



PALLIATIVE AND END-OF-LIFE CARE RESEARCH IN NEURODEGENERATIVE DISEASES

Report of the JPND Action Group

December 2014



JPND
research

EU Joint Programme – Neurodegenerative Disease Research

Executive Summary

The purpose of this report is to recommend options for JPND action in the area of palliative and end-of-life care research. The recommendations highlighted are the result of a consultation with leading European experts in the field, and are based on the analysis of the current status and urgent needs of palliative care research in the field of neurodegenerative diseases (ND). The actions conceived are aimed at facilitating the progress of pan-European research and the improvement of both the quantity and quality of palliative care research in the ND community. The potential actions to be fostered by JPND member countries include among others, transnational/trans-disciplinary networking, coordination of best practices and research funding.

The JPND Management Board (MB) established an Action Group in 2013 to determine the value of JPND actions in this area. Following a mapping consultation with MB members, and a direct consultation workshop with the research community on June 25th, 2014, this report from the Action Group identifies the gaps within current palliative care research on ND, scopes the requirements to be considered in areas of unmet need, and makes specific recommendations for JPND action. The authors believe that JPND should consider this report the first step of a medium-to-long-term, iterative process by which palliative and end-of life care research for ND can be highlighted, prioritised and implemented.

Current ND research landscape

- Palliative care, and the ability of transnational research to address needs and knowledge gaps in this area has not been crystallised for ND before now. In a field currently dominated by cancer care models, the core issues involved are similar to that of cancer, but are complicated by cognitive impairment, variable time periods and the range of people involved.
- Areas that have more recently developed include the growing evidence on palliative care in dementia and a framework for evaluating the practice of palliative care in dementia. There is a need to build on this work and invest in a cumulative, pan-European programme of research, allowing a wider approach to the palliative care of ND to develop.

Opportunities and needs

- There is a clear need for collaborative, pan-European research on the palliative care for people with ND, beyond dementia, and building on the limited research evidence in ND at the present time.
- There is an opportunity for JPND to take advantage of the different health care systems and cultural differences across Europe to gain insight into the best timing of palliative care in ND, and models in which the baseline palliative approach is combined with specialist neurological and dementia care.
- Capitalising on the work of JPND in the area of longitudinal cohorts for ND, there may be an opportunity to make use of the emerging methodological approaches to identify transitions/trigger points of deterioration that occur throughout the ND illness experience that result in a change of care (e.g. diagnosis, decisions to admit to long-term care).

- Assessment of the individual needs of patients, and in particular families and carers, is fundamental to further study the delivery of palliative care for people with ND. Due to the global impact of dementia, the particular needs of these patients and families should be integrated into research on ND patient needs.

Recommendations for Delivery

It is recommended that JPND considers supporting actions in two related domains: *quality improvement* and *research funding*:

Improvement of Quality

1. Support for organization of a transnational networking/engagement infrastructure

JPND should actively support the formation of a medium-to-long term, cohesive infrastructure designed to facilitate multi-professional engagement in palliative care research and enhanced dialogue across JPND member countries. The objective should be to facilitate networking, knowledge brokering and transfer, and training opportunities between current ND-relevant palliative and end-of-life care research approaches being undertaken in JPND member countries and at the EU level.

2. Co-ordination of best practices across EU member states

A number of key areas for best practice which are relevant across the ND spectrum would benefit from the establishment of a JPND “**rapid action**” **call for working groups**. A series of international-level working groups could be established in participating countries, under the JPND umbrella, aimed at providing national-level frameworks for the generation of evidence, and establishing where national barriers can be overcome, in the following areas:

- Advance Care Planning which involves carer and family engagement in planning and decision-making over time.
- The challenges of working with cognitive impairment in terms of communication, symptom management and end of life care.
- The effectiveness of education and training interventions and linked competencies in practice for a range of generalist and specialist practitioners charged with the care of ND patients.
- Engaging primary care providers (e.g. GPs, etc.) in palliative care planning.
- Engaging with national voluntary groups and policy makers on actions and outcomes for ND.

Research Funding

1. Call for collaborative research projects

A joint transnational call for collaborative projects may be launched in two major areas:

- Comparative research across EU member states on different health care systems referring to early timing and anticipating the end of life across ND and how to integrate specialist

knowledge while maintaining or enhancing the continuity of care.

- b. Research to enhance the capabilities of existing population and disease-based longitudinal cohort studies, or link related studies through a synergistic approach, to identify transitions/trigger points of deterioration that occur throughout the ND illness experience that result in a change of care (e.g. diagnosis, decisions to admit to long-term care).

For both transnational call topics:

- Research approaches should take into account the changing needs along the different ND trajectories, as well as the range of people and services involved in decision making and care delivery over the course of the illness.
- Pilot studies and add-ons to longitudinal cohort studies should be encouraged as efficient means to build research capacity in areas of unmet need, and may also raise awareness among experienced research communities who have not yet considered palliative care.

2. Support research into the identification of best practices for needs assessment

Assessment of needs is a fundamental aspect of palliative and end-of-life care research and JPND member countries should support multi-method, interdisciplinary research approaches in this area.

This support may include for example:

- linking up with research on indicators of quality of care or good care practices in dementia and other ND.
- observational studies of needs and preferences of patients and caregivers to provide the empirical evidence for intervention-based studies for individual ND.
- establishing monitoring infrastructures in Eastern and Southern European countries to promote the identification of needs in these countries/regions.

Table of Contents	Page
1. Introduction	4
2. Background and Terms of Reference	4
3. Methodology of work	5
4. Purpose of the report	6
5. State of Play and Current Landscape	7
6. Identifying Needs for Research	11
7. Barriers and Challenges to Implementation	15
8. Core Pillars for Implementation	17
9. Recommendations for Delivery	19
10. References	22
Annex I	Agenda of June 25 th expert-led workshop, Amsterdam airport
Annex II	List of participants at expert-led workshop, including completed questionnaires
Annex III	Collaborative projects supported under the 2012 JPND Transnational call: <i>“European research projects for the evaluation of health care policies, strategies and interventions for Neurodegenerative Diseases”</i>

1. Introduction

This report was prepared by the JPND Action Group on Palliative and End of Life Care, formed in March 2013 by the JPND Management Board. The aim of the Group was to develop JPND actions in line with a key priority within the JPND Strategic Research Agenda (SRA): *“the promotion of research into end-of-life and palliative care - this should include an assessment of the transferability of current hospice care models into social care situations, the ethical issues involved and the core criteria of effectiveness, cost and equity.”*

The JPND SRA Implementation Working Group on Health and Social Care (WG4) also recommended action in this area as a high priority *“due to the lack of knowledge in this field and growing incidence”* and determined: *“a need for more knowledge on the most effective ways of taking care of people with ND, especially in end-of-life care. Research can contribute to the development, evaluation and implementation of palliative and end-of-life-care that enhances the quality in the last phase of life”*.

2. Background and Terms of Reference

Prior to the establishment of the Action Group, the JPND Management Board considered whether this issue was best addressed by the actions of individual member states, rather than through joint

transnational actions within JPND. However, it was felt that an examination of the current status of palliative care research within the JPND member countries should be undertaken as a first step. As such the Action Group was established to:

- Establish and analyse the nature and scale of relevant palliative and end-of-life care research initiatives among JPND member countries and EU-funded initiatives.
- Consult with the research community to establish the value of JPND actions.
- Identify the gaps within current palliative care research on ND and scope the requirements to be considered in areas of unmet need.
- Provide a report and list of recommendations for actions to JPND Management Board.

In addition, four of the six collaborative projects supported under the 2012 JPND Transnational call: *“European research projects for the evaluation of health care policies, strategies and interventions for Neurodegenerative Diseases”* have a palliative/end-of-life care component (see Appendix III of this document). The three-year projects began in early 2014 with the first results expected in 2015. It has been proposed that these results could act as a starting point and/or the nucleus of future JPND initiatives in this area.

3. Methodology of Work

This report builds on the work performed by the JPND SRA Implementation Working Group 4 in 2012 and a number of workshops and consultations in the development of the JPND SRA. A survey of JPND member countries was conducted by the Action Group through the auspices of the Management Board (completed July 2013), seeking information on ongoing initiatives with relevant research focus on palliative care for ND. From a combination of analysis of the survey data, discussions with key researchers, JPND’s own transnational call experience, as well as a review of the [recently published white paper](#) from the EAPC reporting a Delphi study defining optimal palliative care in older people with dementia, it was determined that:

- There is value in JPND initiatives in this area as palliative care research can make substantial impact on ND care and can help to develop ND-focused palliative care research across Europe.
- There is a need to increase awareness and understanding of the importance palliative and end-of-life care research for ND across all member states and to share best practices.
- Growth in the capacity in palliative and end-of-life care research is required, both generically and particularly in ND-focused research, nationally and at regional and international levels.
- There is a need to evaluate different models of service delivery and interventions both nationally and internationally.
- Comparative assessments of approaches across borders would add value to national research, enhance practice development and accelerate the knowledge and understanding amongst member’s states.

In line with this determination, an expert-led workshop was organised by the Action Group in order to provide input into the development of an action plan for this area, in parallel with ongoing work to re-evaluate the JPND approach to Health and Social Care in general. The invite-only workshop took place on June 25th, 2014 in Amsterdam airport and the agenda is contained in Annex I of this document. The overall aim of the workshop was to share knowledge on JPND, to identify capacity-building and integrative research opportunities, and to recommend future JPND actions and

activities in this area. Appropriate chairs and rapporteurs for the workshop sessions were identified and approached. In addition, a questionnaire was prepared in advance of the workshop for those invitees unable to attend. Completed questionnaires were collated and distributed to chairs and rapporteurs prior to the workshop. The workshop minutes were generated by the rapporteurs and chairs and were then re-circulated to all attendees for their comments.



Figure 3.1 Participants at the expert-led workshop, Amsterdam, June 25th, 2014

The current report was generated by a group of workshop chairs, rapporteurs and selected attendees composed of:

- Gian Domenico Borasio, University of Lausanne, Switzerland
- Kevin Brazil, Queens University Belfast, United Kingdom
- Claire Goodman, Centre for Research in Primary and Community Care (CRIPACC), University of Hertfordshire, United Kingdom
- Philip Larkin, University College Dublin and Our Lady's Hospice and Care Services, Ireland
- David Oliver, University of Kent, United Kingdom
- Jenny van der Steen, VU University Medical Center (VUmc), the Netherlands

JPND Representatives:

- Mogens Horder, JPND Executive and Management Board member, Denmark
- Derick Mitchell, JPND Senior Communications Manager
- Enda Connolly, Ireland (since retired as JPND Management Board member)

4. Purpose of the report

The main purpose of this work was to discuss the available evidence and gaps in knowledge in palliative care research in the field of ND, thereby suggesting actions within the reach of the JPND community. These actions may include shared working across JPND member countries, funding of competitive calls, coordination of best practices and organization of transnational initiatives designed to share knowledge and provide training that does not currently exist in this research field.

5. State of Play and Current Landscape

Key points of this chapter

Palliative care in ND is distinct from palliative care in other diseases

- Due to the patient's cognitive decline necessitating substantial family involvement, the protracted trajectory and the difficulty of prognostication in ND

Palliative care in ND also differs from "usual care" as of today in most countries and settings

- It anticipates the end of life ahead of the development of symptoms, problems and needs
- It addresses spiritual needs in addition to physical and psychosocial needs
- It addresses the highly individual needs of patient and families with a multidisciplinary team

Currently, palliative care in ND may be most developed for dementia and ALS/MND

- However, evidence is mostly from individual, nationally-driven studies in the USA and Northern Europe

Important challenges remain

- For example, how to shape palliative care through the stages of the disease, the place of care, and the key transitions more generally
- A key challenge faced by all countries is the appropriate timing and integration of palliative care expertise and expertise in psychiatric, geriatric or dementia care, and of social and medical care to optimize care delivery

The EAN/EFNS and EAPC have recently issued key recommendations to improve care, and for dementia

- The EAPC has also provided a framework for defining palliative care in terms of domains and a research agenda

What is Palliative Care for Neurodegenerative Diseases?

"Palliative care is an approach that improves the quality of life of patients and their families facing problems associated with life-threatening illness, through the prevention and relief of suffering, early identification and impeccable assessment and treatment of pain and other problems, physical, psychosocial and spiritual". (World Health Organization, 2002)

Despite the current agreement on the WHO definition, palliative care is sometimes mistaken for terminal care or end-of-life care. Palliative care may apply to any stage of the disease, and can be provided in conjunction with treatment aimed at delaying disease progression. A number of other terms are being used, such as *hospice care* which refers to a health care system which may provide specialist palliative care, or *supportive care* which refers to specific types of non-specialist palliative care. Patients with cancer and other progressive diseases may benefit from early involvement of palliative care specialists and there is also evidence for its cost-effectiveness (Gomes *et al.*, 2013. Temel *et al.*, 2010).

A particular challenge for palliative care for ND is that whilst these are terminal conditions the trajectory to death is often protracted. Consequently, palliative care interventions that support quality of life and care closely align (or blur) with many of the broader treatment and care goals for people with ND.

It is commonly acknowledged that cancer care models currently dominate and whilst there has been transferable learning from cancer to ND (for example, symptom management for people with amyotrophic lateral sclerosis (ALS) there is a clear need to develop an evidence base that reflects the particular experience of living and dying with ND.

There is also increasing interest in the provision of palliative care for people with ND. This has traditionally been for people with ALS and there is a long involvement with ALS patients and families (Oliver *et al* 2014). Indeed, guidelines on the care of ALS have included palliative care and end-of-life care issues (Miller *et al* 2009; Andersen *et al* 2012).

Several issues, especially relevant to ND, need to be taken into consideration for assessment and management of symptoms during palliative and end-of-life care including cognitive impairment; fluctuating episodes of ill health over an often protracted period of deterioration; lack of speech and other communication skills; increased dependency on carers and families; genetic risk factors. Many ND also have variable progression (ALS prognosis is usually 2-3 years but dementia can be 10+ years), making prognostication difficult for any individual patient. It is suggested that more appropriate care could be provided by considering the idea of ‘transitions’ which occur throughout the illness experience, rather than looking for triggers/points of deterioration to better identify the end-of-life phase. Indeed, there is a distinct lack of evidence regarding the transition experience of many patients with a variety of ND. Elements of palliative care such as advance care planning and symptom management should be provided earlier in the course of the disease with patient and family, especially as this allows fuller discussion ahead of cognitive impairment.

Developments in palliative care in ND

There have been longstanding discussions about the role of palliative care in ND (considering ND such as ALS, multiple sclerosis, Parkinson’s disease, Huntington’s disease, progressive supranuclear palsy and multiple systems atrophy) and the need for neurologists to respond to patients and families, particularly at the end of life (American Academy of Neurology, 1996). This has recently been reinforced by a consensus document looking at ND from the European Association for Palliative Care (EAPC) and the European Academy of Neurology (EAN) / European Federation of Neurological Sciences (EFNS) (Oliver *et al in press*). This consensus covers many core palliative care principles while reflecting the particular experience of living with a long term neurological condition.

Table 5.1 Suggested principles of palliative care for ND (Oliver et al in press 2014)

<ul style="list-style-type: none"> • Palliative care should be considered early in the disease trajectory
<ul style="list-style-type: none"> • The assessment and care should be provided by a multidisciplinary team approach, with access to specialist palliative care
<ul style="list-style-type: none"> • Communication should be open with patients and families and advance care planning is recommended. This needs to be culturally appropriate and may vary across countries within Europe and should be in all settings
<ul style="list-style-type: none"> • Symptoms – physical, psychosocial and spiritual should be managed actively and appropriately
<ul style="list-style-type: none"> • Care needs should be assessed and carers supported before and during the process of dying, and after death. Professional carers should receive education, support and supervision to reduce the risks of emotional exhaustion

- **There should be repeated and continued discussion about end-of-life issues and review of patients' wishes and aims – allowing appropriate management and intervention. This will need to be culturally appropriate and take into account the legal arrangements in each country**
- **Palliative care principles should be included with the training and continuing medical education of neurologists and palliative care professionals should understand the issues for patients with ND**

These recommendations are being considered within both EAPC and the EAN / EFNS and a common curriculum for both palliative care physicians and neurologists based on the consensus is being developed. These principles would be applicable to all ND, including dementia.

Palliative Dementia Care

Dementia is an important global health challenge, and although palliative care in dementia and ALS may be more developed than palliative care in other ND, important challenges remain. Palliative care in dementia was introduced in the US in the 1980s (Volicer et al., 1986). However, until recently it was unclear what palliative care in dementia exactly entails, how it differs from palliative care for patients with cancer, and from usual dementia care. The EAPC white paper provides a framework that defines palliative care in dementia in terms of domains (Table 5.2 of this document), and defines optimal palliative care in terms of recommendations backed up with evidence (van der Steen et al., 2014). The work indicated that palliative care in dementia is distinct. For example, continuity of care / family support and involvement as exemplified by advance care planning is more important than in palliative care for patients with cancer, and anticipating the end of life is also different from usual dementia care.

At a policy level, dementia and palliative care is often not integrated (a paper currently under review examines the content of national dementia strategies in relation to the EAPC white paper). Most national dementia strategies do not refer to palliative or end-of-life care. Although a number of domains of palliative care in dementia are being covered to some extent, prognostication and timely recognition of dying is lacking in all strategies. Also in practice, integration is frequently lacking. For example, palliative care guidelines in Europe often lack guidance on assessment and treatment of pain in dementia, which may differ substantially from assessment and treatment in other patients, while specialists in palliative care may not have experience with dementia patients (Sampson et al., 2013). In practice, palliative care and dementia care are often separate worlds. This is illustrated by the example of the Netherlands, where elderly care physicians provide the care at the end of life of patients with dementia. Until recently, most had not had any formal training in palliative care in dementia, while in practice, specialist palliative care is not being accessed routinely for dementia patients. Ongoing support from geriatricians supports continuity of care, but there is a need for different models of palliative care such as palliative consultation that complements and does not compromise continuity of care. Conversely, in the UK, research on palliative care for people with dementia is complicated by the fact that people with dementia are as reliant on social care as health care and that access to medical care and to specialist palliative care is variable. The challenge of integrating health and social care for people with ND are well documented (Davies et al 2011; Froggat & Goodman, 2014). However, integration can be facilitated through pan-European (EU-FP7)

research projects and also through networking groups where expertise is brought together, an example being the [EU COST action on pain in people with impaired cognition](#), which has a working group on palliative care.

Palliative Care Research for ND?

Compared to cancer, the research agenda for palliative and end-of-life care has been more limited for patients with ND, although the amount and quality of research on the palliative care of people with ND is increasing.

Early studies of palliative care in dementia were from the US and the UK, and these pointed to a large symptom burden in dementia patients at the end of life. Since 2000, research in palliative care in dementia has developed in North-Western Europe, with recent studies in other European countries (van der Steen, 2010; van der Steen et al., 2014). While early studies were mostly small descriptive or retrospective studies, more recent work employs a variety of study designs.

The effectiveness of palliative care in ND has been shown in some limited clinical trials (Higginson *et al* 2009; Veronese and Oliver 2013) with further pan-European studies planned to further develop this evidence (for example the [OPTCARE Neuro study](#), funded in the UK by the NIHR and starting in 2015). There is also a lack of evidence reflecting the best way to manage the involvement of patients and families, with limited evidence for how multi-disciplinary working can support effective palliative care (Traynor et al 2003). Various guidelines (e.g. Andersen et al 2012, Miller et al 2009) have found in reviewing the literature that there is little clear evidence, particularly from Randomised Controlled Trials (RCTs). However the development of European consortia to develop research in ND may help in the development of a stronger research capacity in this area.

It is a limitation that most evidence on palliative care in dementia refers to long-term care settings, where infrastructures are available to facilitate research. The majority of people with dementia live at home but as the disease progresses there is often the need to consider residential care. In the future more people will probably be staying at home as long as possible. This does not negate the fact that there will be for some the need to transition to other care settings, such as residential care and there will be increased risk of hospital referral in the case of an acute event. Therefore research in home care, residential care and hospital settings is urgently needed and recent European research in these settings is available from the UK (Amador et al., 2013) and Italy (Toscani et al., 2013; Sampson et al., 2011). However, most studies are undertaken in a single country or setting, which limits our understanding and it is often unclear if the findings are transferrable to other countries or settings.

The EAPC recently developed a White Paper on palliative care in dementia, based on evidence and consensus through a Delphi study with input from experts of 23 countries (van der Steen, EAPC et al., 2014). It defined palliative care in terms of 11 domains, and optimal palliative care in terms of 57 recommendations, backed up by literature. The experts involved in the Delphi study also prioritized the 11 domains for research. Unresolved issues include when in the course of the disease palliative care should be applied. Further, there is agreement about a baseline palliative approach with care provided by the usual, primary caregivers, and specialist palliative care needed for complex cases. However, the optimal model of care, in which also dementia care specialists such as psychiatrists collaborate, still needs to be given shape in the practice of most European countries.

Table 5.2 The eleven domains of palliative care in dementia in the EAPC white paper on palliative care in dementia.

• Applicability of palliative care
• Person-centred care, communication, and shared decision making
• Setting care goals and advance planning
• Continuity of care
• Prognostication and timely recognition of dying
• Avoiding overly aggressive, burdensome, or futile treatment
• Optimal treatment of symptoms and providing comfort
• Psychosocial and spiritual support
• Family care and involvement
• Education of the health care team
• Societal and ethical issues

6. Identifying needs for research

Key points of this chapter

Needs assessment is fundamental to further study the delivery of palliative care for people with ND

- To date the focus has been on particular patient groups, or people with carers. Needs assessment should also include people that live alone, people from Black Minority and Ethnic groups and people that are socially deprived

Identify transitions in health and carers' capacity to provide support along the disease trajectory

- Longitudinal studies that include the identification of transitions may help in identifying changing needs along different disease trajectories of subgroups of patients with different ND.
- Such studies may start early in the disease, as problems may thus be signaled and preventive factors identified before the end-of-life phase.

The identification of needs should include member states in the East and South of Europe

- It may be particularly helpful to set up studies or a monitoring infrastructure in these countries, which may benefit from experiences in other parts of Europe

Different health care models should be studied for effectiveness in addressing needs in different countries and health care systems

- A particularly promising and pragmatic model involves generalists providing palliative care who are educated and supported by specialists

JPND considers palliative care as part of Health and Social Care

- This should include – but not be limited to – treating symptoms by skilled professionals

Needs assessment should be followed by a mapping of the known enablers and inhibitors to care

- For example, person-centred approaches developed for dementia care, stigma around death and dying) in particular the end of life needs of those with ND, should be considered.

Overview

In palliative care and related approaches for people with ND, such as person-centred care (Kitwood 1993; Chenoweth et al., 2009; Røsvik et al., 2013), identifying needs is fundamental and provides input to any further action. This may refer to clinical practice as well as to research, and setting research priorities.

It is important to focus on the needs of different patient groups living and dying with ND and how experts believe needs can be identified through research. Identified needs may subsequently be addressed through further research on appropriate palliative care interventions, policy or implementation of best practices.

General and individual needs

Some needs are common to virtually all care dependent or end-of-life populations, such as the need to relieve burdensome symptoms and a need for being treated with respect and dignity (van der Steen et al., 2011). This refers to particular physical, social, psychological, and existential/spiritual needs at the end of life. Assessing of needs can be difficult in populations such as patients with severe dementia at the end of life (Perrar et al., 2014). Nevertheless, a few studies have identified such needs from the patients and caregivers. They also had needs that varied and preferences were more individual, such as spiritual and environmental needs, and need for support with daily routines. The universal needs, such as being free from pain as far as possible, are important but for many people the careful identification and management of these individual needs may be more important in maintaining their own quality of life.

Suggested sources for identifying needs

- Involve patients (if still able), and their families in the identification of palliative care needs. However, terminology is important for example; *need for support, preferences at the end of life or for care aimed at comfort, or attributes of a good death* may be better understood than the phrase “palliative care needs”. Patient advocacy groups may also help in the identifying of needs, and may allow discussion of these needs earlier in the disease progression.
- Care providers and staff should provide input in the identifying of the multiple barriers to address the needs, to successfully improve actual practice. For example, palliative care teams may be focused mostly towards care for patients with cancer. The care for patients with ND may be perceived as more protracted and less rewarding for staff. Specialists in providing care to people with ND conditions, on the other hand, may resist focusing on the end of life. The challenge is to achieve continuity of care and integration in practice, to ensure practice is not reduced to a simple re-labelling of care.
- People with ND conditions spend the majority of their life at home and are cared for by primary and community care services. There is often the need for support at the point of transition to other settings - hospital, nursing homes (care homes) or specialist settings – and research is needed in all these areas.
- Assessment of needs at the individual level and level of care organizations (micro- and meso-levels) may be supplemented with a needs assessment at the macro-level (for example, public policy on the management and support of people with long term conditions).

Suggested populations for identifying needs

- Identify needs applicable across different patient groups as well as those specific to particular ND at different stages.
- Patients with dementia require particular attention because of the global impact of the disease. Additionally, research should address needs of the many frail patients who have an ND component to their illness, but a clinical picture that is neither typical nor mono causal.
- The identification of needs should include member states in the East and South of Europe. There is a gap in knowledge in this respect because current reviews are biased through over-inclusion of, for example, studies on highly educated whites who have a family caregiver and reside in Northern or Western European countries or the US (Bunn et al., 2012). In other EU member states, education of families and professionals may be even more important. For example, Italian families were more likely to prefer a family booklet informing them on palliative care in dementia early in the course of the disease than Dutch families (van der Steen et al., 2012). Further, attitudes and perceptions of professional caregivers may differ. For example, physicians and nurses in the Czech Republic, or in Southern European countries such as Italy have difficulties explaining that tube feeding is not beneficial in late-stage dementia (Valentini et al., 2014).

Topics related to health problems

- Acknowledging that a major part of the baseline palliative approach is shaped by psychosocial, existential/spiritual and decision-making aspects, there is still a need to address medical needs such as treating pain, complex neurological symptoms, and behavioural problems in the context of multi-morbidity and frailty.
- Transitions or tipping points are important. The course of progression differs for different ND. For example, dementias are usually chronic-progressive, whereas Creutzfeldt–Jakob disease is acutely progressive and Parkinson disease is characterized by chronic disability with fluctuations (Borasio, 2013). Some diseases such as Huntington’s disease may be close to cancer in terms of trajectory, while dementia and other types are different (longer and very variable, less predictable trajectories). Research around transitions should take such differences into account.

Topics related to Health services provision

- Specialist palliative care does not have the capacity nor the mission to address all patients’ needs at the end of life. Research is required into models of care that emphasise outreach or facilitated support and education from specialists. This may also include the development of tools to evaluate if specialist care is needed, and train first line practitioners such as the GP in its use for appropriate referral.
- With regard to models of care, organizational issues must be considered:
 - Workforce issues - are there sufficient doctors to care for people who increasingly choose to die in community settings? (Sweden);
 - Service delivery models - integrated dementia care in community settings employs regional teams. Palliative care practitioners should become part of such teams (the Netherlands);

- Location of care - the US hospice model is more outpatient oriented than hospice models in Europe, and increasingly includes diseases other than cancer. US experiences may inform developing care models in Europe;
- The comparative efficacy of different palliative care models for people with dementia should be explored – for example, facilitated support for generalist staff, versus medically led care or case management informed approaches for people with complex needs;
- Costings: cross national comparisons may map different health and social care funding for end-of-life care for people with ND.

Identifying best methods and approaches

- Rather than start from scratch, the existing research and knowledge in the area of needs assessment should be built upon. For example, linking up with work on indicators of quality of care or good practice in dementia care or other ND.
- Existing tools (e.g., family booklet – Arcand et al 2013) may be studied for utility and in particular, to address needs in Eastern and Southern European countries.
- To this end, simple registration of characteristics and needs of people at the end of life may be helpful for national monitoring particularly in countries with little research. The experiences in Sweden may be used (www.palliativ.se); The Swedish national quality registry for palliative care, documents the quality of care during the last week of a person's life, including people dying with ND. However, costs and benefits of the registry are unclear for example, when the presence of ND is not the reason for needing palliative care or access to the register is restricted. Underreporting of underlying ND disease as a cause of death is common, and specific consideration is needed for registration of cause of death (not just immediate cause) and which type of diseases should and should not be registered (including extremely rare diseases may not be very helpful).
- As needs may vary along the disease trajectory, longitudinal studies that identify transitions or trigger points to change care, such as diagnosis, and decisions to admit to long-term care are needed for better understanding. Collaborations may be created with teams leading existing cohort studies - such as the [Cognitive Function and Ageing study \(CFAS\)](#) and the English [Longitudinal Study of Aging \(ELSA\)](#). There is a particular need for research approaches that reflect the disease trajectory, the presence of co-morbidities and the range of people and services that are involved in decision making and care delivery over time.
- Observational studies of needs and preferences of patients and their caregivers can provide the foundation and empirical evidence and for more intervention-based studies. There is a lack of information in particular about needs in Parkinson's disease and Huntington's disease.
- Collaboration between the primary health care and universities may facilitate implementation of research findings.
- The research approach should be multi-method and may include further Delphi studies with experts to identify needs in ND; focus group studies with patients, families and staff; survey of problems encountered at the end of life by families and staff; systematic reviews; realist synthesis scoping exercises etc.

7. Barriers and Challenges to Implementation

Key points of this chapter

Social and political factors

- Stigmatization and cultural perception
- Political and professional priorities given to curative disease
- Day to day care management for people with ND is often provided by health and social care workers, which requires appropriate supervision and direction.

Research and methodological factors

- Lack of research funding and limited opportunity for multidisciplinary social science inquiry
- Lack of evidence-based, multicomponent complex interventions

Palliative care-related factors

- Limited opportunity for cross-sector collaboration
- Uncertainty regarding the appropriate involvement of palliative care
- Lack of academic expertise in the research field

Overview

Barriers to the implementation of research in the area of ND and palliative and end-of-life care fall into three distinct categories; social and political factors, research / methodological factors and palliative care-related factors. A key challenge to implementation is a lack of opportunity for integration and cross-working between neurological/palliative care/health and social care services which inhibits research development. The fact that the term 'neurodegenerative disease' reflects a range of life-limiting illness with differing trajectories towards end-of-life indicates complexity in developing a coherent and comprehensive research agenda. On the other hand, this variability, when discussed and acknowledged, can lead to shared learning, strengthen partnership and lead to greater insights into new and complex areas.

Despite some evidence of good models of integration, limited empirical data reflecting palliative care and ND, especially in the area of social sciences, contributes to the limitations and challenges perceived at this time. In this chapter, we offer a brief description of the core issues based on key factors noted above.

Stigmatization and cultural perception. At one level, there is concern amongst both the public and professionals regarding the term 'palliative care', especially when introduced early into the disease trajectory. Alternatively, across some EU member states, there is an insufficient understanding of palliative care, which means that attitudes may be neutral at best. Discussion around end-of-life issues may be culturally prohibitive. The language used to describe the current practice of palliative care in the wider healthcare context can be confusing (supportive, continuing, advanced, complex etc.) and leads to euphemism which fails to denote key transition points along a patient journey towards palliative care involvement. There is a view that ND, especially dementia, is stigmatized in society. With research funders, assumptions may be made that patient-focused research in this area is particularly difficult to carry out. From the perspective of those working in palliative care, there may be a perception that they lack skills and expertise in the area of neurology to contribute to a

shared research agenda. This is a workforce, particularly in social care settings, that often does not receive investment in continuing professional development and whose work is not always recognised as skilled. The management of patients with ND across EU member states is widely variable which makes it problematic to provide a coherent research agenda. However, as many countries are struggling with the same issues, a pan-European approach to research can build capacities and result in greater impact in this area.

Political and professional priorities given to curative disease. When compared to curative disease, the limited availability of research funding for chronic and life-limiting disease in general is indicative of the priorities set by both policy makers and professionals in determining the drive for research. In some EU member states, focus on seeking cure (for example, early-onset dementia) can limit the opportunity to seek funding across the spectrum of disease. It is also suggested that there is reluctance on the part of administrators in championing ND as a focus for research either because of a lack of evidence-based guidelines to support clinical practice (and the need for further research) or the ability to attract stronger funding streams for curative illness.

Lack of research funding, and limited opportunity for multidisciplinary social science inquiry. The complex nature of ND warrants a range of research approaches that capture the evidence as broadly as possible. There is a lack of research funding in this area, particularly in relation to social sciences and research that involved the multidisciplinary team, essential to the care and management of patients with ND. There is a significant gap in research when compared to cancer or other chronic illness where palliative care involvement has been shown to have benefit. There is a growing body of evidence in relation to palliative care in organ failure and dementia but far less in wider neurological conditions such as Cerebro-Vascular Accident, or Parkinson's disease. Nor is it understood when ND such as dementia or Parkinson's complicates the palliative care of patients with other life limiting conditions. It is neither possible nor appropriate to extrapolate findings from other groups in receipt of palliative care (such as cancer) and apply these directly to ND. There is therefore a need for greater creativity in the development of research studies in this field, other than traditional RCTs.

Limited opportunity for collaboration. There are currently limited opportunities to collaborate across disciplines between palliative care and neurology colleagues and across EU member states. To some extent, this is based on a lack of awareness on the part of neurology professionals about the potential benefit of palliative care applied early in disease trajectory, or a misplaced assumption that palliative care is only applicable in the terminal stages of life. This is in spite of a growing body of evidence about the benefit of palliative care to chronic and life-limiting illness beyond cancer. There is an ongoing and often convoluted specialist-generalist debate in terms of the delivery of services that adds to the uncertainty of when to involve palliative care and prevents collaboration and joint research endeavour. It is also acknowledged that academic leadership and expertise in the research field of palliative/neurological disease is weak. Further university professorial chairs and other senior academic posts to lead programmatic research in this area are needed (see below¹). Greater collaboration across member states should be encouraged.

The challenge of involving patients and families in research relative to palliative care is acknowledged, usually due to symptom burden or frailty leading to high levels of attrition. However,

¹ As a good example, an academic chair in geriatric palliative care was recently advertised by the University of Lausanne, Switzerland. The position has the aim of bridging the gap between geriatrics and palliative care, especially in the research field, in close cooperation with the local health care providers (GPs, nursing homes etc.). The chair is expected to start its work in autumn 2015

there is evidence that patients and families would choose to participate in research if asked, albeit for altruistic reasons rather than for personal benefit. One challenge noted in ND is that the support services available to patients and families across the EU are widely variable and often markedly limited. Although patients and families may well value the benefit of research, the need to expend energy in managing and sourcing the best care packages may inhibit their taking part in research studies, irrespective of the potential benefit it may bring for themselves or others.

Uncertainty regarding appropriate involvement of palliative care. The fact that evidence shows that early intervention of palliative care can lead to better patient and organisational outcomes (Gomes et al., 2013; Veronese and Oliver, 2013) only serves to highlight the weaknesses in referral to palliative care services. A key challenge which transcends the palliative-neurological interface are issues of prognostication and diagnosing dying. Time is often spent looking for ‘triggers’ (points of deterioration) which indicate that a palliative care referral may be warranted. However, these are often inaccurate, especially in the context of ND and prognostic tools used in palliative care for cancer patients are not transferable. There is a benefit to consider patient experience in terms of process rather than just the end stage of disease. It is suggested that a better explanation would be to consider the idea of transitions which occur through the illness experience. There is a distinct lack of evidence regarding the transition experience of many patients with a variety of ND.

Importantly, despite the evident barriers to implementation of palliative and end-of-life care research in ND, there are some good examples that demonstrate the possibility of joint working, collaboration and capacity-building. The UK-based ‘[Neighbourhoods and Dementia](#)’ study one of [six that were funded](#) from a £20m joint funding call on dementia by health (NIHR) and Social Science (ESRC) funders will add significantly to the knowledge base regarding dementia care through working alongside patients and families. There is clearly a need for a more cohesive infrastructure to facilitate multi-professional engagement in research and enhances a dialogue across EU states to understand how national barriers can be overcome. There is also an important question as to where responsibility for leadership in this research field lies; palliative care or neurology or geriatrics or psychiatry. Partnership is key to greater and more cohesive implementation.

8. Core Pillars for Implementation

Key points of this chapter
<ul style="list-style-type: none"> • Attract trainees to the research area
<ul style="list-style-type: none"> • Develop effective partnerships with user communities, including decision-makers and patient/family carers.
<ul style="list-style-type: none"> • Promote a better understanding on the experience of dying and what palliative care can offer in the public and policy decision-making communities
<ul style="list-style-type: none"> • Support infrastructure development for palliative care research
<ul style="list-style-type: none"> • Enhance interdisciplinary research collaboration
<ul style="list-style-type: none"> • Encourage the development of career researchers

- Employ effective knowledge transfer strategies that integrates research results into practice guidelines, health professional training, service development, and policy discussions
- Invest in research funding that will support a suite of research funding ranging from pilot studies, operating grants and program grants
- Develop outcome measures and an evaluation plan

Overview

The primary objective is to build research capacity that will facilitate improvement in both the quantity and quality of palliative care research in the ND community. In this chapter, we propose that to achieve this objective a planned, multipronged, integrated initiative is required that addresses the particular experience of dying with a ND. Reasonable timelines for implementation must be identified to enable movement from a small, fragmented research community to a sustainable world class research enterprise. This comprehensive strategy includes the following:

Promote a better understanding on the experience of dying. Palliative care research has received limited attention in the ND community. Leadership is required to inform the public, provider and policy community on the modern experience of dying and the significance of research to examine options available to patients and families and the obligations held by the service, policy community.

Support infrastructure development for palliative care research network. Various formal and informal meetings either as stand alone or in conjunction with other events coupled with internet based technology can facilitate Pan-European networking among a dispersed research community. These activities can enable the research community to collectively take action to develop a long-term research strategy to secure its future.

Enhance interdisciplinary research collaboration. The knowledge base for effective reliable care at the end of life is broad. This knowledge base requires, as examples, symptom-based investigations, epidemiology on dying and death, understanding the role of culture, examining issues in communication and decision-making, and considerations on the social, behavioural and service dimensions of palliative care and what supports the integration of health and social care provision in the ND community. Consequently research questions are complex and require interdisciplinary approaches. A richness of research ideas and a wide range approach to problem solving is seen as an aspect to an interdisciplinary environment. Strategies to establish inter-disciplinarity in the research field are thus required.

Encourage the development of career researchers. Researchers must be equipped and supported to be adaptable and flexible in an increasingly competitive, global research environment. The importance of researchers' career development, and lifelong learning, should be clearly recognised and promoted. A wide range of training, career transition, and development opportunities relevant to palliative care research in the ND community should be identified and made available.

Attract trainees to the research area. A sustainable research enterprise requires recruitment and training of new career researchers. Training opportunities offering mentorship and a wide range of contacts and experiences can facilitate the trainee or new investigator into the competitive edge of job-seeking and securing competitive grants and personal awards.

Develop strong effective partnerships with user communities, including decision-makers and patient/ family carers. Successful initiatives need to plan for the long term and this includes community partnership building and infrastructures that support patient and public involvement. Cultivating partnerships facilitates trust relationships with potential knowledge users ensuring a responsive and responsible research agenda where ‘research makes a difference’.

Employ effective knowledge transfer (KT) strategies that integrates research results into practice guidelines, health professional training, and service development, policy discussions. Multiple and diverse KT strategies must be acknowledged as requirements for ‘research to make a difference’. KT strategies must be incorporated both within public research funding organisations as well as within individual investigator-lead research. An evidence-informed approach on effective KT strategies is required which shapes the framework for local implementation.

Invest in research funding that will support a suite of research activities ranging from pilot studies, operating grants and programme grants. This report is submitted against a background of increasing worldwide research interest and publications in palliative care. Research funding will have a steering effect on established and new researchers to engage in issues of importance to the ND community. A range of funding opportunities enhances the impact of the initiative. Agencies and funding organisations participating in JPND should coordinate their efforts, and partner with external bodies such as foundations, charities and patient organisations to develop a suite of research funding opportunities.

Develop outcome measures and an evaluation plan. An evaluation plan must be developed on the onset to allow assessment on the successful transfer of knowledge. It would be wise to note that the academic definition of excellence can be incompatible with effective knowledge transfer activities. Evaluation outcomes should include both traditional academic outputs as well as assess for public, practice and policy impacts.

9. Recommendations for delivery

The key priority for furthering progress at the JPND/pan-European level should be to support and facilitate research that informs how to give shape to palliative care across the spectrum of ND. It is therefore recommended that JPND considers supporting actions in two related domains: *quality improvement* and *research funding*. In all cases below implementation should take account of ongoing activity in related European and member-state-level initiatives.

In terms of their relative priority, or sequence of implementation, this may be the outcome of an ongoing deliberative process between JPND and relevant stakeholders (patient/ carers, sponsors, decision-makers and researchers) in this area. Indeed, the authors believe that JPND should consider this report the beginning of a medium-to-long-term, iterative process by which palliative and end-of life care research for ND can be highlighted, prioritised and implemented. The authors also recognise the importance of return on investment in the current environment, and that mechanisms for accountability and impact should be incorporated into the recommendations below.

Improvement of Quality

1. Co-ordination of best practices across EU member states

A number of key areas for best practice which are relevant across the ND spectrum would benefit from the establishment of a JPND “**rapid action**” **call for working groups**². A series of international-level working groups could be established in participating countries, under the JPND umbrella, aimed at providing national-level frameworks for the generation of evidence, and establishing where national barriers can be overcome, in the following areas:

- a. Advance Care Planning which involves carer and family engagement in planning and decision-making over time.
- b. The challenges of working with cognitive impairment in terms of communication, symptom management and end of life care.
- c. The effectiveness of education + training interventions and linked competencies in practice for a range of generalist and specialist practitioners charged with the care of ND patients.
- d. Engaging primary care providers (e.g. GPs, etc.) in palliative care planning.
- e. Engaging with national voluntary groups and policy makers on actions and outcomes for ND

Each Working Group should provide a programme of work, with defined outcomes over a limited time period relative to the work and demonstrate multidisciplinary engagement in both process and outcome. It is suggested that the working groups are supported through the provision of national funding to PIs within that country who will act as coordinators for the selected national working groups. Each participating country supports the delivery of one working group, with JPND providing oversight and assistance in the promotion and dissemination of working group outputs to all JPND countries and globally.

2. Support for organization of a transnational networking/engagement infrastructure

JPND should actively support the formation of a medium-to-long term, cohesive infrastructure designed to facilitate multi-professional engagement in palliative care research and enhanced dialogue across JPND member countries. The objective should be to facilitate networking, knowledge brokering and transfer, as well as training opportunities between current ND-relevant palliative and end-of-life care research approaches being undertaken in JPND member countries and at the EU level.

- The infrastructure should aim to encourage and facilitate greater collaboration, innovation and integration between scientific, healthcare and social care researchers from different communities and countries. Such an infrastructure will create opportunities to collaborate across disciplines (e.g. between palliative care and neurology colleagues) and across national borders.
- Efforts should be made to promote interaction and collaboration between JPND member countries with existing experience and practice in the field and those countries (inclusive of Eastern and Central European member states) who would benefit from a strategic development partnership.

² Based on the successful implementation of the [2014 JPND “Rapid Action” Call for working groups in longitudinal cohorts for ND](#). This call was launched in April 2014 with working groups beginning in Autumn 2014 for a 6-month duration, thus aiming to produce their outputs in Spring 2015.

- A key element of the infrastructure should be to promote the recruitment and training of new researchers in this area. A JPND award specific to end-of-life care may be a useful mechanism in this regard. For example, an award to support expert training opportunities for PhD students in different disciplines across Europe (lectures on methodologies, research grant writing, presentation of own work, opportunities for networking).

Research Funding

1. Call for collaborative research projects

A joint transnational call for collaborative projects may be launched in two major areas:

- a. Comparative research across EU member states on different health care systems referring to early timing and anticipating the end of life across neurodegenerative diseases and how to integrate specialist knowledge while maintaining or enhancing the continuity of care.
- b. Research to enhance the capabilities of existing population and disease-based longitudinal cohort studies, or link related studies through a synergistic approach, to identify transitions/trigger points of deterioration that occur throughout the ND illness experience that result in a change of care (e.g. diagnosis, decisions to admit to long-term care).

For both transnational call topics:

- Research approaches should take into account the changing needs along the different ND trajectories, as well as the range of people and services involved in decision making and care delivery over the course of the illness.
- Pilot studies and add-ons to longitudinal cohort studies should be encouraged as efficient means to build research capacity in areas of unmet need, and may also raise awareness among experienced research communities who have not yet considered palliative care.

2. Support research into the identification of best practices for needs assessment

Assessment of needs is a fundamental aspect of palliative and end-of-life care research and JPND member countries should support multi-method, interdisciplinary research approaches in this area. This support may include for example:

- linking up with research on indicators of quality of care or good care practices in dementia and other ND.
- observational studies of needs and preferences of patients and caregivers to provide the empirical evidence for intervention-based studies for individual ND.
- establishing monitoring infrastructures in Eastern and Southern European countries to promote the identification of needs in these countries/regions.

Research approaches in this area should be multi-method, interdisciplinary and may include Delphi studies with experts; Focus group studies with patients, families and staff; Surveys of problems encountered at the end of life by families and staff; Systematic reviews; Realist synthesis scoping exercises etc. A vital component of these initiatives should be the development of evaluation frameworks to assess their impact.

10. References

1. Amador, S., Goodman, C., King, D., Ng, Y. T., Elmore, N., Mathie, E., Machen, I. & Knapp, M. (2013). Exploring resource use and associated costs in end-of-life care for older people with dementia in residential care homes. *International journal of geriatric psychiatry*. DOI: 10.1002/gps.4061
2. American Academy of Neurology. Palliative care in neurology. The American Academy of Neurology Ethics and Humanities Subcommittee. *Neurology*, 1996. 46(3): p. 870-2.
3. Andersen PM, Abrahams S, Borasio GD, de Carvalho M, Chio A, Van Damme P, Hardiman O, Kollwe K, Morrison KE, Petri S, Pradat PF, Silani V, Tomik B, Wasner M, Weber M. The EFNS Task Force on Diagnosis and Management of Amyotrophic Lateral Sclerosis. EFNS guidelines on the Clinical Management of Amyotrophic Lateral Sclerosis (MALS) - revised report of an EFNS task force. *Eur J Neurol* 2012; 19: 360-75
4. Arcand M, Brazil K, Nakanishi M, Nakashima T, Alix M, Desson JF, Morello R, Belzile L, Beaulieu M, Hertogh CM, Toscani F, van der Steen JT. Educating families about end-of-life care in advanced dementia: acceptability of a Canadian family booklet to nurses from Canada, France, and Japan. *Int J Palliat Nurs*. 2013 Feb;19(2):67-74.
5. Borasio GD (2013). The role of palliative care in patients with neurological diseases. *Nat Rev Neurol*;9(5):292-5.
6. Bunn F, Goodman C, Sworn K, Rait G, Brayne C, Robinson L, McNeilly E, Iliffe S. Psychosocial factors that shape patient and carer experiences of dementia diagnosis and treatment: a systematic review of qualitative studies. *PLoS Med* 2012;9(10):e1001331.
7. Chenoweth L, King MT, Jeon YH, Brodaty H, Stein-Parbury J, Norman R, Haas M, Luscombe G (2009). Caring for Aged Dementia Care Resident Study (CADRES) of person-centred care, dementia-care mapping, and usual care in dementia: a cluster-randomised trial. *Lancet Neurology*, 8(4):3,17-25. Erratum in: *Lancet Neurol*;8(5):419.
8. Davies, S. L., Goodman, C., Bunn, F., Victor, C., Dickinson, A., Iliffe, S., ... & Froggatt, K. (2011). A systematic review of integrated working between care homes and health care services. *BMC health services research*, 11(1), 320.
9. Froggatt K and Goodman C (second edition) End of life care for people with dementia. in Downs M, Bauer B. (eds) 2014 Dementia McGraw Hill NY.
10. Gomes B, Calanzani N, Curiale V, McCrone P, Higginson IJ. Effectiveness and cost-effectiveness of home palliative care services for adults with advanced illness and their caregivers. *Cochrane Database Syst Rev*. 2013 Jun 6;6:CD007760. doi: 10.1002/14651858.CD007760.pub2
11. Goodman, Claire, Catherine Evans, Jane Wilcock, Katherine Froggatt, Vari Drennan, Elizabeth Sampson, Martin Blanchard, Maggie Bissett, and Steve Iliffe. "End of life care for community dwelling older people with dementia: an integrated review." *International journal of geriatric psychiatry* 25, no. 4 (2010): 329-337.
12. Higginson, I.J., et al., Is Short-Term Palliative Care Cost-Effective in Multiple Sclerosis? A Randomized Phase II Trial. *Journal of Pain and Symptom Management*, 2009; 38: 816-826.
13. Hussain J, Adams D, Allgar V, Campbell C. Triggers in advanced neurological conditions: prediction and management of the terminal phase. *BMJ Supportive & Palliative Care*. 2014; 4: 30-37.
14. Kitwood T (1993). Towards a theory of dementia care: the interpersonal process. *Ageing Soc*,13(01):51-67.
15. Miller RG, Jackson CE, Karsarkis EJ, England JD, Forshew D, Johnston W et al. Practice Parameter update: The care of the patient with amyotrophic lateral sclerosis: Multidisciplinary care, symptom management, and cognitive / behavioural impairment (an evidence-based review). *Neurology* 2009; 73: 1227-1233;
16. Oliver D, Borasio GD, Johnston W. Palliative care in ALS 3rd edition 2014, Oxford University Press
17. Oliver DJ, Borasio GD, Caraceni A, de Visser M. et al., (2014) EFNS/ENS consensus review on palliative care for patients with progressive neurological disease. European Association for Palliative Care Taskforce Report. *Eur J Neurol* – submitted for publication

18. Palliative and End-of-Life Care Initiative: Impact Assessment' Report prepared for the CIHR Institute of Cancer Research, October 2009.
19. Perrar KM, Schmidt H, Eisenmann Y, Cremer B, Voltz R (2014). Needs of people with severe dementia at the end-of-life: A systematic review. *J Alzheimers Dis*; Aug 5. [Epub ahead of print]
20. Røsvik J, Brooker D, Mjorud M, Kirkevold Ø. (2013). What is person-centred care in dementia? Clinical reviews into practice: the development of the VIPS practice model. *Reviews in Clinical Gerontology*;23(02):155-163
21. Sampson EL, Jones L, Thuné-Boyle IC, Kukkastenvemas R, King M, Leurent B, Tookman A, Blanchard MR. Palliative assessment and advance care planning in severe dementia: an exploratory randomized controlled trial of a complex intervention. *Palliat Med*. 2011 Apr;25(3):197-209. doi: 10.1177/0269216310391691. Epub 2011 Jan 12.
22. Sampson EL, Lewis J, Pautex S, Shvartzman P, Van den Block L, Sacchi V, van der Steen JT, Van Den Noortgate N (2013). How well do European palliative care guidelines consider the assessment and management of pain for people with impaired cognition? *European Journal of Palliative Care* 2013, page 65, abstract PD 1.8
23. Temel JS, Greer JA, Muzikansky A, Gallagher ER, Admane S, Jackson VA, Dahlin CM, Blinderman CD, Jacobsen J, Pirl WF, Billings JA, Lynch TJ. Early palliative care for patients with metastatic non-small-cell lung cancer *N Engl J Med*. 2010 Aug 19;363(8):733-42
24. Toscani F, Di Giulio P, Villani D, Giunco F, Brunelli C, Gentile S, Finetti S, Charrier L, Monti M, van der Steen JT; End of Life Observatory-Prospective Study on Dementia Patients Care Research Group. Treatments and prescriptions in advanced dementia patients residing in long-term care institutions and at home. *J Palliat Med*. 2013 Jan;16(1):31-7. doi: 10.1089/jpm.2012.0165.
25. Traynor, B.J., et al., Effect of a multidisciplinary amyotrophic lateral sclerosis (ALS) clinic on ALS survival: a population based study, 1996-2000. *J Neurol, Neurosurg Psychiatry*, 2003. 74(9): p. 1258-61.
26. van der Steen JT, Radbruch L, Hertogh MPM C, de Boer ME et al., (2013) White paper defining optimal palliative care in older people with dementia; a Delphi study and recommendations from the European Association for Palliative Care. *Palliat Med*;28(3):197-209.
<http://pmj.sagepub.com/content/early/2013/07/05/-269216313493685>
27. van der Steen JT, Arcand M, Toscani F, de Graas T, Finetti S, Beaulieu M, Brazil K, Nakanishi M, Nakashima T, Knol DL, Hertogh CM (2012). A family booklet about comfort care in advanced dementia: three-country evaluation. *J Am Med Dir Assoc*;13(4):368-75.
28. van der Steen JT, Van Soest-Poortvliet MC, Achterberg WP, Ribbe MW, De Vet HC (2011). Family perceptions of wishes of dementia patients regarding end-of-life care. *Int J Geriatr Psychiatry*;26(2):217-20.
29. van der Steen JT, van Soest-Poortvliet MC, Hallie-Heierman M, Onwuteaka-Philipsen BD, Deliëns L, de Boer ME, Van den Block L, van Uden N, Hertogh CM, de Vet HC (2014). Factors associated with initiation of advance care planning in dementia: a systematic review. *J Alzheimers Dis* 40(3):743-57
30. Valentini E, Giantin V, Voci A, Iasevoli M, Zurlo A, Pengo V, Maggi S, Pegoraro R, Catarini M, Andrigo M, Storti M, Manzato E (2014). Artificial nutrition and hydration in terminally ill patients with advanced dementia: opinions and correlates among Italian physicians and Nurses. *J Palliat Med*. Sep 4. [Epub ahead of print]
31. Veronese S, Oliver D. Palliative care for people with neurodegenerative conditions. 2013. Lambert Academic Publishing. Saarbrücken.
32. Volicer L, Rheume Y, Brown J, Fabiszewski K, Brady R (1986). Hospice approach to the treatment of patients with advanced dementia of the Alzheimer type. *JAMA*;256(16):2210-3.
33. World Health Organization. (2002). *Palliative care*. www.who.int/cancer/palliative/definition/en/, accessed October 29th, 2014.

Annex I

**Agenda of June 25th expert-led workshop, Sheraton Hotel,
Schiphol Airport, Amsterdam**



- 09:35am: Welcome**
*Marie Claire de Vries, Department of Long-Term Care,
Ministry of Health, Welfare and Sport, The Netherlands*
- 09:40am: JPND – Tackling the neurodegenerative disease challenge**
Mogens Horder, member of JPND Executive and Management Boards
Including: Overview of JPND current activities and future plans
- 10:00am: Palliative care for patients with progressive neurological diseases**
David Oliver, University of Kent, United Kingdom
Including: Overview of consensus paper on available evidence
- 10:20am: Palliative Care for Neurodegenerative Diseases, in particular Dementia**
Jenny van der Steen, VU University Medical Center, The Netherlands
Including: Overview of EAPC White paper
- 10:40-12:30pm: Sub-group breakouts - Validating Research Priority Areas**
Participants broken up into two subgroups. The same topics/questions will be discussed by each group. Morning session to focus on understanding the research priorities and validating the EAPC agenda.
- 12:30-1:30pm: Networking Lunch**
Chairs and rapporteurs of subgroups to meet
- 1.30 – 3:00pm: Sub-group breakouts - Developing Proposed Actions**
Same subgroups as morning session. Afternoon session includes identification of five-six priority actions that JPND can initiate.
- 3:00 – 3:15pm: Afternoon Refreshments**
- 3:15 - 4:15pm: Summary reports from sub-groups' chairs/rapporteurs**
Discussion on recommendations, leading to overarching report
- 4:15 - 4:30pm: Closing Remarks**
Mogens Horder, member of JPND Executive and Management Boards

Annex II**List of Participants at Exert-led Workshop**

Workshop Attendees:

First name	Last name	Affiliation	Country
Gian Domenico	Borasio	University of Lausanne	Switzerland
Kevin	Brazil	Queens University Belfast	UK
Marie Claire	de Vries	Ministry of Health, Welfare and Sport	Netherlands
Carl Magnus	Edenbrandt	Lund University	Sweden
Yvonne	Engels	Radboud University Nijmegen Medical Centre	Netherlands
Rainer	Girgenrath	PT-DLR	Germany
Julio	Gomez	San Juan de Dios Hospital of Santurce	Spain
Claire	Goodman	University of Hertfordshire	UK
Dianne	Gove	Alzheimer Europe	Luxembourg
Jacqueline	Hoogendam	Ministry of Health, Welfare and Sport	Netherlands
Mogens	Horder	University of Southern Denmark	Denmark
Oyvind	Kirkevold	Norwegian center for research, education and service development	Norway
Raymond	Koopmans	Radboud University Nijmegen Medical Centre	Netherlands
Philip	Larkin	University College Dublin	Ireland
Stephanie	Le Naour	Inserm Transfert	France
Margje	Mahler	Knowledge Through Connection	Netherlands
Sonja	Mcilpatrick	All-Ireland Institute for Hospice and & Palliative Care	UK/Ireland
Derick	Mitchell	JPND	Ireland
David	Oliver	University of Kent	UK
Edward	Richfield	University of Hull	UK
Helle	Timm	National Centre for Palliation	Denmark
Suzanne	Timmons	University College Cork	Ireland
Lieve	Van den Block	Vrije Universiteit Brussel	Belgium
Jenny	van der Steen	VU University Medical Center	Netherlands
Myrra	Vernooij-Dassen	Nijmegen Centre for Evidence Based Practice	Netherlands
Ladislav	Volicer	Univeristy of South Florida	USA / Czech Republic

Completed Questionnaires:

First name	Last name	Affiliation	Country
Paola	Di giulio	Istituto di Ricerche Farmacologiche Mario Negri, Milano	Italy

Shelly	Sternberg	Maccabi Healthcare Services	Israel
Birgit	Rasmussen	Lund University	Sweden
Katharina	Heimerl	University of Klagenfurt, Vienna	Austria
Kelli	Stajduhar	University of Victoria	Canada
Genevieve	Thompson	University of Manitoba	Canada
Franco	Toscani	Istituto di Ricerca in Medicina Palliativa 'L. Maestroni', Cremona	Italy
Nele	Van den Noortgate	University Hospital Ghent	Belgium
Bettina	Husebo	University of Bergen	Norway
Louise	Robinson	Newcastle University	UK

**Annex III Collaborative projects supported under the 2012 JPND Transnational call:
“European research projects for the evaluation of health care policies, strategies
and interventions for Neurodegenerative Diseases”.**

The supported projects are: (CTRL + Click on the project title for a link to an online fact sheet)

*** [Projects with Palliative and End-of-life Care Research Elements](#)**

*** ACTIFCare: [ACces to Timely Formal Care](#)**

Coordinator: Frans Verhey, Maastricht University, Netherlands

*** ALS-CarE: [A Programme for ALS Care in Europe](#)**

Coordinator: Orla Hardiman, Trinity College Dublin, Ireland

*** CLaSP: [Care for Late Stage Parkinsonism](#)**

Coordinator: Anette Schrag, University College London, United Kingdom

*** NEEDS in ALS: [Strategies and interventions for vital decisions in Amyotrophic Lateral Sclerosis in different European countries](#)**

Coordinator: Dorothee Lulé, University of Ulm, Germany

MEETINGDEM: [Adaptive Implementation and Validation of the positively evaluated Meeting Centers Support Programme for people with dementia and their carers in Europe](#)

Coordinator: Rose-Marie Dröes, VU University Medical Center Amsterdam, Netherlands

RHAPSODY: [Research to Assess Policies and Strategies for Dementia in the Young](#)

Coordinator: Alexander Kurz, Technical University Munich, Germany