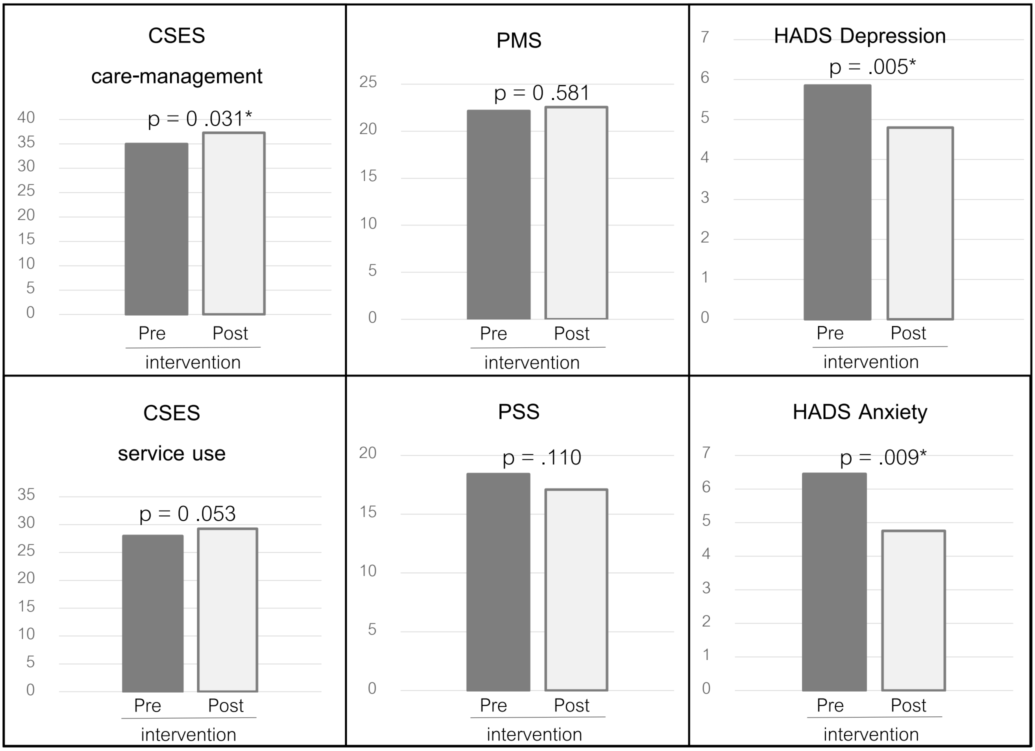

Figure 4. *Average scores on the pre-post questionnaire*

*Note. * = statistically significant*

Discussion

Key results

The tailored Partner in Balance intervention proved a good fit and matched to the expectations of spouses that cared for a spouse with FTD at home. Caregiving spouses positively evaluated the intervention in terms of usability, feasibility, and acceptability. For example, they valued the use of the intervention in daily life and appreciated the quality of the tailored content provided. Post-intervention, spouses felt more at ease and more confident towards the caregiving role. Explorative quantitative findings showed that spouses improved on self-efficacy, anxiety, and depression post-intervention.

Interpretating the findings in the light of previous research

In our study 81.8% of the potential participants started the intervention. This is substantial given that the averaged participation rate in caregiver research is around 27% (Brodaty et al., 2005). In a previous study on Partner in Balance participation rates ranged from 51.9% to 70.2% (Boots et al., 2018). Partly, this may be attributed to the high need of spouses for specific support on FTD (Bruinsma et al., 2020; Rosness et al., 2008). In our study, spouses explained the tailored content on FTD convinced them to participate. They expressed the specific program made them feel recognized. For example, because Partner in Balance gave them the feeling their situation was not an exception. Seeing others talk openly about FTD in the videos also motivated participating spouses to do the same. This is important because spouses of persons with FTD often experience a barrier to discuss FTD with family and friends (Bruinsma et al., 2020). Our findings indicate Partner in Balance helped spouses to mobilize their social network by encouraging them to openly talk about their feelings and actively ask for support. In turn, this may help to create more understanding in family and friends and reduce the risk of social isolation (Bruinsma et al., 2020). The low availability of other appropriate support may clarify why the tailored content on FTD was evaluated highly positive in our study. The overall score on the Program Participation Questionnaire was 217.8, higher than the cut-off of 144. This quantifies the high level of satisfaction spouses expressed during the interviews because in previous studies on Partner in Balance scores ranged from 195 to 211 (Boots et al., 2016; Bruinsma et al., 2021). The only direction for improvement identified, is the development of new videos portraying spouses caring for a spouse with SD. These new videos will be developed in the near future.

Like previous studies, participating spouses qualitatively confirmed that Partner in Balance made them feel more confident as a caregiver (Bruinsma et al., 2021; Boots et al., 2018; Boots et al., 2016; Duits et al., 2020). The questionnaire quantifies this by showing significant improvement in self-efficacy. Additionally, spouses qualitatively felt more at ease, and quantitatively reported lower levels of anxiety and depression post-intervention. Spouses of persons with FTD are at a high risk for burden, distress, and depression (Mioshi et al., 2013; Diehl-Schmid et al., 2013). This has been attributed to the complexity of coping with emotional and behavioral symptoms that accompany FTD (Kaizik et al., 2017). Spouses may perceive symptoms as uncontrollable and intentional (Polenick et al., 2018). Particularly, because persons with FTD often have a low awareness of disease. According to spouses in our study they felt more aware of the influence of their own behavior and felt more confident in managing challenging symptoms. This was attributed to self-reflection and goal-setting assignments embedded at the end of each module. To illustrate, participants felt goal-setting facilitated coping by helping them to set boundaries to prevent discussions, respond less agitated, and feel less guilt after an argument. An aim of Partner in Balance is to facilitate role adaptation by challenging caregivers to be resourceful, reevaluate their expectations, and think in terms of possibilities instead of limitations. This may have helped spouses in our study to become more resilient, explaining why spouses felt more confident and more at ease after the intervention. Resilience is known to mitigate feelings of anxiety and depression (Kobiske and Bekhet, 2018). Anxiety and depression in spouses of persons with FTD may also be attributed to anticipatory grief. The changes in personality accompanying FTD often give spouses the feeling they already lost their loved one, resulting in feelings of grief (Kaizik et al., 2017). Potentially, Partner in Balance may facilitate the grieving process by helping spouses to come to terms with their feelings and the role of informal caregiver. This may explain why they experienced less feelings of anxiety and depression.

Like previous studies, spouses perceived a stronger therapeutic relationship if their healthcare professionals provided coaching (Boots et al., 2017). More specifically, they felt recognized and heard by their coach. They particularly appreciated the constructive feedback, practical advice, and support from the coach. This is important because spouses of persons with FTD often perceive low levels of professional support, undermining their confidence in care and support services (Bruinsma et al., 2020; Johannessen et al., 2017). Currently, only little guidance is available for healthcare professionals aimed at providing psychosocial support to caregivers of persons with FTD (Shnall, 2009). To facilitate coaching in our study, healthcare professionals were recruited via interested spouses, and by spreading information directly to healthcare professionals. Throughout the study, 19 casemanagers were willing to coach. Compared to a previous feasibility study on generic modules on young-onset dementia, the number of casemanagers was higher (Bruinsma et al., 2021). This may reflect a need for tools that healthcare professionals can use in supporting FTD caregivers.

In the Netherlands, dementia casemanagers have a vital role in providing psychosocial support and they facilitate access to information, care, and support services. Therefore, casemanagers are crucial in promoting and offering tailored support to caregivers, also to spouses of persons with FTD. A previous process evaluation already demonstrated casemanagers perceive a generic version of Partner in Balance as relevant, usable, and feasible in daily practice (Boots et al., 2017). Future implementation should therefore focus on getting healthcare professionals acquainted with Partner and Balance and enable them to work with the intervention on a structural basis. It is important for sustainable implementation to adequately position the intervention within the current healthcare infrastructure. In the Netherlands, healthcare organizations receive a budget per patient and can partly allocate this to caregiver support. Therefore, a business model for sustainable implementation was developed enabling healthcare organizations to license Partner in Balance per caregiver (Christie et al., 2020). The non-profit license is used to cover expenses for website maintenance and technological support. Additionally, healthcare organizations purchase tailored trainings for their healthcare professionals who will serve as Partner in Balance coaches throughout the intervention. The training is adapted to the specific organizational context and includes an introduction session, eLearning, and a consultation meeting with experienced Partner in Balance coaches. For caregivers who are supported by healthcare organizations unable to allocate budget for a license, Alzheimer Center Limburg endeavors to cover the licensing costs through crowd-funding initiatives when possible. Additionally, there is a free web-based alternative without coaching provided by the RHAPSODY intervention for caregivers of persons with young-onset dementia (Kurz et al., 2016). Recently, a Dutch version of the skill-building RHAPSODY intervention has been launched on the website of the Dutch Alzheimer's Society [*Alzheimer Nederland*].

Strengths and limitations

In this study we were able to include a diverse sample in age and level of employment. Additionally, spouses of persons with different variants of FTD participated, namely bvFTD, SD, and PPA. Two spouses caring for a spouse with a combination between FTD and amyotrophic lateral sclerosis were also included. The varied sample allowed obtaining a good impression of how the tailored content meets the various needs of FTD caregivers. We substantiated our aim to explore if effects were in line with those of previous studies on Partner in Balance (Boots et al., 2018; Boots et al., 2016; Bruinsma et al., 2021; Duits et al., 2020). We realize that our findings are not sufficient to provide claims about generalized effects given the small sample with limited statistical power. We believe that combining qualitative and quantitative results and relate them to previous findings provides good support for the potentials of Partner in Balance intervention for this target group. Funding is acquired to further estimate long-term effect and health-economic impact of Partner in Balance by conducting a randomized controlled trial. This study will compare an intervention arm of providing Partner in Balance (i.e. both generic and tailored YOD and FTD content) to a control arm providing usual care.

A strength of our study is that most spouses received coaching from their own casemanager, and care as usual continued. Although this may have impacted our findings, this increases the external validity as it resembles how Partner in Balance is perceived in the context of daily practice. For future studies, it would be interesting to evaluate how effects of Partner in Balance may be enhanced by other care and support services, such as peer-support or daycare. According to participating spouses, these services were only limited available during our study due to the Covid-19 pandemic. This may have biased our findings because limited availability of support may influence experienced levels of self-efficacy, mastery, stress, anxiety, and depression. However, our findings also show that web-based support has potential in supporting caregivers during times of the Covid-19 pandemic (Duits et al., 2020).

Conclusions

Partner in Balance is a usable, feasible, and acceptable intervention for spouses of persons with FTD. Spouses qualitatively felt more confident and more at ease following Partner in Balance. Quantitatively they significantly improved on self-efficacy, and experienced lower levels of anxiety and depression. Partner in Balance showed to have substantial benefits for FTD caregivers. Therefore, healthcare organizations could consider adopting Partner in Balance in their daily practice to support spouses of persons with FTD at home, especially as a first step in the support process after diagnosis.

Funding

This study was funded by the Dutch brain foundation [*Hersenstichting*], grant number BG-FTD-SBB.

Declaration of competing interest

None.

Acknowledgements

We want to acknowledge the support of the Dutch Young-onset Dementia Knowledge Center [Kenniscentrum Dementie op Jonge Leeftijd], and Dutch FTD peer-support organization [*FTD lotgenoten*] during the recruitment phase. We also want to express appreciation for Roos Roberts for her help with making transcripts of the interview data.

References

Andreou, E. C., Alexopoulos, C., Lionis, L., Varvogli, C., and Gnardellis, G. P. (2011). Perceived stress scale: reliability and validity study in Greece. *Int J Environ Res Public Health, 8*(8), 3287-3298. doi:10.3390/ijerp8083287

Bakker, C., de Vugt, M. E., van Vliet, D., Verhey, F. R., Pijnenburg, Y. A., Vernooij-Dassen, M. J., and Koopmans, R. T. (2013). The use of formal and informal care in early onset dementia: results from the NeedYD study. *Am J Geriatr Psychiatry, 21*(1), 37-45. doi:10.1016/j.jagp.2012.10.004

Bang, J., Spina, S., and Miller, B. L. (2015). Frontotemporal dementia. *The Lancet, 386*(10004), 1672-1682. doi:10.1016/s0140-6736(15)00461-4

Boots, L. M., de Vugt, M. E., Kempen, G. I., and Verhey, F. R. (2018). Effectiveness of a Blended Care Self-Management Program for Caregivers of People With Early-Stage Dementia (Partner in Balance): Randomized Controlled Trial. *J Med Internet Res, 20*(7), e10017. doi:10.2196/10017

Boots, L. M., de Vugt, M. E., Smeets, C. M., Kempen, G. I., and Verhey, F. R. (2017). Implementation of the Blended Care Self-Management Program for Caregivers of People With Early-Stage Dementia (Partner in Balance): Process Evaluation of a Randomized Controlled Trial. *J Med Internet Res, 19*(12), e423. doi:10.2196/jmir.7666

Boots, L. M., de Vugt, M. E., van Knippenberg, R. J., Kempen, G. I., and Verhey, F. R. (2014). A systematic review of Internet-based supportive interventions for caregivers of patients with dementia. *Int J Geriatr Psychiatry, 29*(4), 331-344. doi:10.1002/gps.4016

Boots, L. M., de Vugt, M. E., Withagen, H. E., Kempen, G. I., and Verhey, F. R. (2016). Development and Initial Evaluation of the Web-Based Self-Management Program "Partner in Balance" for Family Caregivers of People With Early Stage Dementia: An Exploratory Mixed-Methods Study. *JMIR Res Protoc, 5*(1), e33. doi:10.2196/resprot.5142

Brodaty, H., Thomson, C., Thompson, C., and Fine, M. (2005). Why caregivers of people with dementia and memory loss don't use services. *Int J Geriatr Psychiatry, 20*(6), 537-546. doi:10.1002/gps.1322

Bruinsma, J., Peetoom, K., Bakker, C., Boots, L., Millenaar, J., Verhey, F., and de Vugt, M. (2021). Tailoring and evaluating the web-based ‘Partner in Balance’ intervention for family caregivers of persons with young-onset dementia. *Internet Interv, 25*. doi:10.1016/j.invent.2021.100390

Bruinsma, J., Peetoom, K., Bakker, C., Boots, L., Verhey, F., and de Vugt, M. (2021). “They simply do not understand”: a focus group study exploring the lived experiences of family caregivers of people with frontotemporal dementia. *Aging Ment Health.* doi:10.1080/13607863.2020.1857697

Caceres, B. A., Frank, M. O., Jun, J., Martelly, M. T., Sadarangani, T., and de Sales, P. C. (2016). Family caregivers of patients with frontotemporal dementia: An integrative review. *Int J Nurs Stud, 55*, 71-84. doi:10.1016/j.ijnurstu.2015.10.016

Cations, M., Withall, A., Horsfall, R., Denham, N., White, F., Trollor, J., Loy, C., Brodaty, H., Sachdev, P., Gonski, P., Demorkol, A., Cumming, R. G., and Draper, B. (2017). Why aren't people with young onset dementia and their supporters using formal services? Results from the INSPIRED study. *PLoS One, 12*(7), e0180935. doi:10.1371/journal.pone.0180935

Craig, P., Dieppe, P., Macintyre, S., Michie, S., Nazareth, I., and Petticrew, M. (2008). Developing an evaluating complex interventions: the new Medical Research Council Guidance. *BMJ 337*. doi:10.1136/bmj.a1655

Christie, H. L., Boots, L. M. M., Peetoom, K., Tange, H. J., Verhey, F. R. J., and de Vugt, M. E. (2020). Developing a Plan for the Sustainable Implementation of an Electronic Health Intervention (Partner in Balance) to Support Caregivers of People With Dementia: Case Study. *JMIR Aging, 3*(1), e18624. doi:10.2196/18624

Christie, H. L., Boots, L. M. M., Tange, H. J., Verhey, F. R. J., and de Vugt, M. E. (2021). Implementations of Evidence-Based eHealth Interventions for Caregivers of People With Dementia in Municipality Contexts (Myinlife and Partner in Balance): Evaluation Study. *JMIR Aging, 4*(1), e21629. doi:10.2196/21629

Cohen, S. (1988). Perceived stress in a probability sample of the United States. In In S. Spacapan S. Oskamp (Eds.), The Claremont Symposium on Applied Social Psychology. The social psychology of health (pp. 31–67): Sage Publications, Inc.

Diehl-Schmid, J., Schmidt, E. M., Nunnemann, S., Riedl, L., Kurz, A., Forstl, H., Wagenpfeil, S., and Cramer, B. (2013). Caregiver burden and needs in frontotemporal dementia. *J Geriatr Psychiatry Neurol*, 26 (4), 221-229. doi:10.1177/0891988713498467

Draper, B., Cations, M., White, F., Trollor, J., Loy, C., Brodaty, H., Sachdev, P., Gonski, P., Demirkol, A., Cumming, R., and Withall, A. (2016). Time to diagnosis in young-onset dementia and its determinants: the INSPIRED study. *Int J Geriatr Psychiatry, 31*(11), 1217-1224. doi:10.1002/gps.4430

Ducharme, S., Dols, A., Laforce, R., Devenney, E., Kumfor, F., van den Stock, J., . . . Pijnenburg, Y. (2020). Recommendations to distinguish behavioural variant frontotemporal dementia from psychiatric disorders. *Brain 143*(6), 1632-1650. doi:10.1093/brain/awaa018

Duits, A. A., Boots, L. M. M., Mulders, A. E. P., Moonen, A. J. H., and de Vugt, M. E. (2020). Covid proof self-management training for caregivers of patients with parkinson's disease. *Mov Disord, 36*(3), 529-530. doi:10.1002/mds.28457

Edwards, R., Telfair, J., Cecil, H., and Lenoci, J. (2000). Reliability and validity of a self- efficacy instrument specific to sickle cell disease. *Behav Res Ther, 3*(9), 951–963. doi:10.1016/S0005-7967(99)00140-0

Eysenbach, G. (2011). CONSORT-EHEALTH: improving and standardizing evaluation reports of Web-based and mobile health interventions. *J Med Internet Res, 13*(4), e126. doi:10.2196/jmir.1923

Fortinsky, R. H., Kercher, K., and Burant, C. J. (2002). Measurement and correlates of family caregiver self-efficacy for managing dementia. *Aging Ment Health, 6*(2), 153-160. doi:10.1080/13607860220126763

Gossink, F., Pijnenburg, Y., Scheltens, P., Pera, A., Kleverwal, R., Korten, N., Stek, M., Dröes, R-M., and Dols, A. (2018). An intervention programme for caregivers of dementia patients with frontal behavioural changes: an explorative study with controlled effect on sense of competence. *Psychogeriatrics, 18*(6), 451-459. doi:10.1111/psyg.12351

Johannessen, A., Helvik, A-S., Engedal, K., and Thorsen, K. (2017). Experiences and needs of spouses of persons with young-onset frontotemporal lobe dementia during the progression of the disease. *Scand J Caring Sci. 31*(4), 779-788. doi:10.1111/scs.12397

Kaizik, C., Caga, J., Camino, J., O'Connor, C. M., McKinnon, C., Oyebode, J. R., Piguet, O., Hodges, R. R., and Mioshi, E. (2017). Factors Underpinning Caregiver Burden in Frontotemporal Dementia Differ in Spouses and their Children. *J Alzheimers Dis, 56*(3), 1109-1117. doi:10.3233/JAD-160852

Kobiske, K. R., and Bekhet, A. K. (2018). Resilience in Caregivers of Partners With Young Onset Dementia: A Concept Analysis. *Issues Ment Health Nurs, 39*(5), 411-419. doi:10.1080/01612840.2017.1400625

Lindau, M., Almkvist, O., Kushi, J., Boone, K., Johansson, S. E., Wahlund, L. O., Cumming, J. L., and Miller, B. L. (2000). First Symptoms – Frontotemporal Dementia versus Alzheimer's Disease. *Dement Geriatr Cogn Disord, 11*, 286–293. doi:10.1159/000017251

Marshall, G. N., and Lang, E. L. (1990). Optimism, self-mastery, and symptoms of depression in women professionals. *J Pers Soc Psychol, 59*(1), 132

Mioshi, E., Foxe, D., Leslie, F., Savage, S., Hsieh, S., Miller, L., Hodges, J. R., and Piguet, O. (2013). The Impact of Dementia Severity on Caregiver Burden in Frontotemporal Dementia and Alzheimer Disease. *Alzheimer Dis Assoc Disord, 27*(1), 68-73. doi:10.1097/WAD.0b013e318247a0bc

Morgan, D. L. (2014). Pragmatism as a paradigm for social research. *Qual Inq 20*(8), 1045-1053. doi:10.1177/1077800413513733

Nunnemann, S., Kurz, A., Leucht, S., and Diehl-Schmid, J. (2012). Caregivers of patients with frontotemporal lobar degeneration: a review of burden, problems, needs, and interventions. *Int Psychogeriatr, 24*(9), 1368-1386. doi:10.1017/S104161021200035X

Onyike, C. U., and Diehl-Schmid, J. (2013). The epidemiology of frontotemporal dementia. *Int Rev Psychiatry*, *25*(2), 130-137. doi:10.3109/09540261.2013.776523

Pearlin, L. I., and Schooler, C. (1978). The Structure of Coping. *J Health and Soc Behav, 19*(1), 2-21. doi:10.2307/2136319

Polenick, C. A., Struble, L. M., Stanislawski, B., Turnwald, M., Broderick, B., Gitlin, L. N., and Kales, H. C. (2018). "The Filter is Kind of Broken": Family Caregivers' Attributions About Behavioral and Psychological Symptoms of Dementia. *Am J Geriatr Psychiatry, 26*(5), 548-556. doi:10.1016/j.jagp.2017.12.004

Rabinovici, G. D., and Miller, B. L. (2010). Frontotemporal Lobar Degeneration Epidemiology, Pathophysiology, Diagnosis and Management. *CNS Drugs, 24*(5), 375-398. doi:10.2165/11533100-000000000-00000

Rosness, T. A., Haugen, P. K., and Engedal, K. (2008). Support to family carers of patients with frontotemporal dementia. *Aging Ment Health, 12*(4), 462-466. doi:10.1080/13607860802224334

Shnall, A. (2009). Supporting family caregivers of people with frontotemporal dementia. *Can Rev Alzheimers Dis Other Dement, 12*(2), 14-17.

Spinhoven, P., Ormel, J., Sloekers, P. P., Kempen, G. I., Speckens, A. E., and van Hemert, A. M. (1997). A validation study of the hospital anxiety and depression scale (HADS) in different groups of Dutch subjects. *Psychol Med, 27*(2), 363–370. doi:10.1017/s0033291796004382

van Vliet, D., de Vugt, M. E., Bakker, C., Pijnenburg, Y. A., Vernooij-Dassen, M. J., Koopmans, R. T., and Verhey, F. R. (2013). Time to diagnosis in young-onset dementia as compared with late-onset dementia. *Psychol Med, 43*(2), 423-432. doi:10.1017/S0033291712001122

Walford-Kraemer, P., and Light, H. K. (1984). Depression and mastery in women: differences according to personal characteristics. *Psychol Rep, 54* (3), 710. doi:10.2466/pr0.1984.54.3.710

Zigmond, A. S., and Snaith, R. P. (1983). The Hospital Anxiety and Depression Scale. *Acta Psychiatr Scand, 67*(6), 361-370. doi:10.1111/j.1600-0447.1983.tb09716.x

Chapter 8

General discussion

In part one, this dissertation aimed to obtain a better understanding of the lived experiences of YOD and FTD caregivers regarding the disease trajectory to identify directions for support. Therefore, the lived experiences of these caregivers were thoroughly explored by using data from the Needs in Young-onset Dementia (NeedYD) study (chapter 2), by conducting focus group discussions (chapter 3), and performing a case study (chapter 4). Based on the findings of part one, existing web-based support interventions were tailored for YOD and FTD caregivers, namely the RHAPSODY program [*Online training dementie op jonge leeftijd*] (chapter 5) and the Partner in Balance intervention (chapter 6 and 7). Tailored versions contained YOD-specific content for spouse caregivers, children in their puberty or older, and other relatives involved in caregiving. In a cross-sectional study, the tailored version of the RHAPSODY program was evaluated in terms of user acceptability, usability, satisfaction, and user behavior (chapter 5). Using a prospective design, different versions of the Partner in Balance intervention were evaluated in terms of usability, feasibility, and acceptability. This also allowed to examine explorative effects for self-efficacy, sense of mastery, distress, anxiety, and depression (chapter 6 and 7).

The general discussion starts with summarizing the main findings of these studies, followed by a discussion of the methodological considerations and a reflection on the practical implications of the findings. The general discussion ends with a conclusion.

Main findings

Part one: The experiences of YOD and FTD caregivers regarding the disease trajectory

Throughout part one, this dissertation quantitatively and qualitatively explored the experiences of YOD and FTD caregivers regarding the disease trajectory. Using data from the NeedYD study (van Vliet et al., 2010), disease symptoms were identified that were associated with a lower relationship quality as perceived by spouses who lived with a partner with YOD (chapter 2). By building upon a literature review of Holdsworth and McCabe (2018), our results quantified how a diminished sense of couplehood as perceived by spouses was associated with low awareness of deficits, and high levels of apathy, hyperactivity, depression, and anxiety in persons with YOD. While apathy, hyperactivity, and a low awareness of disease occur in many subtypes of dementia, they are characteristic for FTD (Bang et al., 2015). This provides an explanation to why spouses who have a partner with FTD were less satisfied with their relationship (chapter 2). In subsequent studies (chapter 3 and 4), spouses confirmed they lost the sense of couplehood due to specific symptoms of FTD, such as a lack of disease awareness, loss of empathy, low emotional responsiveness, and behavioral symptoms, such as apathy. Our results revealed that personality and coping strategies used by spouses also adversely affected the relationship quality, such as neuroticism or palliative and passive reaction patterns (chapter 2). This suggests that it is important to help spouses to cope emotionally and adaptively with symptoms of YOD, particularly in case of FTD.

Previous results from the NeedYD study showed that the diagnosis of YOD is often delayed, particularly in case of FTD (van Vliet et al., 2013). Often the pre-diagnostic phase is characterized by high levels of uncertainty experienced by caregivers (van Vliet et al., 2011). Our focus group discussions (chapter 3) and case study (chapter 4) allowed for a better understanding of how specific symptoms of FTD complicate the provision of a timely diagnosis and cause pre-diagnostic uncertainty in FTD caregivers.

In the pre-diagnostic phase, caregivers often had difficulty labeling early symptoms as signs of an underlying pathology. This was attributed to the insidious onset of subtle changes in character and behavior accompanying FTD (chapter 3). Early recognition was further hampered because persons with FTD were often capable of presenting well to others and were persuasive in denying symptoms. This induced pre-diagnostic doubts in family caregivers, for example about why their relative with FTD behaved in a selfish or uninterested manner (chapter 3 and 4). Family and friends often did not recognize these early symptoms of FTD and trivialized the concerns of caregivers. This gave caregivers the feeling they overreacted (chapter 3). As a result, caregivers postponed reaching out for professional consultation to address their concerns. When caregivers did consult a healthcare professional, some experienced their concerns were downplayed and symptoms were misattributed to work-related stress or marital problems. According to caregivers this further delayed a timely diagnosis (chapter 3).

Although most caregivers of persons with FTD perceived the diagnosis as devastating, at the same time they sometimes experienced the diagnosis as a relief because it helped them to better understand symptoms and it allowed access to social and professional support (chapter 3 and 4). Some caregivers perceived that more distant family and friends had doubts about the accuracy of the diagnosis, trivialized the severity of symptoms of bvFTD, and questioned care decisions made by family caregivers. This made caregivers of persons with FTD feel frustrated, unrecognized, misunderstood, and socially unsupported (chapter 3). Generally, caregivers also perceived that healthcare professionals often had limited experience with FTD and were unable to provide them with advice on coping with specific symptoms. This undermined the confidence that FTD caregivers had in the support of healthcare professionals (chapter 3).

Only limited insight in the positive aspects of diagnostic disclosure of YOD or FTD is currently available (Tookey et al., 2021). Our findings encourage the development of new insight into positive aspects of the diagnosis for family dynamics. Moreover, the diagnosis helped family members to reevaluate their expectations, redefine new roles for themselves, and redistribute responsibility within the family. The diagnosis also allowed caregivers to transcend the strong opinion and apathetic behavior of their relative with FTD. This facilitated talking about decision-making and helped family members to set goals for their future. Organizing care for their relative with FTD created a sense of cohesion and companionship between the family members that were directly involved (chapter 4). The findings indicate support should account for individual differences in involvement, coping, and bereavement caused by the dynamic interplay between symptoms and family life. A sense of mutual understanding between family members could potentially further strengthen the relationships and help families to cope with organizing care for a relative with FTD who lives at home.

In the post-diagnostic phase, the specific symptoms accompanying FTD made it difficult for caregivers to relate to information and support currently available (chapter 3). It is known that YOD and FTD caregivers can perceive support as not age-appropriate (Cations et al., 2017; Mayrhofer et al., 2018). Our findings extend on this by highlighting the importance of tailored support for FTD caregivers because they can struggle to relate to generic information about YOD (chapter 3 and 6). The low level of adequate professional support made FTD caregivers feel unrecognized, unacknowledged, and misunderstood (chapter 3). Additionally, available support often neglects the perspectives of children, often in puberty, adolescence or (young) adulthood (Barca et al., 2014; Grundberg et al., 2021; Cartwright et al., 2021). Our findings confirm the need for a support approach that accounts for the perspectives of different family members (chapter 3 and 4). The current mismatch between available support and the needs of caregivers, leads to the postponement of the initiation of professional support (chapter 3). This further highlights the importance of acceptable and timely support tailored to the specific needs of caregivers who have a relative with YOD or FTD who lives at home.

Part two: Evaluating web-based support tailored for YOD and FTD caregivers

Building upon the findings from part one (chapter 2 and 3), two existing web-based interventions were tailored to support caregivers of persons with YOD and FTD who live at home. More specifically, the RHAPSODY program (Metcalfe, 2019; Kurz et al., 2016) and the Partner in Balance intervention (Boots et al., 2018; Boots et al., 2016; Duits et al., 2020). These two interventions were selected because they had proven effectiveness in other countries or target populations. The tailoring process was guided by building upon the findings of the NeedYD study, results of part one of this dissertation (chapter 2 and 3), a comprehensive search of international literature, and by involving end-users and stakeholders.

In part two, the RHAPSODY program was evaluated in terms of user acceptability, usability, user satisfaction, and user behavior (chapter 5). Different versions of the Partner in Balance intervention were evaluated in terms of usability, feasibility, acceptability, and explorative effects (chapter 6 and 7). In general, our findings confirm the need for tailored web-based information and support for YOD and FTD caregivers. Like previous studies, caregivers perceived both the RHAPSODY program (Metcalfe, 2019) and the Partner in Balance intervention (Boots et al., 2016) as usable and acceptable. Findings show that the web-based approach allowed for flexibility in the use of the interventions from home at a convenient time (chapter 5, 6 and 7). The web-based approach matched well to the expectations of children aged 16 years or older. For example, they felt it was easier to voice their emotions online via the Partner in Balance intervention, in comparison to in-person meetings with healthcare professionals.

Most caregivers felt that the free and publicly available RHAPSODY program provided a necessary platform containing YOD-specific information (chapter 5). They felt this was useful throughout the disease trajectory and helped them to better understand the symptoms of YOD. They valued the age-appropriate and YOD subtype-specific content and recognized the content from their own daily experiences. When caregivers had become familiar with the RHAPSODY program, they felt more aware of where to find relevant information. Most caregivers indicated that they would likely make use of the program again. The public availability of the RHAPSODY program also facilitated accessibility for healthcare professionals and persons with YOD themselves. Although the RHAPSODY program aims to educate caregivers, the evaluation revealed it also had educational value for healthcare staff and persons with YOD themselves. Only minor suggestions for improvement were identified, such as a clear overview of the program content to facilitate better navigation through the extensive materials incorporated in the RHAPSODY program.

The Partner in Balance intervention (chapter 6 and 7) complements the RHAPSODY program (chapter 5) by offering caregivers a personalized trajectory including online coaching by a healthcare professional, such as a dementia casemanager or psychologist. The Partner in Balance intervention has already proven effective in a more generic population and entails self-management techniques to help caregivers adapt to the caregiving role (Boots et al., 2018; Boots et al., 2016). To achieve this, caregivers follow online modules including videos, psychoeducation, self-reflection, and goal-setting under guidance of an online coach. Chapter 6 evaluated tailored content that was developed for spouses and other family members of persons with YOD, such as children in their puberty or older and other relatives involved in caregiving. This evaluation showed that caregivers of persons with FTD had more need for tailored content, they perceived the content on YOD as too generic. Therefore, content was redeveloped and tailored specifically for spouses of persons with FTD, which elicited high levels of recognition. To illustrate, seeing peers with FTD openly talk about the challenges they faced inspired spouses to actively involve their family and friends (chapter 7). This is relevant because FTD caregivers are at particular risk for feeling socially unsupported (chapter 4).

Our evaluation showed that YOD and FTD caregivers described the Partner in Balance intervention as self-explanatory, easy to use, and logically structured (chapter 6 and 7). Most caregivers appreciated the recognizability, relevancy, usefulness, and quality of the tailored content incorporated. They felt the focus on self-management via self-reflection and goal-setting was essential in translating the Partner in Balance content to daily practice. Feedback from the coach was vital and made caregivers feel heard, helped them to gain new insights, and reevaluate the high expectations they had of themselves. In line with earlier studies evaluating Partner in Balance (Boots et al., 2018; Boots et al., 2016; Duits et al., 2020), caregivers felt more confident, better prepared, and more at ease after the intervention. Some caregivers had improved on measures on self-efficacy after the intervention. Spouses of persons with FTD also reported lower levels of anxiety and depression. Similar to a previous study on the Partner in Balance intervention (Boots et al., 2017), caregivers felt that the therapeutic bond with their casemanager or psychologist had strengthened if they provided the coaching. Altogether these findings indicate that the Partner in Balance intervention can successfully support caregivers in coping with the caregiving role in daily life.

Methodological considerations

This part discusses methodological considerations by reflecting on study designs, data collection methods, and the involvement of end-users and stakeholders. This section ends by discussing endeavors for implementation because they are essential to guarantee caregivers can continue to benefit from the developed web-based support.

Study designs and methodology

A strength of the research documented in this dissertation is the use of diverse research designs and varied methods to collect quantitative and qualitative data. In part one, this allowed a comprehensive understanding of the experiences of YOD and FTD caregivers and helped with identifying directions for support. In part two, the use of different methods to collect data allowed for the thorough evaluation of tailored versions of the RHAPSODY program and the Partner in Balance intervention.

In part one, the research builds upon previous studies to bridge knowledge gaps regarding the quality of the relationship (chapter 2), lived experiences of FTD caregivers (chapter 3), and the interference of FTD with family dynamics (chapter 4). A strength accompanying the research performed is the use of existing data and knowledge that is already available. Building upon the findings and data from the NeedYD study helped us to direct our studies. For example, data from the NeedYD study had sufficient statistical power to explore unique quantitative factors threatening the quality of the relationship as perceived by spouses (chapter 2). A review performed by Holdsworth and McCabe (2018), demonstrated there is only limited understanding of factors that adversely affect the quality of the relationship in YOD.

The NeedYD study also contains qualitative data, that enabled us to explore relevant directions for the topic guides used to structure the qualitative studies reported in part one. Moreover, NeedYD data was used to obtain directions for the focus group discussions (chapter 3) and case study (chapter 4). The findings of such qualitative studies have limited transferability but were designed to obtain a more thorough insight in the lived experiences of FTD caregivers throughout the disease trajectory. In both studies, methods were embedded to increase the credibility of qualitative findings. For instance, in advance to the focus group discussions (chapter 3) caregivers prepared by completing a booklet containing questions about daily life using the context-mapping approach (Visser et al., 2005). Subsequently, findings were interpreted by consulting the chair of the Dutch FTD peer-support organization [*FTD lotgenoten*] to achieve a form of member checking. In the case study (chapter 4), follow-up interviews were organized for member checking. This allowed to deepen the findings, verify preliminary conclusions, and clear ambiguities (Merriam, 2009; Tracy, 2010). To reduce researcher bias, the qualitative data analyses were performed independently by two researchers and the results were discussed extensively with the entire research team to establish investigator triangulation (Carter et al., 2014).

In part two, existing interventions with proven effectiveness were used instead of reinventing the wheel. This enabled us to build upon the initial development process and cost-effectively allocate financial resources to co-designing new content for the RHAPSODY program (chapter 5) and the Partner in Balance intervention (chapter 6 and 7). A strength of using existing interventions is that it enabled verification of our findings in correspondence with earlier findings. By building upon earlier studies, a variety of quantitative and qualitative instruments were selected. Also, since usability, feasibility, and acceptability are broad measures and partly overlapping concepts that are hard to grasp with only quantitative measures (Maramba et al., 2019; Hoddinott, 2015).

The RHAPSODY program was previously tested in Germany, France, and the United Kingdom. A randomized controlled trial showed the RHAPSODY program had high levels of usability and acceptability (Metcalfe, 2019). These earlier versions provided a good starting point for developing the Dutch version of the RHAPSODY program. Generally, the Dutch version showed similar effects as previously documented. Our findings are not sufficient to provide claims about generalized effects, but they do create a certain degree of confidence about the benefits of the RHAPSODY program for YOD and FTD caregivers.

The Partner in Balance intervention was developed using the Medical Council Research (MRC) framework. The MRC-framework is a guide to facilitate the development and evaluation of complex, technological, and health promoting interventions, such as eHealth interventions (Craig et al., 2008). To illustrate, the development was guided by conducting focus group discussions (Boots et al., 2015), co-designing content, feasibility testing (Boots et al., 2016), a randomized controlled trial (Boots et al., 2018), and a process evaluation (Boots et al., 2017). These extensive efforts provided us with a solid evidence-based platform for the tailored versions of the Partner in Balance intervention and enabled us to interpret our findings in light of earlier results. Using a prospective design enabled us to examine explorative effects of the Partner in Balance intervention (chapter 6 and 7). This was needed because the content was extensively revised and there are specific differences between caregivers of persons with YOD or FTD and the caregivers of elderly with dementia. However, given the relatively small samples included in our studies the findings are not sufficient to support earlier claims about generalized effects (Boots et al., 2018). However, like previous findings our qualitative and quantitative results indicate that Partner in Balance had substantial benefits for YOD and FTD caregivers and helped them in coping with the caregiving role. To expand the evidence for the effectiveness of the Partner in Balance intervention, a randomized controlled trial with sufficient power is underway to estimate the long-term effects and health-economic impact.

The involvement of end-users and stakeholders

A strength of this dissertation is the close involvement of the target group and other stakeholders in our research efforts. Throughout different stages of the research, family caregivers of persons having YOD and FTD, healthcare professionals, and field experts were involved. Involving them in our research enabled us to take both caregiver perspectives as well as the healthcare-related context into account. This increased the likelihood that our results and the tailored web-based interventions were accepted. For instance, caregivers helped to design topic lists for interviews (chapter 4) and helped to interpret qualitative findings (chapter 3). Additionally, caregivers, healthcare professionals, and field experts were closely involved to design tailored intervention content. They provided feedback, formulated advice for peers, wrote narrative stories, and helped to develop new videos (chapter 5, 6 and 7). In this process, the Dutch FTD peer-support organization [*FTD lotgenoten*] also gave directions for developing content, reviewed tailored materials, and provided feedback. Additionally, they actively mobilized their network for recruiting participants, invited us to present at their peer-support meetings, and disseminated information via social media and newsletters. On several occasions, caregivers or healthcare professionals joined us to present at (inter)national conferences to share their perspective. Altogether, these joint activities led to a strong collaboration with YOD and FTD caregivers, the Dutch FTD peer-support organization [*FTD lotgenoten*], and healthcare professionals. This enabled us to represent their perspectives, and this increased the acceptance of the developed web-based interventions.

A strength of our research is the collaborations that were established with dementia networks such as the Dutch Young-onset Dementia Knowledge Center [*Kenniscentrum Dementie op Jonge Leeftijd*], the Dutch Alzheimer's Society [*Alzheimer Nederland*], Academic Network of Nursing Homes Nijmegen [*Universitair Kennisnetwerk Ouderenzorg Nijmegen, UKON*], and Radboud University Medical Center [Radboudumc]. To illustrate, the Dutch Alzheimer's Society [*Alzheimer Nederland*] co-designed content and developed the website for the RHAPSODY program. Their expertise on YOD, writing informative texts, and website development ensured that reliable and high-quality content was incorporated. They also hosted the program on their website www.dementie.nl, thereby allowing public and structural access to a large audience. Collaborating with a wide variety of stakeholders helped to effectively recruit participants, for example by spreading information via mailings lists, newsletters, websites, and social media. Particularly effective were the bimonthly meetings with healthcare organizations affiliated with the Dutch Young-onset Dementia Knowledge Center [*Kenniscentrum Dementie op Jonge Leeftijd*]. These meetings allowed us to directly spread information and disseminate findings of our research to affiliated healthcare professionals and care organizations. A consideration to take into account is that early stage caregivers are known to experience difficulty acknowledging their care needs and may postpone accepting support (Boots et al., 2015). As our recruitment mainly occurred through healthcare professionals and peer-support organizations this may have biased our findings because it led to the inclusion of caregivers who are further in the acceptance process.

Endeavors to implement the Partner in Balance intervention

Another methodological consideration is that the perspectives of healthcare professionals and directions for the implementation of the Partner in Balance intervention fall beyond the scope of our research. Implementation is needed to guarantee that caregivers can continue to benefit from the intervention in the future. Therefore, the perspectives of healthcare professionals that provided online coaching using the Partner in Balance intervention were evaluated after our study reported in chapter 6. Additionally, to familiarize healthcare professionals with the intervention a trial was embedded in available (inter)professional training programs on YOD that are offered by Dutch health academies and universities of applied science.

Important to guide the efforts for implementation, was a business model that has been developed based on extensive inventory of the perspectives of healthcare professionals and other stakeholders (Boots et al., 2017; Christie et al., 2020). Based on this, a partnership has been established with technology partner Ilionx. Currently, care organizations are able to license the Partner in Balance intervention per caregiver. The non-profit license is used to cover the expenses for website maintenance, technological support, and consultation sessions for healthcare professionals that provide coaching using the intervention. Additionally, crowd-funding initiatives and fundraisers are organized to cover the costs. A benefit of using this strategy is that the implementation becomes less reliant of external funding to ensure caregivers can continue to use the Partner in Balance intervention in the future.

Practical implications and future directions

Overall, the research in this dissertation highlights the need for a tailored approach to support family members of persons with YOD, in particular in case of FTD. This section will address implications for YOD and FTD caregivers and for the care organizations supporting them. This section ends with suggestions for policy makers, funders of research, and healthcare insurance companies.

Caregivers

Our findings revealed that web-based support was perceived as low threshold, flexible, usable, and acceptable. Therefore a web-based approach may help to bridge the discrepancy between needing and accepting support. Especially in the phase directly after diagnosis caregivers have an explicit need for YOD-specific information and guidance (Millenaar et al., 2018). However, early stage caregivers are known to postpone the initiation of support services while they later regret the postponement of support. This is known as the caregiver needs paradox (Boots et al., 2015).

The publicly available RHAPSODY program might be a valuable tool that allows caregivers to access YOD-specific information in an early stage. This may motivate them to initiate other support services, such as involving a dementia casemanager. A more personalized trajectory is provided by the Partner in Balance intervention that is guided by an online coach. The self-management techniques embedded in the intervention showed to help caregivers to adapt to the caregiving role when supporting a relative with YOD or FTD who lives at home. In order to make caregivers aware of these web-based support tools it is an important direction to raise awareness about their existence.

Healthcare professionals and care organizations

Healthcare professionals and care organizations are vital in supporting caregivers of persons with YOD and FTD who live at home. For example, they facilitate casemanagers, home care, daycare, and provide information or psychoeducation to caregivers (Bakker et al., 2013). Our findings revealed that caregivers would optimally benefit from this if they are strengthened in using adaptive strategies to cope with symptoms of YOD, in particularly in case of FTD. Additionally, caregivers may benefit from support services that are available to the family as whole. To optimize the quality of care and support services, it seems important that care organizations equip dementia casemanagers and psychologists with adequate resources. For example, by providing them with budget and time to offer web-based support. Additionally, allocating experienced healthcare professionals to support YOD and FTD caregivers seems helpful. An important direction to improve current professional support is to invest in the education of healthcare professionals on YOD. In the Netherlands, specific (inter)professional training programs for healthcare professionals are offered by health academies and universities of applied science. As a steppingstone to educate healthcare staff, the publicly available RHAPSODY program may have educational value. Currently, the Dutch Young-onset Dementia Knowledge Center [*Kenniscentrum Dementie op Jonge Leeftijd*] advises care organizations to use the program as an educational tool to train healthcare staff.

Our findings also reveal that caregivers can benefit from tailored information and support provided by the publicly available RHAPSODY program. Therefore, it is important for care organizations to inform early stage caregivers about the program. This may increase the confidence of YOD and FTD caregivers in professional care and may promote timely access to other supportive services, such as a dementia case manager or daycare. To offer caregivers a personalized trajectory, care organizations can consider embedding the Partner in Balance intervention in their daily practice. Therefore, it is important that healthcare professionals feel confident in using digital support tools in daily practice. Our experience taught us that healthcare professionals were persuaded to explore the use of such web-based support tools during the Covid-19 pandemic. However, sustainably using a blended support approach in daily practice requires a different role and technological skills from healthcare professionals. Therefore, we offer training to empower healthcare professionals in using the Partner in Balance intervention. These trainings account for the organizational context, personal experience with coaching, and aim to increase the confidence of healthcare professionals in working with web-based support in daily practice.

Policy makers, funding parties, and healthcare insurance companies

In the Netherlands, the Ministry of Health, Welfare and Sport [*Ministerie van Volksgezondheid, Welzijn en Sport*] recognizes complex medical conditions with a low prevalence that need specific long-term care [*laag volume hoog complex in wet langdurige zorg*]. Currently, YOD and FTD are not recognized. Acknowledging YOD and FTD as complex medical conditions that need specific long-term care would be an important recognition for the low prevalence, high heterogeneity in clinical presentation, and complexity of the symptoms. This would be an important recognition for the context-dependent and disease-specific challenges faced by persons with YOD and FTD, their families, and the care organization supporting them. More recognition is also helpful as it potentially enables healthcare insurance companies to allocate more financial resources to care organizations that support persons with YOD or FTD and their families. Ultimately, this helps to improve the quality of care and support. For example, care organizations would increasingly be able to invest in the education of healthcare staff and would be able to reimburse expenses for using proven effective interventions in daily practice, such as the Partner in Balance intervention.

A positive development in this regard is that *ZonMw* and the Dutch Alzheimer's Society [*Alzheimer Nederland*] have incorporated YOD as a specific target group on their scientific research agenda. A suggestion to them is to recognize the unique challenges faced by FTD caregivers and take this into account when subsidizing future research. Additionally, our findings suggest more support is needed that accounts for the perspectives of different family members, for example by using a system approach to support families. Another suggestion for funders of research, policy makers, and healthcare insurance companies is to allocate sufficient financial resources for the structural implementation of effective interventions. Currently, only 3% of the effective interventions for persons with dementia and their caregivers make it into daily care practice (Christie et al., 2019; Gitlin et al., 2015). Partner in Balance has demonstrated promising effects in several prospective studies (Boots et al., 2016; Duits et al., 2020; Bruinsma et al., 2021a; Bruinsma et al., 2021b) and a randomized controlled trial (Boots et al., 2018). Based on these results, the Dutch knowledge network of long-term care [*Vilans*] has acknowledged Partner in Balance as an effective intervention to support caregivers who have a relative with dementia living at home.

Although endeavors for implementation have been funded, there is a need for structural financial support. A future study on the health-economic impact may provide indisputable evidence on the effects of Partner and Balance and hopefully persuade policy makers and healthcare insurance companies to provide structural reimbursement to healthcare organizations offering the intervention.

Conclusions

The studies documented in this dissertation provide a unique and better understanding of the specific challenges faced by YOD and FTD caregivers throughout the disease trajectory. The findings indicate that there should be more recognition for YOD and FTD as a specific subvariants of dementia. The results advocate for specific and tailored support for caregivers of persons with YOD, especially in case of FTD. This can be achieved by educating healthcare professionals about YOD and by assigning caregivers to experienced healthcare professionals with sufficient knowledge about YOD and FTD. To allow timely access to supportive services, healthcare organizations can consider implementing web-based support in their current strategy. A web-based support approach was acceptable and showed to be beneficial for caregivers who have a relative with YOD or FTD who lives at home. Using a web-based approach early in the caregiving trajectory may eventually lower the threshold for caregivers to accept and initiate professional support. To facilitate the use of web-based support in daily practice there should be structural financial support for the implementation of proven successful interventions, such as the Partner in Balance intervention.

References

Bang, J., Spina, S., and Miller, B. L. (2015). Frontotemporal dementia. *The Lancet, 386*(10004), 1672-1682. doi:10.1016/s0140-6736(15)00461-4

Barca, M. L., Thorsen, K., Engedal, K., Haugen, P. K., and Johannessen, A. (2014). Nobody asked me how I felt: experiences of adult children of persons with young-onset dementia. *Int Psychogeriatr, 26*(12), 1935-1944. doi:10.1017/S1041610213002639

Bakker, C., de Vugt, M. E., van Vliet, D., Verhey, F. R., Pijnenburg, Y. A., Vernooij-Dassen, M. J., and Koopmans, R. T. (2013b). The use of formal and informal care in early onset dementia: results from the NeedYD study. *Am J Geriatr Psychiatry, 21*(1), 37-45. doi:10.1016/j.jagp.2012.10.004

Boots, L. M., de Vugt, M. E., Withagen, H. E., Kempen, G. I., and Verhey, F.R . (2016). Development and initial evaluation of the web-based self-management program “partner in balance” for family caregivers of people with early stage dementia: an exploratory mixed-methods study. *JMIR Res Protoc, 5*(1), e33 doi:10.2196/resprot.5142.

Boots, L. M., de Vugt, M. E., Smeets, C. M., Kempen, G. I., and Verhey, F. R. (2017). Implementation of the blended care self-management program for caregivers of people with early-stage dementia (partner in balance): process evaluation of a randomized controlled trial. *J Med Internet Res, 1*(12), e423 doi:10.2196/jmir.7666

Boots, L. M., de Vugt, M. E., Kempen, G. I., and Verhey, F. R. (2018). Effectiveness of a blended care self-management program for caregivers of people with early-stage dementia (partner in balance): randomized controlled trial. *J Med Internet Res, 20*(7), e10017 doi:10.2196/10017

Boots, L. M., Wolfs, C. A., Verhey, F. R., Kempen, G. I., and de Vugt, M. E. (2015). Qualitative study on needs and wishes of early-stage dementia caregivers: the paradox between needing and accepting help. *Int Psychogeriatr, 27*(6), 927-936. doi:10.1017/S1041610214002804

Bruinsma, J., Peetoom, K., Bakker, C., Boots, L., Millenaar, J., Verhey, F., and de Vugt, M. E. (2021a). Tailoring and evaluating the web-based ‘Partner in Balance’ intervention for family caregivers of persons with young-onset dementia. *Internet Interv, 25.* doi:10.1016/j.invent.2021.100390

Bruinsma, J., Peetoom, K., Boots, L., Daemen, M., Verhey, F., Bakker, C., and de Vugt, M. E. (2021b). Tailoring the web-based ‘Partner in Balance’ intervention to support spouses of persons with frontotemporal dementia. *Internet Interv, 26.* doi:10.1016/j.invent.2021100442

Carter, N., Bryant-Lukosius, D., DiCenso, A., Blythe, J., and Neville, A. J. (2014). The use of triangulation in qualitative research. *Oncol Nurs Forum, 41*(5), 545-547. doi:10.1188/14.ONF.545-547

Cartwright, A. V., Stoner, C. R., Pione, R. D., and Spector, A. (2021). The experiences of those affected by parental young onset dementia: A qualitative systematic literature review. *Dementia,* online ahead of print. doi:10.1177/1471301220988231

Cations, M., Withall, A., Horsfall, R., Denham, N., White, F., Trollor, J., Loy, C., Brodaty, P. S., Gonski, P., Demirkol, A., Cumming, R. G., and Draper, B. (2017). Why aren't people with young onset dementia and their supporters using formal services? Results from the INSPIRED study. *PLoS One, 12*(7), e0180935. doi:10.1371/journal.pone.0180935

Christie, H. L., Martin, J. L., Connor, J., Tange, H. J., Verhey, F. R. J., de Vugt, M. E., and Orrell, M. (2019). eHealth interventions to support caregivers of people with dementia may be proven effective, but are they implementation-ready? *Internet Interv, 18*, 100260. doi:10.1016/j.invent.2019.100260

Christie, H. L., Boots, L. M. M., Peetoom, K., Tange, H. J., Verhey, F. R. J., and de Vugt, M. E. (2020). Developing a plan for the sustainable implementation of an electronic health intervention (partner in balance) to support caregivers of people with dementia: case study. *JMIR Aging 3*(1), e18624. doi:10.2196/18624

Craig, P., Dieppe, P., Macintyre, S., Michie, S., Nazareth, I., and Petticrew, M. (2008). Developing an evaluating complex interventions: the new Medical Research Council Guidance. *BMJ 337*. doi:10.1136/bmj.a1655

Duits, A. A., Boots, L. M. M., Mulders, A. E. P., Moonen, A. J. H., and de Vugt, M. E. (2020). Covid proof self-management training for caregivers of patients with parkinson's disease. *Mov Disord, 36*(3), 529-530. doi:10.1002/mds.28457

Gitlin, L. N., Marx, K., Stanley, I. H., and Hodgson, N. (2015). Translating evidence-based dementia caregiving interventions into practice: state-of-the-science and next steps. *Gerontologist 55*(2), 210–226. doi:10.1093/geront/gnu123

Grundberg, A., Sandberg, J., and Craftman, A. G. (2021). Childrens' and young adults' perspectives of having a parent with dementia diagnosis: A scoping review. *Dementia*, online ahead of print. doi:10.1177/14713012211023653

Hendriks, S., Peetoom, K., Bakker, C., van der Flier, W. M., Papma, J. M., Koopmans, R., Verhey, F. R., de Vugt, and M., Köhler, S. (2021). Global prevalence of young-onset dementia: a systematic review and meta-analysis. *JAMA Neurol*. doi:10.1001/jamaneurol.2021.216

Hoddinott, P. (2015). A new era for intervention development studies. *Pilot feasibility stud, 1.* doi:10.1186/s40814-015-0032-0

Hogan, D. B., Jette, N., Fiest, K. M., Roberts, J. I., Pearson, D., Smith, E. E., Roach, P., Krik, A., Pringsheim, T., and Maxwell, C. J. (2016). The prevalence and incidence of frontotemporal dementia: a systematic review. *Can J Neurol Sci, 43*(1), S96-S109. doi:10.1017/cjn.2016.25

Holdsworth, K., and McCabe, M. (2018). The impact of younger-onset dementia on relationships, intimacy, and sexuality in midlife couples: a systematic review. *Int Psychogeriatr, 30*(1), 15-29. doi:10.1017/S1041610217001806

Kurz, A., Bakker, C., Bohm, M., Diehl-Schmid, J., Dubois, B., Ferreira, C., Gage, H., Graff, C., Hergueta, T., Jansen, S., Jones, B., Komar, A., de Mendonça, A., Metcalfe, A., Milecka, K., Millenaar, J., Orrung Wallin, A., Oyebode, J., Schneider-Schelte, H., Saxl, S., and de Vugt, M. (2016).

RHAPSODY - Internet-based support for caregivers of people with young onset dementia: program design and methods of a pilot study. *Int Psychogeriatr, 28*(12), 2091-2099. doi:10.1017/S1041610216001186

Maramba, I., Chatterjee, A., and Newman, C. (2019). Methods of usability testing in the development of eHealth applications: a scoping review. *Int J Med Inform, 126, 95-104*. doi:10.1016/j.ijmedinf.2019.03.018.

Mayrhofer, A., Mathie, E., McKeown, J., Bunn, F., and Goodman, C. (2018). Age-appropriate services for people diagnosed with young onset dementia (YOD): a systematic review. *Aging Ment Health, 22*(8), 927-935. doi:10.1080/13607863.2017.1334038

Merriam, S. B. (2009). Qualitative research: A guide to design and implementation. San Francisco: Jossey-Bass.

Metcalfe, A., Jones, B., Mayer, J., Gage, H., Oyebode, J., Boucault, S., Aloui, S., Schwertel, U., Böhm, M., Tezenas du Montcel, S., Lebbah, S., De Mendonça, A., De Vugt, M., Graff, C., Jansen, S., Hergueta, T., Dubois, B., and Kurz, A. (2019). Online information and support for carers of people with young- onset dementia: A multi-site randomised controlled pilot study. *Int J Geriatr Psychiatry, 34*(10), 1455–1464. doi:10.1002/gps.5154

Millenaar, J. K., Bakker, C., van Vliet, D., Koopmans, R., Kurz, A., Verhey, F. R. J., and de Vugt, M. E. (2018). Exploring perspectives of young onset dementia caregivers with high versus low unmet needs. *Int J Geriatr Psychiatry, 33*(2), 340–347. doi:10.1002/gps.4749

Tookey, S. A., Greaves, C. V., Rohrer, J. D., and Stott, J. (2021). Specific support needs and experiences of carers of people with frontotemporal dementia: A systematic review. *Dementia*, online ahead of print. doi:10.1177/14713012211022982

Tracy, S. J. (2010). Qualitative Quality: Eight “Big-Tent” Criteria for Excellent Qualitative Research. *Qual Inq, 16*(10), 837-851. doi:10.1177/1077800410383121

van Vliet, D., Bakker, C., Koopmans, R. T., Vernooij-Dassen, M. J., Verhey, F. R. and de Vugt, M. E. (2010a). Research protocol of the NeedYD-study (Needs in Young onset Dementia): a prospective cohort study on the needs and course of early onset dementia. *BMC Geriatr, 10*. doi:10.1186/1471-2318-10-13

van Vliet, D., de Vugt, M. E., Bakker, C., Koopmans, R. T., Pijnenburg, Y. A., Vernooij-Dassen, M. J., and Verhey, F. R. (2011). Caregivers' perspectives on the pre-diagnostic period in early onset dementia: a long and winding road. *Int Psychogeriatr, 23*(9), 1393-1404. doi:10.1017/S1041610211001013.

van Vliet, D., de Vugt, M. E., Bakker, C., Pijnenburg, Y. A., Vernooij-Dassen, M. J., Koopmans, R. T., and Verhey, F. R. (2013). Time to diagnosis in young-onset dementia as compared with late-onset dementia. *Psychol Med, 43*(2), 423-432. doi:10.1017/S0033291712001122

Visser, F. S., Stappers, P. J., van der Lugt, R., and Sanders, E. B. N. (2005). Contextmapping: experiences from practice. *CoDesign, 1*(2), 119-149. doi:10.1080/15710880500135987

Addendum

Summary | Nederlandse samenvatting

Impact paragraph

Author information and list of publications

Dankwoord (acknowledgements)

Thesis defenses from MHeNs

Summary

Young-onset dementia (YOD) refers to dementia with an onset of symptoms before the age of 65 years. The clinical presentation and underlying pathology of YOD is more diverse than in dementia occurring at older age. Partly, this is caused by the higher prevalence of frontotemporal dementia (FTD) in persons with YOD. FTD is a rare and specific dementia subvariant characterized by symptoms such as language problems, loss of semantic knowledge, or profound changes in personality and behavior. The specific symptoms that start at young age are known to delay the diagnosis of YOD, especially in case of FTD. This is problematic because it impedes access to care and support. Throughout the disease trajectory, family members of persons with YOD and FTD face unique challenges, such as combining caregiving with employment or raising children. Additionally, they have to cope with specific symptoms of YOD and FTD that can result in high levels of burden and distress. Problematic is that available support is often not age-appropriate, primarily aimed at Alzheimer’s dementia, and does not account for the perspectives of (adult) children caring for a parent with YOD or FTD. Given the low prevalence of these conditions, a web-based approach may provide a promising direction to offer tailored support to family members of persons with YOD and FTD who live at home. A web-based approach is in line with the current digital trend and may allow service access at a convenient time and place, even in rural areas.

Research documented in this dissertation explored the lived experiences of YOD and FTD caregivers regarding the disease trajectory to identify directions for support (part one). Then, existing web-based support was tailored for this specific group (part two), namely the RHAPSODY program [*Online training dementie op jonge leeftijd*] and the Partner in Balance intervention. The RHAPSODY program was evaluated in terms of user acceptability, usability, user satisfaction, and user behavior. Additionally, different tailored versions of the Partner in Balance intervention were evaluated in terms of usability, feasibility, acceptability, and explorative effects.

Part one: The experiences of YOD and FTD caregivers regarding the disease trajectory

Data from the NeedYD study was used to identify factors that influence the quality of the relationship as perceived by spouses who have a partner with YOD who lives at home (chapter 2). Findings show that spouses perceived lower quality of the relationship if their partner with YOD had symptoms of apathy, hyperactivity, anxiety, depression, and low awareness of disease. While apathy, hyperactivity, and a low awareness of disease can occur in may subtypes of dementia, they are characteristic for FTD. Therefore, this may explain why spouses of partners who have FTD experienced lower levels of satisfaction regarding their relationship. Spouses that used coping characterized by a neurotic personality and palliative or passive reaction patterns were also less satisfied about the quality of their relationship. Therefore, a direction for support is to help spouses to emotionally and adaptively cope with symptoms of YOD, particularly in case of FTD.

Focus group discussions were conducted to explore the lived experiences of caregivers of persons with FTD throughout the disease trajectory (chapter 3). Subsequently, a case study was conducted to examine how symptoms of the behavioral variant of FTD interfered with family dynamics (chapter 4). Both studies helped in obtaining a better understanding of how specific symptoms of FTD complicate establishing a timely diagnosis, and lead to high levels of pre-diagnostic uncertainty in family caregivers. Findings revealed that FTD caregivers often struggled to label early symptoms as signs of a pathology because symptoms had an insidious onset and progressed slowly. Persons with FTD were often capable of presenting well to others and were persuasive in denying symptoms. This led to trivializing responses from family and friends trying to mitigate the early concerns of family caregivers. This increased doubts and led caregivers to postpone reaching out for professional consultation, which would usually take place through the general practitioner. The diagnosis was further delayed if healthcare professionals misattributed early symptoms, for example to work related stress or marital problems. For caregivers this delay in diagnosis was problematic because diagnostic disclosure facilitated understanding of symptoms of FTD and was often the beginning of identifying themselves as a caregiver. A diagnosis also allowed access to professional care and support. Therefore, obtaining the diagnosis was sometimes experienced as a relief. The diagnosis also helped caregivers to comprehend why dynamics within the family had changed. This allowed them to reevaluate expectations, redefine roles, and redistribute responsibilities within the family. This facilitated talking about decision-making and created a sense of companionship among family members involved in organizing care.

Following the diagnosis, some FTD caregivers perceived that more distant family and friends had doubts about the accuracy of the diagnosis, trivialized the severity of symptoms, or questioned care decisions made by caregivers. This was experienced as frustrating and made FTD caregivers feel socially unsupported. FTD caregivers also perceived that healthcare professionals had generally only limited experience with FTD and were unable to provide advice on coping with specific symptoms. They perceived that most post-diagnostic support services were designed with elderly and Alzheimer's dementia in mind. Especially, (adult) children felt there was little relevant and appropriate support available. The mismatch between service availability and need for tailored support made FTD caregivers feel unrecognized, unacknowledged, and misunderstood. This undermined their confidence in professional healthcare and led them to postpone the initiation of supportive services. The low level of professional and social support induced feelings of loneliness in FTD caregivers. Therefore, a direction is to offer tailored and acceptable psychosocial support to family members of persons with YOD, particularly in case of FTD.

Part two: Evaluating tailored web-based support for YOD and FTD caregivers

Existing web-based interventions were tailored and evaluated for caregivers who have a relative with YOD or FTD living at home, namely the RHAPSODY program (chapter 5) and the Partner in Balance intervention (chapter 6 and 7). The tailoring process was guided by the findings of the NeedYD study, results of part one of this dissertation (chapter 2 and 3), a comprehensive search of international literature, and by involving end-users and stakeholders.

In general, caregivers perceived the RHAPSODY program and the Partner in Balance intervention as usable and acceptable. Caregivers particularly valued the flexibility of both online approaches that allowed them to participate from home at a convenient time.

The RHAPSODY program [*Online training dementie op jonge leeftijd*] is a web-based educational program for caregivers that is publicly available via www.dementie.nl. It includes videos with experts and caregivers, medical information about YOD subtypes, advice regarding coping with caregiving, and offers an overview of available care and support services in the Netherlands. According to caregivers, the RHAPSODY program provides a necessary platform containing YOD-specific information that is useful throughout the disease trajectory. They appreciated the usability and valued the age-appropriate content relevant in daily life. Our evaluation revealed the publicly available RHAPSODY program also had educational value for healthcare professionals and persons with YOD themselves. Minor suggestions for improvement were identified, such as a clear overview of the program content, in order to facilitate navigation through the materials incorporated in the RHAPSODY program.

The Partner in Balance intervention complements the RHAPSODY program by offering caregivers a personalized trajectory guided by an online coach, who is usually a healthcare professional. Using self-management principles, the Partner in Balance intervention supports caregivers of persons with YOD and FTD who live at home. Therefore, caregivers follow thematic modules that contain a video, psychoeducation, self-reflection, and goal-setting under guidance of an online coach. Caregivers perceived the intervention as feasible and felt the coach was an essential element of the intervention. The coach was often a dementia casemanager or psychologist and helped caregivers to gain new insights, apply the intervention in daily life, and made caregivers feel heard. According to caregivers, this strengthened the therapeutic bond. Following the Partner in Balance, intervention caregivers felt more confident, better prepared, and more at ease. After the intervention, some caregivers had improved on measures of self-efficacy. Spouses of persons with FTD also reported lower levels of anxiety and depression.

General discussion and conclusions

Our findings provide more understanding of the specific challenges faced by YOD and FTD caregivers throughout the disease trajectory. This helped to identify directions to support this specific group. The findings advocate for more recognition for the context-dependent and disease-specific challenges faced by these caregivers, especially in case of FTD. Support that is tailored to their specific needs would allow them to optimally benefit from post-diagnostic support. To achieve this, care organizations could educate healthcare professionals on YOD and assign caregivers to a healthcare professional experienced with YOD or FTD. Additionally, web-based interventions showed usable, feasible, and acceptable tools to support caregivers who have a relative with YOD or FTD who lives at home. Web-based support helped caregivers to cope with the daily challenge of organizing care for a relative with YOD or FTD. Therefore, care organizations could consider embedding web-based support in daily practice. Potentially this helps to lower the threshold for caregivers to timely initiate other care and support services.

Nederlandse samenvatting

Dementie op jonge leeftijd begint voor het 65^{e} levensjaar. In Nederland hebben 14.000 tot 17.000 jonge mensen dementie. De symptomen en de onderliggende pathologie is meer divers bij jonge mensen met dementie, vergeleken met oudere mensen met dementie. Het sluipende begin zorgt ervoor dat de vroege symptomen lastig te herkennen zijn. Dit geldt in het bijzonder voor jonge mensen met frontotemporale dementie (FTD). FTD is een specifiek subtype van dementie dat relatief weinig voorkomt en wordt gekenmerkt door ingrijpende persoonlijkheids- en gedragsveranderingen, verlies van feitenkennis, of problemen met taalbegrip en -productie. Door de jonge leeftijd en aspecifieke presentatie worden de vroege symptomen van dementie op jonge leeftijd vaak onterecht toegeschreven aan een burn-out of huwelijksproblemen. Dit zorgt voor vertraging in het diagnostisch traject. Voor familieleden van jonge mensen met dementie of FTD is dit problematisch, omdat dit de toegang tot hulp en ondersteuning belemmert.

Dementie op jonge leeftijd begint als mensen nog volop in het leven staan. Het organiseren van mantelzorg vraagt daarom veel van de betrokken gezins- en familieleden. Partners van jonge mensen met dementie combineren bijvoorbeeld een baan met mantelzorg. Regelmatig zijn er ook thuis- of uitwonende kinderen betrokken bij het organiseren van de mantelzorg. Het huidige zorg- en ondersteuningsaanbod is vaak onvoldoende afgestemd op de specifieke behoeften van deze jonge groep mantelzorgers. Dat komt doordat informatie en ondersteuning voornamelijk gericht is op ouderen, veelal gaat over Alzheimer en maar weinig aansluit bij de behoeften van pubers, adolescenten of (jong)volwassenen met een jonge ouder met dementie of FTD. Vanwege de lage prevalentie van dementie op jonge leeftijd, met in het bijzonder FTD, biedt een online interventie wellicht een goede manier om landelijke en gepaste ondersteuning te bieden. Een online benadering sluit eveneens goed aan bij de huidige digitale trend en de vraag naar laagdrempelige en flexibele ondersteuningsvormen.

Onderzoek gerapporteerd in dit proefschrift verkent de ervaringen van mantelzorgers van jonge mensen met dementie en FTD gedurende het ziektetraject. Ook worden suggesties voor passende ondersteuning geformuleerd (deel 1). Voortbouwend hierop zijn er twee bestaande en effectieve online interventies aangepast voor deze specifieke groep, namelijk het RHAPSODY-programma en de Partner in Balans interventie. Onderzoek beschreven in dit proefschrift evalueert deze recent doorontwikkelde interventies in termen van gebruiksvriendelijkheid, toepasbaarheid en tevredenheid (deel 2).

Deel 1: De ervaringen van mantelzorgers gedurende het ziektetraject

Er zijn drie onderzoeken uitgevoerd om de ervaringen van mantelzorgers gedurende het ziektetraject beter in kaart te brengen. Allereerst is er gebruik gemaakt van data afkomstig uit de NeedYD-studie (hoofdstuk 2). Binnen ons onderzoek zijn symptomen geïdentificeerd die invloed hebben op de ervaren relatiekwaliteit van partners van thuiswonende jonge mensen met dementie. Partners blijken een lagere relatiekwaliteit te ervaren als hun geliefde een gebrek heeft aan ziekte-inzicht en symptomen heeft van apathie, hyperactiviteit, angst en depressie. Een gebrekkig ziekte-inzicht en symptomen van apathie en hyperactiviteit komen veel voor bij FTD. Dit verklaart ook waarom partners van mensen met FTD minder tevreden waren over hun relatie. De relatiekwaliteit bleek ook lager bij partners met een neurotische persoonlijkheid en bij partners die gebruik maken van palliatieve en passieve reactiepatronen. De bevindingen benadrukken daarom het belang van praktische handvatten die partners helpen om emotioneel en adaptief om te gaan met symptomen van dementie op jonge leeftijd, en in het bijzonder FTD.

Vervolgens zijn er groepsgesprekken georganiseerd om de ervaringen van mantelzorgers van mensen met FTD beter in kaart te brengen (hoofdstuk 3). Daaropvolgend is er een casestudie verricht om meer zicht te krijgen op hoe symptomen van FTD de gezinsdynamiek beïnvloeden (hoofdstuk 4). Beide onderzoeken hielpen om beter te begrijpen hoe de specifieke symptomen van FTD het diagnostisch traject bemoeilijken en veel onzekerheid veroorzaken bij mantelzorgers. De bevindingen maken verder inzichtelijk hoe het sluipende begin de tijdige herkenning van symptomen belemmerde. Dit kwam bijvoorbeeld, omdat mensen met FTD zich vaak goed konden presenteren aan de buitenwereld en erg overtuigend waren in het ontkennen van symptomen. Volgens mantelzorgers werden hun zorgen hierdoor vaak gebagatelliseerd door familie en vrienden die verder van de situatie afstonden en vroege symptomen niet herkenden. Dit veroorzaakt twijfel en veel mantelzorgers stelden hierdoor een bezoek aan de huisarts uit. Het bleek dat huisartsen of andere zorgprofessionals vaak ook moeite hadden om vroege symptomen van FTD te herkennen. Symptomen werden vaak ten onrechte toegeschreven aan werkgerelateerde stress of huwelijksproblemen. Dit leidde tot verdere vertraging van het diagnostisch traject. Terugblikkend ervaarden veel mantelzorgers de vertraagde diagnose van FTD als problematisch. De diagnose hielp hen namelijk om symptomen van FTD te begrijpen en maakte het mogelijk om hulp en ondersteuning in te schakelen. Veel mantelzorgers zagen zichzelf na de diagnose pas als mantelzorger. Hierdoor werd een diagnose soms als een opluchting beschouwd. In de fase na de diagnose begrepen veel mantelzorgers ook beter waarom de dynamiek binnen het gezin was veranderd. Dit hielp om verwachtingen bij te stellen, rolpatronen opnieuw te definiëren en verantwoordelijkheid binnen het gezin anders te verdelen. De diagnose zorgde ervoor dat er meer gesproken werd over de toekomst en het organiseren van mantelzorg. Dit creëerde een gevoel van samenhorigheid tussen de betrokken gezins- en familieleden.

Sommige mantelzorgers hadden na de diagnose het gevoel dat familie en vrienden de diagnose in twijfel trokken, de ernst van symptomen bagatelliseerden, of kritisch waren over keuzes van mantelzorgers. Dit werd als zeer frustrerend ervaren. Mantelzorgers voelden zich hierdoor sociaal onvoldoende gesteund. Veel mantelzorgers vonden ook dat zorgprofessionals over het algemeen te weinig kennis en ervaring hadden met FTD en hierdoor onvoldoende in staat waren om advies te geven over het omgaan met de specifieke symptomen. Volgens veel mantelzorgers kwam dit omdat FTD niet goed past binnen het generieke beeld dat mensen hebben over dementie. Bestaande informatie en ondersteuning bleek bijvoorbeeld vooral gericht op ouderen en Alzheimer dementie. Met name (jong)volwassen met een ouder met FTD vonden dat er maar weinig geschikte ondersteuning beschikbaar was. Deze discrepantie tussen beschikbare ondersteuning en ondersteuningsbehoeften bleek problematisch, omdat veel mantelzorgers zich hierdoor niet herkend of begrepen voelden. Dit leidde tot gevoelens van eenzaamheid. Het gebrek aan geschikte ondersteuning ondermijnde ook het vertrouwen in de beschikbare ondersteuning. Hierdoor stelden mantelzorgers het gebruik van professionele ondersteuning uit. Een belangrijke conclusie is daarom dat er acceptabele psychosociale ondersteuning op maat nodig is voor mantelzorgers en gezinnen van mensen met FTD.

Deel 2: Het evalueren van specifiek ontwikkelde online ondersteuning

Er zijn drie onderzoeken uitgevoerd om specifieke online ondersteuning te evalueren voor mantelzorgers van jonge mensen met dementie en FTD. Namelijk, het RHAPSODY-programma (hoofdstuk 5) en de Partner in Balans interventie (hoofdstuk 6 en 7). Deze al bestaande en effectieve interventies zijn recentelijk doorontwikkeld en inhoudelijk herzien om ze geschikt te maken voor partners en andere gezins- en familieleden van thuiswonende jonge mensen met dementie of FTD. Bij de doorontwikkeling is er gebruik gemaakt van bevindingen uit de NeedYD-studie, onderzoek gerapporteerd in deel 1 van dit proefschrift en internationale wetenschappelijke literatuur. Daarnaast is er nauw samengewerkt met mantelzorgers en andere belanghebbenden.

Over het algemeen werden het RHAPSODY-programma en de Partner in Balans interventie beide als toepasbaar, gebruiksvriendelijk en acceptabel beschouwd. De online insteek werd hoog gewaardeerd en maakte het voor mantelzorgers mogelijk om vanuit thuis op een geschikt tijdstip deel te nemen. De meeste mantelzorgers waardeerden de hoogwaardige informatie en het advies dat herkenbaar en bruikbaar was in het dagelijkse leven.

Het RHAPSODY-programma [*Online training dementie op jonge leeftijd*] is een online educatieprogramma voor mantelzorgers dat gratis beschikbaar is via www.dementie.nl. Het programma bevat onder andere video's met experts en mantelzorgers, medische informatie over verschillende subtypes van dementie op jonge leeftijd en tips en advies over het omgaan met symptomen en de mantelzorgrol. Het programma bevat ook een overzicht met beschikbare zorg en ondersteuning in Nederland. Het publiekelijk toegankelijke programma werd ervaren als een centraal kennisplatform met hoogwaardige en relevante informatie, zowel kort na de diagnose als in latere fases van het mantelzorgtraject. Ons onderzoek liet zien dat het programma ook educatieve waarde had voor zorgprofessionals en jonge mensen met dementie zelf. Uit de evaluatie kwam naar voren dat een duidelijke inhoudsopgave zou helpen bij het navigeren door de uitgebreide inhoud van het RHAPSODY-programma.

De Partner in Balans interventie is een aanvulling op het RHAPSODY-programma en biedt mantelzorgers een gepersonaliseerd traject onder begeleiding van een online coach. Met zelfmanagementtechnieken biedt de Partner in Balans interventie ondersteuning aan mantelzorgers van thuiswonende jonge mensen met dementie of FTD. Gedurende de interventie volgen mantelzorgers thematische modules, bijvoorbeeld over veranderingen in het gezinsleven, het balanceren van werk en zorgtaken, of over het omgaan met zorgen over erfelijke belasting. Iedere module bevat één video, achtergrondinformatie met persoonlijke verhalen en tips, zelfreflectieopdrachten en het stellen van een persoonlijk doel onder begeleiding van een online coach. In de praktijk werd de coaching vaak gegeven door de eigen casemanager dementie of psycholoog. Volgens mantelzorgers was de coach essentieel en hielp de feedback bij het komen tot nieuwe inzichten en bij het toepassen van de interventie in het dagelijkse leven. Mantelzorgers voelden zich gehoord door de feedback van de coach en dit versterkte de therapeutische relatie. Na de Partner in Balans interventie voelden mantelzorgers zich ook zelfverzekerder, beter voorbereid en rustiger. Op vragenlijsten scoorden ze hoger op zelfeffectiviteit en lager op gevoelens van angst en depressie.

Conclusie

Onze bevindingen bieden een uniek inzicht in de uitdagingen voor mantelzorgers van jonge mensen met dementie of FTD. De bevindingen benadrukken het belang van geschikte ondersteuning voor deze specifieke groep mantelzorgers. Zorgorganisaties kunnen passende ondersteuning leveren door bijvoorbeeld in lotgenotensteun te faciliteren, zorgprofessionals te trainen op gebied van dementie op jonge leeftijd en mantelzorgers toe te wijzen aan ervaren zorgprofessionals. Daarnaast kunnen zorgorganisaties overwegen om online ondersteuning op te nemen in hun zorg- en ondersteuningsaanbod voor mantelzorgers van thuiswonende jonge mensen met dementie of FTD. Dit verlaagt voor mantelzorgers wellicht de drempel om hulp in te schakelen en zorgt mogelijk voor tijdige toegang tot geschikte ondersteuning. Online ondersteuning helpt mantelzorgers ook om zich goed aan te passen aan de uitdagingen die voortvloeien uit de mantelzorgrol. Om mantelzorgers in de toekomst beter te kunnen ondersteunen zou er meer erkenning moeten zijn voor de unieke uitdagingen die een gevolg zijn van dementie op jonge leeftijd, met in het bijzonder FTD. Beleidsmakers, sponsoren van wetenschappelijk onderzoek en zorgverzekeringsmaatschappijen zouden meer financiële middelen ter beschikking moeten stellen ter ondersteuning van deze specifieke groep. Daarnaast is financiële ondersteuning nodig voor de structurele implementatie van wetenschappelijk erkende interventies, zoals de Partner in Balans interventie.

Impact paragraph

The research documented in this dissertation had a practical focus and was conducted close to the daily lives of family members of persons with young-onset dementia (YOD) and frontotemporal dementia (FTD). As a result, the web-based support interventions that were tailored had a direct impact on their functioning and well-being.

This paragraph reflects on the impact of the findings, activities that helped to disseminate the results, and endeavors to implement web-based support to assure it continues to have impact in the future.

Scientific and societal relevance of our findings

Our research provides a unique insight in the experiences of caregivers of persons with YOD and FTD throughout the disease trajectory. Using these insights, two web-based support programs, the RHAPSODY program [*Online training dementie op jonge leeftijd*] and the Partner in Balance intervention [*Partner in Balans*] were tailored and evaluated to support this specific group. The tailored interventions empowered caregivers by improving their ability to cope adaptively with daily challenges. More specifically, the RHAPSODY program was acceptable and useful in daily life and helped caregivers with understanding the symptoms of YOD. The Partner in Balance intervention offered caregivers a personalized trajectory using self-management techniques guided by an online coach, usually a healthcare professional. The coach helped caregivers to use the intervention content in daily life. Caregivers felt more confident, better prepared, and more at ease after the intervention. Some caregivers had improved on measures on self-efficacy after the intervention. Spouses of persons with FTD also reported lower levels of anxiety and depression. The wider societal impact stems in the fact that YOD and FTD caregivers face unique challenges that can lead to high levels of burden, distress, and social isolation. Providing caregiving has a profound impact on their daily functioning and well-being. The developed web-based support tools ensure that caregivers are adequate supported and help them to cope with caregiving in daily life, thereby contributing to their social participation.

Our research is relevant for researchers and intervention developers because the findings bridge existing knowledge gaps. To illustrate, there is only limited understanding in what works to support family members of persons with YOD or FTD who live at home. Our results encourage the development of age-appropriate support that is tailored for different YOD-subtypes, to apply a system approach to support different family members, and to use support that helps caregivers to adaptively cope with symptoms of YOD, especially in case of FTD. This has impact because it provides valuable suggestions to improve currently available professional support services to allow YOD and FTD caregivers to optimally benefit from support. To achieve this, it is important to equip healthcare professionals with sufficient resources. For example, by educating them via specific (inter)professional training programs on YOD. These programs are currently available in the Netherlands and include introductions on the RHAPSODY program and Partner in Balance intervention. Additionally, the free and publicly available RHAPSODY program showed to be a steppingstone for the education of healthcare staff. Ultimately, more knowledge on YOD and FTD among healthcare professionals improves the quality of the care and support they provide to persons with YOD or FTD and their families.

The relevancy and impact of our findings are not restricted to caregivers, researchers or care organizations but also entail valuable suggestions for policy makers, funders of research, and healthcare insurance companies. More specifically, the findings advocate for recognition of YOD and FTD as complex dementia subvariants with a low prevalence that require specific long-term care [*laag volume hoog complex in wet langdurige zorg*]. Ultimately, such recognition would have tremendous impact as it enables to allocate more financial resources to adequately support persons with YOD or FTD and their family members who provide caregiving. A recent and positive development is that research funder *ZonMW* and the Dutch Alzheimer's Society [*Alzheimer Nederland*] have acknowledged YOD on their research agenda. This provides opportunities to further improve and implement proven successful interventions to optimize support for family members of persons with YOD or FTD.

Activities to disseminate findings

The impact of our research grounds in our efforts to disseminate the research findings. Therefore, collaborations with caregivers and healthcare professionals were established. To illustrate, caregivers helped to raise more awareness for their need for tailored support by joining us during presentations at (inter)national conferences, symposia, and meetings with healthcare professionals. To illustrate, the chair of the Dutch FTD peer-support organization [*FTD lotgenoten*] co-presented results from our focus group discussions at the Alzheimer Europe conference. In addition, healthcare professionals accompanied us during presentations to voice their experiences regarding the Partner in Balance intervention. These joint efforts helped to inform researchers, care organizations, and policy makers about the specific challenges for YOD and FTD caregivers and highlight the benefits of tailored web-based support. The high level of involvement of caregivers and healthcare professionals in our research was the result of their exceptional motivation and willingness to contribute to the development of appropriate support. This highlights the high need for adequate support tools to support family members of persons with YOD, especially in case of FTD. Particularly, the enthusiasm and dedication of caregivers affiliated with the Dutch FTD peer-support organization [*FTD lotgenoten*] who actively mobilized their network contributed to the successful recruitment and dissemination of our research.

Creating momentum to disseminate our findings was further facilitated via the collaboration with dementia networks, such the Dutch Young-onset Dementia Knowledge Center [*Kenniscentrum Dementie op Jonge Leeftijd*], the Dutch Alzheimer's Society [Alzheimer Nederland], Academic Network of Nursing Homes Nijmegen [*Universitair Kennisnetwerk Ouderenzorg Nijmegen, UKON*], and Radboud University Medical Center [*Radboudumc*]. Working together and forming an alliance with these network partners strengthened our efforts and helped to disseminate the findings to a large audience of caregivers, care organizations, and policy makers. Again, the Dutch FTD peer-support organization [*FTD lotgenoten*] played a vital role in helping to disseminate findings. To illustrate, the different dementia network partners invited us to give presentations at meetings and spread information via newsletters, mailing lists, and social media. Working together was mutually beneficial and helped with strengthening each other's expertise. This boosted the development of tailored support, facilitated the recruitment for our studies, and provided us with valuable directions for the implementation. In general, other (care) organizations, clinicians, and researchers were always willing to support us in our research efforts. This created a feeling of cohesion about working towards a common goal, namely promoting adequate and tailored support for family members of persons with YOD and FTD. The high level of dedication of all these network partners also creates confidence that these joint activities will continue in the future.

The impact stands with sustainable implementation

The implementation of the developed web-based support is fundamental to ensure it continues to have an impact in the future. Efforts regarding the implementation were strengthened by the established collaborations with dementia network partners. The Dutch Alzheimer's Society [*Alzheimer Nederland*] co-designed the RHAPSODY program. Currently, they ensure free, public, and structural access to the program via their website www.dementie.nl. These joint efforts assure that the program can continue to provide useful information to caregivers and healthcare professionals in the future.

Ongoing endeavors have led to the sustainable implementation of the Partner in Balance intervention. Unfortunately, healthcare insurance companies do not yet reimburse the costs that care organizations have for using the Partner in Balance intervention. Given the limited funding possibilities, our current strategy is therefore focused on becoming less reliant of external funding. Therefore, the implementation of the Partner in Balance intervention is guided by a business model that enables care organizations to license the intervention per caregiver. The non-profit license is used to cover the expense for website maintenance, technological support, and training for healthcare professionals that provide the coaching. Additionally, crowd-funding initiatives and fundraisers are organized to cover the license costs for caregivers without the support of a care provider. In order to help care organizations to facilitate adequate care for caregivers during the Covid-19 pandemic, a *ZonMW* grant was obtained to offer the program free of charge. To familiarize healthcare professionals with the intervention, a short training and free trial of the Partner in Balance intervention is embedded in (inter)professional training programs on YOD that are available via health academies and universities of applied science in the Netherlands. Altogether, these endeavors are impactful as they guarantee that caregivers can continue to benefit from the Partner in Balance intervention in the future.

Author information

Jeroen Bruinsma was born on 20 January 1989 in Delft, the Netherlands. During his childhood, his parents moved to Limburg where he completed primary school and pre-vocational education (*college Rolduc*). Following his passion for healthcare, he graduated as a nurse in 2011 (*Leeuwenborgh College*) and worked at a psychiatric hospital for several years (*Mondriaan voor geestelijke gezondheidszorg*). Subsequently, he obtained a Bachelor's degree (BSc) in podiatry (*Fontys Hogeschool*) and psychology (*Open Universiteit*). He became intrigued with scientific research when finalizing his Master's degree (MSc) in Health Education and Promotion (*Universiteit Maastricht*). At this time, his father-in-law provided caregiving for his wife with young-onset dementia (YOD). This motivated Jeroen to pursue a PhD aiming to develop psychosocial support for YOD caregivers at the Neuropsychology and Psychiatry department of Maastricht University. During his PhD, Jeroen was also involved in different educational activities. Currently, Jeroen enrolled a postdoc position at the department of Health Promotion at Maastricht University and will continue his research and educational activities. His future research concerns the development of a behavior change model to increase brain health for dementia risk reduction.

List of publications

Bruinsma, J., & Crutzen, R. (2018). A longitudinal study on the stability of the need for cognition. *Personality and Individual Differences*, 127, 151-161.

Bruinsma, J., Peetoom, K., Bakker, C., Boots, L., Millenaar, J., Verhey, F., & de Vugt, M. (2021). Tailoring and evaluating the web-based 'Partner in Balance' intervention for family caregivers of persons with young-onset dementia. *Internet Interventions*, 25, 100390.

Bruinsma, J., Peetoom, K., Bakker, C., Boots, L., Verhey, F., & de Vugt, M. (2020). 'They simply do not understand': a focus group study exploring the lived experiences of family caregivers of people with frontotemporal dementia. *Aging & Mental Health*, 1-9.

Bruinsma, J., Peetoom, K., Boots, L., Daemen, M., Verhey, F., Bakker, C., & de Vugt, M. (2021). Tailoring the web-based 'Partner in Balance' intervention to support spouses of persons with frontotemporal dementia*. Internet Interventions*, 26, 100442.

Bruinsma, J., Peetoom, K., Millenaar, J., Köhler, S., Bakker, C., Koopmans, R., Pijnenburg, Y., Verhey, F., & De Vugt, M. (2020). The quality of the relationship perceived by spouses of people with young-onset dementia. *International psychogeriatrics*, 1-10.

Dankwoord (acknowledgements)

De afgelopen vier jaar ben ik onder de vleugels van mijn promotieteam uitgegroeid tot een volwaardig wetenschapper. **Kirsten**, jij hebt een belangrijke rol gespeeld in deze ontwikkeling. Aan je ongezouten mening heb ik veel gehad. Je was precies de sparringpartner die ik nodig had. We maakten er ook altijd een gezellig boel van. Dat gaf me veel positieve energie. **Christian**, je hebt me geleerd om naar het grotere geheel te kijken. Je bulk aan klinische ervaring hielp me om door een andere bril naar mijn onderzoek te kijken. Daar heb ik veel van geleerd. **Frans**, je bent een autoriteit op het vakgebied en hebt een schat aan kennis en ervaring. Je pragmatische instelling en nuchtere blik hielp enorm bij het doorhakken van knopen. **Marjolein**, je visie op onderzoek met maatschappelijke relevantie heeft mijn kijk op de wetenschap verfijnd. Op wetenschappelijk en persoonlijk vlak ben en blijf je een bron van inspiratie. Bedankt voor de prettige samenwerking van de afgelopen jaren!

Mijn dank gaat ook uit naar de **Hersenstichting, ZonMW, Health foundation Limburg** en **Alzheimer Nederland**. Jullie financiële steun en vertrouwen heeft veel voor me betekend. Dankzij jullie komen de resultaten van mijn onderzoek ten goede aan veel mantelzorgers van jonge mensen met dementie en frontotemporale dementie. Bedankt!

De nauwe samenwerking met **(zorg)organisaties, zorgprofessionals en mantelzorgers** heeft een enorme boost gegeven aan het onderzoek gerapporteerd in dit proefschrift. Ik vind het ontroerend om te zien hoe groot de bereidheid van veel mantelzorgers is tot deelname aan wetenschappelijk onderzoek. In het bijzonder gaat mijn waardering uit naar **FTD lotgenoten** en ook aan het **Kenniscentrum Dementie op Jonge Leeftijd**. Vanaf dag één hebben jullie me warm ontvangen en ik voelde me als kind aan huis. De samenwerking met jullie heeft niet alleen deuren voor me geopend maar ik heb er als mens ook veel van geleerd. Mijn dank is groot!

Ook de warme sfeer bij **Alzheimer Centrum Limburg** heeft de afgelopen jaren veel voor me betekend. De 'sharing is caring' -mentaliteit zorgt voor een vertrouwde en gezellige werkomgeving. Ik heb altijd het gevoel gehad dat iedereen voor me klaarstond. De gezelligheid ga ik ongetwijfeld missen maar ik ben dankbaar dat ik onderdeel mocht zijn van deze inspirerende omgeving. Een belangrijke factor in mijn werkplezier was de nauwe samenwerking binnen het **Partner in Balans Team**. Met veel bevlogenheid hebben we samengewerkt bij de doorontwikkeling en de landelijke uitrol van Partner in Balans. Met plezier blik ik terug op de brainstorm sessies, de PiB-heidag en onze maandelijkse overleggen. Dankzij het goede leiderschap van **Lizzy** en de prettige samenwerking met IT-partner **Ilionx** weet ik zeker dat Partner in Balans een prachtige toekomst tegemoet gaat!

Hoewel ik **al mijn collega's** dankbaar ben voor de prettige werksfeer wil ik er een aantal persoonlijk bedanken. **Hannah**, gedurende mijn PhD heb ik veel van je geleerd. Ook je onvoorwaardelijke hulp bij het doorlezen van mijn proefschrift waardeer ik enorm. Daarnaast wil ik ook mijn waardering uitspreken naar de onderzoeksassistenten **Esther, Anne-Marieke en Roos**. Jullie hebben met veel toewijding een belangrijke bijdrage geleverd bij de praktische uitvoering van mijn onderzoek. Bedankt! **Maud**, jij begon als onderzoeksassistent en bent inmiddels gegroeid tot promovendus. We vormden een goed duo! De prachtige resultaten van de laatste hoofdstukken uit dit proefschrift zijn mede jouw verdienste. Daarnaast wil ik ook **Ron, Danielle** en **Els** bedanken. Jullie staan altijd en voor iedereen klaar. Dat draagt enorm bij aan de warme sfeer binnen Alzheimer Centrum Limburg. Bedankt daarvoor!

In wisselende samenstelling heb ik met verschillende collega's een kantoorruimte gedeeld. Ik wil al mijn '**Roomies**' bedanken voor de leuke tijd. In het bijzonder **Irene, Julia, Lieke en Stevie**. Zonder jullie steun, de humor en de enorme hoeveelheden chocolade had ik dit traject niet overleefd. Zonder de afleiding van **Stevie** was ik waarschijnlijk wel een jaar eerder klaar geweest. Alle gekheid op een stokje, **Stevie** tussen ons is een ware vriendschap opgebloeid. Het klikt erg goed en we delen dezelfde interesses (lees: we zijn allebei gek op bier). Daarnaast was ik zonder jou al lang verhongerd omdat ik regelmatig te weinig saldo heb voor de lunch. De soep bij Bandito's en biertjes bij Thembi houden we erin!

Inmiddels heel wat jaren geleden heb ik in Eindhoven gestudeerd aan de opleiding podotherapie. In deze periode zijn er blijvende vriendschappen ontstaan en heb ik mezelf leren kennen. Daarnaast is ook het fundament gelegd voor mijn academische carrière. Ik werd namelijk al snel lid van het studiegenootschap '**Geeks & Freaks**'. Dankzij **Alper, Arjan, Tan, Astrid en Nicolai** heb ik destijds gekozen voor een academisch vervolg. Bedankt daarvoor!

Na het afronden van mijn Master (MSc) 'Health Education and Promotion' twijfelde ik aanvankelijk over een carrière binnen de wetenschap. **Rik**, jij begeleidde me destijds bij het schrijven van mijn thesis. Jij was ook degene die me inspireerde en motiveerde om te zoeken naar een academisch vervolg. Daar ben ik je enorm dankbaar voor want ik kan me momenteel geen carrière voorstellen buiten de academische wereld.

Kort heb ik ook het privilege gehad om met **Kevin** en **Lars** samen te werken na het afronden van mijn master (MSc). Destijds hebben we veel gelachen en 'geminimaliseerd-gemaximaliseerd'. Ook hebben we diepgaande gesprekken gehad over onze carrières. Mede hierdoor heb ik uiteindelijk gekozen voor het academische pad. Voor mijn gevoel zijn we nog steeds als drie musketiers: "Een voor allen, allen voor een"!

Buiten kijf staat dat dit proefschrift nooit tot stand was gekomen zonder de onvoorwaardelijke steun van het thuisfront. **Sabine**, zonder jou had ik dit niet voor elkaar kunnen boksen. Jij hebt altijd vertrouwen in me en bent mijn steun en toeverlaat. Samen krijgen we alles voor elkaar!

In het bijzonder wil ik ook mijn **ouders** bedanken voor hun steun de afgelopen jaren. Jullie staan altijd klaar voor mij en mijn gezin. Jullie onvoorwaardelijke hulp bij oppassen op Pepijn en steun bij de eindeloze verbouwing is niet met woorden te beschrijven. Bedankt! Daarnaast wil ik ook mijn dank uitspreken naar mijn **schoonouders** voor al de sfeervolle avonden vol kookkunsten van George. Jullie betrokkenheid bij Pepijn geeft me veel energie. Ook wil ik **mijn zusjes** bedanken voor de gezellige weekendjes op de Veluwe en spelletjesavonden.

Ook mijn **grootouders en andere familieleden** wil ik bedanken voor al de leuke familiedagen. In het bijzonder doe ik een shout-out naar mijn neef **Michael** (also known as the prodigy). Je bent de favoriete neef en grote held van Pepijn. Ik hoop dat we nog vaak appeltaart met bitterballen gaan eten.

Hoewel ik mijn vrienden voornamelijk moet bedanken voor de studievertraging op de lagere en middelbare school, vind ik het noemenswaardig dat we al die jaren bevriend zijn. Ik ga niet teveel lof aan jullie toeschrijven in dit dankwoord want dat blijf ik horen. Maar stiekem toch bedankt!

Thesis defenses from MHeNs

2022

Ellis Nelissen | 23 February, 2022
Soluble guanylate cyclase as a novel target for cognition enhancement.
Supervisor: Prof. Dr. J. Prickaerts;
Co-supervisors: Dr. T. Vanmierlo, Dr. P. Sandner, Bayer AG, Germany.

Manon te Dorsthorst | 17 February, 2022
Next Level Tibial Nerve Stimulation in the Treatment of Overactive Bladder Syndrome.
Supervisor: Prof. Dr. J.P.F.A. Heesakkers;
Co-supervisors: Dr. M.R. van Balken, Rijnstate ziekenhuis, Arnhem, Dr. F.M.J Martens, RadboudUMC, Nijmegen.

Maartje Colaris | 11 February, 2022
Breast Implant Illness in Silicone Breast Implant patients.
Supervisors: Prof. Dr. R.R.W.J. van der Hulst, Prof. Dr. J.W. Cohen Tervaert.

Bernice Doove | 8 February, 2022
Personalizing Preschool Preventive Child Health Care; The MOM study.
Supervisors: Prof. Dr. F.J.M. Feron, Prof. Dr. J. van Os, MUMC en Universiteit Utrecht;
Co-supervisor: Dr. M. Drukker.

Supriya Dabir| 27 January, 2022
High resolution retinal imaging.
Supervisor: Prof. Dr. C.A.B. Webers;
Co-supervisors: Dr. T.T.J.M. Berendschot; Dr. J.S.A.G. Schouten.

Evgeniy Svirin | 11 January, 2022
Excessive aggression, ADHD- and ASD-like phenotypes in TPH2- and brain ganglioside- deficient mice: mechanisms and gene-by-environment interaction.
Supervisor: Prof. K.-P. Lesch;
Co-supervisors: Dr. T. Strekalova; Prof. E. Ponomarev, the Chinese University of Hong Kong.

2021

Linda Pagen | 17 December, 2021

Stress and Worry during cognitive aging: behavioral and neural correlates.

Supervisors: Dr. Heidi I.I. Jacobs, Prof. Dr. Benedikt A. Poser;

Co-supervisor: Prof. Dr. Frans R.J. Verhey.

Christian Rauschenberg | 17 December, 2021

Transdiagnostic Approaches to Mental Health: Linking adversity, cognition, candidate mechanisms, and novel digital interventions.

Supervisor: Prof. Dr. J. van Os, UM, Utrecht University Medical Centre;

Co-supervisor: Prof. Dr. U. Reininghaus, Heidelberg University.

Christian Nogales Calvo | 16 December, 2021

Network Modules as Novel Molecular Disease Definitions for Precision Theranostics.

Supervisors: Prof. Dr. Harald H.H.W. Schmidt, Dr. Ana I. Casas Guijarro.

Margot Heijmans | 10 December, 2021

Track and treat Parkinson's disease using wearable sensors and MRI.

Supervisor: Prof. Dr. Yasmin Temel;

Co-supervisors: Dr. Pieter Kubben, Dr. Mark Kuijf.

Jessica Bruijel | 1 December, 2021

Tired of being tired: Fatigue and sleep following traumatic brain injury.

Supervisor: Prof. Dr. C.M. van Heugten;

Co-supervisors: Dr. A. Vermeeren, Dr. S.Z. Stapert.

Naomi Daniëls | 1 December, 2021

Bringing experience-sampling technology to family medicine: Feasibility, usability and lessons-learned.

Supervisors: Prof. dr. P.A.E.G. Delespaul, Prof. dr. A.J. Beurskens;

Co-supervisor: Dr. M.A. van Bokhoven.

Soraya Jonker| 19 November, 2021

Safety and Efficacy of Intraocular Lenses in Cataract - and Refractive Surgery.

Supervisor: Prof. Dr. R.M.M. A. Nuijts;

Co-supervisors: Dr. N.J.C. Bauer, Dr. T.T.J.M. Berendschot.

Ana Maria Alzate Sànchez| 9 November, 2021
Microelectrode recordings for deep brain stimulation: Patient specific variables yields fundamental and clinical insights into the human basal ganglia.
Supervisor: Prof. Dr. Y. Temel;
Co-supervisors: Dr. M.L.F. Janssen, Dr. M.J. Roberts.

Paula Bartholomeus| 9 November, 2021
ReAttach - A transdiagnostic intervention for adults and children with mental health problems.
Supervisor: Prof. Dr. T.A.M.J. van Amelsvoort;
Co-supervisors: Prof. Dr. M. Fitzgerald, Trinity College Dublin, Prof. Dr. D. Marazziti, University of Pisa.

Rik Schalbroeck| 5 November, 2021
Orderly chaos: social defeat as a risk factor for psychosis in autism.
Supervisors: Prof. Dr. J.P.Selten, Prof. Dr. L.F. de Geus-Oei, Leids Universitair Medisch Centrum, Prof. Dr. J. Booij, Amsterdam UMC.

Renzo Riemens| 5 November, 2021
Neuroepigenomics in Alzheimer's disease: The single cell Adds.
Neuroepigenomik bei der Alzheimer-Krankheit: Die Einzelzell ADds.
Supervisors: Prof. dr. D. van den Hove; Prof. dr. B. Rutten; Prof. dr. T. Haaf, Julius- Maximilans-Universität Würzburg;
Co-Supervisor: Dr. G. Kenis; Prof. dr. C. Förster, Julius-Maximilans- Universität Würzburg; Prof. Dr. K.P. Lesch, Julius-Maximilans- Universität Würzburg.

Vera Marsman-Bonekamp| 3 November, 2021
Beyond dis-ease and dis-order - Exploring the long-lasting impact of childhood adversity in relation to mental health.
Supervisor: Prof. Dr. J.J. van Os;
Co-supervisor: Dr. H.B. Lousberg.

Kim van der Linden| 11 October, 2021
Stress, anxiety and psychotic experiences in adults with autism spectrum disorder; and oberservational study in the context of daily life.
Supervisors: Prof. Dr. M.C. Marcelis, Prof. Dr. T.A.M.J. van Amelsvoort;
Co-supervisor: Dr. C.J.P. Simons.

Jeroen Habets| 1 October, 2021
Prediction and real-life monitoring of DBS motor response in Parkinson's disease.
Supervisor: Prof. dr. Y. Temel;
Co-supervisor: Dr. P. Kubben, Dr. M. Kuijf.

Mohammed Alahmari| 15 September, 2021
Radiological and radio-therapeutic nuances in skull base tumours.
Supervisors: Prof. Dr. Y. Temel; Prof. Dr. Ir. F.J.W. Verhaegen;
Co-supervisor: Dr. D. Eekers.

Michaël Veldeman| 15 September, 2021
Diagnosis and treatment of early and delayed cerebral injury after aneurysmal subarachnoid hemorrhage.
Supervisors: Prof. Dr. Y. Temel; Prof. Dr. H. Clusmann, RWTH Aachen;
Co-supervisor: Dr. R. Haeren.

Siyu Wu| 9 September, 2021
Modulation of myelin phagocytosis by means of anti-inflammatory treatment as a therapy of spinal injury.
Supervisors: Prof. Dr. B.W.W. Kramer; Dr. J. Mey, Toledo, Spain.

Wouter Hubens| 2 September, 2021
Glaucoma biomarkers in aqueous humor and blood.
Supervisor: Prof. Dr. C.A.B. Webers;
Co-supervisor: Dr. T.G.M.F. Gorgels.

Sjors van de Weijer| 1 September, 2021
Digital technology-enabled home health care - Gamification in online cognitive therapies for Parkinson's disease.
Supervisor: Prof. Dr. B.R. Bloem, RU Nijmegen;
Co-supervisors: Dr. A.A. Duits; Dr. M.L. Kuijf; Dr. N.M. de Vries, RU Nijmegen.

Lisanne Canjels| 31 August, 2021
Morphological and functional Magnetic Resonance Imaging at ultra-high field.
Supervisors: Dr. J.F.A. Jansen; Prof. Dr. Ir. W.H. Backes; Prof. Dr. A.P. Aldenkamp;
Co-supervisor: Dr. C. Ghossein-Doha.

Yentl van der Zee | 30 August, 2021
Novel Insights into the Neurophysiological and Epigenetic Changes in Major Depressive Disorder.
Supervisor: Prof. Dr. B.P.F. Rutten;
Co-supervisors: Dr. O. Issler; Dr. L. de Nijs; Dr. L.M.T. Eijssen, New York, USA.

Judith Lionarons| 9 July, 2021
Nonmotor comorbidities and somatic manifestations of Duchenne Muscular Dystrophy.
Supervisor: Prof. Dr. C.G. Faber;
Co-supervisors: Dr. G. Hoogland, Dr. J.G.M. Hendriksen, Kempenhaghe, Dr. S. Klinkenberg.

Gusta van Zwieten| 9 July, 2021
Silencing neural symphonies with deep brain stimulation.
Supervisors: Prof. Dr. Y. Temel, Prof. Dr. R.J. Stokroos;
Co-supervisor: Dr. M.L.F. Janssen.

Shuo Zhang| 7 July, 2021
A dark field illumination probe linked to Raman spectroscopy for non-invasivety etermination of ocular biomarkers.
Supervisors: Prof. Dr. C.A.B. Webers, Dr. T.T.J.M. Berendschot;
Co-supervisor: Dr. R.J. Erckens.

Ahmed Hassan| 7 July, 2021
FAIR and bias-free network modules for mechanism-based disease redefinitions.
Supervisors: Prof. Dr. H.H.H.W. Schmidt, Prof. Dr. M.J. Dumontier.

Anouk Geraets| 2 July, 2021
Biological determinants of depression, the role of cerebral damage, microvascular dysfunction, and hyperglycemia: a polulationbased approach.
Supervisors: Dr. M.T. Schram, Prof. Dr. F.R.J. Verhey;
Co-supervisor: Dr. S. Köhler.

Le Guo | 23 June, 2021
No, They Didn't? Oh, They Did!; Advancing Insights on Social Norm Interventions in Consumer Financial Decision-Making.
Supervisor: Prof. Dr. H.J.M. Smeets;
Co-supervisors: Dr. F.H.J. van Tienen; Dr. M. Gerards.

Yvonne van der Zalm | 18 June, 2021
An inquiry into various aspects of clozapine use: prescription, monitoring and mortality.
Supervisor: Prof. Dr. J.P. Selten, Prof. Dr. I.E. Sommer, UMC Groningen;
Co-supervisors: Dr. P.F.J. Schulte, GGZ N.H., Dr. F. Termorshuizen, GGZ Rivierduinen.

Onur Alptekin | 18 June, 2021
Methodological aspects of deep brain stimulation: the untold story behind DBS surgery.
Supervisor: Prof. Dr. Y. Temel;
Co-supervisors: Dr. E. Kocabicak, Samsun, Turkeydr., L. Ackermans.

Elaine Schepers | 17 June, 2021
The role of white noise speech illusions in indicating risk for psychotic disorders.
Supervisor: Prof. Dr. J.J. van Os;
Co-supervisor: Dr. R. Lousberg.

Christian Bertens | 7 June, 2021
Development of a non-invasive ocular drug delivery device.
Supervisor: Prof. Dr. R.M.M.A. Nuijts;
Co-supervisors: Dr. M. Gijs, Dr. F.J.H.M. van den Biggelaar.

Remco Santegoeds | 3 June, 2021
A journey of skull base chordoma: where imaging meets molecular biology.
Supervisor: Prof. Dr. Y. Temel;
Co-supervisors: Dr. L. Jacobi-Postma, Dr. D. Eekers.

Inge Verheggen | 1 June, 2021
Imaging blood-brain barrier function in aging.
Supervisors: Prof. Dr. W.H. Backes, Prof. Dr. F.R.J. Verhey;
Co-supervisor: Dr. A. Jahanshahianvar.

Douwe van der Heide | 27 May, 2021
On the assessment of symptom validity in refugee mental health.
Supervisors: Prof. Dr. H.L.G.J. Merckelbach, Prof. Dr. P.N. van Harten.

Gowoon Son | 26 May, 2021
Olfactory system pathology in Alzheimer's disease: evidences from rodent and human studies.
Supervisors: Prof. dr. H.W.M. Steinbusch, Prof. Dr. C. Moon, DGIST, South Korea;
Co-supervisor: Dr. A. Jahanshahianvar

Markos Xenakis | 26 May, 2021
Molecular complexity of voltage-gated sodium channels; theory and applications in mutation-response prediction.
Supervisor: Prof. Dr. H.J.M. Smeets;
Co-supervisors: Dr. P.J. Lindsey, Dr. R.L. Westra.

Alix Thomson | 21 May, 2021
From Micro to Macro: Unravelling the Underlying Mechanisms of Transcranial Magnetic Stimulation (TMS).
Supervisor: Prof. A.T. Sack;
Co-supervisors: Dr. T.A. de Graaf, Dr. T. Schuhmann, Dr. G.R.L. Kenis.

Ozan Cinar | 19 May, 2021
Combining In formation: Model Selection in Meta-Analysis and Methods for Combining Correlated p-Values.
Supervisor: Dr. W. Viechtbauer;
Co-supervisors: Dr. I.S. Gülöksüz.

Anne Koopmans | 12 May, 2021
CYP2D6 and CYP2C19 genotyping in psychiatry - Bridging the gap between practice and lab.
Supervisors: Prof. Dr. P.N. van Harten, Prof. Dr. H.W.Hoek, RUG;
Co-supervisor: Dr. D.J. Vinkers.

Ashwin Mohan | 29 April, 2021
Retinal oximetry in health and disease.
Supervisor: Prof. C.A.B. Webers;
Co-supervisors: Dr. T.T.J.M. Brendschot, Dr. R. Shetty, Bangalore, India.

Danique Hellebrekers | 23 April, 2021
Neurocognition and behaviour: diagnostic work-up and interventions in Duchenne and Becker muscular dystrophy.
Supervisors: Prof. J.S.H. Vles, Dr. J.G.M. Hendriksen;
Co-supervisor: Dr. S. Klinkenberg.

Milaine Roet | 16 April, 2021 *Modulating microcircuits in depression.*
Supervisor: Prof. Y. Temel;
Co-supervisor: Dr. A. Jahanshahianvar.

Julia van Tuijl | 16 April, 2021
Post-stroke epilepsy.
Supervisor: Prof. A.P. Aldenkamp;
Co-supervisors: Dr. R.P.W. Rouhl, Dr. E.P.M. van Raak.

Hans de Munter | 9 April, 2021
The patient's own bone marrow-derived stromal cells: disease modifiers in (neuro) degenerative disorders.
Supervisors: Prof. B.W.W. Kramer, Prof. E.CH. Wolters;
Co-supervisors: Dr. T. Strekalowa, Dr. J. Mey.

Sophie Leijdesdorff | 9 April, 2021
Ain't no mountain high enough - How to improve access to youth mental health care.
Supervisors: Prof. T.A.M.J. van Amelsvoort, Prof. A. Popma, AMC;
Co-supervisor: Dr. R.M.C. Klaassen, AMC.

Laura Vergoossen | 1 April, 2021
Brain Network Alterations due to Cardiometabolic Risk Factors; Insights from Population Magnetic Resonance Imaging.
Supervisor: Prof. W.H. Backes;
Co-supervisors: Dr. J.F.A. Jansen, Dr. M.T. Schram.

Ranjana J. Jairam | 24 March, 2021
Sacral nerve stimulation for lower urinary tract dysfunctions: towards better outcome.
Supervisors: Prof. Ph.E.V.A. Van Kerrebroeck, Prof. G.A. van Koeveringe; Co-supervisor: Dr. D.M.J. Vrijens.

Luiz Kae Sales Kanazawa | 22 March, 2021
The antimanic-like effects of andrographolide and quercetin
Supervisors: Prof. J. Prickaerts;
Co-supervisor: Dr. Roberto AnDr.eatini, Paraná, Brasil.

Maria Ferrarac | 12 March, 2021
Early Intervention in Psychosis A data-driven population health approach to reduce the duration of untreated psychosis.
Supervisors: Dr. S. Gölöksüz, Prof. J.J. van Os;
Co-supervisor: Prof. V.H. Srihari (Yale University, USA).

Talakad Narasappa Sathyaprabha | 1 February, 2021
Cardiovascular autonomic regulation in health and neurological disorders.
Supervisors: Pof. B.W.W. Kramer, Prof. H.W.M. Steinbusch;
Co-supervisor: Dr. T.R. Raju, Bangalore.

Raoul Stevens | 15 January, 2021
Unboxing the Brain; Development of Technologies for Non-Invasive Assessment of Cerebral Pathologies.
Supervisor: Prof. T. Delhaas, Prof. W.H. Mess;
Co-supervisor: Dr. W. Huberts.

Clara Snijders | 8 January, 2021
Post-traumatic stress disorder epigenetic signatures of differential susceptibility to combat trauma.
Supervisor: Prof. B.P.F. Rutten;
Co-supervisors: Dr. L. de Nijs, Dr. G. Kenis.