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The views of adults with neurodegenerative diseases on end-of-life care: a metasynthesis

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Abstract

Objective: Improving end-of-life care for people with neurodegenerative diseases is seen as a clinical priority. In order to do this, it is important to take into account the views expressed by people with these conditions on their experiences of this care. The purpose of this review was to provide a thematic synthesis of the views of adults with neurodegenerative diseases on end-of-life care.

Methods: After a systematic search, 13 articles were included and thematic synthesis was used to collate and interpret findings.

Results: Four analytical themes were identified; 1) Importance of autonomy and control; 2) Informed decision-making and the role of healthcare professionals; 3) Contextual factors in decision-making; 4) The pitfalls of care.

Conclusion: Participants' views were framed by the context of their lives and experience of their illness and these shaped their engagement with end-of-life care. Given the varying disease trajectories, care needs to be individualised and needs-based, implementing palliative care in a timely way to prevent crises and loss of autonomy.

Keywords: Neurodegenerative disease, end-of-life care, palliative care, qualitative, metasynthesis,

Introduction

Improving end-of-life care (EOLC) is a significant health care priority worldwide, particularly as individuals are living longer with conditions which previously significantly shortened lifespan. Palliative care offers a theoretical model of EOLC for people with life-limiting conditions and is promoted by the World Health Organization as the preferred approach (Wolrld Health Organization, 2002). Conceptually, the aim of palliative care is to improve the quality of life of patients with life-threatening illnesses and their families by providing compassionate care that emphasises the importance of attending to individuals' psychological and spiritual needs as well as the physical aspects of dying (Morrison and Meier, 2004). Accordingly, central importance is placed on the wishes of the dying individual. Effective palliative care has three main aims: management of physical symptoms and side effects; continuing communication of treatment goals between doctor, patient and family; and psychological, spiritual and social support for patient and family.

Neurodegenerative diseases

A systematic review by Zimmermann, Riechelmann, Krzyzanowska, Rodin, and Tannock (2008) demonstrated that palliative care approaches improve family satisfaction with care for people with chronic conditions. However, access to palliative care is predominantly restricted to people with cancer (Solano, Gomes, & Higginson, 2006). Indeed an important group of patients who have not benefitted fully from effective EOLC are those with neurodegenerative diseases such as Alzheimer's disease (AD), Parkinson's disease (PD), Huntington's disease (HD) and motor neuron disease (MND), also known as amyotrophic lateral sclerosis (ALS). Although distinct conditions, these can be described as largely adult-onset and characterised by progressive loss of structure or function of neurons, including neuronal cell death (Ross and Poirier, 2004).

They also share a number of characteristics, such as progressive loss of functioning in a range of domains, although rate of deterioration is varies.It is estimated that, since 2002, chronic conditions (including degenerative diseases) have been the largest cause of death globally (World Health Organization, 2008). Despite their capacity to cause significant morbidity and shorten life, neurodegenerative diseases are not typically associated with palliative care (Luddington, Cox, Higginson, & Livesley, 2001)

Moreover, research suggests that the quality of EOLC for people with neurodegenerative diseases is often poor (Bede et al., 2011; McGarva, 2001). This is in spite of the fact that people with neurodegenerative diseases are often aware in advance that their cognitive abilities and capacity for communication will decline and for some (e.g. MND) the time from diagnosis to death is relatively short. Of course it is possible that there are significant challenges with the implementation of palliative care in this population. Disease trajectories for neurodegenerative diseases are more variable, ranging from months to decades, and so prognosis can be uncertain (Lindenberger and Meier, 2013). A further barrier may be the belief that palliative care should only be offered at the end stage of an illness (Kristjanson, Toye, & Dawson, 2003). However, due to the broad scope of palliative care, it can in principle be integrated with curative treatment and need not preclude life-prolonging treatments.

However, in order for care to be best provided, a range of research is needed to create an effective evidence base and this includes qualitative research as this can focus on views and experiences in a way that meta-analyses cannot. It is particularly important that views from patients (as opposed to professionals) are emphasised as it has been shown in other research (e.g., in the assisted dying debate: see Duncan, & Parmelee, 2006) that views on what is seen as optimal care differ between groups. It is also useful to note that as the push for palliative care to

be provided for people with NDDs is increasing, producing a synthesis on patient views is timely. Services need to develop which meet the needs of patients. As some conditions (e.g., HD) are relatively rare, the synthesis can also help provide evidence in the absence of a separate and sufficient corpus of research for each condition. Consequently, it was decided that it was timely and necessary to conduct a qualitative meta-synthesis to explore the views of people with neurodegenerative diseases on EOLC.

Method

Data collection

A systematic search of four electronic databases (Academic Search Complete, PubMed, Cumulative Index to Nursing and Allied Health Literature (CINAHL) and PsycINFO) was conducted in January 2016. In order to identify search terms, the research question was separated into its essential parts: neurodegenerative diseases and EOLC. Search terms were generated based on these two areas (see Table 1) and refined using the index terms for each database (for example, subject headings, APA descriptors, database thesauruses and the MeSH 'explode' function was used in databases where this was an option). The search terms were combined using the Boolean terms 'and' and 'or'.

Insert Table 1 about here

Inclusion and exclusion criteria

The selected studies were screened using the following inclusion criteria: papers must include (a) participants with an adult onset neurodegenerative disease (multiple sclerosis, despite being more controversial, was also included (e.g., Chaudhuri, 2013). (b) explored participants' views in relation to EOLC (either as a whole experience or in relation to specific

care practices), either through their actual experience or their views on its availability and/or quality; (c) reported findings of qualitative research using recognised methods of data collection and analysis, (d) been available in English and (e) been published in a peer-reviewed journal.

Neurodegenerative disorders are one of the most problematic classifications of disease (Du and Pertsemlidis, 2011). The focus here was on progressive disease processes which affect specific neurons in specific functional anatomical systems (Przedborski, Vila, & Jackson-Lewis, 2003) and are not due to an external cause. Consequently, studies were excluded which focused on a) neoplasm, oedema, haemorrhage, and trauma of the nervous system or infection or poisoning (Przedborski, et al., 2003).

The first author initially assessed the titles and abstracts, and full text studies were obtained and screened. Studies that focused on the perspective of caregivers, family members, or healthcare professionals were excluded unless patients' views were reported separately. Sandelowski and Barroso (2003) suggest a classification system for appraising the depth of analysis. Consistent with the recommendations of this system, 'no-finding' studies (e.g. articles consisting of uninterpreted narratives or diaries) and topical survey studies were excluded. To illustrate, a single case report (Mitsumoto and Rabkin, 2007) was excluded, despite its use of rich quotes, because no qualitative analysis was evident and it was considered a 'no-finding' study. Figure 1 summarises the application of the inclusion and exclusion criteria to the papers found.

Insert Figure 1 here

Critical appraisal

The quality of each study was appraised using the Critical Appraisal Skills Programme (CASP) checklist for qualitative research (Critical Appraisal Skills Programme, 2017) to identify potential limitations. A three-point numerical scoring system was used, developed by Feder,

Hutson, Ramsay and Taket (2006), whereby papers were given a score of 0-2 on each of the CASP's 10 items. . The purpose of this appraisal was not to exclude studies (Bondas and Hall, 2007) but to give weighting to their contribution in the synthesis (Topcu, Buchanan, Aubeeluck, & Garip, 2016). However, all papers scored sufficiently highly and none of the papers were given less emphasis (see Table 2).

Characteristics of the selected studies

The 13 selected studies had sample sizes ranging from 2-34 people. Diagnoses included HD (*n* = 1), MND (*n* = 7), MS (*n* = 2), dementia (*n* = 1) and PD (*n* = 2) and participants varied widely in stage of disease. Seven of the selected studies used semi-structured interviews, one study used narrative interviews (employing a variety of modes of communication including email, diary and telephone), three studies did not detail the structure of their interviews, describing them as 'in-depth interviews', one study reportedly used 'episodic face-face interviews' and one study used autobiographies. Four of the selected studies used grounded theory, three studies thematic analysis, three studies content analysis, one study used the 'constant comparison method' and two studies used phenomenological based approaches to analysis.

The data collection for the studies took place in a variety of settings, predominantly individuals' homes, but also hospitals, outpatient clinics, hospices and nursing homes. Two studies used the same participants (Foley, Timonen, & Hardiman, 2013, 2014); however, they each provided a novel set of findings that were complementary and, consequently, both studies were included in the metasynthesis. Studies came from a number of countries, perhaps not surprisingly all more associated with a more advanced approach in palliative care: United Kingdom (*n* = 3), Australia (*n* = 2), Canada (*n* = 2), Germany (*n* = 2), Ireland (*n* = 2),

Netherlands (n = 1), and Sweden (n = 1). See Table 2 for more in-depth characteristics of the studies.

Insert Table 2 about here

Data extraction, analysis and synthesis

An inductive thematic synthesis approach (Thomas and Harden, 2008) was used to synthesise the original content of the studies and produce a more advanced understanding through interpretive themes. This approach was chosen due to its emphasis on synthesising the content of the individual papers' themes as opposed to assessing their linguistic structure. The approach also privileges the development of more analytic themes as opposed to the collection of similar content. The three conceptual stages of thematic synthesis were followed: the 'line-by-line' coding of text; the development of descriptive themes; and the generation of 'analytical themes'. Each paper was read and re-read so that the first author became familiarised with and immersed in the data. Reflective notes were kept at this stage to enhance depth of understanding. The studies were then coded, much as they would be in primary qualitative research, to capture the themes within the original studies relating to the review question on people's views on end-of-life care.

The codes were then analysed, identifying differences and similarities between them, to generate an initial set of descriptive themes. These descriptive themes did not attempt to reinterpret the primary findings; closeness to the original data sets is essential in this secondary analysis. These themes were considered to extract the meaning of studies in a consistent way that was close to the original interpretations of the respective authors.

Finally, more interpretative analytical themes were refined from the descriptive themes by the first author and then developed by the fourth author. These analytical themes are more

tacit, abstract entities and attempt to capture the essence of the data (DeSantis & Ugarriza, 2000). The contribution of each paper to each analytical theme is provided in Table 3.

Insert Table 3 about here

Results

Four analytical themes were identified in the analysis of the interview data.

Importance of autonomy and control

A strong sense of the importance of autonomy and control ran through participants' accounts. For example, participants wanted to take an active role in decision-making about their care. Although there was wide variation in individuals' wishes (for example, for life-prolonging treatments, for hastened death, or for preferred place of death), participants were clear that these views should be respected and they should be involved in reaching a final decision. 'And I would like to know how it will be managed and what my choices are' (Whitehead, O'Brien, Jack, & Mitchell, 2012, p. 372). Desire for autonomy and control occurred in the context of the participants' feeling that they had reduced control over many aspects of their lives as a result of their illnesses. Participants were acutely aware of the progression of their neurodegenerative disease and spoke about this affecting their ability to make active decisions. For example, some people felt that their choices about medical interventions were restricted because of their condition. 'It's not a choice, you either use it or can't breathe of a night, so there is no choice.' (Greenaway et al., 2015, p. 1005). Conversely, other participants felt reassured by making active decisions about their care, though sometimes they had to fight to obtain this.

> I felt absolutely no control so I said to them [healthcare professionals] 'I feel a complete disconnect ... [that] I'm kind of apprehensive about a lot of things' ... now I am connected and it's a team effort. Now I'm able to sit down and discuss it [EOLC] with them ... make decisions when you are in control [of care] (Foley, et al., 2014, p. 321)

Loss of autonomy was associated with diminished quality of life and meaningfulness.

> I asked about a possible trache [tracheotomy] and ventilator for the future. He said a definite 'NO', he wouldn't advise it and neither would the other consultant there….But what about what I want!!!! That didn't seem to matter… I feel useless and as if my life isn't even worth talking about (Whitehead, et al., 2012, p. 372)

Other participants questioned the value of life-prolonging treatments given the life-limiting nature of their neurodegenerative diseases, perceiving that interventions could potentially prolong suffering. This was seen as unacceptable and participants talked about disengaging from care, for example, by refusing life-prolonging treatments and supportive care, when this was anticipated. For this group, autonomy over when and how to engage with their care was more important than extending their life.

There were a range of views on assisted dying and euthanasia. Ending life in this way was chosen by some participants in countries where these were legal, and wished for in countries where these were not. For others, assisted dying and euthanasia were antithetical to their principles and therefore rejected. However, for all self-determination was important, regardless of their beliefs or personal intentions.

Informed decision-making and the role of healthcare professionals

This theme conceptualised the information needs of participants and their, often implicit, belief that health care professionals were responsible for meeting these. Many participants spoke about a lack of information about their care, which inhibited their ability to make fully informed choices. 'I didn't get the brochures or anything from the doctors… there's really not much there to help (Giles and Miyasaki, 2009, p.121). Similarly, some participants felt that there was a lack of awareness of their neurodegenerative disease, even among health professionals, which added

to their feelings of frustration. Some participants talked about managing the lack of information from healthcare professionals by actively seeking out information (e.g. from the internet). A smaller number of participants, in contrast, felt that they had all the information they needed, which enabled them to make confident decisions. 'They covered everything I wanted to know and the questions they asked were the right questions' (Poppe, Burleigh, & Banerjee, 2013, p. 3).

Generally, there was ambivalence within and across studies over how much information was enough. Many participants wanted information to be able to make decisions about their care but reported anxiety about the prospect of a bleak prognosis. Fear of confronting their potential deterioration and death meant that some participants rejected advanced care-planning discussions, finding them futile or dispiriting. This variability in individuals' information needs illustrates the complex task facing healthcare professionals.

Most participants valued their experiences of advanced care planning, finding it a helpful experience that gave them important information, the opportunity to have significant conversations with loved ones, alleviated their worries about their care in the future and reassured them that their wishes would be honoured.

> I decided to make arrangements and things so if anything happens now they all know, what I want and what's happening so it saves me worrying about it (Poppe, et al., 2013, p. 3)

Similarly, participants disclosed their reticence in discussing EOLC. Many participants expressed their wish to have open conversations about EOLC but not knowing what to ask for or feeling able to raise this difficult topic. Accordingly, their expectations of care were not met.

Participants wanted a trusting relationship with their healthcare professionals and felt more reassured about their care when this was realised. Trust was placed in healthcare professionals when support and empathy were offered as well as sensitivity to their needs.

> I was concerned that it was actually something that would help me but could weaken my ability in the daytime. And it really reassured me that actually it would make it better. That was what I wanted to hear (Greenaway, et al., 2015, p. 1007).

It is possible that trust is a key determinant of participants' satisfaction and presupposes other factors that comprise good care. Noticeably, when trust was not present participants across studies appeared to be silent and disengaged from their care.

Contextual factors in decision-making

Participants across studies communicated the importance of contextual factors in their EOLC. Family, for example, played a central role in decision-making. Most participants saw their neurodegenerative disease in a family context and frequently discussed their preferences for EOLC with family members and came to decisions endorsed by the family. A prominent desire for participants was not to be a 'burden' on family members. However, it was also important for participants to 'be there' for families to support and be supported by them. This demonstrates the complexity of decision-making. Family could be supportive but also demanding and participants often struggled to balance their personal needs with those of the family unit. At times this resulted in complex emotions and participants feeling restricted in their decision-making about care. For example, on considering assisted suicide one participant commented:

> I had to consider my family and the implications. It's like a suicide… the pebble in the water. It spreads out. (Foley, et al., 2014, p. 72).

Family influenced not only decision-making but practical care. For example, some participants refused paid carers because family members occupied that role. These participants spoke about the importance of reciprocal family support. Alternatively, where participants had no family to take into account, this influenced their choices. ‘You see, I don’t feel I have the need to hang on at any cost. If I had a husband, wife, children, it might be different. So I’m free to make the choice.’ (Foley, et al., 2014, p. 72).

Another important contextual factor was participants’ perceived status in comparison with others. Frequent social comparisons were made in order to form opinions on care. These were often first-hand experiences, such as family members’ experiences of the end stages of neurodegenerative disease: ‘Not the way my mother suffered’ (Booij, Rödig, Engberts, Tibben, & Roos, 2013, p. 326). Having witnessed a parent or other family member affected by the same neurodegenerative condition seemed to shape participants’ wishes for end of life.

The variability of disease progression complicated decision-making for participants. Many participants were acutely aware of situations, points in their trajectory, that they felt would be unbearable and lead them to seek means to hasten their death: ‘As soon as I become dependent on others, then it is time to pull the plug’ (Booij, et al., 2013, p. 326). The concept of time therefore influenced participants’ views on EOLC. Some participants made the conscious choice to ‘live in the moment’, which meant decisions on interventions represented that particular situation in time. However, participants were aware that they might change their mind in the future and revisit these decisions. ‘I was going to write a living will but I’ve decided with my GP [general practitioner], now that there’s no point really, if I wrote a living will now you might want to change your mind.’ (Whitehead, et al., 2012, p. 327).

Some participants held on to hope that the future might hold different options for care. This sense of hope sustained them in adjusting to the ramifications of their neurodegenerative diseases and engaging with their EOL care. '[I've] just got to stay fit and healthy and exercise, take my medication and hope… they have some luck with the stem cell testing' (Hudson, Toye, & Kristjanson, 2006, p. 89).

The pitfalls of care

Participants described varied and individualised care needs, including emotional, physical and practical support. Often the care received was inadequate and viewed as difficult to access and a demanding experience: 'And now I get pats on the cheek or, worst of all, on the head, like a child. So I hate it. That patronizes compassion. So far from compassion and empathy' (Rosengren, Gustafsson, & Jarnevi, 2015, p. 79).

Often the promise of care was different from the reality and participants were left disappointed by the service they received. One participant spoke about how support groups, designed to provide psychosocial support, were unhelpful.

> I guess I don't really need to be around people like that at this stage… what are you going to do? Sit around and talk about how much you shake at night… it might tend to be a little depressing for the type of person that I am at the moment (Hudson, et al., 2006, p. 90).

Accordingly, specialist support services were highly valued, where healthcare professionals were well informed about their disease and able to provide continuity of care. 'If I have any questions I visit the professor. And my GP, he doesn't really know about the disease' (Booij, et al., 2013, p. 328).

Often negative emotions were associated with the inadequacy of care. However, many participants seemed resigned to the fact that care could not meet all of their needs. For example, some participants expressed a need for meaningfulness. This involved staying connected to other people and their own values. This was least commonly met by healthcare professionals and perceived as an inappropriate conversation topic.

> Those are pretty private things, like attitude towards life or further progress or whatever, and all those things you can talk about for hours, and I think that's why the doctor doesn't even bother getting into all that. Instead it's just acute problems, and that was it (Galushko et al., 2014, p. 278).

Many participants expressed a wish to die at home. For them, home was a meaningful place of care where family could support them and sadness was expressed at the thought of this not being possible. 'With a tracheostomy, I'm stranded to a nursing home.' (Lemoignan and Ells, 2010, p. 211). This demonstrates how participants questioned the meaning of interventions and made decisions about them based on their own values and goals.

Discussion

The aim of this synthesis was to collate the views of adults with neurodegenerative conditions on end of life care; end of life care for this group is a growing clinical priority and it is important to understand and position these individuals as key stakeholders in the development of appropriate services. A systematic search process found 13 articles suitable for inclusion and these were analysed using a form of thematic synthesis.

This synthesis has revealed a number of important empirical and clinical insights into how people with neurodegenerative diseases viewed EOLC. Moreover, while no claims are made of generalisability across all neurodegenerative diseases, the views within and across studies and disease conditions were remarkably similar. Empirically, the themes 'Importance of autonomy and control' and 'Informed decision-making and the role of healthcare professionals are consistent with existing research into patient preferences for healthcare. These findings confirm the results of a systematic review (Kiesler and Auerbach, 2006) exploring matching patients' preferences for information, decisional control, and consultative interpersonal style. This found that patients vary in how active a role they play in their healthcare and how much responsibility they wish to take over decisions made. When the interactional styles of healthcare professionals and their patients do not align, outcomes such as effectiveness of treatment and patient satisfaction are negatively affected. The researchers suggest that healthcare professionals should not interact with patients in a rigid, standardised way but in a manner that matches patients' preferences for information and involvement in decision-making. This key finding in this metasynthesis - that many participants were dissatisfied with the information given about their EOLC - fits with research suggesting that healthcare professionals tend to be poor at communicating about end-of-life issues (Milberg and Strang, 2000).

Empirical evidence on neurodegenerative diseases supports the significance of autonomy and the notion that patients wish to be informed and actively involved in their care (e.g., Joffe, 2003; Tramonti, Bongioanni, Di Bernardo, Davitti, & Rossi, 2012). However, it is noteworthy that some people want little or no role in decision-making (Benbassat, Pilpel, & Tidhar, 1998; Say, Murtagh, & Thomson, 2006) and a minority of participants in this synthesis preferred healthcare professionals to make decisions about their care. It is possible to view this in a

positive way, as individuals paradoxically exercising their autonomy and control by giving their responsibility for decision-making to healthcare professionals. However, Rodin, Timko and Harris (1985) suggest that people who perceive less self-control tend to take less accountability for their health, be less likely to make use of health protective behaviours and have lower immunological response. Certainly perceived loss of control is a feature in neurodegenerative diseases and the need to reassert control when uncertainty and lack of control are so evident is a prominent characteristic for many (Eccles and Simpson, 2011).

From a clinical perspective, the shortcomings in care identified by participants were largely related to their psychosocial needs rather than unmet biological or medical needs. Despite the widespread acceptance of the biopsychosocial model of health (Engel, 1980) and subsequent move towards the delivery of holistic healthcare, Brown, Alaszewski, Swift, and Nordin (2011) argue that the purpose of medicine is still perceived to be to correct the 'problematic body'. Yet evidence from this review suggests that this conceptualisation is at odds with the needs of patients. For example, recent studies (Chiò et al., 2004; Ho, Gilbert, Mason, Goodman, & Barker, 2009; Simpson, Lekwuwa, & Crawford, 2014; Yamout et al., 2013) have found that mental health variables were more influential than physical ones in determining health-related quality of life in people with neurodegenerative disorders. Consequently, a palliative care approach would be beneficial to many people with neurodegenerative disease. Timing is important as some participants reflected that they wished to 'live in the moment' and the disease trajectory of many neurodegenerative conditions is long. Therefore, although it is generally beneficial to involve care early, an individualised approach is needed, with high quality care provided over time at the degree to which it is required. More widely, this will require a shift in care provision from the more common model of crisis management to a framework of chronic

care, incorporating palliative care, which is often lacking, particularly for non-cancer conditions (Siouta, van Beek, Preston, et al., 2016; Siouta, Van Beek, van der Eerden, et al., 2016). This should include anticipating disease milestones and addressing them in a timely way to prevent crises and loss of autonomy.

In terms of review limitations, the studies included were restricted to those in the English language; relevant studies in other languages may exist. However, Doyle (2003) argues that the sample for qualitative review is justifiably purposive, not exhaustive because the aim is interpretation, not prediction.

Additionally, although the synthesis found consistency across papers and neurodegenerative diseases share many features, they are heterogeneous conditions. For example, the median life expectancy for MND is 2–5 years after diagnosis (Mitchell & Borasio, 2007) whereas HD is around 20 years (Simpson, 2007). It is possible that some views on end-of-life care, such as around desire for hastened death, might differ between these populations and it would be useful to explore this further. At the moment, given the greater number of studies involving people with MND – which has the shortest life expectancy after diagnosis - in this review, it is possible that this has overly influenced the analysis.

In conclusion, this synthesis offers an enhanced understanding of the views of people with neurodegenerative diseases on EOL care by providing a higher level of analysis than what would be gleaned from individual studies. This synthesis highlights that the care needs of people with neurodegenerative diseases are often not met, that autonomy and a sense of control are key, that views are contextual and local, and that specific illness belief factors (such as the meaning participants make, their beliefs and their preferences for information) shape engagement with

care. Also, this synthesis promotes the importance of social context and recommends that care should be individualised, placed in the context of the life of the individual and implemented in a needs-based, integrated way across the disease trajectory as opposed to a traditional, symptom-led approach. From these studies it is not possible to determine if it is a lack of palliative care services that is the difficulty for individuals or if palliative care services need to improve (there is certainly some evidence of both across the papers). Indeed an area for more research would be in focusing only on the experiences of those who are receiving palliative care and a focus on those for whom the system has worked well.

Disclosure of interest

The authors report no conflicts of interest.

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Tables

Table 1. *Final search terms for each database.*

Database	Search terms
Academic Search Complete	(DE "NEURODEGENERATION") OR (DE "HUNTINGTON'S chorea") OR (DE "PARKINSON'S disease") OR (DE "AMYOTROPHIC lateral sclerosis") OR (DE "ALZHEIMER'S disease") OR (DE "MULTIPLE sclerosis-- Treatment") OR (DE "MOTOR neuron diseases") AND (DE "TERMINAL care" OR DE "HOSPICE care" OR DE "PALLIATIVE treatment")
CINAHL	(MM "Neurodegenerative Diseases+") OR (MM "Multiple Sclerosis+") AND (MM "Palliative Care") OR (MM "Terminal Care+") OR (MM "Hospice Care") OR (MM "Hospice and Palliative Nursing")
PubMed	("Neurodegenerative Diseases"[Mesh]) OR "Multiple Sclerosis"[Mesh]) AND ((((("Palliative Care"[Mesh] OR "Hospice and Palliative Care Nursing"[Mesh]) OR "Terminal Care"[Mesh]) OR "Hospice Care"[Mesh]))
PsycInfo	("Neurodegenerative Diseases"[Mesh] OR "Multiple Sclerosis"[Mesh]) AND ((("Palliative Care"[Mesh] OR "Hospice and Palliative Care Nursing"[Mesh]) OR "Terminal Care"[Mesh]) OR "Hospice Care"[Mesh]) AND ("Neurodegenerative Diseases"[Mesh] OR "Multiple Sclerosis"[Mesh]) AND ((("Palliative Care"[Mesh] OR "Hospice and Palliative Care Nursing"[Mesh]) OR "Terminal Care"[Mesh]) OR "Hospice Care"[Mesh])

Table 2. *Summary information of the studies included in the metasynthesis*

1st Author, Title, Year and Country	Aims	Method	Analysis	(Total) number, gender and age range of participants	Key findings/themes relevant to the review	CASP score
Booij. Euthanasia and advance directives in Huntington's disease: Qualitative analysis of interviews with patients (2013, The Netherlands) (Booij, et al., 2013)	To obtain in-depth information about patients' thoughts on and attitudes to euthanasia, physician-assisted suicide and the use of advance directives in Huntington's Disease	Semi-structured in-depth interviews	Qualitative analysis based on grounded theory	(14) 8 women, 28-70	Background of EOL issues, presence of wishes, knowledge of advance directives, role of family and physician	18
Burchardi. Discussing living wills. A qualitative study of a German sample of neurologists and amyotrophic lateral sclerosis patients (2005, Germany)(Burchardi, Rauprich, Hecht, Beck, & Vollmann, 2005)	To investigate how neurologists provide information about living wills to Amyotrophic Lateral Sclerosis patients. To explore if discussions met patients' needs and expectations.	Semi-structured interviews	Grounded theory techniques	(15), 5 women, 43-78, *M*= 59	Healthcare professional biases, timing of interventions, importance of choice, importance of trust, desire to hasten death, aim to reduce suffering, future decline	16
Foley. Acceptance and decision making in amyotrophic lateral sclerosis from a life-course perspective (2013 Ireland) (Foley, et al., 2013)	To identify key psychosocial processes that underpin how and why people with Amyotrophic Lateral Sclerosis engage with services.	In-depth interviews	Grounded theory	(34) 17 women, 37-81,	Life and death, acceptance, family context to decision making, views on assisted dying,	18
Foley. Understanding psycho-social processes underpinning engagement with services in motor neurone disease: A qualitative study (2014, Ireland) (Foley, et al., 2014)	To identify key psychosocial processes that underpin how and why people with motor neurone disease engage with services.	In-depth interviews	Grounded theory	(34) 17 women, 37-81,	Control over care, reassurance from healthcare professionals, importance of trust, meaning of life, meaning of interventions	18
Galushko. Unmet needs of patients feeling severely affected by multiple sclerosis in Germany: A qualitative study (2014, Germany)(Galushko, et al., 2014)	To explore the subjectively unmet needs of patients feeling severely affected by multiple sclerosis.	Semi-structured in-depth interviews	Inductive qualitative content analysis	(15) 9 women, 23-73, *M*= 47	Inadequacy of care, family context, trust/lack thereof in healthcare professionals, meaning and identity	15
Giles. Palliative stage Parkinson's disease: patient and family experiences of health-care services (2009, Canada)(Giles and Miyasaki, 2009)	To understand the lived healthcare experiences of people with Parkinson's and their families and the needs flowing from these experiences.	Semi-structured in-depth interviews with family groups	Interpretative phenomenological analysis	(2) 2 women, 75-77, *M*= 76	Missing information, being on your own, meaning and identity	17
Greenaway. Accepting or declining non-invasive ventilation or gastrostomy in amyotrophic lateral sclerosis: patients' perspectives (2015, United Kingdom)	To identify factors associated with decisions made by patients with amyotrophic lateral sclerosis to accept or decline non-invasive ventilation or gastronomy	Semi-structured interviews	Thematic analysis	(21) 8 women women, 41-76	Perceptions of choice, fear, perceived need and acceptance, influence and support of healthcare professionals, trust,	18

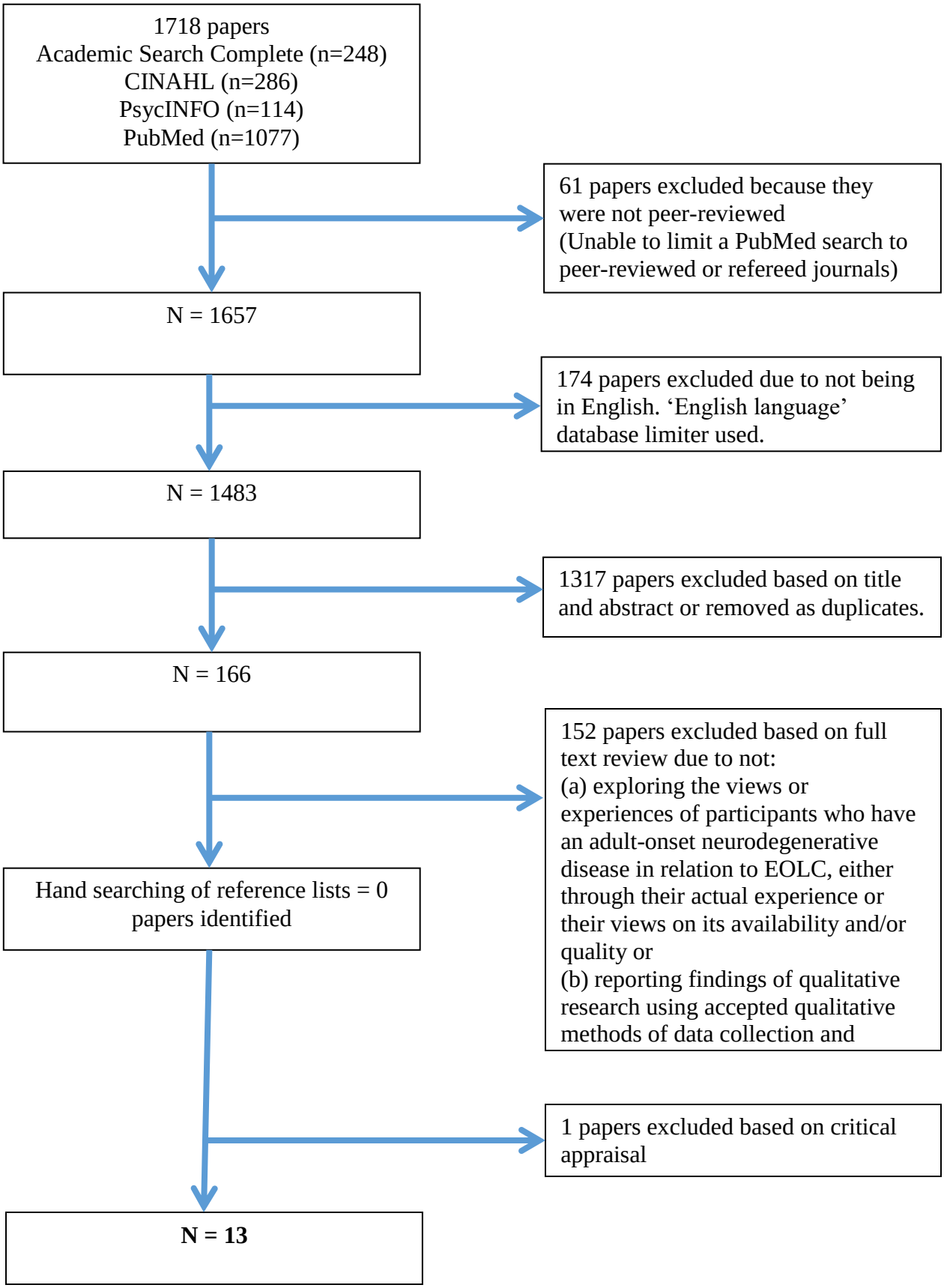
(Greenaway, et al., 2015)					family context, information needs	
Hudson. Would people with Parkinson's disease benefit from palliative care? (2006, Australia) (Hudson, et al., 2006)	To describe the experience of Parkinson's disease and consider the relevance of palliative care for this population	Semi-structured interviews	Thematic analysis	(8) 4 women, 40->80	Contextual factors, emotional impact, staying connected, managing physical challenges, finding help	19
Lemoignan. Amyotrophic lateral sclerosis and assisted ventilation: how patients decide (2010, Canada) (Lemoignan and Ells, 2010)	To better understand the experience of decision-making about assisted ventilation for amyotrophic lateral sclerosis patients	Semi-structured interviews	Qualitative phenomenology methodology	(9) 3 women, 36-72	Meaning of intervention, importance of context, importance of values (autonomy), effect of fears, need for information, adaptation and acceptance	18
Poppe. Qualitative evaluation of advanced care planning in early dementia (ACP-ED) (2013, United Kingdom) (Poppe, et al., 2013)	To explore the acceptability of discussing advanced care planning with people with memory problems and mild dementia shortly after diagnosis	In-depth interviews	Constant comparison method	(12) 8 women, 68-88, *M*= 79	Information needs, anxiety about future, reassurance from healthcare professionals,	14
Rosengren. Every second counts: Women's experience of living with amyotrophic lateral sclerosis in the end-of-life situations (2015, Sweden) (Rosengren, et al., 2015)	To describe patients' experiences of living with ALS in the end-of-life situations	Biographies	Manifest content analysis	No demographic information available	Suffering, meaningfulness, unmet care needs, context to decision making	12
Whitehead. Experiences of dying, death and bereavement in motor neurone disease: a qualitative study (2012, United Kingdom) (Whitehead, et al., 2012)	To explore the experiences of people with motor neurone disease, current and bereaved carers in the final stages of the disease and bereavement period	Narrative interviews	Thematic analysis	(24) 16 women, 25-84	Fears for the future, Information seeking, social and family context, life and death, wishes and decision making, importance of choice, influence of healthcare professionals, euthanasia	19
Wollin. Supportive and palliative care needs identified by multiple sclerosis patients and their families (2006, Australia) (Wollin, Yates, & Kristjanson, 2006)	To identify the supportive needs of individuals with multiple sclerosis and their families	In-depth semi-structured interviews	Content analysis	(13) 6 women, 23-55	Lack of support, tracking down services and information,	17

Table 3: Identified analytical themes and contributing findings and concepts from each study

Analytical themes	Booij (2013)	Buchardi (2005)	Foley (2014a)	Foley (2014b)	Galushko (2014)	Giles (2009)	Greenaway (2015)	Hudson (2006)	Lemoignan (20015)	Poppe (2013)	Rosengren (2015)	Whitehead (2011)	Wollin (2006)
Importance of autonomy and control	✓	✓	✓	✓			✓	✓	✓	✓		✓	
It's the role of HCPs to get the balance of information right (and provide context for discussions)	✓	✓		✓	✓	✓	✓	✓		✓	✓	✓	✓
Decision making occurs in context	✓	✓	✓		✓		✓	✓	✓	✓	✓	✓	
Care can't meet all of one's needs		✓		✓	✓	✓	✓	✓	✓	✓	✓	✓	✓

Figure captions

Figure 1. Search process flow chart


1718 papers
Academic Search Complete (n=248)
CINAHL (n=286)
PsycINFO (n=114)
PubMed (n=1077)
61 papers excluded because they were not peer-reviewed (Unable to limit a PubMed search to peer-reviewed or refereed journals)
N = 1657
174 papers excluded due to not being in English. 'English language' database limiter used.
N = 1483
1317 papers excluded based on title and abstract or removed as duplicates.
N = 166
152 papers excluded based on full text review due to not:
(a) exploring the views or experiences of participants who have an adult-onset neurodegenerative disease in relation to EOLC, either through their actual experience or their views on its availability and/or quality or
(b) reporting findings of qualitative research using accepted qualitative methods of data collection and
Hand searching of reference lists = 0 papers identified
1 papers excluded based on critical appraisal
N = 13