

Helpful post-diagnostic services for young onset dementia: Findings and recommendations from the Angela Project

Vasileios Stamou¹, Jenny La Fontaine, Mary O'Malley², Bridget Jones, Jacqueline Parkes, Janet Carter, Jan R Oyeboode.

Corresponding author: Jan R Oyeboode, Centre for Applied Dementia Studies, Faculty of Health Studies, University of Bradford, Bradford, BD7 1DP, UK, email: j.oyeboode@bradford.ac.uk, tel.: 0044 1274 236 330

Author affiliations:

Stamou. V., La Fontaine J., Oyeboode, J.R.: Centre for Applied Dementia Studies, Faculty of Health Studies, University of Bradford

O'Malley, M., Parkes, J.: Faculty of Health, Education and Society, University of Northampton.

Jones, B.: Surrey Health Economics Centre, Department of Economics, University of Surrey.

Carter J: Division of Psychiatry, Faculty of Brain Sciences, University College London,

Funding

This work was supported by the Alzheimer's Society [grant number 278 AS-PG-15b-034].

Authors' contribution

JO was involved in study design, analysis, drafting and re-drafting of the article. VS was involved in study design, data collection, analysis, drafting and re-drafting of the article. JLF was involved in study design, data collection and analysis, and commenting on article drafts. MOM, BJ, JP and JC were involved in study design and commenting on article drafts.

Acknowledgements

The researchers acknowledge the support of the National Institute for Health Research Clinical Research Network (NIHR CRN). The research team would also like to acknowledge the significant contribution of Professor Heather Gage, University of Surrey, who conducted analysis of the socio-demographic data; the members of our Patient and Public Involvement

Group and Steering Committee; the NHS and charitable organisations and Join Dementia Research which facilitated study recruitment.

Data availability

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

Disclosure of interest

We have no conflict of interest to declare.

¹ Now at Department of Psychology, Manchester Metropolitan University

² Now at Department of Psychology, University of West London.

Abstract

There is a significant lack of evidence regarding optimum models for service provision in young onset dementia. Our study aim was to gather detailed information about services experienced as helpful by those with young onset dementia and family carers. The objective was to identify the core features of these services to inform service design, delivery and improvements. A qualitative approach based on appreciative inquiry was used, posing open-ended questions about services experienced as helpful, as part of a national UK survey of people with young onset dementia and carers. We used inductive thematic analysis to analyse the free-text responses. The resulting template was used as a basis for analysis of in-depth follow-up interviews, conducted to acquire greater in-depth understanding.

Two hundred and thirty-three survey respondents provided 856 examples of helpful support. Twenty-four follow-up interviews were conducted (two with dyads, so 26 participants in total: 8 with people with young onset dementia, 14 with carers, two with dyads). Twelve themes capturing the features of helpful services were clustered into three super-ordinate themes. ‘Person-centredness’ reflects micro-levels of person-professional interaction (positive attitude, flexibility, collaborative, user-friendly materials, in-person). ‘Functional consistency’ captures the meso-level, demonstrating that services were helpful when organised consistently with needs (age-appropriate, holistic, responsive, accessible). ‘Organisational coherence’, at the macro-level, emphasises the need for service integration, specialist services and service continuity.

Key conclusions are: that the needs for flexibility and a collaborative stance may be particularly important for those under 65 years with dementia, who have full lives and are used to being in control; to be age-appropriate, helpful services need to provide activities and

opportunities suitable for active middle-aged people; and to be holistic, services need to provide for needs associated with rare dementias and be family-centred. Specialist services need to be commissioned and arrangements need to be stable over time to enable continuity.

What is known about this topic

- People with young onset dementia have distinctive needs.
- Most dementia services are ‘all-age’ and do not provide well for people with young onset.
- There is little UK research evidence on what sort of services are helpful.

What this paper adds?

- People with young onset dementia:
 - find person-centred interaction helpful: when staff have a positive attitude, are collaborative and flexible, and provide in-person support and user-friendly information.
 - find services helpful when they take younger age into account, consider the whole family, are accessible, and respond proactively to changing needs.
 - find service systems helpful when there is specialist young onset dementia provision, continuity over time and integration across services.

Keywords: younger people with dementia, carers, qualitative research, service delivery and organisation, person-centred planning

Introduction

Young Onset Dementia (YOD) refers to dementia with onset under 65 years (Draper & Withall, 2016). Approximately 42,500 people live with YOD in the UK (Prince et al., 2014), accounting for 5-9% of dementia prevalence. YOD is significantly different from dementia with late onset (LOD), due to the earlier stage of life and higher prevalence of rarer types (Vieira et al., 2013), resulting in distinctive needs. Those with YOD need active ways to continue involvement and contribution to life; support to manage difficulties associated with vision, communication, social cognition and movement; advice on employment and finances; and support for families with young people (Chaplin & Davidson, 2016; Mayrhofer et al., 2018; Millenaar et al., 2016; Roach, Drummond, & Keady, 2016; Stamou et al., 2020a).

Despite the distinctive needs, specifically-commissioned YOD services are few and short-term commissioning means many are later discontinued (Cations et al., 2017; Mayrhofer et al., 2018). Most people with YOD who receive post-diagnostic support, access this through all-age dementia services (Stamou et al., 2020b), with the exception, in the UK, for dementia caused by Down's syndrome, traumatic brain injury, HIV, Huntington's disease or alcohol-related dementia, whose services are usually provided through other specialist services. People with YOD are not well served by most 'all-age' services, which are geared towards older people with amnesic symptoms, resulting in lack of support for those with rarer dementias and provision of therapeutic activities that do not suit younger generations (Cations et al., 2017; Giebel et al., 2020a; Mayrhofer et al., 2018; Stamou et al., 2020b; Sansoni et al., 2016). Sansoni et al. (2016) report a catalogue of shortcomings: lack of a care pathway, poor provision of information, unmet needs for social companionship, day-time activities and lack of help regarding intimate relations, behavioural issues and

communication. This contributes to lower quality of life (Bakker et al., 2010; Baptista et al., 2016; Chemali et al., 2012), and potentially preventable service costs (Kandiah et al., 2016).

There is little evidence regarding optimum service provision (Carter, Oyebode & Koopmans, 2018). In Australia, Westera and Fildes (2014)'s literature review and stakeholder consultation suggested services should be individualised, have staff with seven key attributes (e.g. flexibility) and care organisations should fulfil 12 key attributes (e.g. timely provision). Also, in Australia, Hutchinson et al. (2018) consulted people with YOD, families and care providers and concluded provision should be family-centred. In the UK, Mayrhofer et al. (2018) consulted 11 people with YOD, 20 family members and 10 service providers about service and support priorities, recommending community hubs would provide a good basis for provision.

Knowledge about what works in practice is crucial to inform services. Appreciative Inquiry (AI; Cooperrider & Srivastva, 1987), originally developed to create ideas for organisational change by focusing on what works well, is consistent with solution-focused research aims. McCarthy (2017) suggested AI as potentially useful for resolving care issues usually labelled as demonstrating 'behavioural and psychological symptoms'. He illustrated the application of all four phases (discovery, dream, design, destiny) of AI to find a helpful solution in a fictional case. Scerri, Innes and Scerri (2015) applied an adapted version of AI using only the discovery phase, to establish care processes that led to positive experiences for people with dementia on in-patient wards; then built on this, through staff workshops addressing the remaining phases, to gain improvements in care (Scerri, Innes & Scerri, 2019).

Bearing in mind the lack of evidence on the nature of positive services, and the importance of hearing the voices of people with YOD (Brown et al., 2012; Johannessen & Muller, 2013; Mayrhofer et al., 2018), we aimed to contribute to the literature on how to

provide effective YOD services by using an adapted AI approach to gather and analyse examples of helpful service support, from people with YOD and family carers.

Design and Methods

We used a qualitative design based on the *discovery* and *dream* phases of AI. The discovery phase involved gathering and analysing data from people living with YOD and carers to understand what works well in support services; and the dream phase involved interpreting these findings to inform recommendations for good practice.

Recruitment

Following ethical approval (South Central Berkshire Research Ethics Committee, REC ref.: 17/SC/0296), we recruited through 14 English National Health Service (NHS) sites, purposively selected to include breadth of diagnostic pathways (YOD-specialist, neurology, all-age dementia services), Join Dementia Research (a UK initiative encouraging people to register interest in research participation) and a wide range of charitable organisations. The study was exploratory and aimed for maximum recruitment.

Participants

Those with dementia needed to have received their diagnosis before the age of 65 and could have any dementia diagnosis, defined by DSM-5 (American Psychiatric Association, 2013), except for those dementia types usually served by other specialist services (see the introduction. Diagnosis was confirmed by services for those recruited via NHS sites and by self-report for others. Carers could be any age and needed to self-identify as a ‘family member/supporter’ of a person with YOD, with details gathered about the amount of care provided (see Stamou et al., 2020b for details).

Data Collection

Survey

We used a cross-sectional nationwide survey, to gather socio-demographic data, information on service use and satisfaction, and experiences of helpful support, between August 2017 and September 2018. The survey was developed by the research team. It was piloted twice using cognitive interviews (Willis, 2004) with members of the Patient and Public Involvement (PPI) Group to refine wording, content, length and format. This article reports on the analysis of helpful support services, with other aspects reported elsewhere (Stamou et al., 2020a; 2020b)

Participants were provided with briefing notes, followed by questions (table 1). The briefing notes were developed with the PPI group to help participants to recall their experiences and gave examples of general outcomes that might be helpful. Questions were presented in pairs, combining a closed question, to direct attention to a time-period, and an open question asking for a free-hand description of “when the help and assistance you received was helpful”. The questions addressed three time periods: (i) between seeking help and gaining a diagnosis, (ii) during the year after diagnosis and (iii) between the first-year post-diagnosis and the present. Participants were invited to give one or more examples for each time period, and to give examples about both the experience of the person with YOD and those of families/carers.

Table 1 ABOUT HERE

The survey was made available through Bristol Online Surveys (<https://www.onlinesurveys.ac.uk>) and on paper. It could be completed by people with YOD, alone or with help, or by a carer alone. Capacity to consent was assumed for survey self-completions. Where a researcher assisted, via Skype, phone, or face-to-face, capacity was

assessed and written consent obtained. A list of national sources of support was provided to all participants.

Interviews

Semi-structured follow-up interviews were conducted with purposively selected survey respondents, to acquire greater in-depth understanding. Sampling was based on the need for further exploration of particular survey themes and to capture diverse voices, e.g., of those with rarer dementias, from minority ethnic communities or living alone. Interviews took place from September 2018 to January 2019. No participants were deemed to lack capacity to consent and written consent was obtained from all.

The interview schedule (Table 2) was developed by three researchers in consultation with the PPI Group. To accommodate different degrees of cognitive impairment, it included two different approaches. Which approach was taken was determined through a prior telephone conversation with each participant and depended on whether the person appeared to recall the survey. A direct approach, focused on in-depth discussion of participants' survey responses, was followed with those who were able to recall their survey responses. For others, a more open approach was employed, that avoided participants having to recall their survey responses, by focusing on discussion of what was important in the participant's life now and on forms of support that helped to meet related needs.

Table 2 ABOUT HERE

Analysis

Survey

Socio-demographic data were analysed using SPSS 23. Inductive thematic analysis (Braun & Clarke, 2013) was used to analyse the free-text responses. Emergent codes were

clustered into three groups concerning *what* was provided, *how* it was provided and *why* it was helpful. This article reports on the *how* codes (see (Stamou et al. 2020a) for an analysis of *what* codes). Manual coding was carried out by two researchers (VS, JLF) after repeated reading of the data, with coding consistency achieved via constant comparison and research team discussions. The codes were then clustered by consensus among the researchers (VS, JLF, JO) into sub-themes and themes, based on relevance and similarities in meaning. Two researchers (VS, JLF) further revisited the dataset to confirm that the final themes accurately captured participants' accounts.

Interviews

Interviews were audio-recorded, transcribed verbatim, anonymised and analysed via template analysis (Brooks et al., 2015), using the survey themes as the initial template. Meaningful segments were labelled using the template and new emergent codes were added to existing themes or clustered into additional themes. Interview analysis took place alongside data collection. Initially, three co-authors (VS, JLF, JO) independently read three transcripts and engaged in preliminary coding, leading to modification of the template, which was then applied to successive sets of three interviews. Further refinements were made by consensus as necessary, following coding of each set. Interviews and analysis continued until data saturation was reached. The final template was then re-applied to the whole dataset by VS to check no further refinements were required. The inductively-derived themes addressed the *discovery* phase of AI, which appreciates what is done well.

Derivation of superordinate themes

In the *dream* phase of AI, we considered the themes from the perspective of their implications for YOD service provision. A researcher (VS), and two experienced clinical-academic researchers (JLF, JO), interpreted the implications of each theme for service provision and a set of superordinate themes was agreed through discussion and consensus.

Quality assurance

Specific quality criteria for qualitative research are contested (Bryman et al., 2008). However, to enhance confirmability, we paid attention to reflexivity through keeping detailed reflexive notes and holding regular team discussions and coding seminars. We established credibility by discussing the themes with the PPI group, wider research team, Steering Committee and external audiences to obtain feedback on plausibility.

Results

Of 233 survey respondents, 40 did not provide examples of helpful post-diagnostic services, because there were none or they had only been recently diagnosed. The remaining 193 gave 856 examples between them (see Stamou et al., 2020b) for socio-demographic details). The survey coding template was finalised after analysis of 502 examples (appendix 1), as thematic saturation had been reached, confirmed via coding of the remaining examples, which did not reveal new meanings or themes (Bowen, 2008). Twenty-four follow-up interviews took place: eight with people with YOD, two with dyads and nine with carers using the open approach, and 5 with carers using the semi-structured approach (see table 3 for their socio-demographic data). As two interviews were with dyads there were 26 participants in total.

Table 3 ABOUT HERE

Analysis led to twelve themes, grouped into three superordinate themes reflecting the level at which they could be best enacted in practice: ‘Organisational coherence’, ‘Functional consistency’, and ‘Person-centredness’ (see Table 4). Figure 1 gives a diagrammatic representation, with the superordinate themes presented as reflecting macro, meso and micro levels of practice. Each superordinate theme and its associated themes is described below.

Table 4 and figure 1 ABOUT HERE

Organisational coherence

This superordinate theme incorporates three themes with implications for overall coherence of YOD services: The helpfulness of integrated services, of care being managed in specialist services and of service consistency over time.

The helpfulness of integrated care was reflected through examples of collaboration between services and through the value placed upon having a tailored, coherent care pathway. Participants stressed the significance of collaboration between different arms of the NHS. One dyad, in their joint response, wrote: *“During this period, we moved to 'shared care' between Memory Clinic and GP. They liaise and co-ordinate in terms of our joint care and very much treat us as a couple who both need support and help.”* [Survey Participant (SP) 128]. A carer wrote about the value of contact between mental and physical health services: *“The assigned dementia nurse was able to meet with my husband’s diabetic nurse, so that they both had a better understanding and the approach is 'joined up' [...]. It was very helpful.”* [SP79]. These responses illustrate the value placed on inter-organisational collaboration.

Participants stressed the value of a tailored care pathway, often facilitated by a designated professional who helped the person with YOD and/or the carer to navigate a relevant pathway through services. A carer explained:

Our link worker, that one person that was immediately designated to be with us [...]. [Link worker] was able to offer the information at the right time. She offered us [access to] carer’s education, to a psychologist for me, and a counsellor for [person with dementia]. If we had to go through the GP for this, it would have been a long wait. [Interview Participant (IP) 21]

In this extract, the link worker introduced this dyad to information, services and professionals, steering their route through an appropriate timely pathway as their needs changed.

Participants emphasised the benefits of YOD specialist services. A carer wrote: *“Once we reached the Young Onset [locality] team everything changed for the better. The emotional support and care by the local young onset community team was great.”* [SP42], and a carer, in a dyad interview, spoke of his discontent at the prospect of his wife being discharged to primary care:

They are trying to move this special care that my wife needs into the GP and that’s just not going to work. [...] It’s lack of experience, it’s lack of understanding, they just can’t give the right advice, you need specialist professionals in this place to give people the right advice [...], you don’t get that from a GP. [IP17]

Here, the husband stresses the need for specialist care from professionals experienced in YOD.

The majority underlined the importance of service consistency across time. A person with YOD stated: *“My strongest and most reliable professional support has come from [charity]. These are the people who have shown true concern and have remained interested in my physical and mental well-being and have been available for me to talk to.”* [SP157] and a carer wrote:

When my husband retired most of his friends forgot him. [...] [Dementia Companion] started by just going for walks with my husband, but these gradually morphed from a few hours to complete days. [...] He became his best friend and, 7 years later, still is. [...] This is the single most thing that has affected us both and I cannot stress this enough. [SP1]

These quotes with their emphasis on services ‘remaining’ interested and ‘still’ being involved, show the value placed on continuity over time.

Functional consistency

This super-ordinate theme groups together four themes (age-appropriate, holistic, responsive, accessible) related to the way helpful services were organised and delivered to meet the needs of those affected by YOD.

Participants emphasised the significance of services that provided age-appropriate support:

It's important to get support and care that is age-appropriate; when you find people who are willing to talk to you on a level appropriate to your needs instead of trying to apply your needs to fit old people services. [IP5]

This quote illustrates the relief of finding services that fitted a younger age rather than having to accommodate to old age services. Activities that were age-appropriate were often quite adventurous, including canoeing, cycling and, in this example, boating: “(Professional) even organised a boat trip [...] [they had] a specially designed boat [...]. It's something different that takes people out of themselves and it's another relaxing thing that you can do.” [IP18]

Participants appreciated a holistic approach that addressed all aspects of this multi-faceted condition and all members of affected families. One couple wrote:

[Charity] has been amazing. [...] [They] provide an 'all family' service. They have provided practical support and advice for the family as a whole. They have put on workshops for the person affected and for the carer to give useful information around the condition and practical issues to consider. They also provided an evening with a solicitor to explain some of the legal considerations. The children have been given the opportunity to learn about the illness in a friendly environment while taking part in 'fun' stuff. [...] Without them, it would be very difficult. [SP50]

It is apparent that this service was valued due to its provision for different aspects of the condition, as well as addressing the needs of all family members, including children.

Participants also emphasised the responsiveness of helpful services, through providing timely responses as needs changed:

I think in the very beginning we got three hours a week of a support worker [...]. The help was for [person with YOD], but it was such a huge benefit to me as well. [...] As the disease progressed the amount of support hours increased. [...] Having that level

of support coming in very early on, it prepares the pathway for accepting help [...] it just seemed natural, it didn't seem like I was putting my hands up and saying I am failing. [IP21]

In this example, the carer emphasises consistency and responsiveness, and expresses appreciation of the service proactively stepping up the level of support.

Participants highlighted the importance of services being accessible, in terms of being local, available when needed, provided in dementia-friendly ways and affordable. A person with YOD described the importance of local services:

On the whole, I feel I am lucky in the area I live as I have heard stories from people from other areas who are not as fortunate as me. [Local YOD services] staff are amazing. [...] I would not be where I am emotionally or mentally without them. [SP67]

One person with YOD implied that being able to call on the service as needed gave a sense of security, stating: *"We could call our memory clinic at any time and there would be someone on the other end of the line to help with any information we needed"* [SP55]. Carers stressed the need for services to be at an appropriate time, as employment could otherwise prevent them from attending. A carer said: *"[There are] carers support networks that I could go to, but they're all during the day, and I'm 51 and I've got a mortgage to pay, and we've lost my husband's income. [...] I can't go to things during the day."* [IP19]. Affordability was frequently mentioned. A carer said: *"I found a weekly exercise class specifically for people with dementia. [...] My husband can work hard and achieve aerobic exercise without fear of falling. [...] We are lucky because we can afford the £3 entrance fee."* [SP30]

Person-centredness

The third superordinate theme contains five themes (positive attitude, collaboration, flexibility, in-person support, user-friendly material) that captured aspects of person-centred interaction with care providers.

Professionals were seen as being helpful when they conveyed a respectful, compassionate, attentive and genuine attitude. Participants valued interactions with respectful professionals. A person with YOD explained:

[I value] people who are willing to listen and give me credit. [...] I still know my own mind, I can make my own choices, and to feel like I am in partnership with my own treatment and support rather than just being doled out. [IP5]

In this example, it is apparent that the participant appreciated being treated as an active partner in treatment. A carer described:

My husband has been attending the [Hospital Balance Centre]. The physiotherapist understands the effects of frontotemporal dementia well. She has made a good bond with my husband. [...] [She] treats him with respect and gives him a set of exercises tailored to his needs which have helped him to gain confidence and have kept him walking. [SP30]

She illustrates how a respectful approach enabled a trusting relationship to develop. A compassionate attitude was also valued. A carer said: *“The support worker [...], the understanding and the compassion that they brought into my house got into our lives and the whole family’s life” [IP21]*, giving a sense that the support worker’s attitude enabled her to provide huge emotional support to the heart of this family. Similarly, a person with YOD described a sense of warmth from staff, which seems to have been very important to their well-being:

After the diagnosis, [professional] came out from [YOD NHS service] and the next week I went. I was nervous about going. [...] The staff were friendly and helpful, lovely. They care and if I needed a hand, they always help me. They kept me going. [SP159]

Participants also found it helpful when professionals took time to listen attentively to expressed concerns. A person with YOD stated: *“Professionals were great listeners and gave good explanation and advice.” [SP163]*, and a dyad wrote: *“Our GP and hospital consultant [...], give time when [person with dementia] has any difficulty. We never feel rushed.” [SP128]*. Participants spoke of appreciating genuineness conveyed via professionals’ honesty.

A person with YOD stated: *“The best help I got was an honest and frank discussion with the*

psychiatrist [...]. I said, 'Cards on the table, let's be honest here'. So, facts were best and that's how I moved forward." [SP151]. In other cases, professionals went beyond what was expected to ensure needs were met. A dyad wrote: "[The] local GP and pharmacist, [they] go the extra mile if needed. The pharmacy sort out any problems with medication. [...] [They] reduced waiting times and any uncertainty about what to do." [SP181]

These positive attitudes appeared to lay the foundation for therapeutic collaboration, expressed here by a carer:

The social worker assigned to us began to think creatively about the care needed rather than following the normal path associated with an Alzheimer's diagnosis. We are forging a new path together to think more creatively about [our] care needs. This allowed us to put in place care funding. [SP78]

The use of the word 'we' in this excerpt is significant, indicating the sense of partnership and the use of the expression 'forging a new path together' shows a strong sense of collaboration.

Descriptions of tailored support often referred to professionals' flexibility as they adjusted their approach to the individual, illustrated in the written response from a person with YOD:

I did not want to go to [peer support] groups. [...] I had outreach visits twice weekly. We talked about diagnosis and response from family. Staff came to house originally, then we met for coffee to talk, alternating with [peer support group] fortnightly. [SP163]

Seeing professionals in-person, rather than online, was preferred, as personal contact generated a stronger sense of connectedness, especially when related to social support. A carer explained:

I could look on the internet and look at a website that let you chat with other people, but it's not the same as talking to somebody and meeting the other person with dementia as well. That's very important, I like meeting others. [IP18]

Finally, participants appreciated the use of clear, concise, understandable user-friendly material. A carer said:

It was pretty difficult and swamped with information about where to go and what to do. [...] It was just confusing, there was no single place, no kind of focus of where to go to actually get everything I needed. [...] I met this guy from [charity], he was very good. [...] Best way to describe it is he was a signpost service, he said, 'If you want this you go there'." [IP17]

Discussion

In this study, we used an adapted version of AI. Through the *discovery* phase, we inductively developed twelve themes about the nature of helpful YOD services and, through the *dream* phase, we interpretively grouped these into superordinate themes at micro, meso and macro levels, to draw out practice implications. These levels have been widely applied to conceptualise facilitators and barriers to successful service delivery, including in dementia care (Meiland, Dröes, De Lange, & Vernooij-Dassen, 2004; van Mierlo, Meiland, van Hout, & Dröes, 2014).

The micro-level relates to the person-centred nature of helpful interaction between professionals and people with YOD/carers. The helpful facets of positive attitude that we identified correspond closely to those that have been found important to people of all ages with dementia, resonating with the theory of person-centred dementia care (Brooker, 2019) the human-rights approach (Cahill, 2018) and the dimensions of a core outcome set for assessing the quality of psychosocial community-based interventions (Reilly et al. 2020). It is therefore not surprising that these qualities emerged as central in our study or that they have been found within other YOD research (Johannessen & Muller, 2013; Westera et al., 2016). Possibly more particular to the YOD population was the emphasis on flexibility, also identified in Australian studies (Brown et al., 2012; Cations et al., 2017; Westera et al., 2016). This may be especially important to those under 65 years, who usually have many ongoing responsibilities, varied needs and ambivalence about receiving support, due to stigma attached to untimely experience of dementia. Collaborative partnerships between those with YOD/carers and professionals, also identified as important by Mayrhofer et al.

(2018) and Johannessen and Müller (2013), may equally be particularly valued by those in middle-age, who have not yet reached an age when they are reconciled to relinquishing control to younger generations. Support delivered via a collaborative, continuing relationship has been found in previous research to engender trust and prevent isolation (Roach & Drummond, 2014) and enables services to be responsive to changing needs (Carone, Tischler & Dening, 2016). The value placed on in-person support was unexpected and has become particularly noteworthy at this time of the Covid-19 pandemic (British Psychological Society, 2020; Giebel et al, 2020b) when social distancing has led to the suspension of social groups and activities.

At meso-level, we grouped aspects of support that reflected functional aspects of services. The need for age-appropriate, holistic support has been stressed over many years in previous YOD research (Beattie et al., 2002; Hutchinson et al., 2020; Mayrhofer et al., 2018) and the needs we identified for services to be responsive and accessible have also been previously reported (Bentham & La Fontaine, 2005; Brown et al., 2012; Cations et al, 2016; Mayrhofer, et al., 2018; Westera et al., 2014). However, only two studies were UK-based of which one was localised (Bentham & La Fontaine, 2005) and the other had relatively small samples (Mayrhofer et al, 2018). Our study therefore amplifies existing knowledge by virtue of the large set of examples provided directly by people with YOD and carers.

At macro-level, our themes are consistent with valued organisational attributes in the Australian context (Westera et al., 2014). Other findings of the Angela project highlight that specialist services are not only subjectively valued but also outperform other service delivery models on quality indicators (Stamou et al., 2020b). Consistency of professional and team input was very highly valued by participants, who usually require support over a number of years. Yet this can be interrupted by service re-organisations due to short term funding or commissioning (Mayrhofer et al., 2017) or be prevented when only short-term follow-up is

commissioned from memory services (Giebel et al., 2020a). While this factor could have been placed at meso-level, we included it at macro-level as, without long-term commissioning, it is not possible to have the stability needed for service consistency over time.

Limitations

Our survey carried a risk of selection bias, towards educated participants in contact with services. Despite targeting recruitment towards Black, Asian and Minority Ethnic (BAME) communities via social media and specialist third-sector organisations, we were only able to recruit a few participants from BAME communities. The briefing notes, used in the survey, may have primed recall of particular features of services. However, the notes were general and the majority of participants' responses were specific and detailed. Additionally, no one under 50 years when diagnosed was interviewed, although YOD prevalence is higher in the 50s than at earlier ages (Vieira et al., 2013).

Implications

The YOD-related essence of our themes may become diluted when raised to high level labels. However, the first-hand words and examples demonstrate that these vital service attributes apply distinctively in the context of YOD. The findings bring research to a point where the qualities needed to provide helpful YOD services are now well established. Future research needs to move beyond rehearsing what is needed to co-production and evaluation of interventions. Our envisioning of changes needed at each of three levels has something in common with the circles of influence outlined by Westera et al. (2014). Our findings imply that at the level of professional development, it would improve service quality to develop and test interventions to ensure staff in all-age services have the awareness, knowledge and competence needed to enact the flexible collaboration that is required to provide for those affected by YOD. Such research would help to operationalise the recommendations of the

UK National Institute for Clinical Excellence Guidelines (NICE, 2018) to involve people with YOD and carers in decisions about their care and would be consistent with NICE emphasis on the importance of staff having YOD-specific training. Additionally, at the present time of social restrictions due to covid-19, and given our findings show the central importance of continuity and in-person support, services are challenged to find new ways of maintaining contact with those with YOD, for example via the Internet. At the meso-level, future research could focus upon co-producing and evaluating service improvements based on complex interventions that ensure YOD services are age-appropriate, holistic, responsive and accessible; e.g. testing the impact of family-centred approaches. Our study emphasises that long-term commissioning is needed to give stability to those with YOD, given its long-term progressive nature, and that commissioners need to find ways of achieving service integration (Department of Health, 2019). People affected by YOD find specialist services particularly helpful. Such services have been recommended (RCPsych, 2018) and would provide a gold-standard service equivalent to specialist services for other rare conditions, such as Motor Neurone Disease (NICE, 2016). This has been recognised in the Netherlands (Carter, Oyebode & Koopmans, 2018) and should be emulated in the UK.

Conclusions

To the best of our knowledge, this is the first large-scale UK study to report on specific features of helpful services from the perspective of people with YOD and carers. Over the past five years, research has moved from identifying needs in YOD to considering what needs to change to meet these needs. Our findings consolidate and expand on this work. The AI approach allowed us to take inductively-derived themes and use them to envision the implications for how they could be enacted, leading to a three-tiered set of recommendations for good practice at the levels of service design, function and delivery.

References

- American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders*. Arlington: American Psychiatric Publishing.
- Bakker, C., de Vugt, M. E., Vernooij-Dassen, M., Van Vliet, D., Verhey, F. R., & Koopmans, R. T. (2010). Needs in early onset dementia: A qualitative case from the NeedYD study. *American Journal of Alzheimer's Disease & Other Dementias*, 25(8), 634-640. doi: 10.1177/1533317510385811
- Baptista, M. A. T., Santos, R. L., Kimura, N., Lacerda, I. B., Johannessen, A., Barca, M. L., ... & Dourado, M. C. N. (2016). Quality of life in young onset dementia: an updated systematic review. *Trends in Psychiatry and Psychotherapy*, 38(1), 6-13. doi: 10.1590/2237-6089-2015-0049
- Beattie, A., Daker-White, G., Gilliard, J. and Means, R. (2004). 'How can they tell?' A qualitative study of the views of younger people about their dementia and dementia care services. *Health and Social Care in the Community*, 12, 359–368. doi: 10.1111/j.1365-2524.2004.00505.x.
- Bentham, P., & La Fontaine, J. (2005). Services for younger people with dementia. *Psychiatry*, 4(2), 100-103. doi: 10.1016/j.mppsy.2007.12.008
- Bowen, G. A. (2008). Naturalistic inquiry and the saturation concept: A research note. *Qualitative Research*, 8(1), 137-152. doi: 10.1177/1468794107085301
- Braun, V., & Clarke, V. (2013). *Successful qualitative research: A practical guide for beginners*. Sage.
- British Psychological Society (2020) *Guidance for health professionals supporting groups with specific complex needs who are or have been shielding*. BPS: Leicester.
Retrieved from:
<https://www.bps.org.uk/sites/www.bps.org.uk/files/Policy/Policy%20-%20Files/Guidance%20for%20Health%20Professionals%20Supporting%20Groups%20with%20Specific%20Complex%20Needs.pdf>
- Brooker, D. (Ed.). (2019). *Dementia reconsidered revisited: The person still comes first*. London: Open University Press.
- Brooks, J., McCluskey, S., Turley, E., & King, N. (2015). The utility of template analysis in qualitative psychology research. *Qualitative Research in Psychology*, 12(2), 202-222. doi: 10.1080/14780887.2014.955224
- Brown, J-A., Sait, K., Meltzer, A Fisher, K.R., Thompson, D., & Faine, R. (2012) *Service and Support Requirements of People with Younger Onset Dementia and their Families*, for NSW Department of Family and Community Services, Ageing, Disability and Home Care, Sydney.
- Bryman, A., Becker, S., & Sempik, J. (2008). Quality criteria for quantitative, qualitative and mixed methods research: A view from social policy. *International Journal of Social Research Methodology*, 11(4), 261-276. doi:10.1080/13645570701401644
- Cahill, S. (2018). *Dementia and human rights*. Policy Press.
- Cations, M., Withall, A., Horsfall, R., Denham, N., White, F., Trollor, J., . . . 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
- Carone, L., Tischler, V., & Dening, T. (2016). Football and dementia: A qualitative investigation of a community based sports group for men with early onset dementia. *Dementia*, 15, 1358–1376. doi: 10.1177/1471301214560239
- Carter, J. E., Oyebode, J. R., & Koopmans, R. T. C. M. (2018). Young-onset dementia and

- the need for specialist care: a national and international perspective. *Aging & Mental Health*, 22(4), 468-473. doi: 10.1080/13607863.2016.1257563
- Chaplin, R., & Davidson, I. (2016). What are the experiences of people with dementia in employment?. *Dementia*, 15(2), 147-161. doi:10.1177/1471301213519252
- Chemali, Z., Schamber, S., Tarbi, E. C., Acar, D., & Avila-Urizar, M. (2012). Diagnosing early onset dementia and then what? A frustrating system of aftercare resources. *International Journal of General Medicine*, 5, 81. doi:10.2147/IJGM.S26523
- Cooperrider, D. L., Srivastva, S., Woodman, R. W., & Pasmore, W. A. (1987). Research in organizational change and development. *Research in Organizational Change and Development*, 1, 129-169. doi:10.1108/S0897-301620170000025003
- Department of Health. (2019). The NHS Long Term Plan. Retrieved from www.longtermplan.nhs.uk
- Draper, B., & Withall, A. (2016). Young onset dementia. *Internal Medicine Journal*, 46, 779-786. doi: 10.1111/imj.13099
- Giebel, C., Eastham, C., Cannon, J., Wilson, J., Wilson, J., & Pearson, A. (2020a). Evaluating a young-onset dementia service from two sides of the coin: staff and service user perspectives. *BMC health services research*, 20(1), 1-7.
- Giebel, C., Cannon, J., Hanna, K., Butchard, S., Eley, R., Gaughan, A., ... & Gabbay, M. (2020b). Impact of COVID-19 related social support service closures on people with dementia and unpaid carers: a qualitative study. *Aging & Mental Health*, 1-8.
- Hutchinson, K., Roberts, C., Roach, P., & Kurrle, S. (2020). Co-creation of a family-focused service model living with younger onset dementia. *Dementia*, 19, 1029-1050. doi:10.1177/1471301218793477
- Johannessen, A., & Möller, A. (2013). Experiences of persons with early-onset dementia in everyday life: A qualitative study. *Dementia*, 12(4), 410-424. doi: 10.1177/1471301211430647
- Kandiah, N., Wang, V., Lin, X., Nyu, M. M., Lim, L., Ng, A., ... & Wee, H. L. (2016). Cost related to dementia in the young and the impact of etiological subtype on cost. *Journal of Alzheimer's Disease*, 49(2), 277-285. doi:10.3233/JAD-150471
- Mayrhofer, A., Mathie, E., McKeown, J., Bunn, F., & Goodman, C. (2017). Age-appropriate services for people diagnosed with young onset dementia (YOD): A systematic review. *Aging & Mental Health*, 1-9. doi:10.1080/13607863.2017.1334038
- Mayrhofer, A., Mathie, E., McKeown, J., Bunn, F., & Goodman, C. (2018). Age-appropriate services for people diagnosed with young onset dementia (YOD): A systematic review. *Aging & Mental Health*, 22(8), 927-935. doi:10.1080/13607863.2017.1334038
- McCarthy, B. (2017). Appreciative inquiry: an alternative to behaviour management. *Dementia*, 16(2), 249-253. doi.org/10.1177/1471301216634921
- Meiland, F. J. M., Dröes, R. M., De Lange, J., & Vernooij-Dassen, M. J. F. J. (2004). Development of a theoretical model for tracing facilitators and barriers in adaptive implementation of innovative practices in dementia care. *Archives of Gerontology and Geriatrics*, 38, 279-290. doi: 10.1016/j.archger.2004.04.038
- Millenaar, J. K., Bakker, C., Koopmans, R. T., Verhey, F. R., Kurz, A., & de Vugt, M. E. (2016). The care needs and experiences with the use of services of people with young-onset dementia and their carers: A systematic review. *International Journal of Geriatric Psychiatry*, 31(12), 1261-1276. doi:10.1002/gps.4502
- National Institute for Health and Care Excellence. (2018). *Dementia: Assessment, management and support for people living with dementia and their carers* (NICE Quality Standard No. 97). Retrieved from <https://www.nice.org.uk/guidance/ng97>

- National Institute for Health and Care Excellence. (2016). *Motor neurone disease: Assessment and management* (NICE guideline NG42). Retrieved from <https://www.nice.org.uk/guidance/ng42/evidence/full-guideline-pdf-2361774637>
- Prince, M., Knapp, M., Guerchet, M., McCrone, P., Prina, M., Comas-Herrera, A., ... Salimkumar, D. (2014). *Dementia UK: Overview* (2nd ed.). London: Alzheimer's Society.
- Reilly, S. T., Harding, A. J., Morbey, H., Ahmed, F., Williamson, P. R., Swarbrick, C., ... & Keady, J. (2020). What is important to people with dementia living at home? A set of core outcome items for use in the evaluation of non-pharmacological community-based health and social care interventions. *Age and Ageing*, *49*(4), 664-671.
- Roach, P., & Drummond, N. (2014). 'It's nice to have something to do': Early-onset dementia and maintaining purposeful activity. *Journal of Psychiatric and Mental Health Nursing*, *21*, 889-895. doi.org/10.1111/jpm.12154
- Roach, P., Drummond, N., & Keady, J. (2016). 'Nobody would say that it is Alzheimer's or dementia at this age': Family adjustment following a diagnosis of early-onset dementia. *Journal of Aging Studies*, *36*, 26-32. doi:10.1016/j.jaging.2015.12.001
- Royal College of Psychiatrists, *Young-onset dementia in mental health services. Recommendations for service provision*. 2018. Available from: https://www.rcpsych.ac.uk/docs/default-source/improving-care/better-mh-policy/college-reports/college-report-cr217.pdf?sfvrsn=31e04d9f_2
- Sansoni, J., Duncan, C., Grootemaat, P., Capell, J., Samsa, P., & Westera, A. (2016). Younger onset dementia: A review of the literature to inform service development. *American Journal of Alzheimer's Disease & Other Dementias*, *31*(8), 693-705. doi: 10.1177/1533317515619481
- Scerri, A., Innes, A., & Scerri, C. (2015). Discovering what works well: exploring quality dementia care in hospital wards using an appreciative inquiry approach. *Journal of Clinical Nursing*, *24*, 1916-1925. doi: 10.1111/jocn.12822.
- Scerri, A., Innes, A., & Scerri, C. (2019). Using appreciative inquiry to implement person-centred dementia care in hospital wards. *Dementia*, *18*, 190-209. Doi: 10.1177/1471301216663953
- Stamou, V., Fontaine, J. L., O'Malley, M., Jones, B., Gage, H., Parkes, J., ... & Oyebode, J. (2020). The nature of positive post-diagnostic support as experienced by people with young onset dementia. *Ageing & mental health*, 1-9.
- Stamou, V., La Fontaine, J., Gage, H., Jones, B., Williams, P., O'Malley, M., ... & Oyebode, J. (2020b). Services for people with young onset dementia: The 'Angela' project national UK survey of service use and satisfaction. *International journal of geriatric psychiatry*.
- van Mierlo, L. D., Meiland, F. J. M., van Hout, H. P. J., & Dröes, R. M. (2014). Towards personalized integrated dementia care: A qualitative study into the implementation of different models of case management. *BMC Geriatrics*, *14*(1), 84. doi: 10.1186/1471-2318-14-84
- Vieira, R. T., Caixeta, L., Machado, S., Silva, A. C., Nardi, A. E., Arias-Carrión, O., & Carta, M. G. (2013). Epidemiology of early-onset dementia: A review of the literature. *Clinical Practice and Epidemiology in Mental Health: CP & EMH*, *9*, 88-95. doi: 10.2174/1745017901309010088
- Westera, A.B. & Fildes, D. L. (2014). National younger onset key dementia worker program national evaluation: Final report. Centre for Health Service Development, Australian Health Services Research Institute. Wollongong: University of Wollongong. Retrieved from:

https://pdfs.semanticscholar.org/7642/2a62011724dd656b9452f79461b37dc3f1d4.pdf?_ga=2.32497854.1804026028.1590359585-1326864848.1590359585

Westera, A., Fildes, D., Duncan, C., Samsa, P., Capell, J., Grootemaat, P., & Sansoni, J. (2016). Literature review and needs and feasibility assessment of services for people with younger onset dementia. Australian Health Services Research Institute. 371. <https://ro.uow.edu.au/ahsri/371>

Willis, G. B. (2004). *Cognitive interviewing: A tool for improving questionnaire design*. London: Sage Publications.

Table 1: Survey briefing notes and a sample question on helpful services

Section 1. Positive experiences of services and support

We would like to know about good examples of help and assistance that have benefited **younger people with dementia and/or family members/supporter(s)**. These could include:

1. A service or form of support which has:

- helped you to maintain or improve your quality of life or,
- made your life easier or,
- addressed a concern you may have

This could include, for example:

- help with your finances,
- your health or,
- maintaining activities that are important to you.

2. Advice or information. This could include, for example:

- information about dementia,
- about how to cope with specific symptoms or,
- how to find the right services to meet your needs.

3. The approach or attitudes of a person who has provided help and assistance that has benefited you.

Survey sample question:

<p>During the year after diagnosis, can you think of times when the help and assistance you received was helpful?</p> <p style="text-align: center;">Yes No</p>
<p>If you answered yes, please provide a detailed description of these experiences. We would like to know:</p> <p>a) what was helpful? b) who was it helpful for? c) why was it helpful? d) who was involved (e.g. social worker, family doctor, nurse, etc.)? e) what was the name of the service(s)?</p>

Table 2: Interview guides used to elicit in-depth information on the nature of helpful services for young onset dementia

Interview Version 1 (used with those at ease recalling survey information)

Open discussion about the service examples provided in the survey. Where possible explore the needs of the person with young onset dementia and the family members/supporters separately. Then explore any additional examples of helpful experiences with services.

For each example given ask:

Can you tell me about the events/ experiences you had leading up to being referred to/receiving this service? (prompts include)

- i. What prompted the referral?
- ii. What were you feeling/ needing at the time?
- iii. Tell me about the process that you went through to get the service.
- iv. What happened when you saw them/ began to receive the service?
- v. What specifically was good about it?
- vi. Why was it helpful to you?
- vii. How was the person's/ services approach helpful?
- viii. How did it make you feel?
- ix. How was this different from how you felt before?

Version 2 (used with those who found it hard to recall survey information)

1. I would like to understand what is important in the lives of people living with young onset dementia and their families and supporters since receiving the diagnosis of dementia. Some people find that life changes quite significantly and that some aspects of their lives become more important. Others describe how important it is to keep living life the way you have always done.

I would like to know what is important to you now in your life?

2. I would like to discuss what helps you to achieve the things in your life that are important to you. Some people find that they require help and assistance to live well with dementia and achieve the things that are important to them.

I would like to know what help and assistance you have received that has helped to you achieve the things that are important to you?

- i. What did they do for you?
- ii. How have they helped you?
- iii. What exactly was helpful about that?
- iv. Why has it been helpful to you?

Table 3: Socio-demographic characteristics of the 26 interview participants

Younger people with dementia (N=10)				
Age distribution				
	Mean	Std. Dev.	Min	Max
Age at diagnosis	57.4	4.09	51	65
Age at study participation	60.6	4.3	55	68
Gender				
	Frequency		Percent	
Female	7		70.0	
Male	3		30.0	
Diagnosis				
	Frequency		Percent	
Alzheimer's	4		40.0	
Frontotemporal dementia (language and behavioural variants)	1		10.0	
Vascular dementia	1		10.0	
Mixed (Alzheimer's + Cerebrovascular Disease)	1		10.0	
Posterior Cortical Atrophy	1		10.0	
Don't know	2		20.0	
Time living with the condition				
	Frequency		Percent	
1-2 years	2		20.0	
5-6 years	5		50.0	
7-9 years	2		20.0	
> 9 years	1		10.0	
Carers (N=16)				
Age distribution				
	Mean	Std. Dev.	Min	Max
Age at diagnosis	52.63	8.16	35	67
Age at study participation	56.13	8.99	37	72
Gender				
	Frequency		Percent	
Female	9		56.2	
Male	7		43.8	
Carer's relationship with younger person with dementia				
	Frequency		Percent	
Daughter	1		6.3	
Son	1		6.3	
Partner	1		6.3	
Spouse	13		81.1	
Carer living with younger person with dementia				
Yes	14		87.5	
No	2		12.5	

Table 4: The nature of helpful services for young onset dementia: Super-ordinate themes, main themes and theme descriptions derived from a survey (n=233) and interviews (n=24)

Super-ordinate themes	Themes	Brief theme description
Organisational coherence	Integrated Care	Services provided collaboratively, rather than in silos, organised in a way that enables a coherent tailored care pathway.
	Care managed by specialists	Care provided and co-ordinated by specialist services and professionals who are trained and experienced with YOD.
	Consistent	Services which give the person regular and continuing contact with a known person and/or service, who remains in touch as needs changed over time.
Functional Consistency	Age-appropriate	Services which provide support to fit the younger age profile and needs of people with YOD and their families, and involve approaches/activities appropriate to age, life stage and lifestyle.
	Holistic	Services which consider and address the multiple facets of YOD and the needs of all in the family who are affected, including children and young people.
	Responsive	Services that are proactive in response to changing needs and which respond to these in a timely manner.
	Accessible	Services that are easily accessible in terms of being local, affordable, dementia friendly and delivered at convenient times.
	Positive attitude	Professionals who interact with service recipients in an attentive, respectful, compassionate, genuine manner, going beyond the minimum to ensure needs are met.
Person-centredness	Collaborative	Professionals who treat service recipients as active and equal partners, forming a collaborative partnership regarding current and future support.
	Flexible	Professionals who adjust the support they offer to the needs of service recipients.
	In-person	Support offered through personal contact, rather than on-line.
	User-friendly material	Clear, concise material about YOD, how to cope, and available services.

Figure legends

Figure 1: Overview of the framework of good practice in young onset dementia service design and delivery, as reported by younger people with dementia and carers.

Appendix 1: Themes and sub-themes on the nature of helpful services, derived from survey responses from people living with young onset dementia and family carers (233 survey responses, 856 written free-text responses)

Themes	Sub-themes
Sensitive engagement with services	Professionals' sensitive approach Active listening Professional's supportive attitude Honesty
Continuity and Consistency	Continuity of services Regular provision of support Developing rapport
Accessible and Responsive to Needs	Timely responsive Collaboration between services Holistic approach Proximity to services Cost-effectiveness of services