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Family experience of young-onset dementia: the perspectives of spouses and children

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Abstract

Objectives: Although young-onset dementia (YOD) affects the whole family system, this population is still under-represented in literature, and no progress in care provision has been made. Hence, additional evidence is necessary to understand how family and social relationships are affected by YOD and care challenges, as to provide recommendations for clinical practice and service improvement from a family perspective.

Method: Family carers were recruited via one memory clinic and the local Alzheimer's Associations in Italy. Semi-structured interviews explored their experiences with YOD, the impact of the condition on their lives, family and social relationships, and the support and care they received. Transcripts were coded by three researchers and analysed using inductive thematic analysis.

Results: Thirty-eight interviews were conducted with 26 spouses and 12 adult children. Three themes emerged: 1) Problems around diagnosis, 2) Lack of post-diagnostic support, and 3) Living with YOD as a family. Overall, problems occurred across the dementia pathway. Without appropriate support, it was difficult for families to adjust to living with YOD and to the associated changes in family roles and relationships.

Conclusions: Since optimal care depends on good family relationships, better support for families in the adaptation to condition would likely benefit patient care while ensuring social inclusion and health equity for vulnerable groups.

Keywords: young onset dementia; patients; spouses; adult children; caregivers; family relations; delayed diagnosis; adaptation, psychological; social isolation; delivery of health care

Introduction

Young-onset dementia, commonly known as YOD, affects people's behaviours and cognitive functions under the age of 65 years (Draper & Withall, 2016). Although dementia mainly affects older than younger adults, increasing attention is being devoted to YOD due to the profound impact of the condition on patients and their families (Richardson et al., 2016).

The prevalence of YOD is challenging to estimate, as epidemiological studies are scarce, and most of them are registry-based (Millenaar et al., 2016). On their basis, the proportion of YOD varies from 5% to 10% of all dementias (Prince et al., 2014). However, these estimates might be lower than actual since many people do not ask early for help, thus preventing timely access to treatment and care (Carter et al., 2018; van Vliet et al., 2013). Furthermore, making a diagnosis of YOD is not easy since it is more likely to be inherited and has a variety of aetiologies and clinical presentations (Rossor et al., 2010). Indeed, younger people living with dementia are less likely to have memory problems as their first symptoms (Koedam et al., 2010). In the early stages of the condition, neuropsychiatric symptoms (e.g., personality changes, loss of empathy, diminished motivation) are common and can be difficult to recognise (Woolley et al., 2011). Misdirected referrals and misdiagnosis can occur and are associated with a lack of specialist knowledge by healthcare practitioners (Bakker et al., 2013; Mendez, 2006). Notwithstanding, the progression of YOD seems to be relatively rapid, thus underlining the importance of earlier diagnosis and intervention (Kuruppu & Matthews, 2013).

Once obtained the diagnosis, people living with YOD and their families need specialist counselling and support to adapt to the condition (Millenaar et al., 2016). Indeed, people living with YOD are often working and are physically active, without any other serious or long-term health conditions (Mayrhofer et al., 2018). They can have financial and family responsibilities, such as mortgages, young children, and/or dependent parents to care for (Svanberg et al., 2011). Dementia symptoms can adversely impact all these roles and responsibilities, thus leading to difficulties surrounding employment, changing social roles and family relationships, poor self-esteem, and a reduced sense of purpose in life (Richardson et al., 2016).

Family carers, mostly spouses, often provide an intensive level of caring which can last several years, thus leading, in the absence of support, to adverse consequences on their emotional and physical health (Cabote et al., 2015; van Vliet et al., 2010). In accomplishing various daily tasks, such as working, raising children, and managing finances, they may not feel confident and prepared to play the role of carers (Wawrziczny et al., 2018). Furthermore, dementia at a younger age, because of its non-normative nature, impacts the entire family unit, including children and young people who are at risk of lasting psychological distress (Chirico et al., 2021a). Indeed, they can struggle to balance caring tasks and their own needs, thus compromising their development, future planning and success in life (Chirico et al., 2021a).

It seems evident that both people living with YOD and their carers experience various psychosocial problems that require specialist support and appropriate treatment and care throughout the entire course of the condition (Stamou et al., 2021a, b). Indeed, due to their age, people living with YOD can prefer less sedentary activities or want more help surrounding the environment. At the same time, carers can need improved access to respite care and psychological support (Giebel et al., 2020, 2021). Despite it, no standard model of care provision exists, and most services are designed for older people and, therefore, are inappropriate for this population’s needs (Ottoboni et al., 2021a). In this context, insufficient attention has been paid both by Higher Education and current policies, thus resulting in a lack of specialist knowledge on how to manage the psychosocial consequences of YOD, as well as in a low level of public awareness (Chirico et al., 2021b; Ottoboni et al., 2021a, b).

Despite the potential and multiple adverse outcomes of dementia in midlife, this population is still under-represented in literature, and no progress in care provision has been made (Livingston et al., 2020; Roach et al., 2008). Since the impact of YOD on families is enormous, it is crucial to understand how family relationships are affected by YOD in order to provide adequate care and services designed upon both patients and families’ needs (Roach et al., 2012). Hence, this study aimed to investigate the experience of dementia in midlife through the perspectives of family members. Findings will inform both researchers and practitioners about the needs of this population by providing recommendations for clinical practice and service improvement from a family perspective.

Methods

Participants and recruitment

Family carers were eligible to take part if they had a relative with a diagnosis of YOD (i.e., under the age of 65 years) of any type. Participants were recruited via convenience sampling via one memory clinic and the local Alzheimer's Association of five urban cities in three regions of Italy. At first, professionals and volunteers contacted potential participants directly via telephone or met them in person, thus sharing information about the study and asking whether they wanted to be involved. Once researchers obtained a list of participants, they called them to set up interview details (date, time, interviewer, location).

Recruitment ceased once no additional themes emerged during the analysis of three consecutive interviews, at which point it was established that saturation had been reached (Saunders et al., 2018). Participants were thanked for their involvement in the study singularly and offered the options to receive a brief research report and be contacted for future research.

Data collection

Ethical approval was obtained from the Ethics Committees of the University of Bologna [Ref: 116887], and of the University Hospital Sant'Orsola-Malpighi of Bologna (Italy) [Ref: 18148]. Written informed consent, including confidentiality and the participant's right to withdraw at any time, was obtained before participation in the study.

Data were collected between October 2019 and February 2020 through semi-structured interviews which were conducted face-to-face or by telephone with geographically dispersed participants. Interviews were carried out by two of the study authors (IC and EM) who have experience in qualitative data collection. During each interview lasting between 30 and 45 minutes, participants were encouraged to describe their experience with YOD, the impact of the condition on their lives, family and social relationships, and the support and care they received. When necessary, the interviewer asked participants to add some details or to clarify their answers. Prior to the interviews, socio-demographic characteristics about participants and their relatives living with YOD (e.g., age, gender, education), as well as information about the condition (e.g., first onset symptoms, time and type of diagnosis) were collected.

Data analysis

The audiotapes of all interviews were transcribed verbatim and analysed via inductive thematic analysis (Braun & Clarke, 2006). Data analysis was performed independently by three researchers (IC, RC, GO) experienced in analysing qualitative data. This analysis consisted of firstly gaining familiarity with the transcripts through repeated readings of the interviews. Each researcher individually employed a feed-forward strategy to extract relevant codes representing the main units of content. Codes about the same issue were clustered into subthemes and, then, into themes by each researcher. The clustering process evolved until it was finalised. It was discussed after each interview with the remaining study authors, until a consensus about its consistency was reached and any discrepancies were solved (Hickey & Kipping, 1996).

Results

Background characteristics

Thirty-eight interviews were undertaken. These included 26 spouses and 12 adult children coming from different families. Spouses' ages ranged between 55 and 75 years (M = 65.1, SD = 5.5), while children were on average 35.5 (±9.4) years old (range 21 to 47). Most participants were female (68.4%) and married (78.9%). They finished secondary (44.7%) or middle (26.3%) school, while smaller percentages obtained an academic degree (23.7%) or just completed primary school (5.3%). Most spouses (65.4 %) were retired, while the majority of children were working (83.3%) and living outside the family home (75%).

Concerning their relatives living with YOD (n=38), their ages ranged between 50 and 76 years (M = 62.03, SD = 5.3), and more than half were male (55.3%). They finished middle (47.4%) or secondary (31.6%) school, while smaller percentages obtained an academic degree (18.4%) or just completed primary school (2.6%). Most of them (81.6%) were retired. The mean time lag between symptom onset and diagnosis was 1.6 (± 1.2) years (range 6 months to 5 years). Diagnoses of YOD were Alzheimer's disease (65.8%) or frontotemporal dementia (23.7%), and the remaining were unknown (10.5%). All people living with YOD were under medication, and 34.2% did not receive any benefits related to their condition. While 71% of them were attended by unpaid family carers only, smaller percentages received home paid care (15.8%), followed day-care centers (7.9%) or lived in a nursing home (5.3%).

Qualitative findings

Three themes were identified from the interviews: 1) Problems around diagnosis; 2) Lack of post-diagnostic support; 3) Living with YOD as family. Each theme is described in detail below, with verbatim extracts of participants' responses.

Table 1. List of themes and sub-themes from interviews

Themes	Sub-Themes
1. Problems around diagnosis	Difficulty in pre-diagnostic phase Delayed diagnosis Inappropriate communication Need of counselling
2. Lack of post-diagnostic support	Financial barriers Lack of case management Geographic barriers Care needs
3. Living with YOD as family	Couple relationships Parent-child relationships Social isolation

THEME 1: Problems around diagnosis

Difficulty in pre-diagnostic phase

The pre-diagnostic phase was characterised by changes in mood, personality and behaviours or, sometimes, by increasing memory problems. It was difficult for people living with YOD and family members to seek help with symptoms being ascribed to stress due to issues related to work, ageing and retirement, and life events.

"It took over 1 year before we sought help. He didn't accept the condition, he still used to drive and ride the bike. <My friends forget things too> he kept saying. I'm not ill, I don't want to go to the psychiatrist, I'm not crazy" **(wife, 69 yrs)**.

"We were aware that there was something wrong, but we didn't want to accept it. Those signs that I initially misinterpreted as due to work overload were instead symptoms of the condition" **(husband, 70 yrs)**.

Sometimes, a point of crisis such as economic losses forced participants to ask for a diagnostic consultation.

"We couldn't understand this condition until we realised we were in an economic crisis because of him" **(wife, 68 yrs)**.

Delayed diagnosis

Participants reported that people living with YOD experienced delays before receiving a correct diagnosis due to having their concerns neglected or being misdiagnosed. General practitioners tended to underestimate symptoms and give misdiagnoses such as depression or stress. Age seemed to be a factor contributing to the missed diagnosis of dementia.

"Our general practitioner has learnt about YOD thanks to me. She's a doctor, but she was asking me for information about this condition since she had no previous experience with it" **(wife, 73 yrs)**.

"[...] I was crying in front of the doctor <Doctor, what can I do with my husband? He's worsening day by day and no one can tell what condition it is. Could it be Alzheimer's? There's another man with Alzheimer's and his symptoms remind me of my husband>. <Can't be, your husband's too young for Alzheimer's, he's probably just tired> the doctor said - he was 60 years old at that time" **(wife, 69 yrs)**.

Before receiving the correct diagnosis, participants often sought the advice of more than one professional (psychiatrists, neurologists) with additional costs due to private visits and time off from work. At the same time, relatives were increasingly frustrated and worried about their loved one's health and were uncertain about the future. Furthermore, misdiagnoses often led to inappropriate drug prescriptions.

"Medical visits were a journey of hope" **(son, 38 yrs)**.

"At first, the doctor visited her ensuring me that it was just anxiety or depression. He gave her psychiatric drugs, anxiolytics and antidepressants but, instead of improving, she was worsening" **(son, 46 yrs).**

Inappropriate communication

Many participants reported that the the YOD condition was not named clearly to their relatives, but rather euphemisms and vague words such as memory problems were used. Such a choice aimed to minimise the emotional impact due to receiving a diagnosis of dementia at a younger age.

"She thinks she's depressed and has some problems with memory and language. That's what she knows, nothing more" **(daughter, 21 yrs)**.

"We haven't clearly told him it, we've tried to sweeten the deal" **(wife, 55 yrs)**.

In a few cases, when diagnosis was clearly stated, participants reported that people living with YOD lacked awareness or capacity to remember it.

"He does realise that he has something…but he doesn't want to know it, or he doesn't realise, or he's pretending, who never knows" **(wife, 58 yrs)**.

"He was there when they told us the diagnosis, but he doesn't remember it" **(wife, 72 yrs)**.

Need of counselling

Receiving the diagnosis came as a shock, and information and guidance by professionals on how to manage this condition were minimal.

"When the diagnosis was told, we didn't know anything about dementia. We didn't know what it would have implied, how the condition would have developed and how much time it would have lasted. It was shocking. I had to learn all by myself, and it wasn't easy at all. There's the need for someone teaching you how to cope with this condition" **(husband, 70 yrs)**.

"Practitioners are not informed enough and they don't have the necessary empathy to manage the situation. Information is just the first step, someone should help us with practical implications and suggestions. How can we do if he doesn't take the pills? Such ordinary things are the most important ones to deal with this condition in the everyday life" **(wife, 62 yrs)**.

Some participants acknowledged that, without any advice, they made wrong choices, thus increasing their psychological distress.

"I had to do all by myself with him and I made many mistakes. I wasn't prepared for it and no one helped me" **(wife, 68 yrs)**.

Whether some information was provided by professionals, it seemed quite narrow in scope and, therefore, was combined with other sources of information, such as the Internet or asking people with similar experiences.

"The doctor helped me with a small number of issues related to the condition. I had to study on my own even during night-time. Internet is a pot of knowledge nowadays, but you have to know how to use it" (**husband, 62 yrs)**.

THEME 2: Lack of post-diagnostic support

Financial barriers

Participants experienced great frustration accessing financial aids or funding assessments due to a slow and excessive bureaucracy. They had to wait several months and filled out plenty of documents before obtaining the help they needed. These challenges were difficult to reconcile with carers' work and life commitments.

"[...] I'm still waiting for the assessment by social workers...He's 65 years old, so they're having trouble comparing him to older people" (**wife, 63 yrs)**.

"The economic support comes too late. Many families haven't received it yet, even though their relative with dementia is already at a moderate stage. The support should come earlier to cope with the first condition challenges" (**wife, 73 yrs)**.

Financial barriers also occurred when people affected by YOD had to stop working some years before being eligible for an age pension.

"At the time of the diagnosis he was currently working as a teacher, that was the main problem. It was hard for him to get retired due to the education policy, so he had to be off sick for a couple of years. Finally, after that, they let him retire" (**wife, 70 yrs)**.

Lack of case management

Participants complained about the lack of a 'case manager' aimed to coordinate health and social care services, and to link people with service upon request.

"There is a lack of coordination among services. There is the need of a person helping you with every aspect of the condition" (**daughter, 42 yrs)**.

Only few participants received the information they needed to identify service resources and facilitation, although it appeared to be due to chance.

"I was really lucky because a neurologist gave me all the information about the available services and resources for the condition" (**wife, 62 yrs)**.

Geographic barriers

Participants perceived geographical disparities in the distribution of services across the city. This limited access to services, since many people living with YOD stopped driving, and they relied upon the carer, in the absence of dedicated transports. While some participants experienced extra-burden, others ultimately gave up due to a difficult balance between daily duties and their loved ones' needs.

"We have to consider another service because it's too far from home. It's on the other side of the city and it's unrealistic for me to reach it twice a week, considering that we've got already many visits and everyday issues" (**wife, 65 yrs)**.

Care needs

Participants spoke about the little and expensive help provided by the national health services, which was partially compensated by local associations. These latter offered a range of social activities such as Alzheimer cafés and carer support groups.

"Public services are ridiculous [...] Associations are much better, they compensate for the lacking public health system" (**son, 38 yrs)**.

Although associations played an important role in supporting families making them fell socially connected, there appeared to be limitations in the activities provided to younger people living with dementia. Participants mainly complained about the low frequency of activities and the group composition. The latter was largely represented by older people with activities less convenient for younger people or, alternatively, it was quite heterogeneous in terms of age and level of impairment. Furthermore, some participants perceived a sort of gender imbalance in groups, with men being under-represented.

"I've tried to bring her in 2 or 3 groups, but she was embarrassed because of the elderly participants" (**husband, 68 yrs)**.

"More time is needed and groups should be homogeneous. My wife cannot write, while the other participants can do it. The group includes people in the 50-70 age range and 20 years are too much. Someone is 50 years old, there are 2-3 men and now there are almost 8 people aged 60 years old" (**husband, 66 yrs)**.

For what concerns the transition to residential care, participants complained about the suitability of facilities and staff unwillingness to accept younger residents as being physically agile yet.

"[...] We looked for appropriate facilities, but most of them were not suitable for him [...] Even the manager told us they couldn't manage someone like him, as he was able to walk yet" (**daughter, 32 yrs)**.

THEME 3: Living with YOD as family

Couple relationships

Since symptom onset, couple relationships seemed to experience several challenges. At the beginning, a lack of understanding of the spouse’s personality changes or the delays in receiving the diagnosis put couple relationships under significant pressure. This, sometimes, caused conflicts thus making it harder to get on, as they previously were used to.

“I was angry because he didn’t want to take care of himself, our relationship was deteriorating… well I wanted to understand that it was the condition, he had nothing against me, knowing earlier about the condition would have helped me a lot. I would have managed it differently, instead of leaving home temporarily” (**wife, 58 yrs)**.

As the condition progressed, spouses had to assume increasing caring tasks to compensate the multiple losses (autonomy, driving license) experienced by people living with YOD. In doing so, spouses tried to protect their children, while having to learn new skills aimed to accomplish those daily tasks that were previously carried out by people living with YOD. Providing intensive care could even lead to symbiosis and feelings of being emotionally overwhelmed.

“She had to stay with her boyfriend, she had her own life…I’m here for the dad. She couldn’t sacrifice her life to stay at home with her dad, for what? Washing and dressing her dad?” (**wife, 72 yrs)**.

“In 2014, when I realised that my husband’s orientation was compromised and he couldn’t drive anymore, what did I do? I was 64-65 years old, I attended the driving school, I said <let’s see if I can do it>” (**wife, 69 yrs)**.

“We’ve become addicted to each other because of this lack of social life, lack of recreation to break the routine. The bond between us is like an umbilical cord” (**wife, 70 yrs)**.

The challenges of dealing with YOD radically changed the husband-wife roles as they were before this condition. Spouses highlighted that, as symptoms worsened, romantic and intimate relationships changed in favour of care relationships only (nurse-patient, mother-child). This seemed to affect the marital quality and was accompanied by feelings of anticipated grief, loneliness, lack of reciprocity and emotional responsiveness by the person living with YOD, and sexual intimacy. Unlike in the past, participants had to make choices regarding family life without their spouses’ support. They also had to give up on their family dreams, plans for the future and live day by day.

“I’m grieved because I don’t have a husband anymore. He complains when I touch him. I’ve become his mother, his nurse. He gives me orders, he needs me. I’m there when he needs me, when he needs drugs, when he needs everything, but I never receive satisfaction, never a sweet

gesture. I don't have a husband anymore, your life changes, the person with you is totally different than was before, your life is different" (**wife, 58 yrs**).

"It's strange…You've been living for years, 40 years, with a person you could talk with, discuss with, even argue with but then, out of the blue, you're alone. You suddenly have to choose just by yourself, and you don't know if you're making the correct decision" (**husband, 70 yrs**).

Only very few spouses were able to be still engaged in social life, and this allowed them to better manage caring tasks thus preserving their mental health.

"I've started to meet friends again. Actually, I've got my own space away from home, I would probably freak out otherwise. My own space lets me survive and I can do things that I enjoy, think less about the condition" (**wife, 56 yrs**).

Parent-child relationships

In the beginning, children struggled to accept the diagnosis, thus experiencing negative emotions such as anger, frustration and family conflicts. Hence, it took some time to re-establish a relationship with their parent or, in extreme cases, they failed to accept the diagnosis.

"At the beginning, my daughter couldn't accept her mother's condition. There was a sort of war and she left home. Then, slowly, she came back and now she's growing little by little" (**husband, 68 yrs**).

"My son doesn't understand it. I mean, yes, he does understand his father's condition, but he cannot accept it" (**wife, 65 yrs**).

Children with caring responsibilities found it challenging to cope with the situation, as these tasks were perceived as unexpected in relation to the life cycle stage. In accomplishing duties, they were concerned about their healthy parent and made efforts to alleviate their emotional discomfort. Relationship roles with their parent with YOD were reversed from what they were previously, i.e. children became parents to their parents while communication gradually decreased. Some children acknowledged that they lived in a limbo and even experienced an 'anticipatory grief' as their parent's condition worsened.

"Honestly, I would have never thought to change my father's diaper at 25 years old…I would accept it if I was 50-60 years old with my needy elderly parents, as it should usually occur" (**daughter, 28 yrs**).

"He's not there anymore. He's out of our lives but, actually, he's always there and you always think about him" (**daughter, 42 yrs**).

Caring responsibilities strongly impacted children's lives concerning education/career, interpersonal relationships and future life planning.

"All these changes caused trouble in my personal life since my mother lives with me and my partner. That brought a lot of problems, and now the relationship with my partner is completely compromised. [...] I've lost my friends too. I hope I won't lose my job because I'm always tired and nervous at work" (**son, 38 yrs)**.

"You can't make any long-term plans, you have to live day by day and face the upcoming demands. You don't know how it will carry on" (**daughter, 32 yrs)**.

However, some positive aspects seemed to coexist with the adverse outcomes of caring at younger age, such as increased maturity, self-awareness and self-worth.

"I feel stronger now, this experience brought out my determination. I was surprised about myself. Despite this bad situation, I feel like my self-esteem has increased, I'm more aware of myself now" (**daughter, 34 yrs)**.

Social isolation

Participants faced significant challenges in maintaining close social relationships. Conflicts with family members often occurred due to having different views on how to manage this condition. Simultaneously, most friends gradually disappeared due to the emotional impact of this condition, difficulty in communication, and lack of knowledge on how to relate to a person living with YOD.

"I lost contact with his family. This condition broke our bonds. I've been accused of not caring him enough by her sisters" (**wife, 68 yrs)**.

"My mother suffered a lot because many people disappeared after the diagnosis. They had this group of friends since they were 18 years old [...] but they drifted apart little by little (**daughter, 32 yrs)**.

"They don't know how to manage the relationship. It's embarrassing because they sometimes ask something and then she answers about something else" (**husband, 62 yrs)**.

"People that I know well genuinely told me <I'm scared she may not recognise me>. They're in good faith, but they don't help us in this way. This increases family burden, our perceived isolation, and my mother's complete loneliness" (**son, 38 yrs)**.

Participants felt stigmatised by society at large due to the lack of awareness about YOD, its nature and strong impact on families. This could even lead them to avoid social interactions because they felt ashamed of their relatives' behaviours, and were afraid of being bullied or rejected.

"I think that there is a social stigma yet due to lack of information about the condition. It's a social problem that exists from a long time. You couldn't even mention oncological diseases around 20-30 years ago, it's not that different from the stigma about homosexuality" (**wife, 55 yrs**).

"I notice a lack of knowledge, people don't know anything about this condition, how it destroys our lives, how it breaks families and friendships. There is no adequate knowledge and support for this specific condition, there are no advertisements as for other conditions, it's a bastard condition" (**husband, 64 yrs**).

"It was annoying going outside with him because he acted weirdly and people chattered about it. That's what annoyed me the most. I don't know if embarrassment is the correct word to describe it [...] I didn't want him to be addressed as a dumb person, that's all" (**wife, 68 yrs**).

Discussion

This study explored the experience of dementia in midlife from diagnosis to post-diagnostic support. It provides evidence on how families touched by YOD face significant challenges to adjust to living with such a long-term condition. Indeed, problems occurred across the course of dementia, from the time required to obtain the diagnosis, to receiving post-diagnostic support. Our results support previous studies (Giebel et al., 2020, 2021; Millenaar et al., 2017; Rossor et al., 2010; van Vliet et al., 2013), suggesting several diagnostic challenges associated with a lack of specialist knowledge by general practitioners and healthcare professionals. A delayed diagnosis does imply that the person and their family don't get the support they need at the right time. Furthermore, we found that the diagnosis was often not clearly told to people living with YOD. This aspect should be carefully considered since there is empirical evidence (Werbrouck et al., 2018) suggesting the need for patients to be told, since the early stages of YOD, all relevant aspects of their condition, including its nature, expected outcomes and available treatments. If such disclosure is made appropriately with sensitivity and tact, it empowers patients to adapt to their situation while avoiding family tension due to a secret being kept (Millenaar et al., 2017).

Families felt emotionally neglected from the time of diagnosis throughout the condition. As found in the previous research (Millenaar et al., 2016; Roach et al., 2008, 2012), professionals did not understand the enormous burdens associated with the experience of dementia in midlife. Indeed, it was difficult for families to receive appropriate information and timely support to adapt to the condition. They often faced a lonely journey of multiple and overlapping difficulties. Among them, a lengthy bureaucratic process often hindered families in their efforts to receive support and help, even modest, at the right time. Furthermore, families were proactive in looking for information and support on their own, through the Internet or by having the chance to meet people with similar experiences. However, without any advice, they made some mistakes that increased their psychological distress while reducing the quality of care for patients. In this regard, counselling for carers should be focused on improving their resilience and empowerment as to deal with symptoms and cope with psychosocial problems associated with YOD, while knowing how and where seeking help (Cabote et al., 2015; Millenaar et al., 2017; Pinquart & Sörensen, 2006; Wawrziczny et al., 2018).

Even when some support was available, geographic barriers prevented access to care. With previous studies (Bauer et al., 2019; Innes et al., 2011) suggesting that rural residents, due to geographic distance, may be limited or unable to access healthcare services, similar evidence was found even on a smaller scale. Indeed, services were not equally distributed across the city, causing an additional burden on care provision, as patients had to rely on their family members in the absence of dedicated transport. These gaps should be adequately addressed by existing policies to design interventions for patients taking into consideration family needs, thus avoiding that treatment course would cause a greater burden to patients' relatives (Golics et al., 2013).

Interestingly, as compared to the little and expensive help provided by the national healthcare system, local Alzheimer's Associations were able to provide support to families and activities for people living with YOD, although with some limitations. As found in previous studies (Cations et al., 2017; Giebel et al., 2020, 2021; Stamou et al., 2021a, b), many participants highlighted the need for their relatives living with YOD to attend activities specifically designed for their age. Indeed, groups composed of older people or mixed groups seemed to create a mismatch in physical abilities and leisure interests. This even led to service users' emotional discomfort and drop-out due to not having received the support they really needed. Therefore, attention should be paid to provide age-appropriate respite care through a careful design of therapeutic group composition, as well as appropriate activities for people living with YOD aimed to strengthen their capacities while enhancing their social connectivity (Aplaon et al., 2017; Green & Kleissen, 2013; Ottoboni et al., 2021a). Our study also highlighted the need of accommodating residential facilities to younger residents as to increase the chance of access. At this regard, the national health system should guarantee the continuity of high-quality care in those situations where families cannot provide any support (Bakker et al., 2013).

This study provides evidence on how YOD affects family and social relationships. As increasing evidence points out (Golics et al., 2013), family members can be seriously emotionally affected by this condition, even more than patients themselves. In our study, family relationships were significantly challenged since the diagnostic period, during which delays in diagnosis increased family conflicts with symptoms being blamed on the patient rather than on dementia. In this regard, it is necessary to provide timely diagnosis and specialist support to families to avoid the early potential worsening of family relationships and to minimise the emotional impact of such a diagnosis (Roach et al., 2012). Indeed, we found that radical changes occurred in spousal relationships throughout dementia. Among them were the person living with YOD's increasing dependence on the spouse and the gradual shift of roles and responsibilities. This led to lower quality of the relationship, increasing social isolation and loneliness, lack of reciprocity and sexual intimacy. In extreme cases, there was risk of symbiosis, which was fostered by social isolation. With lack of support, spouses felt emotionally overwhelmed because of caring tasks and multiple roles associated with their specific life stage. Future family plans were abandoned, and life was mostly restricted to patients' needs with other vital areas of life being sacrificed. Furthermore, sexual activity, which represents a crucial aspect of couple functioning, significantly reduced. Whilst for older people, sexual intimacy often shifts towards other modalities of exchanging affection, for a younger population, sexual activity is still important, with declines likely affecting self-esteem and the emotional health of couples (Holdsworth & McCabe, 2018). Hence, beyond practical needs, counselling should be provided to couples concerning relationships, intimacy, and sexuality to ensure the adaptation of spousal relationships to the profound changes related to YOD.

In such a family scenario, we found that children struggled to deal with the diagnosis so much so that they could drift apart from families. This points out the importance of providing tailored support since the early stages of this condition to safeguard the family unit. Over time, children can assume increasing responsibilities as adults in the attempt to help their healthy parents and keep the family together. However, as compared to adult carers, for a younger population caring tasks and developmental needs can be difficult to reconcile (European Association Working for Carers, 2017). Indeed, our results suggest that YOD impacts on different areas of children's lives including education/career, interpersonal relationships, and future life planning. Despite it, as a recent systematic review highlights (Chirico et al., 2021a), caring at a younger age can be viewed as a natural extension of family relationship thus not receiving specialist support and care. For example, the children's experience of 'anticipatory grief' of their sick parent is often neglected or not recognised both by professionals and society. Therefore, support is required, and psychoeducation, face-to-face groups, online forums and/or blogs are recommended to ease children's burden (Chirico et al., 2021a; Pappadà et al., 2021).

A prominent theme emerging from interviews was the growing social isolation and loneliness due to the loss of relationships, including with the extended family and friends, and to increasing responsibilities. As previously discussed, dementia at a younger age is challenging to cope with for both patients and families due to its non-normative nature. Its psychosocial consequences are broader and less evident than in late-onset dementia. Therefore, effective support and care should be based on a 'whole family' approach taking into consideration both patients and family needs at the same time (Cabote et al., 2015; Roach et al., 2012). This would require a network of collaborative services, including adult, child, health and voluntary services (Chirico et al., 2021a). Professionals should be provided with YOD-specific training and education and linked by the mean of a link contact, knowing the family situation and able to coordinate care at the right time and in the right ways (Cations et al., 2017; Ottoboni et al., 2021a). By providing support, positive aspects of caring such as personal growth or improved self-efficacy could balance the adverse outcomes of care. In this view, greater public knowledge and understanding about YOD, through the involvement of mass media, may contribute to avoid family self-isolation while promoting social connectivity, increased support at the policy level, and better emotional and practical support for patients and families.

Limitations and proposals for future research

This is a study exploring the experience of YOD and, more specifically, its impact on family and social relationships from the perspectives of spouses and children. Despite our findings support and extend previous evidence from this field, findings are based on a small number of interviews which were also conducted throughout three regions in Italy only. Therefore, since dementia care is health system and culture dependent, the generalizability of this study is limited by the small representativeness of participants.

Another limitation stems from the fact that diagnoses reported by participants were only Alzheimer's disease and frontotemporal dementia. It would be interesting to obtain quantitative longitudinal data on how YOD impacts on family relationships within different phases of child development, type of YOD and parental condition stages. To gain a more comprehensive view about the impact of YOD on family relationships, data might be collected from people living with YOD as well as from their family members. Furthermore, family interventions could be empirically evaluated to provide evidence on what works and why.

Conclusions

The impact of YOD is both profound due to the specific life cycle stage, and widespread as it regards the overall family system. Despite it, family members' concerns could be neglected or not understood. As known, one patient could create several more 'hidden patients' in the family. In this regard, this study provides evidence for the need for age-appropriate support for people living with YOD, which would take into consideration family needs and changing roles and responsibilities within the family. Overall, results are in line with those from a recent, large scale UK study (Stamou et al., 2021c) that identified the core features of optimum models for service provision for people living with YOD and their families. Firstly, services staff should adopt a person-centred approach, i.e. have a positive attitude, be collaborative and flexible, and provide in-person support and user-friendly information for active middle-aged people. Secondly, services should be organised taking into account the specific needs of this younger population as well as those of the whole family, and be accessible and respond proactively to their changing needs. Finally, specialist services should be guaranteed as well as the continuity of care over time and integration across services. Since optimal care depends on good family relationships, better support for families would likely benefit patient care while ensuring social inclusion and health equity for vulnerable groups.

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References

Aplaon, M., Belchior, P., Gélinas, I., Bier, N., & Aboujaoudé, A. (2017). Interventions for individuals with young-onset dementia. A review of the literature. *Journal of Aging Research & Clinical Practice, J Aging Res Clin Practice 20176,* 28–31. http://dx.doi.org/10.14283/jarcp.2016.123

Bakker, C., de Vugt, M. E., van Vliet, D., Verhey, F. R. J., Pijnenburg, Y. A., Vernooij-Dassen, M. J. F. J., & Koopmans, R. T. C. M. (2013). The Use of Formal and Informal Care in Early Onset Dementia: Results From the NeedYD Study. *The American Journal of Geriatric Psychiatry, 21*(1), 37–45. https://doi.org/10.1016/j.jagp.2012.10.004

Bauer, M., Fetherstonhaugh, D., Blackberry, I., Farmer, J., & Wilding, C. (2019). Identifying support needs to improve rural dementia services for people with dementia and their carers: A consultation study in Victoria, Australia. *The Australian Journal of Rural Health*, *27*(1), 22–27. https://doi.org/10.1111/ajr.12444

Braun, V., & Clarke, V. (2006). Using thematic analysis in psychology. *Qualitative Research in Psychology*, *3*(2), 77–101. https://doi.org/10.1191/1478088706qp063oa

Cabote, C. J., Bramble, M., & McCann, D. (2015). Family Caregivers' Experiences of Caring for a Relative With Younger Onset Dementia: A Qualitative Systematic Review. *Journal of Family Nursing*, *21*(3), 443–468. https://doi.org/10.1177/1074840715573870

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. https://doi.org/10.1080/13607863.2016.1257563

Cations, M., Withall, A., Horsfall, R., Denham, N., White, F., Trollor, J., Loy, C., Brodaty, H., Sachdev, P., Gonski, P., Demirkol, A., Cumming, R. G., & 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. https://doi.org/10.1371/journal.pone.0180935

Chirico, I., Ottoboni, G., Valente, M., & Chattat, R. (2021a). Children and young people's experience of parental dementia: A systematic review. *International Journal of Geriatric Psychiatry*, *36*(7), 975–992. https://doi.org/10.1002/gps.5542

Chirico, I., Chattat, R., Dostálová, V., Povolná, P., Holmerová, I., de Vugt, M. E., Janssen, N., Dassen, F., Sánchez-Gómez, M. C., García-Peñalvo, F. J., Franco-Martín, M. A., & Ottoboni, G. (2021b). The Integration of Psychosocial Care into National Dementia Strategies across Europe: Evidence from the Skills in DEmentia Care (SiDECar) Project. *International Journal of Environmental Research and Public Health*, *18*(7), 3422. https://doi.org/10.3390/ijerph18073422

Draper, B., & Withall, A. (2016). *Young onset dementia—Draper—2016—Internal Medicine Journal—Wiley Online Library*. https://doi.org/10.1111/imj.13099

European Association Working for Carers. (2017). *EUROCARERS POLICY PAPER YOUNG CARERS*. https://eurocarers.org/publications/eurocarers-policy-paper-on-young-carers/

Giebel, C., Eastham, C., Cannon, J., Wilson, J., Wilson, J., & Pearson, A. (2020). Evaluating a young-onset dementia service from two sides of the coin: Staff and service user perspectives. *BMC Health Services Research*, *20*(1), 187. https://doi.org/10.1186/s12913-020-5027-8

Giebel, C., Sutcliffe, C., Darlington-Pollock, F., Green, M. A., Akpan, A., Dickinson, J., Watson, J., & Gabbay, M. (2021). Health Inequities in the Care Pathways for People Living with Young- and Late-Onset Dementia: From Pre-COVID-19 to Early Pandemic. *International Journal of Environmental Research and Public Health*, *18*(2), E686. https://doi.org/10.3390/ijerph18020686

Golics, C. J., Basra, M., Salek, & Finlay. (2013). The impact of patients' chronic disease on family quality of life: An experience from 26 specialties. *International Journal of General Medicine*, 787. https://doi.org/10.2147/IJGM.S45156

Green, T., & Kleissen, T. (2013). Early onset dementia: A narrative review of the literature. *Indian Journal of Gerontology: A Quarterly Journal Devoted to Research on Ageing*, *27*(1), 1–28.

Hickey, G., & Kipping, C. (1996). A multi-stage approach to the coding of data from open-ended questions. *Nurse Researcher*, *4*(1), 81-91. https://doi.org/10.7748/nr.4.1.81.s9

Holdsworth, K., & McCabe, M. (2018). The impact of younger-onset dementia on relationships, intimacy, and sexuality in midlife couples: A systematic review. *International Psychogeriatrics*, *30*(1), 15–29. https://doi.org/10.1017/S1041610217001806

Innes, A., Morgan, D., Kosteniuk, J., & Kostineuk, J. (2011). Dementia care in rural and remote settings: A systematic review of informal/family caregiving. *Maturitas*, *68*(1), 34–46. https://doi.org/10.1016/j.maturitas.2010.10.002

Koedam, E. L. G. E., Lauffer, V., van der Vlies, A. E., van der Flier, W. M., Scheltens, P., & Pijnenburg, Y. A. L. (2010). Early-Versus Late-Onset Alzheimer's Disease: More than Age Alone. *Journal of Alzheimer's Disease*, *19*(4), 1401–1408. https://doi.org/10.3233/JAD-2010-1337

Kuruppu, D. K., & Matthews, B. R. (2013). Young-onset dementia. *Seminars in Neurology*, *33*(4), 365–385. https://doi.org/10.1055/s-0033-1359320

Livingston, G., Huntley, J., Sommerlad, A., Ames, D., Ballard, C., Banerjee, S., Brayne, C., Burns, A., Cohen-Mansfield, J., Cooper, C., Costafreda, S. G., Dias, A., Fox, N., Gitlin, L. N., Howard, R., Kales, H. C., Kivimäki, M., Larson, E. B., Ogunniyi, A., … Mukadam, N. (2020). Dementia prevention, intervention, and care: 2020 report of the Lancet Commission. *The Lancet*, *396*(10248), 413–446. https://doi.org/10.1016/S0140-6736(20)30367-6

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. https://doi.org/10.1080/13607863.2017.1334038

Mendez, M. F. (2006). The accurate diagnosis of early-onset dementia. *International Journal of Psychiatry in Medicine*, *36*(4), 401–412. https://doi.org/10.2190/Q6J4-R143-P630-KW41

Millenaar, J., Bakker, C., Koopmans, R. T. C. M., Verhey, F. R. J., 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 caregivers: A systematic review. *International Journal of Geriatric Psychiatry*, *31*(12), 1261–1276. https://doi.org/10.1002/gps.4502

Millenaar, J., Hvidsten, L., Vugt, M. E. de, Engedal, K., Selbæk, G., Wyller, T. B., Johannessen, A., Haugen, P. K., Bakker, C., Vliet, D. van, Koopmans, R. T. C. M., Verhey, F. R. J., & Kersten, H. (2017). Determinants of quality of life in young onset dementia – results from a European multicenter assessment. *Aging & Mental Health*, *21*(1), 24–30. https://doi.org/10.1080/13607863.2016.1232369

Ottoboni, G., Stamou, V., Chirico, I., De Riso, L., Allione, L., Johannessen, A., & Chattat, R. (2021a). Needs-appropriate services for people with young onset dementia: The perspectives of healthcare professionals. *Dementia (London, England)*, 14713012211009340. Advance online publication. https://doi.org/10.1177/14713012211009340

Ottoboni, G., Chirico, I., Povolná, P., Dostálová, V., Holmerová, I., Janssen, N., Dassen, F., de Vugt, M., Sánchez-Gómez, M. C., García-Peñalvo, F., Franco-Martin, M. A., & Chattat, R. (2021b). Psychosocial care in dementia in European higher education: Evidence from the SiDECar ("Skills in DEmentia Care") project. *Nurse Education Today, 103,* 104977. https://doi.org/10.1016/j.nedt.2021.104977

Pappadà, A., Chattat, R., Chirico, I., Valente, M., & Ottoboni, G. (2021). Assistive Technologies in Dementia Care: An Updated Analysis of the Literature. *Frontiers in Psychology, 12,* 644587. https://doi.org/10.3389/fpsyg.2021.644587

Pinquart, M., & Sörensen, S. (2006). Helping caregivers of persons with dementia: Which interventions work and how large are their effects? *International Psychogeriatrics*, *18*(4), 577–595. https://doi.org/10.1017/S1041610206003462

Prince, M., Knapp, M., Guerchet, M., McCrone, P., Prina, A. M., & Comas-Herrera, A. (2014). *Dementia UK Second edition—Overview*. 62. alzheimers.org.uk/dementiauk

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

Roach, P., Keady, J., & Bee, P. (2012). 'It's easier just to separate them': Practice constructions in the mental health care and support of younger people with dementia and their families. *Journal of Psychiatric and Mental Health Nursing*, *19*(6), 555–562. https://doi.org/10.1111/j.1365-2850.2011.01836.x

Roach, P., Keady, J., Bee, P., & Hope, K. (2008). *Subjective experiences of younger people with dementia and their families: Implications for UK research, policy and practice | Reviews in Clinical Gerontology | Cambridge Core*. https://doi.org/10.1017/S0959259809002779

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

Saunders, B., Sim, J., Kingstone, T., Baker, S., Waterfield, J., Bartlam, B., Burroughs, H., & Jinks, C. (2018). Saturation in qualitative research: Exploring its conceptualisation and operationalisation. *Quality & Quantity*, *52*(4), 1893–1907. https://doi.org/10.1007/s11135-017-0574-8

Stamou, V., La Fontaine, J. L., Gage, H., Jones, B., Williams, P., O'Malley, M., Parkes, J., Carter, J., & Oyebode, J. (2021a). Services for people with young onset dementia: The 'Angela' project national UK survey of service use and satisfaction. *International Journal of Geriatric Psychiatry*, *36*(3), 411–422. https://doi.org/10.1002/gps.5437

Stamou, V., La Fontaine, J. L., O'Malley, M., Jones, B., Gage, H., Parkes, J., Carter, J., & Oyebode, J. (2021b). The nature of positive post-diagnostic support as experienced by people with young onset dementia. *Aging & Mental Health*, *25*(6), 1125–1133. https://doi.org/10.1080/13607863.2020.1727854

Stamou, V., La Fontaine, J., O'Malley, M., Jones, B., Parkes, J., Carter, J., & Oyebode, J. R. (2021c). Helpful post-diagnostic services for young onset dementia: Findings and recommendations from the Angela project. *Health & social care in the community*, 10.1111/hsc.13383. Advance online publication. https://doi.org/10.1111/hsc.13383

Svanberg, E., Spector, A., & Stott, J. (2011). The impact of young onset dementia on the family: A literature review. *International Psychogeriatrics*, *23*(3), 356–371. https://doi.org/10.1017/S1041610210001353

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

van Vliet, D., Vugt, M. E. de, Bakker, C., Koopmans, R. T. C. M., & Verhey, F. R. J. (2010). Impact of early onset dementia on caregivers: A review. *International Journal of Geriatric Psychiatry*, *25*(11), 1091–1100. https://doi.org/10.1002/gps.2439

Wawrziczny, E., Berna, G., Ducharme, F., Kergoat, M.-J., Pasquier, F., & Antoine, P. (2018). Characteristics of the spouse caregiving experience: Comparison between early- and late-onset dementia. *Aging & Mental Health*, *22*(9), 1213–1221. https://doi.org/10.1080/13607863.2017.1339777

Werbrouck, A., Swinnen, E., Kerckhofs, E., Buyl, R., Beckwée, D., & De Wit, L. (2018). How to empower patients? A systematic review and meta-analysis. *Translational Behavioral Medicine*, *8*(5), 660–674. https://doi.org/10.1093/tbm/iby064

Woolley, J. D., Khan, B. K., Murthy, N. K., Miller, B. L., & Rankin, K. P. (2011). *The diagnostic challenge of psychiatric symptoms in neurodegenerative disease: Rates of and risk factors for prior psychiatric diagnosis in patients with early neurodegenerative disease—PubMed*. https://doi.org/10.4088/jcp.10m06382oli