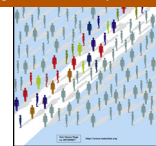




Maturitas

journal homepage: www.elsevier.com/locate/maturitas

Review article

The experiences of people with young-onset dementia: A meta-ethnographic review of the qualitative literature



Nan Greenwood (PhD)*, Raymond Smith (PhD)

Kingston University and St. George's University, London, United Kingdom

ARTICLE INFO

Article history:

Received 18 July 2016

Accepted 26 July 2016

Keywords:

Dementia

Young onset

Experiences

Review

Meta-ethnography

ABSTRACT

Dementia is usually diagnosed in later life but can occur in younger people. The experiences of those with older-onset dementia are relatively well understood but little is known about the experiences of those with young-onset dementia (aged less than 65 years).

This meta-ethnography therefore synthesised qualitative literature investigating the experiences of people with young-onset dementia (YOD). Six electronic databases were searched and 1155 studies were identified, of which eight fitted the inclusion criteria.

These studies were all from Western countries, were mostly recent (2004–2015) and included the experiences of 87 people with YOD. Participants were generally in their fifties or early sixties and were living at home with others. Many reported difficulties both in the process of receiving a diagnosis and afterwards. Diagnosis felt unexpected, 'out of time' and led to changes in self-identity, powerlessness and changes in relationships. Social exclusion was common. Loss of meaningful activity exacerbated a difficult situation. However, the diagnosis did not mean people's lives were over and many with YOD try to regain control by seeking connections with others with the same condition – sometimes a very important source of support.

Overall, people living with YOD face unique social challenges which go beyond those of older people living with dementia and which result in an even greater negative impact on their lives. Interventions that facilitate peer support and allow people with YOD to engage in meaningful activity should be developed and could perhaps be provided by the voluntary sector.

© 2016 The Author(s). Published by Elsevier Ireland Ltd. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

Contents

1. Background	103
2. Review methodology	103
3. Methods	103
3.1. Inclusion criteria and study selection	103
3.1.1. Inclusion criteria	103
3.1.2. Exclusion criteria	103
3.2. Electronic search strategy	104
3.3. Other sources searched	104
3.4. Data extraction and management	104
3.5. Quality assessment	104
3.6. Data synthesis	104
3.7. Articles coming close to inclusion but excluded	104

* Corresponding author at: Reader in Health and Social Care Service Research, Faculty of Health Social Care and Education, St George's University of London and Kingston University, Room 61, 2nd Floor Grosvenor Wing, Cranmer Terrace, Tooting, London SW17 0RE, United Kingdom.

E-mail address: nan.greenwood1@gmail.com (N. Greenwood).

3.8. Study and participant characteristics.....	104
3.9. Overall themes.....	105
3.10. Line of argument.....	105
4. Discussion.....	107
4.1. Review strengths and limitations.....	107
5. Conclusions, service implications and future research.....	108
Contributors.....	108
Conflict of interest.....	109
Funding.....	109
Provenance and peer review.....	109
References.....	109

1. Background

Although dementia mostly affects older people, it can occur in younger people aged 65 years or less where it is variously known as young-onset dementia [1], early-onset dementia [2] or working-age dementia [3].

Young-onset dementia (YOD) is relatively rare. Estimates of prevalence vary but a recent review reported that registry based studies suggest the prevalence of dementia in people aged between 45 and 64 years old lies between 81 and 113 per 100,000 [4]. However, this may be an underestimate because of unexpected timing and relative rarity of YOD [5]. As a result for many people diagnosis may come comparatively late in the disease's progression [6]. Indeed, it is thought that, on average, it takes over one and a half years longer to be diagnosed for people with YOD compared with people with later onset dementia [7,8]. Furthermore, misdiagnosis is not unusual, resulting in further delays in diagnosis [9] with subsequent delays in receiving appropriate support.

The three most common types of YOD are Alzheimer's disease, vascular dementia and frontotemporal dementia (FTD) [10]. Compared to those with later onset dementia, FTD is more common in younger people [11]. Early on, FTD may involve personality changes, challenging behaviour and reduced motivation [12]. Such changes in the person with dementia can be particularly difficult both for those living with the condition and those supporting them [13].

The challenges for unpaid carers of someone with dementia are well documented [14] but arguably the situations of those caring for someone with YOD are particularly difficult because of its timing. For example, most people with later onset dementia have already retired and any children are likely to be grown up, whilst those with YOD are more likely to have relatively young children posing particular challenges for the entire family [15]. In addition, the age of people with YOD means that the symptoms of dementia may lead to loss of employment [2,16]. The implications of unemployment are manifold and include financial, psychological and social consequences, such as changed or difficult family relationships, poor self-esteem and reduced sense of competency and purpose [2,16–18].

Recognition of the challenges faced by those with dementia in general and their families [19] and more specifically YOD has led to the development of support services including education, support groups and counselling [17,20,21]. A recent review focussing on interventions for younger people with dementia and their carers highlighted the value of purposeful activity (often employment based) for both people with YOD and their carers [22]. Benefits included enhanced self-esteem, sense of purpose and increased social contact.

However, although there is a considerable body of synthesised literature exploring the needs and experiences of both people with dementia in general [23] and their family carers [14], there appears

to be no systematic synthesis of the literature exploring the experiences of people with YOD.

This review therefore aimed to synthesise and evaluate qualitative literature relating specifically to the experiences of people with YOD. The primary research question was: What are the experiences of people diagnosed with YOD?

2. Review methodology

There are now several well recognised approaches for conducting systematic reviews of qualitative literature. Meta-ethnography [24], a form of interpretative synthesis, is one of the earliest methods and is a well-accepted form of qualitative review [25,26]. It was selected here for several reasons but primarily because it integrates concepts, goes beyond simply summarising data and is intended to develop further concepts and theories. It requires both induction and interpretation and makes explicit direct comparisons between studies and 'translates' concepts across the selected studies [27]. Furthermore, it has been used in syntheses of health research looking at a variety of issues ranging from, for example, medication taking [26] and the experiences of family carers of people with stroke [28].

3. Methods

The review followed the Centre of Reviews and Dissemination (CRD) guidelines [29] and was reported using the Preferred Reporting Items for Systematic Reviews and Meta-Analysis (PRISMA) guidelines [30].

3.1. Inclusion criteria and study selection

3.1.1. Inclusion criteria

1. Qualitative or mixed methods, primary research
2. Investigations of the experiences and perceptions of people with YOD (aged less than 65 years)
3. Published in English in peer reviewed journals

3.1.2. Exclusion criteria

1. Quantitative
2. Participants diagnosed with YOD aged 65 years or over
3. Investigations of the experiences of people with LOD and other conditions
4. Experiences of people with YOD and those of family or paid carers not separately described
5. Published in grey literature, non-peer reviewed journals, reviews or opinion publications

Following duplicate removal, all titles and abstracts were screened. Full texts were sourced for all articles fitting the inclusion criteria. This process was conducted by both authors and where disagreements occurred, consensus was achieved by discussion.

Table 1
Example electronic search strategy conducted in Medline (OVID).

	Search concept	Search terms
1	Experiences	Experienc\$ OR Perception\$ OR Perspective\$
2	Condition	<i>Alzheimer Disease</i> OR Alzheimer\$ OR <i>Dementia</i> OR Dement\$ OR presenile dement\$ OR <i>Frontotemporal dementia</i> OR <i>Frontotemporal lobar degeneration</i> OR <i>Frontotemporal\$</i> OR <i>Pick Disease of the Brain</i> OR Pick's disease OR Frontal lobe dement\$ OR Behavior?ral variant OR Progressive non-fluent aphasia OR Semantic dementia OR <i>Dementia, Vascular</i> OR Vascular dement\$ OR <i>Korsakoff syndrome</i> OR Korsakoff\$ OR Alcoholic dement\$ OR <i>Lewy bodies</i> OR Lewy bod\$ OR Mixed dement\$ OR <i>Dementia, Multi-Infarct</i> OR <i>Huntington disease</i> OR <i>Huntington\$</i> OR <i>Creutzfeldt-Jakob syndrome</i> OR CJD OR AIDS Dementia Complex OR HIV related dement\$ OR Parkinson's disease dementia OR Down? Syndrome OR <i>Down syndrome</i>
3	Population	Under 65 OR Young onset OR Young-onset OR Younger-onset OR Younger onset OR YOD OR Early onset OR Early-onset OR EOD OR Young person OR Young people OR Younger person OR Younger people OR Working age

Note: The MeSH (Medical Subject Heading) terms used are reported in italics and key words with truncation where appropriate.

3.2. Electronic search strategy

Six electronic databases were searched: Medline (1945 – March 2016), Embase (1980 – March 2016), PsychINFO (1967 – March 2016), Cumulative Index to Nursing and Allied Health Literature (CINAHL, 1937 – March 2016), Applied Social Sciences Index and Abstracts (ASSIA, 1987 – March 2016) and Scopus (1960 – March 2016).

The example search strategy used in Medline is available in [Table 1](#). Similar search strategies were developed according to specific database requirements and consisted of both MeSH terms and key words. Key words and combinations were the same throughout the database searching.

3.3. Other sources searched

Dementia: The International Journal of Social Research and Practice was hand searched for potentially relevant articles as many articles were identified from this journal during electronic searches. Reference searching of both relevant systematic reviews retrieved from electronic searches and included articles was also conducted.

3.4. Data extraction and management

A standardised data extraction form was developed and data were extracted from all included articles independently by both authors. Data extracted included, for example: author details, year of publication, participant demographics, sample size and the main themes identified by the study authors.

3.5. Quality assessment

Quality assessment was conducted independently by the authors using the rating scale developed by Greenwood et al. [31] The scale consists of 11 questions with 'yes' or 'no' answers. The maximum score possible is 11. Included articles were independently scored with disagreements resolved by discussion. Studies were not excluded based on quality scores but this assessment process enhanced study interrogation.

Box 1: Seven steps in conducting a meta-ethnography [24]

1. Getting started: determining the research questions
2. Deciding what is relevant to the initial interest: defining the focus of the synthesis, locating relevant studies, inclusion decisions and quality assessment of included studies
3. Reading the studies: reading the articles multiple times to identify the main concepts
4. Determining how the studies are related: listing the emerging concepts and identifying how they are related
5. Translating the studies into one another: listing the concepts in a table and then looking for these concepts in each of the included articles
6. Synthesising translations: building a line of argument by exploring the relationships between concepts
7. Expressing the synthesis: How the synthesis is reported

3.6. Data synthesis

Data were synthesised using the meta-ethnographic approach developed by Noblit and Hare [24]. Both reviewers were involved in all stages of the analysis. An overview of the process is available in [Box 1](#).

Electronic searches identified 1125 articles (with 122 duplicates). Twenty-four full-texts were retrieved from the remaining 1003 and six were included in the data synthesis [2,16–18,20,32]. Hand searching reference lists of six relevant systematic reviews identified from the electronic searches revealed 13 articles for scrutiny. After reviewing their abstracts, three full-texts were retrieved with one fitting the inclusion criteria [34]. Further hand searching in *Dementia: The International Journal of Social Research and Practice* identified six articles which fitted the inclusion criteria of which one was included in the synthesis [33]. Finally, hand searching the reference lists of the eight included articles produced a potential further 11 articles. After this screening, three full-text articles were retrieved and scrutinised but excluded.

Thus from 36 full-text articles retrieved, eight were included in the final synthesis. A full breakdown of the process of retrieving articles with reasons for their exclusion is available in [Fig. 1](#).

3.7. Articles coming close to inclusion but excluded

Four articles were deemed to come close to inclusion by the researchers but were excluded because they contained insufficient data from people with YOD [35–38].

3.8. Study and participant characteristics

Study publication dates spanned more than a decade dating from 2004 to 2015 but with most published since 2011. All were from Western countries, with half from the United Kingdom (UK) [17,18,20,33]. All were cross-sectional and the majority used purposive sampling. Participants were recruited from both statutory and voluntary sector services and most people were interviewed at home or in services such as day centres. Data were collected using face-to-face semi-structured in-depth interviews with research focussing on investigating participants' experiences of living with YOD, although two also specifically looked at perceptions of services. There was a total of 87 participants. Samples sizes ranged from five to 23 with a mean average of 12.4 participants (median nine). There were more male (51) than female (36) participants and most were in their fifties or sixties. Ethnicity was infrequently reported but in the two studies providing this information [17,32] white participants were predominant. The type of YOD was also not

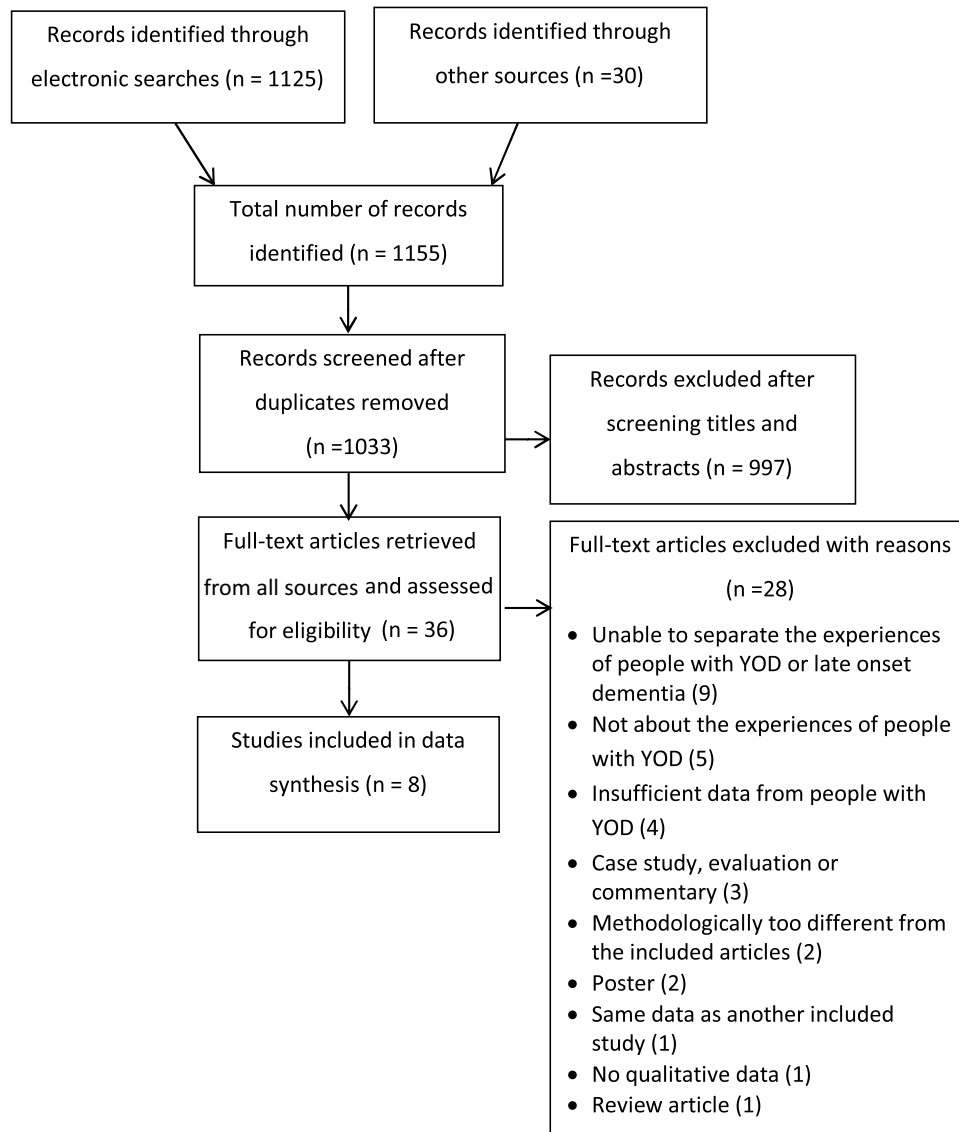


Fig. 1. PRISMA [30] flow diagram showing the process of including and excluding retrieved articles.

always reported but where it was, Alzheimer's disease (39 participants) and FTD dementia (6 participants) were most common. Most studies reported place of residence and the vast majority of participants were living at home, mostly with other people. Two studies described the sample as coming from a mixture of urban and rural environments [17,20] and one as urban [33]. The remaining studies did not mention this. Employment status was rarely reported but where it was, most participants were not working.

In some cases, data analysis was framed by theoretical approaches such as interpretative phenomenological analysis (IPA) and grounded theory [2,18,20,32,33,43], whereas other authors used framework or thematic analysis [16,17]. Full details of study characteristics and participants are available in Tables 2 and 3.

Study quality was variable (Table 3). Scores ranged between six and 10 out of a maximum possible score of 11. Weaknesses included not adequately describing the context for the study and not stating if data analysis involved more than one researcher.

3.9. Overall themes

Perhaps not surprisingly given the stated aims of these investigations, the themes frequently identified in the studies highlighted

participants' overall experiences of living with YOD (Table 4). However, within this more specific themes were identified. These included difficult experiences associated with receiving a diagnosis of YOD, feeling 'too young' to have dementia, and changes in memory and capacity. The diagnosis and symptoms were seen as threatening participants' sense of self, their autonomy and their identity. Their experiences were made more challenging by other people's lack of understanding and the resultant sense of social isolation.

3.10. Line of argument

The diagnosis of dementia at a younger age disrupts the 'normal' life cycle and is 'out of time' with people's expectations and hopes. The diagnosis and the symptoms of YOD with associated losses in abilities lead to changes in self-identity and feelings of disempowerment or powerlessness. Changes in abilities and the stigma of dementia lead to losses and changes in social relationships and avoidance of others. This in turn leads to isolation and feelings of social exclusion. The loss of meaningful activity, often the result of losing their job, is particularly challenging for people with YOD. This and others' sometimes over-protective behaviour

Table 2
Study and participant characteristics.

Author (year) [Country]	Aims	Sample size	Age (yrs)	Gender ratio (M:F)	YOD type	Time since diagnosis (yrs/mths)	Living alone/with others
Beattie, Daker-White, Gilliard & Means (2004) [UK] [20]	Explore the experiences of people with YOD (memory problems, care needs & views of services).	14	Mean: 59.43 Median: NR Range: 41–66	9:5	NR	NR	10 with others, 3 alone
Clemerson, Walsh & Isaac (2013) [UK] [18]	Explore subjective experiences of people with YOD & personal, social & psychological impact of living with YOD.	8	Mean: 55.6 Median: NR Range: 35–63	7:1	AD	Mean: 28.75 mths Median: 18 mths Range: 12–72 mths	6 with others, 2 alone
Griffin, Oyebede & Allen (2015) [UK] [33]	Explore the experiences of people living with bvFTD.	5	Mean: 57.6 Median: 60 Range: 46–62	3:2	BvFTD	3 participants <1 yr, 2 participants 1–2 yrs	All with others
Harris (2004) [USA] [32]	Explore the lived experiences of younger people with dementia.	23	Mean: 56 Median: 54 Range: 43–68	10:13	14 AD, 6 FTD, 3 others	Mean: 3.3 yrs Median: 3 yrs Range: 1–7 yrs	All with others
Johannessen & Moller (2011) [Norway] [2]	Investigate the experiences of living with YOD, & assess the implications for practice service development.	20	Mean: NR Median: 62 Range: 54–67	12:8	NR	Mean: NR Median: NR Range: 4–36 mths	15 with others 5 alone
Pipon-Young, Lee, Jones & Guss (2011) [UK] [17]	Investigate the experiences of people with YOD including: receiving the diagnosis; social challenges; beneficial support; areas in need of change; key problems.	8	Mean: 63.1 Median: 63 Range: 60–67	1:7	7 AD, 1 mixed dementia	Mean: 2.25 yrs Median: 2 yrs Range: 1–5 yrs	NR
Roach & Drummond (2014) [Canada] [16]	Examine the experiences of transition & health expectations in YOD.	9	Mean: 62 Median: 62 Range: 58–68	9:0	7 AD, 2 others	Mean: 2.67 yrs Median: 2 yrs Range: 0–7 yrs	All with others
Rostad, Hellzen & Enmarker (2013) [Norway] [34]	Understand the experiences of people with YOD living at home.	4	Mean: 58.75 Median: 59 Range: 55–62	2:2	3 AD, 1 vascular dementia	1 diagnosed recently, others 24–48 mths	3 with others, 1 alone

Key: AD-Alzheimer's disease; BvFTD-behavioural variant FTD; FTD-frontotemporal dementia; mths- months; NR-not reported; YOD-young onset dementia; yrs-years.

Table 3
Study methods.

Author (year)	Experiences investigated	Sampling method	Inclusion criteria	Data collection (face-to-face unless specified otherwise)	Approach to data analysis	Quality rating (max 11)
Beattie et al. (2004) [20]	Memory problems; service perceptions/use; needs & whether met.	Purposive & snowballing	<65 yrs; YOD diagnosis; service user	In-depth interviews	Grounded theory	8
Clemerson et al. (2013) [18]	Personal, social & psychological impact of living with YOD.	Purposive	<65 yrs; AD diagnosis; British; know diagnosis; mild to moderate YOD	Semi-structured interviews	IPA	10
Griffin et al. (2015) [33]	Perspectives of living with bvFTD.	Purposive	BvFTD diagnosis; know diagnosis	Semi-structured interviews	IPA	9
Harris (2004) [32]	How YOD impacts on the daily life focusing on the social effects.	Purposive	<65 yrs	In-depth, semi-structured interviews, focus group, online interview	Grounded theory	6
Johannessen & Moller (2011) [2]	Living with YOD.	Theoretical	Diagnosis of YOD	Semi-structured interviews	Grounded theory	6
Pipon-Young et al. (2011) [17]	Living with YOD, what is helpful & changing support needs	Purposive	6+ mths since diagnosis of YOD;	Semi-structured interviews	Phase 1: thematic Phase 2: framework	9
Roach & Drummond (2014) [16]	Transitions & health expectations.	Purposive	Diagnosis of YOD, <65 yrs	In-depth semi-structured interviews	Framework analysis	7
Rostad et al. (2013) [34]	Living with YOD	NR	Diagnosis of YOD, <65 yrs, living at home	Narrative interviews	Phenomenological hermeneutic	8

Key: mths-months; NR-not reported; YOD-young onset dementia; yrs-years.

Table 4
Concepts and studies identifying them.

Concepts	Beattie et al. [20]	Clemerson et al. [18]	Griffin et al. [33]	Harris [32]	Johannessen & Moller ²	Pipon-Young et al. [17]	Roach & Drummond [16]	Rostad et al. [34]
Biographical disruption		Feel too young for diagnosis	Perceived discrepancy between age & dementia diagnosis			Perceived difficulty at age of diagnosis		
Diagnosis	Process confusing & distressing	Shock, disbelief	Overwhelming, difficulty understanding diagnosis	Misdiagnosed, fears dismissed	Difficulty obtaining diagnosis, shock	Surprise, shock		
Activity	Need for meaningful activity		Reduced participation	Lack of meaningful activity	Unable to do previous activities	Importance of keeping active, maintain hobbies	Want meaningful activity	Lack of meaningful activity
Loss	Deteriorating memory	Losing competencies		Loss of friends and family, stopping work	Loss of independence	Loss of abilities	Loss of employment	
Self-identity	Treated differently	Loss of skills and self-worth	Dementia label	Loss of sense of self	Feeling 'outside themselves'	Importance of maintaining sense of self		Loss of identity, self-esteem, self respect
Relation-ships	Lack of understanding from others	Lack of understanding and avoidance by others	Treated differently by friends	Lack of understanding from others	Stigma			
Social isolation	Excluded, isolated	Disconnected		Extreme isolation			Spending a lot of time alone	Reduced sense of belonging to society
Emotions		Anger	Sadness	Anger, sadness, frustration	Frustration		Sadness & anxiety	
Coping		Accepting changing abilities	Blaming others			Talking to others with YOD		Preserve hope and willpower

further disempowerment and social isolation. Some people with YOD cope with this by not disclosing their diagnosis and avoiding others which, in turn, exacerbates their social isolation. However, others with YOD opt to try and regain control by reviewing their lives and reconnecting with others. Being with others in similar situations can be a very important means of support (Table 5).

Overall, for these reasons, people with YOD face unique social challenges which arguably go beyond those of people with later onset dementia and result in an even more negative impact on their lives [32].

4. Discussion

The relatively young age of the person with YOD is critical to their experiences. Their age means the diagnosis is much less expected than for older people. This has two main ramifications. Firstly, as it is unexpected, it makes obtaining a diagnosis especially difficult and means that at times the concerns of people with YOD are dismissed by professionals causing anxiety and frustration. Secondly, it also means that others are less likely to understand what the diagnosis means adding to their frustration, and sometimes resulting in social isolation. This difficult situation is compounded by lost or reduced abilities and the associated reductions in meaningful activity. This has huge implications for their sense of agency and self-worth and is strongly associated with disempowerment. Being with others with YOD was suggested by some authors as particularly valuable here.

The participants in these studies faced many challenges and had found the process of diagnosis and coming to terms with their reduced abilities very difficult. Many identified a sense of loss of agency which related both to reduced abilities and to others' at times over-protective behaviour which often led to feeling disem-

powered. However, there was a strong sense that people with YOD did not believe that a dementia diagnosis meant their lives were over [17]. Many wanted to stay engaged with others, to take part in meaningful activity and to be included in society. In short, they wanted to be treated as 'normal human beings' [20].

There were several, perhaps surprising, features of the findings in this review. Firstly, the financial implications of loss of employment in those with YOD have been highlighted in the past [2,16] but were seldom highlighted here. Indeed the focus here appeared to be on loss of meaningful activity, disempowerment and reduced social contact after job losses, rather than financial concerns. Similarly, there was little focus on the impact of the diagnosis of YOD on others. Other research, often from the perspectives of families, has highlighted this, particularly the effect on partners and children [15]. Possible explanations for not identifying this here include the focus on the experiences of the person with dementia or perhaps the reduced insight or empathy often associated with people with dementia [39]. Either way, these are areas for further specific exploration.

4.1. Review strengths and limitations

The review's strengths include its robust and reproducible search strategy, and rigorous assessment of the included studies methodological quality [31]. However, it was limited by not searching the grey literature and only including studies published in English, potentially missing research exploring the experiences of people with YOD from non-English speaking cultures. Nonetheless, the findings help to further our knowledge surrounding the experiences specific to people living with YOD and highlight where areas of support could be increased.

Table 5
Key concepts, second and third order translations.

Key concepts	2nd order interpretations	3rd order interpretations
Age at diagnosis & biographical disruption	The diagnosis of dementia is out of time disrupting their expected life cycle, hopes & expectations [18] Great care needs to be taken in how assessment & diagnosis are undertaken [20]	This biographical disruption exacerbates the negative experience of receiving a diagnosis of dementia. The opportunity to associate with others in similar situations (both those with YOD & LOD) can be very beneficial as it provide social contact with people in a similar position to understand better the challenges of living with YOD. It can help to reduce social isolation.
Emotional responses including anger, sadness & bewilderment Loss of meaningful activity e.g. employment & life-style restrictions e.g. driving. Lack of meaningful activity also leads to boredom	Distress at a diagnosis of YOD is distressing & often related to it being 'out of time' Loss of meaningful activity has a negative impact on their sense of self [33], purpose & role fulfilment [16]. Keeping up with activities can help maintain a positive identity [17] Restrictions in activities increase their dependence on others [34].	The restrictions in activities experienced by people with YOD have far reaching implications for their autonomy & sense of worth. Providing appropriate support (e.g. from employers) to maintain meaningful activity for as long as possible would help increase their sense of agency & reduce social isolation. Maintaining autonomy is very important to people with YOD. Although some abilities are reduced or lost, those supporting people with YOD should avoid being over-protective.
Loss of abilities & disempowerment	Loss of abilities & risk aversion by others results in a sense of loss of control & agency. Reduced ability to communicate leads to increased isolation.	
The diagnosis & symptoms of YOD lead to changes in self-identity & loss of self	Having dementia is stigmatising & can lead to avoidance of social situations [2]	The stigma of dementia is perceived as even greater in people with YOD than those with LOD which means that people with YOD may be more unwilling to disclose their diagnosis. This combined with avoiding social situations increases social isolation & exclusion. The symptoms of dementia & the diagnosis can lead to a vicious circle of avoidance of other people leading to greater social isolation & reduced sense of self-worth. Some people with YOD strive to continue to live a 'normal' life with activities meaningful to them but others with YOD avoid others which increases their social exclusion. Encouraging those with YOD to disclose their diagnosis where appropriate & to continue with meaningful enjoyable activities & being with others, may be beneficial.
Changes in social relationships, social isolation & social exclusion Coping	Disconnection from others leads to loneliness & powerlessness [18] People with YOD may cope by avoiding others & not revealing their diagnosis. Others review their life & try to reconnect with others. Some people search out meaningful activity whilst others take the initiative to preserve faith & control over their lives [34]	

5. Conclusions, service implications and future research

Although they have much in common with people diagnosed with dementia in later life, people with YOD have additional challenges. Receiving a diagnosis of dementia is a difficult process for anyone but is arguably even harder for those with YOD. Not only is obtaining a diagnosis more challenging for younger people but it also feels particularly unexpected and out of time with their biography both to them and those who know them. Particular care needs to be taken by clinicians in explaining the diagnosis both to the person with YOD and their families. Negative reactions and lack of understanding by others, including employers, mean that people with YOD may avoid social contact. Combined with loss of interaction with others through loss of employment this can result in social isolation and social exclusion. Several authors here highlighted the value of peer support [20,32] either from others with YOD or more generally others with dementia. There is evidence that peer support can benefit family or informal carers [40] but this suggests that research focussing solely of the impact of peer support specifically on those with YOD and their family carers is warranted. In addition, all the studies here were cross-sectional and longitudinal research is also needed perhaps focussing on dyads rather than people with YOD and their carers separately.

The voluntary sector may be able to play an important role here. In the UK and elsewhere, there has been a relatively recent inter-

est in peer support offered in the voluntary sector both for people with long term conditions and their family carers. This interest may be, in part, because it can avoid medicalising the condition [41] which is perhaps particularly pertinent for people with YOD. Studies included here suggest that people with YOD may enjoy and benefit from interacting with others with YOD and with people such as their carers who are well placed to understand their situation and challenges. In addition, there may be a role for the voluntary sector to support work-based interventions for people with YOD to allow meaningful activity to continue as long as possible [22]. Programmes that offer people with YOD opportunities to be involved in such activities can increase their self-worth [22] but also allow both the person with YOD and their families to continue to feel 'normal'. It may also simultaneously provide a break for the family carer.

Contributors

With the support of RS, NG conceived the focus of the review and together they developed the search strategy.

RS undertook the literature searches and both authors selected the relevant articles, undertook the data extraction and performed the quality ratings.

NG led on writing the paper but both authors developed the final manuscript.

Conflict of interest

The authors have no conflict of interest to declare.

Funding

No funding was received for this article.

Provenance and peer review

This article has undergone peer review.

References

- [1] M.N. Rossor, N.C. Fox, C.J. Mummery, J.M. Schott, J.D. Warren, The diagnosis of young-onset dementia, *Lancet Neurol.* 9 (8) (2010) 793–806, [http://dx.doi.org/10.1016/s1474-4422\(10\)70159-9](http://dx.doi.org/10.1016/s1474-4422(10)70159-9).
- [2] A. Johannessen, A. Moller, Experiences of persons with early-onset dementia in everyday life: a qualitative study, *Dementia* 12 (4) (2011) 410–424, <http://dx.doi.org/10.1177/1471301211430647>.
- [3] N. Rudman, J. Oyeboode, C. Jones, P. Bentham, An investigation into the validity of effort tests in a working age dementia population, *Aging Ment. Health* 15 (1) (2011) 47–57, <http://dx.doi.org/10.1080/13607863.2010.508770>.
- [4] M. Lamberta, H. Bickelb, M. Prince, L. Fratiglionid, E. von Straussd, D. Frydeckae, A. Kiejnae, J. Georges, E. Reynishg, Estimating the burden of early onset dementia; systematic review of disease prevalence, *Eur. J. Neurol.* 21 (4) (2014) 563–569, <http://dx.doi.org/10.1111/ene.12325>.
- [5] R. Harvey, M. Skelton-Robinson, M. Rossor, The prevalence and causes of dementia in people under the age of 65 years, *J. Neurol. Neurosurg. Psychiatry* 74 (2003) 1206–1209, <http://dx.doi.org/10.1136/jnnp.74.9.1206>.
- [6] S. Lockeridge, J. Simpson, The experience of caring for a partner with young onset dementia: how younger carers cope, *Dementia* 12 (5) (2013) 635–651, <http://dx.doi.org/10.1177/1471301212440873>.
- [7] D. van Vliet, M.E. de Vugt, C. Bakker, Y.A.L. Pijnenburg, M.J.F.J. Vernooij-Dassen, R.T.C.M. Koopmans, F.R.J. Verhey, Time to diagnosis in young-onset dementia as compared with late-onset dementia, *Psychol. Med.* 43 (2) (2013) 423–432, <http://dx.doi.org/10.1017/S0033291712001122>.
- [8] D. van Vliet, M. de Vugt, P. Aalten, C. Bakker, Y.A. Pijnenburg, M.J. Vernooij-Dassen, R.Y. Koopmans, F.R. Verhey, Prevalence of neuropsychiatric symptoms in young-onset compared to late-onset Alzheimer's disease – part 1: findings of the two-year longitudinal NeedYD-study, *Dement. Geriatr. Cogn. Disord.* 34 (5–6) (2012) 319–327, <http://dx.doi.org/10.1159/000342824>.
- [9] M.F. Mendez, The accurate diagnosis of early-onset dementia, *Int. J. Psychiatry Med.* 36 (4) (2006) 401–412, <http://dx.doi.org/10.2190/q6j4-r143-p630-kw41>.
- [10] R.T. Vieira, L. Caixeta, S. Machado, A.C. Silva, A.E. Nardi, Epidemiology of early-onset dementia: a review of the literature, *Clin. Pract. Epidemiol. Ment. Health* 9 (2013) 88–95, <http://dx.doi.org/10.2174/1745017901309010088>.
- [11] R. Davies, M. Doran, A.J. Larner, Diagnosis and management of early-onset dementia, *Prog. Neurol. Psychiatry* 15 (4) (2011) 12–16, <http://dx.doi.org/10.1002/pnp.205>.
- [12] K. Jefferies, N. Agrawal, Early-onset dementia, *Adv. Psychiatr. Treat.* 15 (5) (2009) 380–388, <http://dx.doi.org/10.1192/apt.bp.107.004572>.
- [13] F. Ducharme, M.J. Kergoat, P. Antoine, F. Pasquier, R. Coulombe, The unique experience of spouses in early-onset dementia, *Am. J. Alzheimers Dis. Other Dement.* 28 (2013) 634–641, <http://dx.doi.org/10.1177/1533317513494443>.
- [14] M. McCabe, E. You, G. Tantangelo, Hearing their voice: a systematic review of dementia family caregivers' needs, *Gerontologist* (2016), <http://dx.doi.org/10.1093/geront/gnw078>, Epub ahead of print.
- [15] K. Hutchinson, C. Roberts, S. Kurrle, M. Daly, The emotional well-being of younger people having a parent with younger dementia, *Dementia* 0 (2014) 1–20, <http://dx.doi.org/10.1177/1471301214532111>.
- [16] P. Roach, N. Drummond, 'It's nice to have something to do': early onset dementia and maintaining purposeful activity, *J. Psychiatr. Ment. Health Nurs.* 21 (10) (2014) 889–895, <http://dx.doi.org/10.1111/jpm.12154>.
- [17] F.E. Pipon-Young, K.M. Lee, F. Jones, R. Guss, I'm not all gone, I can still speak: the experiences of younger people with dementia. An action research study, *Dementia* 11 (5) (2012) 597–616, <http://dx.doi.org/10.1177/1471301211421087>.
- [18] n.S. Walsh Clemerso, C. Isaac, Towards living well with young onset dementia: an exploration of coping from the perspective of those diagnosed, *Dementia* 13 (4) (2014) 451–466, <http://dx.doi.org/10.1177/1471301214247419>.
- [19] A.E.H. Dam, M.E. de Vugt, I.P.M. Klinkenber, F.R.J. Verhey, M.P.J. van Boxtel, A systematic review of social support interventions for caregivers of people with dementia: are they doing what they promise? *Maturitas* 85 (2016) 117–130, <http://dx.doi.org/10.1016/j.maturitas.2015.12.008>.
- [20] A. Beattie, G. Daker-White, J. Gilliard, R. Means, 'How can they tell?' A qualitative study of the views of younger people about their dementia and dementia care services, *Health Soc. Care Community* 12 (4) (2004), <http://dx.doi.org/10.1111/j.1365-2524.2004.00505.x>.
- [21] E.L. Goldberg, Filling an unmet need: a support group for early stage/young onset Alzheimer's disease and related dementias, *W. V. Med. J.* 107 (3) (2011) 66–68.
- [22] A. Richardson, G. Pedley, F. Akhtar, F. Pelone, J. Chang, W. Muleya, N. Greenwood, Psychosocial interventions for people with young onset dementia and their carers: a systematic review, *Int. Psychogeriatr.* 13 (2016) 1–14, <http://dx.doi.org/10.1017/S1041610216000132>.
- [23] M. von Kutzleben, W. Schmid, M. Halek, B. Holle, S. Bartholomeyczik, Community-dwelling persons with dementia: what do they need? What do they demand? What do they do? A systematic review on the subjective experiences of persons with dementia, *Aging Ment. Health* 16 (3) (2012) 378–390, <http://dx.doi.org/10.1080/13607863.2011.614594>.
- [24] G.W. Noblit, R.D. Hare, *Meta-Ethnography: Synthesizing Qualitative Studies*, Sage, Newbury Park, 1988.
- [25] R. Campbell, P. Pound, M. Morgan, G. Daker-White, N. Britten, R. Pill, L. Yardley, C. Pope, J. Donovan, Evaluating meta-ethnography: systematic analysis and synthesis of qualitative research, *Health Technol. Assess.* 15 (43) (2011), <http://dx.doi.org/10.3310/hta15430>.
- [26] P. Pound, N. Britten, M. Morgan, L. Yardley, C. Pope, G. Daker-White, R. Campbell, Resisting medicines: a synthesis of qualitative studies of medicine taking, *Soc. Sci. Med.* 61 (1) (2005) 133–155, <http://dx.doi.org/10.1016/j.socscimed.2004.11.063>.
- [27] M. Dixon-Woods, S. Agarwal, D. Jones, B. Young, A. Sutton, Synthesizing qualitative and quantitative evidence: a review of possible methods, *J. Health Serv. Res. Policy* 10 (1) (2005) 45–53, <http://dx.doi.org/10.1258/1355819052801804>.
- [28] N. Greenwood, A. Mackenzie, Informal caring for stroke survivors: meta-ethnographic review of qualitative literature, *Maturitas* 66 (3) (2010) 268–276, <http://dx.doi.org/10.1016/j.maturitas.2010.03.017>.
- [29] Centre for Reviews Dissemination (CRD), *Systematic Reviews: CRD's Guidance for Undertaking Reviews in Health Care*, CRD: University of York, 2009.
- [30] D. Moher, A. Liberati, J. Tetzlaff, J. et al., Preferred reporting items for systematic reviews and meta-analyses: the PRISMA statement, *Ann. Intern. Med.* 151 (4) (2009) 264–269, <http://dx.doi.org/10.7326/0003-4819-151-4-200908180-00135>.
- [31] N. Greenwood, A. Mackenzie, G.C. Cloud, N. Wilson, Informal primary carers of stroke survivors living at home—challenges, satisfactions and coping: a systematic review of qualitative studies, *Disabil. Rehabil.* 31 (5) (2009) 337–351, <http://dx.doi.org/10.1111/10.1080/09638280802051721>.
- [32] P.B. Harris, The perspective of younger people with dementia, *Soc. Work Ment. Health* 2 (4) (2004) 17–36, http://dx.doi.org/10.1300/j200v02n04_02.
- [33] J. Griffin, J.R. Oyeboode, J. Allen, Living with a diagnosis of behavioural-variant frontotemporal dementia: the person's experience, *Dementia* 1–21 (2015), <http://dx.doi.org/10.1177/1471301214568164>, Epub ahead of print.
- [34] D. Rostad, O. Hellzén, I. Enmarker, The meaning of being young with dementia and living at home, *Nurs. Rep.* 3 (1) (2013) e3, <http://dx.doi.org/10.4081/nursrep.2013>.
- [35] L. Carone, V. Tischler, T. Denning, Football and dementia: a qualitative investigation of a community based sports group for men with early onset dementia, *Dementia* 1–19 (2014), <http://dx.doi.org/10.1177/1471301214560239>, Epub ahead of print.
- [36] D. Edvardsson, D. Fetherstonhaugh, R. Nay, Promoting a continuation of self and normality: person-centred care as described by people with dementia, their family members and aged care staff, *J. Clin. Nurs.* 19 (17–18) (2009) 2611–2618, <http://dx.doi.org/10.1111/j.1365-2702.2009.03143.x>.
- [37] J.M. Kinney, C.S. Kart, L. Reddecliff, 'That's me, the Goother': Evaluation of a program for individuals with early-onset dementia, *Dementia* 10 (3) (2011) 361–377, <http://dx.doi.org/10.1177/1471301211407806>.
- [38] E. Wawrzynicz, P. Antoine, F. Ducharme, M.J. Kergoat, F. Pasquier, Couples' experiences with early-onset dementia: an interpretative phenomenological analysis of dyadic dynamics, *Dementia* 1–18 (2014), <http://dx.doi.org/10.1177/1471301214554720>, Epub ahead of print.
- [39] D. Morhardt, S. Weintraub, B. Khayum, J. Robinson, J. Medina, M. O'Hara, M. Mesulam, E.J. Rogalski, The CARE Pathway Model for Dementia: psychosocial and rehabilitative strategies for care in young-onset dementias, *Psychiatr. Clin. North Am.* 38 (2) (2015) 333–352, <http://dx.doi.org/10.1016/j.psc.2015.01.005>.
- [40] R. Smith, R. N. Greenwood, The impact of volunteer mentoring schemes on carers of people with dementia and volunteer mentors: a systematic review, *Am. J. Alzheimers Dis. Other Dement.* 29 (1) (2014) 8–17, <http://dx.doi.org/10.1177/1533317513505135>.
- [41] G. Charlesworth, K. Burnell, J. Beecham, Z. Hoare, J. Hoe, J. Wenborn, M. Knapp, I. Russel, R. Wood, M. Orrell, Peer support for family carers of people with dementia, alone or in combination with group reminiscence in a factorial design: study protocol for a randomised controlled trial, *Trials* 12 (2011), <http://dx.doi.org/10.1186/1745-6215-12-205>.