

Online information and support for carers of people with young-onset dementia

Citation for published version (APA):

Metcalfe, A., Jones, B., Mayer, J., Gage, H., Oyebode, J., Boucault, S., Aloui, S., Schwertel, U., Bohm, M., du Montcel, S. T., Lebbah, S., De Mendonca, A., De Vugt, M., Graff, C., Jansen, S., Hergueta, T., Dubois, B., & Kurz, A. (2019). Online information and support for carers of people with young-onset dementia: A multi-site randomised controlled pilot study. *International Journal of Geriatric Psychiatry*, 34(10), 1455-1464. <https://doi.org/10.1002/gps.5154>

Document status and date:

Published: 01/10/2019

DOI:

[10.1002/gps.5154](https://doi.org/10.1002/gps.5154)

Document Version:

Publisher's PDF, also known as Version of record

Document license:

Taverne

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.

Online information and support for carers of people with young-onset dementia: A multi-site randomised controlled pilot study

Anna Metcalfe¹  | Bridget Jones²  | Johannes Mayer³ | Heather Gage²  |
 Jan Oyeboode⁴  | Sarah Boucault¹ | Sabrina Aloui¹ | Uta Schwertel⁵  | Markus Böhm³ |
 Sophie Tezenas du Montcel⁶  | Said Lebbah¹  | Alexandre De Mendonça⁷  |
 Marjolein De Vugt⁸  | Caroline Graff^{9,10}  | Sabine Jansen¹¹ | Thierry Hergueta^{1,12} |
 Bruno Dubois¹ | Alexander Kurz³

¹Hôpital de la Pitié-Salpêtrière, Sorbonne University, Assistance Publique – Hôpitaux de Paris AP-HP, Paris, France

²Surrey Health Economics Centre, Department of Clinical and Experimental Medicine, University of Surrey, Guildford, UK

³Department of Psychiatry, Klinikum rechts der Isar, Technical University of Munich, Munich, Germany

⁴Centre for Applied Dementia Studies, University of Bradford, Bradford, UK

⁵IMC, Information Multimedia Communication AG, Saarbrücken, Germany

⁶Sorbonne Universités, INSERM, Institut Pierre Louis d'Epidémiologie et de Santé Publique, Assistance Publique—Hôpitaux de Paris AP-HP, Hôpitaux Universitaires Pitié-Salpêtrière—Charles Foix, Paris, France

⁷Instituto de Medicina Molecular, Universidade de Lisboa, Lisbon, Portugal

⁸School for Mental Health and Neuroscience, Maastricht University, Maastricht, Netherlands

⁹Karolinska Institutet, Department NVS, Center for Alzheimer Research, Division of Neurogeriatrics, Bioclinicum J10:20, Solna, Sweden

¹⁰Karolinska University Hospital, Theme Aging, Unit for Hereditary Dementias, Solna, Sweden

¹¹Deutsche Alzheimer Gesellschaft, Berlin, Germany

¹²Laboratoire de Psychopathologie et Processus de Santé, Institut de Psychologie Université Paris Descartes—Sorbonne Paris Cité, Paris, France

Correspondence

A. Metcalfe, Sorbonne University, Assistance Publique—Hôpitaux de Paris AP-HP, Paris, France.
 Email: anna.metcalfe@aphp.fr

Objectives: The European RHAPSODY project sought to develop and test an online information and support programme for caregivers of individuals diagnosed with young onset dementia. The objectives were to assess user acceptability and satisfaction with the programme and to test outcome measures for a larger effectiveness study.

Design: A pilot randomised controlled trial in England, France, and Germany was conducted with 61 caregivers for adults with young onset Alzheimer's disease or frontotemporal degeneration. Evaluations at baseline, week 6, and week 12 assessed user acceptability and satisfaction. Use of the programme was measured from online back-end data. Qualitative feedback on user experiences was collected via semi-structured interviews. Measures of caregiver well-being (self-efficacy, stress, burden, frequency of patient symptoms, and caregiver reactions) were explored for use in a subsequent trial.

Results: Participants logged in online on average once a week over a 6-week period, consulting approximately 31% of programme content. Seventy percent of participants described the programme as useful and easy to use. Eighty-five percent expressed intent to use the resource in the future. Reductions in reported levels of stress and caregivers' negative reactions to memory symptoms were observed following use of the programme.

Conclusions: Results indicated that the RHAPSODY programme was acceptable and useful to caregivers. The programme may be complementary to existing services in responding to the specific needs of families affected by young onset dementia. Distribution of the programme is underway in England, France, Germany, and Portugal.

Funding information

United Kingdom, Economic and Social Research Council (ESRC); Swedish Research Council (SRC); Fundação para a Ciência e Tecnologia (FCT); The Netherlands Organization for Health Research and Development (ZonMW); Federal Ministry of Education and Research (BMBF); French National Research Agency (ANR)

KEYWORDS

Alzheimer's disease, caregiver support, family caregiver, frontotemporal degeneration, online intervention, psycho-education, randomised controlled trial, technology acceptance, thematic analysis, young onset dementia

1 | INTRODUCTION

Young onset dementia (YOD), defined by first symptoms of cognitive or behavioural decline occurring before the age of 65, is associated with particularly challenging consequences for individuals affected and their caregivers. Although the prevalence of YOD is much lower than that of dementia in later life,¹⁻³ it has a more severe impact on families. YOD is associated with higher frequency of behavioural symptoms⁴; disruption of family relationships, partnership, and intimacy^{5,6}; adverse impacts on children⁷; conflicts between caring roles and family or professional responsibilities⁸; and economic consequences of early retirement or reduced working hours.⁹ Caregivers of people with YOD report high levels of depression, stress, and burden associated with the psychosocial consequences of disease onset.⁹⁻¹¹

People with YOD and their caregivers are an underserved group, because accurate and timely diagnosis as well as appropriate counselling and treatment may be difficult to obtain.¹² Existing health and social care structures for people with dementia rarely meet the needs of this particular group of patients and carers, having typically been designed for older adults.¹³ Services specifically designed for people with YOD are available at only a few centres across Europe,^{14,15} therefore access may be hindered by geographical and logistic barriers. Moreover, a lack of information resources dedicated to younger onset forms of dementia has been identified.¹⁶

Support programmes for informal caregivers have proven to be an effective component of dementia management, enhancing outcomes for both caregivers and care recipients.¹⁷⁻¹⁹ The relative rarity of young onset disease variants, however, presents a logistical challenge to the organisation of in-person groups in a given geographical area, so availability of face-to-face support is sparse. Web-based interventions may present an accessible, low-cost alternative form of support. Advantages of internet-based programmes over traditional formats of carer support include convenience of use, overcoming of geographical barriers, variety of information delivery formats, and low delivery costs.^{20,21}

Web-based multicomponent interventions for dementia caregivers have been reported by users to be educational, convenient, beneficial, and interesting.^{22,23} Such interventions have been shown to reduce caregiver depression and stress,²⁴⁻²⁸ improve illness-related knowledge and self-efficacy,²⁹ and strengthen empathy and understanding.³⁰

1.1 | Online intervention

The RHAPSODY project (Research to Assess Policies and Strategies for Dementia in the Young), a consortia of six European countries

Key points

- Access to appropriate information and support is a challenge for people with young onset dementia and their caregivers, who are particularly impacted by the onset of disease at a younger age.
- An online information and skill-building programme developed specifically for young onset dementia carers was evaluated in a pilot randomised controlled trial with 61 participants to assess user acceptability and satisfaction.
- Overall caregivers reported finding the programme easy to use, useful, and relevant. Qualitative results provided insights into caregivers' user experiences and useful suggestions for future enhancements to the online resource.
- User activity on the programme was moderate (one visit per week on average); results indicate modest improvements in reported stress and negative reactions to patient memory symptoms among caregivers using the programme compared with a wait-list control group after 6 weeks.

(England, France, Germany, Netherlands, Portugal, and Sweden), set out to develop a web-based multimedia, information, and skill-building programme for carers of people with YOD and to evaluate it in a pilot study.¹⁵ The finished programme features seven modules covering the nature of YOD, medical explanations, common problems and solutions, management of cognitive and behavioural symptoms, adapting to relationship changes, available care and support, and self-care suggestions. The multimedia format combines written and video content, case-studies, presentations from professionals, and downloadable materials. The format is based on the successful Resources for Enhancing Alzheimer's Caregiver Health (REACH) II Study.^{31,32} Evidence on care needs was taken from a systematic review,³³ from qualitative interviews conducted as part of the Needs in Young Onset Dementia (NeedYD) Study in the Netherlands,³⁴ and from focus groups with caregivers undertaken in five RHAPSODY countries. The programme was produced in English, French, German, and Portuguese.

This paper presents the results of the pilot evaluation, which aimed to explore acceptability of the programme to caregivers and the potential for a larger trial of effectiveness.

2 | MATERIALS AND METHODS

2.1 | Study design

A pilot unblinded randomised wait-list controlled trial design was used in order to anticipate the methodology of a potential subsequent effectiveness (Technical University of Munich, Germany; Pitié-Salpêtrière Hospital, Paris, France; and University of Surrey, England). It was approved by the relevant ethics committees in each country and registered on the German Clinical Trials Register (DRKS00009891).

2.2 | Inclusion and exclusion criteria

Participants were informal carers aged 18 years or more, supporting someone with symptom onset before the age of 65 and a diagnosis of either Alzheimer's disease (AD) (criteria of McKhann et al.³⁵) or behavioural variant frontotemporal degeneration (FTD) (criteria of Rascovsky et al.³⁶) within three years of recruitment. Dementia cases unrelated to AD or FTD were excluded. Basic computer skills and literacy in English, French, or German were stated as necessary for caregivers.

2.3 | Recruitment, consent, and baseline data collection

In Germany and France, recruitment took place in memory clinics of partner sites. Researchers identified carer-patient dyads meeting eligibility criteria and provided initial details about the study. Where caregivers expressed interest, information sheets were provided and a subsequent appointment arranged for obtaining informed consent and baseline information. In England, the study was promoted via national not-for-profit organisations (the Alzheimer's Society and Young Dementia UK) and listed on the National Institute of Health Research Join Dementia Research database. Carers expressed interest in participating by contacting researchers directly and providing contact details. Eligibility of volunteers was confirmed and full information provided by email. Volunteers were asked to return a consent form in advance of a telephone appointment to collect baseline information. Data gathered at baseline included sociodemographic information on both the caregiver and person with YOD, including diagnosis, symptoms, treatment, health status, service use, and available support.

2.4 | Randomisation

Computer-generated randomisation and closed envelopes were used to assign participants to group A (immediate access to the online programme) or group B (wait-list control with delayed access to the programme).

2.5 | Trial process

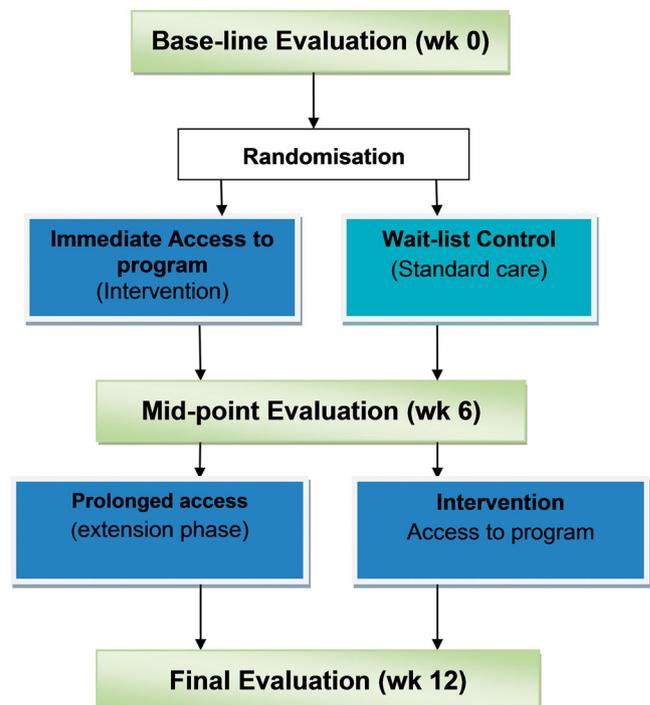
The research protocol design is represented in Figure 1.

Following completion of baseline evaluations, participants in group A (immediate access) in France and Germany were given a short demonstration of the programme on-site and a user guide containing their unique log-in details. Those in group A in the UK were sent the user guide with log-in details and were phoned to make sure they were able to gain access. The programme was made available online 24 hours a day for 6 weeks. In order to assess spontaneous usage, no guidelines on how much or when to use the programme were offered. Contact details for the research team were provided in case of technical difficulties (all sites). After 6 weeks, members of group B (wait-list control) were also given access to the programme, with technical support if needed, for 6 weeks. During this time, group A retained programme access, but no technical support was offered.

Follow-up assessments were conducted at week 6 (midpoint) and at week 12 (final). Assessments were in person at research centres or by telephone, according to participants' preference in France and Germany. All follow-up assessments in England were by phone as participants were widely spread geographically.

2.6 | Outcome measures

Outcome measures were selected to assess user acceptability and satisfaction with the programme, with a view to gaining information for future programme refinement. In addition, outcome measures



Wk : Week

FIGURE 1 Schema of research protocol [Colour figure can be viewed at wileyonlinelibrary.com]

that might reflect the effectiveness of the programme on caregiver well-being were tested to inform the subsequent development of a larger follow-up trial. A list of outcome measures is presented in Table 1.

2.7 | Economic evaluation

Direct costs of delivering the intervention were estimated through research logs kept at each site. These records detailed time spent on inducting participants and providing technical support. The costs of designing and producing the programme were not considered since these were one-off costs that would be discounted as the number of users increased.

2.8 | Sample size, data management, and analysis

The current study was planned as a pilot study and was not powered to explore efficacy. A sample size of 60 participants was considered adequate for a pilot study (30 per group, 20 in each country)^{43,44}. Researchers received training on data entry into an electronic Client

Record Form and resolving data queries. The collective database was locked following data cleaning, before analysis.

Quantitative data were analysed using SPSS Statistics and R version 3.3. The characteristics of participants were compared between the two groups using appropriate statistical tests. Responses to acceptability and satisfaction questionnaires were analysed descriptively. Back-end usage data from the online platform were analysed descriptively to ascertain the number of logins and programme components accessed by participants. Group score changes for caregiver well-being measures from week 0 to week 6 were compared between groups using the Wilcoxon rank sum test (without correction for multiple testing). Results were used to identify a potential primary outcome for a larger follow-up trial.

2.9 | Qualitative analysis

A semi-structured interview guide was developed and translated in all working languages. Interviews were conducted in person or by phone, transcribed or notated, and then translated into English for thematic analysis, taking an inductive approach to the dataset. Two researchers coded all interviews and identified potential themes, which were

TABLE 1 Outcome measures applied at each evaluation point

Evaluation Domain	Measure	Baseline	At 6 weeks	At 12 weeks
User satisfaction and acceptability	Technology Acceptance Model (TAM). ³⁷ Four subscales of this measure were selected and adapted with reference to the online programme: Perceived usefulness, Ease of use, Behavioural intention to use, and Computer self-efficacy.	-	Group A only	Group B only
	Individual semi-structured interviews based on a topic guide regarding user experiences. Topics included satisfaction with programme content and mode of delivery and likelihood of recommending to others.	-	Group A only	Group B only
	User behaviour web metrics from the online platform: number and duration of participant log-ins, percentage of programme reviewed, and number of posts on discussion forum.	-	Group A only	Groups A and B
Caregiver well-being	Revised Scale for Care-giving Self-Efficacy (RSCSE). ³⁸ A 19 item scale covering three domains: obtaining respite, responding to disruptive behaviours, and controlling upsetting thoughts about caregiving.	Groups A and B	Groups A and B	Groups A and B
	Perceived Stress Scale (PSS). ³⁹ A quick-to-administer and widely used 10 item indicator of subjectively evaluated stress with well-established psychometric properties.	Groups A and B	Groups A and B	Groups A and B
	Burden Scale for Family Caregivers (BSFC) 10-item short form. ⁴⁰ Assesses carer's global subjective burden relating to the care of a chronically ill person.	Groups A and B	Groups A and B	Groups A and B
	Revised Memory and Behaviour Checklist (RMBC). ⁴¹ A 24 item self report measure providing 1 total score, and 3 subscores for patients' problems (memory related n=7; depression n=9; disruptive behaviours n=8), and parallel scores for caregivers' reactions.	Groups A and B	Groups A and B	Groups A and B
	EQ-5D-5L. ⁴² A standardised measure of health status in five dimensions (mobility, self-care, usual activities, pain/discomfort, and anxiety/depression) used in health-economic analyses to indicate health-related quality of life and calculate quality-adjusted life years (QALYs).	Groups A and B	Groups A and B	Groups A and B

TABLE 2 Characteristics of participants and care recipients

Participant	Variable	Group A, Immediate Access	Group B, Wait-List Control
Caregiver	Age, mean (SD) in years	57.6 (10.5)	57.2 (9.9)
	Caregiving years, mean (SD)	1.8 (1.1)	2.6 (1.7)
	Females, %	60.0	61.3
	Higher education, %	46.7	41.9
	Full/part time employment, %	50.0	67.7
	Retired, %	23.3	22.6
Person with YOD (care recipient)	Age, mean (SD) in years	61.6 (3.9)	61.9 (5.7)
	Females, %	43.3	54.8
	Diagnosis (AD/FTD), %	56.7/43.3	64.5/35.5
	Years since diagnosis (1-2/3-4/5+), %	26.7/43.3/30	22.6/41.9/35.5

Abbreviations: AD, Alzheimer's disease; FTD, frontotemporal degeneration; YOD, young-onset dementia.

reviewed and refined in collaboration with a third researcher. A thematic map demonstrating relationships between identified themes and subthemes was developed.

3 | RESULTS

3.1 | Characteristics of participants and care recipients

Baseline characteristics of participants and care recipients are shown in Table 2. A total of 61 carers (20 in England and Germany and 21 in France) were randomised (30 to group A, 31 to group B). 58 participants completed the protocol, with one at each intervention site dropping out during the study. There were no statistically significant differences in sociodemographic variables between the two treatment groups.

3.2 | User acceptability

Analysis of four dimensions of the Technology Acceptance Model³⁷ questionnaire (perceived usefulness, ease of use, computer self-efficacy, and behavioural intention) are shown in Table 3. Most participants agreed that they had good levels of computer literacy and found the programme easy to use. Although the mean score for perceived usefulness was moderate (4.7 on a scale of 1 [*strongly disagree*] to 7 [*strongly agree*] that the programme is useful), most respondents agreed strongly that they would use it again (behavioural intention). There was no statistically significant difference between groups on technology acceptance measures.

TABLE 3 Technology acceptance scores

Technology Acceptance Model	Mean (SD)	Range	N
Perceived usefulness	4.70 (1.32)	1-7	55
Perceived ease of use	6.26 (0.93)	1-7	55
Computer self-efficacy	4.79 (1.27)	1-7	55
Behavioural intention	6.06 (1.33)	1-7	55

3.3 | User satisfaction

Feedback on the quality of the programme at the end of the study was generally good. Of 55 respondents, 39 (70.9%) rated it as very good/excellent and 51 (92.7%) rated it as good/very good/excellent. Similarly, for likelihood to recommend to others, ease of understanding, relevance of information, and putting learning to use, responses were generally very positive. Regarding views on the level of detail in the programme, 38 of 55 participants (69.1%) felt the information was just right, 12 (21.8%) thought it was too general, and five (9.1%) thought it was too detailed. Around 80% thought the number of individual sections, duration of sections, and overall length of the programme was about right. See online supplementary information.

3.4 | User behaviour

On average, participants accessed the programme just over once a week (average of 7.5 visits over 6-week access periods) and consulted 31% of its overall content. The number of chapter views decreased progressively from chapter one ("What is young onset dementia?") to chapter seven ("Looking after yourself"). No difference was identified between the two groups in their patterns of use. Following the initial 6-week access period, approximately 60% of group A (immediate access) participants logged on to the programme again at least once.

3.5 | Qualitative feedback

A thematic map (Figure 2) demonstrating relationships between identified themes and subthemes was developed. Thematic analysis identified two major themes relating to participants' experiences of the programme: *Usability* and *Impact*.

Within the theme of *Usability*, subthemes of *Feasibility* and *Functionality* referred to participants' real-life experiences of using the programme in context. Overall qualitative feedback was very positive, stating that the programme was easy to use, featured accessible language, and the online format was helpful.

"It's particularly helpful not having to go somewhere (to receive information)."

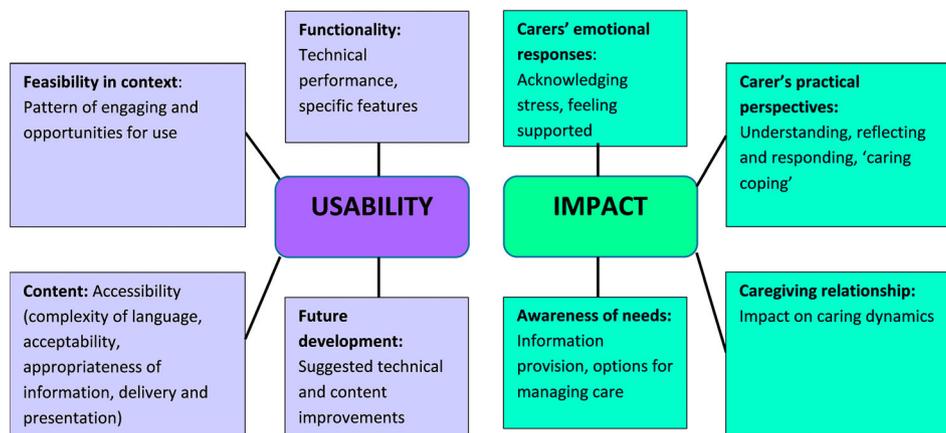


FIGURE 2 Thematic map from qualitative analysis of carers' experiences of using the programme [Colour figure can be viewed at wileyonlinelibrary.com]

TABLE 4 Outcome scores at baseline and midpoint evaluation (N= 61 carers)

Variable	N	Group A, Intervention, mean (SD)		Group B, Wait list control, no access to programme mean (SD)		P value ^a Difference between groups in change scores (week 0 to week 6)
		Baseline Week 0	Mid-point Week 6	Baseline Week 0	Mid-point Week 6	
Revised Scale for Caregiving Self Efficacy (RSCSE)³⁸						
Higher is better self efficacy: Range 0 (certain cannot); 50-60 (moderately certain can); 100 (certain can);						
Obtain respite	56	62.7 (28.4)	59.7 (30.4)	57.5 (31.2)	57.5 (31.7)	0.97
Respond to disruptive behaviour	54	67.5 (22.6)	66.9 (21.9)	73.2 (21.8)	69.3 (17.8)	0.08
Control upsetting thoughts about caregiving	59	63.1 (23.5)	62.1 (22.7)	62.3 (21.9)	66.27 (21.6)	0.41
Caregiver Perceived Stress Scale (PSS)³⁹ Range 0 to 40 (highest stress)						
Overall score	60	17.3 (6.3)	15.6 (8.03)	17.0 (6.3)	18.6 (6.3)	0.03*
Burden Scale for Family Caregivers (BSFC)⁴⁰						
Ten statements rated 0 (strongly disagree) to 3 (strongly agree), Range 0 (lowest) to 30 (highest burden)						
Overall score, from 10 items	60	12.5 (7.8)	11.8 (6.69)	14.9 (7.8)	14.9 (7.3)	0.47
Revised Memory & Behaviour Checklist (RMBC)⁴¹: Patients' symptoms & Caregivers' reactions						
Higher scores are more frequent / more bothersome. Subscore means. Range 0-4.						
Memory problems (7 items) frequency	58	2.4 (1.0)	2.4 (1.0)	2.7 (1.9)	2.9 (1.0)	0.18
Memory problems reactions	57	1.3 (0.7)	1.0 (0.8)	1.5 (1.0)	1.5 (0.8)	0.04*
Depression (9 items) frequency	59	0.8 (0.7)	0.8 (0.7)	1.1 (0.8)	1.0 (0.7)	0.61
Depression reaction	47	1.8 (1.0)	1.7 (1.0)	2.2 (0.9)	2.3 (1.1)	0.72
Disruption (8 items) frequency	59	0.7 (0.6)	0.7 (0.9)	0.6 (0.5)	0.9 (0.7)	0.01*
Disruption reaction	42	2.0 (1.1)	1.8 (1.1)	2.2 (1.0)	2.2 (1.1)	0.66
Total symptom frequency	60	1.3 (0.6)	1.2 (0.6)	1.4 (0.6)	1.5 (0.5)	0.04*
Total symptom reaction	59	1.6 (0.7)	1.3 (0.8)	1.9 (0.6)	1.9 (0.7)	0.11
Caregiver's Health-related quality of life (EQ-5D-5L)⁴²						
Range -0.59 to 1.00 (best possible health related quality of life)						
Overall score	61	0.82 (0.17)	0.84 (0.17)	0.87 (0.16)	0.86 (0.16)	0.28

^aWilcoxon Rank sum test

*Differences are significant with $P < 0.05$

"I liked the convenience of being able to pick it up when I could."

The subtheme *Content* described this aspect as good, comprehensive, and relevant, with many participants stating that they appreciated the practical focus on concrete situations and the use of videos.

"The case studies (videos) with explanations, instead of a theoretical speech, it's a concrete reality, more easy to relate to."

Several participants reported that not all content was relevant to them at the current stage in the condition as either they had already received such information or it was not yet applicable. The subtheme of *Future Development* encompasses the participants' suggestions for enhancing the programme content and functionality. These included adding a keyword search facility, more video content, and videos in the same language as written content rather than using voice-overs to create multiple language versions.

The second major theme *Impact* related to changes resulting from programme use for carers, the caregiving relationship, and for people with YOD. Many participants reported *Emotional Responses* to the online resource such as identifying with case studies, feeling understood or "heard," and an alleviated sense of guilt and isolation. Negative emotional responses were also noted; for some participants, certain aspects of the programme were stressful or worrying to read.

"Confrontation with future stages of the illness was stressful."

Many participants described improvements in the *Caregiving Relationship* due to an enhanced understanding of the care recipient's symptoms.

"There is less conflict in our relationship ... he is less anxious and I am less angry."

"Finding explanations for things has helped."

While the programme appeared to respond to certain needs for many caregivers, barriers to use and limitations of the programme were also identified. Several participants communicated that while helpful, an online resource should not replace the individualised advice that face-to-face consultations with a professional allow. Barriers to programme use included lack of time, lack of opportunity to access the programme due to the presence of the person with YOD, and unwillingness to devote more time to the illness than was necessary.

3.6 | Economic analysis

Running costs for the programme were minimal as the induction process proved sufficient for participants to use the programme with little or no technical support during the study period.

3.7 | Caregiver well-being outcomes

Scores at baseline (week 0) and midpoint (week 6) for group A (immediate access to the programme) and group B (wait-list control/

no access to the programme) are shown in Table 4. Comparison of group score changes from week 0 to week 6 indicate modest but statistically significant changes in favour of the intervention group A, compared with control group B, in the overall score of the Perceived Stress Scale and three items in the Revised Memory and Behaviour Checklist (caregivers' reactions to the patient's memory problems, and the frequency of disruptive behavioural symptoms and of total symptoms in people with YOD). No statistically significant differences in change scores between groups were noted regarding caregiving self-efficacy, caregiver burden, or health-related quality of life (EQ-5D-5L) (Table 4).

Among the measures of well-being tested for possible use in a follow-up trial, the Perceived Stress Scale appeared sensitive to the intervention and was identified as a potential instrument.

4 | DISCUSSION

In response to a lack of dedicated information and support for the caregivers of people with YOD, a multimedia online programme was developed based on research into the information and support needs of this group. The programme covered a range of topics including causes and treatments, common difficulties and suggested strategies, and where to access appropriate care and support. An advantage of the online resource is the ease of access for caregivers from any location at any time.

First testing of this resource with 61 caregivers across three European countries (England, France, and Germany) showed that almost all participants (93%) rated the programme as good, very good, or excellent. Ease of understanding, perceived helpfulness, practical relevance of the information, and willingness to recommend the programme to others was high in all three countries. However, the frequency and intensity of online user activity were moderate. Modules presented later within the programme were visited less, indicating a possible order effect. Qualitative interviews identified that shortage of time and reluctance to engage with difficult aspects associated with progression of symptoms were possible reasons for this. Therefore, an advanced search function and indexing of content may be helpful future refinements, allowing caregivers to identify topics of most interest and avoid those they may find unhelpful.

Previous studies have shown that information and support interventions for caregivers of people with dementia, including web-based approaches, can help reduce carer burden and improve coping skills.^{17-19,22,23} The current evaluation was a pilot study with a small sample and not powered to show effectiveness. However, caregiver well-being outcomes showed some beneficial results with respect to stress and reacting to memory symptoms. These findings should be treated with caution but may point to the Perceived Stress Scale as a potentially sensitive instrument for use in a larger effectiveness trial with sample size calculations based on the parameters from this study.

Strengths of the present study include its value in demonstrating the acceptability of a widely accessible information resource for carers of people with YOD, a traditionally underserved group. It also provided

useful experience to inform the design and conduct of a larger randomised controlled trial of effectiveness. A multinational cohort was recruited using strict diagnostic criteria. Care was taken to ensure that programme content and research procedures were as consistent as possible across the three investigation sites, in each working language. Well-being measures selected not only allowed assessment of outcomes for caregivers but also considered symptoms of people with YOD using the RMBC. Feedback from participants provided useful suggestions for improving the programme, eg, more video content and a keyword search facility.

The study was limited in several respects. The relatively small sample size limited the study's statistical power, and the statistically significant findings in favour of the immediate access group may not necessarily translate to clinically significant outcomes. Confounding variables such as disease progression, changes in family circumstances, and the caregiving situation could not be controlled for. Feedback from the participants direct to the research team may have encouraged socially desirable responding, particularly where participants were recruited from the local clinics. While standardisation of the content was maintained across different language versions of the programme, there were differences in linguistic nuances and presentation when the resource was translated. However, these adaptations might be seen as a strength as they accommodate cultural differences, enhancing the programme's acceptability for use in a range of contexts. Technical design limitations meant that the length of time participants used the programme (overall and by module) could not be recorded, as there was no "time out" setting after a period of inactivity. Hence, it is not known how much time individuals spent engaging with the programme, and patterns of access across content type (eg, video and text) could not be mapped.

The duration of access to the programme was restricted within the pilot study. The true value of an online resource may be through its continuing availability, as some participants highlighted the utility of being able to "dip in and out" of the resource as and when information is needed. There was indication that participants in group A continued to access the programme beyond the first 6 weeks, although no apparent effect on well-being outcomes was observed. A longer intervention and follow-up period in future study designs would allow fuller exploration of acceptability, utility, and effectiveness. Similarly, the economic evaluation was not able to explore the possible impact of the programme in terms of reducing demands made by caregivers and people with YOD on health and social care services because of the short follow-up period. Further evaluation should incorporate these considerations.

5 | CONCLUSION

The results of the pilot study suggest the online information and support programme is acceptable and rated as useful by informal caregivers of people with YOD. It is also inexpensive to deliver. Participant feedback will enhance refinements to the programme before wider dissemination. Experience from the pilot will inform the

design of a future study to investigate effectiveness and possible impact on service use.

ACKNOWLEDGEMENTS

The authors wish to thank the research participants who contributed to the development and testing of the RHAPSODY online resource. Thank you to NIHR Join Dementia Research who assisted with recruitment in the UK. RHAPSODY is an EU Joint Programme—Neurodegenerative Disease Research (JPND) project. The project is supported through the following funding organisations under the aegis of JPND: France, The French National Research Agency (ANR); Germany, Federal Ministry of Education and Research (BMBF); the Netherlands, The Netherlands Organization for Health Research and Development (ZonMW); Portugal, Fundação para a Ciência e Tecnologia (FCT); Sweden, Swedish Research Council (SRC); United Kingdom, Economic and Social Research Council (ESRC). www.jpnd.eu.

CONFLICT OF INTEREST

None declared.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

CLINICAL TRIAL REGISTRATION

German Clinical Trials Register, DRKS00009891.

ORCID

Anna Metcalfe  <https://orcid.org/0000-0002-5727-8914>

Bridget Jones  <https://orcid.org/0000-0001-9709-1794>

Heather Gage  <https://orcid.org/0000-0002-2049-9406>

Jan Oyebode  <https://orcid.org/0000-0002-0263-8740>

Uta Schwertel  <https://orcid.org/0000-0001-5804-5867>

Sophie Tezenas du Montcel  <https://orcid.org/0000-0002-2866-4330>

Said Lebbah  <https://orcid.org/0000-0002-4657-3352>

Alexandre De Mendonça  <https://orcid.org/0000-0002-0488-1453>

Marjolein De Vugt  <https://orcid.org/0000-0002-2113-4134>

Caroline Graff  <https://orcid.org/0000-0002-9949-2951>

REFERENCES

- Harvey RJ, Skelton-Robinson M, Rossor MN. The prevalence and causes of dementia in people under the age of 65 years. *J Neurol Neurosurg Psychiatry*. 2003;74(9):1206-1209.
- Mercy L, Hodges JR, Dawson K, Barker RA, Brayne C. Incidence of early-onset dementias in Cambridgeshire, United Kingdom. *Neurol*. 2008;71(19):1496-1499. <https://doi.org/10.1212/01.wnl.0000334277.16896.fa>
- Vieira RT, Caixeta L, Machado S, et al. Epidemiology of early-onset dementia: a review of the literature. *CP & EMH*. 2013;9(1):88-95. <https://doi.org/10.2174/1745017901309010088>
- Sansoni J, Duncan C, Grootemaat P, Samsa P, Capell J, Westera A. Younger onset dementia: a literature review. *Am J Alzheimers Dis*

- Other Dement.* 2016;31(8):693-705. <https://doi.org/10.1177/1533317515619481>
5. Svanberg E, Spector A, Stott J. The impact of young onset dementia on the family: a literature review. *Int Psychogeriatr.* 2011;23(3):356-371. <https://doi.org/10.1017/S1041610210001353>
 6. Harris PB. Intimacy, sexuality, and early-stage dementia: the changing marital relationship. *Alzheimer's Care Today* 2009; April-June 10: 63-77.
 7. Millenaar J, van Vliet D, Bakker C, et al. The experiences and needs of children living with a parent with young onset dementia: results from the NeedYD study. *Int Psychogeriatr.* 2013;26(12):2001-2010. <https://doi.org/10.1017/S1041610213001890>
 8. Ducharme F, Kergoat M, Antoine P, Pasquier F, Coulombe R. The unique experience of spouses in early-onset dementia. *Am J Alzheimers Dis Other Dement.* 2013;28(6):634-641. <https://doi.org/10.1177/1533317513494443>
 9. Van Vliet D, de Vugt ME, Bakker C, Koopmans RTCM, Verhey FRJ. Impact of early onset dementia on caregivers: a review. *Int J Geriatr Psychiatry.* 2010;5(11):1091-1100.
 10. Rosness TA, Haugen PK, Gausdal M, Gjøra L, Engedal K. Carers of patients with early-onset dementia, their burden and needs: a pilot study using a new questionnaire—Care-EOD. *Int J Geriatr Psychiatry.* 2012;27:1095-1097. <https://doi.org/10.1002/gps.2818>
 11. Freyne A, Kidd N, Coen R, Lawlor BA. Burden in carers of dementia patients: higher levels in carers of younger sufferers. *Int J Geriatr Psychiatry.* 1999;14(9):784-788.
 12. Mendez MF. The accurate diagnosis of early-onset dementia. *Int J Psychiatry Med.* 2006;36(4):401-412. <https://doi.org/10.2190/Q6J4-R143-P630-KW41>
 13. Bakker C, de Vugt ME, Vernooij-Dassen M, van Vliet D, Verhey FRJ, Koopmans RTCM. Needs in early onset dementia: a qualitative case from the NeedYD study. *Am J Alzheimers Dis Other Dement.* 2010;25(8):634-640. <https://doi.org/10.1177/1533317510385811>
 14. Hooghiemstra AM, Eggermont LHP, Scheltens P, et al. Study protocol: EXERCISE and Cognition In Sedentary adults with Early-ONset dementia (EXERCISE-ON). *BMC Neurol.* 2012;12(1):75.
 15. Kurz A, Bakker C, Böhm M, et al. RHAPSODY—Internet-based support for caregivers of people with young onset dementia: program design and methods of a pilot study. *Int Psychogeriatr.* 2016;28(12):2091-2099. <https://doi.org/10.1017/S1041610216001186>
 16. Jones B, Gage H, Bakker C, et al. Availability of information on young onset dementia for patients and carers in six European countries. *Patient Educ Couns.* 2018;101(1):159-165.
 17. Parker D, Mills S, Abbey J. Effectiveness of interventions that assist caregivers to support people with dementia living in the community: a systematic review. *Int J Evid Based Healthc.* 2008;6(2):137-172.
 18. Livingston G, Johnston K, Katona C, Paton J, Lyketsos CG, Old Age Task Force of the World Federation of Biological Psychiatry. Systematic review of psychological approaches to the management of neuropsychiatric symptoms of dementia. *Am J Psychiatry.* 2005;162(11):1996-2021.
 19. Brodaty H, Green A, Koschera A. Meta-analysis of psychosocial interventions for caregivers of people with dementia. *J Am Geriatr Soc.* 2003;51(5):657-664.
 20. Godwin KM, Mills WL, Anderson JA, Kunik ME. Technology-driven interventions for caregivers of persons with dementia: a systematic review. *Am J Alzheimers Dis Other Dement.* 2013;28(3):216-222.
 21. Serafini JD, Damianakis T, Marziali E. Clinical practice standard and ethical issues applied to virtual group intervention for spousal caregivers of people with Alzheimer's. *Soc Work Health Care.* 2007;44(3):225-243.
 22. Boots LMM, de Vugt ME, van Knippenberg RJM, Kempen GIJM, Verhey FRJ. A systematic review of internet-based supportive intervention for caregivers of patients with dementia. *Int J Geriatr Psychiatry.* 2014;29(4):331-344.
 23. Lewis ML, Hobday JV, Hepburn KW. Internet-based program for dementia caregivers. *Am J Alzheimers Dis Other Dement.* 2010;25(8):674-679.
 24. Blom MM, Zarit SH, Groot Zwaafink RBM, Cuijpers P, Pot AM. Effectiveness of an internet intervention for family caregivers of people with dementia: results of a randomized controlled trial. *PLoS ONE.* 2015;10(2):e0116622. <https://doi.org/10.1371/journal.pone.0116622>
 25. Kajiyama B, Thompson LW, Eto-Iwase T, et al. Exploring the effectiveness of an internet-based program for reducing caregiver distress using the iCare stress management e-training program. *Aging Ment Health.* 2013;17(5):544-554.
 26. Beauchamp N, Irvine AB, Seeley J, Johnson B. Worksite-based Internet multimedia program for family caregivers of persons with dementia. *Gerontologist.* 2005;45(6):793-801.
 27. Finkel S, Czaja SJ, Martinovich Z, Harris C, Pezzuto D, Schulz R. E-Care: A telecommunications technology intervention for family caregivers of dementia patients. *Am J Geriatr Psychiatry.* 2007;15(5):443-448.
 28. Schulz R, O'Brien A, Czaja S, et al. Dementia caregiver intervention research: in search of clinical significance. *Gerontologist.* 2002;42(5):589-602.
 29. Cristancho-Lacroix V, Wrobel J, Cantegreil-Kallen I, Dub T, Rouquette A, Rigaud A. A web-based psychoeducational program for informal carers of patients with Alzheimer's disease: a pilot randomized controlled trial. *J Med Internet Res.* 2015; May;17(5):e117. <https://doi.org/10.2196/jmir.3717>
 30. Hattink B, Meiland F, van der Roest H, et al. Web-based STAR e-learning course increases empathy and understanding in dementia caregivers: results from a randomized controlled trial in the Netherlands and the United Kingdom. *J Med Internet Res.* 2015;17(10):e241. <https://doi.org/10.2196/jmir.4025>
 31. Belle SH, Burgio L, Burns R, et al. for the Resources for Enhancing Alzheimer's Caregiver Health (REACH) II Investigators. Enhancing the quality of life of dementia caregivers from different ethnic or racial groups: a randomized, controlled trial. *Ann Intern Med.* 2006;145(10):727-738.
 32. Burgio LD, Collins IB, Schmid B, Wharton T, McCallum D, DeCoster J. Translating the REACH caregiver intervention for use by area Agency on Aging Personnel: the REACH OUT Program. *Gerontologist.* 2009;49(1):103-116.
 33. Millenaar JK, Bakker C, Koopmans RTCM, Verhey FRJ, Kurz A, de Vugt M. 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.* 2016;31(12):1261-1276. <https://doi.org/10.1002/gps.4502>
 34. Millenaar JK, de Vugt ME, Bakker C, et al. The impact of young onset dementia on informal caregivers compared to late onset dementia: results from the NeedYD study. *Am J Geriatr Psychiatry.* 2015;24:467-474.
 35. McKhann GM, Knopman DS, Chertkow H, et al. The diagnosis of dementia due to Alzheimer's disease: recommendations from the National Institute on Aging-Alzheimer's Association workgroups on diagnostic guidelines for Alzheimer's disease. *Alzheimers Dement.* 2011;7(3):263-269.

36. Rascovsky K, Hodges JR, Knopman D, et al. Sensitivity of revised diagnostic criteria for the behavioural variant of frontotemporal dementia. *Brain*. 2011;13:2456-2477.
37. Venkatesh V, Bala H. Technology Acceptance Model 3 and a research agenda on interventions. *Decis Sci*. 2008;39:2.
38. Steffen AM, McKibbin C, Zeiss AM, Gallagher-Thompson D, Bandura A. The Revised Scale for Caregiving Self-Efficacy: reliability and validity studies. *J Gerontol*. 2002; *Series B*;57(1):74-86. <https://doi.org/10.1093/geronb/57.1.P74>
39. Cohen S, Williamson G. Perceived stress in a probability sample of the United States. In: Spacapan S, Oskamp S, eds. *The social psychology of health: Claremont Symposium on applied social psychology*. Newbury Park, CA: Sage; 1988.
40. Graessel E, Chiu T, Oliver R. *Development and Validation of the Burden Scale for Family Caregivers (BSFC)*. Toronto: Ontario Comprehensive Rehabilitation and Mental Health Services; 2003.
41. Teri L, Truax P, Logsdon R, Uomoto J, Zarit S, Vitaliano PP. Assessment of behavioral problems in dementia: the revised memory and behavior problems checklist. *Psychol Aging*. 1992;7(4):622-631.
42. EuroQuol-Group. *EQ-5D-5 L*. Rotterdam: EuroQuol Group; 2009.
43. Lancaster GA, Dodd S, Williamson PR. Design and analysis of pilot studies: recommendations for good practice. *J Eval Clin Pract*. 2004;10(2):307-312.
44. Teare MD, Dimairo M, Shephard N, Hayman A, Whitehead A, Walters SJ. Sample size requirements to estimate key design parameters from external pilot randomised controlled trials: a simulation study. *Trials*. 2014;15:264. <https://doi.org/10.1186/1745-6215-15-264>

SUPPORTING INFORMATION

Additional supporting information may be found online in the Supporting Information section at the end of the article.

How to cite this article: Metcalfe A, Jones B, Mayer J, et al. Online information and support for carers of people with young-onset dementia: A multi-site randomised controlled pilot study. *Int J Geriatr Psychiatry*. 2019;34:1455-1464. <https://doi.org/10.1002/gps.5154>