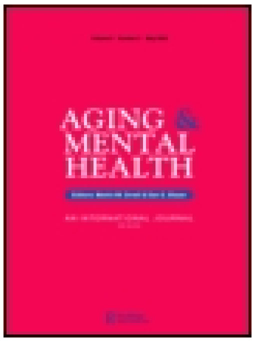

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# Age-appropriate services for people diagnosed with young onset dementia (YOD): a systematic review

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**ABSTRACT**

**Background:** Literature agrees that post-diagnostic services for people living with young onset dementia (YOD) need to be age-appropriate, but there is insufficient evidence of ‘what works’ to inform service design and delivery.
**Objective:** To provide an evidence base of age-appropriate services and to review the perceived effectiveness of current interventions.
**Methods:** We undertook a systematic review including all types of research relating to interventions for YOD. We searched PubMed, CINHAL Plus, SCOPUS, EBSCO Host EJS, Social Care Online and Google Scholar, hand-searched journals and carried out lateral searches (July–October 2016). Included papers were synthesised qualitatively. Primary studies were critically appraised.
**Results:** Twenty articles (peer-reviewed [*n* = 10], descriptive accounts [*n* = 10]) discussing 195 participants (persons diagnosed with YOD [*n* = 94], caregivers [*n* = 91] and other [*n* = 10]) were identified for inclusion. Services enabled people with YOD to remain living at home for longer. However, service continuity was compromised by short-term project-based commissioning and ad-hoc service delivery.
**Conclusion:** The evidence on the experience of living with YOD is not matched by research and the innovation needed to mitigate the impact of YOD. The inclusion of people with YOD and their caregivers in service design is critical when planning support in order to delay institutional care

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**KEYWORDS**
Young onset dementia (YOD); age-appropriate services; effectiveness; social connectedness; inclusion

## Introduction

In the UK, approximately 5% of people living with dementia (*n* = 42,500) have been diagnosed with young onset dementia (YOD) (age <65 at diagnosis) (Prince et al., 2014). Their support needs differ considerably from those of people with dementia in later years (AM Beattie, Daker-White, Gilliard, & Means, 2002; Sansoni et al., 2016; Sansoni et al., 2014; Thompson, 2011; Tyson, 2007; Westera et al., 2014) as they still have comparatively high levels of physical fitness and are not well-matched to join more sedentary services designed for older age groups (Pamela Roach, Drummond, & Keady, 2016; Tolhurst, Bhattacharyya, & Kingston, 2014). They also need to negotiate changing family dynamics with a particular focus on children and teenagers (Chow, Pio, & Rockwood, 2011; Gibson, Anderson, & Acocks, 2014; Hutchinson, Roberts, Daly, Bulsara, & Kurrle, 2016; Johannessen, Bruvik, & Hauge, 2015; Lockeridge & Simpson, 2013; Morhardt et al., 2015; P. Roach & Drummond, 2014; Shnall, 2015; Svanberg, Spector, & Stott, 2011; Thompson, 2011), accommodate changing care needs along the disease trajectory (Bakker et al., 2013; Bakker et al., 2010) and need to seek financial and legal advice as employment is discontinued and spouses give up work to become the main caregiver or take on work to compensate for lost income (Alzheimer’s Society, 2015; Chaplin & Davidson, 2016; Picard, Pasquier, Martinaud, Hannequin, & Godefroy, 2011; Ritchie, Banks, Danson, Tolson, & Borrowman, 2015). The need for age-appropriate service design for people diagnosed with YOD has been discussed extensively in a number of comprehensive, non-systematic literature reviews of YOD (Baptista et al., 2016; Beattie et al., 2002; Cabote, Bramble, & McCann, 2015; Millenaar et al., 2016; Richardson et al., 2016; Sansoni et al., 2016; Sansoni et al., 2014; Westera et al., 2014), reports (Austin A, 2016; Brown et al., 2012; Clayton-Turner et al., 2016) and papers across various disciplines (Armari, Jarmolowicz, & Panegyres, 2013; Bakker et al., 2014; Bakker et al., 2010; Ducharme, Kergoat, Antoine, Pasquier, & Coulombe, 2014a; Ducharme et al., 2014b; Gibson et al., 2014; Hvidsten et al., 2014), but there is little evidence of ‘what works’ in order to inform service design and delivery. The aim of this systematic review was to identify service outcomes that were perceived as important and effective by people living with YOD and their caregivers. The specific outcomes we looked for were (a) what is the current range of service provision for people diagnosed with YOD and their caregivers/supporters? (b) What are the referral mechanisms involved? (c) How is effectiveness defined and evaluated? (d) What are the associated costs?

## Methodology

This systematic review had three stages: to understand the range of post-diagnostic service needs and service provision, we undertook a comprehensive scoping review. To understand service users’ perceived effectiveness of services, we held group discussions with patient and public involvement (PPI) representatives, who were people living with YOD and their caregivers, in the two study sites. The findings of the scoping review and group discussions informed the focus of the systematic review (Box 1). This paper reports on the

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systematic review, which was registered on the International Prospective Register of Systematic Reviews (PROSPERO) (PROSPERO, 2016), registration number 42016043103.

**Box 1:**

***Themes in the scoping review:*** The need for age-appropriate services; absence of clarity around referral mechanisms; a lack of knowledge about how to find/access age-appropriate services; only a few post-diagnostic, age-appropriate services had been evaluated; some suggestions for service development.

***Conversations with PPI representatives*** verified and corroborated findings from the scoping review.

Findings from the scoping review and PPI discussions will be reported elsewhere.

### *Inclusion and exclusion criteria*

Inclusion criteria were services, interventions and approaches to care for people diagnosed with YOD. Of primary interest were types of interventions, referral mechanism, cost-indicators, perceived service user satisfaction, and health and well-being and quality of life to address the gaps identified by the scoping review. Exclusion criteria: if inclusion criteria were not met. All types of empirical studies published in English were included. We also included descriptive accounts of the development and outcomes of YOD services.

### *Search strategy*

Electronic searches were performed on PubMed, CINHAL Plus (Cumulative Index to Nursing and Allied Health Literature), SCOPUS, EBSCO Host EJS, Social Care Online and Google Scholar (July-October 2016). Search queries are shown in Box 2. Google and Google Scholar were used to search for grey literature such as reports and non-peer reviewed publications. We also used the 'citation option' on Google Scholar. Lateral searches were performed by searching reference lists of peer-reviewed papers and literature reviews. The subject specific Journal 'Dementia: the international journal of social research' was hand searched from 2009–2016 as it is currently not included in any of the databases searched.

**Box 2: Search query**

"Early onset" OR "young onset" AND Dementia OR Alzheimer* OR "Lewy body" OR vascular OR frontotemporal OR PCA OR CADASIL AND Service OR referral OR intervention OR care OR support AND Evaluation AND "Co-production" OR "co-design" OR participation OR engagement OR "co-creation" OR "co-delivery" OR involvement AND "Social return on investment" OR SROI.

No restrictions were applied to study design or date.

### *Screening and data extraction*

Papers were downloaded into EndNote bibliographic software (Reuters, 2016). Duplicates were removed and three researchers (AM, EM and JM) double-screened (AM and EM; AM and JM) the remaining papers for inclusion eligibility. Disagreements were resolved by discussion.

### *Data analysis and thematic synthesis*

Data addressing the primary and secondary outcomes stipulated for this review were coded, analysed conceptually and synthesised thematically using QSR software NVivo 11 (NVivo, 2012). As only one quantitative study was found, the results are presented narratively.

### *Quality appraisal*

Primary studies were assessed for quality. The Critical Appraisal Skills Programme (CASP, 2014) was used to assess qualitative and mixed methods papers (Box 3). The paper using quantitative methods was assessed using the Cochrane risk of bias tool (Higgins & Green, 2011).

**Box 3: Quality assessment**

A paper was scored 'Low' if it scored 5 or <5 of 10 items on the CASP (2014) checklist on credibility, reliability and transferability;

Medium: scored 6-8 items

High: an overall score (including ethics and value of research) of 9 -10 out of 10

The paper using quantitative methods was assessed using the Cochrane risk of bias tool [41].

### *Ethical approval*

Permission for discussions with PPI representatives who attended support groups was granted by the Alzheimer's Society Research Office. The University of Hertfordshire Ethics Protocol Number is HSK/SF/UH/02340.

## Results

Database searches and hand-searched articles produced 403 records for screening (Figure 1). Twenty articles were included, 10 of which were primary studies (Beattie, Daker-White, Gilliard, & Means, 2004; Carone, Tischler, & Dening, 2016; Hewitt, Watts, Hussey, Power, & Williams, 2013; Kelly & Innes, 2016; Kinney, Kart, & Reddecliff, 2011; O'Connell et al., 2014; Parahoo, Campbell, & Scoltock, 2002; Perkins & Poynton, 1990; Reed, Cantley, Clarke, & Stanley, 2002; Robertson & Evans, 2015). Ten were descriptive accounts of interventions (Barker & Johnson, 2008; Chaston, Pollard, & Jubb, 2004; Craig & Strivens, 2016; Davies-Quarrell et al., 2010; LaFontaine, 2004; Parkes & Ward, 2015; Ramluggun & Ogo, 2016; J. Robertson, Evans, & Horsnell, 2013; Tonga, Arnevik, Werheid, & Ulstein, 2016; Wheeler et al., 2015).

### *Study characteristics*

The 20 included articles spanned a time frame of 26 years (1990–2016) (Table 1). They covered five countries. All publications were in English. Seven papers used qualitative methodologies, one study used standardised measures (Perkins & Poynton, 1990) and two used mixed methods (Hewitt et al., 2013; Kelly & Innes, 2016). The remaining 10 papers provide descriptive accounts and were treated as qualitative data. The 195 study participants included were individuals diagnosed with YOD (*n* = 94), caregivers (*n* = 91) and health care

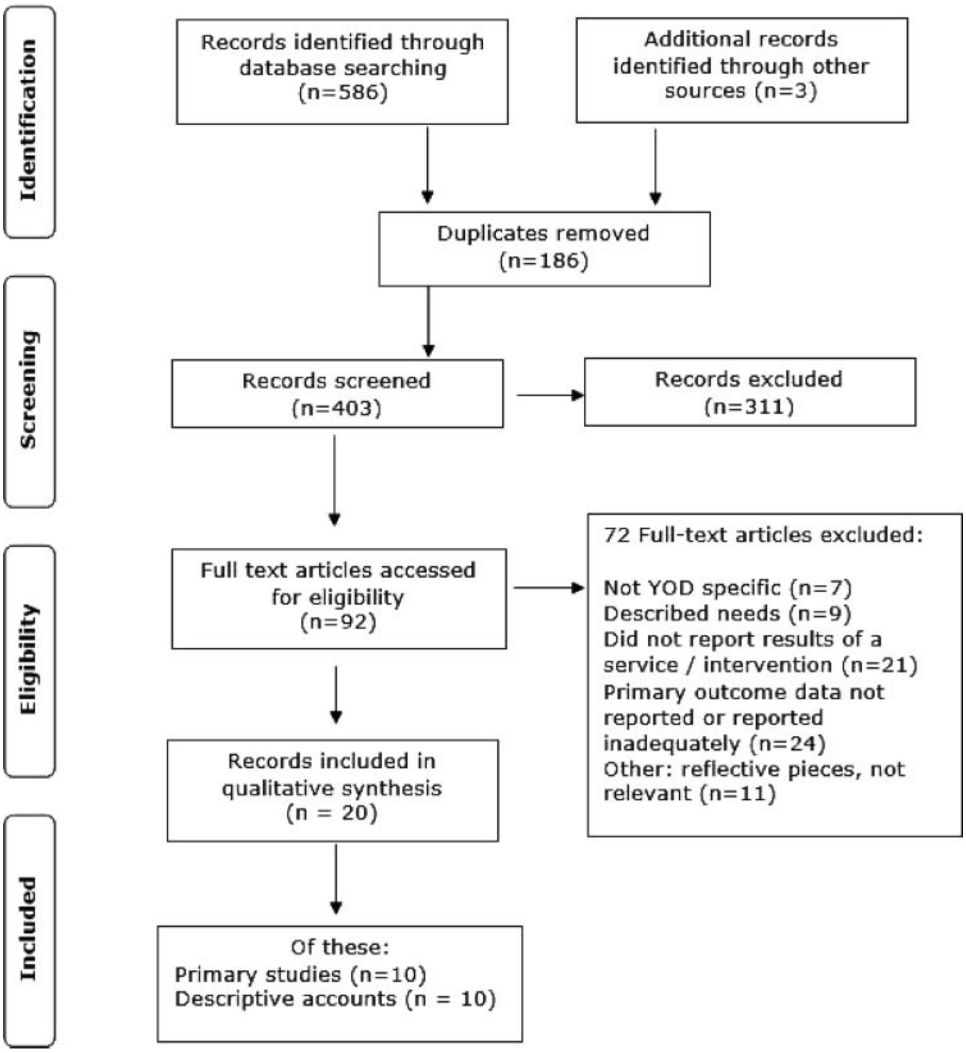

**Figure 1.** Flow diagram of literature review.

professionals and/or service providers (*n* = 10). Eighteen of the reported services were YOD specific. The remaining two interventions were not specifically designed for YOD, but applied to people diagnosed with YOD and reported as such (Kelly & Innes, 2016; Tonga et al., 2016). Six interventions showed evidence of user-involvement in the design of services (Barker & Johnson, 2008; Chaston et al., 2004; Davies-Quarrell et al., 2010; Kinney et al., 2011; O'Connell et al., 2014; Parkes & Ward, 2015).

As indicated, only 10 peer-reviewed papers were available for inclusion. Of these, only one paper (Perkins & Poynton, 1990) used standardised measures. Consequently, this systematic review is unable to report on the 'effectiveness' of interventions. Instead, we refer to 'perceived effectiveness' as reported in the peer reviewed papers. This refers to all outcomes. The results presented in this systematic review are therefore presented thematically. Types of interventions, referral mechanisms and outcomes are presented in Table 2.

### *Types of services/interventions*

Post-diagnostic support services designed specifically for people diagnosed with YOD and their caregivers varied considerably and comprised community services, hospital-based services and a mix of statutory and non-statutory services.

Community-based services included day centres for people with YOD (Beattie et al., 2004), respite services offering YOD-specific support (Parahoo et al., 2002), community-based support groups (Carone et al., 2016; Chaston et al., 2004; Davies-Quarrell et al., 2010), volunteer-led activities (Kinney et al., 2011), work-based programmes (Robertson & Evans, 2015; J. Robertson et al., 2013) and on-line support groups (Craig & Strivens, 2016; O'Connell et al., 2014). Day centres provided activities facilitated by trained staff for a number of hours per week (Beattie et al., 2004), whilst domiciliary respite services were designed to support caregivers, enabling families to keep people living with YOD in their own homes for longer (Parahoo et al., 2002). Community-based and volunteer-led support groups networked with local NHS Trusts (Chaston et al., 2004), local Alzheimer's Society services (Carone et al., 2016; Davies-Quarrell et al., 2010; Kinney et al., 2011) and organisations such as a local zoo (Kinney et al., 2011), a football club (Carone et al., 2016) and organisations hosting structured gardening programmes (Hewitt et al., 2013). Work-based programmes were organised between a respite service, a local hardware store and volunteers (Robertson & Evans, 2015; J. Robertson et al., 2013) and participants diagnosed with YOD were de-briefed daily by trained staff (Robertson et al., 2013).

Hospital-based services included group counselling for relatives of hospitalised young-onset dementia patients in the

**Table 1.** Overview of study characteristics.

| | | | | | |
|---|---|---|---|---|---|
| Year | 1990–2008 ($n = 7$) | | | | |
| | 2010–2016 ($n = 13$) | | | | |
| Country | AUS ($n = 3$) | | | | |
| | CAN ($n = 1$) | | | | |
| | NOR ($n = 1$) | | | | |
| | UK ($n = 14$) | | | | |
| | USA ($n = 1$) | | | | |
| Language | English | | | | |
| Primary study | Yes ($n = 10$) | | | | |
| | No ($n = 10$) | | | | |
| Methods used | Qualitative ($n = 7$) | | | | |
| | Quantitative ($n = 1$) | | | | |
| | Mixed methods ($n = 2$) | | | | |
| | Descriptive (qualitative) accounts ($n = 10$) | | | | |
| Types of participants | Individuals diagnosed with young onset dementia (YOD), their caregivers, support staff, clinicians | | | | |
| Number of participants | | YOD | Caregiver | Other | Total |
| | 10 primary studies | 76 | 78 | 10 | 164 |
| | 10 descriptive papers | 18 | 13 | 0 | 31 |
| | Total | 94 | 91 | 10 | 195 |
| Service YOD specific | Yes ($n = 18$) | | | | |
| | No ($n = 2$) Not designed for YOD, but applied to YOD and reported as such | | | | |
| Evidence of user involvement in the design of services | Yes ($n = 6$) | | | | |
| | No/not applicable ($n = 14$) | | | | |
| Quality assessment of primary studies | | Low | Medium | High | |
| | Qualitative studies (7) | 0 | 4 | 3 | |
| | Mixed methods (2) | 0 | 0 | 2 | |
| | Quantitative study (1) | 1 | | | |

UK (Perkins & Poynton, 1990), cognitive therapy applied to people diagnosed with YOD in Norway (Tonga et al., 2016), and telehealth on-line support groups for remote rural groups in Canada (O'Connell et al., 2014).

In the UK, National Health Service (NHS) Trusts were also part of a mix of statutory and non-statutory post-diagnostic services. NHS Trusts liaised with a range of agencies that offer social, financial and practical dementia support ranging from occupational therapy and buddy systems to bus passes and worked with local and district councils, job centres and debt recovery services to offer help with legal issues and financial guardianship (Ramluggun & Ogo, 2016). They also partnered with a Local Citizen's Bureau (Wheeler et al., 2015), worked with local agencies and the Alzheimer's Society (Parkes & Ward, 2015), offered a therapeutic support group for women (Barker & Johnson, 2008), or offered their NHS Trust hospital garden for gardening programmes (Hewitt et al., 2013). Local Councils in Scotland liaised with the Alzheimer's Society and Health and Community Health Partnerships to offer YOD-specific services (Kelly & Innes, 2016).

### *Referral mechanisms*

Post-diagnostic services offered by NHS Trusts could only be accessed by people with YOD via referrals from memory clinics (Ramluggun & Ogo, 2016; Tonga et al., 2016; Wheeler et al., 2015) and old-age psychiatry/psycho geriatricians or neurologists (Reed et al., 2002). Partnerships between Local Councils, community-based organisations and agencies such as the Alzheimer's Society required referrals from health and social care professionals (Kelly & Innes, 2016), local clinics (O'Connell et al., 2014) or local dementia services (Hewitt et al., 2013). Referrals to community-based services were made primarily via non-statutory, third-sector agencies such as the Alzheimer's Society (Carone et al., 2016; Davies-Quarrell et al., 2010; Kelly & Innes, 2016; Kinney et al., 2011; LaFontaine, 2004; Parahoo et al., 2002; Parkes & Ward, 2015) and local dementia services (Hewitt et al., 2013). On-line support groups (Craig & Strivens, 2016) and local projects run by volunteers (Chaston et al., 2004; Jacinta Robertson & Evans, 2015) were publicised via leaflets and word of mouth. The reviewed papers reported no evidence of formal self-referral, but people with YOD were free to attend community-based services of interest.

### *Perceived service user satisfaction, health and well-being, quality of life*

Outcomes such as perceived service user satisfaction, health and well-being and quality of life were not reported as discrete entities, but described in terms of concepts such as social connectedness, participation and inclusion, adjustment, normalisation and support for caregivers.

### *Social connectedness and participation, adjustment and normalisation*

The scoping review and discussions with people living with YOD identified that services should support inclusion and ongoing participation in society. The systematic review found several examples of this. For instance, participation in activities such as art work or physical work (Beattie et al., 2004) and doing 'ordinary things' like going down to the pub (Beattie et al., 2004) reportedly created a sense of continuation of 'normal life'. Projects were also mindful of gender. One football club offered an 'all male environment' that benefited from a high staff-YOD ratio (Carone et al., 2016) which in turn provided an opportunity for family caregivers to have conversations during that time. Another project supported a forum for women diagnosed with YOD to meet, have lunch and learn about emotion focused coping (Barker & Johnson, 2008). Interventions also included work-based programmes such as gardening (Hewitt et al., 2013), working in a Zoo (Kinney et al., 2011) or in a hardware store (Robertson et al., 2013), all of which facilitated a sense of leading a 'normal' life. The anticipation of engaging in meaningful activities was reported as leading to improved motivation more generally, which was

Table 2. Types of interventions and concepts related to primary outcomes.

| Primary study | Author, year, country | Type of intervention | | Primary outcomes: perceived service user satisfaction, health and wellbeing, quality of life were discussed in terms of the following concepts: *Social connectedness, social participation and inclusion; processes of adjustment; normalisation; support for caregivers* |
|---|---|---|---|---|
| Yes | Beattie et al_2004_UK (A. Beattie et al., 2004) | Community based | Day centres for YOD | Service users felt comfortable in specialist, age-appropriate day service; a place to meet new friends/able to socialise; opportunities to engage in meaningful activities (art work, material work); doing 'ordinary' things, like going down to the pub; service contributes to Quality of Life as service users feel included and accepted; no need to keep explaining to others |
| Yes | Carone et al_2014_UK (Carone et al., 2016) | Community based | Local football club activities | The opportunity for people to feel 'normal' (in the company of others with a diagnosis); availability of skilled coaches; can offer highly personalised and person-centred approach to intervention due to high staff-PWD ratio; provides the 'all male' environment appreciated by men; a sense of 'normality', a sense of 'belonging' to a sports group.<br>Family carers/wives: peer support, time for self (reported and observed) |
| Yes | Hewitt et al_2013_UK (Hewitt et al., 2013) | Community based + NHS | Gardening programme | Benefits: enhanced self-identity (having a purpose); companionship; improved orientation (knew when it was Friday – gardening day); safe physical activity; respite for carer—knowing that loved one was 'looked after'; enjoyment (both person diagnosed with YOD and carer); feeling useful (achievement); feeling valued; carers' reduced anxiety; improved mood of person with YOD; an increase in mean weekly well-being scores |
| Yes | Kelly & Innes_2016_UK (Kelly & Innes, 2016) | Mixed; Statutory and non-statutory | Post diagnostic support service in Scotland | Evaluation of pilot project; Facilitated independence and increased self-confidence and motivation; increased quality of life via support in claiming benefits; having a named person to contact; facilitated social contact by providing transport and offering a choice of events being planned; this created a sense of normalcy; learning how to go about organising 'self-directed support' |
| Yes | Kinney et al_2011_US (Kinney et al., 2011) | Community-based (volunteer-led) | Volunteer programme, Zoo | Programme provided a break from the day-to-day routine for participants and spouses; offered opportunities for social interaction for both groups with other families in the same situation; safe environment, provided cognitive stimulation, participants come home in good spirits; the only weakness of the program identified: it was only run one day per week |
| Yes | O'Connell et al_2014_CA (O'Connell et al., 2014) | Hospital | Telehealth online support sessions; remote rural groups | "Members found it helpful to witness and work through processes of grief, loss, and eventually a healthy emergence following death of a spouse" (p388). Socialisation was greatly appreciated; on-line meetings more often than once a month would have been welcome; imparting and sharing of information provided a sense of having made a positive contribution to the group; for some, this developed into advocacy with the local Alzheimer's Society |
| Yes | Parahoo et al_2002_UK (Parahoo et al., 2002) | Community based | Domiciliary Respite Service | Respite in the form of help with bathing and dressing the person with dementia. Carers reported they used the respite time to catch up with household chores and shopping. Well trained staff attempted to fit into the 'normal' way of life of the family. Caregivers perceived domiciliary respite as keeping things 'as normal as possible' for the person with dementia and the family; carers were reassured they could get away for short while, in the knowledge that relative was being looked after by someone reliable and experience; carers felt that caring for their relative with dementia at home contributed to the person's quality of life; "the cohesion and integrity of the household were preserved." |
| Yes | Perkins & Poynton_1990_UK (Perkins & Poynton, 1990) | Hospital/NHS | Group counselling for relatives of hospitalised younger patients with dementia | Counselling increased morale; showed a significant increase in knowledge about YOD; increased the number of physical and social activities that relatives performed with the patient; this helped to 'decrease the helplessness and distress that relatives experienced during visiting periods, because they actually elicited a response from the patient' (p294). |
| Yes | Reed et al_2002_UK (Reed et al., 2002) | Mixed NHS & Community Statutory and non-statutory | NHS team offers advice; Community Psy Nurse; Social Worker; OT | Promptness of response; appreciation of interaction; reassurance; making help available in relation to financial worries and retirement plans (NHS team as intermediary) |
| Yes | Robertson & Evans_2015_AUS (Jacinta Robertson & Evans, 2015) | Community based (work based) respite home + hardware store | Work-based activities in a hardware store | Improved self-esteem, self-worth and confidence; 'participants are more interactive at home after a day at the store and sleep better'; this impacts on the whole family; 'enjoys being able to do something worthwhile'; 'he knows he has helped others and it's helped him, so it's a win-win' (p2335); 'opportunity to mix with other younger people with dementia'; 'he developed a good relationship with everyone, he can walk in to the store and everybody knows him and wants to help him'; 'to meet people outside of their family' (most had experienced a decline of social networks following their diagnosis of dementia' (p2337); 're-established connection with wider community post diagnosis' (p2338); Work as therapy: 'the opportunity to talk about their dementia with other similar people was also a positive aspect of |

(continued)

**Table 2.** (*Continued*)

| Primary study | Author, year, country | Type of intervention | | Primary outcomes: perceived service user satisfaction, health and wellbeing, quality of life were discussed in terms of the following concepts:<br>*Social connectedness, social participation and inclusion; processes of adjustment; normalisation; support for caregivers* |
|---|---|---|---|---|
| | | | | the programme' (p2336); 'we often talk about where we are at with the dementia' (p2336). |
| No | Barker & Johnson_2008_ UK (Barker & Johnson, 2008) | Mixed NHS & Community Statutory and non-statutory | Therapeutic support group; *Ladies who lunch* | Considers gender mainstreaming<br>Provides a forum for women with YOD to meet others; promotes inclusion, social networking where activities can be 'normalised'; emotion focused coping; |
| No | Chaston et al_ 2004_UK (Chaston et al., 2004) | Mixed NHS & Community Statutory and non-statutory | PROP – local support group | PROP (people relying on people) was a small local group formed in partnership with Rotherham Doncaster and South Humber NHS Foundation Trust (RDaSH) Young Onset Dementia Service.<br>Outcomes: activism and advocacy; the group helped to re-design the plan documents<br>Discontinued – replaced with other services |
| No | Craig & Strivens_2016_ AUS (Craig & Strivens, 2016) | On-line | On line facility | Increased interaction with others; accessible, tailored information; broad access to health information; peer, social and emotional support |
| No | Davies-Quarrell_2010_ UK (Davies-Quarrell et al., 2010) | Community based | Club | Outcomes: increased self-esteem; companionship with others; considerable support; gained a measurement of control of their lives; as participants gained insights and understanding of their condition, they shared this with others; individuals regained and 'remodelled' their sense of self; status: funding discontinued after approx. Ten years; as expressed by the authors: 'arguably, it is up to the commissioning agents to view quality of life as the target goal' |
| No | La Fontaine_2004- UK (LaFontaine, 2004) | Mixed NHS and community Statutory and non-statutory | Dedicated community service | Provides pre and post assessment counselling; multi-agency interventions;<br>Outcomes: symptom management; increased social interaction through activities such as art classes, horse riding, gardening scheme… . |
| No | Parks & Ward_2015_UK (Parkes & Ward, 2015) | Mixed NHS and community Statutory and non-statutory | Forget Me Not Social Group in Northamptonshire | Participants perceive group as very positive; strong peer support; encouraging, supportive, informative, socially engaging; interactive; benefits for H&W and quality of life; *An independent evaluation into the effectiveness of the group was completed in the autumn 2016, too late for inclusion in the systematic review, but will be incorporated into the scoping review*) |
| No | Ramluggun et al_2016_UK (Ramluggun & Ogo, 2016) | Mixed NHS and community Statutory and non-statutory | YPDS (young people dementia service) | Younger People Dementia Service in North Cambridgeshire—liaising with statutory and non-statutory organisations and services to provide support and assistance. A large part of the role is to ensure they receive relevant work and health benefits and entitlements, including personal independence payment; psychological support; |
| No | Robertson et al_2013_AUS(J. Robertson et al., 2013) | Community based (work-based) | Work-based activities Hardware store | Demonstration project: family carers have reported a positive impact on self-esteem and life satisfaction as a result of the workplace experience; family carers report increased mental alertness in participants, increased interest in life in general; improved self-esteem; the project offers regular breaks for family carers; increased community awareness of YOD<br>'This evaluation of feasibility has exceeded all expectations and has demonstrated that it is possible to offer meaningful activities for people with mild dementia if an appropriate framework of support is provided.' |
| No | Tonga et al_2016_NOR (Tonga et al., 2016) | Hospital | Behavioural and cognitive rehabilitation therapy | Single case study; participant outcomes: improved mood, decrease in depressive symptoms, increased participation in activities; this also benefited the caregiver |
| No | Wheeler et al_2015_UK (Wheeler et al., 2015) | Mixed NHS and Community Statutory and non-statutory | Welfare advice and advocacy service (Citizens Advice Bureau) | Main support: help with legal and financial concerns; entitlements; debt advice, telephone helpline, support for cares<br>'…If it hadn't been for the advocate my life would have been much more difficult… .the bureaucratic complexity was just seemed too much…the CAB advisor was able to interpret questions on the forms… (p23).'<br>Funding discontinued. |

seen as beneficial for the whole family (Hewitt et al., 2013). Transport arrangements (Kelly & Innes, 2016) made it possible for individuals diagnosed with YOD and their caregivers to meet families in similar situations, which created a sense of companionship for both groups (Kelly & Innes, 2016; Kinney et al., 2011). A sense of normalcy created through socialising with others made participants feel included and accepted as they no longer felt they had to keep explaining to others (Carone et al., 2016).

### *Emotional well-being/quality of life*

Social interaction through worked-based programmes made participants feel useful, conferred a sense of making a positive contribution to others (Hewitt et al., 2013), gave them a sense of purpose (Hewitt et al., 2013) and achievement and enhanced their self-identity (Kelly & Innes, 2016). Participants were reported to be more interactive at home after working in the hardware store and to sleep better, which impacted positively on the whole family (Jacinta Robertson & Evans, 2015; J. Robertson et al., 2013). Caregivers also reported an improved mood in the person with YOD (Kelly & Innes, 2016), a proven increase in weekly well-being scores (Kelly & Innes, 2016), a self-reported increase in self-confidence and motivation (Kinney et al., 2011), and a general increase in the quality of life for both caregivers and people living with YOD.

### Support for family caregivers

Some studies specifically considered the needs of family carers/supporters by offering group counselling for relatives (Perkins & Poynton, 1990), domiciliary respite (Parahoo et al., 2002) and help to claim benefits, (Kelly & Innes, 2016; Reed et al., 2002; Wheeler et al., 2015). This included having a named person to contact and learning how to go about organising 'self-directed support' (Kelly & Innes, 2016). Support that targeted people with YOD had the additional benefit of providing respite for family carers as they were not expected to attend gardening programmes (Hewitt et al., 2013) or work-based programmes (Robertson & Evans, 2015; Robertson et al., 2013), but could join programmes such as social clubs (Davies-Quarrell et al., 2010) or the football club and meet other family carers looking after a person with YOD (Carone et al., 2016).

### Cost indicators

The scoping review and PPI discussions had identified the importance of services being sustained. Cost-related challenges of achieving this were only addressed in two studies, one of which stated that 'specialised programmes can be costly' (Kinney et al., 2011) or, in sharp contrast, that services provided were 'safe and low-cost' (Carone et al., 2016). How costs were assessed was not reported. Four projects ended or were not implemented more widely due to a discontinuation of funding (Davies-Quarrell et al., 2010; Kelly & Innes, 2016; Robertson et al., 2013; Wheeler et al., 2015). One such example was that of a welfare service that helped and supported 178 people with YOD and their families and achieved 'financial gains in excess of £196 210 over 12 months' (Wheeler et al., 2015) (p 20), but funding was withdrawn. The paper did not report how these figures were derived or how gains were made.

### Barriers to accessing services

Reported barriers to accessing services were the hiring of venues, transport and staff-participant ratios (Barker & Johnson, 2008) and the discontinuation of programmes (Davies-Quarrell et al., 2010; Kelly & Innes, 2016; Robertson et al., 2013; Wheeler et al., 2015). Where services continued, it was reported that get-togethers more often than once a month would have been welcome (Kinney et al., 2011; O'Connell et al., 2014; Reed et al., 2002).

## Discussion

Although the wider literature on YOD recognises the need for age appropriate services, only 10 peer-reviewed papers reporting services for this group were eligible for inclusion in this systematic review. The included articles and descriptive accounts span a timeframe of approximately 26 years, but findings relating to age-appropriate services for people diagnosed with YOD and their caregivers have remained largely unchanged. Services are still fragmented, geographically dispersed, variable and often short-term due to project-based commissioning. Some services were run as pilot studies or demonstration projects.

Newly diagnosed individuals perceived services at the point of diagnosis as useful if they were informative, educational and able to signpost to community-based support. Whilst this is consistent with the experience of being diagnosed with dementia regardless of age, the particular experience of living with YOD added extra significance to being able to access community-based services. Community-based services were perceived as effective if they facilitated social interaction, provided a sense of normalcy, belonging and purpose, accommodated changing care needs along the disease trajectory and, importantly, offered continuity over extended periods of time.

Post-diagnostic support concerning employment, legal and financial issues was recognised as critical, but is currently under researched. Similarly, although widely recognised in the literature, approaches to involving individuals living with YOD and their caregivers in the design of age-appropriate services remain underexplored. Only six of the ten included peer-reviewed papers evidenced involvement of people living with YOD in service design. Examples included spousal caregivers and families who participated in the design of support groups (Kinney et al., 2011; O'Connell et al., 2014), or were members of a project steering group or committee (Barker & Johnson, 2008; Chaston et al., 2004; Davies-Quarrell et al., 2010; Kelly & Innes, 2016). This impacted on the design of individualised support (Kinney et al., 2011; O'Connell et al., 2014), which in turn led to increased empowerment and independence (Kelly & Innes, 2016), and to increased self-esteem, well-being and social inclusion (Barker & Johnson, 2008). Inclusion in service design also allowed individuals and families to regain a measure of control over their lives and the lives of those closest to them (Davies-Quarrell et al., 2010). This also increased activism and advocacy (Chaston et al., 2004).

In order to promote the sustainability and continuity of services that are able to accommodate changing care needs along the disease trajectory future research needs to address the known problems of project-based commissioning (which currently is ad-hoc), undertake cost-of-illness studies, chart changing care needs across the disease trajectory to inform service design, produce robust evaluations against identified service outcomes, investigate support concerning employment, legal and financial issues, and establish prevalence and incidence figures.

## Limitations

The systematic review was limited by the small number of peer-reviewed publications available for inclusion. This indicates a dearth of primary studies on this subject and reflects limited service availability and evaluation.

## Conclusion

Although the wider literature reports on the needs of people living with Young Onset Dementia there is little evidence of service development or delivery for this group. The review findings demonstrate a need for research that can inform service design and implementation with a view to establishing a post-diagnostic care pathway. Research into age-appropriate service provision needs to address the dementia policy's overarching aim, which is to enable individuals to continue living at home for as long as possible.

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