Early Onset Dementia: A Systematic Review of the Literature to Inform Qualitative Experiences

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Abstract

There is increasing recognition that EOD (Early Onset Dementia) represents an important social problem affecting economic and social impacts (Campbell et al., 2008; Johannessen et al., 2018). Recent research calls for greater efforts to be made in consulting with the PwD (people with dementia) directly (Allen 2001; Bamford & Bruce 2000). The condition is understood to occur between the ages of 45-65 (Mercy, 2008). This makes EOD a sub-group of dementia with numerous differences when compared to later onset dementia. These include the likelihood of still being in work and having a family to raise. Being responsible for an income and for dependent others is particularly difficult for those affected. Additionally, the social and psychological context for younger people is different (Beattie, 2004). PwEOD (people with Early Onset Dementia) are more likely to be physically fitter than those with later onset dementia which may impact on their physical care needs. The existing expectation within health and social care agencies for PwEOD is in keeping within an older person's framework of care which may well be inappropriate. This may have occurred in the past due to biomedical assumptions regarding the condition (Kitwood, 1997; van Vliet et al., 2017). This also suggests that little attention has been paid to subjective experiences (van Vliet, 2017). The need to elicit the views and subjective experiences of PwEOD is therefore gaining increasing recognition within health and social care research (van Vliet, 2010). Literature has been modestly growing in the subject area to demonstrate how PwEOD have expressed their views and experience of dementia successfully (Page and Keady, 2010; Ohman et al., 2001). However, most of the extant literature is based on family carers (Cabote, 2015; Kobiske and Bekhet, 2018). Whilst carers' views are important they should not be used as a substitute for the views of younger people with dementia (Goldsmith 1996, Whitlatch 2001). Given the limited research available, both the views of PwEOD and their family carers are incorporated into the systematic literature review.

Key words: Early onset dementia (EOD), qualitative, people with early onset dementia (PwEOD), family kin, meta-ethnography, stigma, liminality and chronicity, biographical disruption, losses, coping

Introduction

Personal accounts describing EOD have potential to inform clinical and care provision as well as informing other dementia subsets. Examining first person accounts makes this a valuable exercise. This may be assumed as PwEOD (People with Early Onset Dementia) possess more faculties with which to tell about lived experience from first person accounts.

Method

Study aim: This systematic review paper sought to address the following question:

‘How do PwEOD as a diagnosed sub group of other dementias and their immediate family experience living with EOD?’

Study inclusion: Studies were included and excluded according to the following criteria: a diagnosis of EOD between the ages of 45-65 [1]; research dated between 1998-2018;[2] in English language; qualitative and peer reviewed papers. The key aim of study inclusion was to capture the experience of living with EOD through the available literature in the field. Personal experiences were sought in the literature on PwEOD and their immediate family living with the experience post-diagnosis. The scoping review uncovered the relative lack of studies to date on the experiences of PwEOD, therefore studies were inclusive of spouses, partners, children and adult dependents as people living with the PwEOD. Searches were kept broad and unconstrained by further filters in order to capture a fuller picture of the issues and experiences connected to EOD.

Study exclusion: Studies focusing solely on people presenting with dementia younger than 45; studies with a predominant interest in; dementia caused by HIV, traumatic brain injury, Down’s syndrome, congenital birth conditions likely to include dementia, Huntington’s chorea and alcohol-related dementia were excluded. Systematic literature reviews were excluded.

Scoping: A scoping exercise of the literature took place prior to the systematic literature review which identified EOD as a sub-group of dementia under-represented in the literature. Google Scholar and Abertay’s Library Search including serendipitous searches using prior knowledge of the research field extended the search in preparation for the systematic review.

Search strategy: The author then searched databases which were selected for their social and clinical perspectives through EBSCOhost; Web of Science and Cinahl plus with text, Psychology and Behavioural Sciences Collection, Scopus and Sage. The search terms were: dement*, early onset dementia, young onset dementia, presenile-Alzheimer* and working age dementia. These were searched as single terms using Boolean phrasing; ‘OR’ then once the searches were captured, refined with; ‘AND’ then stored for scrutiny at the next stage.

Selection of papers: The search located five-hundred-and-fifty-two (522) papers. Duplicates were removed (n=22). The remaining studies’ (n=500) abstracts and titles were screened. Twenty-two studies (n=22) were retained and full texts read. This left sixteen studies (n=16) to be included. The reference lists of the twenty-two studies were also examined. Although two were added from references, they were finally excluded for failing to meet the criteria. With reference to the final six studies excluded, these are listed in the appendices (Appendix 1).

Approach to systematic and meta-analysis synthesis of studies

The review was guided by the systematic approach preferred by PRISMA (Reporting Systematic Reviews and Meta-Analysis Studies (Liberati, 2009). Figure 1 illustrates the process of papers being excluded or included for the systematic review based upon the study question. This process sets a standard for the assessment and critique of health focussed studies and interventions assisting the processes for summarising evidence accurately and reliably. However, it is the case that the methods of meta-analysis are not transferable to qualitative health research for a number of pragmatic and epistemological reasons; for example, computer literature searches, statistical data and priorities in quantitative research may fail to capture forms of qualitative research which lack the appeal of more clinical protocols and interventions (Britten et al., 2002). As such, criteria for judging the quality of published research whilst contested in the past have since found established qualitative protocols for comparing studies (Britten et al., 2002). The potential audiences for viewing research through this lens include practitioners across a broad health practice background as well as policy-makers and qualitative researchers (Britten et al., 2002). Therefore, there exists several well recognised methods by which to conduct a systematic review of qualitative literature (Greenwood & Smith, 2016).

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[1] This definition is in keeping with Mercy (2008), excluding two other studies limiting EOD to 45-60. All others searched for the systematic review including the scoping review referred to 45-65 as the most commonly used criteria for EOD.

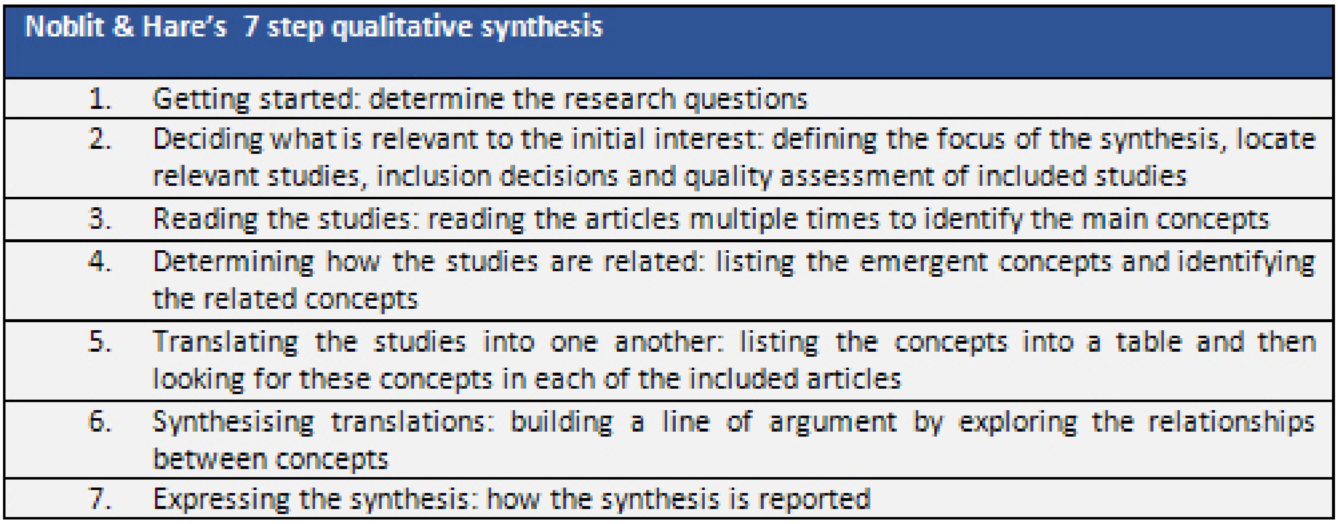
[2] Searches between 1998-2018 captured the advent and widespread prescription of anti-cholinesterase inhibitor treatment and linked with a key driver as laid out in the Scottish Government’s 2009 report making dementia a healthcare priority for development.

The role of meta-ethnography in qualitative research

The impetus for developing methods of qualitative synthesis has arisen from a need to complement quantitative research. This looked to gain a more complete understanding of phenomena, especially in terms of organisational processes and provision of services (Greenhalgh, 1998). Therefore a need existed to bring together isolated studies for comparison (Sandelowski et al., 1997). Meta-ethnography provides a way to compare qualitative studies accommodating induction and interpretation (Greenwood & Smith, 2016). It also can synthesise conceptual innovations such as metaphorical and emotionally relevant phenomena (Strike and Posner, 1983). It has origins in the interpretive paradigm and as such, it possesses an alternative to traditional aggregative methods of synthesis which retain qualities or concepts of the qualitative method of study it aims to synthesise.

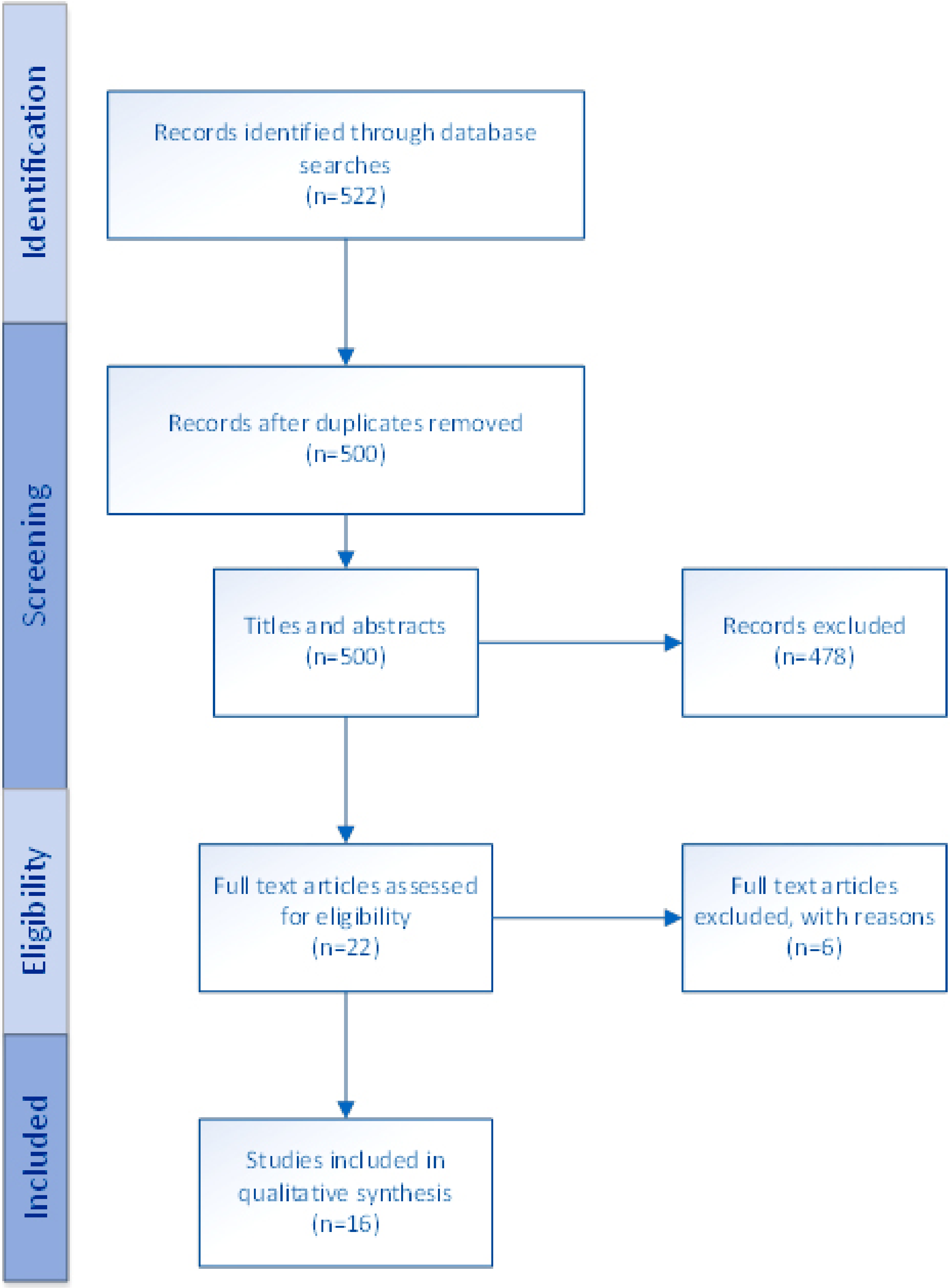
The benefit of applying meta-ethnography to the synthesis of qualitative research and suitability for this study was informed by Noblit and Hare's (1988) seven-step process (Table 1). Narrative literature reviews capture concepts and information in a more eclectic fashion but have in the past been criticised for being singular descriptive accounts based upon the implicit bias of the researcher (Fink, 1998). They have also been condemned for lacking critical assessment. Adopting a more systematic approach to the literature was therefore useful in order to approach a more comprehensive contemporary review of the field. This approach was particularly helpful in investigating EOD as a lesser known sub-group of dementia. Meta-ethnography has proven a sound technique for synthesising qualitative research in health studies (Paterson et al., 1998). It has been successfully employed in publications to date including: lay meanings of medicines (Britten et al., (2002); lay experiences of diabetes and diabetes care (Campbell et al; 2003); what values people seek when they provide unpaid care for an older person (Al-Janabi et al., 2008) and locating how coping experiences appear in chronic fatigue syndrome sufferers (Larun and Malterud, 2007).

Table 1:



Noblit & Hare's 7 step qualitative synthesis	
1.	Getting started: determine the research questions
2.	Deciding what is relevant to the initial interest: defining the focus of the synthesis, locate 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 emergent concepts and identifying the related concepts
5.	Translating the studies into one another: listing the concepts into 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

Figure 1: PRISMA flowchart



Results

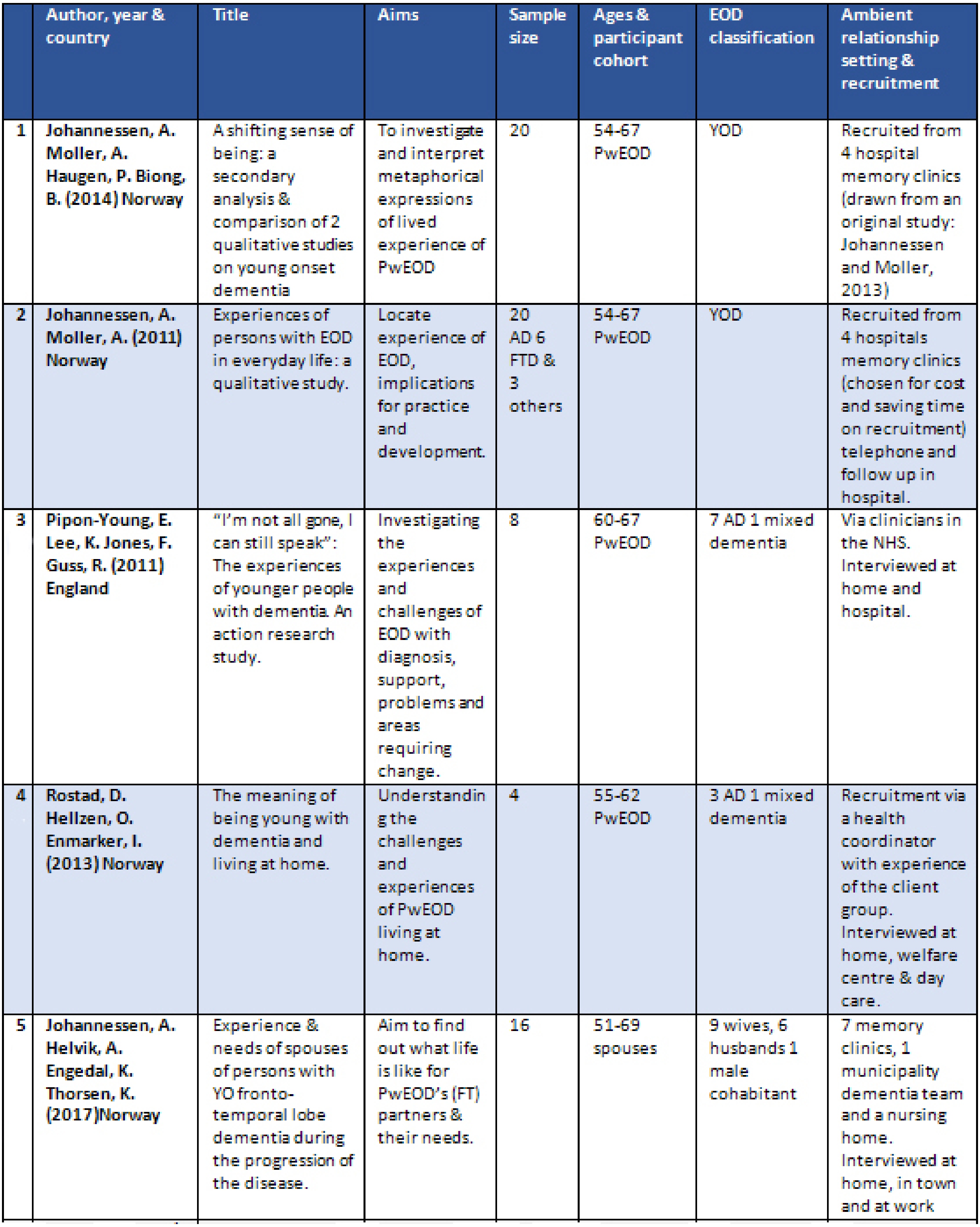
Participant and study design

The inclusion criteria sought studies spanning 1998-2018. However, the studies ranged from 2009 to 2018. The mean date was 2015. All studies were performed in Western countries (Norway-6; America-1; England-; 6; The Netherlands-1; Ireland-1 & Australia-1). Where English was not the dominant language it was widely taught and well spoken (Norway and The Netherlands). The participants were predominantly drawn from health environments or services structured to assist PwD or PwEOD such as statutory or voluntary bodies. There were a total of 229 participants after making amendments for those participants drawn from the same sample groups where multiple study authors were included. Johannessen et al., (2014) and Johannessen and Moller (2011) used the same participants. Johannessen et al., (2016) and Johannessen et al., (2017) also shared participants databases throughout the studies. Data were collected through face to face interview mostly using a semi-structured format. These were situated within the statistics of the studies quoted above; (PwEOD (4); their family members (2); both spouses (2) and dependents (8) whether still regarded as children living at home or adult children living independently elsewhere). These studies drew together the theoretical approaches to the data founded in: grounded theory (5); autobiographical life story narrative (3); phenomenological hermeneutic analysis (2); Thematic Analysis (TA) (2); qualitative semi structured interview (1); conceptual model (1); action research study (1) Interpretative phenomenological analysis (IPA) (1).

Ethnicity was referred to infrequently (n=1) and where ethnic origins were detailed, the sample groups were white/Western. Allen et al., (2009) was the only study to include 25% Asian participants within an English sample. Other studies made no attempt to refer to ethnicity and so a presumption is made that natives of the country of origin satisfied the sample cohorts. This is excepting Sikes and Hall (2017) which reported that the sample participant group was ‘mainly white, British, middle-class, participants’.

Type of dementia was not a focus except for Johannessen et al., (2017) which focused on people with fronto-temporal lobe dementia. Other data reported were related to whether participants (both PwEOD and family) were working, living at home, in studies, in a care home, retired or medically signed off work and living on retirement funds or state benefits. The source for participants overwhelmingly arose from clinical or health focused environments. This particular feature was examined in the discussion of the studies. Having noted the brief characteristics of the studies above, the following tables and sub-sections developed overall themes along with the development of the line of argument.

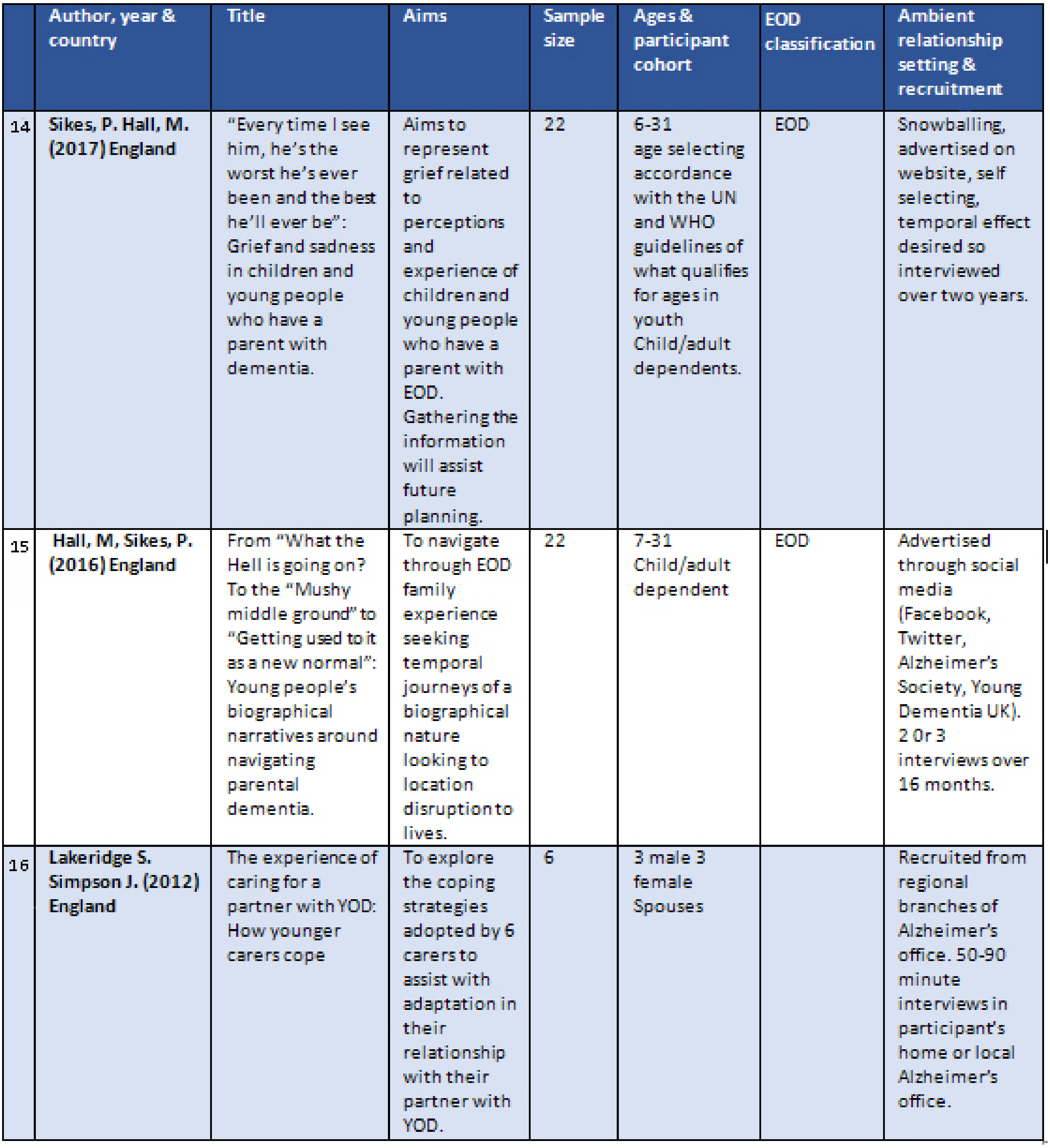
Table 2: Participant and study design



	Author, year & country	Title	Aims	Sample size	Ages & participant cohort	EOD classification	Ambient relationship setting & recruitment
1	**Johannessen, A. Moller, A. Haugen, P. Biong, B. (2014) Norway**	A shifting sense of being: a secondary analysis & comparison of 2 qualitative studies on young onset dementia	To investigate and interpret metaphorical expressions of lived experience of PwEOD	20	54-67 PwEOD	YOD	Recruited from 4 hospital memory clinics (drawn from an original study: Johannessen and Moller, 2013)
2	**Johannessen, A. Moller, A. (2011) Norway**	Experiences of persons with EOD in everyday life: a qualitative study.	Locate experience of EOD, implications for practice and development.	20 AD 6 FTD & 3 others	54-67 PwEOD	YOD	Recruited from 4 hospitals memory clinics (chosen for cost and saving time on recruitment) telephone and follow up in hospital.
3	**Pipon-Young, E. Lee, K. Jones, F. Guss, R. (2011) England**	"I'm not all gone, I can still speak": The experiences of younger people with dementia. An action research study.	Investigating the experiences and challenges of EOD with diagnosis, support, problems and areas requiring change.	8	60-67 PwEOD	7 AD 1 mixed dementia	Via clinicians in the NHS. Interviewed at home and hospital.
4	**Rostad, D. Hellzen, O. Enmarker, I. (2013) Norway**	The meaning of being young with dementia and living at home.	Understanding the challenges and experiences of PwEOD living at home.	4	55-62 PwEOD	3 AD 1 mixed dementia	Recruitment via a health coordinator with experience of the client group. Interviewed at home, welfare centre & day care.
5	**Johannessen, A. Helvik, A. Engedal, K. Thorsen, K. (2017)Norway**	Experience & needs of spouses of persons with YO fronto-temporal lobe dementia during the progression of the disease.	Aim to find out what life is like for PwEOD's (FT) partners & their needs.	16	51-69 spouses	9 wives, 6 husbands 1 male cohabitant	7 memory clinics, 1 municipality dementia team and a nursing home. Interviewed at home, in town and at work

	Author, year & country	Title	Aims	Sample size	Ages & participant cohort	EOD classification	Ambient relationship setting & recruitment
6	**Hoppe, S. (2018) The Netherlands**	A sorrow shared is a sorrow halved: the search for empathetic understanding of family members of a person with EOD.	How Dutch family members find empathy for PwEOD in their family and assess barriers to providing care	7 EOD +41 family members, = 48 partners, PwEOD, 11 group constellations.	55-65Family kin	EOD	Recruited from the Alzheimer's Society and 3 care institutions. Interviews at home at researcher's office in cafes for about 1 to 2 hours.
7	**Flynn, R, Mulcahy, H. (2013) Ireland**	EOD: the impact on family caregivers	Explore caring from family perspective in looking after PwEOD.	7 (over 16)	Under 65 when diagnosed. Family kin	EOD	Regional manager recruited via letter to volunteer participants meeting criteria. At offices of Alzheimer's Society Ireland.
8	**Johannessen, A. Engdal, K. Thorsen, K. (2016) Norway**	Coping efforts & resilience among adult children who grow up with a parent with YOD: a qualitative follow up study.	How adolescent/adult dependents experience their parents' EOD and how they coped with life situations.	14	18-30 Adult dependents	EOD	7 memory clinics, 1 municipality dementia team and a nursing home and from Norwegian National Support Group for Adult Children. Interviews held at convenience of respondent.
9	**Johannessen, A. Engdal, K. Thorsen, K. (2015) Norway**	Adult children of parents with young onset dementia narrate the experience of their youth through metaphors.	To investigate & interpret metaphorical expression of adult dependents of PwEOD. What sense can be made?	14	18-30 Adult dependents	EOD	Heterogeneity attained through diverse geography in Norway/2 memory clinics/1 municipality/nursing home & support group for PwEOD. Interviewed at convenience at home or place of choice

	Author, year & country	Title	Aims	Sample size	Ages & participant cohort	EOD classification	Ambient relationship setting & recruitment
10	**Hutchinson, K. Roberts, C. Kurrle, S. Daly, M. (2016) Australia**	The emotional well-being of young people having a parent with early onset dementia.	To explore the lived experiences of young people with parents with EOD from the perspective of a social model of disability.	12	19-33 + 1 x 10 year old Child/adult dependents	YOD	Advertising via Alzheimer's Australia for volunteers. Interviewed at work, at home or in local library.
11	**Gelman, C. Rhames, K. (2018) America**	In their own words: the experience and needs of children in younger onset Alzheimer's disease & other dementia families.	What are the experiences & needs of children with YOD families & the impact of a diagnosis on children of those families.	(14) 4 families 4 mothers 10 children	10-25 Child/adult dependents	YOD	Flyers were displayed at support organisations $25 payment for information. Interviews lasted an hour at participant's preferred location.
12	**Allen, J. Oyebade, J. Allen, J. (2009) England**	Having a father with young onset dementia. The impact on well being of young people	To investigate and explore the impact on young people's well being in light of them having a PwEOD	12	8-31 (mean 19) Child/adult dependents	British 75% Asian 25% Families of EOD	Recruited from Alzheimer's Society throughout the Midlands. Interviewed from 45-90 minutes
13	**Sikes, P. Hall, M. (2018) England**	"It was then that I though what? This is not my dad". The implications of the 'still the same person' narrative for children & young people who have a parent with dementia	To investigate the 'still the same' narrative through children of PwEOD to assist future plans to support others in the same situation.	19	8-31 age selecting accordance with the UN and WHO guidelines of what qualifies for ages in youth Child/adult dependents	EOD	2 interviews based upon self selecting participants from web advertisement.

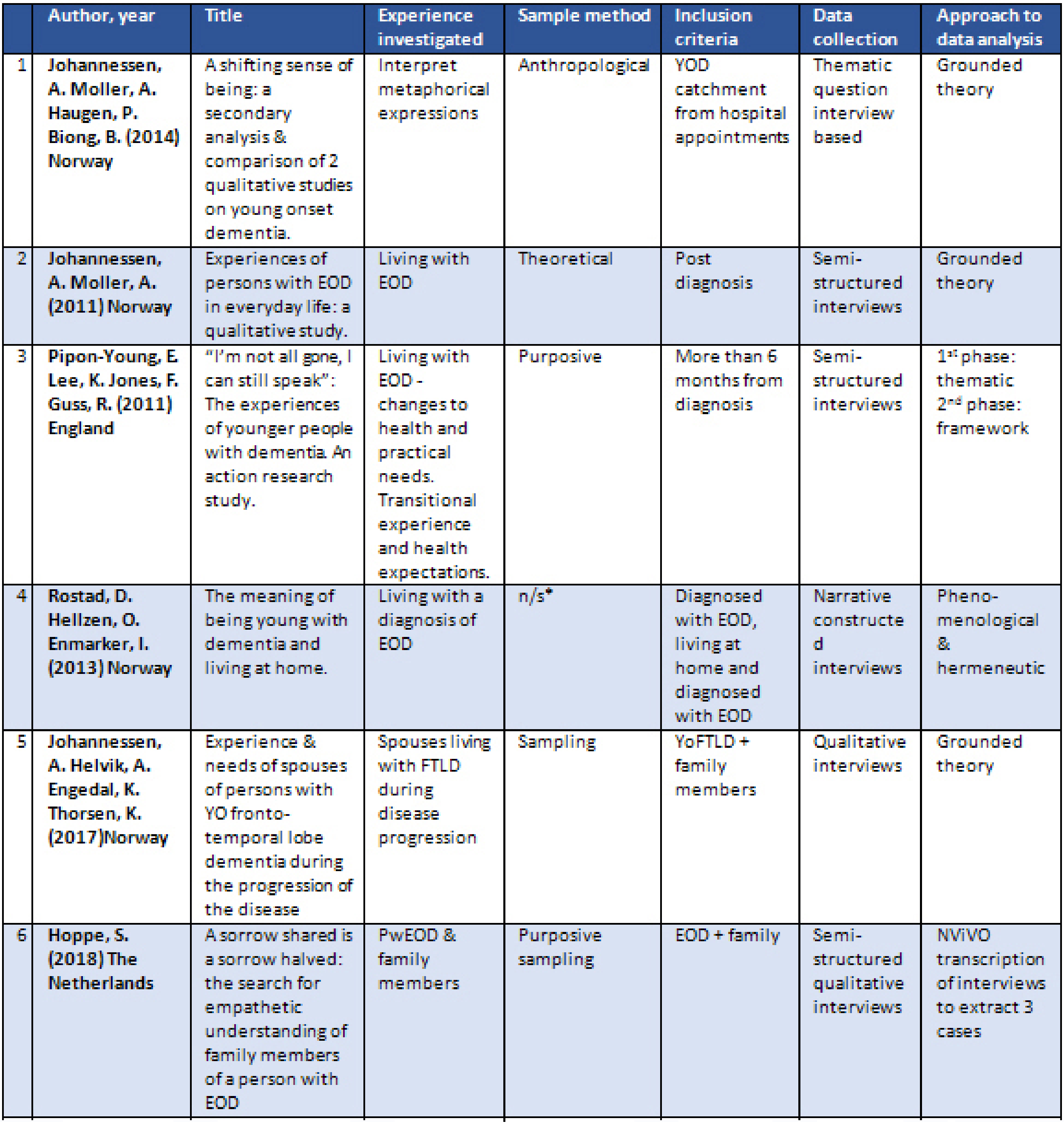


	Author, year & country	Title	Aims	Sample size	Ages & participant cohort	EOD classification	Ambient relationship setting & recruitment
14	**Sikes, P. Hall, M. (2017) England**	"Every time I see him, he's the worst he's ever been and the best he'll ever be": Grief and sadness in children and young people who have a parent with dementia.	Aims to represent grief related to perceptions and experience of children and young people who have a parent with EOD. Gathering the information will assist future planning.	22	6-31 age selecting accordance with the UN and WHO guidelines of what qualifies for ages in youth Child/adult dependents.	EOD	Snowballing, advertised on website, self selecting, temporal effect desired so interviewed over two years.
15	**Hall, M, Sikes, P. (2016) England**	From "What the Hell is going on? To the "Mushy middle ground" to "Getting used to it as a new normal": Young people's biographical narratives around navigating parental dementia.	To navigate through EOD family experience seeking temporal journeys of a biographical nature looking to location disruption to lives.	22	7-31 Child/adult dependent	EOD	Advertised through social media (Facebook, Twitter, Alzheimer's Society, Young Dementia UK). 2 Or 3 interviews over 16 months.
16	**Lakeridge S. Simpson J. (2012) England**	The experience of caring for a partner with YOD: How younger carers cope	To explore the coping strategies adopted by 6 carers to assist with adaptation in their relationship with their partner with YOD.	6	3 male 3 female Spouses		Recruited from regional branches of Alzheimer's office. 50-90 minute interviews in participant's home or local Alzheimer's office.

Drawing a line of argument from the seven step process

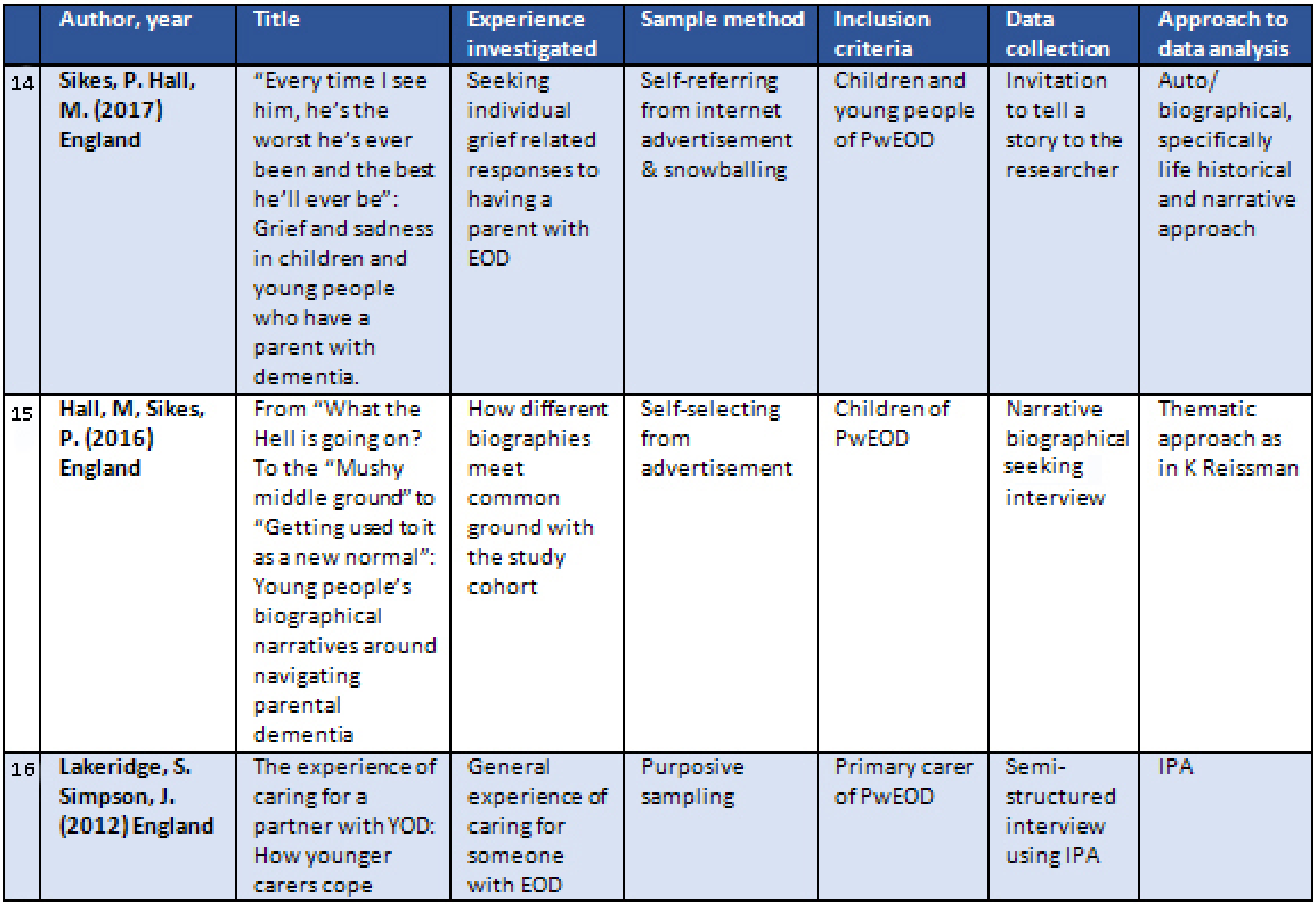
Noblet and Hare (1988) refer to a meta-ethnographic line of argument which emerges to articulate a larger phenomenon drawn from the data. This was achieved by following the steps. After selecting an aim and study question (steps one and two), the studies were read to fulfil step three. This was followed by populating the tables with typical broad characteristics (Table 2). Investigation of experiences were then described (Table 3). Following this, steps four, five and six produced more concepts (Table 4) and themes (Table 5) were populated taking care to ensure the data remained true to the original studies. Step seven provided for a discussion through the line of argument of what fresh data was discovered.

Table 3: Investigation of experiences



	Author, year	Title	Experience investigated	Sample method	Inclusion criteria	Data collection	Approach to data analysis
1	**Johannessen, A. Moller, A. Haugen, P. Biong, B. (2014) Norway**	A shifting sense of being: a secondary analysis & comparison of 2 qualitative studies on young onset dementia.	Interpret metaphorical expressions	Anthropological	YOD catchment from hospital appointments	Thematic question interview based	Grounded theory
2	**Johannessen, A. Moller, A. (2011) Norway**	Experiences of persons with EOD in everyday life: a qualitative study.	Living with EOD	Theoretical	Post diagnosis	Semi-structured interviews	Grounded theory
3	**Pipon-Young, E. Lee, K. Jones, F. Guss, R. (2011) England**	"I'm not all gone, I can still speak": The experiences of younger people with dementia. An action research study.	Living with EOD - changes to health and practical needs. Transitional experience and health expectations.	Purposive	More than 6 months from diagnosis	Semi-structured interviews	1st phase: thematic 2nd phase: framework
4	**Rostad, D. Hellzen, O. Enmarker, I. (2013) Norway**	The meaning of being young with dementia and living at home.	Living with a diagnosis of EOD	n/s*	Diagnosed with EOD, living at home and diagnosed with EOD	Narrative constructed interviews	Phenomenological & hermeneutic
5	**Johannessen, A. Helvik, A. Engedal, K. Thorsen, K. (2017)Norway**	Experience & needs of spouses of persons with YO fronto-temporal lobe dementia during the progression of the disease	Spouses living with FTLD during disease progression	Sampling	YoFTLD + family members	Qualitative interviews	Grounded theory
6	**Hoppe, S. (2018) The Netherlands**	A sorrow shared is a sorrow halved: the search for empathetic understanding of family members of a person with EOD	PwEOD & family members	Purposive sampling	EOD + family	Semi-structured qualitative interviews	NViVO transcription of interviews to extract 3 cases

	Author, year	Title	Experience investigated	Sample method	Inclusion criteria	Data collection	Approach to data analysis
7	**Flynn, R, Mulcahy, H. (2013) Ireland**	EOD: the impact on family caregivers	Physical, social, emotional and financial experience sought	Purposive	Open invitation to those within criteria	Face to face semi-structured in-depth	Chou (2000) conceptual analysis tool
8	**Johannessen, A. Engdal, K. Thorsen, K. (2016) Norway**	Coping efforts & resilience among adult children who grow up with a parent with YOD: a qualitative follow up study	Transition and resilience	Purposive sampling	EOD diagnosis made 6 months include adult children	Interview face to face	Grounded theory
9	**Johannessen, A. Engdal, K. Thorsen, K. (2015) Norway**	Adult children of parents with young onset dementia narrate the experience of their youth through metaphors	Emotional trauma and distress	Purposive sampling	Family of EOD	Individual qualitative interviews Seeking metaphors from interview	Phenomenological, hermeneutic. T.A.
10	**Hutchison, K. Roberts, C. Kurrle, S. Daly, M. (2016) Australia**	The emotional well-being of young people having a parent with early onset dementia	Purposive sampling	Children of PwEOD	Semi-structured interviews		Social model of disability
11	**Gelman, C. Rhames, K. (2018) America**	In their own words: the experience and needs of children in younger onset Alzheimer's disease & other dementia families	Experience of social impact	Purposive sampling	Children of PwEOD	Face to face interviews	Thematic narrative analysis
12	**Allen, J. Oyebade, J. Allen, J. (2009) England**	Having a father with young onset dementia. The impact on well-being of young people	Emotional impact and psychological experience	Purposive sampling	Children of PwEOD	Face to face interviews	Grounded theory
13	**Sikes, P. Hall, M. (2018) England**	"It was then that I thought what? This is not my dad". The implications of the 'still the same person' narrative for children & young people who have a parent with dementia	Autobiographical meanings	Purposive and self-referring	Children and young people of PwEOD	Collection of in depth personal stories and biographies	Autobiographical/life histories interviews &T.A



	Author, year	Title	Experience investigated	Sample method	Inclusion criteria	Data collection	Approach to data analysis
14	**Sikes, P. Hall, M. (2017) England**	"Every time I see him, he's the worst he's ever been and the best he'll ever be": Grief and sadness in children and young people who have a parent with dementia.	Seeking individual grief related responses to having a parent with EOD	Self-referring from internet advertisement & snowballing	Children and young people of PwEOD	Invitation to tell a story to the researcher	Auto/biographical, specifically life historical and narrative approach
15	**Hall, M, Sikes, P. (2016) England**	From "What the Hell is going on? To the "Mushy middle ground" to "Getting used to it as a new normal": Young people's biographical narratives around navigating parental dementia	How different biographies meet common ground with the study cohort	Self-selecting from advertisement	Children of PwEOD	Narrative biographical seeking interview	Thematic approach as in K Reissman
16	**Lakeridge, S. Simpson, J. (2012) England**	The experience of caring for a partner with YOD: How younger carers cope	General experience of caring for someone with EOD	Purposive sampling	Primary carer of PwEOD	Semi-structured interview using IPA	IPA

Overall themes

By the time Table five was completed at stages five and six in accordance with Noblit and Hare's seven step process, new data emerged to realise conceptual themes crystallised into themes which formed the expression of the new information. The expression of the synthesis followed the tables discussed theme by theme.

Table 4: Conceptual themes

	Author/Title/ Year	Concepts							
		Losses	Chronicity and lost time	Biographical disruption	Stigma & judgment	Diagnosis	Isolation & social identity	Emotions	Coping
1	Johannessen, A. Moller, A. Haugen, P. Biong, B. (2014) Norway	Loneliness	Life changes will appear		Masking & hiding	Difficult and lengthy to achieve	Sliding away & falling from life once taken for granted	Anger Fear Reticence Living in limbo	Coping Claiming dignity
2	Johannessen, A. Moller, A. (2011) Norway	Lack of normality. Confusion in every day contexts. Dignity lost	Dislocation & lack of certainty. Constraint s on time and planning		Dismissed by others. Status lost	Diagnostic struggles & shock	Feeling outside others and time Relationship changes	Diminished & rejected. Irritation	Stronger together, going on
3	Pipon-Young, E. Lee, K. Jones, F. Guss, R. (2011) England	Losing ideas of how to relate		Calm for now but afraid of the future	Saving face with others, shielding truth	Diagnosis feels in the wrong time & stage	'Out of step'	Resilience	Keeping 'in the swim' & retaining new social identities
4	Rostad, D. Hellzen, O. Enmarker, I. (2013) Norway	Loss of power, Loss of humanity, Loss of identity,	Wrong time for diagnosis		Reduced sense of belonging in the eyes of others,		Trapped by circumstances		Preservation of hope and resilience. Desiring to live well and managing.
5	Johannessen, A. Helvik, A. Engedal, K. Thorsen, K. (2017) Norway			Moving apart	Stigma	Distanced emotional reactions		Coping	Calm & resilience
6	Hoppe, S. (2018) The Netherlands	From certainty of health to the uncertainty of disease		Lifestyle burnout when too young	Barriers to acceptance of EOD condition	In between ideas of health and illness	Living in a state of confusion	Trepidation Guilt	Communing with friends increasing strength
7	Flynn, R, Mulcahy, H. (2013) Ireland	No means to support and care	Waiting times for news and change			Different diagnoses given. Big delays. Diagnosis denial	Doubts over how to go forward differently	Fear Loneliness	

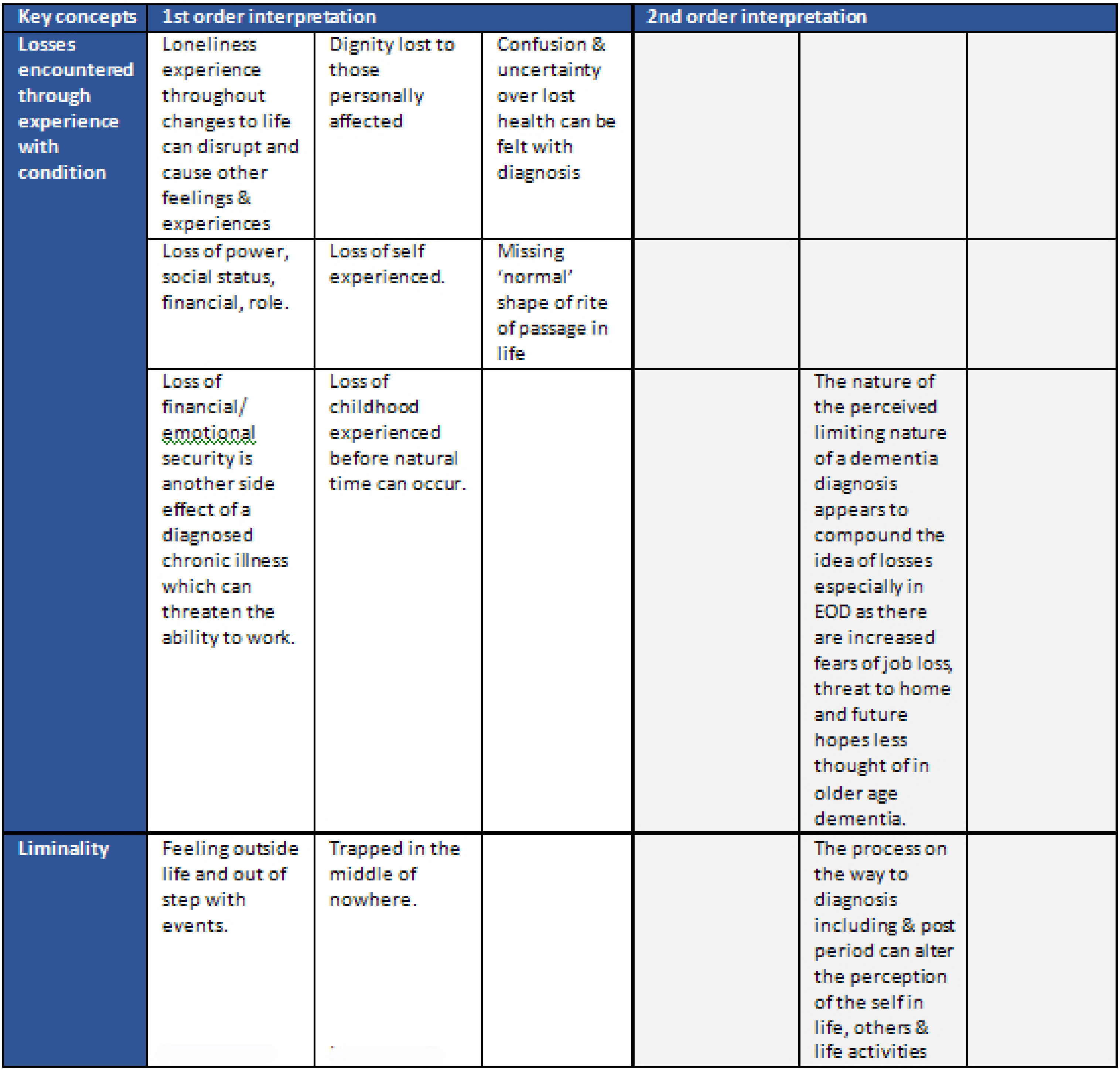
	Author/Title/ Year	Concepts							
		Losses	Chronicity and lost time	Biographical disruption	Stigma & judgment	Diagnosis	Isolation & social identity	Emotions	Coping
8	**Johannessen, A. Engdal, K. Thorsen, K. (2016) Norway**	Lost childhood of child dependent						Feeling freedom. Feeling like themselves. Guilt on top of freedom	Relief. Making a stand
9	**Johannessen, A. Engdal, K. Thorsen, K. (2015) Norway**	The loss of a parent sliding away. The shock of losing the parent inside & no one left behind.		Stilted development	Covering up the diagnosis		Waiting to begin life	Confused	Emotional chaos. Escaping from doubt to cope
10	**Hutchison, K. Roberts, C. Kurrle, S. Daly, M. (2016) Australia**	Loss compounded with grief			Being judged			Fear of being trapped, hopelessness, frustration, 24/7 nature of caring is overwhelming. Normality of family weighted against normality of self	24/7 nature of caring is overwhelming. Normality of family weighted against normality of self.
11	**Gelman, C. Rhames, K. (2018) America**	Abruption – interruption of child/adolescent development. Losing the natural order of parents & children.	Waiting for others to 'get it'-disbelief of diagnosis	Living with lies, terror, dread & 'freak-outs				Fear, Embarrassment exasperation	Adaption, acceptance, growth & change
12	**Allen, J. Oyebade, J. Allen, J. (2009) England**	Loss of status; working parent, credible parent, comforted child.	Waiting to become a child but needing to be a parent instead. Parentification.	Loss of parent loss of childhood & youth	Disjunction of childhood experience.	Delays in diagnosis.		Fear, fright, unease Despair.	Engaged, Knowledgeable. Better understanding of way forward.

	Author/Title/ Year	Concepts							
		Losses	Chronicity and lost time	Biographical disruption	Stigma & judgment	Diagnosis	Isolation & social identity	Emotions	Coping
13	**Sikes, P. Hall, M. (2018) England**	The loss and receding of the self (parent) Leaving the idea of the parent, loss of role, 'mum is an utter bitch'	The slipping away of parent & child	Disconnect of parenting	The 'Hollywooding' of the demented-the romanticising of dementia		No warning of losses & happenings	Crumbling spirit,	
14	**Sikes, P. Hall, M. (2017) England**	Missing important stages and rites of passage in life.	Life on hold, Worrying about death & funerals 'out of time'				Liminal grief. Uncertainty of future.	Envy of other families in health deemed normal. Post-carer symptoms of depression.	Relief after death or after caring
15	**Hall, M, Sikes, P. (2016) England**	Shock realisation of death after illness for children of PwEOD	Irrational behaviour only takes on meaning after time. Time lag of diagnosis.	Disruption to the natural expectation of life with dementia-children don't understand dementia is terminal.		Epiphanic moment of realisation. Time lag in diagnosis	Stuck in the middle ground of not knowing what may happen next	Shock sadness of unexpected death. Sadness of loss of parent in life & death.	
16	**Lakeridge, S. Simpson, J. (2012) England**	Loss of social status. Uncertainty over position in future life beyond caring. Threat to self over EOD being diagnosed.			Rejected by social life and society Left out of the loop as carers of EOD which is not understood by other health care roles/services	Lack of awareness of EOD as a condition. Futility of giving a negative diagnosis.	Denial of what is happening in everyday life.	Anxiety, Self denial, In denial, Shock, Disappointment Distress, Anger, Bitterness at treatment as carers, Feeling bullied by service providers into accepting unwanted services.	Finding help from others in same position.

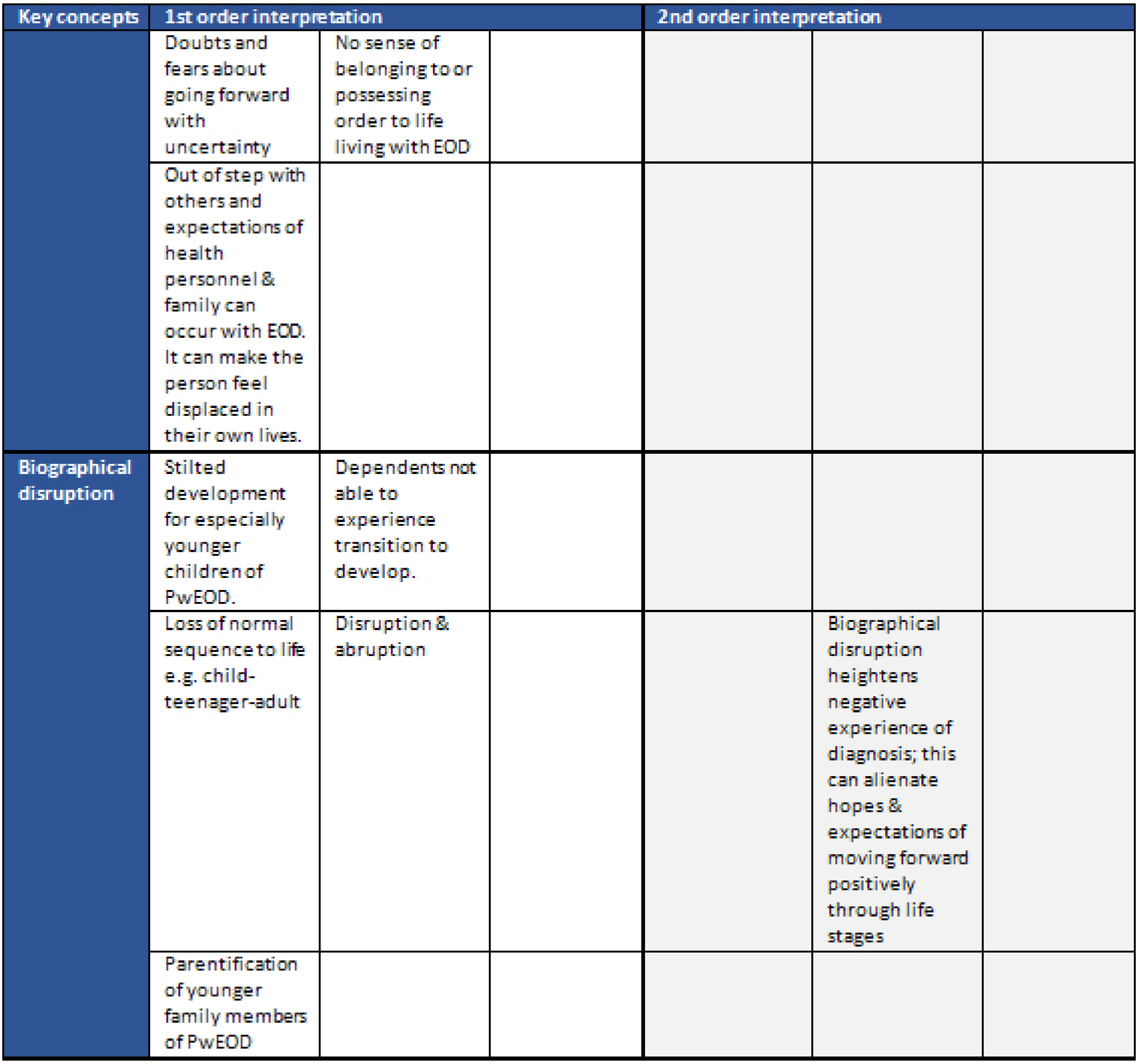
Conceptual themes and Schultz's first and second order constructs

The table below concluded the development of the line of argument. Noblit and Hare adopted Schutz's notion of first and second order constructs assisting the progression of themes. Schutz utilised the term first-order construct in referring to the everyday constructs and understandings of ordinary lay people. The second-order construct referred to those constructs familiar to social science researchers. The table below (Table 5) reveals how the themes took their place within the constructs drawn in accordance with Schutz's terms. The final themes that arose were: i) biographical disruption ii) diagnosis, iii) losing life, friends and competences, iv) liminality and chronicity, v) stigma, and vi) coping with cautious optimism. The table below finalised the creation of new concepts which are followed by discussion of the themes.

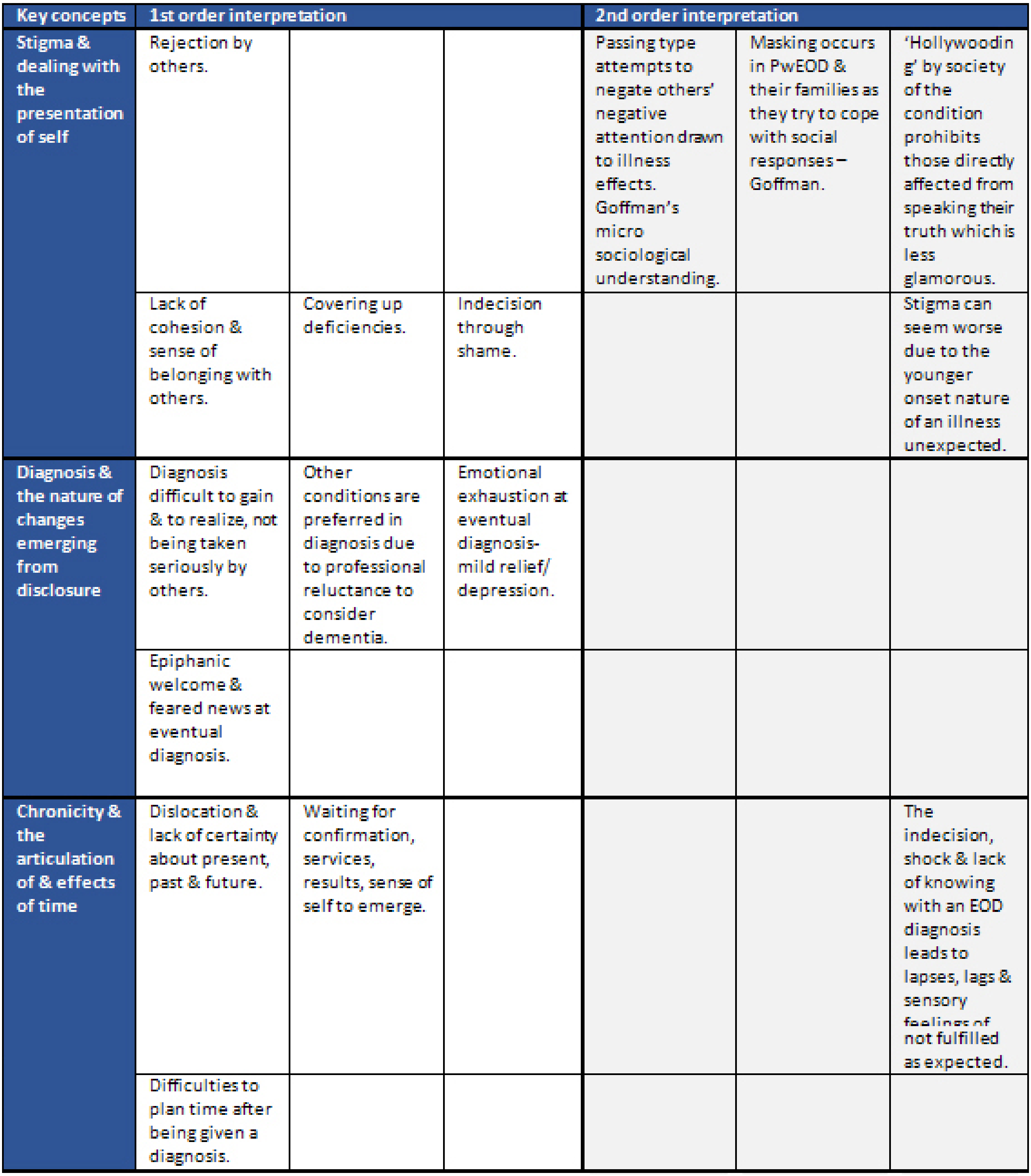
Table 5: Second and third order interpretations



Key concepts	1st order interpretation			2nd order interpretation		
Losses encountered through experience with condition	Loneliness experience throughout changes to life can disrupt and cause other feelings & experiences	Dignity lost to those personally affected	Confusion & uncertainty over lost health can be felt with diagnosis			
	Loss of power, social status, financial, role.	Loss of self experienced.	Missing 'normal' shape of rite of passage in life			
	Loss of financial/ emotional security is another side effect of a diagnosed chronic illness which can threaten the ability to work.	Loss of childhood experienced before natural time can occur.			The nature of the perceived limiting nature of a dementia diagnosis appears to compound the idea of losses especially in EOD as there are increased fears of job loss, threat to home and future hopes less thought of in older age dementia.	
Liminality	Feeling outside life and out of step with events.	Trapped in the middle of nowhere.			The process on the way to diagnosis including & post period can alter the perception of the self in life, others & life activities	



Key concepts	1st order interpretation			2nd order interpretation		
	Doubts and fears about going forward with uncertainty	No sense of belonging to or possessing order to life living with EOD				
	Out of step with others and expectations of health personnel & family can occur with EOD. It can make the person feel displaced in their own lives.					
Biographical disruption	Stilted development for especially younger children of PwEOD.	Dependents not able to experience transition to develop.				
	Loss of normal sequence to life e.g. child-teenager-adult	Disruption & abruption			Biographical disruption heightens negative experience of diagnosis; this can alienate hopes & expectations of moving forward positively through life stages	
	Parentification of younger family members of PwEOD					



Key concepts	1st order interpretation			2nd order interpretation		
Stigma & dealing with the presentation of self	Rejection by others.			Passing type attempts to negate others' negative attention drawn to illness effects. Goffman's micro sociological understanding.	Masking occurs in PwEOD & their families as they try to cope with social responses – Goffman.	'Hollywooding' by society of the condition prohibits those directly affected from speaking their truth which is less glamorous.
	Lack of cohesion & sense of belonging with others.	Covering up deficiencies.	Indecision through shame.			Stigma can seem worse due to the younger onset nature of an illness unexpected.
Diagnosis & the nature of changes emerging from disclosure	Diagnosis difficult to gain & to realize, not being taken seriously by others.	Other conditions are preferred in diagnosis due to professional reluctance to consider dementia.	Emotional exhaustion at eventual diagnosis- mild relief/ depression.			
	Epiphanic welcome & feared news at eventual diagnosis.					
Chronicity & the articulation of & effects of time	Dislocation & lack of certainty about present, past & future.	Waiting for confirmation, services, results, sense of self to emerge.				The indecision, shock & lack of knowing with an EOD diagnosis leads to lapses, lags & sensory feelings of not fulfilled as expected.
	Difficulties to plan time after being given a diagnosis.					

Key concepts	1st order interpretation			2nd order interpretation		
Coping & emerging from suffering positively	There can be 'room to live with it' helps to see life more positively after.	Collaboration from others helps to be positive.				
	With understanding plans can be made to bond with post diagnosis life.	Lessons to teach others emerge; can occur with acceptance			Once a stage of acceptance has been met, beyond this is a reintegration with a normal life once more but as changed, informed with an optimistic outlook of self & others.	
	Desiring to manage & flourish with new knowledge					

Line of argument

PwEOD – Biographical disruption

A prevailing theme within eleven of the synthesised sixteen papers reflected disruption of future life plans for PwEOD and their families (Pipon-Young et al., 2011; Hall and Sikes, 2018; Gelman and Rhames, 2016; Johannessen et al, 2016; Johannessen et al, 2017;Johannessen et al, 2015; Johannessen and Moller, 2011; Rostad and Hellzen, 2013;Allen et al, 2009; Sikes and Hall, 2017; Sikes and Hall, 2018).

A synthesis of these studies showed how participants' awareness of feeling too young to face a dementia diagnosis prevailed (Pippon-Young et al., 2011). For dependents, living with a parent with EOD could alter the experience of ideas of childhood. Hall and Sikes (2017) suggested how life could be on hold. From this position, Hall and Sikes (2018) noted how there was no parental template to draw upon for support. Sikes and Hall (2018) expanded upon their own authorship of other papers and described the 'Hollywooding' of the experience of dementia. This referred to the way film and media positioned dementia to a lay public. However, participants reported that this perspective did not equate to their lived reality. Gelman and Rhames (2018) reflected upon the chaos and uncertainty of living with EOD and how life lacked any sense of continuity. Johannessen et al., (2016) and Johannessen et al, (2017) noted that the overall burden of EOD on the family was a major biographical consideration. Shock was recorded prior to and beyond diagnosis with the reshaping of life plans (Johannessen and Moller, 2011; Rostad and Hellzen, 2013). Sikes and Hall (2017) suggested that life could be experienced as hectic prior to diagnosis. Following this, post diagnosis held no changes to a life continuing in crisis or fear of crisis. These feelings extended to include both children and teenagers who were immersed in the difficulties in trying to achieve, enjoy and establish lifetime goals that others took for granted (Hutchinson et al., 2016; Hoppe, 2018; Hall and Sikes, 2016). Extending from this, Allen et al., (2009) and Johannessen et al., (2015) described levels of parentification which became normal ways to react to living within a family. Gelman and Rhames, (2016), spoke of developmental hitches occurring to both emotional and psychological development. The ways that this could be reported lay with noticing that a sibling had stopped smiling and laughing. Hall and Sikes (2016) reported a lack of awareness in children that dementia was terminal.

Some studies discussed how being in the workplace represented feelings of exposure with others noticing changes (Johannessen and Moller, 2011). This could lead to queries by colleagues over observable drops and changes in performance (Johannessen and Moller, 2011). Although early retirement caused by EOD could be a life changing departure it could come as a relief when struggles became overwhelming (Johannessen and Moller, 2011). Johannessen et al., (2016) reported how the younger members of families with a PwEOD struggled to define themselves. This had consequences for their own identity. Johannessen et al., (2015) suggested that the integrity and identity of the family as a whole was diminished in certain participant groups. The parental role was eroded through illness as was the child/teenager's role interrupted. Therefore, this could feel like, as discussed, how life changing situations at home felt like a continuous life threat (Hall and Sikes, 2018).

In conclusion it appeared that although biological disruption is a common theme, it can be experienced differently. Participants could experience it with a sense of not knowing what the future held thus making plans and expectations hard to follow or to enjoy. This could arise from not understanding the trajectory of the illness and at the same time understanding that changes would be unpredictable making life plans difficult to gauge and direct.

Diagnosis

A dominant theme appeared throughout fourteen of the studies indicating that the periods before and after diagnosis could be pivotal (Flynn and Mulcahy, 2013; Johannessen and Moller, 2011; Allen et al, 2009; Hall and Sikes 2018; Johannessen et al., 2017; Lockeridge and Simpson, 2012; Sikes and Hall, 2017; Sikes and Hall, 2018; Johannessen et al., 2016; Johannessen et al., 2015; Pipon-Young, 2011; Rostad and Hellzen 2013; Gelman and Rhames, 2018;and Hutchinson et al, 2016).
A summary and synthesis of similarities reflected that the period running up to diagnosis could be triggered by major crisis events attracting the intervention of authorities as well as minor events which were not exposed to public scrutiny (Lockeridge and Simpson, 2012). It could prove an exhausting journey (Sikes and Hall, 2018). Diagnosis could provide the catalyst for lifestyle changes (Gelman and Rhames, (2018). However, Lockeridge and Simpson (2012) reported that the road towards the clarity and certainty provided by diagnosis was not always welcomed. When dementia was feared or suspected it could take some time to convince a family member to address the matter through a clinical diagnosis (Lockeridge and Simpson, 2012). It was also acknowledged (Allen et al., 2009) that family carers felt despondent over the quality of diagnosis; the competence of the doctor being able to diagnose dementia and ability to offer any supportive strategy thereafter. Johannessen et al., (2017) reflected how each member of the triad reacted to diagnosis; the clinician, the PwEOD and family. Some evidence uncovered how participants' credibility was affected (Johannessen et al., 2017; Johannessen and Moller, 2011) as participants felt they were not believed by a clinical audience. However, Johannessen et al., (2017) and Sikes and Hall (2017) reported relief at diagnosis as it gave a rational account of the recent past. But contrasted to this was experiences of loneliness could set in with no one to talk to about the diagnosis (Johannessen et al., 2015). In Johannessen and Moller (2013), diagnosis meant that an explanation was provided for odd behaviour or withdrawal from previous interests. However, this was tempered by the fact that EOD still represented a terminal illness (Johannessen and Moller, 2011).

Experiences and feelings of blame did permeate the diagnostic period. Such conflicts meant that there was a variety in responses from those affected personally by EOD (Hall and Sikes, 2018). Because doctors were unlikely to suspect EOD often subjective criticism and blame directed towards the family members occurred (Johannessen et al., (2017). Some spouses were not often welcomed in the consulting room and their input was not valued (Johannessen et al., 2017). Often diagnosis was finally achieved after investigation of a string of plausible conditions had proven fruitless (Allen, 2009).

The certainty of diagnosis could allow for exploration of potential future pathways. Attempts could be made to incorporate new approaches to health like new treatment therapies and healthier lifestyles (Hall and Sikes, 2018). While some participants felt relief in reaching a diagnosis, suicidal feelings could manifest immediately after diagnosis (Hutchinson et al, 2016). However, diagnosis could also provide the catalyst for making life changes such as giving up career plans for both the PwEOD and their family (Gelman and Rhames., 2018). Acceptance helped make the best of the situation (Rostad et al., (2013). Contrasting with acceptance was the way that the diagnosis period was experienced outside the clinician's consulting room with one participant being outpaced by his wife's determination to locate a diagnosis. This meant that the participant lagged in their knowledge and acceptance of the diagnosis (Rostad et al., 2013) Lockeridge and Simpson (2012) illustrated how spouses felt conflicted whilst pursuing a diagnosis. They reported that they felt a level of deceit in reporting symptoms at home.

To conclude, when facing a diagnosis of EOD, there were obstacles. These were present prior to diagnosis with lack of realisation of EOD and a desire to not acknowledge a life changing condition. Medical authorities often failed to identify the condition and as a coping mechanism blamed the family who were consulting. Families coped in different ways where optimism could be a feature but predominantly negative feelings were voiced. Whether positive or negative experiences prevailed, families found their lives following very different paths after the delivery of a diagnosis.

Losing life, friends and competencies

Eight studies reflected themes connected to losses; (Rostad et al., 2013; Gelman and Rhames,2018; Flynn and Mucahy, 2013; Allen et al.,2009; Sikes and Hall, 2018; Hutchinson et al., 2016; Sikes and Hall, 2017 and Johannessen et al., 2014).

The synthesis located the diverse effects which could be considered as losses. Rostad et al., (2013) represented ideas expressed over losses to humanity. This was described as loss of self-identity, esteem, determination and respect which extended to becoming dependent upon others. This led to increased inactivity and passivity. Gelman and Rhames (2018) stated that a lack of finite resources to provide for and recognise needs in the family led to permanent feelings of loss. Sikes and Hall (2018) reported loss of the PwEOD and bereavement and grieving occurring before the death occurred and discussed how life was lived alongside these feelings. In Allen et al., (2009) there was a reported loss of a father role in the family and the loss of expectation of being parented. In Hutchinson et al., (2016) a loss was recorded in opportunities due to mandatory care responsibilities being required to be covered in the household. Sikes and Hall (2017) reported a downward spiral effect of losing a normal perspective. In the Johannessen et al., (2014) study, losses were discussed as losing social role, health, quality of life, ability to work and cognitive capacity.

Flynn and Mulcahy, (2013) positioned their study to understand losses as being service driven and they often fell short of sufficiency for the PwEOD and their family carers. Restrictions were financial, social, emotional and physically felt by the participant group; the family carers (Sikes and Hall, 2017).

To conclude, losses could be understood personally as well as socially.

Experiences around how 'normal' life was now dislocated, particularly after the certainty of a diagnosis, were notable and diverse. These experiences could venture into aspects of life which were connected to feelings and perceptions of identity, autonomy and selfhood. This could reflect how it was difficult to isolate claims into disparate concepts. Instead they tended to rely upon linking and merging into each other. Conceptual losses could thus be understood within a number of interlinking experiences.

Liminality and chronicity

Themes drawn from eleven of the sixteen studies revealed experiences around liminality. This effect can arise when people feel 'out of this world', 'betwixt and between' and in situations divorced from their perception of normal life. Notions of chronicity prevailed with feeling that time did not follow a normal pattern: (Rostad et al., 2013;Johannessen and Moller,2011;Johannessen et al., 2014; Lockeridge and Simpson, 2012; Gelman and Rhames,2018; Hutchison et al., 2016; Johannessen et al., 2015; Sikes and Hall, 2017; Sikes and Hall. 2018;Flynn and Mulcahy,2013 and Johannessen et al., 2017).

The synthesised results led to the following understanding: Johannessen and Moller (2011) noted feelings of confusion and not knowing what was happening to them in the interim period prior to diagnosis. Gelman and Rhames (2018) reported experiences of chaotic feelings of 'otherness' and 'outsiderhood' in relation to change as well as leading to feelings of failure to cope (Rostad et al., 2013; Johannessen et al., 2014; Johannessen and Moller, 2011). These experiences read to reflect a sensory experience which was experienced as a fracturing away from others and perceived normality (Allen et al., 2009). Spouses could feel rejected and confused over how to cope (Johannessen et al., 2017). Lockeridge and Simpson (2012) outlined being lost with feelings of rejection and how carers could feel bullied into accepting services they did not wish or feel necessary. Allen et al (2009) reported in accordance with the Gelman paper feelings of disconnection whereby other family members could appear co-dependent with the PwEOD. This could lead to epiphanic, revelationary and euphoric traits present whereby certain family members could advocate for the day when the PwEOD recovered or different causes could be found for symptoms. Johannessen et al., 2017; Sikes and Hall (2017) reported how family carers observed feelings of rejection and confusion about everyday life. Johannessen et al., (2016) articulated lifetime rites of passage and aggregated experiences that were under threat such as marriage, graduations and childbirth. Therefore, there was an articulated experience of achievements going unnoticed. Within this failed recognition, a detachment could form with dependents based upon feelings of exclusion and indifference (Hutchinson et al., (2016). This led to background fears of impending decline and death. Sikes and Hall (2017) reported further reflections around the experiences of time and waiting. By the time a diagnosis did arrive, emotional exhaustion prevented diagnosis from providing a 'Eureka moment'. There nonetheless remained a feeling that life was on hold and there was no destination end to the journey. Themes related to chronicity were noted. Time could feel more burdensome in the way that every declaration and event had to be waited upon. Such experiences were provoked by waiting for news, diagnosis, service provision and uncertainty about the future (Johannessen et al., (2016); Sikes and Hall (2018). Johannessen et al., (2015) articulated the conceptual meme of feeling 'zombified' by the experience and taint of EOD. The family reported coping in ways that were more; more dislocated, spatially lost and remote (Flynn and Mulcahy, 2013; Johannessen et al., 2017. Johannessen et al., (2015) recorded metaphorical experiences of 'slipping away' of being 'in another world' and 'spinning in a centrifugal machine'.

These concluded experiences of life in limbo including thematic descriptions of dealing with the often chaotic experiences of EOD. The experiences were overarching the confusion that EOD symptoms often provoked in PwEOD and reflected how family felt wrong-footed over daily routines and how to react in a 'new normal' life. These led to situations of feeling 'betwixt and between' with regard to not finding or locating a comfortable place to occupy in the family as the roles of child/adult, young adult/parent lost definition.

Stigma

Eleven of the sixteen studies reflected stigma was prevalent being reported by both PwEOD and with spouses and dependents; Hutchinson et al., (2016);Pipon-Young et al., 2011, Allen et al.,(2009); Johannessen and Moller, (2011);Sikes and Hall, (2017); Johannessen et al., (2016); Lockeridge and Simpson,(2012); Hoppe (2018); Gelman and Rhames, (2018); Johannessen et al., (2014) and Hoppe,(2018).

Johannessen and Moller (2011) reported how suicidal thoughts could be the outcome of not being confident about being in company with others due to their cognitive abilities. This could be shown in self-harm (Allen et al., 2009). However, most stigma emerged with interaction with others. This is where stigma could manifest obviously. Pipon-Young et al., (2011) discussed 'saving face' as a way by which people covered up their dementia through embarrassment and coped socially with potentially discriminating features of their lives. Johannessen and Moller (2011) described how PwEOD could feel marginalised through having no control over what others knew about them. Stigma by association (3) affected participants across some studies (Allen et al.),

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(3) Stigma by association is the placing of strain on family members arising from the connection

2009; Hutchinson et al., 2016). Dependents felt stigmatised by having a parent with EOD. At any time the participants reflected how difficult it was to manage their accounts to others often changing information dependent upon to whom they were directing their conversation (Hutchinson et al., 2016) Participant family carers vacillated between guilt and a form of coping. This form of coping allowed family carers to operate daily life whilst trying to shield the PwEOD from difficulties in the aftermath of a dementia diagnosis. Some studies reported how strategies were found to cope and deflect stigma both in the self and other family members (Sikes and Hall, 2017; Johannessen et al., 2016); Lockeridge and Simpson (2012) and Hoppe (2018). Crucially these experiences of being judged negatively emerged when interfacing with medical services (Sikes and Hall, 2017; Johannessen et al., 2016). Gelman and Rhames (2018) reported stigma like experiences reported through loss of status, self-efficacy and credibility. As a result of stigma, dependents could be left with feelings of irrecoverable lack of confidence.

The experiences concluding EOD stigma demonstrated how PwEOD, their spouses and children were affected in different ways; some of which were rooted in the micro-relationships within the family and physician relationships, others emerged within social life in society.

Coping with cautious optimism.

Under the theme of coping, eleven of the sixteen studies reported evidence of participants thriving with EOD (Johannessen et al., (2017); Johannessen et al., (2016); Rostad et al., (2013); Pipon-Young et al., (2011); Lockeridge and Simpson (2012); Millenaar et al., (2016); Johannessen et al., (2015);Gelman and Rhames (2018) Allen et al., (2009); Hutchinson et al., (2016) and Hall and Sikes, (2018).

Pipon-Young et al., (2011) related how PwEOD negotiated positive ways to live well with dementia; ; keeping active and involved and sharing a social life. Rostad et al., (2013) Lockeridge and Simpson (2012) and Johannessen. Rostad et al., (2013) Lockeridge and Simpson (2012) and Johannessen et al., (2017) reported participants as being able to reach acceptance and live with their diagnosed condition. Mental health issues affected coping resulting from unmet needs and lack in provision of support (Millenaar et al., 2016) with reference to young carer literature (4). Johannessen et al., (2016) and Johannessen et al., (2015) showed that part of coping was enabled by detaching from everyday suffering in life to gain a safe perspective. Getting used to a 'new normal' was possible alongside negotiating the difficulties of life drawing parallels with Parsonian obligations to work and contribute to self and society (Hall and Sikes, 2018). This impacted upon coping themes related to competing in a job market. Gelman and Rhames (2018) and Allen et al., (2009) drew upon claims that coping meant adjusting to living with fear and taking one day at a time. Hutchinson et al., (2016) reported how the identity could be developed which enhanced strategies of coping. Overarching the views on coping was securing a blend of normality which could be achieved provided economic pressures could be minimised or set aside. Resilience helped dependents distance themselves from traumatic family events and overwhelming tension. This meant that part of being a 'new normal' could result in not being economically competitive.

Conclusion

The review focused on the experiences of living with EOD. The results indicated that there was variation in the sample sizes, patient type, models and study design. Meta-ethnography was the selected approach to systematically review the literature. The main themes extracted were; i) biographical disruption ii) diagnosis, iii) losing life, friends and competences, iv) liminality and chronicity, v) stigma, and vi) coping with cautious optimism. The findings do reflect broad consensus that EOD prompts some different needs within the dementias' classifications. To this end, further research and development in the field is suggested in order to provide more knowledge for those providing and receiving care and services with particular sensitivity to each personal perspective in the triad of PwEOD, spouse and children.

Gaps in the literature and rationale for study

The literature review conducted in this chapter reflected a gap in the literature capturing the lived experience of PwEOD as reported in their own voice. As a result, only four studies were located reflecting the views of PwEOD were expanded with the experiences of family kin reporting on life at home with the PwEOD. This study aim therefore intended to investigate the direct experiences of PwEOD as they chose to reveal what living with the condition was like in everyday life. This review contributes to the modest extant knowledge of PwEOD. Of the available literature meeting criteria, there is still a tendency for the literature to seek family kinship views as opposed to drawing upon the PwEOD's views where these opinions and views could be provided. Given the gaps in current research, it was important to investigate further the challenges and opportunities for researchers and practitioners in researching, planning, implementing, and evaluating appropriate educational, supportive, and therapeutic services for individuals affected by EOD. Without a firm policy steer, PwEOD in the UK may be at risk of not receiving age-sensitive appropriate care.

Strengths and limitations of the study

Methodological strengths of this review include its reproducible and systematic nature including the application of a meta-ethnographic tool (Noblit and Hare, 1988). This review contributes to the modest extant knowledge of PwEOD. The process allowed similar concepts to be compared through a system of values enabling a full review of all available literature meeting the criteria.

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(4)Domestic estimates in Scotland suggest mental health difficulties amongst young carers indicate are reported twice as much as is reported by non-carers (Scottish Government, 2017).

There were a number of limitations with the review. Weaknesses were located around the paucity of studies located highlighting experience of having EOD. This was dealt with by expansion to encompass family kin. This allowed inclusion of a wide variation in age ranges and eclectic family role dyads. The literature illustrated that there are many legitimate perspectives affected by EOD. However, as thevariation in the studies was so wide, the synthesis could not fully account for the influence of these factors in the findings. Finally, included studies were conducted within Western cultures and within traditional 'nuclear families'. This had the effect of restricting generalisability of findings to other family structural systems. Studies were included from clinical settings which may distort some of the findings and assumptions.

Clinical implications

Participant views are deemed to be important for the planning of service provision and delivery. The results suggest strongly that planners of health and social care will be required to be answerable to an audience which is differently populated. Within any population of EOD there may likely be people who form part of the working population with children who still require support from parents both financially and emotionally. Care providers and planners must be cognisant that EOD is a chronic condition which whilst terminal, occurs in individuals and families with acute and vital ongoing needs. It is critical that qualitative research takes on board the nature of biographical disruption in an EOD diagnosis and considers the needs of a younger population. This is critical to the needs of other workers in the family including the vulnerability of children.

One noteworthy consideration in qualitative studies gathering person-centred-views is awareness of location and channel through which studies are performed. The studies relied upon hospital based or clinically led studies. Most made provision to ensure participant convenience and comfort. However, it is the case that ethical concern must be drawn to what extent participant groups may suspect that there are right answers or wrong answers. Medicine still is situated as the dominant hegemony governing care in times of medical frailty where people fear demise and death. It may be imagined that by complying with a particular view it may help participants to gain access to medication, clinical trials or enhanced service provision. For these reasons such concern should be taken over recruitment paths. This will ensure that participants are not unclear about what their data may realise for them personally in the short to medium term. In order to ethically and respectfully manage the expectations of participants, the aims and likely outcomes of studies should very clearly be illustrated to potential participants.

Future research implications

Further contributions to the research body including qualitative studies on experience could sensitise policy-makers, health and social care providers to the factors likely to enhance service provision and care. EOD is a classification which brings its own separate concerns from those presenting in later onset dementias. Therefore, it may be fruitful for future research to explore further the experience of PwEOD. This would relate to both the personal impacts as a distinct condition with different symptoms to that of older onset conditions as well as experiences of coping within work and family settings.

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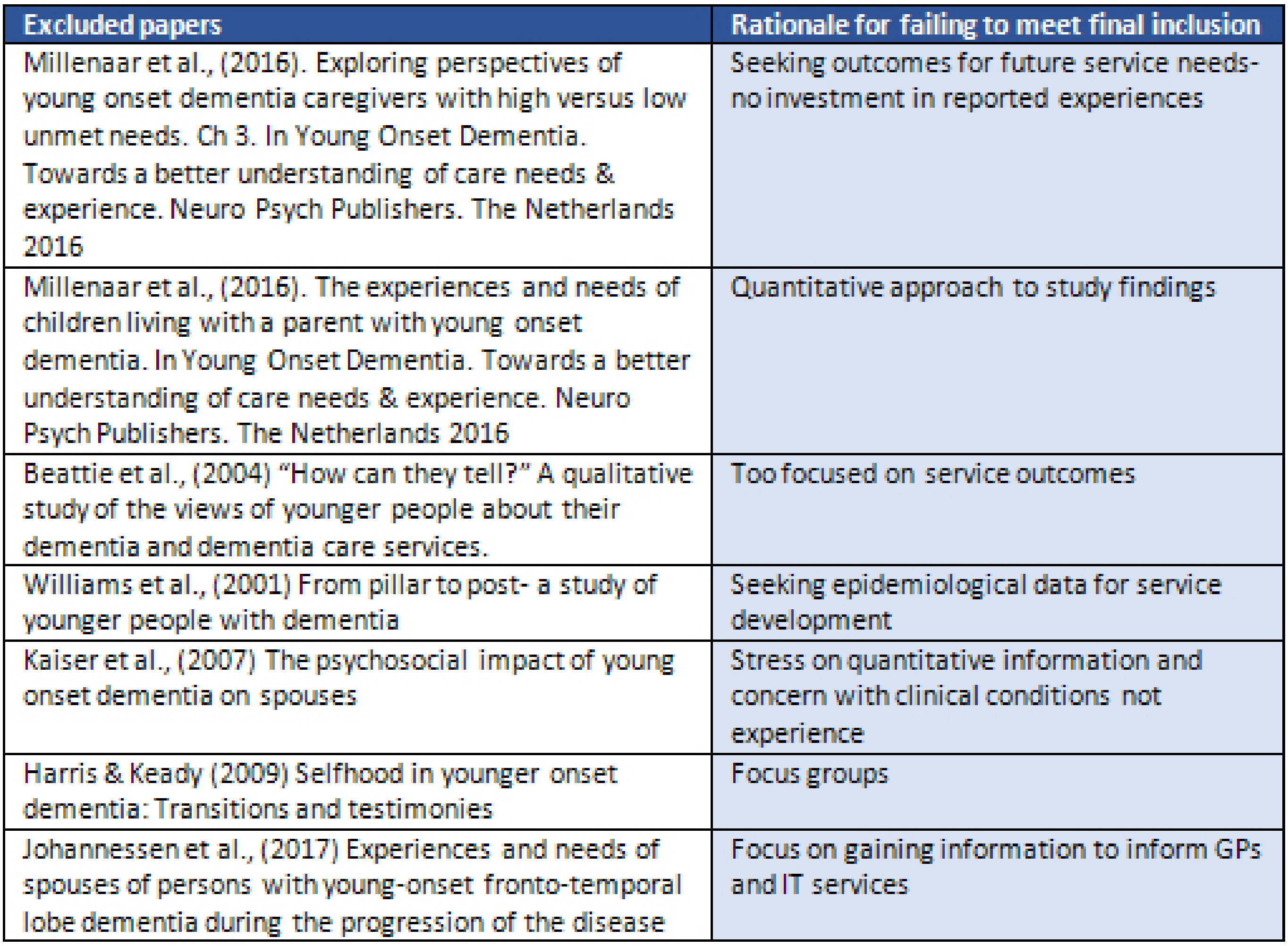
Appendices

Appendix 1



Excluded papers	Rationale for failing to meet final inclusion
Millenaar et al., (2016). Exploring perspectives of young onset dementia caregivers with high versus low unmet needs. Ch 3. In Young Onset Dementia. Towards a better understanding of care needs & experience. Neuro Psych Publishers. The Netherlands 2016	Seeking outcomes for future service needs- no investment in reported experiences
Millenaar et al., (2016). The experiences and needs of children living with a parent with young onset dementia. In Young Onset Dementia. Towards a better understanding of care needs & experience. Neuro Psych Publishers. The Netherlands 2016	Quantitative approach to study findings
Beattie et al., (2004) "How can they tell?" A qualitative study of the views of younger people about their dementia and dementia care services.	Too focused on service outcomes
Williams et al., (2001) From pillar to post- a study of younger people with dementia	Seeking epidemiological data for service development
Kaiser et al., (2007) The psychosocial impact of young onset dementia on spouses	Stress on quantitative information and concern with clinical conditions not experience
Harris & Keady (2009) Selfhood in younger onset dementia: Transitions and testimonies	Focus groups
Johannessen et al., (2017) Experiences and needs of spouses of persons with young-onset fronto-temporal lobe dementia during the progression of the disease	Focus on gaining information to inform GPs and IT services