Nienke Bekkema

# The challenge of caring together

END-OF-LIFE CARE FOR PEOPLE
WITH INTELLECTUAL DISABILITIES

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## End-of-life care for people with intellectual disabilities

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# The challenge of caring together
## End-of-life care for people with intellectual disabilities

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# Contents

# 1

# General introduction

*Joe, a man aged 40 with a severe intellectual disability (ID), suffered from incurable lung cancer. He lived in a small-scale residential home on the premises of an ID care provider. Joe's situation worsened and communicating with him became an even greater challenge. Joe's sister: "I really wanted to talk to him at the same level for just one day – so that he could tell me what he wanted and how he felt". The question was raised where Joe would stay. Joe's social worker was emotional about this: "Clients often move to a more specialized care setting, but the wish of the whole team was: Joe should stay here. We wanted to do it ourselves [...] We all loved Joe. He was more than just a client to us". Although Joe's sister was happy that Joe could stay with his familiar care staff, she struggled to cooperate with the staff and was critical about their experience of end-of-life care: "I think they lack a lot of experience, which they cannot be blamed for, as they don't often have one of their clients dying. You really miss that experience though, as relatives".*

Joe's case highlights how challenging end-of-life care for people with ID can be. Communicating with Joe, discovering what his needs and wishes are, the team's expertise and cooperation between his sister and care staff - none of these things are self-evident. These challenges, amongst other topics, will be addressed in this thesis in an attempt to explore further the nature of end-of-life care for people with ID. This introduction first considers the characteristics of people with ID, their care and the way they age. Then, key literature on the challenges in end-of-life care for people with ID is presented as well as the role of relational aspects within this care. At the end of the chapter, the aim and structure of the thesis are set out.

## Characteristics of people with intellectual disabilities

Individuals with ID are characterized by significant limitations, from before the age of 18, in both intellectual functioning and adaptive behaviour (Schalock *et al.* 2010). Usually, an IQ below or around 70 indicates a limitation in intellectual functioning (AAIDD 2015a). There are no exact numbers of the prevalence of ID in the Netherlands, but recent estimates suggest there are approximately 142,000 people with an ID with an IQ < 70, of whom 74,000 people with an IQ between 70 and 50 and 68,000 people with an IQ below 50 (Woittiez *et al*. 2014). Traditionally, the level of intellectual functioning is classified as follows: mild IQ (IQ 50/55-70), moderate ID (IQ 35/40-50/55), severe ID (IQ 20/25-35/40) and profound ID (IQ below 20/25) (Buntinx *et al.* 2014).

## Care for people with intellectual disabilities

In 2013, 163,615 people in the Netherlands had an indication for care based on their intellectual disability (monitor langdurige zorg 2015). In the Netherlands, the care for people with ID is often provided by 'ID care services'. These are usually services that primarily provide care for people with ID, or a service offering combined care for people with intellectual and/or other disabilities, such as sensory and physical disabilities (Kwartel 2013). In 2013, 72,200 people with ID lived in accommodation provided by an ID care service (VGN 2015).

Since the de-institutionalization of the care for people with ID in the 1970s, more people with ID live within the community, in group homes or ordinary homes. As in other Western countries, the trend towards integration in society and community-based care is strong in the Netherlands (United Nations 2006; WHO 2011). Yet European countries do differ in their policies related to the process of de-institutionalization. In countries such as Sweden and Norway, large residential institutions no longer exist and the process of de-institutionalization in the UK is well advanced too (Beadle-Brown *et al*. 2007; Mansell & Beadle-Brown 2010). In the Netherlands and countries such as Belgium, Germany, Spain and Greece, de-institutionalization is progressing, but residential settings still exist (Beadle-Brown *et al.* 2007; Mansell *et al.* 2007). Although exact numbers are lacking, this may mean that relatively more people with ID live within institutional or semi-institutional settings in the Netherlands compared to other countries such as the UK.

In the Netherlands, care staff for people with ID have varying backgrounds, but the large majority are social workers (in Dutch: *agogisch begeleiders*), followed by nurses and care and nursing assistants (VGN 2015). As reflected in data for 2004 and 2014, the proportion of nurses working in the care for people with disabilities in the Netherlands is decreasing, while the proportion of social workers is increasing (van der Windt & Bloemendaal 2015; van der Windt & Talma 2005). Internationally, the background, training and qualifications of care staff in ID care are not well defined. An expert consultation among several European countries suggests that ID care staff in Europe differ in their qualifications, and encompass e.g. staff with limited educational qualifications, social workers and nurses specialized in ID (Bekkema *et al.* 2015). Reports from the UK suggest that many UK care staff members who support people with ID on a day-to-day basis have no formal qualifications (Mansell 2006).

As regards medical care, people with ID in the Netherlands are usually cared for by general practitioners (GPs) or ID physicians. Dutch ID physicians have received three years of specialist training in the care for people with ID and are generally employed by an ID care service. GPs care mostly for people with mild ID and ID physicians mostly for people with moderate to severe/profound ID. European countries differ in the types of doctors that offer medical care to people with ID. They include general practitioners and psychiatrists (Bekkema *et al.* 2015).

### People with intellectual disabilities: a vulnerable ageing group

Health care has improved to such an extent that people with intellectual disabilities are also living longer than in previous decades (Coppus 2013). Yet adults with ID often age while suffering from various life-limiting chronic illnesses, such as progressive cancer, chronic cardiovascular diseases, chronic lung diseases and dementia (Janicki *et al.* 1999; Tuffrey-Wijne *et al.* 2007). A Dutch study among 1050 people with ID aged 50 and older showed that their frailty is comparable to the frailty of people aged 75 and older in the general population (Evenhuis 2014). Also, the prevalence of multimorbidity is high: 79.8% of the people with ID aged over 50 in the study suffered from two or more chronic conditions (Hermans *et al.* 2014). An Irish study on multimorbidity showed similar results and even showed that multimorbidity was high (63%) in a younger age group of people with ID (40-49 years) (Mccarron *et al.* 2013). Appropriate identification and management of increasing comorbid conditions is further complicated by the variety of syndrome-related ageing issues (Perkins & Moran 2012). Their poor health status in combination with their intellectual disabilities makes people with ID a vulnerable ageing group.

### End-of life care for people with intellectual disabilities

As people with ID live longer and age with chronic illnesses, they suffer longer from conditions that eventually lead to their death. As a result, more people with ID are in need of end-of-life care for a longer period.

There are several definitions of end-of-life care. In this study, end-of life care is defined as the care aimed at the quality of life of a person with a life-threatening illness that is expected to result in the person's death. The quality of life is enhanced by relieving pain and other distressing symptoms, incorporating the holistic needs of people as reflected in physical, psychological and spiritual support. End-of-life care parallels ‘palliative care’. Palliative care is defined as “an approach that improves the quality of life of patients and their families facing the problem associated with life-threatening illness, through the prevention and relief of suffering by means of early identification and impeccable assessment and treatment of pain and other problems, physical, psychosocial and spiritual” (WHO 2015). Palliative care may start early in the trajectory of a life-threatening illness, and curative and life-prolonging interventions may initially co-occur with palliative care (Lynn & Adamson 2003). [1]

End-of-life care for people with intellectual disabilities may be particularly difficult because of the combination of complex medical problems, behavioural problems (in some cases) and limitations to verbal communication. Research literature indicates challenges in several areas:

- *The challenge of communication with people with ID*. Communicating with people with ID about their illness and their wishes and needs is found to be challenging (Tuffrey-Wijne *et al.* 2007). E.g. adequately assessing symptoms in people with severe/profound ID and severe communication problems is found to be very difficult (Tuffrey-Wijne *et al.* 2007). Moreover, for staff in palliative care services, communicating with people with ID can generally be difficult as these professionals are not trained in this or used to doing it (Stein 2008; Tuffrey-Wijne *et al*. 2008). Barriers to communication can hamper a good provision of end-of-life care (Tuffrey-Wijne *et al.* 2009 and 2010).
- *Lack of involvement of people with ID*. Studies suggest that people with ID are not always adequately involved in communication about their illness and in decisions about end-of-life care (Tuffrey-Wijne *et al.* 2009 and 2010; Wagemans *et al*. 2010 and 2013). E.g. Tuffrey-Wijne found that although most participants with ID were told they were ill, many were not helped to understand what this meant. Furthermore, in a retrospective study of medical files, Wagemans *et al.* (2010) found no evidence in the files that people with ID were involved in end-of-life decision making. The studies highlight how complex it is to involve people with ID who are near the end of their life in communication and decisions.
- *Lack of expertise in end-of-life care*. To provide comprehensive, multidisciplinary end-of-life care to this challenging population, care staff and other professionals need various kinds of knowledge and skills. Yet studies suggest that care staff working with people with ID often lack expertise in

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[1] In English, the term ‘end-of-life care’ is sometimes associated exclusively with terminal care (care in the final days or hours). We use ‘end-of-life care’ (and the Duch equivalent ‘*zorg aan het levenseinde*’) as a broader concept describing care that may start long before death (even months or years earlier). In this sense, end-of-life-care is a synonym for palliative care as defined by the WHO (www.WHO.int)

end-of-life care (Ng & Li 2003; Botsford 2004; Ryan *et al*. 2010; McCarron *et al*. 2010; Tuffrey-Wijne *et al.* 2007; Dunkley & Sales 2014). E.g. Ryan *et al.* (2010) found that Irish ID care staff often lacked confidence and training in end-of-life care. Likewise, a study in Australian ID community group homes revealed that less than one third of the professionals concerned received any training in end of-life care (Wiese *et al*. 2012). A review by Dunkley and Sales (2014) highlights that the training requirements of care staff need to be identified and that staff confidence needs attention.

- *Suboptimal cooperation between palliative care services and ID care services*. The literature suggests there is too little collaboration between ID care services and specialist palliative care services, such as hospices or palliative care units in hospitals or nursing homes (Dunkley & Sales 2014). Effective collaboration may be hampered by differences in care approaches between these services and by skill limitations (Friedman *et al.* 2012; Mccarron *et al*. 2010; Ryan *et al.* 2010; Tuffrey-Wijne *et al*. 2008).
- *Late recognition of needs for end-of-life care.* The above may contribute to the fact that the end-of-life care needs of people with ID are often recognized only at a late stage, when death is very close (Heslop *et al*. 2013). This is alarming, as late recognition may lead to sub-optimal care and unrelieved suffering in the final stage of life.

The challenges in end-of-life care for people with ID pose the risk that end-of-life care is not being properly adapted to the needs of people with ID. End-of-life care for people with ID should therefore be further explored and improved by incorporating the perspectives of the most important stakeholders: people with ID, relatives and professionals (in particular care staff and physicians).

### Relational aspects and care ethics

The abovementioned challenges reveal that various people are involved in end-of-life care for people with ID and that many of the challenges are relational issues at heart. Taking Joe's case, cited at the start of the Introduction, as an example: uncovering the needs and wishes of Joe, who was severely disabled, inevitably required good relational skills. Honouring the wishes and autonomy of people with ID at the end of their lives is generally seen as an important principle (AAIDD 2015b). Yet uncovering the wishes of people like Joe may be complex and not self-evident. Besides, Joe's relatives and care staff had to build a relationship in order to find ways to cooperate and provide proper end-of-life care for him. Hence, caring for someone like Joe is not easy. It likely raises ethical issues, such as what is the best care for Joe and who should decide about this care.

In order to explore such ethical issues within the practice of end-of-life care for people with ID, *care ethics* may provide a useful approach. There are two relevant reasons why care ethics is well suited to a better interpretation and understanding of end-of-life care for people with ID and its relational aspects: 1) care ethics is based on a relational human-centred vision and thus aligns well with end-of-life care, focusing both on patients and their significant others (Leget 2013); 2) care ethics also aligns well with the care for people with ID, as care ethics is based on the fundamental vulnerability and dependence of people, and most people with ID have always been more or less dependent on others. At the end of life, when people with ID become increasingly frail, this dependency may be even more pronounced.

By homing in on the dependency of people and the importance of care relationships, care ethics may contribute to our understanding of the practice of end-of-life care for people with ID. Core elements of care ethics are for example: *attentiveness* to recognize the need for care*, responsibility* for taking on the care when the need for care is recognized, *competence* (the moral quality, knowledge and resources) to provide actual care, and the *responsiveness* of the care receiver to the care (Tronto 1993). Care ethics has become important in the thinking about people with ID (e.g. Verkerk & Maeckelberghe 2003; Kittay 2011). Care ethicist Kittay for example states "It is only with care, and care of the highest quality, that she can be included, loved, and allowed to live a joyful and dignified life", when she writes about her own daughter with severe ID (Kittay 2011, p. 52).

## Aim and structure of this thesis

End-of-life care for people with ID is a relatively young research topic (Todd 2003; Todd *et al*. 2013). Only in recent years has the volume of research and the scientific body of knowledge slowly started to increase. The general aim of this thesis is to further explore end-of-life care for people with ID from the perspectives of people with ID, relatives, care staff and physicians. This thesis aims to gain a better understanding in three areas: 1) the care, needs and wishes of people with ID at the end of life; 2) decision making at the end of life of people with ID; and 3) the end-of-life care expertise and training needs of ID care staff. Six research questions were formulated, and several different research methods were used to address these questions. This thesis is set up as follows:

*Part 1: Care, needs and wishes of people with ID at the end of life*

Part 1 concentrates on the care, needs and wishes of people with ID at the end of life. It describes whether and how relatives, care staff and physicians change their care approach and attitudes when the death of a person with ID is imminent, as end-of-life care may not be naturally embedded in ID care services. It also deals with the dimensions of the care relationship that people with ID themselves find important for good end-of-life care, as people with ID may have different views on care than care staff, physicians and relatives. Finally, it studies how relatives, care staff and physicians shape respect for autonomy in the end-of-life care for people with ID. The following main research questions will be addressed in Chapters 2 to 4:

- Chapter 2. Do relatives, care staff and physicians perceive a shift in their care approach and attitudes when the death of a person with ID is imminent? And if so, what shifts in care approaches and attitudes do they perceive? And what values underlie these shifts?
- Chapter 3. Which dimensions of the care relationship in end-of-life care are perceived as relevant from the perspective of people with ID?
- Chapter 4. How do relatives, care staff and physicians shape respect for autonomy in the end-of-life care for people with ID?

Two qualitative studies were conducted to answer these research questions. The first study (Chapters 2 and 4) consisted of a retrospective multiple-case study design, interviewing those relatives, care staff members and physicians involved in the care for a person with ID at the end of life.

The second qualitative study (Chapter 3) consisted of group interviews with participants with ID, using the nominal group technique to guide the discussion.

*Part 2: Decision making at the end of life of people with ID*

Part 2 addresses decision making in end-of-life care for people with ID. Two types of decisions that often have to be made and may lead to dilemmas are decisions regarding the place of end-of-life care and the use of medical interventions. Is it better to move a person with ID to a place offering more specialized care, but which is not familiar to the person? And should one start a medical intervention that is potentially burdensome for a person due to their limited level of understanding and communication difficulties? To enhance well-considered decision making, we need insight into the beliefs and actual considerations of care staff and physicians in making such decisions. The following research questions will be addressed in

Chapters 5 and 6, based on a quantitative survey study among ID care staff, ID physicians and general practitioners:

- Chapter 5: What considerations and beliefs do care staff and physicians have concerning decisions on the place of end-of-life care for people with ID?
- Chapter 6: What considerations and beliefs do care staff and physicians have concerning decisions about the use of potentially burdensome medical interventions in the end-of-life care for people with ID?

*Part 3: Expertise and training needs of ID care staff*

Part 3 focuses on the expertise and training needs of care staff working in ID care services in the Netherlands. Although growing attention has been paid in the Netherlands to the quality of end-of-life care in ID care services, earlier studies suggest that ID care staff may have insufficient expertise in end-of-life care (Bekkema *et al.* 2011). In order to further promote good care for incurably ill people with ID, more insight is needed into vocational training in end-of-life care, the training needs and the consultation options of ID care staff (nurses as well as social workers). The following research question will be addressed in Chapter 7, based on a quantitative survey study among care staff:

- Chapter 7: What views and needs do care staff in ID care services have with regard to their expertise and training in end-of-life care?

Finally, Chapter 8 concerns the *general discussion*, in which reflections are made concerning the main findings, methodological considerations are discussed, and the implications for practice, policy and research are examined.

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# 2

# ‘From activating towards caring’: shifts in care approaches at the end of life of people with intellectual disabilities; a qualitative study of the perspectives of relatives, care-staff and physicians

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## Abstract

### Background

Professionals and relatives increasingly have to deal with people with intellectual disabilities (ID) who are in need of end-of-life care. This is a specific type of care that may require a different approach to the focus on participation that currently characterizes the care for people with ID. This paper describes the shifts in care approaches and attitudes that relatives and professionals perceive as the death of a person with ID approaches, as well as the values underlying these shifts.

### Methods

A qualitative design was used to reconstruct the cases of twelve recently deceased people with ID. Relatives and professionals who were closest to the person at the end of their life were interviewed. Interviews were transcribed verbatim and data were analyzed inductively, using elements of thematic analysis.

### Results

Five shifts were found: 1) adapting to a new strategy of comforting care, taking over tasks and symptom relief, 2) interweaving of emotional and professional involvement, 3) stronger reliance on the joint interpretation of signals expressing distress and pain, 4) magnified feeling of responsibility in medical decisions, 5) intensified caring relationship between ‘two families’: relatives and care staff. Six relational values were behind these shifts: ‘being there’ for the person with ID, ‘being responsive’ to the person’s needs, ‘reflection’ on their own emotions and caring relationships, ‘attentiveness’ to the ID person’s wishes and expressions of distress, ‘responsibility’ for taking joint decisions in the best interests of the person, and ‘openness to cooperation and sharing’ the care with others.

### Conclusions

End-of-life care for people with ID involves curtailing expectations of participation and skill acquirement, and an increase in teamwork featuring intensified comforting care, symptom management and medical decision making. Three caring relationships need to be fostered: the relationship with the person with ID, relationships among professionals and the relationship between relatives and professionals. ID care services should invest particularly in the emotional support and expertise level of care staff, and in the collaboration between relatives and professionals.

Keywords
End-of-life care, Intellectual disabilities, Professionals-family relationship, Values

## Background

In line with the ongoing process of the de- institutionalization of people with Intellectual Disabilities (ID), the focus is increasingly on the self-reliance of people with ID and their participation in society (Nations U 2006; WHO 2011). Individuals with ID are characterized by significant limitations, from before the age of 18, in both intellectual functioning and adaptive behavior (Schalock *et al.* 2010). Many policy and healthcare initiatives are undertaken to foster equal opportunities for people with ID, endorsed by the UN's stated principles of equal rights for people with disabilities so that they can live, participate and be fully included in the community (Nations U 2006). People with ID are encouraged to acquire new social and practical skills and to live as independently as possible (Dusseljee *et al.* 2010). Several methods are used to enhance social inclusion, such as personal goal setting (McConkey & Collins 2010). People with ID increasingly live in community houses rather than in segregated institutions, which gives them more opportunity to participate in the community. This development can be typified as a shift towards a citizenship paradigm (Van Gennep 1997). In the citizenship paradigm, relatives and professionals encourage the selfdetermination and personal development of people with ID (van Loon & van Hove 2001). The 'new' professional has become an 'enabler', a companion who enables a person with ID to participate in society and reach their full potential (Baart 2004). The dominant values underlying the citizenship paradigm are e.g. helping the person with ID to realize self-actualization and reach autonomy.

Another relevant development for people with ID is the improvement in health care over recent decades. Consequently, the life expectancy of people with ID has increased and they form an aging population. This epidemiological development is connected with growing incidences of life-limiting illnesses, such as progressive cancer, chronic cardiovascular diseases, chronic lung diseases and dementia (Ellison & Rosielle 2008; Janicki *et al.* 1999; Tuffrey-Wijne *et al.* 2007a). As a result, professionals and relatives increasingly have to deal with people with ID who need end-of-life care. End-of-life care may start early on in the trajectory of a life-threatening illness and can be typified as multidisciplinary care aimed at enhancing the quality of life by assessing and relieving pain and other distressing symptoms, while taking account of the physical, psychological and spiritual needs of patients and their families. End-of-life care for people with ID is known to be particularly challenging due to their limited level of understanding and communication difficulties (e.g. Tuffrey-Wijne *et al.* 2009; Stein 2008). Several studies have indicated that ID care professionals have a lack of knowledge and expertise in end-of-life care, for example in pain and symptom management (McCarron *et al. 2010*; Bekkema *et al.* 2014a; Ng & Li 2003). Todd interviewed ID care staff and found that although care staff are willing to provide end-of-life care for their clients (with whom they often have strong bonds), they can be very overwhelmed by caring for a dying person. This is amplified by a lack of knowledge and a lack of the kind of support that would enable them to provide this care properly (Todd 2004, 2005). In an earlier study, we found that insufficient expertise among care staff within ID care services often prompts the decision to move a terminally ill client to another care setting (Bekkema *et al.* 2014b).

End-of-life care may require a different care approach and attitude to what caregivers are used to. Kirkendall argues that when death is imminent, people with ID need an approach covering all aspects of end-of-life care, an approach that may not be naturally rooted in ID care services (Kirkendall *et al.* 2012). Thus far, it is unclear whether and how relatives and professionals change their care approach and attitudes when the death of a person with ID is imminent. To gain more insight into these processes, this paper will address the following research questions: do relatives, ID care staff and ID physicians perceive a shift in their care approach and attitudes when the death of a person with ID is imminent? And if so, what shifts in care approaches and attitudes do they perceive? And what values underlie these shifts?

## Methods

### Design

This study used a retrospective case study design. We held semi-structured interviews with professionals and relatives who were close to a person with ID at the end of life. In this way, end-of-life care of twelve recently deceased people with ID were reconstructed from different perspectives. Such a qualitative design is particularly well suited to comprehending complex issues.

This paper is part of a wider study set up to increase knowledge about end-of-life care for people with ID in the Netherlands. From the inductive analyses of the data it appeared that some major shifts in care approaches were taking place. These shifts are described in this paper. Another paper, based on the same qualitative data, describes another main theme emerging from the analyses, namely how care providers try to respect the autonomy of people with ID at the end of life (Bekkema *et al.* 2014c).

### Participants

Twelve recently deceased people with ID were selected by the contact persons at ten ID care services organizations. In total, 45 professionals and relatives who were close to one of these recently deceased people were interviewed. Table 1 shows that the cases comprised six men and six women with different degrees of severity of ID. Half of them died of cancer. They reached a mean age of 53 years (range from 13 to 74). Six died in their own living environment, two in an intensive-care unit of their ID care service, three in a hospital and one in a hospice. Nine of the twelve people with ID lived in community houses and three in houses in a residential complex. All received care or support from an ID care service. All but one person died less than a year before the interview was held, allowing us to retrieve the participants' recent memories. One person died two-and-a-half years before the time of the interview and was included because of his specific situation, dying in a hospice.

Table 1 General overview of the cases: the deceased people and the interviewees

| | **Relatives interviewed** | **Professional caregivers interviewed** |
|---|---|---|
| Case 1: female, died from dementia, moderate intellectual disability | 1 mentor<br>(a friend) | 1 nurse<br>1 ID physician<br>1 psychologist |
| Case 2: female, died from metabolic disease, severe/profound intellectual disability | 1 mother | 2 social workers [1]<br>1 nurse<br>1 GP |
| Case 3: female, died from kidney failure and heart failure, mild intellectual disability | 1 sister<br>1 brother-in-law | 1 social worker<br>1 GP |
| Case 4: male, died from cancer, mild intellectual disability | 1 sister<br>1 brother-in-law | 1 social worker<br>1 nurse specialized in palliative care<br>1 GP |
| Case 5: female, died from cancer, mild intellectual disability | | 1 social worker<br>1 nurse |
| Case 6: male, died from cancer, severe/profound intellectual disability | 1 sister | 1 social worker<br>1 ID physician |
| Case 7: male, died from metabolic disease, severe/profound intellectual disability | 1 mother | 1 social worker<br>1 nurse<br>1 ID physician |
| Case 8: male, died from cancer, mild/moderate intellectual disability | 1 sister | 2 social workers<br>1 ID physician |
| Case 9: male, died from cancer, mild intellectual disability | 2 sisters | 1 social worker<br>1 hospice coordinator |
| Case 10: female, died from dementia, moderate intellectual disability | 1 brother | 1 nurse<br>1 ID physician |
| Case 11: male, died from frailty in old age, severe/profound intellectual disability | 1 brother | 1 social worker<br>1 end-of-life care consultant [2] |
| Case 12 female, died from cancer, moderate intellectual disability | 1 sister | 2 social workers<br>1 ID physician |
| **Total** | **16** | **29** |

[1] In the Netherlands, social workers who work in ID care services provide support to people with ID in their daily lives. These social workers usually have an associate degree in socio-pedagogical support.

[2] The end-of-life care consultant in this case was a nurse from the ID care service, specialized in end-of-life care, who advises teams of nurses and social workers on how to provide end-of-life care.

### Data collection

Individual in-depth semi-structured interviews were held at the place of preference of the interviewees. Interviews were conducted by the first author and lasted one to two hours. Open questions were used, encouraging interviewees to describe the case in their own words. A topic list was used as a guide for formulating interview questions. The topic guide was developed in two stages: 1) the first draft of the topic guide was inspired by elements in the WHO definition of palliative care (WHO 2015) and literature regarding end-of- life care for people with intellectual disabilities (Tuffery-Wijne *et al.* 2007b; Tuffrey-Wijne & McEnhill 2008; Wagemans *et al.* 2010). 2) This draft guide was discussed within a group of six experts working in the field of end-of-life care for people with intellectual disabilities, after which the guide was refined and finalized. The first question in the topic guide was what had made the strongest impression on the interviewee during the period of end-of-life care. Subsequent topics or questions concerned the care provided, communication with the person with ID, communication among professionals and between professionals and family, end-of-life decisions and dealing with loss. Table 2 presents an overview of the main topics. The order in which topics were discussed varied, depending on the course of the interview.

**Table 2** Overview of the main interview topics

| **Opening question** | **What made the strongest impression on the interviewee during the period of end-of-life care?** |
|---|---|
| Topic 1 | Marking the start of palliative phase, communication about the diagnosis |
| Topic 2 | Primary care process (e.g. pain, other physical symptoms, physical care, psychosocial well-being and spiritual well-being) |
| Topic 3 | Decisions at the end of life |
| Topic 4 | Communication with the person with ID |
| Topic 5 | Autonomy of the person with ID |
| Topic 6 | Transitions (e.g. changes in place of care or caregivers) |
| Topic 7 | Communication and cooperation with relatives |
| Topic 8 | Care for relatives (e.g. dealing with loss) |
| Topic 9 | Fellow residents |
| Topic 10 | The funeral |
| Topic 11 | Aftercare |
| Topic 12 | Care for carers (e.g. dealing with loss) |

Participants were selected purposively in order to obtain a relatively diverse set of client cases with regard to age, living situation (own apartment, group home or residential home), place of death (own home, parents' home, intensive care facility provided by an ID care service, hospital or hospice), kind of disease and severity of ID (mild, moderate or severe ID). In this way, a variety of situations could be taken into account in the study. The number of cases was not predetermined. Rather, data collection and analysis were conducted in a cyclical process in accordance with key principles of qualitative research (Green & Thorogood, 2004). Data saturation (Ando *et al.* 2014) was attained after twelve cases.

### Analysis

Interviews were taped and transcribed verbatim. Data were analyzed inductively, using elements of thematic analysis (Braun & Clarke 2006). Important elements of thematic analysis are familiarizing with the data, generating initial codes, searching for themes, reviewing and defining themes and writing down the results. Thematic analysis was applied as follows: interviews were re-read and codes were ascribed to those text fragments potentially reflecting shifts and values. We used descriptive codes directly based on words that the interviewees used (such as having two families), as well as interpretative codes (such as attentiveness). Interview fragments with the same codes were constantly compared. Written memos were made concerning the interpretation of text and the relationships between codes throughout the analysis process. All authors were involved in the entire process from the analyses of interviews to the generation of concepts. All interviews were analyzed by the first author. To ensure reliability, each co-author individually analyzed all interviews for at least one case. Correspondence between interpretations and the original interviews was continuously verified and differences in analysis and interpretations were discussed. After the first author had discussed a third of the interviews with the co- authors, consensus was reached about the chosen codes and their interpretation. After coding all transcripts, potential themes were formulated. These themes were then reviewed in relation to the coded text fragments and transcripts. Finally, themes were refined and written down. MAXQDA 2007 was used to facilitate the analysis (MAXQDATA 2015).

### Ethical consideration

All interviewees were mentally competent individuals and no interventions were performed. In such cases, no approval by an ethics committee is required in the Netherlands, according to the Medical Research Involving Human Subjects Act (CCMO 2015). All respondents received a letter informing them of the aim of the study and gave informed consent. Study participation was voluntary. The responses were anonymous and non-traceable to individuals.

## Results

Relatives, care staff and ID physicians perceived several shifts in their care approaches and attitudes as the death of a person with ID approached. Five shifts were distinguished. They are described below, including the values behind these shifts. To illustrate the shifts, we present the cases of Josh, Eleanor and Joe (see Table 3; the names used in the case descriptions are not the subjects' real names). We also discuss the extent to which they resemble the other cases studied.

**Table 3** Cases illustrating the perspectives of care staff, physicians and relatives

| |
|---|
| Josh was a man aged 58, with moderate intellectual disability (Down syndrome). He died from dementia. Josh lived in a group home in the community. |
| Eleanor was a frail woman aged 74, with Parkinson's disease and heart failure. Eleanor was in a wheelchair and had a severe intellectual disability. Eleanor lived in a group home in the community. She died in a hospital. |
| Joe was a man aged 40, with a severe intellectual disability and lung cancer. Joe lived in a small-scale living facility on the residential premises of an ID care service. |

### From activating towards comfort care and taking over tasks

For care staff, providing care for a person with ID in their last phase in life generally meant a shift to a different care approach. Care staff were used to focusing on encouraging the person to undertake activities. However, as the terminal condition progressed and the person with ID became less and less able to participate in daily activities (such as day care, work and hobbies), care staff took over more and more daily tasks. Meanwhile, care staff increasingly put the emphasis on paying more loving attention to the person with ID and providing comfort care, such as holding hands, giving massages, offering favorite foods, listening to favorite music, having reassuring talks, or undertaking small favorite activities such as blowing bubbles or watching football. In the last stage in life, even regular care, such as showering or eating, was sometimes burdensome and new solutions needed to be found, such as reducing time taken showering, washing in a bath chair and offering favorite foods in a liquid form. Josh's nurse described how she experienced the transition towards end-of-life care:

> Josh's Nurse: *'There are no 'musts' anymore. It's more about letting go, letting Josh have a fine last week. Let him snuggle in bed. He doesn't have to sit up if he doesn't have the power to do so. But it was very difficult to let it all go. <> I often wondered: when am I going to take it all over from him? We have learned to encourage clients in all their daily activities and even encourage them to do more activities. Now you have to let go. You should ask yourself: who do I keep offering the activities for? <> We are not doing him a favor. Josh will never be the same person. <> This awareness came slowly, but it was very hard.'*

As Josh's case illustrates, for most care staff, the shift from care as usual to end-of-life care implied a continuous search for a new balance between offering activities (perhaps adapted) and taking over tasks from the person with ID. This could be quite difficult, as it implied 'letting go' of parts of a care strategy that care staff identified with as being the essence of their work, namely expanding the activities and encouraging self-reliance.

For most relatives, it was particularly hard to see the decline in the health of their relative, particularly when this was a process of rapid deterioration. In some cases, the person ended up in a wheelchair and eventually in bed in just a matter of days, not being able to eat or drink properly. In other cases, the deterioration went more slowly, for example in the case of dementia or frailty due to old age. But even then, the person could become weaker quickly in the final weeks. Joe had cancer and his sister described how she experienced the shift:

> Joe's sister: *'Joe always liked to go outside. He had a custom-made bike and we often went out together. He could also really shake his head and say: "I don't want to go out". But I thought: he should go out, it's good for his health, and he will see some different things.<> Then I heard of a woman who also had cancer*

> *and who complained that her friends would always take her out, but she had to recover for a week from these outings, and she actually did it more for them than for herself. I thought: why do I still take Joe out? Only to tell myself he is doing fine? Then I decided: if he doesn't want to go, we won't go. We don't have to keep him in shape, he's going to die. I have to look at his abilities and should not assume that shaking his head is just a gesture, it really means no.'*

Joe's sister reflected on her activity planning with her brother and came to the conclusion that she needed to respond better to his signals. Like Joe's sister, many relatives were used to encouraging their relative to be active and now increasingly had to let go of these expectations, adapting to what was still possible.

'Being there' and making more time available to be close to the ill person and subsequently 'being responsive' to the person's real needs and possibilities were identified as values underlying the shift towards comfort care and taking over tasks.

### Interweaving of emotional and professional involvement becomes a struggle

The second shift dealt with the interweaving of the emotional and professional involvement of care staff with their clients. This interweaving had always been there, but increasingly became a struggle as the death of a client came near. In ID care services, many care staff members have built long-term caring relationships with their clients, which distinguishes this setting from many other healthcare settings. Their compassionate caring relationships were sometimes even described as a 'special bond', 'love' or 'friendship'. The prospect of losing this client combined with the often hectic period of intense care for the dying client and their co-residents led to a rise in emotions that could be hard to deal with. Eleanor's social worker described how she experienced her emotions:

> Eleanor is in the hospital and she is expected to die soon. Her social worker and brother are visiting her as often as they can. The social worker is about to go on holiday. She is stressed about this: *'I didn't want Eleanor to be alone in the hospital. I found it hard to go home. <> Eleanor's brother told me several times I could go home. Then I decided: I can only stay this long tonight, then I have to go home, I am going on holiday the next day. <> There are boundaries, I could not have stayed all night. I told myself: I need to transfer my care for Eleanor to others now. But that was very hard. I couldn't get that act together. I was so deeply involved in her care. It was hard to decide to leave. <> So I wrote a card for*

> *Eleanor, for in her coffin and for her brother to wish him strength. And then I went home, to pack for my holiday. But I did not feel like holiday at all'.*

Joe's social worker also talked about her last time with Joe:

> *'We always had a special bond. I was really fond of Joe. He was really one of my special clients. <> (in tears) In the team, we decided to care for him ourselves. He wanted to be with us. It felt like family. Caring for him at the end of his life was hard, but rewarding. <> I tried to keep in mind that he was my client, to help me cope. But I didn't let him notice that, I was very involved with him. I didn't want him to be in any distress.*

Like Eleanor's and Joe's social workers, many care staff members struggled to find ways to provide warm, tender end-of-life care but at the same time keep an emotional distance. In this process, most care staff members needed the emotional support of their colleagues and superiors. This period could be particularly intense for care staff members who were providing end-of-life care for the first time. A lack of experience could make care staff insecure and anxious about what was going to happen. Reflection was identified as a value for dealing with the interweaving of emotional and professional involvement; being able to reflect on your own emotions, capabilities and caring relationship helped care staff to find a balance between warm care and emotional distance.

### Symptom relief: stronger reliance on the joint interpretation of signals

Symptom relief became very important during end-of- life care. In particular it was often a challenge to identify pain, anxiety and other signals of distress in people with severe ID. Often these symptoms only became visible through small changes in behavior or signals such as grimaces or stretching the neck. These changes were not easily recognized. This meant that physicians now relied heavily on information from people closely involved with the person, mostly relatives and care staff. The case of Joe is an example of this:

> Joe's ID physician: *'It took a while before we had the idea that Joe was indicating pain. <> The idea was that Joe would not have to suffer in any case. <> My role as a physician was mainly on demand. It's the care staff who know him best, they pick up signals such as pain. Then they call me. <> But the identification of pain is*

> *largely done by the care staff and the family. I think in Joe's case it was his sister who first said to me he's in pain now. And over time everyone agreed that he was in pain. <> You could tell that Joe had pain by his moaning and grabbing. He used to clutch his chest or neck. And make groaning noises. And he would wince. He was also very affectionate towards his care staff and to his mother or anyone who was familiar for him at that time. Whether that expressed real pain I don't know but I felt he was deteriorating. He sought more proximity and couldn't be left alone.'*

Although many physicians were already used to incorporating the views of others in their regular care, the reliance on third-party information became more pronounced in end-of-life care. Symptom relief became a real team effort. The joint interpretation of signals could be a hurdle as relatives, care staff and physicians did not always proactively seek contact and did not always find a common language to talk about what they saw. Two values were essential in this shift: 1) 'attentiveness' to the person's signals and expressions of needs, distress and pain, often best done by permanent care staff and relatives who had a life-long relationship with the person, and 2) 'openness to cooperation and building a shared understanding' of the interpretation of signals and expressions.

### Magnified dependency and responsibility in medical decisions

In most of the cases studied, many medical decisions needed to be made during the end-of-life care, for example about life-prolonging treatments, tube feeding, providing oxygen or pain medication by infusion pump. In most cases, the role of the people with ID themselves was unclear in the decision-making process, especially for people with more severe ID. Physicians usually discussed medical information with relatives and mentors, and tried to come to shared decisions with them. Relatives and mentors often felt overwhelmed by the dependency of the person with ID, which seemed magnified during end-of-life care, in particular concerning decisions about prolonging treatment, tube feeding or palliative sedation. The case of Josh is an example:

> Josh's physician and mentor are discussing whether they should place a tube to facilitate Josh's feeding. Josh would need to go to the hospital to have the tube placed. Josh's mentor: *'I said I don't want him to go to the hospital to get the tube. If Josh is in the hospital he gets very scared by all those white coats, you wouldn't make him happy by doing that. <> So the doctor told me they may be*

> *able to do it in the ID care center. But then again, the tube would only prolong his life a little bit. Would that be right? I wanted to know, am I doing him harm if he doesn't get the tube? Will he starve to death and feel awful? The doctor said that that was not going to happen, he gave me the confirmation that they would relieve his suffering. That was what I needed to hear. I wanted to be able to look at myself in the mirror and tell myself that I did not do anything very burdensome to him at the end of his life.'*

Like Josh's mentor, many relatives were insecure, often because their sick relative was unable to adequately indicate what his or her wishes were. They had to deal with the emotions of feeling highly responsible, and sometimes even felt they were deciding about the other person's life and death. Four values were behind this shift: 1) 'responsibility' for taking joint decisions in the best interests of the person, 2) 'attentiveness' to the person's wishes, 3) relatives' 'reflection' on their own emotions, and 4) 'openness to cooperation and making shared decisions'.

### Growing awareness of having two 'families'

In most cases, the imminence of the end of life led to an intensified caring relationship between relatives and care staff. Relatives visited more often, became more involved in the caring process and sometimes even gave care jointly with care staff. Some relatives and care staff were easily able to coordinate their activities and make clear agreements, aiming for the same goal of being there and providing the best possible warm, comforting care for the sick person. In these cases, relatives were often full of praise for the support they received from the professionals in the ID care service. However, it was not always easy for relatives to deal with the professionals during this emotional period. In some cases, cooperation did not run smoothly and relatives and care staff had different perceptions of what good end-of-life care entailed. In one case, the relative experienced the close involvement and advice of the care staff member as an infringement of her privacy and her rights as a representative. Other relatives struggled initially in cooperating with the care staff, but eventually found ways to properly establish shared care. As Joe's sister declared: 'I came to realize he had "two families"':

> Joe's sister: *'I drove back from my holiday because I had received a call that Joe was rapidly becoming weaker <>. I thought, gosh, if he dies, then there will only be six of us at his funeral. Well, we all love him, not a lot of people need to be*

> *there. <> And while driving I thought: But actually his family is much bigger than our family. Joe has two families. Our family (the relatives) sees him as a man with an intellectual disability, but at his home at the ID care service he has another family who see him as Joe, as a person with a sense of humor and jokes. They (the care staff) talk in a very different way to him than we did. That was good to realize.<> After Joe died, it felt like we (the relatives and care staff) were really one family, all care was finished, and we were there for each other.'*

'Openness to cooperation and sharing' the care was an important value to accompany the intensified presence of two 'families'.

## Discussion

### Specifics of end-of-life care for people with ID

Although some of the shifts found in this study may also apply to other client groups and care settings, several aspects seem particularly relevant for the end-of-life care for people with ID. First, ID care staff, more than professionals in other healthcare sectors, are trained to activate their client and are used to doing this. At the end of life, their usual strong focus on improving the client's quality of life by maximizing the activation and participation of people with ID in society has to change to a strong focus on the quality of life through comfort care, taking over tasks and good symptom relief. Second, the increasing focus on the identification and relief of symptoms is particularly challenging as verbal communication with a person with ID is often hampered. Hence, professionals and relative rely strongly on their joint interpretation of signals of distress in the person with ID. This can be problematic as ID care staff, and in particular social workers, are known to have a shortage of knowledge and skills regarding e.g. the use of instruments to measure pain or other symptoms at the end of life (Bekkema *et al.* 2014a). Third, another typical factor is that decision making about, for instance, whether to continue or forgo life-prolonging treatments at the end of life is particularly complex because people with ID often have difficulties with clearly expressing personal care needs or wishes. Relatives feel highly responsible as a 'proxy', and feel that their relative with ID becomes increasingly dependent on them. Fourth, the fact that both the family and the care staff at the ID care service are often deeply emotionally involved over a long period of time is quite specific to the end-of- life care for people with ID. Among care staff, the interweaving of emotional and professional involvement often leads to struggles in trying to achieve a balance between warm care and professional distance.

### Comparison with existing literature

The impact on relatives of 'having to let go' and being overwhelmed by the feeling of being responsible for 'deciding for somebody else' has been described earlier in papers, for example in our and other studies on end-of- life care for people with ID (Bekkema *et al.* 2014c; Wagemans *et al.* 2013), and also in papers on children with cancer (Kars *et al.* 2011). Care staff, the other 'family' in our study, experienced a rise in emotions when they had to let go of a client they had often taken care of for a long period of time. Similar emotions during the end-of-life care among ID care staff were also found by Wiese (Wiese *et al.* 2012). These 'family-like' relationships between care staff and clients have also been observed in other settings, such as dementia care (Livingston *et al.* 2012). In a nursing home study, care staff pinpointed the loss of the close attachment to their client as the biggest challenge to overcome in the transition to end-of-life care (Waldrop & Nyquist 2013). Yet for care staff in ID care services 'letting go' might be even more intense, as providing end-of-life care is not regular care for them. Generally, ID care staff serve clients of all ages. Indeed, many care staff in our study (in particular inexperienced staff) struggled to channel their feelings in order to foster professionalism and secure the provision of tender end-of-life care. These struggles may jeopardize the caring relationships between care staff and clients. Wagemans (2013) also found such struggles and described them as 'balancing involvement and distance'. The ID care staff in her study suggested that keeping too much distance might hamper good care. The struggles seem to reveal an inner conflict within ID care staff members, which calls for more attention than is currently given in ID care services.

Having 'two families' (relatives and care staff) come together and jointly provide care for you as a dying person with ID can lead to warm, loving care in the proximity of the very people who are most important to you and who know you and your needs best. Yet, if perceptions of what constitutes good care differ and communication fails, the good intentions to provide joint care may turn into a burden and source of distress for the dying person. Problems in the joint provision of end-of-life care between relatives and care staff are not unique to ID care: they were also found in nursing homes and care homes (Livingston *et al.* 2012; Thompson *et al.* 2012; Gjerberg *et al.* 2011). A difference is that people with ID have generally lived much longer in their care setting than people in care homes or nursing homes. Relationships between relatives and ID care services often go back decades, making the comparison of 'two families' much stronger. Moreover, in contrast to most nursing homes, ID care staff often lack expertise and experience in end-of-life care (Bekkema *et al.* 2014a;Ryan *et al.* 2010; Botsford 2004), which puts them more on a level with relatives in terms of end-of-life care expertise. Care staff may be just as insecure about what to do as relatives. This can put existing care relationships between relatives and care staff under pressure and may jeopardize good end-of-life care. ID care services should pay timely attention to this relationship, and to the expertise of their staff.

### Values underlying the shifts at the end of life of people with ID

Six values were behind the shifts in end-of-life care for people with ID: 1) 'Being there' and making extra time available to be close to the sick person. Being there was a prerequisite for the second value. 2) 'being responsive' to the person's real needs and possibilities. Being there and being responsive both refer to the acknowledgment of what the person is capable of and acting on both the durable and the changing capabilities. 'Being there' has been recognized as an important theme in other end-of- life care studies, e.g. of people with ID (Todd 2013) and children with cancer (Kars *et al.* 2008). 'Reflection' on your own emotions, caring capabilities and the relationship with the person with ID. Reflection proved particularly helpful in achieving a balance between the emotional involvement and professional distance of care staff, and in enabling relatives to channel their emotions when confronted with medical decisions. 4) 'Attentiveness' to the person's needs and wishes and expressions of distress and pain. Attentiveness is generally found to be important for end-of-life care. E.g. Leget stressed the importance of listening attentively to dying patients in order to unravel their real needs and questions ('inner space'; (Leget 2007). Unravelling the real needs and distress can be extremely complex in people who have ID, and yet is therefore so very essential. In particular, people with severe ID rely heavily on deep, long-lasting relationships with close caregivers. 5) 'Responsibility' for taking joint decisions in the best interests of the person with ID, particularly evident among relatives who felt a great sense of responsibility for taking the right decision. 6) Finally, 'openness to cooperation and sharing' comprised the intensified cooperation among professionals and between relatives and professionals in providing good end-of-life care: e.g. being able to communicate adequately about the person's signals and needs, the openness required to take joint medical decisions and the openness of the 'two families' to enable the joint provision of care. The six aforementioned values are all highly relational. They are associated with a care ethics perspective, which recognizes that all care is relational (Tronto 1993). Care for people with ID inevitably builds on relationships, as helping people to participate and get a job, for instance, requires caregivers to have many relational qualities. At the end of life, the value of good caring relationships seems to increase. Three caring relationships need to be fostered: the relationship with the person with ID, relationships among professionals and the relationship between relatives and professionals.

### Strengths and weaknesses

We were able to reconstruct the story of twelve deceased people with ID by using a multi perspective design, incorporating the viewpoints of all the people most closely involved. A limitation is that we conducted this study retrospectively, so the experiences of the interviewees may have been subject to a recall bias. On the other hand, retrospective studies make people's statements less susceptible to day-to-day emotions.

This study did not explore shifts in the end-of-life care approaches to people without ID. Future research could reveal the extent to which the shifts found in this study match shifts in care approaches among other client groups in long-term care settings, such as people with dementia and residents in nursing homes and elderly care homes.

## Conclusion

As the death of a person with ID comes near, several shifts are perceived: care staff and relatives have to let go of their usual care strategy aimed at activation; the interweaving of emotional and professional involvement becomes a challenge for care staff; the joint interpretation of signals expressing distress becomes increasingly important; the dependency of the person with ID rises in the eyes of relatives (their 'proxy' decision makers), in particular with regard to medical decisions; and at the end of life it becomes increasingly evident that the ID care staff have also become a 'family' for the client. Highly relational values are behind these shifts: 'being there' for the person with ID, 'being responsive' to the person's needs, 'reflection' on their own emotions and caring relationships, 'attentiveness' to the ID person's wishes and expressions of distress, 'responsibility' for taking joint decisions in the best interests of the person, and 'openness to cooperation and sharing' the care with others.

### Practice implications

End-of-life care for people with ID requires a different care approach and attitude to participation-focused care. It requires an allowance for decreasing expectations as regards activities and skill acquirement, and an increase in teamwork featuring intensified comforting care, symptom management and medical decision making. Cooperation among close caregivers is fundamental, as is emotional support for care staff. As more and more people with ID will need end-of-life care in the future, ID care services should be better prepared. With regard to supportive care for carers, services should invest particularly in three areas. First, sufficient emotional support should be given for care staff who struggle to find a balance between their emotional involvement and professional distance. Informal support from colleagues alone may not be sufficient. These inner conflicts require professional support sessions, including time for intrapersonal and interpersonal reflection. Secondly, improving care staff's level of expertise in end-of-life care is necessary, as care staff can be inexperienced and insecure in dealing with both clients and relatives. Some end-of-life care training interventions for ID care staff have already been developed (e.g. (Reddall 2010; Hahn & Cadogan 2011; McLaughlin *et al.* 2012; McLaughlin *et al.* 2014). Thirdly, the relationship between relatives and care staff needs attention, as good cooperation is essential for the dying person with ID. Moreover, relatives need the support of professionals as they go through the process of letting go and have to make difficult decisions. Care staff need to learn how to build strong, good working relationships with relatives, preferably at a much earlier stage in the lives of people with ID as emotions usually run high when death is imminent.

### Competing interests

The authors declare that they have no competing interests.

### Authors' contributions

AV and AF wrote the research proposal and contributed to the design of the study, the analyses, the interpretation of data and the critical revision of the paper. NB contributed to the design of the study, conducted and analyzed the interviews and drafted the manuscript. CH commented on the design of the study and contributed to the analyses, the interpretation of data and the critical revision of the paper. All authors read and approved the final manuscript.

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# 3

# Perspectives of people with mild intellectual disabilities on care relationships at the end of life. A group interview study.

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## Abstract

### Background

Care relationships are crucial in tailoring the end-of-life care of a person with intellectual disabilities (ID) to their needs. Yet, almost all studies on end-of-life care for people with ID have been conducted among caregivers. The views of people with ID about care relationships at the end of life have not been a specific focus of research.

### Aim

To explore relevant dimensions of the caring relationships in end-of-life care from the perspectives of people with mild ID in the Netherlands.

### Design

Group interviews were conducted using nominal group technique. Interviews were inductively analysed by two researchers.

### Setting/participants

Seven groups participated (33 people). Inclusion criteria: having mild ID, being able to decide about participation and give informed consent, not receiving end-of-life care. All groups were interviewed twice.

### Results

Two dimensions of care relationships were found: 1) 'Ascertain, record and honour wishes' of the ill person. Adequately dealing with care wishes, 'last wishes' and funeral wishes was of central importance: 'it's about their life'. We found an emphasis on control that seemed to reflect the participants' experience that respecting autonomy does not always happen. 2) 'Being there': the ill person needs people who are there for him/her, practically, as well as emotionally, socially and spiritually. Participants placed specific emphasis on providing positive experiences.

### Conclusions

The views of people with mild ID highlight the high demands which end-of-life care imposes on relational qualities of caregivers. This study shows the added value and potential of involving people with ID in studies on end-of-life care.

**Keywords:** Palliative Care, Intellectual Disability, Professional-Patient Relations, Family Relations, Focus Groups

### What is already known about the topic?

- Care relationships are crucial in tailoring the end-of-life care of people with intellectual disabilities (ID) to their needs.
- Almost all studies on end-of-life care for people with ID have been conducted among care professionals or relatives.
- The views of people with ID themselves about care relationships at the end of life have not been explored.

### What this paper adds.

- According to people with mild ID, ascertaining, recording and honouring wishes is a central dimension of care relationships at the end of life.
- The prior experiences of people with mild ID highlight the fact that respecting their autonomy does not happen as a matter of course.
- Terminally ill people with ID need people who are there, in a practical, emotional, social, and spiritual sense.
- Creating opportunities and positive experiences in end-of-life care is important for people with mild ID.

### Implications for practice, theory or policy.

- End-of-life care places high demands on the relational qualities of caregivers, such as being able to ascertain the ill person’s wishes, console them and empathise with them.
- People with mild ID are able to discuss ideas about end-of-life care and should be involved more systematically in research and practice development.

## Introduction

Individuals with ID are characterised by significant limitations, from before the age of 18, in both intellectual functioning and adaptive behavior (Schalock 2014) Many people with ID rely on others for care and support. People with ID form a diverse and special group of care recipients in end-of-life care, for example because of their limited level of understanding and the challenges of communicating with them (Bekkema *et al.* 2014a; Tuffrey-Wijne *et al.* 2009; Stein 2008). End-of-life care for people with ID needs improvement as palliative care needs are often not recognised until late (Heslop *et al.* 2013), cooperation between palliative care services and ID care services is often suboptimal, and many care professionals in ID care lack expertise in end-of-life care (Ng & Li 2003; Botsford 2009; Ryan *et al.* 2010; McCarron *et al.* 2010; Tuffrey-Wijne *et al.* 2010; Bekkema *et al.* 2014b; Dunckley & Sales 2014). Moreover, people with ID are not always involved in communication about their illness and decision making (Tuffrey-Wijne *et al.* 2009 & 2010; Wagemans *et al.* 2010; Wagemans *et al.* 2013; Bekkema *et al.* 2014c & 2014d). This poses the risk that end-of-life care is not adequately adapted to their needs.

Caring and trusting relationships contribute to a better quality of life for patients in need of end-of-life care (Mok & Chiu 2004). As many people with ID become even more dependent on others due to the combination of existing cognitive impairments and physical impairments resulting from a life-limiting illness, good caring relationships are of paramount importance for terminally ill people with ID. Several studies among care professionals and relatives have shown the importance of relational aspects in end-of-life care for people with ID, such as being responsive to the person's needs, having a trusting relationship, and giving them space to show their wishes and preferences (Bekkema *et al.* 2014a & 2015a; Ryan *et al.* 2014; Wiese *et al.* 2012; Todd 2013).

Almost all scientific studies on end-of-life care for people with ID have been conducted among care professionals or relatives (e.g. Bekkema *et al.* 2014a & 2014b; Ryan *et al.* 2010; McCarron *et al.* 2010; Wagemans *et al.* 2013; Wiese *et al.* 2012). Studies exploring the views of people with ID about end-of-life care are rare. Among the few exceptions is the work of Tuffrey-Wijne, who used an observational approach to study the experiences and needs of people with ID who had cancer. She found e.g. a lack of power to make decisions among people with ID, and a lack of adequate information about their illness (Tuffrey-Wijne *et al.* 2006, 2009 & 2010). Tuffrey-Wijne also tested and adapted the nominal group technique, where a single question is used to generate, record, discuss and rank ideas, to elicit the views of people with ID on the provision of end-of-life care (Tuffrey-Wijne *et al.* 2007). Another relevant study was performed by McLaughlin, who held focus group interviews with people with ID about end-of-life care with the aim of using the findings to improve education for professionals (McLaughlin *et al.* 2014a). These studies demonstrate that people with ID are indeed capable of participating in studies on end-of-life care. But although their findings did include some relational elements, such as 'having family and friends around', they did not focus specifically on care relationships in end-of-life care.

It is evident that end-of-life care should be tailored to the person with ID's needs, and that care relationships are crucial in achieving this goal. So far, it is not known which dimensions of the care relationship people with ID themselves find important for good end-of-life care. People with ID may have different perspectives to care professionals and relatives, or place different emphases, on how people should take care of the incurably ill. Hence, the aim of this study is to explore the relevant dimensions of the care relationship in end-of-life care from the perspectives of people with mild ID.

## Methods

### Design

Seven groups of people with mild ID were interviewed twice, using qualitative group interviews. An adapted nominal group technique was used to structure the discussion. For this purpose, we further adapted the technique as used by Tuffrey-Wijne (Tuffrey-Wijne *et al.* 2007) to suit our specific goal and piloted it successfully (Bekkema *et al.* 2014c & 2014d).

### Participants

This study was conducted in the Netherlands in 2013 to 2014. Thirty-three people participated. Inclusion criteria were having a mild ID, being able to decide about participation and give informed consent, and not receiving end-of-life care. Although the inclusion criterion about not receiving end-of-life care was mentioned in the information letter for care professionals, it became clear during an interview that one participant had recently been diagnosed with incurable cancer. At his own request, he continued to join the discussions, which he enjoyed. Careful precautions were taken to ensure that the discussions were not stressful for him or the other participants.

Six groups were recruited through six ID care services, and one group was recruited through a theatre company. To provide a safe environment, group sizes were kept small and interviews were held at a familiar location. All participants within a group knew one other. One group consisted of actors in a theatre company, three groups of members of the local client council, and three groups of co-residents. All participants received support from an ID care service. Most participants had (voluntary) work and/or hobbies.

**Table 1** participants' background details

| | No. of participants | Age (range) | Male | Female |
|---|---|---|---|---|
| Group 1 | 5 | 29-71 | 4 | 1 |
| Group 2 | 7 | 62-82 | 3 | 4 |
| Group 3 | 4 | 45-73 | 4 | - |
| Group 4 | 3 | 54-68 | 3 | - |
| Group 5 | 5 | 31-84 | 1 | 4 |
| Group 6 | 4 | 39-65 | 3 | 1 |
| Group 7 | 5 | 21-63 | 2 | 3 |
| **Total** | **33** | **Mean age: 58** | **20** | **13** |

Ages ranged from 21 to 84; the mean age was 58 and 76% of the participants were over 50. 61 % were men.

## Data collection

The number of groups was not predetermined. In accordance with qualitative principles, data collection and analysis were conducted in a cyclical process. Data saturation - the point at which researchers did not observe any new information or themes - was attained after seven groups had been interviewed. In total, fourteen group interviews of approximately one hour took place. NB facilitated twelve interview discussions and NB and AdV jointly facilitated two interviews. Field notes were written after each interview. A familiar care professional was present during all interviews to help create a safe environment. We applied nominal group technique as follows:

1) During the first interview, a short picture story was told about a woman with ID who heard from her doctor that she was incurably ill. Participants were asked the following question: what can people do to take good care of her? The interviewer explained that all sorts of ideas were welcome and equally important. Participants were first asked to think for themselves and were then invited to share their thoughts. Ideas were discussed in the group and the interviewer encouraged the participants to come up with more ideas. All ideas were written down. Once no more new ideas emerged, the ideas were summarised. Sometimes one or two new ideas were added. Finally, the first interview was evaluated and brought to a close.

2) After two to three weeks, the second interview took place. In the meantime, the interviewer had ordered the ideas of the group, and had made a card illustrating each idea. The number of ideas per group varied from 8 to 13. The goal of the second interview was three-fold: to validate the results of the first interview, add any in-depth insights and rank the ideas. The ideas were presented and participants were asked if they recognised the ideas. They were invited to add insights and further explain their ideas. Each participant then received a set of cards and was asked to divide the ideas into two piles, one for the 'best' ideas and one for other ideas - until four best ideas were left over. Some participants needed assistance in this selection process. Participants then voted using ballot boxes; the top best idea received four points, the next best idea three etc. Votes were counted and the three ideas with the most points were announced to the group. Finally, the second interview was evaluated and brought to a close. Participants' reactions to the interviews were very positive in all groups.

Aftercare was discussed with the care professional to ensure that support was available. Participants were told who they could turn to within the care service if they needed emotional support, and were given the contact details of the researcher if they wished to talk to the researcher. All participants received a feedback report listing the ideas, the outcome of the voting and the interviewer's contact details.

## Analysis

Interviews were audiotaped and transcribed verbatim. Data were analysed inductively, using thematic analysis (Braun & Clarke 2006). Important elements of thematic analysis are familiarizing with the data, generating initial codes, searching for themes, reviewing and defining themes and writing down the results. Thematic analysis was applied as follows: interviews were re-read and codes were ascribed to those text fragments potentially reflecting dimensions of care relationships. Descriptive codes were used directly based on interviewees' wordings (such as ask what she wants to eat), as well as interpretative codes (such as providing consolation). Interview fragments with the same codes were constantly compared. To ensure reliability, all interviews were coded independently by the first author and by one of the co-authors. Correspondence between interpretations and the original interviews was continuously verified, and differences in analysis and interpretations were discussed. After coding all transcripts, potential themes were formulated. These themes were then reviewed in relation to the coded text fragments and transcripts. Finally, themes were refined and written down. MAXQDA 2011 was used to facilitate the analysis (www.MAXQDA.com).

The quantitative outcomes of the voting gave direction to the prioritising of ideas, but were not part of the qualitative analyses. The voting outcomes have also been used to compile a practical book about end-of-life care for people with ID (Bekkema *et al*. 2015b).

### Ethical consideration

The protocol and topic list for conducting the group interviews were approved by the Medical Ethical Committee of the VU University Medical Centre. Prior to the interviews, all participants received a pictorial information letter informing them of the aim and course of the study, as well as an accessible informed consent form. The information letter explained that study participation was voluntary. If needed, a care professional helped the participants with reading the information. The study goal and procedure were repeated orally before the start of the interview. All participants gave written informed consent. The transcripts of the interviews were made anonymous and non-traceable to individuals.

## Results

Two main dimensions of care relationships in end-of-life care could be identified from the interviews with people with ID: ascertaining, recording and honouring the wishes of the ill person and being there to provide practical, as well as emotional, social and spiritual support (figure 1). The dimensions are explained below, illustrated by participants' quotes (names used are not the participants' real names).

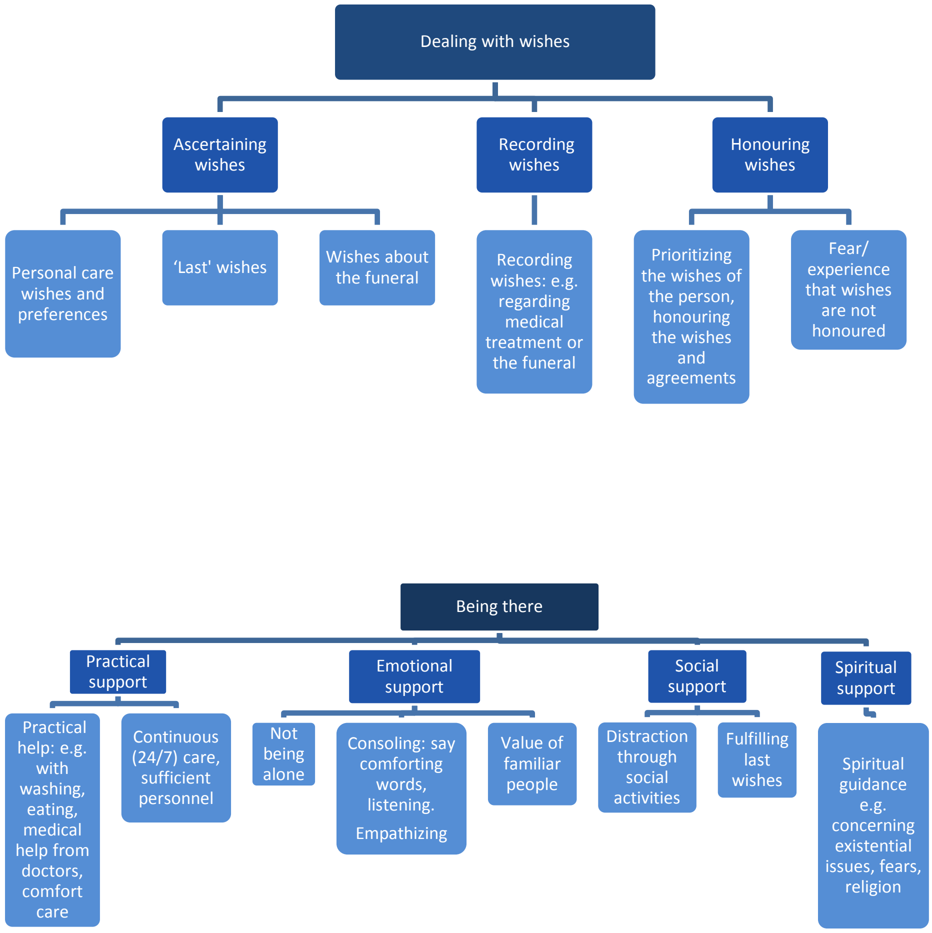

**Figure 1** main dimensions of care relationships in end-of-life care

## Ascertain, record and honour wishes

### *Ascertaining wishes*

Participants indicated that ascertaining the incurably ill person's personal care wishes and preferences is very important. Care professionals and relatives should find out what the person's practical care wishes are, such as which food they like or if they would like television or music in their room. The same applies to wishes regarding place of care: either home or another setting, and the person's medical care preferences, e.g. regarding pain medication or medical treatments. Knowing these wishes helps ensure that the person feels as good as possible; as one respondent put it: 'it is about their life'. Talking to the person and asking what he or she wants was mentioned as a way of ascertaining their care wishes. Or, if he or she is unable to talk, having preferably familiar people closely observe their behaviour or facial expressions.

> Paula: *'They should ask: "Can I get you anything? What can I do for you? Can I do some grocery shopping for you? Do you want to go outside? In a wheelchair, or…?"'* (I6R1)
>
> Bert: *(If the person is unable to talk):'I think the people who know her, her family and friends, the ones who were always around her, looked after her and sympathised with her, they should be able to see from her behaviour what her wishes are.'* (I1R3)

Participants emphasised that the last phase in life should also bring some joy and peace. The person's 'last wishes' should therefore be ascertained, by e.g. talking about what trips they would still like to make, or what unfulfilled dreams they have. The ill person may want to go on holiday to somewhere sunny, go on a boat trip, or go back to a favourite holiday destination. Participants stressed that knowing last wishes is important as these would be the last nice things that a dying person could do; it may provide satisfaction and peace.

> Mick: *'There are 'wish' ambulances (ambulances that take incurably ill people on a trip under medical supervision). You can choose where you want to go. Going to the beach for example. Doing something you like. The last thing you can do, as it were. I really like that idea.'* (I2R1)
>
> Isaac: *'You want the person to stay cheerful (…) Maybe you can talk to her about her wishes. Maybe she wants to meet her favourite pop singer. (…). I find these things very important. That she is not only sad, but also has some joy.'* (I7R4)

Furthermore, participants indicated that ascertaining the person’s wishes about the funeral is important as well. Many participants had attended funerals and had already thought about their own wishes regarding the burial or cremation, funeral service and what should happen to their personal belongings. Some participants even advocated that a person should be able to choose his/her own moment of death. Yet, they were concerned at the same time that this was not a realistic option.

> Marcel: *‘Nowadays they have donor cards, but you can’t say: I had a good life, I can’t get better and I don’t want to live anymore. I think that’s a pity.’* (I5R4)

> Carol: *‘It is important that you tell others what you want, your last wishes. (…) That the funeral service is done by a vicar. And that the co-residents can say goodbye. You can say what kind of service you want. A cheerful one or a sad one. (…) what I would like is being carried away by a horse and carriage. (…) just like my Dad. (…) my personal belongings should go to charity, to homeless people. I want to decide about this myself.’* (I4R1)

### Recording wishes

The discussions about wishes also seemed to express a deep desire that the person with ID should be in control. Participants stressed that wishes should not only be ascertained but also be recorded. This applied to wishes regarding medical treatments and even more to funeral wishes. Early recording of wishes, ´on paper´ or on the computer, was acknowledged as a way of ensuring that the wishes were known and honoured, including after death. Several participants had filled in a ‘wish book’, a book regularly used in Dutch ID care to write down someone’s preferences for the funeral.

> Leo: (about having a wish book). *‘Well, if people die, they have a wish: being buried or cremated. They can fill in a wish book.’* (I7R1)

> Aron: (about the person’s wishes regarding medical treatment) *‘It could be that you can’t speak anymore at the end, but if you have it on paper, then your doctor would know and then they wouldn’t make a wrong decision afterwards.’* (I5R3)

### Honouring wishes

Yet ascertaining and recording wishes does not automatically mean that they are honoured, a point made vigorously by some participants. Some referred to times in their life when their wishes had not been honoured, or expressed a general distrust regarding the honouring of the wishes of people with ID.

> Bert: *'If you put your wishes on the computer, are you still in control?' Maria: 'I put everything on paper: in the Catholic Church and then cremated. I wrote that down on the computer in the office.' Bert: 'How are you so sure they will look there when you're dead?' Maria: 'Because they will look in the computer.' Bert: 'But you don't know that. You'll be dead. (…)' Bert: 'People don't want us to think for ourselves and make decisions. They want to arrange everything themselves, they think: we know best, and we will decide. (…) that's their attitude. (…).' Maria: 'Anne (the woman in the pictorial story) should know for herself what she wants. Not a care staff member. (..) We have a right to tell them our wishes and say yes or no. A care staff member shouldn't decide. If they tell me: you should be buried, I'll tell them: no, I don't want that. I want to be cremated. That is my choice.'* (I1R3 & I1R4)

According to the participants, the most important aspect of dealing with wishes in the care relationship was that people should make sure that the person with ID is in control, that their wishes take priority and are actually honoured.

> Aron: *'It is important to do what was agreed. Else the care staff will fill in things for her, and not stick to the agreements they made. (….) What the person wants is the most important thing.' (Interviewer: so what the person wants should get priority?) Aron: 'If possible and after agreeing things. (….) if it isn't possible, they should say that honestly, and explain why it isn't possible.' (*I5R3)

> Marcel: *'You should listen to her wishes. Meet her wishes. It is about her. What she wants. And how she wants it. That's the last thing you can do for her.'* (I5R4)

## Being there

### *Being there in a practical sense*

Participants emphasised that when a person is incurably ill, (s)he needs care professionals or relatives to be there for practical help, and doctors for medical help. This help is necessary as people may be very ill and unable to take care of themselves, for example with regard to getting out of bed, washing, eating and drinking, and feeling comfortable. Participants also stressed the importance of continuous care and the availability of sufficient personnel. Regular physical presence is important, as well as 24/7 availability, e.g. via an alarm system. Noteworthy, several participants had already contemplated whether they would be able to stay in their residence if they became ill. Some suggested that there would be insufficient staff where they lived.

> Theo: *'Somebody should be there. Someone who can help her. (…) I don't know if they have 24/7 care where she lives. That would be good. Good help.'* (I5R1)

> Joe: *'She should have an alarm bell. So she can call. And then they can come right away.'* (I6R2)

### *Being there in an emotional sense*

Participants associated 'being there' with emotional support as well. It meant not letting the person be alone and letting them know you are there to help them deal with their situation. Participants stressed that the person should be encouraged and consoled, e.g. by saying comforting words, providing a listening ear and by empathising with their situation. This may help them calm down and not feel too sad. Familiar people who know and understand the person -in particular relatives and friends- were the best people to provide emotional support.

> Aron: *'You should visit, so she knows she isn't facing this alone.'* (I5R3)

> Carol: *'Say comforting words. That she doesn't need to be afraid. (….). Tell her that it will be all right. That we are thinking about her.'* (I4R1)

*Being there in a social sense*

Participants also strongly related 'being there' to social support. They described this in two different ways. The first way was instrumental: others should try to take the person's mind off his/her difficult situation so that (s)he is not always thinking about the disease; e.g. by paying him/her a visit, reminiscing about shared memories, watching television, playing games together or getting some fresh air. The second way related to fulfilling the person's last wishes. The person should be enabled to still experience fun and pleasure; e.g. by accompanying the person on a day out. Both approaches were directed at interacting with the person and helping him/her feel better and have more positive thoughts. Friends, co-residents and relatives were the best people to provide this social support.

> Paula: *'People should visit. The time will go quicker if someone is there, then you don't need to think about it.'* (I6R1)

> Mick: '*Take somebody outside if the weather is nice, and go for a walk. Try to make them think of other things, distract them. When my sister was ill, we took her on a trip in a wheelchair. On a boat with music. She really enjoyed that. That's something you could do for somebody, if it's still possible.'* (I2R1)

*Being there in a spiritual sense*

Participants also talked about the value of the support of spiritual caregivers, such as pastors, chaplains or general counsellors. Several participants said that spiritual caregivers are unable to cure you but they can provide spiritual guidance, bring peace and reduce fear. Some examples of how they could do this were looking back on your life, talking about fears, existential issues or religion and preparing the person for the terminal phase and imminent death.

> Marcel: *'You could talk to the pastor about how you want to be buried. (…) He could help you prepare for your death.' Aron: 'And also bring up the nice things, the nice things that happened.'* (I5R4 & I5R3)

> Bert: *'If you're about to die, then it may help to hear about other people's experiences, or near-death-experiences of other people. (…). Because many people are afraid. What will happen to you when you die (….). If you talk about*

> *that, than you know that death is not something to be afraid of. That could be a personal victory.' (*I1R3)

## Discussion

In this study seven groups of people with mild ID were interviewed twice. Two main dimensions of care relationships were found: 1) *'Ascertaining, recording and honouring wishes*' and 2) '*Being there'*. The first dimension is based on the view that adequately dealing with a person's wishes is of central importance in providing good end-of-life care: it is about their life. In caring for the person, their care wishes, last wishes and funeral wishes should not only be ascertained but also recorded and honoured. The second dimension is based on the participants' opinion that an ill person needs dedicated people who are there for him/her at the end of life. This support can be practical (providing practical and medical help), emotional (not letting the person feel alone and helping them deal with their situation), social (taking the person's mind off their situation and accompanying them in the fulfilment of their 'last wishes'), and spiritual (providing spiritual guidance, bringing peace and reducing fear).

### Comparison with other studies

'It is about what she wants and how she wants it': this statement reflects people with ID's desire that properly handling their wishes should be at the heart of caring relationships at the end of life. This is in line with the Caring at the End of Life position statement by the American Association on Intellectual and Developmental Disabilities (aaidd.org/news-policy/policy/position-statements/caring-at-the-end-of-life), which sees honouring wishes and autonomy as an important principle. Autonomy is also a major principle in end-of-life care within the general population (e.g. Proot *et al*. 2004; Claessen *et al*. 2011). Yet what seems different here is the emphasis on control, which seems influenced by the prior experiences of the participants with ID. This was reflected in the fervent way in which participants discussed honouring wishes, their emphasis on advance directives, and in the distrust, scepticism and fear of some regarding whether their wishes would be honoured. Thus, respecting autonomy of people with ID does not seem to happen as a matter of course. This shows how crucial trusting caring relationships throughout life are for acknowledging and respecting the wishes of someone with

ID, including at the end of life. This supports the view that autonomy is best seen as a relational concept (Bekkema *et al*. 2014a; Tronto 1993) and that adequately dealing with people with ID's wishes requires continuous attention.

Being there, the other important dimension, was also felt to be important by caregivers of people with ID (Todd 2013; Bekkema *et al*. 2015a). By referring to practical support as well as emotional, social and spiritual support, people with ID showed that they believe that dying people with ID should be treated as whole persons. This is in line with international definitions of end-of-life care and palliative care (WHO 2015). Noteworthy was the strong emphasis participants placed on being able to 'live life'. Ill people should have their mind taken off their situation, have positive experiences and be enabled to fulfil their last wishes. This focus on 'life' – which is occasionally highlighted in studies among the general population (Cannaerts *et al*. 2004) – was not found so clearly in research among care professionals and relatives. Hence, this calls for more awareness of the need to create opportunities and positive experiences in end-of-life care for people with ID.

### Strengths and weaknesses

To our knowledge, this is the first study exploring the views of people with ID on care relationships in end-of-life care. The relevant dimensions add new insights to the few other studies in this area, e.g. on cancer experiences of people with ID (Tuffrey-Wijne *et al*. 2006, 2009 & 2010). Their specific emphases, e.g. the stress they put on honouring wishes and their fear that this might not happen, and their focus on positive experiences, demonstrate the added value of interviewing people with ID themselves, in addition to studies among caregivers and the general population. The nominal group technique elements gave the discussion direction, and structured the second meeting where ideas were further explored and validated. A clear explanation of the study, the presence of familiar care professionals, the use of a pictorial story, and holding two separate meetings also seemed to contribute to its success. A limitation of this study is that all except one participant were not receiving end-of-life care. As such, their views may differ from people who are receiving end-of-life care. Another limitation is that we only included people with a mild ID. Further research is needed to determine whether the nominal group technique –with some modifications- could be used for people with moderate ID. Including the perspectives of people with severe ID is complicated, yet of vital importance. Other methods, such as participant observations, may help to capture their views (Tuffrey-Wijne *et al*. 2009 & 2010).

### Conclusion and implications

The aim of the study was to explore the relevant dimensions of the care relationship in end-of-life care from the perspectives of people with ID. The two main dimensions found were dealing with wishes and being there. These dimensions and specifically the fear of people with ID that their wishes will not be respected, show that end-of-life care imposes high demands on the relational qualities of caregivers - e.g. their ability to uncover wishes, listen carefully, empathise and closely observe the behaviour of the person with ID. The results confirm the need for continuous knowledge acquisition and expertise building in this area, and for a better awareness of the good practices that have already been developed (e.g. McLaughlin *et al.* 2014b; Tuffrey-Wijne 2012). This study also shows the possibility and added value of involving people with ID in studies of end-of-life care. This highlights the potential for a more systematic involvement of people with ID in research and practice development, even when difficult topics are involved.

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# 4

# Respecting autonomy in the end-of-life care of people with intellectual disabilities: a qualitative multiple-case study

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## Abstract

### Background

The aim of this article was to describe how caregivers and relatives shape respect for autonomy in the end-of-life care for people with intellectual disabilities (ID) and to discuss to what extent this corresponds with a relational concept of autonomy, such as described in care ethics.

### Method

This study consisted of a qualitative design in which the cases of 12 recently deceased people with ID were reconstructed by interviewing the caregivers and relatives who were closest to the person at the end of their life. A cyclic process of data collection and analysis was used. Interviews were transcribed verbatim and data were analysed inductively.

### Results

Respect for autonomy in the end-of-life care of people with ID was mainly reflected in helping the person with ID familiarise with three transitions: new information on the diagnosis and prognosis, changing care needs and wishes, and important decisions that were at stake. In respecting autonomy, relatives and caregivers encountered several challenges. These concerned ascertaining information needs, communicating about illness and death, inexperience in end-of-life care, eliciting current and hidden last wishes, the dependence of people with ID and conflicting wishes. Several qualities were important for respecting autonomy: attention to information needs, connecting, recognising end-of-life care needs, giving space to show wishes and preferences, and discussing dilemmas.

### Conclusions

If caregivers and relatives embrace autonomy as a relational construct, attained through an open, active and reflective attitude, and have more access to knowledge about communication and how to identify end-of-life care needs, this could lead to improved respect for the ID persons’ autonomy at the end of life. We discuss the view that a relational concept of autonomy is useful for describing respect for autonomy in end-of-life care for people with ID, but that more reflection and openness is needed to sufficiently employ relationships and regard autonomy as a product of joint work.

Keywords autonomy, decision-making, end-of-life care, ethics, intellectual disabilities, truth-telling

## Introduction

As an important value in healthcare, respect for autonomy is also embedded in the care for people with intellectual disabilities (ID). The American Association on Intellectual and Developmental Disabilities (AAIDD) views autonomy as a major principle in disability policy and states that caregivers should always try to discover what the person with intellectual or developmental disabilities wants (as much as possible) and honour those wishes (see the position statement on 'Caring at the End of Life' issued in 2005). In general, respecting autonomy at the end of life can be particularly strenuous as most people become increasingly dependent on others (Proot *et al.* 2004; Winzelberg *et al.* 2005; Claessen *et al.* 2011). Several autonomy-related challenges may arise, such as unwanted dependence, losing control and limitations to activities (Proot *et al.*2004; Vernooij-Dassen *et al.* 2005). The majority of people with ID have already been heavily dependent on others in previous phases of their life. Respecting autonomy at the end of life may be an especially challenging issue in this target group as people with ID become even more dependent in the last phase of their life because of existing cognitive impairments combined with physical impairments resulting from a life-limiting illness. Their wishes and needs at the end of life may therefore not be self-evident.

In modern ethics, respect for patient autonomy is approached in many different ways. Patient autonomy is often described in 'liberal' terms where people know what is best for themselves. In the well-known 'Principles of Biomedical Ethics', for example, the ethicists Beauchamp & Childress (2001) define autonomy in terms of being able to choose and think for oneself, based on a notion of the 'independent and free self' as the highest ideal. Respecting autonomy here means that people must be given the opportunity to decide for themselves without interference from others. One could question, however, whether a liberal notion of autonomy adequately suits the context of end-of- life care, as many people are strongly reliant on others then, and even more, whether it would suit the end-of-life care of highly dependent people, such as people with ID.

A notion of autonomy that gives more space to the dependence of people may be found in care ethics, which represents a more relational conception of autonomy.

For example, in her book Moral Boundaries, Tronto (1993, p. 162) states:

> . . . humans are not fully autonomous, but must always be understood in a condition of interdependence. While not all people need others' assistance at all times, it is a part of the human condition that our autonomy occurs only after a long period of dependence, and that in many regards, we remain dependent on others throughout our lives.

Tronto emphasises that we humans are never independent of others and that we will always need others to 'reach' a notion of autonomy. Other authors have elaborated on this relational concept of autonomy. Verkerk, for example, argues that autonomy cannot be seen in isolation from other people and relationships, that both caregiver and care receiver have responsibilities in maintaining the relationship, and by maintaining that relationship, they guard the autonomy of the patient (Verkerk 1999, 2001). A relational notion of autonomy can also be found in Agich's concept of actual autonomy. Agich defines autonomy as the ability to identify with changing circumstances and he views dependence as an essential feature of human development (Agich 2003). A relational concept of autonomy has been incorporated in empirical studies of people with ID, for example on overeating behaviour in Prader–Willi syndrome (Van Hooren *et al.* 2002). The relational concept of autonomy has also been incorporated in the ethical reflections of Verkerk & Maeckelberghe (2003), who argue that respecting the autonomy of people with ID requires responsible, attentive caregivers who are able to listen and empathise with the view of the world taken by people with ID, and who constantly reflect on their work. According to this vision, autonomy is an achievement rather than an assumption, and is therefore not easily accomplished (Maeckelberghe 2003; Verkerk & Maeckelberghe 2003).

As medical care has improved over recent decades, the life expectancy of people with ID is approaching that of the general population and causes of death are comparable as well (Janicki *et al.* 1999; Ellison & Rosielle 2008). More and more attention is given to the quality of end-of-life care for people with ID (McCallion *et al.* 2012). Although autonomy is seen as an important value, it has rarely been a subject in relation to healthcare for this group (Wullink *et al.* 2009) and we found no studies that specifically address respect for the autonomy of people with ID in relation to end-of-life care. Yet there have been studies that indirectly address elements of respect for autonomy, such as truth-telling, the involvement of people with ID in decision-making and life preferences. These studies show that people with

ID are often not (fully) informed and involved in decision-making (Friedman 1998; Tuffrey-Wijne *et al.* 2005, 2006, 2009, 2010; Watchman 2005; Ryan & McQuillan 2006; Wagemans *et al.* 2010). People with severe ID in particular were often protected from the truth and not helped to understand the consequences of their diagnosis. Withholding the truth may severely obstruct end-of-life care for people with ID (Tuffrey-Wijne & McEnhill 2008). Stein & Kerwin (2010) emphasise that the values and life preferences of people with ID must be well documented, in order to prevent these values from staying unnoticed when end-of-life decisions are at stake.

Although the significance of respecting autonomy in end-of-life care is not disputed, the way in which this is achieved for people with ID remains unclear. This article describes how caregivers and relatives shape respect for autonomy in the end-of-life care for people with ID. We do this by exploring the challenges caregivers and relatives face and the qualities they consider as important for respecting autonomy. In the discussion, we will reflect on the extent to which the results correspond with a relational concept of autonomy as described in care ethics.

## Methods

### Design

This study consisted of a qualitative design in which the cases of 12 recently deceased people with ID were reconstructed from different perspectives. We held semi-structured interviews with several caregivers and relatives who were close to the person at the end of life. Such a qualitative design is particularly well suited to comprehending complex issues such as respect for autonomy, and for grasping the concepts that underlie respect for autonomy in the last phase of life.

### Participants

The interviewees were recruited through contacts with 10 ID care provider organisations in different parts of the Netherlands. Caregivers and relatives of recently deceased people with ID were identified by the contacts, who then asked them whether they would be willing to participate in the study and whether they gave permission to have their names and addresses passed on to the researcher. In total, 47 people (see Table 1) who were close to one of the 12 recently deceased people with ID were interviewed. We included the perspectives of several professional caregivers and trained volunteers as well as relatives (Table 1). In all 12 cases, one or more social workers or nurses were interviewed. In all but one case, at least one relative was interviewed. In nine cases we interviewed physicians: three general practitioners and six physicians specialised in people with ID (ID physicians).

**Table 1** General overview of the cases: the deceased people and the interviewees

| | **Relatives** | **Professional or volunteer caregivers** |
|---|---|---|
| Case 1: female, died from dementia, moderate intellectual disability | 1 friend | 1 nurse<br>1 ID physician<br>1 psychologist |
| Case 2: female, died from metabolic disease, severe/profound intellectual disability | 1 mother | 2 social workers<br>1 home-nurse<br>1 GP |
| Case 3: female, died from kidney and heart failure, mild intellectual disability | 1 sister<br>1 brother-in-law | 1 social worker<br>1 GP |
| Case 4: male, died from cancer, mild intellectual disability | 1 sister<br>1 brother-in-law | 1 social worker<br>1 nurse specialised in palliative care<br>1 GP<br>1 volunteer |
| Case 5: female, died from cancer, mild intellectual disability | | 1 specialised social worker<br>1 nurse |
| Case 6: male, died from cancer, severe/profound intellectual disability | 1 sister | 1 social worker<br>1 ID physician |
| Case 7: male, died from metabolic disease, severe/profound intellectual disability | 1 mother | 1 social worker<br>1 nurse<br>1ID physician |
| Case 8: male, died from cancer, mild/moderate intellectual disability | 1 sister | 2 social workers<br>1 ID physician |
| Case 9: male, died from cancer, mild intellectual disability | 2 sisters | 1 social worker<br>1hospice coordinator<br>1 volunteer |
| Case 10: female, died from dementia, moderate intellectual disability | 1 brother | 1 nurse<br>1 ID physician |
| Case 11: male, died from old age frailty, severe/profound intellectual disability | 1 brother | 1 social worker<br>1end-of-life care consultant |
| Case 12 female, died from cancer, moderate intellectual disability | 1 sister | 2 social workers<br>1 ID physician |
| **Total** | **16** | **31** |

All interviews concerned the experiences with the care for one of 12 recently deceased people. Relatively diverse cases were selected to take into account the variety of possible end-of-life care circumstances. All 12 people with ID received care from a Dutch ID care provider. All but one person died less than a year before the interview was held, allowing us to retrieve the participants' recent memories. One person died 2.5 years before the time of the interview and was included because of his specific situation, living in a hospice with the assistance of a volunteer. Table 1 shows that the cases comprised six men and six women with different levels of severity of ID. Half of them died of cancer. They reached a mean age of 53 years. Half of them died in their own home, two in an intensive care unit of their ID care provider, three in a hospital and one in a hospice. We asked participants when they realised the death of the person was imminent; this ranged from 2 days to 6 months before the death.

### Data collection

Individual in-depth semi-structured interviews were held at the place of preference of the interviewees. Interviews were conducted between December2010 and April 2011 by the first author and lasted 1–2 h. Open questions were used, encouraging interviewees to describe the case in their own words. A topic list was used as a guide for formulating interview questions. Topics included communication, the care provided, the living situation and end-of-life decisions. Data collection was part of a cyclic process of data collection and analysis. This cyclic process was repeated until data saturation was reached.

### Ethical consideration

All interviews were treated confidentially and anonymously. As we only held interviews with care professionals and relatives, no formal medical- ethical approval was required according to Dutch law.

### Analysis

Interviews were taped and transcribed verbatim. Data were analysed inductively, in the sense that the content of the transcribed interviews was given priority in identifying key themes. For the analysis we used descriptive codes, often directly based on words that the interviewees used, and interpretative codes (labels of text fragments that function as concepts). Interview fragments with the same codes were constantly compared during the analysis. Written memos were made concerning the interpretation of codes and the relationships between codes throughout the analysis process. All authors were involved in the entire process from the analyses of interviews to the generation of concepts. All inter- views were analysed by the first author. In addition, each co- author individually analysed all the interviews for at least one case. Differences in analysis and interpretations were discussed, for example concerning whether or not relatives and caregivers shared views on end-of-life care with the person with ID or concerning possible interpretations of a situation in which information was withheld from a person with ID. Because the co-authors analysed the interviews in consecutive order, we were able to jointly refine the coding system over time. The correspondence between interpretations and the original interviews was continuously verified using constant comparative analysis. After the first author had discussed a quarter of the interviews with the co-authors, consensus was reached about the interpretation of the codes addressing respect for autonomy. The first author manually checked the interpretations with the existing interviews and then analysed all interviews with MAXQDA 2007 to facilitate coding (http://www.MAXQDA.com). To validate the results, we discussed the findings in two focus groups with ID caregivers, trainers and policy experts (peer debriefing).

## Results

The way respect for autonomy was shaped became especially eminent during three kinds of transitions at the end of life: (1) when new information became available about the diagnosis, prognosis or treatment of the person with ID; (2) when the care needs and wishes of the person with ID changed in the last phase of life; and (3) when important decisions needed to be made, for example about whether or not to begin treatment. In the perception of relatives and caregivers, respect for autonomy mainly concerned the question of how to help the person with ID feel familiar with these transitions.

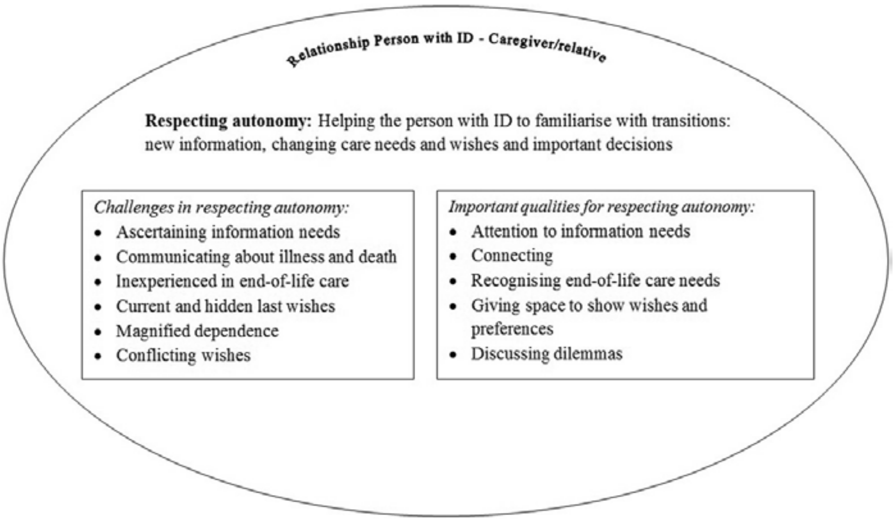

**Figure 1** Respecting autonomy: challenges and important qualities. ID, intellectual disability.

Figure 1 summarises the challenges that caregivers and relatives met in respecting autonomy during these transitions as well as the qualities they regarded as important for respecting autonomy. These challenges and qualities are explained in more detail below. To illustrate the results we use the cases of Joe, Sabine, Cassandra and Charles because autonomy played an important role in their last phase in life (Table 2). We also discuss the extent to which they resemble the other cases studied.

**Table 2** cases illustrating the role of autonomy in end-of-life care

The names used in the case descriptions are not the subjects' real names.

*Joe* was a man aged 40 with a severe intellectual disability and lung cancer. Joe lived in a small-scale residential home on the premises of an ID care provider.

*Cassandra* was a woman aged 49 with a moderate intellectual disability and colorectal cancer. Before her illness, Cassandra lived in her own apartment on the premises of an ID care provider. Cassandra worked on a farm, together with other people with ID. When she became ill, she moved to an intensive care department.

*Sabine* was a woman with a mild intellectual disability and behavioural problems; she was regularly aggressive. When Sabine was 71, she got stomach cancer. Sabine had her own apartment in a center for supervised living. Sabine had daily supervision for several hours.

*Charles* was a man with a mild intellectual disability and serious physical limitations. He had deformities and hemiplegia. Charles used to communicate with a speech computer. When he was 69 he got cancer. Before his illness he lived independently in an apartment with daily supervision.

### Challenges in respecting autonomy

#### *Ascertaining information needs*

It was usually relatives and caregivers, not the person with ID, who first heard about the prognosis. For relatives and caregivers, this raised the difficult question of how to communicate to the person with ID about the situation, estimate what he/she would want to know and how much information he/she was able to handle. The interviewees thought that the person with ID had the right to be informed. However, in the case of very ill people with severe ID in particular, what they understood about the situation was largely unknown, making it very difficult to ascertain their need for information. In these situations, communication mainly started in the here and now, for example by explaining what kind of examination the doctor wanted to perform. With some people with severe ID no communication took place at all, according to their relatives and caregivers: communication was regarded as impossible because of the low cognitive level.

#### *Communicating about illness and death*

There was more potential for involving people with mild ID in communication about the illness; however, this did not automatically lead to more communication:

> *Cassandra was informed about her illness and the approaching end of life by her brother only shortly before she died. The social worker of the farm where*

> *Cassandra used to work thought, however, that the illness should have been discussed at a far earlier stage in her disease: 'I think that Cassandra would have understood it. Now Cassandra was confused; she felt that something was wrong, everybody behaved strangely, but she did not understand what was going on. It was also confusing for the other workers at the farm, who had to guess why Cassandra was suffering.' The social worker did not regard it as her task to communicate to Cassandra about her illness since she was not her primary caregiver. Moreover, she would not have known how to start communication on such a difficult subject. After Cassandra had passed away, the social worker was still bothered by this.*

The social worker observed a need for information in Cassandra, but she did not act upon it. Uncertainty about how to handle difficult subjects combined with the fear of upsetting the person with ID was seen in many cases, especially among social workers, nurses and relatives.

### *Inexperienced in end-of-life care*

Caregivers and relatives considered it their task to make the person with ID feel as comfortable as possible in the last phase of life. This made them primarily focus on finding out what needs the person had and how to respond to these. The physical care needs of the people with ID changed dramatically in all the cases studied. They encountered more physical problems, were bedridden and needed more help with their daily care. It was especially difficult for social workers to respond adequately, as they lacked nursing skills and basic knowledge of end-of-life care. For some it was the first time they had a client who was dying. Moreover, social workers and nurses had to take over tasks from the person with ID, which tended to conflict with their feelings of respecting autonomy, as they were used to encouraging their clients' independence.

### *Current and hidden last wishes*

People with mild ID were, more or less, able to communicate their last wishes, like the place of the funeral and the colour of the coffin. However, there remained a need to find out whether these wishes were still applicable:

> *Charles and his caregivers had previously filled out a book of last wishes regarding the farewell process and the funeral. When he became ill he indicated*

> *that he was not happy with his book of wishes: he felt it had been mostly filled in by others. Together with his social worker, Charles filled out the book of wishes again. 'Charles decided what was going to happen . . . Because he was very ill, we could not finish the book in one go, we had to do it in steps. . . . Every time he said "let's work on the book", we worked right through the whole book all over again.' When the book of wishes was finished it turned out to be exactly the same as the previous version. 'It was exactly the same, but he wanted to do it his way.' Charles died the evening the book of wishes was finished.*

In contrast to Charles, the wishes of people with severe ID were less clear: they were often hidden. Their wishes were often discussed without involving the person with ID, because of communication difficulties and a lack of discussion of wishes in a healthier stage in life.

*Magnified dependence*

Important decisions at the end of life, such as whether or not to start life-prolonging treatment or whether to move to another home, were often taken in multidisciplinary meetings together with relatives. There was no sign from the interviews that the person with ID was present at these meetings. Decisions were based on what were thought to be the wishes of the person with ID or what seemed to be best for that person. In some cases where communication was possible, the wishes of the person with ID were discussed with him or her before the multi-disciplinary meeting, but in other cases the person was only informed afterwards, for example about the use of a probe tube or why it was better to move to another home. For people with severe ID, medical decisions were taken by a physician together with relatives. Many relatives found it very hard to take such decisions for a person with ID; as the case of Joe illustrates:

> *A decision was needed on whether or not to start chemotherapy to lengthen Joe's life. Because of his low cognitive ability, Joe was not able to take this decision for himself. Deciding for Joe was a real burden for his relatives. Joe's illness led to a reflection about their interaction patterns with Joe. His sister came to realize that she had always taken a lot of Joe's preferences for granted, as it turned out that Joe was able to do things she would have never imagined. For example, Joe did not mind so much being in the hospital anymore, and food had become less important to him. She found this hard to accept. 'Joe adapted his desires without*

> *us noticing it. It is so frustrating when you have to take decisions for someone else and you find out that you have been wrong about his wishes several times. Joe being intellectually disabled has never been a problem to me, but when he became ill I hated it. I really wanted to talk to him at the same level for just one day – that he could tell me what he wanted and how he felt.'*

Especially in his last phase in life, Joe's sister became very aware of Joe's dependence. The decisions about prolonging life and about death magnified Joe's dependence: his relatives started to wonder what they had already filled in for Joe earlier in his life.

*Conflicting wishes*

People with mild ID were better able to express their wishes. Although this made communication easier, their wishes could conflict with the wishes of others:

> *All her life, Sabine had had an aversion to hospitals and doctors. She persisted in her desire for independence and did not want to go to the hospital. She suffered severely from fears and restlessness. Her relatives and caregivers offered her medicines to reduce her fears. But Sabine did not want these; she wanted to decide for herself what was good for her. She was afraid that her caregivers would administer the medication secretly, which is not farfetched as this option had been discussed by her relatives and caregivers. In the end, the choice was given to Sabine herself. It was explained to her clearly what the medicines were for; however, she left the medicines on her bedside table untouched until she died. Her caregivers found it difficult to accept her refusal. Social worker: 'You have to let go and accept that Sabine has her own say. . . . But it is always difficult to determine whether she is capable of making these decisions or whether it is better to interfere . . . It is hard that you cannot see what she truly understands.' Physician: 'You have the feeling that Sabine does not realize the consequences of not taking the pills.'*

Sabine's case shows the struggle her relatives and caregivers had with the dilemma between respect for her own wishes and the moral obligation to provide good care and therefore intervene to reduce her fear symptoms. Respecting Sabine's own wishes probably resulted in her keeping her fears. Because of her ID, there were doubts about her decision-making capacity and judgement. Her caregivers

wondered if they did enough to help Sabine understand her situation and if they had sufficiently respected her autonomy. Sabine herself seemed very aware of her dependence, as she was afraid the medicines were administered secretly.
Similarly, in Charles's case, his wishes were compromised:

> *Charles's wish was to die in his own apartment, which was very dear to him. However, his social workers felt that he had the right to receive good, continuous care, including at night, and they felt they were unable to provide this in his apartment. Charles was moved to an intensive care unit. He missed his own apartment a lot. Charles's new nurse thought that his social workers were just too afraid to care for someone with a tumour and possible bleedings. Charles was depressed due to the transition. Charles's new nurse helped Charles understand why he could not go back and tried to make the new situation as comfortable as possible. They took pictures of his apartment and hung them up in his new room. Charles talked proudly about his apartment. After he died, his body was brought back to his apartment and laid out there.*

Charles's wish to stay in his apartment led to a dilemma between respecting Charles's wish and the desire to provide high-quality care around the clock. Ultimately, Charles's wish was not answered and he had to accept the new situation. He was helped to accept this transition by his new nurse, who tried to make him feel at home.

### Important qualities in respecting autonomy

The people with ID in the cases studied needed the help of others to meet their wishes and needs and feel familiar with new situations. According to relatives and caregivers, the following qualities were important in respecting autonomy.

#### *Attention to information needs*

Although the interviewees endorsed the ID people's right to be informed, they did not think that all information should be shared; that depended on their estimation of the individual ID person's information needs. Relatives and caregivers were better able to consider information needs if they knew the person well: knew how much that person already understood about the illness and what their attitude towards illness and death had been in the past. This helped carers recognise the signals implying requests for more information or requests to be left alone.

*Connecting*

Communication was not only about sharing information but was also a means to help the person feel familiar with his illness. Not everyone had access to these skills, according to the interviewees. An important quality was the ability to connect with the person with ID in such a way that it enabled the person with ID to be accessible to the help offered. Joe's case is one such example:

> *Joe's sister: 'We talked to him, not really with him, because he was unable to ask questions. Joe could sort of "talk" by pointing. He also pointed at the stars. Perhaps he only meant: "where is daddy now?" Or "the stars are beautiful", but we interpreted it as: yes that is where daddy is now, daddy is dead. We did not say that daddy is asleep or anything like that. No, we said "daddy is gone and Joe gave his teddy bear to daddy when he died". I thought Joe had the right to know what was going on . . . In the end I said to him: "Just as you can show us the direction where you want to walk, you can also tell us the way you want to die. It's your call. Joe is a big boy and Joe can say for himself how he wants to die. It is okay, you may go." You don't know whether he understands it, but you hope that you are able to communicate a feeling of safety, subconsciously, that it is all right and that there is nothing to be afraid of.'*

Joe's sister connected with him by making a link to Joe's previous experiences and by connecting to Joe's way of communicating: pointing. Although Joe's sister did not need any assistance in connecting with her brother, several other interviewees had a need for tools, such as pictures, to facilitate connecting and communication.

*Recognising end-of-life care needs*

Caregivers strived to respect autonomy by making the people with ID feel as comfortable and familiar with the situation as possible. They therefore wanted to act on their care needs in the best possible way. This required basic knowledge and skills in end-of-life care that enabled caregivers to recognise a care need, to interpret symptoms like pain, restlessness and fatigue, and to know what type of care should be offered.

*Giving space to show wishes and preferences*

Although tracing the preferences of people with a severe ID was very difficult, it was not always impossible:

> *Joe's sister: 'Actually, we never talked to Joe, but in the last period of his life he expressed himself a lot, just because we gave him the space he needed . . . we thought we knew it all, how he was and how he would react. But in fact, this was only half true and he was very well able to show that he was capable of more than we always assumed.' Joe's sister learned from this experience: 'I think that as a caregiver of a relative, you have to look further. Do not assume that the person who you have known all his life, will also die in the same way. People can change in that last period and are able to adapt their preferences. You will be surprised. You will restrict yourself if you don't look beyond the person and try things out.'*

Although Joe had severe ID, he communicated a lot with his sister in his last stage in life. The sister noticed a change in Joe's preferences at the end of his life. She learned to keep an eye open for the needs of people with ID, as needs can change at the end of life. Joe's sister tried out new moves and activities, and closely observed Joe's reaction to actions, which enabled her to notice, for example, that Joe did not mind so much going to the hospital anymore.

*Discussing dilemmas*

In some cases, the wishes of people with ID came into conflict with the wishes and options of others. Sabine's and Charles's cases show the importance of freely discussing the dilemma with the person with ID in order to find a solution that can be accepted by all parties. Although the decisions were difficult and Charles's wish was not granted, Charles and Sabine were both involved in discussions to resolve the dilemmas, as well as the consequences, which shows an attempt to respect their autonomy.

## Discussion

Respect for autonomy in the end-of-life care of people with ID, as summarised in Fig. 1, was mainly reflected in helping the person with ID familiarise with three transitions: new information on the diagnosis and prognosis, changing care needs and wishes, and important decisions that were at stake. The challenges and qualities that were found in this study are discussed below, as well as their correspondence to a relational concept of autonomy.

### Challenges in respecting autonomy

Respecting autonomy implied hard work. The hurdles that caregivers and relatives needed to overcome were diverse and often related to the persons' cognitive level. It was very hard to identify information needs, elicit care needs and uncover wishes in people with severe ID. In people with milder ID, relatives and caregivers struggled to resolve dilemmas with conflicting wishes and strived to find ways to communicate without upsetting the person with ID.

As patients, people with ID have a basic right to be informed about diagnosis and treatment options. It is the foundation for informed decision making. For our interviewees, the challenge in respecting autonomy meant making a careful balance between protecting and giving more information. Moral challenges around truth-telling have also been found in advanced dementia care (Hertogh *et al.* 2004) and have been extensively addressed by Tuffrey-Wijne for the care for people with ID. Tuffrey-Wijne states that breaking bad news to people with ID is a process, and that truth-telling should be facilitated so it can contribute to making people with ID better informed (Tuffrey-Wijne 2012). Some authors suggest that communication about end-of-life wishes and preferences should take place earlier in life, when people are healthier and better able to communicate, for example by using a life story, values assessment, or a preferred places-of-care plan (Watchman 2005; Foster *et al.* 2006; Stein & Kerwin 2010). We do think that early attention to wishes and preferences can contribute to a respect for autonomy if it helps people with ID to make their own choices about end-of-life care. It is, however, still essential that caregivers and relatives keep an open eye for the current needs and wishes, as needs and wishes can change at the end of life. Relatives in particular struggled to involve the person with ID in important decisions. Lack of involvement in end-of-life decision-making clearly remains an issue that may severely undermine the autonomy of people with ID (Stein 2008; Wagemans *et al.* 2010, 2012). Although several decision-making approaches have been described for aiding the involvement of people with ID in end-of-life decisions (Beltran 1996; Friedman 1998; Kleespies 2004; Stein & Kerwin 2010), they have been insufficiently investigated with regard to the actual participation of people with ID.

### Qualities in respecting autonomy

The cases show that respecting autonomy at the end of life is about opening up and understanding the person's perspective, as attention to information needs, connecting with the person with ID and giving space to show wishes and preferences were important qualities in relatives and caregivers when helping the person with ID familiarise with transitions. The cases also show that respecting autonomy at the end of life should focus on addressing the specific needs of people with ID in their last phase of life, as acknowledging an end-of- life care need and discussing dilemmas were important qualities as well. Even though the lives of the people in the cases studied were characterised by lifelong dependence, their dependence was particularly evident and burdensome for their relatives who needed to make important decisions on behalf of a person with severe ID who lacked decision-making capacity. The Netherlands has a model of substitute decision-making for incompetent people in which representatives (usually a close relative) have the legal power to make substitute decisions (Welie *et al.* 2005). Emotions ran high and relatives felt overpowered by the responsibility of taking weighty decisions at the end of life. It made them wonder what they had already unconsciously filled in for the person with ID earlier in life. In most of the cases studied, many things changed rapidly for the people with ID, which underscored the importance of actively addressing the sick person's needs and may explain why respect for autonomy was mainly an issue in transitional situations. The qualities we found all stress the importance of an overall active attitude to 'attaining' autonomy. This makes respecting autonomy sound almost passive in comparison with attaining, ensuring or enhancing autonomy.

### Respecting autonomy: a relational concept?

Our results indicate that a relational concept of autonomy is useful for describing respect for autonomy in end-of-life care for people with ID, but that caregivers and relatives do not always sufficiently employ the relationship to attain autonomy. Relatives and caregivers made numerous references to efforts to help the sick person with ID get through transitions by utilising their relationship. These efforts fit within a relational notion of autonomy, such as in Agich's work where autonomy has been defined as the ability to identify with changing circumstances (Agich 2003). Our results are in accordance with Agich's 'actual autonomy', as Agich explains that when people are impaired, caregivers have the responsibility to help a sick person identify with his or her autonomy, a notion that is also supported by some other authors on end-of-life care (Proot *et al.* 2004; Vernooij-Dassen *et al.* 2005). The qualities we found to be important also show similarities to the core values of 'good care' in care ethics, such as the attentiveness, responsibility, competence and responsiveness of Joan Tronto (Tronto, 1993) and to the work of Verkerk and

Maeckelberghe who wrote that respecting the autonomy of people with ID implies listening to their story, empathising with their view of the world and helping them stay close to themselves (Verkerk & Maeckelberghe 2003). Respecting autonomy sometimes led to severe burdens, especially for relatives of people with a severe ID who had to decide for their beloved one and wished the person was able to decide for himself/herself. These wishes appear to tie in with liberal concepts of self-determining individuals and independence, but in view of the cases studied they are better interpreted as a deep desire for more togetherness and for enlarging the role of the person with ID in the decision-making process. However, some relatives and caregivers regarded communication and involvement simply as impossible, which may mean that at that point the autonomy of the people with ID in these cases was not fully respected. In such cases, relatives and caregivers were far less open to employing their relationship with the person with ID, and they would need more reflection and open- ness to regard autonomy as a product of 'joint work'.

### Strengths and limitations

By using a multiperspective design, we were able to reconstruct the story of 12 deceased people with ID, incorporating the viewpoints of all people who were most closely involved, such as their relatives, social workers, nurses and physicians. This helped avoid giving too much emphasis to the viewpoint of one single person.

The study was performed retrospectively, which is a limitation as the experiences of the interviewees at the actual time that end-of-life care took place remain unknown. On the other hand, a retrospective study makes interviewees' statements less susceptible to day-to-day emotions. Another limitation of our retrospective design is that the experiences of the people with ID themselves could not be studied. A more in-depth study, including participative observation and interviews with people with ID themselves, could give more insight into the qualities of caregivers and relatives that contribute to or undermine autonomy.

### Conclusion and implications

Our material shows a lot of potential for respecting autonomy. If relatives and caregivers embrace autonomy as a relational construct, to be attained by an open, active and reflective attitude, and have more knowledge on communication and identifying end-of-life care needs, this could lead to greater respect for the ID persons' autonomy at the end of life. In practice this implies that ID care providers should encourage team meetings that enable caregivers to jointly discover the options for involving people with ID in communication and decision- making. This may expand the opportunities for helping an ID person become at ease with transitions at the end of life. However, fully respecting the autonomy of people with ID in their last phase in life will be an illusion if caregivers lack the basic skills to provide good end-of-life care. Care providers should therefore also invest seriously in educating their caregivers in recognising and addressing end-of-life care needs.

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# 5

# ‘To move or not to move’: a national survey among professionals on beliefs and considerations about the place of end-of-life care for people with intellectual disabilities

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## Abstract

### Background

The aim of this article was to investigate the beliefs and considerations of professionals concerning an appropriate environment for end-of- life care for people with intellectual disabilities (ID).

### Method

A survey questionnaire was composed based on a scan of relevant literature and the results of group interviews with professionals, family members and people with ID. The questionnaire focused on the respondents' general beliefs about an appropriate environment for end-of-life care and their specific considerations regarding the place of care of the last client for whom the respondent provided end-of-life care. The questionnaire was sent to 294 care staff members recruited from a nationally representative panel of nurses and social workers, 273 ID physicians recruited from the members list of the Dutch professional association of ID physicians, and 1000 general practitioners (GPs) recruited from a nationally representative sample from a national registration of GPs.

### Results

The overall response was 46%. Professionals predominantly believe that all efforts should be made to ensure that a person with ID receives end- of-life care in their own home environment, and that 24/7 care is available. Respondents indicated that most of the clients (79%) who had last received end-of-life care from an ID care service were able to stay in their own home environment. Decisions to keep the client in their own home environment were primarily based on the familiarity of the environment and the expertise of the team in end-of-life care. Insufficient expertise and a lack of adequate equipment were the main considerations in decisions to move a client. Despite the belief of care staff in particular that the wishes of the person with ID should always be leading when deciding on the place of end-of-life care, only 8% of the care staff and ID physicians explicitly mentioned that the client's wishes were taken into account in actual decision-making.

**Conclusions**
Professionals agree that end-of-life care for people with ID should preferably take place in the client's home environment, even when nursing expertise, experience and adequate equipment are not (yet) in place. Nonetheless, a lack of expertise in end-of-life care is the foremost consideration in decisions to move a client. If ID care services want to promote end-of-life care in the client's own home, we recommend formulating a policy on how to realise end-of-life care in the client's own home environment and provide sufficient training and support for staff. To involve people with ID as much as possible, we recommend that professionals integrate more collaborative principles in decision- making about the place of end-of-life care.

**Keywords** decision-making, end-of-life care, intellectual disabilities, place of end-of-life care

## Introduction

Most people prefer to remain in their own home environment up to the end of their life (Gomes et al. 2012; Fischer et al. 2013; Wilson et al. 2013). However, many people actually die in another place, such as a hospital or nursing home (Gomes & Higginson 2008; Van der Velden et al. 2009). Transitions at the end of life can be related to several factors, such as multimorbidity, the availability of health care, a lack of social support or the patient's wishes (Cohen et al. 2006; Van den Block et al. 2007; Abarshi et al. 2010). It is thus far from certain people will receive end-of-life care (aimed at improving the quality of life of people with a life- threatening illness) in their own environment, whether patients in general or terminally ill people with an intellectual disability (ID). However, being cared for in a familiar surrounding may be even more important for people with ID.

Authors who argue in favour of end-of-life care in the client's own home environment point to the importance of familiarity and routine for people with ID (Kirkendall et al. 2012). Professionals in ID care services wish to care for their clients with ID themselves up to the very end (Todd 2004; McCarron et al. 2010a,b; Ryan et al. 2011; Wiese et al. 2012). However, several other studies show that staff working in ID care services often lack expertise and education in end-of-life care (Ng & Li 2003; Botsford 2004; Stein 2008; McCarron et al. 2010a; Tuffrey-Wijne et al. 2010; Bekkema et al. 2014b). Ryan et al. (2010, 2011), for instance, found that Irish professionals caring for people with ID often lack confidence and training in end-of-life care, which led to considerable stress when they cared for terminally ill persons.

In other care services, residential elderly care organisations or hospices, professionals may have more expertise and equipment to deliver high-quality end-of-life care. However, professionals working in such care services say that they have insufficient knowledge and communication skills to provide the specific attention that people with ID need (Todd 2005; Cartlidge & Read 2010). Moreover, studies reveal the difficulties that people with ID encounter when they move to general elderly care services, such as difficulties with participating in social activities and with building relationships with other residents (Thompson et al. 2004; Bigby et al. 2008).

This raises the question of what the best place is for providing end-of-life care for people with ID and what considerations professionals have regarding decisions about the place of care. Only one study specifically addressed the decision-making concerning the place of end-of-life care for people with ID: Tuffrey-Wijne (2009) found that although dying in their own home environment may be the ideal option for many people with ID, resources (e.g. the availability of personnel with relevant expertise and appropriate housing facilities) for supporting end-of-life care in the home environment were not always available. There have been other studies addressing decision-making about the place of care for ageing people with ID. These studies also found that arguments for moving a person to another setting were often related to limited resources for the care of aged clients in their own home environment (Janicki et al. 2002; Bigby et al. 2008). Moreover, Janicki et al. (2002) found that professionals' arguments that were used in decisions to keep an ageing person in their own home situation were often comparable with arguments used in decisions to move them to another setting. These studies suggest that resources (e.g. the expertise of professionals and housing facilities) may be important arguments in deciding upon the place of end- of-life care. Yet the considerations underlying decisions whether or not to move a person with ID to another care setting have only been loosely described so far.

Hence decision-making about the place of end-of- life care for people with ID remains unclear: where should end-of-life care be provided and why should it be provided there? To gain a broad insight, this study will examine both the general beliefs and the considerations in actual decision-making about the best place of end-of-life care. Moreover, we will investigate whether different care professionals have distinct a priori beliefs about the best place of end- of-life care. The following research questions will be addressed:

1. To what extent do people receiving care from an ID care service move to another care setting to receive end-of-life care?
2. What considerations do the professionals concerned actually take into account when deciding on the place of end-of-life care for people with ID? Are decisions to provide end-of-life care in the client's own home environment based on different considerations compared with decisions to provide end-of- life care elsewhere?
3. What beliefs do professionals have with regard to an appropriate environment for end-of-life care for people with ID? Do care staff, ID physicians and general practitioners (GPs) differ in these beliefs?

## Method

### Research subjects

A pre-structured questionnaire was sent to three groups of professionals:

1. Care staff employed by care services for people with ID. The care staff were recruited from an existing nationally representative research panel of care professionals in the Netherlands (De Veer & Francke 2010; De Veer et al. 2011). This research panel consists of a group of registered nurses (mainly associate or Bachelor's level), certified nursing assistants (associate level) and social workers (associate or Bachelor's level) who are prepared to fill in questionnaires on current topics in health care. Nurses generally receive more basic or post-basic vocational training in end-of-life care than social workers (Bekkema et al. 2014b). All registered nurses, certified nursing assistants and social workers who were members of the panel and who worked in ID care services (a total of 294 people) were sent a questionnaire.
2. ID physicians. ID physicians receive a 3-year specialist education in the care for people with ID. They are generally employed by an ID care service. All members of the Dutch professional association of ID physicians (a total of 273) were sent a questionnaire.
3. GPs. In the Netherlands, people with ID receive medical care from either an ID physician or a GP, with GPs commonly caring for people with mild ID living in the community. GPs were recruited by sending a questionnaire to a nationally representative sample of 1000 GPs, drawn from the national registration of about 9000 GPs in the Netherlands (Van Hassel & Kenens 2013).

### Survey questionnaire

The questionnaire focused on considerations and beliefs concerning an appropriate environment for end-of-life care for people with ID. The questions were based on a scan of relevant literature (e.g. Todd 2005; Cartlidge & Read 2010; McCarron et al. 2010a,b; Tuffrey-Wijne et al. 2010), the results of two group interviews that were conducted with professionals (n = 6) and family members (n = 7), and two group interviews with people with mild or moderate ID (n = 10). Professionals and family members were interviewed about their considerations in decision-making concerning the place of end-of-life care, using the technique of moral case deliberation (Molewijk et al. 2008). People with ID were interviewed twice about the best place of end-of-life care using nominal group technique (Tuffrey-Wijne et al. 2007). The considerations that emerged from the literature and the group interviews were used to formulate the questions used in the survey questionnaire. The questionnaire was tested for comprehensibility and completeness among six ID physicians and six ID care staff members.

A specific part of the questionnaire addressed the place of care of the last client with ID for whom the respondent had provided end-of-life care. Respondents were asked where the client lived before the start of the illness, where end-of-life care was actually provided (using the response categories shown in Table 2) and what considerations were actually taken into account in deciding on the place of end- of-life care (open text field).

Beliefs about an appropriate environment for providing end-of-life care were measured by 11 statements that incorporated different aspects of the place and provision of end-of-life care (the response categories were ‘strongly agree’, ‘agree’, ‘neutral’, ‘disagree’ and ‘strongly disagree’). ‘Home’ was defined as the original home situation of the person with ID, referring to all forms of accommodation (living independently, in a community group home or in a residential setting). Also, some questions were included on the respondent’s background characteristics and work-related characteristics.

For GPs, the questions on end-of-life care formed a part of a broader questionnaire on their experiences with patients with ID. GPs usually have only a few patients with ID in their practice (Straetmans et al. 2007) and will therefore have little or no experience with end-of-life care for people with ID. Therefore, GPs were not asked the specific questions about their last patient with ID who needed end-of-life care.

### Analysis

In order to answer research question 1, crosstabs were calculated to measure the frequencies and percent- ages for the combination of original home situation before the start of the illness and the place where end-of-life care was actually provided.

In order to answer research question 2, all considerations that were mentioned by the respondents were coded using a coding system that was developed on the basis of the prior scan of relevant literature and group interviews (see the ‘questionnaire’ section). Two authors (NB and AdV) coded the answers (Cohen’s Kappa of 0.86 for inter-rater reliability). Fisher’s exact tests were used to explore

differences in considerations between decisions to provide end-of-life care in the client's own home environment and decisions to provide end-of-life care elsewhere.
In order to answer research question 3, the percent- age selecting 'agree' or 'strongly agree', means and standard deviations (SD) for the belief questions were calculated to measure the beliefs of professionals concerning the provision of end-of-life care in the client's own home environment. ANCOVAs were performed to explore differences in beliefs between groups of professionals. The ANOCVAs included an adjustment for the possible influence of experience in end-of-life care and for the interaction between experience in end-of-life care and professional background. All analyses were performed using Stata 12.1.

### Ethical considerations

The protocol for conducting the group interviews with people with ID (which were held to construct the survey questionnaire) was approved by the Medical Ethical Committee of the VU University Medical Center. The protocol for the questionnaire study and the group interviews with professionals and family members was in accordance with the Dutch Medical Research Involving Human Subjects Act (see ccmo-online.nl). All respondents received a letter informing them of the aim and goal of the study. Study participation was voluntary. The responses were anonymous and non-traceable to individuals.

## Results

### Background of the respondents

A total of 718 questionnaires were completed. The overall response rate was 46%. The response rate was 67% (n = 196) for care staff, 53% (n = 145) for ID physicians and 38% (n = 377) for GPs. The 196 care staff comprised 85 registered nurses, 8 certified nursing assistants and 103 social workers. The 377 responding GPs all had people with ID in their practice. However, reflecting the limited expertise GPs have with terminally ill people with ID, only 114 (30% of the respondents) completed the questions on end-of-life care.

**Table 1** Background characteristics of the respondents

| | **Total (n=455)** | **Care staff (n=196)** | **ID physicians (n=145)** | **GPs (n=114)** |
|---|---|---|---|---|
| Work experience as a professional (years) | 17.4 | 20.3 | 12.8 | 18.6 |
| Has experience in end-of-life care for people with ID (%) | 77.5 | 63.0 | 93.7 | n.a. |

Table 1 shows that the responding ID physicians have on average less work experience as a professional (12.8 years) than the care staff (20.3 years) and GPs (18.6 years). ID physicians have more experience in providing end-of-life care for people with ID (94%) than care staff (63%).

**Table 2** Breakdown by place where end-of-life care was provided to the last client concerned, per original home situation (the situation before the onset of the illness), n=255 (table numbers are percentages)

| | **Place where end-of-life care was provided** | | | | | | |
|---|---|---|---|---|---|---|---|
| | In original home situation (n=201) | Other department of ID care service (n=36) | Family's home (n=6) | Hospice (n=5) | Nursing home (n=4) | Other (n=3) | Total (n=255) |
| *Original home situation* | | | | | | | |
| **Apartment or group home in a residential setting** (n=142) | 88.0 | 10.6 | 0.7 | - | - | 0.7 | 100 |
| **Group home in the community** (n=86) | 68.6 | 20.9 | 3.5 | 2.3 | 3.5 | 1.2 | 100 |
| **Own apartment or home in the community (either living independent or with family)** (n=18) | 55.6 | 16.7 | 5.6 | 16.7 | - | 5.6 | 100 |
| **Other/unknown** (n=9) | 77.8 | - | 11.1 | - | 11.1 | - | 100 |
| ***Total*** *(n=255)* | *78.8* | *14.1* | *2.4* | *2.0* | *1.6* | *1.2* | *100* |

### Moves to another care setting

A total of 255 care staff and ID physicians responded to the questions about the last client with ID for whom they provided end-of-life care. Seventy-eight per cent of the clients concerned died less than 2 years ago. Table 2 shows that most lived in a residential setting or group home in the community. The table also shows that most (79%) received end-of-life care in their original home environment. This was particularly the case for people who lived in a residential setting. The people who had their own apartment in the community or lived with family were least likely to receive end-of-life care in their original home environment. Most people who changed care settings moved to another department of the ID care service. Transitions to the home of a family member, a hospice or nursing home were less common. The three people who received end-of-life care in another setting all stayed in a hospital.

**Table 3** Considerations that were taken into account in deciding upon the place of end-of-life (EOL) care (% of respondents mentioning consideration). (Percentages do not add up to 100% as respondents could indicate multiple considerations per decision.)

| | **Total** (n= 237) | **EOL care in own home environment** (n=184) | **EOL care in another place** (n=53) |
|---|---|---|---|
| **Expertise in end-of-life care and continuity of care** | | | |
| Expertise of the team in end-of-life care *(e.g. nursing skills and knowledge about end-of-life care)* | 47.7 | 45.1 | 56.6 |
| Possibility to employ extra caregivers (*from inside or outside the own organisation, e.g. nursing, behavioural or palliative care experts, home care or volunteers)* | 11.8 | 13.0 | 7.6 |
| Burden on the team of professionals *(*e.g. the team's emotional stability and self-confidence)* | 6.8 | **4.4** | **15.1** |
| Number of professionals available / intensity of care needs ** | 5.1 | **2.7** | **13.2** |
| Availability of 24/7 care ** | 5.1 | **1.6** | **17.0** |
| **Familiarity** | | | |
| Familiarity with the environment ** (*e.g. the person with ID has lived for years in the same setting, and been cared for by the same care staff, which provides the person with emotional security)* | 46.4 | **59.2** | **1.9** |

*- Table 3 continues –*

*- Table 3 continued –*

| | **Total** (n= 237) | **EOL care in own home environment** (n=184) | **EOL care in another place** (n=53) |
|---|---|---|---|
| **Equipment** | | | |
| Equipment within the residence (home environment) * *(e.g. the possibility of placing a bed downstairs, the presence of patient lifters and available space for medical supplies or a customised bed)* | 16.5 | **13.0** | **28.3** |
| **Wishes/preferences** | | | |
| Wishes/preferences of family members | 15.2 | 15.8 | 13.2 |
| Wishes/preferences of the person with ID | 8.4 | 9.8 | 3.8 |
| Wishes/preferences of professionals * | 6.8 | **8.7** | **0.0** |
| **Other residents** | | | |
| Impact on other residents with ID *(both positive impact, e.g. supporting the ill person and getting the opportunity to say goodbye, and negative impact e.g. lack of time for the care for other residents and stress)* | 5.1 | 3.8 | 9.4 |
| **Other** | | | |
| Expected direct (emotional/physical) burden of the (intended) move on the client | 5.9 | 7.1 | 1.9 |
| Policy/vision of the ID care service regarding the place of end-of-life care | 5.1 | 6.5 | 0.0 |
| Pace of deterioration of the client's health | 2.5 | 3.3 | 0.0 |
| Tranquillity of the home environment | 3.0 | 2.7 | 3.8 |
| Other considerations | 3.0 | 2.7 | 3.8 |

* Fisher exact test = $p<0.05$

** Fisher exact test = $p<0.01$

## Considerations in decisions about the place of end-of-life care

The considerations underlying actual decisions about the place of end-of-life care were described for a total of 237 people (respondents' last client with ID for whom they provided end-of-life care). Table 3 shows that the top three considerations underlying decisions to provide end-of-life care in the client's own home environment were the client's familiarity with the environment (59%), the team's expertise in end-of-life care (45%) and the wishes/ preferences of family members (16%). The top three considerations underlying decision to move the client to another place were the team's lack of expertise in end-of-life care (57%), the lack of equipment within the residence (28%) and the unavailability of 24/7 care (17%).

We found several differences in how often different considerations were taken into account: the familiarity of the environment and the wishes/ preferences of professionals were taken into account more often in decisions to provide end-of-life care in the client’s own home environment than in decisions to provide end-of-life care elsewhere. The burden on the team, the number of professionals available, the availability to provide 24/7 care and the equipment in the residence were taken into account more often in decisions to provide end-of-life care elsewhere than in decisions to provide end-of-life care in the client’s own home environment.

### Beliefs concerning the provision of end-of-life care in the client’s own home environment

Table 4 shows that almost all professionals believe that every effort should be made to ensure that the client can stay in their own home environment (89%) and that care should be offered 24 h a day, 7 days a week (82%). Most respondents do not believe that a team without nursing experience cannot provide good end-of-life care. Absence of experience in end-of-life is also not considered to be an obstacle to providing end-of-life care in the client’s own home environment. The client’s wishes are considered to be leading (72%), but only 41% believe that this preference outweighs the quality of care that can be given at that place. Professionals generally do not believe that the equipment in a residence is an obstacle to the client staying in their own home environment, nor is the presence of other residents.

Care staff are more likely than ID physicians and GPs to believe that the wishes of the client are always leading and they are more likely to stress the importance of 24/7 care. Fewer GPs than ID physicians believe that every effort should be made to ensure that the client can stay in their own home environment, and more GPs worry about the experience of professionals. GPs are also more likely than ID physicians and care staff to believe that professionals from other care organisations are able to care for seriously ill people with ID. ID physicians are least likely to believe that the presence of other residents is an obstacle to providing end-of- life care in a group home.

**Table 4** Beliefs regarding an appropriate environment for providing end-of-life care for people with ID

| | **%<br>Agree or strongly agree<br>Total** | **Mean (SD)<br>Total *** | **Mean:<br>Care staff**<br>(n=192) | **Mean:<br>ID physicians**<br>(n=143) | **Mean:<br>GPs**<br>(n=114) |
|---|---|---|---|---|---|
| **Expertise** | | | | | |
| A team without nursing expertise cannot provide good end-of-life care. | 43.2 | 3.11 (1.10) | 3.18 | 2.94 | 3.21 |
| At the end of life, a client should be housed somewhere that can provide continuous 24-hour care. [1] | 82.4 | 3.96 (0.79) | **4.10** | **3.86** | **3.84** |
| If a team of care staff has no prior experience in providing end-of-life care, it would be better for the client to move elsewhere. [2] | 5.9 | 2.30(0.69) | 2.32 | **2.20** | **2.40** |
| **Familiarity** | | | | | |
| Every effort should be made to enable the client to remain in their own home environment. [3] | 88.9 | 4.16 (0.67) | 4.18 | **4.24** | **4.03** |
| Professionals in other organisations (such as hospices and nursing homes) cannot provide the same level of care for a seriously ill client with an intellectual disability as professionals in intellectual disability care services. [4] | 44.2 | 3.23 (0.94) | **3.35** | **3.41** | **2.81** |
| **Equipment** | | | | | |
| If a place is not fitted with the appropriate equipment and furnishings for the provision of end-of-life care, a client would be better off moving to another place. | 31.4 | 2.9 (0.90) | 2.95 | 2.87 | 2.86 |

*- Table 4 continues –*

*- Table 4 continued -*

| | **%** **Agree or strongly agree** **Total** | **Mean (SD)** **Total *** | **Mean:** **Care staff** (n=192) | **Mean:** **ID physicians** (n=143) | **Mean:** **GPs** (n=114) |
|---|---|---|---|---|---|
| **ID persons' wishes** | | | | | |
| The client's wishes as to where they want to live at the end of life should always be leading. [5] | 72.2 | 3.83 (0.84) | **4.07** | **3.70** | **3.59** |
| A client's preferences regarding where they wants to live at the end of life should be given greater weight than the quality of the care that can be provided in that place. | 40.6 | 3.23 (0.84) | 3.43 | 3.09 | 3.06 |
| **Presence of other residents** | | | | | |
| Care staff in a group home who are providing end-of-life care have insufficient time left for the care of the other residents. [6] | 27.7 | 2.90 (0.92) | **3.05** | **2.62** | **2.99** |
| End-of-life care in a group home is too disruptive for the other residents. [7] | 8.9 | 2.30 (0.80) | **2.46** | **2.03** | **2.36** |
| A group home is too disruptive for a client who receives end-of-life care. [8] | 5.5 | 2.32 (0.72) | **2.44** | **2.08** | **2.45** |

* Range = 1-5, 1 = strongly disagree - 5 = strongly agree

P values reported for post-hoc Bonferroni test, after ANCOVA, $p<0.05$:

[1] Care staff versus ID physicians: p=0.016. Care staff versus GPs: p=0.015

[2] ID physicians versus GPs: p=0.042

[3] ID physicians versus GPs: p=0.037

[4] Care staff versus GPs: p<0.001 ID physicians versus GPs: p<0.001

[5] Care staff versus ID physicians: p<0.001 Care staff versus GPs: p<0.001

[6] Care staff versus ID physicians p<0.001 ID physicians versus GPs: p=0.005

[7] Care staff versus ID physicians: p<0.001 ID physicians versus GPs: p=0.004

[8] Care staff versus ID physicians: p=0.003. ID physicians versus GPs: p<0.001

## Discussion

Most of the professionals in this study believed that every effort should be made to ensure that a person with ID can receive end-of-life care in their own home environment. In practice, most people (79%) who received care from an ID care service in our study were able to remain in their own home environment, with the familiarity of the environment being the foremost consideration. This is a larger number than the figures we have from studies of the general population (Cohen et al. 2006; Van den Block et al. 2007; Van der Velden et al. 2009; Abarshi et al. 2010).

Our study also revealed that although a lack of nursing expertise and adequate equipment is not generally believed to be an obstacle to providing end-of-life care in the client's own home environment, still the lack of expertise and equipment were the main considerations in decisions to move a client. This may explain why people living in their own apartment in the community or with family were more likely to be moved than people who lived in a residential setting. Despite the belief of care staff in particular that the wishes of the person with ID should always be leading in deciding on the place of care, only 8% of the care staff and ID physicians explicitly mentioned that the wishes of the client were taken into account in actual decisions.

### Familiarity and relationships

Care staff and ID physicians said that staying in a familiar environment with care staff who know the person well and have known them for a long time provides that person with emotional security. This sense of security is important for people in the general population too. Yet it may be even more important for people with ID to be close to familiar caregivers, as it can be very difficult to 'read' their needs and identify signals such as distress and pain (Ryan et al. 2010; Bekkema et al. 2011). McCarron et al.'s study also demonstrated the importance of long-term relationships at the end of life, as for the staff in their study the maintenance of the relationship with the person with ID and dementia even outweighed the burden of the care on the team (McCarron et al. 2010a). Strong wishes of professionals to care for a client until the very end may also play a role. The wishes and preferences of professionals mentioned in our study were all in favour of keeping the client in their own home environment. Likewise, Wiese et al. (2012) found that staff were unanimous in their preference for end-of-life care at home where possible, including staff with negative past end-of-life care experiences.

As for relationships with other residents, their presence is not believed to be an obstacle to providing end-of-life care in the client's own home environment.

### Resources needed to provide end-of-life care

A lack of expertise was the foremost consideration in decisions to move a client. This emphasises the need for continuing training in end-of-life care, particularly as an earlier study revealed that only 15% of care staff in ID care services feel adequately trained to provide good end-of-life care (Bekkema et al. 2014b). Nevertheless, professionals generally do not believe that a lack of nursing expertise or experience in end-of-life care is an obstacle to providing end-of-life care. This may be a reflection of the professionals' strong belief that the client should preferably stay in their own home environment. GPs (who work outside ID care services) are somewhat more critical; they worry more about a lack of experience and are more likely to believe that professionals from other care organisations are also able to care for seriously ill people with ID. On the other hand, expertise in end-of-life care was also an important consideration in decisions to keep the client in their own home environment. In these cases, the teams' expertise in end-of-life care may have been sufficient or supplemented sufficiently by the expertise of others, although the possibility of employing extra caregivers was not often mentioned as a consideration.

Care staff in particular believe the availability of 24/7 care to be very important in determining where end-of-life care is provided. Indeed, the unavailability of 24/7 care was the third most common consideration in decisions to move a client. However, the availability of 24/7 care was rarely a consideration in decisions to keep a client in their own home environment. This may be explained by the fact that many of the clients in our study who stayed in their own home environment already lived in a residential setting, in which 24/7 care may be seen as a natural part of care.

### Wishes of people with ID

Being able to die according to your own preferences is generally found to be an important value in end-of-life care (Patrick et al. 2003). The majority of the professionals in our study (72%) also believe that the wishes of the person with ID should always be leading in deciding on the place of end-of-life care. Care staff in particular (who work on a day-to- day basis with people with ID) seem in favour of giving the client's wishes considerable weight in decision-making. This is in line with the findings of Williams et al. (2010), who found that listening to the person with ID is central to the decision-making of ID nurses. Nonetheless, only 8% of the care staff and ID physicians explicitly mentioned that the wishes or preferences of the client were taken into account in actual decisions, while the wishes and preferences of family members were mentioned twice as often (15%). It is unclear what this means. The difference may imply that decisions about the place of care are often driven in practice by professional and practical arguments, such as expertise and equipment, and that the wishes of the client can only be followed when they do not conflict with these professional and practical arguments. This would be in line with the belief that the client's preferences about where they want to spend the end of their life do not outweigh the quality of care that can be given at that place. It points to the responsibility of professionals to provide good-quality end-of-life care and confirms the weight given to resources as important considerations, which is in line with what has been found by Tuffrey-Wijne (2009). It may also be that the wishes of the person with ID were indeed taken into account in the actual decisions, but more implicitly, incorporated in other considerations such as the familiarity of the environment. It is also possible that the wishes were unclear, as it has been found that respecting the autonomy and eliciting the wishes of dying people with ID is very difficult (Bekkema et al. 2014a). In that case, new ways to enhance the involvement of people with ID in decision-making need to be explored.

### Strengths and weaknesses

An important strength of this study is that we studied both the considerations in actual decision-making and general beliefs. We were therefore able to provide a more profound insight into the decision-making on the place of end-of-life care for people with ID. Another strength is that we were able to recruit a broad group of professionals who work with people with ID by using two nationally representative samples and one members list that covers almost all practising Dutch ID physicians. A possible limitation of this study is the recall of the last client for whom care staff and ID physicians provided end-of-life care, although the majority of these clients died quite recently. Another limitation is that more than half of the actual decisions concerned people living in a residential setting, which may have resulted in an overrepresentation of people who were already receiving intensive care. Therefore, future studies should try to include a larger group of people who live in the community. Furthermore, future research should study the beliefs and considerations of relatives, as relatives are also part of the decision-making process. Finally, more knowledge is needed on how to enhance the involvement of people with ID in decision-making, as their wishes about the place of end-of-life care were rarely mentioned as a consideration in actual decisions.

### Recommendations

According to professionals, end-of-life care for people with ID should preferably take place in their own home environment. In this regard, it is positive that most of the clients reported on actually seemed able to remain in their familiar environment. This study also indicates that the home environment needs to be able to provide adequate resources, such as personnel with expertise in end-of-life care, adequate equipment and 24/7 care, as a lack of such resources were the main considerations in decisions to move a client. If ID care services want to promote end-of-life care in the clients' own home, we therefore recommend that they (1) formulate a policy on how to realise end-of-life care in the client's own home environment; (2) encourage professionals to reflect on their own expertise and provide sufficient (continuing) training and expert consultation opportunities; and (3) provide emotional and practical support for staff. To enhance the involvement of people with ID in decision-making about the place of end-of-life care, professionals could learn from newly proposed models, such as the ASK ME model, that use collaborative principles to involve people with cognitive disabilities as much as possible in decision-making related to their care (Peisah et al. 2013).

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# 6

# Decision making about medical interventions in the end-of-life care of people with intellectual disabilities: A national survey of the considerations and beliefs of GPs, ID physicians and care staff

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## Abstract

### Objective

This paper explores the personal beliefs and specific considerations of professionals regarding decisions about potentially burdensome medical interventions in the end-of-life care for people with intellectual disabilities (ID).

### Methods

A survey questionnaire covering decision making about potentially burdensome medical interventions was sent to nationally representative samples of 294 ID care staff-members, 273 ID physicians and 1000 GPs.

### Results

Professionals predominantly believed that considerations about quality of life are most important. Quality of life and wellbeing were also frequently considered in both decisions to start/ continue an intervention and decisions to forgo/withdraw an intervention. Seventy percent believed that people with ID should always be informed about interventions, and 61% would respect a refusal by the person. The family's wishes were explicitly considered more often than the wishes of the person with ID.

### Conclusion

Although respondents agree that the quality of life is highly important, the wishes of people with ID (especially of those with severe/profound ID) were often not considered in decisions about potentially burdensome medical interventions.

### Practice implications

To enhance the active involvement of people with ID in decision making we recommend that professionals integrate collaborative principles in decision making and make use of pictorial and easy reading resources.

### Keywords

End-of-life care Intellectual disabilities Decision making Medical interventions

## 1. Introduction

End-of-life care is the total care that aims to maintain the best possible quality of life until death. According to the WHO (WHO 2014), end- of-life care provides relief from pain and other distressing symptoms, and may start early in the course of a life-threatening illness. The focus on quality of life has consequences for choice of medical interventions (Higginson *et al.* 2013; Raijmakers *et al.* 2012; Lorenz *et al.* 2008). Medical interventions can improve wellbeing but may also be quite burdensome for somebody with a life-threatening illness. Decisions about whether to use or forgo medical interventions are therefore of paramount importance. These decisions may be especially difficult when it comes to people with intellectual disabilities (ID).This article presents a study of the beliefs and considerations that influence such decisions.

There are several reasons why decision making about medical interventions at the end of life can be more difficult in the case of people with ID. First, it may take more time before palliative care needs are recognised. A study in the UK showed that people with ID had more delays in the diagnosis and treatment of their final illness and received less opioid analgesia than a group without ID (Heslop *et al.* 2013). Second, many people with ID do not fully understand the reasons, impact and consequences of medical interventions.

Communication about medical interventions and assessing their wishes and needs is often difficult (Bekkema *et al.* 2014a; Tuffrey-Wijne *et al.* 2009; Stein 2008; Lohiya *et al.* 2003). Heslop et al. (2013) found almost no documentation on what people with ID understood about their condition and medical interventions. Similarly, Wagemans et al. (2010) found no evidence in medical files that people with ID were involved in end-of-life decision making. Not understanding their illness and intervention options can lead to considerable distress in people with ID (Tuffrey-Wijne *et al.* 2009). Third, determining the wellbeing of people is an important element in end-of-life care. The identification and management of pain and other symptoms can be complicated in people with ID, as signals that express pain or distress are hard to recognize (Bekkema *et al.* 2011; Ryan *et al.* 2010; Crawford 2010). Difficulties in determining wellbeing may lead professionals to rely on biased presumptions about a person's needs and values (Stein & Kerwin 2010; Flynn *et al.* 2009).

So far, most studies addressing medical interventions in the end-of-life care of people with ID have either been case descriptions (Stein & Kerwin 2010; Flynn *et al.*

2009) or have focused specifically on decisions with a possible life-shortening effect (Wagemans *et al.* 2013a & 2013b). Yet many medical intervention may be potentially burdensome for people with ID who need end-of-life care due to their limited level of understanding and communication difficulties. As far as we know, these decisions have not been explored. Neither do we know whether decisions to start an intervention are based on different considerations than decisions to forgo an intervention. To enhance well- considered decision making, we need insight into the beliefs and actual considerations of professionals in such situations. The research questions are:

1. What considerations do the professionals concerned take into account in actual decision making about the use of potentially burdensome medical interventions? Are decisions to start or continue a medical intervention based on different considerations than decisions to withdraw or forgo a medical intervention?
2. What beliefs do professionals have with regard to the use of potentially burdensome medical interventions in the end-of-life care for people with ID? Do care staff, ID physicians and GPs differ in these beliefs?

## 2. Methods

### *2.1. Research subjects*

A pre-structured questionnaire was sent to three groups of professionals:

(1) Care staff employed by care services for people with ID, recruited from an existing national research panel of care professionals in the Netherlands (Albers *et al.* 2014; de Veer *et al.* 2011). This research panel consists of a national sample of registered nurses, certified nursing assistants and social workers who are prepared to fill in questionnaires on current topics in health care. All panel members who worked in ID care services (a total of 294 people) were sent a questionnaire.

(2) ID physicians, who had received three-year specialist education in the care for people with ID and who are generally employed by an ID care service. All members of the Dutch professional association of ID physicians (a total of 273) were sent a questionnaire.

(3) General practitioners. In the Netherlands, people with ID receive medical care from either an ID physician or a GP, with GPs commonly caring for people with mild ID living in the community. GPs were recruited by sending a questionnaire to a nationally representative sample of 1000 GPs, drawn from the national registration of about 9000 GPs in the Netherlands (van Hassel & Kenens 2013).

*2.2. Survey questionnaire*

The questionnaire focused on (1) the respondents' background characteristics, (2) the case-related considerations that played a role in decisions about transitions in the place of care and medical interventions and (3) personal beliefs about transitions in the place of care and potentially burdensome medical interventions, such as chemotherapy, life-prolonging surgery, radiotherapy, tube feeding, pain medication by infusion pump, further diagnostic investigations or punctures that may in some sense be burdensome for the person with ID. This paper focuses on decisions regarding medical interventions.

The questionnaire items relevant for this paper were based on a scan of the relevant literature (e.g. about decisions regarding chemotherapy, tube feeding and further diagnostic investigation in the end-of-life care for people with ID (Tuffrey-Wijne *et al.* 2009; Wagemans *et al.* 2010; Stein & Kerwin 2010), on the results of two group interviews with professionals and family members, and on two group interviews with people with mild and moderate ID.

1. Questions were included on the respondent's background and work-related characteristics.
2. The considerations used in decisions on medical interventions were explored by open questions about the last person with ID for whom the respondent provided end-of-life care. Respondents were asked if a potentially burdensome medical intervention was considered (using response categories, see Table 1) and whether it was decided to start/continue or forgo/ withdraw this intervention. If more than one intervention was considered, the respondent chose one intervention to answer the questions about. Respondents were asked what considerations in the case of that specific person led to the decision to start/ continue or forgo/withdraw the intervention (open text field).
3. Personal beliefs about the use of potentially burdensome medical interventions were measured by eleven statements that incorporated different considerations about the use of medical interventions in end-of-life care (the response

categories were 'strongly agree', 'agree', 'neutral', 'disagree' and 'strongly disagree').

For GPs, the questions on end-of-life care formed part of a broader questionnaire on their experiences with patients with ID. GPs usually have only a few patients with intellectual disabilities in their practice (Straetmans *et al.* 2007) and will therefore have little or no experience with end-of-life care for people with ID. Therefore, GPs were not asked the specific questions about their last patient with ID who needed end-of-life care.

The questionnaire was tested for comprehensibility and completeness among six ID physicians and six ID care staff members.

### *2.3. Analysis*

To answer research question 1, all considerations that were mentioned by the respondents in the open text field were coded using a code system partly based on the preliminary scan of relevant literature and group interviews (see questionnaire section) and extended with new codes that emerged from the data. Two authors (NB and AdV) coded the answers (Cohen's Kappa for inter-rater reliability: 0.93). Fisher's exact tests were used to explore differences in considerations between decisions to start/ continue a medical intervention and decisions to forgo/withdraw a medical intervention. The cases reported were checked for possible double counting: all cases included are unique cases.

To answer research question 2, the percentages selecting 'agree'/ 'strongly agree', means and standard deviations (SD) were calculated to measure the beliefs of professionals. ANCOVAs were performed to explore differences in beliefs between groups of professionals. The ANCOVAs included an adjustment for the possible influence of experience (yes/no) in end-of-life care and for the interaction between experience in end-of-life care and professional back- ground. All analyses were performed using Stata 12.1.

### *2.4. Ethical considerations*

The protocol for conducting the preliminary group interviews with people with ID (used to compose the questionnaire) was approved by the Medical Ethical Committee of the VU University Medical Center. The preliminary group interviews with professionals and family members and the protocol for the questionnaire study and did not involve imposing interventions or actions, and in such cases no approval by an ethics committee is required in the Netherlands (according to the Medical Research Involving Human Subjects Act, see http://ccmo-online.nl). All respondents received a letter informing them of the aim of the study. Study participation was voluntary. The responses were anonymous and non-traceable to individuals.

## 3. Results

### *3.1. Background of the respondents*

The overall response rate was 46% with 718 questionnaires being completed. The response rate among care staff was 67% (n = 196), among ID physicians 53% (n = 145) and among GPs 38% (n = 377). The group of care staff (n = 196) was comprised of 85 registered nurses, 8 certified nursing assistants and 103 social workers. All responding GPs (377) had patients with ID in their practice and 107 (28% of the 377 GPs) filled in the questions on end-of-life care. The responding ID physicians had on average less work experience as a professional (12.8 years) than care staff (20.3 years) and GPs (18.5 years). ID physicians had more experience of providing end-of-life care for people with ID (94%) than care staff (63%). Of the 377 GPs with patients with ID in their practice, 24% had experience with end-of-life care for this patient group.

### *3.2. Background medical interventions*

A total of 248 care staff and ID physicians completed the questions about the last person with ID for whom they provided end-of-life care. A potentially burdensome medical intervention was considered in 145 of the 248 cases described (58%). Of these 145 persons, 85% died less than two years ago, 52% had mild/ moderate ID and 48% severe/profound ID, and 37% died of cancer, 17% of an overall decline due to old age, 14% of dementia, 6% of heart failure, 5% of stroke, 2% of asthma/COPD and 19% of another/ unknown disease.

Table 1 shows that decisions to start or continue an intervention often concerned pain medication by infusion pump or tube feeding. Decisions to forgo or withdraw an intervention often concerned further diagnostic investigations, tube feeding or pain medication by infusion pump.

**Table 1** Potentially burdensome medical interventions that were considered (%)

| | **Decision: start/ continue** (%) | **Decision: forgo/ withdraw** (%) | **Total** (%) |
|---|---|---|---|
| Pain medication by infusion pump | 38.1 | 13.4 | 24.1 |
| Tube feeding | 15.9 | 22.0 | 19.3 |
| Further diagnostic investigation (e.g. origination carcinoma, heart failure tests) | 4.8 | 26.8 | 17.2 |
| Other intervention (e.g. placing a suprapubic catheter or stoma, providing oxygen and dialysis). | 14.3 | 8.5 | 11.0 |
| Chemotherapy | 6.4 | 11.0 | 9.0 |
| Life-prolonging surgery | 4.8 | 7.3 | 6.2 |
| Administering antibiotics | 3.2 | 3.7 | 3.5 |
| Pain medication (other than infusion pump; e.g. by injection) | 6.4 | 0.0 | 2.8 |
| Radiotherapy | 3.2 | 2.4 | 2.8 |
| Unknown intervention | 1.6 | 3.7 | 2.8 |
| (Veni)puncture | 1.6 | 1.2 | 1.4 |
| **Total** | **100 (n=63)** | **100 (n=82)** | **100 (145)** |

**Table 2** Considerations in decisions about potentially burdensome interventions (% of cases in which a consideration was taken into account)

| | **Total** (n=140) (%) | **Decision: start/ continue** (n=61) (%) | **Decision: forgo/ withdraw** (n=79) (%) |
|---|---|---|---|
| Quality of life and wellbeing (e.g. mobility, minimising suffering, no pain, tranquillity, comfort, dignity) | 43.6 | **52.5** | **36.7** |
| Wishes/preferences of family members | 25.7 | **23.0** | **27.8** |
| Medically meaningful intervention/ medical futility ** | 22.9 | 3.3 | **38.0** |
| Direct emotional strain of the intervention on the person (e.g. understanding of the intervention, fear/stress due to the intervention/hospital) | 19.3 | 11.5 | **25.3** |
| Direct physical strain of the intervention on the person (e.g. weak health, fast decline in physical condition) ** | 17.9 | 4.9 | **27.9** |
| Wishes/preferences of person with ID | 15.7 | **21.3** | 11.4 |
| Wishes/preferences of professionals | 12.1 | 14.8 | 10.1 |
| Availability of an alternative intervention (e.g. less invasive intervention such as oral pain relief instead of by infusion pump) * | 4.3 | 0.0 | 7.6 |
| Prolonging the life of the person with ID * | 3.6 | 8.2 | 0.0 |
| Curative goal * | 3.6 | 8.2 | 0.0 |
| Possibility of the person with ID to cooperate with intervention (e.g. leave tube unattached, cooperate during diagnostic investigation) | 3.6 | 0.0 | 6.3 |
| Age of the person with ID | 2.9 | 1.6 | 3.8 |
| Diagnostic uncertainty | 2.1 | 3.3 | 1.3 |
| Professionals involved not authorised to work with intervention (e.g. providing tube feeding) | 1.4 | 0.0 | 2.5 |
| Costs of the intervention | 0.7 | 0.0 | 1.3 |

Please note; in 10.7% of the cases other remarks were made that could not be clustered into a meaningful category. Numbers in bold refer to considerations mentioned in at least 20% of the decisions.
* Fisher's exact test: $p<0.05$
** Fisher's exact test: $p<0.01$

### *3.3. Considerations in decisions about the use of potentially burdensome medical interventions*

Considerations that were taken into account in decisions about the use of potentially burdensome medical interventions care were described for a total of 140 people. Table 2 shows that three types of considerations were taken into account in at least 20% of the decisions to start or continue an intervention: considerations regarding the quality of life and wellbeing (52.5%), the wishes/ preferences of family members (23.0%) and the wishes/preferences of the person with ID (21.3%). Five types of considerations were taken into account in at least 20% of the decisions to forgo or withdraw an intervention: considerations regarding the futility of the intervention (38%), the quality of life and wellbeing (36.7%), the direct physical strain of the intervention on the person (27.9%), the wishes/preferences of family members (27.8%) and the direct emotional strain of the intervention on the person (25.3%). The wishes/preferences of people with mild/moderate ID were taken into account more often (27.8%) than the wishes/preferences of people with severe/profound ID (2.9%).

Prolonging the life of the person with ID and curative goals were taken into account more often in decisions to start or continue the intervention than in decisions to forgo or withdraw the intervention. The futility of the intervention, the direct physical strain of the intervention and the availability of an alternative intervention were taken into account more often in decisions to forgo or withdraw the intervention than in decisions to start or continue the intervention.

Table 3 Beliefs about the use of potentially burdensome medical interventions in the end-of-life care for people with ID

| | % Agree or strongly agree Total (n=434) | M (SD) Total * (n=434) | M Care staff (n=187) | M ID physicians (n=140) | M GPs (n=107) |
|---|---|---|---|---|---|
| **Quality of life** | | | | | |
| The client's quality of life is the most important consideration in deciding on a medical intervention. [1] | 92.6 | 4.28 (0.70) | 4.14 | 4.47 | 4.25 |
| **Emotional strain on the person with ID** | | | | | |
| The potential unease the client may feel because of an intervention outweighs the potential prolongation of life. | 72.0 | 3.83 (0.86) | 3.94 | 3.77 | 3.73 |
| If a client has to be admitted to hospital for a medical intervention, you should be wary about using that intervention. [2] | 44.4 | 3.23 (0.91) | 3.13 | 3.18 | 3.46 |
| **Cooperation of the person with ID** | | | | | |
| If a client is unable to cooperate properly with the medical intervention, it is better not to start on that intervention. [3] | 33.1 | 3.06 (0.81) | 2.96 | 3.01 | 3.31 |
| **Wishes/preferences of the person with ID** | | | | | |
| Clients should always be informed about options for intervention, even if the possibilities for communication are limited. [4] | 69.5 | 3.71 (0.85) | 3.75 | 3.54 | 3.89 |
| The client's wishes about whether to use the medical intervention are always leading. [5] | 43.6 | 3.28 (0.87) | 3.49 | 3.15 | 3.09 |
| If a client does not want an intervention, this wish must be respected. [6] | 60.8 | 3.61 (0.75) | 3.73 | 3.51 | 3.53 |
| If a client's wishes are not in line with the opinion of the caregivers, the client's wishes prevail. [7] | 56.5 | 3.58 (0.75) | 3.76 | 3.50 | 3.36 |

*-table 3 continues –*

*- table 3 continued -*

| | % Agree or strongly agree Total (n=434) | M (SD) Total * (n=434) | M Care staff (n=187) | M ID physicians (n=140) | M GPs ( n=107) |
|---|---|---|---|---|---|
| **Decision-making capacity of the person with ID** | | | | | |
| If a client refuses an intervention, their decision-making capacity in this matter should always be investigated. [8] | 60.1 | 3.54 (0.88) | 3.31 | 3.91 | 3.43 |
| If a client is unable to decide for themselves, the family's opinion about whether to use the medical intervention should be the deciding factor. [9] | 52.1 | 3.43 (0.78) | 3.65 | 3.15 | 3.39 |
| If a client is unable to decide for themselves, the physician's opinion about whether to use the medical intervention should be the deciding factor. [10] | 39.8 | 3.23 (0.77) | 3.04 | 3.43 | 3.28 |

* range = 1-5, 1 = strongly disagree - 5 = strongly agree

p values (p<.05) reported of post hoc Bonferroni test, after ANCOVA:

[1] Care staff versus ID physician: p=0.000, ID physician versus GPs: p=0.042

[2] Care staff versus GPs: p=0.009, ID physician versus GPs: p=0.048

[3] Care staff versus GPs: p=0.001, ID physician versus GPs: p=0.013

[4] ID physician versus GPs: p=0.004

[5] Care staff versus ID physician: p=0.002, care staff versus GPs: p=0.001

[6] Care staff versus ID physician: p=0.031

[7] Care staff versus ID physician: p=0.005, care staff versus GPs: p<0.001

[8] Care staff versus ID physician: p<0.001, ID physician versus GPs: p<0.001

[9] Care staff versus ID physician: p<0.001

[10] Care staff versus ID physician: p<0.001, care staff versus GPs: p=0.034

### *3.4. Beliefs about the use of potentially burdensome medical interventions*

Table 3 shows that almost all professionals (92.6%) believe that considerations regarding the quality of life are the most important considerations in decisions about potentially burdensome medical interventions. Most professionals (72%) believe that the client's possible unease outweighs prolongation of life. The client not being able to cooperate with a medical intervention is generally not perceived as a reason to forgo the intervention. A majority (69.5%) believe that people with ID should always be informed about intervention options, even when the possibility of communication is limited. Less than half of the professionals (43.6%) believe that the wishes of the person with ID should be leading, yet rather more professionals (56.5%) believe that these wishes should prevail when they are not in line with the opinion of professionals. A slight majority (60.8%) believe that a refusal of the intervention should be respected, yet a similar proportion (60.1%) believe that if a person with ID refuses an intervention, their decision-making capacity should be tested. If a person with ID is unable to decide for themselves about a medical intervention, more professionals believe that the opinion of relatives is decisive (52.1%) than believe the opinion of the physician should prevail (39.8%). ID physicians are the only professional group giving more weight to the opinion of the physician than of the relatives.

There are differences, albeit small, in beliefs between the three groups of professionals: care staff are most likely to believe that the wishes of the person with ID should be leading and they are also more likely to follow the opinion of relatives rather than the opinion of the physician. ID physicians are most likely to believe that the quality of life is the most important consideration in decisions, are most likely to believe that if a person with ID refuses an intervention their decision-making capacity should be tested, and are less inclined to always inform the person with ID about intervention options. GPs are most likely to believe that hospital admission should be avoided and that an intervention should be avoided if it is not possible for the client to cooperate properly.

## 4. Discussion and conclusion

### *4.1. Discussion*

This study explored actual considerations and personal beliefs of professionals regarding decisions about potentially burden- some medical interventions in the end-of-life care for people with ID.

Regarding actual considerations, the study shows that decisions about potentially burdensome medical interventions are often made in the end-of-life care of people with ID. Most of the decisions described in our study resulted in the rejection or withdrawal of a potentially burdensome medical intervention, mainly based on considerations regarding the futility of the intervention and the quality of life and wellbeing of the person. Considerations about the quality of life and wellbeing were the commonest considerations in decisions to start or continue an intervention. Generally, an important aspect of quality of life in end-of-life care is pain relief (WHO 2014). Decisions about pain medication by infusion pump were the decisions most frequently mentioned in this study. The majority of these decisions resulted in the start or continuation of the infusion pump, which indicates that in these cases the relief of pain outweighed the possible distress that the infusion pump might cause the person. As other studies have indicated, decision making about pain interventions should always be preceded by careful identification of pain, as assessing pain in people with ID can be very complicated (Heslop *et al.* 2013; Bekkema *et al.* 2011; Ryan *et al.* 2010; Bekkema *et al.* 2014b).

Regarding personal beliefs, almost all professionals, and ID physicians in particular, found the quality of life of the person with ID the most important consideration. This finding is in line with studies within the general population on decision making about medical interventions at the end of life (Higginson *et al.* 2013; Raijmakers *et al.* 2012; van Leeuwen *et al.* 2004). Yet what wellbeing actually means can best be determined by the people with ID themselves. Many people with ID will need the help of relatives and professionals to clarify their values and wishes, for example by eliciting the life story of the person with ID as a means of clarifying their values and preferences, preferably before the onset of the illness (Stein & Kerwin 2010).

### *4.1.1. Right to know*

Although most respondents (69.5%) agreed with the statement that people with ID should always be informed about intervention options, a minority (30.5%) had another opinion. The 69.5% that agreed with this statement may be seen as a low percentage given the patient's legal right to be informed about medical interventions. Several explanations may account for this finding: professionals may want to protect people with ID from complex information they presumably cannot (fully) understand. This may reflect moral challenges in truth-telling, which have e.g.

also been found in advanced dementia care (Hertogh *et al.* 2004). Moreover, informing people with ID about medical interventions can be hampered by communication barriers (Bekkema *et al.* 2014a; Tuffrey-Wijne *et al.* 2009; Stein 2008; Lohiya *et al.* 2003). It may also be that professionals prefer to inform a family member or other representative. Yet even people who lack decision-making capacity have a right to be informed. The decision not inform the person with ID should therefore not be based on fear or prejudgments.

### *4.1.2. Wishes of the person with ID*

Only 43.6% of the professionals believe that the wishes of the person with ID should always be leading in decisions about medical interventions, whereas we described in another paper that 72% believe that this wish is leading in decisions about the place of end-of-life care (Bekkema *et al.* 2014c). Probably, professionals believe that people with ID are less able to decide about complex medical matters than about more practical matters such as where they want to spend the end of their life. The wishes of people with severe/profound ID in particular were rarely taken into account (2.9%).

Care staff appear to be more in favour than physicians of giving the clients' wishes priority in decisions about medical interventions. This finding is in line with the findings presented in the paper about decisions regarding the place of end-of-life care (Hertogh *et al.* 2004), indicating that care staff find it important to encourage self- reliance in clients.

### *4.1.3. Role of the family*

The family's wishes were explicitly considered more often (25.7%) than the wishes of the person with ID (15.7%) and professionals' wishes (12.1%) when making decisions about potentially burdensome interventions. Moreover, professionals, especially care staff, frequently believe that if people are unable to decide for themselves, the opinion of relatives should be decisive rather than the opinion of the physician. Wagemans et al. (2013a) also found that substantial weight is given to the opinions of the relatives of people with ID in the case of end-of-life decisions. According to the Dutch Medical Treatment Contract Acts (WGBO), a patient should give informed consent to a proposed medical intervention. If a person lacks decision-making capacity, a representative (usually a relative) can give consent to the medical intervention. Making medical decisions in end-of-life care is the joint responsibility of all involved, with physicians having ultimate formal responsibility. It is noteworthy that only 60.8% would respect a refusal by a person with ID of a potentially burdensome intervention, and that 60.1% believe decision-making capacity should be tested after a refusal. This indicates that refusals are complicated and raise questions about decision-making capacity.

### *4.1.4. Strengths and weaknesses*

A strength of this study is that we studied both actual considerations and personal beliefs, which gave a more profound insight into decisions on medical interventions in the end-of-life care of people with ID. Another strength is that we were able to incorporate the perspectives of three different groups of professionals who work with people with ID: care staff, ID physicians and GPs.

A limitation is that we framed the respondents' choice of medical interventions by giving them a set list to choose from, which may have influenced the respondents' choices. A second limitation is the possible recall bias regarding the last client with ID for whom the respondent provided end-of-life care: professionals may be more likely to recall clients who made a great impact on them. A third limitation is that we do not know what the personal input and role of the respondent was in the decision-making process. Different professionals may have other ideas about which considerations are relevant or decisive in decisions about medical interventions. Moreover, the actual considerations of GPs, as well as the perspectives of people with ID and their relatives were not included in this study. More in-depth qualitative research on the course of decision-making processes is necessary, including different perspectives and responsibilities.

### *4.2. Conclusion*

Quality of life and wellbeing were frequently considered in both decisions to start or continue an intervention and decisions to forgo or withdraw an intervention. Although respondents agree that quality of life is highly important, the results give a diffuse picture of decision making about medical interventions in the end- of-life care for people with ID. Professionals hold varying beliefs on whether people with ID should always be informed about medical interventions and whether a refusal by the person with ID should be respected. Moreover, the wishes of people with ID (especially of those with severe/profound ID) were often not considered in decisions about potentially burdensome medical interventions.

### *4.3. Practice implications*

The results of this study raise questions about how to actively involve people with ID in decision making and how to acknowledge their wishes: e.g. should all people with ID always be informed, how should refusals be tackled, whose opinion should prevail when decision-making capacity is lacking and how to make shared decisions with people with ID, including those with severe/ profound ID? This highlights another question, namely whether standard informed-consent procedures can adequately incorporate the preferences of people with ID. These procedures assume a liberal notion of patient autonomy where people should be able to decide without interference from others. Yet, many people with ID will have difficulties expressing themselves and need the help of others to decide about medical interventions (Bekkema *et al.* 2014a; Tuffrey-Wijne *et al.* 2009; Tyffrey-Wijne 2012). A relational notion of patient autonomy where people reach autonomy with the assistance of others may therefore be more appropriate (Bekkema *et al.* 2014a; Tronto 1993; Verkerk 1999 & 2001). To improve the active involvement of all people with ID in decisions, resources such as pictorial guides and easy read books or websites could be used to assist in this process (for resources also see http://www.pcpld.org; the Palliative Care for People with Learning Disabilities Network). Moreover, professionals can learn from existing models such as (1) the 'ASK ME' collaborative model of supported decision making for involving people with cognitive disabilities; which includes steps such as assessing the person's strengths and deficits, simplifying the task, understanding their values and maximising their ability to understand (Peisah *et al.* 2013) and (2) the model of Tuffrey–Wijne about for breaking bad news to people with ID in which small pieces of information about e.g. the intervention are added one by one to the current knowledge of the person (Tuffrey-Wijne 2012).

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# 7

# Training needs of nurses and social workers in the end-of-life care for people with intellectual disabilities: A national survey

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## Summary

**Background**
Nurses and social workers caring for people with intellectual disabilities are increasingly confronted with clients in need of end-of-life care. Previous studies, however, suggest that professionals in intellectual disability care services lack knowledge and experience concerning end-of-life care. Moreover, the proportion of nurses within the staff of intellectual disability services has declined in recent years, while the proportion of social workers has increased, which may have consequences for the quality of end-of-life care.

**Objectives**
To gain insight into the quality of end-of-life care, past vocational training, training needs and expert consultation opportunities of nurses and social workers working in intellectual disability care services.

**Design**
Survey questionnaire study conducted in the Netherlands.

**Settings**
Intellectual disability care services.

**Participants**
The study sample was recruited from an existing nationally representative research panel of care professionals. In 2011, all 181 nurses and social workers in the research panel who worked in intellectual disability care services were sent our survey questionnaire.

**Methods**
Postal survey addressing education, views and needs regarding end-of-life care.

**Results**
The response was 71.8%. Respondents positively evaluated the quality of end-of-life care. However, most respondents felt inadequately trained in end-of-life care issues. Nurses had received more training in end-of-life care and had fewer training needs than social workers. Respondents wished for additional training, especially in

supporting clients in dealing with the impending death and farewell process. Half of the respondents were unaware of the availability of external consultation facilities.

### Conclusions

This study shows that although nurses and social workers positively appraise the quality of end-of- life care for people with intellectual disabilities, the majority feel inadequately trained to provide good end-of-life care. As the number of people with intellectual disability in need of end-of-life care grows, organizations need to offer additional relevant training and must give information about the availability of external expert consultation for nurses and social workers.

### Keywords

Education Intellectual disability Nursing End-of-life care Palliative care Social work

## Introduction

Professionals caring for people with intellectual disabilities (ID) are confronted with an aging population. Since health care has improved over recent decades, the life expectancy of people with ID has increased. However, the increased life expectancy is connected with growing incidences of life-limiting illnesses such as progressive cancer, chronic cardiovascular diseases, chronic lung diseases and dementia (Ellison & Rosielle 2008; Janicki *et al.* 1999; Tuffrey-Wijne *et al.* 2007). As a result, professionals increasingly encounter people with ID who are in need of end-of-life care, a specific type of care requesting specific expertise and training.

End-of-life care focuses heavily on the quality of life by relieving pain and other distressing symptoms, incorporating the holistic needs of people, as reflected in physical, psychological and spiritual support. Professionals need a variety of knowledge and skills in order to provide comprehensive, multidisciplinary end-of-life care to the person and their family (de Vlieger *et al,* 2004). End-of-life care for people with ID can be especially challenging due to pre-existing intellectual disabilities combined with physical impairments resulting from a life-limiting illness. Professionals encounter difficulties, for example, in communicating about end-of-life issues with their clients (Bekkema *et al*. 2013; Stein 2008; Tuffrey-Wijne *et al.* 2010). Furthermore, the identification and management of pain and other symptoms can be hard at the end of life, as signals that express pain or other distress can be very small and hard to recognize in people with ID (Bekkema *et al*, 2011; Ryan *et al.* 2010). As the work of professionals caring for people with ID has a strong focus on activating people rather than on 'caring' and symptom relief, end-of-life care may not be a matter of course (Kirkendall *et al.* 2012; McCallion *et al.* 2012; Mccarron *et al.* 2010). Previous studies suggest that professionals working in ID care services may lack knowledge and skills concerning end-of-life care. Ng and Li (2003) found that in the UK, 50% of the professionals who care for incur- ably ill people with ID were unable to identify signs and symptoms of the illness. Ryan *et al.* (2010 & 2011) found that Irish professionals caring for people with ID often lacked confidence and training in end-of-life care, and that providing end-of-life care therefore led to considerable stress. Lack of training in end-of-life care was also rated as an obstacle by 72% of the directors of ID care services in the USA (Botsford 2004). Likewise, a recent study in Australian ID community group homes revealed that less than one third of the professionals concerned received any training in end of-life care (Wiese *et al.* 2012a).

Also, research performed in the Netherlands among managers and physicians in ID care services suggested that professionals often have insufficient expertise with end-of-life care (Bekkema *et al.* 2011). In the Netherlands, professionals caring for people with ID have varying backgrounds, but the large majority are social workers, followed by nurses (RNs) and certified nursing assistants (CNAs) (Van der Windt *et al.* 2009). The de-institutionalization of the care for people with ID that started in the 1970s and the disappearance of specific vocational training in ID nursing in the 1990s led to a decrease in the number of nurses working in this field in the Netherlands that is still ongoing (AVVV 2004; CBS Statistics Netherlands 2012). This necessitated a redefinition of the position of nurses working in ID care services in relation to other disciplines such as social workers; a development that also took place in other countries such as the UK and Ireland (Sheerin & McConkey 2008; Sweeney & Mitchell 2009). A survey performed in 2002 in the Netherlands revealed that 81% of the nurses working in Dutch ID care services expressed a need for additional training in end-of-life care (De Veer *et al.* 2004). No social workers were involved in the 2002 survey, but it seems probable that social workers will have received less training in end-of-life issues in their vocational training programs than nurses and that therefore their training needs will be also more pronounced. Indeed, since then there has been growing attention in the Netherlands for the quality of end- of-life care in ID care services, seen for example in the provision of training courses and consultation facilities on end-of-life care. In principle, these consultations facilities are available to all health-care professionals and may complement missing knowledge, especially for teams with no first-hand expertise in end-of-life care.

In order to further promote good care for incurably ill people with ID, more insight is needed into past vocational training in end-of-life care, the training needs and the consultation options of nurses and social workers. It is also important to know how nurses and social workers evaluate the quality of the end-of-life care provided by their team. Given their educational background, we expect that nurses will have received more training in at least the physical side of end-of-life care, will have fewer training needs and will be more aware of end-of- life care consultation facilities than social workers. The research questions are:

1. How do nurses and social workers in ID care services appraise the quality of end-of-life care provided by their team or department?
2. What views or needs do nurses and social workers have with regard to:

- training in end-of-life care?
- facilities that enable consultation of internal and external experts related to end-of-life care?

## Methods

*Sample*

The study sample was recruited from an existing nationally representative research panel of care professionals in the Netherlands (De Veer & Francke 2010; De Veer *et al.* 2011). This research panel consists of a group of care professionals who are prepared to fill in questionnaires on current topics in health care. In 2011, all 181 participants (RNs, CNAs or social workers) in the research panel who worked in care services for people with ID were sent our survey questionnaire.

*Data Collection And Survey Questionnaire*

The questionnaire contained questions on education, actual experiences and attitudes regarding end-of-life care. The questions were derived from an existing survey questionnaire which was proven to be valid and useful in earlier research (De Veer *et al.* 2004). Recent experience in end-of-life care was measured by asking the respondents: have you cared for a client who died from chronic illness or old age during the last two years? To measure the evaluation of the quality of end-of-life care of the team, respondents were asked: how do you appraise the quality of end-of-life care in your team or department? (The response categories were ‘very good’, ‘good’, ‘moderately good’, ‘moderately bad’, ‘bad’, plus ‘not applicable’ if end-of-life care was not provided by the team/department.) To measure past training in end-of-life care, the respondents were asked: what topics or skills related to end-of-life care were covered in your vocational training (basic and post basic)? A list of 15 different education topics was given, which are described in Table 3. Respondents could also indicate whether they were able to take a separate module addressing end-of-life care. In a separate question we asked: do you feel adequately trained to provide good end- of-life care? To measure current training needs in end-of-life care, the respondents were asked: in what topics or skills related to end-of-life care would you like additional training? A list of 16 different training needs was given, which are described in Table 4. To measure consultation facilities related to end-of-life care, the respondents were asked whether experts on end-of-life care could be consulted within and outside their organization, and if this was not the case, whether they would like such consultation facilities to become available. Also, some questions on background characteristics and work-related characteristics were included.

### *Ethical Consideration*

As this was a questionnaire-based study without any intervention, no approval by an ethics committee is required in the Netherlands.

### *Analysis*

T-tests and Chi-square tests were used to describe background characteristics of the respondents. In order to answer research question 1, multiple linear regression was performed to explore differences in perceived quality of end-of-life care by professional background, corrected for the influence of recent experience in end-of-life care. In order to answer research question 2, multiple logistic regression analysis was performed to explore differences in past vocational training and training needs by professional background, corrected for the influence of recent experience in end-of-life care. Odds ratios and 95% confidence intervals were calculated. Poisson regression was performed to explore differences in the mean number of end-of-life care topics covered in vocational training and the mean number of different training needs by professional background, corrected for the influence of recent experience in end-of-life care. Differences in the perceived availability of consultation facilities by professional background were tested using Chi-square tests.

An adjustment weight was assigned to nurses and social workers in order to calculate total group percentages that more accurately represent the population distribution of nurses and social workers working in Dutch ID care services. The tables present percentages for nursing staff and social workers separately, and weighted percentages for the total group of respondents. All analyses were performed using Stata 12.1.

## Results

### *Background Of The Respondents*

130 questionnaires were completed (a response of 71.8%). The group of nursing staff is comprised of nurses with an associate degree or Bachelor's degree (n = 87),

and also includes a small group of certified nursing assistants (n = 6). The group of 37 social workers is comprised of social workers with an associate degree or Bachelor's degree. Table 1 shows that the nurses in this study are on average older and have more work experience as a qualified care professional than the social workers. One in five was involved in end-of-life care in the last two years. Only a small percentage (4%) work in a team or department specialized in end-of-life care. The responding nurses have more experience in providing end-of-life care in the last two years (38%) than social workers (14%).

Table 1 background of the respondents, by profession

| | Total (weighted) | Nursing staff (n=93) | Social workers (n=37) |
|---|---|---|---|
| Type of professionals (%) | | 71.5 | 28.5 |
| Age (mean) [1] | 44.0 | **47.3** | **42.7** |
| Work experience in years (mean) [2] | 17.4 | **22.2** | **15.2** |
| Recent experience in end-of-life care: cared for terminally ill clients during the last two years (%) [3] | 21.0 | **37.6** | **13.9** |
| Works in a team or department specialized in end-of-life care (%) | 4.3 | 6.0 | 2.9 |

differences p<.05 :

[1] t = 2.6657, p=0.009

[2] t = 4.2247, p=0.000

[3] Chi-square = 6.8405, p=0.009

### *Appraisal Of The Quality Of End-Of-Life Care*

Table 2 shows that the quality of end-of-life care is highly valued. Almost 80% of the respondents perceived the quality of end-of-life care delivered by their team or department to be good or very good. No relationship was found between the professional background and appraisal of end-of-life care.

**Table 2** Appraisal of the quality of end-of-life care by own team/department

| **Evaluation** (n=92) | **Total (weighted)** | **Nursing staff (n=67)** [1] | **Social workers (n=25)** [1] |
|---|---|---|---|
| (Very) good (%) | 79.7 | 79.1 | 80.0 |
| Moderately good (%) | 19.4 | 17.9 | 20.0 |
| (Moderately) bad (%) | 0.9 | 3.0 | 0.0 |
| Evaluation score for quality of end-of-life care (mean, range 1-5, 5 = very good) | 3.86 | 3.99 | 3.80 |

[1] Only respondents whose team/department provided end-of-life care answered this question.

### *Past Training In End-Of-Life Care*

Table 3 shows that only 15% of the respondents felt adequately trained to provide good end-of-life care. No differences related to professional background were found. In general, most respondents (78%) covered at least one aspect of end-of-life care in their vocational training (basic and/or post basic). However, only 6% took a separate module on end-of-life care. Topics concerning psychosocial and spiritual care were most likely to be covered, but no end-of-life topic was covered in the vocational training of more than 42% of the respondents. With the exception of the physical care of a deceased client, end-of-life topics related to the physical and medical care, in particular the use of instruments to measure pain or other symptoms, were barely covered in the vocational training. Nurses were more likely to receive training in the physical care of a deceased client than social workers. Within the category psychosocial and spiritual care, the topics most likely to be covered were support in dealing with the impending death and conducting 'bad–news' conversations. Religious and spiritual support, and conversations about the meaning of life were less likely to be covered. Nurses were significantly more likely to be trained in the provision of support in dealing with the impending death than social workers. Within the category practical and complementary care, more nurses were trained in practical issues surrounding the dying process and the funeral than social workers. Complementary care such as mas- sage and end-of-life care in other cultures were covered less often. Within the category personal and team development, nurses were more likely to have been educated in cooperating with other professions in end-of-life care than social workers.

The mean number of different end-of-life care topics covered in vocational training was 2.9 (range of 0–13). Nurses received training on more topics (3.8) than did social workers (2.5) (Coef: − 0.3894 [− 0.632 to − 0.147]).

Table 3 Past vocational training (basic and post basic) in end-of-life care received by nurses and social workers

| | Total (weighted) % | Nursing staff % | Social workers % |
|---|---|---|---|
| **Perception of end-of-life care training** | | (n=80) | (n=30) |
| Feels adequately trained to provide good end-of-life care | 15.4 | 27.5 | 10.0 |
| | | (n=84) | (n=34) |
| **Took separate module on end-of-life care** | 5.9 | 13.1 | 2.9 |
| **End-of-life care topics covered in vocational training (basic and/or post basic)** | | (n=93) | (n=37) |
| Received at least one end-of-life care topic | 77.8 | 82.8 | 75.7 |
| *Physical and medical care* | | | |
| Physical care of deceased client [1] | 20.6 | **50.5** | **8.1** |
| End-of-life decisions (e.g. starting or forgoing treatment) | 14.6 | 17.2 | 13.5 |
| Palliative sedation | 7.6 | 12.9 | 5.4 |
| Law and regulations on euthanasia and assisted suicide | 7.3 | 11.8 | 5.4 |
| Use of instruments to measure pain and other symptoms | 2.5 | 8.6 | - |
| *Psychosocial and spiritual care* | | | |
| Support in dealing with the impending death [2] | 36.8 | **53.8** | **29.7** |
| Conducting 'bad–news' conversation | 42.2 | 39.8 | 43.2 |
| Religious and spiritual support | 21.9 | 16.1 | 24.3 |
| Talking about the meaning of life | 19.1 | 12.9 | 21.6 |

*- Table 3 continues -*

*Table 3 continued -*

| | Total (weighted) % | Nursing staff % | Social workers % |
|---|---|---|---|
| *Practical and complementary care* | | | |
| Practical issues surrounding the dying process and the funeral (e.g. clothing of the client) [3] | 25.7 | **41.9** | **18.9** |
| Complementary care (e.g. massage) | 6.0 | 14.0 | 2.7 |
| Knowledge about end-of-life care in other cultures | 9.9 | 7.5 | 10.8 |
| *Personal and team development* | | | |
| Cooperation with other professions in end-of-life care [4] | 18.4 | **36.6** | **10.8** |
| Dealing with own grief | 28.6 | 25.8 | 29.7 |
| Reflecting on own ideas and thoughts on end-of-life care | 22.9 | 25.8 | 21.6 |

Odds ratios (OR) are corrected for the influence of recent experience in end-of-life care (0= nurses, 1= social workers) $p<.05$:

[1] OR: 0.104 [0.029-0.368], p=0.000

[2] OR: 0.353 [0.150-0.829], p=0.017

[3] OR: 0.379 [0.148-0.971], p=0.043

[4] OR: 0.183 [0.051-0.654], p=0.009

### *Training Needs*

Table 4 shows that almost 70% felt a need for additional training in end-of-life care. In general, respondents were most interested in training on how to support clients in dealing with the impending death and the farewell process, knowledge about end-of-life care in other cultures, and law and regulations on euthanasia and assisted suicide. There were no significant differences between nurses and social workers regarding training needs.

Table 4: Needs for additional training among nurses and social workers

| | Total (weighted) % | Nursing staff (n=86) (%) | Social workers (n=36) (%) |
|---|---|---|---|
| Has end-of-life care training need(s) | 67.9 | 70.9 | 66.7 |
| **Topics in which training is needed** | | | |
| *Physical and medical care* | | | |
| Law and regulations on euthanasia and assisted suicide | 35.4 | 33.7 | 36.1 |
| Palliative sedation | 25.8 | 27.9 | 25.0 |
| End-of-life decisions (e.g. starting or forgoing treatment) | 24.5 | 23.3 | 25.0 |
| Technical instrumental care (e.g. catheter or ostomy care, wound care) | 9.9 | 20.9 | 5.6 |
| Conducting conversations on physical condition and care needs | 17.2 | 11.6 | 19.4 |
| Basic physical care (e.g. washing, feeding, clothing) | 13.3 | 4.7 | 16.7 |
| Physical care of deceased client | 7.3 | 4.6 | 8.3 |
| *Psychosocial and spiritual care* | | | |
| Support in dealing with the impending death and the farewell process | 43.7 | 34.9 | 47.2 |
| Conducting meaning of life conversations | 22.9 | 17.4 | 25.0 |
| Conducting 'bad-news' conversations | 22.2 | 15.1 | 25.0 |
| Religious and spiritual support | 15.6 | 5.8 | 19.4 |
| *Practical and complementary care* | | | |
| Knowledge about end-of-life care in other cultures | 39.4 | 33.7 | 41.7 |
| Complementary care (e.g. massage) | 15.6 | 19.8 | 13.9 |
| Practical issues surrounding the dying process and the funeral (e.g. clothing of the client) | 9.0 | 3.5 | 11.1 |
| Home care (e.g. cleaning, making dinner) | 0.7 | 2.3 | - |
| *(Inter)personal development* | | | |
| Dealing with own grief | 21.9 | 14.0 | 25.0 |

No differences were found between the professions (correcting for the influence of recent experience in end-of-life care).

Concerning physical and medical care, respondents were most likely to want to be educated in law and regulations on euthanasia and assisted suicide, palliative sedation and end-of-life decisions such as starting or foregoing treatment. Concerning psychosocial and spiritual care, respondents primarily wished to be trained in how to support a client in dealing with the impending death and the farewell process. Concerning practical and complementary care, respondents primarily wished for training in knowledge on end-of-life care in other cultures. One in five respondents wished to be trained in dealing with their own grief. The mean number of training needs was 3.3 (range of 0–13). Social workers had more training needs (3.6) than nurses (2.8) (Coef: 0.275 [0.048 to 0.503]).

*Perceived Availability Of Consultation Facilities And Need For Them*
79% of the respondents said that internal expert consultation on end-of-life care is already available in their organization, whereas 8% expressed a need for internal consultation facilities. However, only 50% of the respondents stated that external consultation facilities are already in place, whereas 24% expressed a need for this type of consultation. We found no differences between nurses and social workers in this regard.

## Discussion

Nurses and social workers working in ID care services positively evaluate the quality of end-of-life care provided by their team or within their department. Nurses are more involved in end-of-life care, received more training in end-of-life care and have fewer training needs than social workers. Although most professionals received training in at least one end-of-life care topic, many topics were not covered well in (basic and/or post basic) vocational training. Moreover, the large majority of care professionals feel inadequately trained to provide end-of-life care. The top training priority is how to support clients in dealing with the impending death and the farewell process. Although external consultation on end-of- life care is supposed to be available to all healthcare professionals, half of the respondents said that these facilities were unavailable to them.

The positive evaluation of the quality of end-of-life care seems to contradict the finding that most respondents feel inadequately trained and have extensive training needs when it comes to end- of-life care. Since most of the respondents had not cared for a dying client in the past two years, an explanation may be that although they feel insufficiently equipped to provide end-of-life care themselves, they have a high opinion of the quality of end- of-life care provided by more experienced colleagues. The positive evaluation of the quality of end-of-life care may also have been influenced by emotions, as studies suggest that care professionals often experience end-of-life care as very rewarding and honorable (Bekkema *et al.* 2011; Wiese *et al.* 2012a).

### *Education Falls Short When It Comes To End-Of-Life Care*

Only a small percentage of the respondents had taken a special course on end-of-life care, and in general end-of-life care was not covered very well in the (basic and/or post basic) vocational training they had received. The topics covered most frequently concerned support in dealing with the impending death, conducting 'bad-news' conversations, the physical care of a deceased client and practical issues surrounding the dying process and the funeral. The topic that received the least attention was the use of instruments to measure pain and other symptoms, a shortage in education which has also been found among nursing home nurses (Whittaker *et al.* 2006). This is alarming, since pain and symptom relief are important parts of end-of-life care (WHO 2012). Earlier research suggests that the identification and management of pain and other symptoms can be hard in people with ID, especially at the end of their life (Bekkema et al., 2011; Ryan et al., 2010). Since almost no respondents received training in the use of assessment instruments, more awareness is needed of the complexities of assessing pain and distress in people with ID (Symons *et al.* 2008; Regnard *et al.* 2007).

Strikingly, 85% of the respondents feel inadequately trained to provide end-of-life care. This is in line with the lack of training which has been found in other countries (Ng & Li 2003; Ryan *et al.* 2010 & 2011; Wiese *et al.* 2012a). Here is a task for organizations, teachers and policy makers to investigate what can be done to increase the knowledge and expertise of current and future nursing staff and social workers. End-of-life care is still a rather infrequent type of care in people with ID, although the numbers in need of end-of-life care will expand in the years to come. Studying the knowledge of nurses in the US, Schlairet (2009) found that only nurses with continuing education in end-of-life care (and not the ones with only formal education in end-of-life care) scored better on objective knowledge/skills concerning end-of-life care. It may therefore be most effective to provide continuing education on end-of-life care issues to those professionals who are most likely to be confronted with clients with end-of-life care needs, e.g., those who work with older people with ID. There is a need for education interventions to reduce the gaps in knowledge and skills. For instance, Reddall (2010) developed a palliative care resource booklet for professionals caring for people with intellectual disabilities, aimed at providing easy and accessible information on end-of-life care. Other work is being done by McLaughlin *et al.* (2012) who developed a multi- media educational resource about end-of-life care which stimulates partnerships between ID care services and specialist palliative care services. Also, promising results came from a recent evaluation of a training program on palliative care for staff working with people with ID: staff members' levels of preparation and confidence in their ability to provide palliative care improved due to the program (Hahn & Cadogan 2011).

### *Training Priorities*

The many needs for additional training in end-of-life care illustrates a willingness among professionals to provide good end-of- life care, and is in line with the willingness found in other studies (Ryan *et al.* 2010 & 2011; Wiese *et al.* 2012a). Compared to the 2002 survey (De Veer *et al.* 2004), the overall need for end-of- life training decreased by 10 percentage points, which may be due to the growing attention for the quality of end-of-life care and the development of supporting material in Dutch ID care services in recent years. However, training needs are still very high. Just as in 2002, the top training priority remains training in how to support clients in dealing with the impending death and the farewell process. This training priority is a reminder that communication between care professionals and people with ID remains a challenge in end-of-life care, as has also been found in other studies (Stein 2008; Tuffrey-Wijne *et al.* 2007 & 2010). Fortunately, promising research is currently being done in this matter (Tuffrey-Wijne 2012; Wiese *et al.* 2012b), although new ways for communication in the end-of-life care for people with ID still need testing in practice.

### *Lack Of Access To External Experts*

End-of-life care should be comprehensive and multidisciplinary. This is reflected in our results as the majority of nurses and social workers confirmed the availability of consultation with experts on end-of-life care within the organization. This exchange of knowledge seems particularly important since professionals in ID care services have varying education backgrounds and may not feel adequately trained. Within the context of knowledge exchange, a specialized palliative care consultant may function as a key person in the transfer of internal expertise. However, half of the professionals said they cannot consult relevant experts outside their organization. This is surprising as multidisciplinary consultation teams are available in all regions in the Netherlands and can be consulted by all kinds of healthcare professionals with questions about physical, psychosocial or spiritual aspects of end-of-life care. Hence, professionals working in ID care services are often not aware of these consultation facilities. Partner- ships between ID care services and specialist palliative care services may considerably improve the quality of end-of-life care for people with ID. Collaborative working can increase the knowledge on end-of-life care of professionals working in ID care services and it may improve the access of people with ID to generic palliative care services. Such partnerships require overcoming differences in care approaches and skill limitations (Friedman *et al.* 2012; Kirkendall *et al.* 2012; Mccarron *et al.* 2010; Read & Cartlidge 2012; Ryan *et al.* 2010; Tuffrey-Wijne *et al.* 2008).

### *Nurses And Social Workers: Differences And Opportunities For Holistic Care*

Nurses received more training in several aspects of end-of-life care. As for the physical care of a deceased client, this may be a more natural fit with nursing curriculum than in social-work curriculum. Yet, the other topics in which nurses scored higher – support in dealing with the impending death, practical issues surrounding the dying process and the funeral, and cooperating with other professions in end-of-life care – could also fit well within social-work education. The question arises whether nurses and social workers should receive the same training in end-of-life care, and whether this is feasible. In selecting their basic vocation- al training, most social workers did not choose to provide physical and medical care to clients. Nevertheless, end-of-life care usually involves a lot of physical care tasks. In Dutch ID care services, nurses and social workers generally work in similar positions. The division of tasks between the professions is not clear, but in practice nurses may be more engaged with clients who have special nursing care needs, and may therefore more readily opt to care for a client who becomes ill. This would be in line with the results of our study that nurses are more involved in end-of-life care. However, as clients get ill in all types of care services, social workers will be confronted with end-of-life care needs as well. A positive finding is that social workers are aware of their responsibility to have sufficient expertise in end-of-life care, as they expressed more training needs in terms of end-of-life care than nurses. Moreover, social workers seem to acknowledge that their task goes beyond the psychosocial part, seeing that they also wish for training in the physical and medical aspects of end-of- life care. As end-of-life care is eminently holistic, multidisciplinary teams in ID care services, composed of nurses as well as social workers, may allow for the ideal combination of psychosocial, spiritual and physical expertise.

### Strengths and Weaknesses

An important strength of this study is that respondents were recruited from an existent large, national panel of nurses and social workers. We were therefore able to provide insights into how staff working in ID care services perceive their knowledge and skills in end-of-life care. Another strength is the relatively high response rate; this might be due to the fact that all participants in the panel are in principle committed to completing survey questionnaires regularly. However, using the existing panel also has a disadvantage in the sense that staff who are highly committed and dedicated to their profession will probably be over-represented in our study sample. Additionally, there is a limitation in that most questions and statements in the current study were about perceptions. Moreover, recall of basic vocational training topics may have been a problem for older respondents. Future research should therefore focus more on their actual knowledge and skills.

### Implications

This study raises the question of whether end-of-life care for a person with ID can best be provided within an ID care service, taking into account the lack of knowledge of the nurses and social workers. As the number of people with ID and end-of-life care needs is expected to grow, organizations must take adequate measures in order to equip care staff with the necessary expertise to answer the end-of-life care needs of their clients. Dying in one's own home environment is the preferred option for many dying people (Escobar Pinzón *et al.* 2011; Higginson & Sen-Gupta 2000), and also for many incurably ill people with ID. Organizing end-of-life care is challenging, perhaps especially since many people with ID now live in small-scale housing facilities. If we want to promote end-of-life care in the client's own home environment, good quality end-of-life care must be guaranteed. Combining opportunities for expert consultation with continuing education on end-of-life care for those professionals who are most likely to be confronted with incurably ill clients may be most effective. Being a well-skilled professional in end-of-life care also entails reflecting on one's own expertise and engaging the support of experts from outside and inside the organization for good end-of-life care for people with ID.

## Conflicts Of Interest

The authors have declared no conflicts of interest.

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# 8

# General discussion

The aim of this thesis was to explore end-of-life care for people with ID from the perspectives of people with ID, relatives and professionals. In this final chapter, first the principal findings in Chapters 2 to 7 are presented. This is followed by reflections on these results, methodological considerations and discussion of the implications for practice, policy and research.

### Findings about care approaches, needs and wishes

Two qualitative studies were conducted to explore shifts in care approaches and attitudes among professionals and relatives, the important dimensions of the caring relationship at the end of life from the perspectives of people with ID, and how the autonomy of people with ID is respected at the end of life.

#### *Towards wellbeing and comfort care by being there for the person*

In qualitative interviews with relatives and care staff, we found that at the end of life of a person with ID, the care approach shifted from a focus on activation and social participation towards a focus on wellbeing and comfort, taking over tasks and symptom relief (Ch. 2). In doing this, relatives and care staff increasingly had to let go of their usual care strategy and adapt to what was still possible.

Within end-of-life care, being there and making more time available to be close to the ill person were regarded as important by both care staff and relatives, as well as by people with ID themselves. Care staff and relatives regarded being there as an important value underlying the shift towards comfort care and taking over tasks. In group interviews, people with mild ID also highlighted being there as one of the most important dimensions of caring relationships at the end of life (Ch. 3). In the view of people with ID, an ill person needs dedicated people who are there for him/her at the end of life, to provide adequate practical, emotional, social and spiritual support. This highlighted the opinion of people with ID that being there should cover all aspects of end-of-life care.

People with ID also emphasized the importance of creating positive, active experiences while being there for the ill person; e.g. going on day trips or a longer holiday, 'living life to the full' and fulfilling last wishes. So although activating the person becomes less important, opportunities to undertake positive and active experiences in the remaining life of an ill person with ID should still be created.

### *'Reading' needs and jointly interpreting signals*

Another shift in the care approach of relatives and professionals concerned a stronger reliance on the joint interpretation of signals expressing distress, as verbal communication with the person with ID was often hampered (Ch. 2). Physicians who were interviewed about cases of people with ID receiving end-of-life care indicated that they relied heavily on information from people closely involved with the person, mostly relatives and care staff. Finding a common language to talk about signals could be a hurdle.

Reading needs was particularly demanding for relatives of people with severe/profound ID: the dependency of these persons became magnified, leading to another shift in their care approach. Properly interpreting symptoms could be further hampered by a lack of knowledge among ID care staff, e.g. about basic end-of-life care and the use of instruments to measure pain or other symptoms (Chs. 4 and 7).

'Attentiveness' was regarded as important for reading needs by both professionals and relatives and by people with ID. Attentiveness to the signals of distress and pain, preferably by people who are closely involved, was recognized as a value underlying the shift towards stronger reliance on joint interpretations (Ch. 2). People with mild ID also recognized that a person who cannot talk needs familiar people who can closely observe his/her behaviour and facial expressions (Ch. 3).

### *The involvement of care staff and relatives*

When the death of a person becomes imminent, the interweaving of emotional and professional involvement increasingly becomes a struggle for ID care staff, leading to another shift in their care approach (Ch. 2). Care staff have often built strong long-term caring relationships with their clients, and struggle to achieve a balance between warm care and professional distance. Being able to reflect on their own emotions, capabilities and caring relationships was an important value underlying this shift.

The growing awareness that the person with ID actually has two 'families' - relatives and the care staff - constituted the last shift made in end-of-life care (Ch. 2). The proximity of these two 'families', who know the person best and wish to offer him/her warm and loving care, could be very valuable. Yet if perceptions of what constitutes good care differ between care staff and relatives and communication fails, the good intentions to provide joint care can turn into a burden and source of distress for the dying person. The lack of end-of-life care expertise may put ID care staff on a same level in terms of knowledge as relatives (Chs. 2 and 7). Yet some relatives expected care staff's expertise to be on a higher level, which could put extra pressure on the relatives' cooperation with care staff (Ch. 2).

*Respecting autonomy and wishes*

The qualitative studies also highlighted the importance of respecting the autonomy of people with ID at the end of life, as well as the challenges that are inherent to such a difficult task. According to people with mild ID, properly handling a person's wishes is a central element of caring relationships at the end of life (Ch. 3). This handling of wishes concerned care wishes and wishes for a final pleasant event as well as wishes regarding the funeral. 'Handling wishes' referred not only to ascertaining wishes but also to recording and - especially - honouring them.

Yet, dealing with wishes and respecting autonomy does not always happen as a matter of course, as indicated by both people with ID and the people caring for them. Some people with ID were sceptical about the honouring of wishes or talked intensely about their fear that their end-of-life care wishes would not be honoured. This seemed influenced by their prior life experiences (Ch. 3). In addition, relatives and professionals encountered numerous challenges in respecting the autonomy of a person with ID, such as eliciting current and hidden last wishes, dealing with conflicting wishes, ascertaining information needs and communicating about illness and death (Ch. 4). Many relatives and professionals struggled for example to find a balance between protecting the person with ID and giving them more information.

Trusting, caring relationships were found to be crucial for acknowledging and respecting the wishes of a person with ID. Autonomy could best be seen as a product of joint work that imposes high demands on relational qualities, such as attention to information needs, connecting with the person and giving them the space to show their wishes and preferences (Chs. 3 and 4).

### Findings about decision making

*Place of care: familiarity of the environment and end-of-life care expertise*

A quantitative survey study was conducted to investigate the considerations and beliefs of care staff, ID physicians and GPs when confronted with difficult end-of-life decisions. Receiving end-of-life care in the person with ID's own home environment, and the 24/7 availability of end-of-life care were important considerations in decisions about the place of end-of-life care (Ch. 5). Almost all professionals were in favour of keeping the person at home, which may also relate to the strong bonds between care staff and their clients (Ch. 2). Indeed, most of the clients described in the case section of the survey were able to stay in their own home environment, which is a larger number than in the general population. The main considerations to keep the person at home were the familiarity of the environment and the end-of-life care expertise of the team. Insufficient expertise and a lack of adequate equipment were main considerations to transfer a person to a different setting.

*Use of medical interventions: quality of life and wellbeing*

Quality of life was the consideration most frequently mentioned in decisions about potentially burdensome medical interventions, such as chemotherapy, life-prolonging surgery, pain medication by infusion pump or further diagnostic investigations (Ch. 6). In addition, the client's wellbeing was frequently considered in both decisions to start or continue an intervention and decisions to forgo or withdraw an intervention. Most decisions concerned the rejection or withdrawal of the intervention.

*Involving the person with ID in decisions*

The beliefs of professionals regarding the wishes of the person with ID differed between decisions about the place of care and decisions about the use of medical interventions. Professionals predominantly believed that the wishes of the person should always be leading when deciding upon the place of end-of-life care (Ch. 5). Yet only a minority of professionals believed that the wishes of the person with ID should always be leading in decisions about medical interventions (Ch. 6). It may be that professionals believe that people with ID are less able to decide about complex medical matters than about more practical matters.

In both type of decisions, explicit involvement of the person’s wishes was low in practice and relatives’ wishes were more likely to be explicitly considered. Only a small minority of professionals explicitly mentioned that the wishes of the clients were taken into account in practice (Chs. 5 and 6). In particular the wishes of people with severe/profound ID were often not explicitly taken into account. Involving people with ID in decisions and respecting their wishes seemed not to happen as a matter of course, which is in line with the findings of Chapter 3 and Chapter 4.

### Findings about the training needs, expertise and resources of ID care staff

Expertise is needed to ensure good end-of-life care for clients with ID. A quantitative survey was conducted to evaluate the quality of end-of-life care as perceived by social workers and nurses working in ID care services, their training experiences and training needs, and consultation options regarding end-of-life care.

Although ID care staff positively evaluated the quality of end-of-life care provided by their team or within their department, only 15% felt adequately trained to provide good end-of-life care (Ch. 7). Care staff wished for additional training, particularly in supporting clients in dealing with the impending death and the farewell process. Expertise is important to provide end-of-life care in the home environment, as we found that lack of expertise was the foremost argument for transferring a client to another care setting (Ch. 5).

Although palliative care consultations teams are available for consultation all over the Netherlands for all healthcare professionals, half of the respondents were unaware of the availability of these services. As expected, nurses were more involved in end-of-life care, had received more training in end-of-life care and had fewer training needs than social workers. This is in line with the finding in Chapter 4 that social workers struggled to respond adequately to a client’s needs because of a lack of nursing skills and basic end-of-life care knowledge.

## Reflections on the quality of end-of-life care

### *The value of familiar people being there*

It is generally known that people with ID are a vulnerable ageing group because of their poor health status (e.g. Tuffrey-Wijne *et al.* 2007a; Hermans & Evenhuis 2014; Mccarron *et al*. 2013). This thesis indicates that at the end of life of people with ID, their dependency and vulnerability seems even more pronounced, for instance because of the complexity of assessing their needs. Being close to familiar caregivers and being in a familiar surrounding may be particularly important for this vulnerable group. The findings of this thesis highlight that it is vital for people with ID to be surrounded by familiar care staff and relatives, who know how to 'read' their needs and identify signals such as distress and pain. Relatives and care staff 'who are there' and take time to acknowledge and address these needs can therefore be a big strength in enhancing the quality of life, as indicated by both people with ID and relatives and professionals in our studies. The power of 'being there' has also been acknowledged by Todd (2013), who found that ID care staff experienced a client's death as a 'good death' when they were able to 'be there' for the person with ID in the different phases of end-of-life care.

'Being there' and providing good end-of-life care impose high demands on the relational qualities of professionals and relatives, as also highlighted by this thesis. Qualities that were found to be important included the ability to uncover wishes, listen carefully, empathize and closely observe the behaviour of the person with ID. These qualities fit with the core values of 'good care' in care ethics, such as attentiveness to recognize the need for care (e.g. Leget 2007; Tronto 1993). Leget for example emphasized the value of 'inner space': an attitude of listening attentively to dying patients with an open heart and mind in order to unravel their real needs and questions (Leget 2007). The relational qualities also align with the vision that care is reciprocal (Hertogh 2009; Hertogh 2010) and the awareness that care should be provided in such a careful way that the person receiving the care is able to be responsive to this care (Hertogh 2009). Recognizing care needs and responses to care in ill people with ID can be quite complex. The proximity of ID care staff who are experienced in communicating with people with ID, and who have often built a long-term relationship with a person with ID, can therefore provide people with ID with a sense of security that may be particularly important for them.

### *Responsibility and expertise in providing good end-of-life care*

Caring also implies taking responsibility for providing this care, another core value of 'good care'. The care staff who participated in our studies predominantly felt strongly about their wish to remain the primary caregiver until the end. This strong willingness to provide end-of-life care has also been found in other studies among ID care staff (Wiese *et al.* 2012; Mccarron *et al*. 2010). This indicates that ID care staff do wish to take on the responsibility for providing end-of-life care.

Yet, in line with care ethics, competence is a key core value of end-of-life care in addition to responsibility. Tronto (1993), a care ethicist, reasons for instance that although there may be an intention to provide good care, care needs cannot be met if there is a shortage of knowledge or skills. And it is precisely this competence that seems to be a weakness within ID care services: only 15% of the care staff respondents in our study felt adequately trained to provide good end-of-life care. This lack of expertise has also been found in several other studies of end-of-life care for people with ID (e.g. Ng & Li 2003; Ryan *et al*. 2010; Botsford 2004, Wiese *et al.* 2012, Mccarron *et al.* 2010). We found that in particular the use of instruments to measure pain or other symptoms was barely covered in the vocational training of care staff. This is alarming since pain and symptom relief is a very important part of end-of-life care (WHO 2015), and the identification and management of pain and other symptoms can be particularly hard in the case of people with ID, especially at the end of their life (e.g. Ryan *et al.* 2010).

Although the care staff in our studies were often willing to provide end-of-life care, some were also insecure or felt incompetent, in particular if they had never provided end-of-life care before. Possible reasons for a lack of expertise are a general deficiency in the body of knowledge with regard to end-of-life care for people with ID (e.g. symptom and pain management and communication), and a shortage of end-of-life care training for ID care staff. If care staff do not have the expertise to recognize end-of-life care needs, they may have trouble identifying when care is required. A lack of knowledge and skills among ID care staff may therefore lead to poor end-of-life care, e.g. because they miss signs of pain, are not able to recognize or correctly treat particular symptoms such as pain, anxiety or swallowing disorders, or do not know how to comfort the client. A telling point is our finding that a lack of expertise was the most frequently cited consideration in decisions to transfer the person with ID to another care setting.

*Opportunities for providing good quality end-of-life care within ID care services*

Although this thesis highlights the aspiration among ID care staff to take on end-of-life care for their clients, additional relevant training is essential for ensuring good end-of-life care for people with ID. Additional training should take into account the holistic nature of end-of-life care, as people with ID have physical, emotional, social and spiritual needs that are similar to those of the rest of the population (Tuffrey-Wijne & McLaughlin 2015). As ID care staff teams are often multidisciplinary, composed of nurses as well as social workers, a well-trained care staff team could potentially allow for a good 'skills mix' within the team.

Moreover, intensified partnerships between ID care services and specialist end-of-life care services could also considerably improve the quality of end-of-life care within ID care services. End-of-life care is still a rather infrequent type of care in ID care services, and ensuring that all ID care staff have sufficient knowledge to address the full scope of possible end-of-life care needs would be impossible, and indeed not desirable. That is where the input of end-of-life care specialists may make a big difference. As ID care staff are experienced in supporting people with ID and identifying their particular needs, and end-of-life care specialists can bring in specialized end-of-life care skills, collaboration between these services is key to the successful provision of end-of-life care for people with ID (EAPC European white paper on end-of-life care for people with ID; Tuffrey-Wijne & McLaughlin 2015). But although cooperating with specialized end-of-life care services and drawing on their expertise may help to significantly improve the quality of end-of-life care for people with ID, our study shows that many ID care staff members are still unaware of these external facilities. So in order to improve the access of people with ID to these services, more information should be provided about the option of partnerships between ID care services and specialist end-of-life care services.

## Reflections on the wishes of people with ID

Being able to die according to your own preferences is generally found to be an important value in end-of-life care (Patrick *et al.* 2003). Also, the 'Caring at the End of Life Position Statement' by the American Association on Intellectual and Developmental Disabilities sees honouring wishes and autonomy as an important principle (AAIDD 2015). Yet, although the significance of these principles is undisputed, they do not tell us how they relate to the complexity of the daily care for people with ID who have an incurable illness.

*"You should listen to her wishes. Meet her wishes. It is about her. What she wants. And how she wants it. That's the last thing you can do for her"*

This quote by Marcel (mild ID) reflects the finding of this thesis that people with ID, in line with the general principles, indeed believe that honouring the wishes of the incurably ill person is central to good end-of-life care. Professionals and relatives also predominantly believe that the person's wishes are important and that the person should be involved. Yet it is not simple, as shown by the complexity of involving people with ID in decisions and by the apparent lack of trust in the ability of the person to become involved.

*The complexity of involving people with ID in decisions*

This thesis highlights the fact that professionals hold varying beliefs regarding people with ID's involvement in decisions. For instance, professionals predominantly believed that the wishes of the person are leading in decisions about the place of end-of-life care, but not in decisions about medical interventions. Moreover, although being informed about the diagnosis and treatment options is the basis for informed decision making, less than 70% of professionals believed that people with ID should always be informed about medical intervention options. What may play a role is that professionals wish to protect people with ID from complex medical information that they presumably cannot fully understand, or that they prefer to inform a relative or other representative.

This thesis also showed that the explicit consideration of people with ID's wishes in practice was rare, in particular of the wishes of people with severe/profound ID. The wishes of relatives were more likely to be explicitly considered than the wishes of the person with ID. This is in line with other studies on end-of-life care for people with ID, which showed that people with ID are not always involved in communication and decisions (Tuffrey-Wijne *et al*. 2009; Tuffrey-Wijne *et al.* 2010; Wagemans *et al.* 2010; Wagemans *et al*. 2013). Although our research did not provide insight into the person's decision-making capacity, it may be that the people with ID in our decision-making studies were believed to be incapable of making decisions, and that therefore their wishes were not prioritized. However, the incapacity to decide should not be assumed beforehand, as also stated by the European White Paper on end-of-life care for people with ID: people with ID should be assumed to have the capacity to make decisions concerning their care and treatment, unless this is demonstrated not to be the case (EAPC: Tuffrey-Wijne & McLaughlin 2015). Moreover, even when a person is incapable of making decisions, that does not absolve professionals and relatives from the responsibility to try to uncover his/her wishes and take these into account.

*Trust in the person’s ability to be involved*

Our findings on respecting wishes and autonomy suggest that trust in the person´s ability to be involved in end-of-life care seems to be a central issue. Many professionals and relatives in our studies found it hard to talk about illness and death to people with ID (for example, Cassandra’s caregivers in Chapter 4). They often oscillated between between providing information and protecting the person from information, as they felt that information might harm them. These professionals and relatives may lack trust in the person’s ability to be involved, or they may lack trust in their own ability to involve the person with ID. This lack of trust was also visible among the people with mild ID who were interviewed, as some of them were quite sceptical and displayed distrust when talking about the honouring of their own wishes. This distrust might be a reflection of the daily life experiences of people with ID within an ID care setting, as this can to some extent be characterized by supervision and a lack of freedom of choice.

A study by Tuffrey-Wijne *et al.* (2010) highlighted that when people with ID are not involved (or not enough) in communication or decisions related to their end-of-life care, they can feel distressed because they do not understand what is happening, and this lack of understanding affects their ability to make decisions. Although not involving someone may be motivated by a desire to protect them and by caring intentions, *protecting*, according to Tronto, does not take the needs of that person as a starting point for what must be done (Tronto 1993, p. 105). A risk is that by protecting the person with ID, their real end-of-life care needs and wishes are lost sight of. Hence, it is highly important to find the trust in the person’s ability to be involved, at a level that suits their capabilities.

*Respecting autonomy as a relational concept*

Respecting the autonomy of people with ID at the end of their lives is best seen as a relational concept, as is shown by this thesis. When talking about respecting the autonomy of people with ID, their dependency always comes to the fore. People with ID, like other people with decision-making disabilities, are vulnerable and dependent on others to maximize their participation in decisions (Peisah *et al.* 2013). A relational concept of autonomy gives more space to this dependence of people. It fits with care ethics, which regards dependency as essential to human relationships (Agich 2003; Tronto 1993; Verkerk 1999 & 2001) and in which asymmetrical relationships are seen to be as morally relevant as symmetrical relationships (Kittay 2011). Our findings suggest that respecting autonomy can be attained by relatives and professionals adopting an open, active and reflective attitude. In doing this, much can be learned from existing models, such as supported decision making for involving people with cognitive disabilities, and for breaking bad news to people with ID (Peisah *et al.* 2013; Tuffrey-Wijne 2012). When there is a basic trust that the person can be involved in his or her own way, and when relatives and professionals proactively seek means to achieve this, by e.g. assessing the capabilities of the person with ID, simplifying the information and maximizing their ability to understand, the person with ID's wishes may actually take centre stage in their end-of-life care.

## Reflections about relationships based end-of-life care for people with ID

The nature of end-of-life care for people with ID and the involvement of many different people call for good cooperation and good caring relationships. The results of this thesis confirm for example that symptom relief and uncovering the needs of a person with ID become a real team effort at the end of life: relatives, care staff and physicians have to cooperate to find a common language to talk about what they see. The European White Paper highlights the fact that collaboration between those who know the person well and those who are experts in symptom management is crucial to ensure adequate symptom management for people with ID (EAPC: Tuffrey-Wijne & McLaughlin 2015). Yet, this thesis also demonstrates that cooperation and safeguarding a professional caring relationship is not always easy.

### *About feeling one family and the ownership of the person*

Experiences of relatives regarding the cooperation with care staff varied. On the one hand, co-caring was experienced as special, powerful and constructive by relatives, and as a support for the whole family. In some cases it even felt as if care staff and relatives were acting as one family in the support for the ill person with ID. Yet, in other cases communication failed and co-caring became stressful for relatives. Some relatives experienced the end of life as something intimate that should only be shared with relatives. They experienced the input of care staff as an infringement of their privacy or rights as a representative. Others had mixed feelings regarding the quality of care in the ID organization, or the level of end-of-life care expertise among care staff. Other care settings such as nursing homes and care homes also encounter challenges in shared end-of-life care by relatives and professionals (e.g. Livingston *et al*. 2012; Thompson *et al*. 2012; Gjerberg *et al*. 2011). Yet what is different here is that relationships between relatives and ID care service staff often go back decades. The fact that both the family and the care staff at the ID care service are often deeply emotionally involved over a long period of time is probably unique to ID care. Therefore, old feelings may surface again and may lead to conflicts about the 'ownership' of the person with ID.

*About being overwhelmed, friendship, reflection and caring for carers*

Providing end-of-life care can be quite overwhelming for care staff members, as this thesis highlights. Strong emotional, long-term bonds between care staff and clients make ID care unique across the healthcare sector. These bonds also appear in e.g. attachment studies and social network studies, which demonstrate the significant role that care staff often fulfil in the lives of people with ID (de Schipper *et al*. 2005; van Asselt-Goverts *et al.* 2013). In our studies, some care staff members even described their relationship with their client in terms of 'a special bond', 'love' or 'friendship'. In contrast to nursing home staff, for example, ID care staff had often cared for their client in the active stages of his/her life. We found that it could be particularly hard for ID care staff to let go of a person they previously helped to fully participate in life.

Precisely because of these friendship bonds between care staff and clients, reflection on this caring relationship becomes tremendously important for ID care staff who provide end-of-life care. The value of reflection has become visible in earlier writings. For instance, Nouwen (1997) describes the importance of presence, trust and time for building a friendship in caring for a person with severe/profound ID, and sees reflecting on this caring relationship as an inner process that could ensure good care. Tronto reasons that one needs to be able to postpone one's own concerns in order to be attentive to others: it is a great moral task to "stand back from the ongoing processes of care and ask: what is going on here?" This requires e.g. honesty and a continuous evaluation of the care provided (Tronto, 1993 p. 141).

Yet are ID care staff able to reflect on their caring relationships? This thesis suggests that in order to deal with their emotions, ID care staff need to reflect on their caring relationship with their client, their relationship with relatives, their own emotions and thoughts on end-of-life care and the possible intertwinement of the interests of the client and their own interests. This is a lot to ask from care staff, who are often not highly educated and for whom end-of-life care is not regular care. This is why the support and 'care for carers' (care staff) is so essential. Caring for your own needs is important in order to be alert to the needs of others. The necessity of care for carers has also been stressed in studies within other settings, such as dementia care. In dementia care, carers e.g. encounter moral challenges around truth-telling (Hertogh *et al.* 2004). Furthermore, providing person-centred care on a daily basis is found to be highly demanding for dementia carers, as this asks for a continuous deep involvement and confronts them with the loss of a common shared world of meaning (Hertogh 2004; Hertogh & The 2008). In demanding care situations, such as dementia care and end-of-life care for people with ID, care for carers is indispensable. Yet we hardly encountered any formal implementation of care for carers in our studies, which may mean that this is lacking. Other papers have also seen a gap in care for ID carers and noted that ID care staff may (unjustly) not be seen as 'mourners' (Tuffrey-Wijne & McLaughlin 2015). Care for carers should therefore receive more attention, as it may vastly improve the confidence and qualities of ID care staff in providing end-of-life care.

### Reflection on the challenge of caring together: fostering three caring relationships

End-of-life care for people with ID is a challenge of caring together. Although care for people with ID inevitably builds on relationships, this thesis highlights the fact that the value of good caring relationships seems amplified at the end of life. In encouraging caring together in end-of-life care for people with ID, three caring relationships need to be fostered: the relationship with the person with ID, relationships among professionals and the relationship between relatives and professionals.

In the first place, caring together means involving the person with ID. Building a form of end-of-life care that meets the wishes and preferences of the person with ID means regarding autonomy as a product of joint work, and regarding end-of-life care as shared care. There needs to be trust that the person can be involved in his or her own way and means to achieve this need to be sought proactively, e.g. by focusing on and assessing their capabilities, simplifying the information and maximizing their ability to understand.

Second, caring together also means sharing the care among professionals. ID care staff, physicians and other professionals need to share their expertise and knowledge about the person with each other, as well as with professionals outside

ID care. This implies training ID care staff in end-of-life care topics so that they are more able to provide this care and feel more confident in doing so. It also implies cooperation with services outside ID care, such as specialized end-of-life care services, which may increase the access of people with ID to generic end-of-life care services. It also means providing good 'care for carers'; adequate support for those care staff members who are confronted with the care and loss of their incurably ill client.

Third, relatives and professionals need to join forces to provide care together. If relatives and professionals are able to come together and communicate openly about their views on good end-of-life care, co-caring by both relatives and professionals can lead to warm, loving care in the proximity of the very people who are most important to the person with ID.

## General methodological considerations

This thesis built upon four studies that explored the perspectives of the main stakeholders in end-of-life care: people with ID themselves, relatives and professionals. The thesis combined the insights of the four studies and presented an overall reflection on end-of-life care for people with ID. It has added value to existent studies, which often paid attention exclusively to the staff's perspective. Another advantage was that the four studies were performed in consecutive order. As researchers learn along the way, this enabled us to integrate the insights and lessons from one study and incorporate them into the next study.

The combination of quantitative and qualitative designs is also a strength. By performing mixed-methods research, we were able to gain insights into the perspectives of representative groups of professionals, as well as in-depth insights into the experiences and stories of professionals, relatives and people with ID.

Another strength is that the studies were performed in close cooperation with experts directly involved in ID care. The studies delivered many practical insights, which were validated and refined in consultation with experts in the field. The close cooperation with practitioners also enabled us to develop practical guides and tools.

However, the explorative character of the qualitative studies in particular means that caution should be exercised in generalizing the results, as ID care is a hugely diverse care setting, both with regard to the people with ID themselves and with regard to their caring contexts.

## Methodological considerations per study

### *Retrospective multiple-case study*

Chapter 2 and Chapter 4 were based on a retrospective multiple-case study. The cases of recently deceased people with ID were reconstructed by interviewing 47 relatives and professionals who were closest to the person in question at the end of their life. From this multiperspective focus, we were able to reconstruct the stories of 12 deceased people with ID, incorporating the viewpoints of all the people who were most closely involved. This helped avoid giving too much emphasis to one viewpoint, for instance the viewpoint of the professional.

In the multiple-case study we therefore explored 12 cases. Several measures were taken to enhance the scientific rigour of the research:

1) Participants were selected purposively in order to obtain a relatively diverse sample of client cases with regard to age, living situation (own apartment, group home or residential home), place of death (own home, parents' home, intensive care facility of an ID care service, hospital or hospice), kind of disease and severity of ID (mild, moderate or severe/profound ID). In this way, client cases in a variety of situations could be studied.
2) The principle of data saturation was used: we reached data saturation after studying 12 cases, meaning that we did not identify any new relevant information or themes in the last interviews. Data collection and analysis alternated in a cyclical process in accordance with key principles of qualitative research (Green & Thorogood 2004), until we attained this data saturation point (Ando *et al.* 2014).
3) To ensure reliability and to enrich the data analyses, all interviews were analysed by the principle researcher and at least one co-researcher. The different analyses were then discussed.
4) To validate and enrich the analyses, we discussed the findings in two focus groups with ID caregivers, trainers and policy experts.

However, a limitation is that we studied the client cases retrospectively, so the experiences of the interviewees may have been subject to a recall bias. A second limitation and consequence of this retrospective design is that the experiences of the people with ID themselves could not be studied.

*Group interview study among people with mild ID*

Chapter 3 was based on a group interview study with people with mild ID, using the nominal group technique to structure the discussion. This technique was specifically adapted for people with ID by Tuffrey-Wijne *et al.* (2007b). These group interviews have added value since studies exploring the views of people with ID about end-of-life care are very rare. The nominal group technique adapted for this specific target group appeared to be very useful in eliciting the perspectives of people with mild ID. A clear explanation of the aim of the group interviews, the presence of familiar care professionals, the use of a pictorial story, holding two separate group interview meetings and the structured process of validation and ranking all seemed to contribute to its success. In this study we held interviews with seven groups and a total of 33 people with mild ID who were not receiving end-of-life care. We sought to enhance the scientific rigour and richness of the analyses through:

(1) Purposive sampling of clients from diverse groups (members of the local client council, residents from ID care services and members of a theatre group for people with ID);

(2) Using the principles of data saturation and the cyclic process of data collection and analysis;

(3) Independent analyses and subsequent discussions in pairs consisting of the principle researcher and one of the co-researchers.

The decision to only include people with ID who were not at the end of life themselves was carefully made after considering our responsibility in safeguarding the emotional security of client participants. Yet the views of our respondents may differ from clients who are actually receiving end-of-life care. Another limitation is that we only included people with mild ID, so the views of people with moderate and severe/profound ID remain unknown.

*Quantitative survey study about decision making*

A total of 718 professionals took part in the survey study on decision making about the place of end-of-life care and the use of burdensome medical interventions (Chapters 5 and 6). An important strength is that we were able to recruit a nationwide sample of professionals by using one nationally representative sample of GPs (Van Hassel and Kenens 2013) and one nationally representative sample of nurses and social workers (Albers *et al.* 2014; De Veer *et al*. 2011), and one members' list that covered almost all practicing Dutch ID physicians. We were therefore able to incorporate and compare the perspectives of three different groups of professionals who work with people with ID. A limitation is the possible recall bias, as the considerations in decision making were explored by open questions about the last person with ID for whom the respondent provided end-of-life care. Hence, professionals may be more likely to recall clients who made a great impact on them. Another limitation is that we do not know what the personal input and role of the respondent was in the decision-making process. Different professionals may have different ideas about which considerations are relevant or decisive in decisions. Moreover, the actual considerations of GPs, as well as the perspectives of people with ID and their relatives, were not included in this study.

*Quantitative survey study about expertise and training needs*

Chapter 7 was based on a survey study about the quality of end-of-life care, past training, needs for training, and expert consultation opportunities among 130 nurses and social workers working in ID care services. An important strength of this survey study is the relatively high response rate (71.8%). However, a limitation is that most questions in the study were about perceptions, so we cannot be sure about the actual end-of-life care knowledge and skills of ID care staff.

## Implications for practice and policy

End-of-life care for people with ID is about caring together, and in many ways a real team effort. This can be translated into the following recommendations for practice and policy.

*1. Improve the involvement of people with ID in end-of-life care*

This thesis underscores the complexity of involving people with ID in end-of-life care and the importance of proactively seeking ways to involve the person in end-of-life care on his/her own terms. As highlighted by this thesis, it is hard for many relatives and professionals to uncover and respect their wishes and involve the person with ID in decisions regarding the place of care or the use of medical interventions. In order to involve the person with ID and respect their autonomy, professionals and relatives should aim to adopt an open, active and reflective attitude.

Much can be learned from existing models that were developed in other studies. One example is the ´ASK ME´ collaborative model of supported decision making for involving people with cognitive disabilities; which includes steps such as assessing the person's strengths and deficits, simplifying the task, understanding their values and maximizing their ability to understand (Peisah et al., 2013). The model of Tuffrey-Wijne about breaking bad news to people with ID is also an important tool. Within this model, relatives or professionals break up information about e.g. a medical intervention into small pieces, and these pieces are added one by one to the current knowledge of the person (Tuffrey-Wijne 2012).

To further improve the active involvement of people with ID, resources such as pictorial guides and easy-read books or websites could be used to assist in this process. Resources can be found for example on www.pcpld.org: the Palliative Care for People with Learning Disabilities Network. In the Netherlands, several versions of 'wish books' exist. These are books for recording the wishes of the person with ID concerning the funeral service and the contact details of relatives and other closely involved people. A version of a wish book (in Dutch) is included in a 'signpost' with information on end-of-life care[1]. A practical 'working book' for people with ID was developed on the basis of our group interview study (Chapter 3). This practical book can be used to assess and record the wishes of people with mild ID regarding their end-of-life care (Bekkema *et al.* 2015a).

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[1] http://www.kennispleingehandicaptensector.nl/kennispleindoc/showcases/WEGWIJZERpz-vg.pdf (in Dutch)

*2. Increase the end-of-life care knowledge and skills of ID care staff*

This thesis showed that ID care staff often lack knowledge and expertise regarding end-of-life care, which may lead to a) poor recognition of end-of-life care needs and b) poor treatment. Care staff should know how to appraise the end-of-life care needs of their clients, assess their own skills and assess who (within or outside their organization) is best able to provide this care. ID care services must therefore take adequate measures in order to equip care staff with the necessary expertise to be able to identify and sufficiently meet the end-of-life care needs of their clients. These needs are holistic, as confirmed by e.g. the people with mild ID in our study, so training should focus on symptom management as well as practical, emotional, social and spiritual support. Due attention should also be paid in training to 'soft' skills, as end-of-life care imposes high demands on the relationship qualities of caregivers - e.g. their ability to uncover wishes, listen carefully, empathize and closely observe the behaviour of the person with ID.

Training should focus particularly on the assessment of pain and other symptoms. People with ID, in particular those with severe/profound ID, have an increased risk of pain, related to problems with their physical health and general health (van der Putten & Vlaskamp 2011). This thesis underscores how challenging the identification of pain, anxiety and other signals of distress in people with ID is, and reveals that ID care staff receive almost no training in the use of assessment instruments. An awareness of the complexities of assessing pain and distress in people with ID is highly important, as also indicated by the White Paper on palliative care for people with ID: symptoms in people with ID may be masked, or expressed through behavioural changes (EAPC: Tuffrey-Wijne & McLaughlin 2015). Moreover, professionals should be conscious of the risk of 'diagnostic overshadowing': making diagnoses in which symptoms are incorrectly attributed to the intellectual disability instead of the disease. A systematic review of pain in people with ID shows that behavioural indicators of pain may include motor activity, facial activity, nonverbal vocal expression and socio-emotional indicators (De Knegt *et al.* 2013). Instruments have been developed to assess pain in people with communication difficulties and may contribute to a better management of pain and other symptoms (e.g. the DisDAT: Regnard *et al.* 2007; the Rotterdam Elderly Pain Observation Scale (REPOS): van Herk *et al.* 2009 and STOP-ID!, a self-reporting tool on pain in people with ID: De

Knegt *et al*. 2015). Also, guidelines have been developed in the Netherlands to improve the identification of pain and physical problems in people with ID[2].

Several other actions have been undertaken in the Netherlands to improve end-of-life care for people with ID:

- A number of ID care services have joined the national palliative care improvement programme of ZonMw (The Netherlands Organization for Health Research and Development), implementing end-of-life care interventions within their service. Examples of these interventions are 'Signalling by carers', a training course designed to improve the identification of changes in the condition, daily living activities or behaviour of clients receiving end-of-life care; 'Stem-inspiration cycle', a training course designed to make professionals and managers aware of their own values and norms when providing end-of-life care; and 'End-of-life care consultants', a programme for training nurses from ID care services in advising ID care teams about end-of-life care.
- Guidelines have been developed. They include a general guideline on end-of-life care for people with ID for professionals in this field[3], covering e.g. diagnostics, treatment of symptoms, the farewell process and aftercare. Another guideline is about medical decisions at the end of life for physicians[4], covering legal frameworks, ethical assumption, roles and responsibilities of stakeholders and including an aid for taking decisions at the end of life.
- Training has been developed, for example the blended learning education programme about end-of-life care for people with ID and a training course on palliative sedation for people with ID by the Netherlands Comprehensive Cancer Organization (www.iknl.nl), and training for end-of-life care volunteers in supporting people with ID (www.vptz.nl).
- Practical tools have been developed, such as: a 'signpost', to provide guidance in the care for people with ID[5], covering e.g. communication issues, ethical questions and the coordination of care. Two practical tools for professionals have been developed on the basis of the studies underlying this thesis: 'Caring until the last day' (Bekkema *et al.* 2011), covering stories and

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[2] http://www.venvn.nl/Berichten/ID/90630/Richtlijnen-VVN-voor-signaleren-pijn-en-lichamelijk-ongemak (in Dutch).

[3] http://www.pallialine.nl/verstandelijke-beperking (in Dutch)

[4] http://nvavg.nl/wp-content/uploads/2014/upload/standaarden/medische-beslissingen-rond-het-levenseinde---def.pdf (in Dutch)

[5] http://www.kennispleingehandicaptensector.nl/kennispleindoc/showcases/WEGWIJZERpz-vg.pdf (in Dutch)

advice about end-of-life care for ID professionals, and 'Decision-making about end-of-life care for people with ID' (Bekkema *et al*. 2013a-c; Bekkema *et al*. 2015b), practical tools on how to make decisions about the best place for end-of-life care and the use of medical interventions in end-of-life care for people with ID.

Internationally, some end-of-life care training interventions for ID care staff have been developed (e.g. Reynolds *et al*. 2008; Reddall 2010; Hahn & Cadogan 2011; McLaughlin *et al*. 2012). Hahn and Cadogan (2011) for example developed, implemented and evaluated a palliative care curriculum and educational programme for ID care staff that covers e.g. general end-of-life care principles, values and choices, case-based scenarios, the spiritual and cultural context, the assessment of pain, and distress and symptom management. Reddall (2010) developed a palliative care resource for professional carers of people with learning disabilities, covering e.g. hospice care, cancer and symptom management. Other resources can be found at www.pcpld.org; the Palliative Care for People with Learning Disabilities Network.

*3. Cooperate with end-of-life care experts outside ID care*

Cooperating and sharing expertise with specialized end-of-life care services is vital for improving the quality of end-of-life care for people with ID. End-of-life care is still a rather infrequent type of care in ID care services and it is not possible for ID care staff teams to have all the necessary expertise to meet all end-of-life care needs. ID care staff should, however, know the boundaries of their own end-of-life care expertise and know when the input of other experts is necessary. Experts in end-of-life care can e.g. bring in specialized knowledge on the management of pain and medication, and expertise in advanced nursing treatments. ID care services as well as specialist end-of-life care services should invest in intensified partnerships and make ID care staff aware of the availability of consultation facilities.

Several developments are directed towards better cooperation within end-of-life care:

- In the Netherlands, ID care services can join the 'palliative care networks', a collaboration between healthcare providers in a particular region (www.netwerkpalliatievezorg.nl). These networks aim to optimize the organization of the end-of-life care for people in the region in question.
- A new development in the Netherlands is the creation of seven palliative care consortia to optimize end-of-life care throughout the Netherlands. Consortia are partnerships between expert centres, palliative care networks and the

Netherlands Comprehensive Cancer Organisation. End-of-life care for people with ID may also benefit from these new networks.

- Services may learn from best practices models that can promote partnerships between ID care and specialist end-of-life care services, e.g. from the partnership framework developed by McLaughlin *et al.* (2014).

### *4. Provide care for carers*

If ID services wish to invest in the quality of end-of-life care, they should make care for care staff a priority. The emotional struggles of ID care staff who provide end-of-life care seem to reveal an inner conflict, which is currently given insufficient consideration as we hardly encountered any formal implementation of care for carers in our studies. Sufficient emotional support should be given to ID care staff who provide end-of-life care, with due attention to intrapersonal and interpersonal reflection. Care for carers could for example cover sufficient information for care staff concerning the illness and prognosis of the illness, information on the organization’s policy and procedures regarding end-of-life care, emotional support from colleagues and supervisors, and the intrapersonal search for the meaning of suffering, death and end-of-life care.

Some general guidelines (in Dutch) on end-of-life care (about care in the last days of life and palliative sedation) pay attention to care for carers[6].

Moreover, Buijssen *et al.* compiled a book on care for carers (Buijssen *et al.* 2003). This book features stories by carers about providing end-of-life care and provides insights into how a ‘care for carers’ programme might be set up.

### *5. Foster close cooperation between professionals and relatives*

Good cooperation between care staff and relatives can improve the quality of end-of-life care. This thesis shows that a successful co-caring relationship between relatives and care staff is powerful and a big strength in providing good end-of-life care for people with ID. Yet, in some cases, communication and cooperation fails, e.g. due to the inexperience of care staff, different perceptions on what good end-of-life care entails or friction based on earlier disputes. It is therefore recommended that care staff, as well as ID care services, continuously invest in strong, good co-

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[6] www.oncoline.nl/richtlijn/item/pagina.php?id=32589&richtlijn_id=770

www.oncoline.nl/index.php?pagina=/richtlijn/item/pagina.php&id=28947&richtlijn_id=632

caring relationships with relatives, preferably at a much earlier (and therefore less emotional) stage in the lives of people with ID.

*6. Better prepared ID care services: set up an end-of-life care policy*

As more and more people with ID will need end-of-life care in the future, ID care services should be better prepared and should set up an end-of-life care policy within their organization. In line with the findings of this study, ID care services should acknowledge the following aspects in developing this policy:

- ID care services should recognize that end-of-life care is an indispensable part of the ID care provision within the organization. This thesis shows that end-of-life care is not a core activity within ID care. Hence, only when managers make end-of-life care a priority and take responsibility for providing this care will it be possible to achieve an improvement in the quality of end-of-life care. If the value of end-of-life care is openly advocated within an ID care service, end-of-life care policies can be co-created that inspire employees to take their care to the next level.
- ID care services should endorse the vision that end-of-life care is about caring together. In line with the abovementioned recommendations an end-of-life care policy should at least cover ways to involve the person with ID, roles and skills of the different professions within the service, ways to increase the expertise of care staff, cooperation with specialized end-of-life care services, co-caring with relatives and care-for carers.
- Finally, ID care services should make it easier for relatives and care staff to actually be there for the person. End-of-life care ultimately comes down to satisfactorily meeting the end-of-life care needs of the person with ID. Assessing if the right needs are being met is complex in people with ID, as this thesis stresses. Hence, services should prioritize and facilitate having care staff and relatives actually be there for the person with ID, so they can closely observe their needs and find out if their care has been effective.

## Implications for further research

As end-of-life care for people with ID is a 'young' research topic, many topics deserve further exploration. The following recommendations are made for future research.

*1. Explore barriers in equal access to end-of -life care provided by general care providers*

People with ID have the right to enjoy the highest attainable standard of health, and the right to use the same community services and facilities as are available for the general population (UN 2006). People with ID therefore have the right to equal access to end-of-life care provided by general care providers outside ID care, such as hospice care and home care teams. It is as yet unknown how accessible end-of-life care by general care providers is for people with ID in the Netherlands, and what experience these providers have in caring for people with ID. International studies do indicate that people with ID are underrepresented in end-of-life care by general care providers (Tuffrey-Wijne & McLaughlin 2015; Heslop *et al.* 2013). Future studies should therefore provide insight into the accessibility of end-of-life care by general care providers for people with ID, and into ways in which these providers can deliver proper care for this special group. Moreover, studies should explore how ID care providers and general care providers can cooperate sufficiently to improve the quality of end-of-life care for people with ID, irrespective of the setting in which the person resides.

*2. Explore the strengths and challenges of the triadic relationship between the client, relatives and care staff*

Although the value of relationship-based care is well documented in ID care, the strengths and challenges of the triadic client-relatives-care staff relationship, in particular in times when emotions run high, needs further exploration. This thesis indicates how complex and sometimes family-like care relationships between relatives and care staff can be, and how these relationships may positively or negatively influence the quality of end-of-life care. Further investigations are therefore recommended into the underlying values of people with ID, relatives and care staff regarding end-of-life care. A useful method might be to offer participants of all groups the same vignettes representing a hypothetical end-of-life care situation or dilemma, and subsequently ask them questions about their perspectives and values. Vignettes have been used previously in research on sensitive subjects, e.g. on violence amongst children (Barter & Renold 2000).

*3. Study support and aftercare for relatives of people with ID*

Care for relatives is a relevant part of end-of-life care (WHO 2015, Tuffrey-Wijne & McLauglin 2015). From our qualitative multiple-case study we know for example that the family can feel overwhelmed when confronted with decisions at the end of life. They often feel that the person’s dependency is magnified, which makes them feel highly responsible in their role of ‘proxy’. We also found that it could be challenging for relatives to cooperate and co-care with care staff. Moreover, having a relative with ID can generally have a great impact on your family life (Tuffrey-Wijne & McLauglin 2015). Yet, professional support and aftercare for relatives of clients with ID was not the focus of our study, nor has it been of other studies of end-of-life care for people with ID. Further research could therefore provide insight into 1) relatives’ own support needs, and 2) how proper support and aftercare for relatives could be developed and implemented within end-of-life care for people with ID.

*4. Incorporate the perspectives of co-residents in end-of-life care research*

If people with ID live in a group home or residential setting, they have relationships with co-residents with ID. When someone in the group becomes incurably ill, co-residents have to deal with this, which may not be easy. Co-residents have their own feelings of loss and grief, which has been the subject of research by Todd (2010) and Read & Elliott (2007). Co-residents may in some cases also fulfil a valuable supportive role in end-of-life care. According to the people with ID who we interviewed, co-residents could play an important role in e.g. providing psychosocial support for the ill person. Yet, although co-residents might have a close relationship with the ill person, our studies have not incorporated their views. It is therefore recommended that future studies focus on the perspectives of these important stakeholders and study what they feel is important in end-of- life care and how they could be involved.

*5. Develop models on how to involve people with ID in decision making*

The wishes of people with ID are often not explicitly taken into account in decisions about the place of care and the use of burdensome medical interventions. Many issues concerning decision making arise from our studies, such as the following. Can standard informed-consent procedures adequately incorporate the preferences of people with ID? What factors influence decision making in end-of-life care for people with ID? How can the interests of the person with ID be distinguished from the interests of relatives and care staff when they are so deeply involved? Are existing models for decision making, such as *shared decision making* (Bélanger *et al.* 2011) and *supported decision making for involving people with cognitive disabilities* (Peisah *et al.* 2013), suitable for (all) people with ID? Future studies should further investigate how people with ID could be involved in decision making in end-of-life care, including the exploration of which types of decision making models and frameworks are appropriate given the diversity of people with ID.

*6. Continue research on identifying and interpreting pain and other symptoms in people with ID*

Managing pain and other physical, psychosocial and spiritual problems and symptoms is of central importance in end-of-life care (WHO 2015). Pain can have a severe impact on the wellbeing and quality of life of people with ID (Walsh *et al*. 2011). Important research is being done (e.g. De Knegt *et al*. 2013 & 2015, van der Putten & Vlaskamp 2011). As highlighted by this thesis and many other studies, identifying and interpreting symptoms in people with ID is often difficult. It is therefore of vital importance that research is continued to better identify, interpret and relieve symptoms.

*7. Conduct more research on how to uncover the wishes and needs of people with severe/profound ID*

The complexity of uncovering the wishes and needs of people with ID in end-of-life care was a recurring finding in our studies, in particular regarding the care wishes of people with severe/profound ID. It appears to be hard to find out what they value in good end-of-life care, for example what makes them comfortable, what could help make them feel better, whether they are in pain or distress, and how they feel about transfers and the use of medical interventions. Hence, there is a need for insights into how wishes and needs could be uncovered. Future research could focus for example on the development of instruments or communication strategies to identify the wishes of people with severe/profound ID as well as their responses to end-of-life care.

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# Summary

Providing good care for people with an intellectual disability (ID) at the end of life does not happen as a matter of course, for example because the intellectual disability makes it difficult for people to clearly express their wishes and care needs. Various people are involved in providing this care and they all have their own outlook on it. The aim of this doctoral research is to obtain a better picture of end-of-life care for people with ID, from the viewpoints not only of people with ID but also of relatives, care staff and physicians.

## Introduction

**Chapter 1** introduces the theme of the research. Intellectual disabilities are characterized by significant limitations in both intellectual functioning and adaptive behaviour that have arisen before the age of eighteen. People receive care and support above all from ID care services, mostly from care staff trained in social work, as well as nurses and nursing assistants. Medical care is generally provided by GPs and physicians specializing in the intellectually disabled (ID physicians). People with ID who are ageing are a vulnerable group: they often suffer from chronic diseases and multi-morbidity is more common than in the general population. Because the population group with intellectual disabilities is ageing and they often suffer from chronic, progressive diseases over lengthier timeframes, increasing numbers of people with ID are receiving end-of-life care.

Care at the end of life, also known as palliative care, is care that focuses on the quality of life for people with an incurable and ultimately fatal condition. It is known that care at the end of life for people with ID can be a challenge, for example because people with ID often have difficulty getting their wishes and needs across. The net result of this can be that they are not involved in the decisions about the final stage of life. In addition, there is a lack of knowledge among care staff in the ID care sector about somatic conditions and end-of-life care; that a client is suffering from an incurable condition is often only diagnosed at a relatively late stage. Often there is also relatively little cooperation between professionals in the ID care sector and professionals with specific expertise in end-of-life care.

The involvement of many different people means that attention needs to be paid to the relational aspects of this care. The challenges and bottlenecks mentioned earlier can also result in ethical questions coming to the fore, such as what is actually good care for someone with an intellectual disability at the end of their life, and who can best take the decisions about such care. Care ethics, which is based on a view of human relationships and assumes the fundamental vulnerability and dependence of people, can help think through these ethical questions and relational aspects. Key ethical values such as attentiveness, responsibility and competence can help interpret research findings.

## Part 1: Care, needs and wishes of people with ID at the end of life

**Chapter 2** presents the results of a retrospective case study. Forty-five interviews with relatives and professionals in twelve cases of people with ID who had recently died provided the basis for the analysis. This chapter shows five important shifts at the end of life for people with ID:

1) Care staff and relatives develop a new care strategy focused on offering comfort, tackling the symptoms and taking over tasks from the ill person. Many care staff members and relatives struggle to let go of the care strategy that they were used to, which was one of encouraging participation and independence.
2) In the final phase of life, finding a balance between keeping a professional distance and providing warm and tender care increasingly becomes a struggle for care staff (who are often closely involved emotionally).
3) Mutual cooperation among professionals, and between professionals on the one hand and relatives on the other, becomes increasingly important for recognizing and interpreting signs of pain or other symptoms.
4) Relatives experience more responsibility for taking medical decisions, which is related to the ill person's increasing degree of dependence.
5) Awareness grows at the end of life that the ill person often has two 'families' who are looking after them: their biological family and the care staff.

Within these shifts, six values in the way relatives and care staff act appear to be important: 'being there' for the ill person, being responsive to their needs, being attentive to personal wishes and needs, reflecting on their own emotions and care relationships, being responsible for taking joint decisions in the interests of the ill person, and being open for cooperation, and sharing the care with others.

**Chapter 3** presents the results of qualitative group interviews held with people with mild ID. These were aimed at studying what people with ID themselves find important within the care relationships for end-of-life care. Seven groups of people with mild ID (33 people in total) who were not receiving end-of-life care and were not incurably ill participated twice each in group interviews that were based on a nominal group technique adapted for the target group and the objectives of this study.

People with mild ID find two elements important within the care relationships at the end of life: dealing with the ill person's wishes, and ‘being there’. Dealing with people's wishes covers not only finding out and recording what they want (such as wishes about the care, final wishes for things that the ill person would still like to do, and what they want for the funeral) but also respecting those wishes. People with ID are sometimes afraid that their wishes will not be respected, something that seems to be based on their previous experiences in the care sector. ‘Being there’ is about offering practical support (for example with getting dressed, eating and drinking, as well as getting medical help from a physician), as well as emotional support (comfort, empathy, listening), social support (such as offering distractions) and spiritual support (such as talking about the impending death, their beliefs, or life). People with mild ID believe it is important to be able to have positive experiences and live comfortably to the very end.

The modified nominal group technique seems suitable for finding out what ideas people with ID have about end-of-life care. It is important here to give the participants a thorough explanation of the purpose of the study, to make sure that a trusted person is present during interviews, to use (supporting) image material and to hold two successive meetings so that the sessions do not last too long and the participants have time to prepare themselves for the second meeting. It is also important to make sure there is support outside the sessions, should any of the participants feel a need for it.

**Chapter 4** is also based on the retrospective case study, describing how care staff and relatives actually respect the autonomy in the care at the end of life. Respecting the autonomy of people with ID comes to the fore in particular during transitions, for instance when helping the ill person understand new information about their condition, finding out about any changes to the needs and wishes, and in important decisions. Professionals and relatives however often find it difficult to work out just how much information the ill person wants about the disease, particularly in people with severe ID. Care staff and relatives often do not really know how they should communicate about severe illness and death, and social workers in particular do not know enough about care at the end of life. Finding out what people want is complex, particularly because wishes can change at the end of life and because the wishes of people with severe ID are often ‘hidden’. In addition, the wishes of the person who is ill can sometimes conflict with the wishes of others. On top of that, the increasing dependency of the ill person is difficult, in particular for the relatives of people with severe ID, especially when they have to take important decisions.

A variety of skills of the care staff and relatives turn out to be important for respecting autonomy: paying attention to the ill person's need for information, connecting, recognizing care needs, creating scope for the wishes and preferences to be expressed, and being able to discuss dilemmas.

Respecting the autonomy of people with ID at the end of their lives is, in terms of care ethics, a ‘relational construct’ that can be achieved together with the ill person.

## Part 2: Decision making at the end of life of people with ID

**Chapters 5 and 6** are about a survey in the form of a questionnaire for care staff about decisions involving the place of care and potentially burdensome medical interventions, such as further diagnostic investigations, chemotherapy, an operation to extend the lifespan or pain control using an infusion pump. The questionnaire was sent to 294 ID care staff (67% response), 273 ID physicians (53% response) and 1000 GPs (38% response).

The professionals surveyed believe that everything possible should be done to ensure that the client can remain in their own home until the very end (Chapter 5). They also believe it is important that 24-hour care should be available. When deciding not to move the client, the familiarity with their own residential environment is an important consideration, as is the expertise of the team in providing care at the end of life. Considerations in favour of moving a client are largely related to the lack of expertise in their own team and the client's home not being suitably laid out. Although many professionals believe that the client's wishes about where they want to live should always be paramount, the client's wishes were not often listed as one of the considerations. This discrepancy suggests that more attention should be paid to involving people with ID in such decisions.

Furthermore, professionals listed the quality of life and the wellbeing of the ill person the most often as considerations in decisions about whether or not to start or continue a potentially burdensome medical intervention (Chapter 6).

Professionals have a range of opinions about whether people with ID should or should not always be informed about a medical intervention. Opinions also differ as to whether a person's rejection of a medical intervention should always be respected. The wishes of relatives are explicitly listed more often as a consideration than the wishes of the ill person themselves. The best way to involve people with ID in medical decision-making needs further investigation.

## Part 3: Expertise and training needs of ID care staff

**Chapter 7** presents the results of a survey study among care staff in the ID care sector. It examines what care staff think of the quality of end-of-life care within their team or department, what training they have had about care at the end of life, and what additional training they would like to have for it. The questionnaire was sent to 181 care staff members (72% response).

The majority of care staff members are positive about the quality of the end-of-life care within their team or department. Nevertheless, 85% of them do not feel they are sufficiently well trained to provide good end-of-life care. Instruments for helping to measure pain, for instance, are rarely discussed in their training. Care staff would like additional training, particularly for assisting the client in dealing with the impending death and saying their farewells. In comparison with social workers, those with nursing backgrounds have had more training about care at the end of life and have less of a need for additional training. The questionnaire survey also shows that care staff are often not aware of the options for consultations about end-of-life care.

## General discussion

**Chapter 8**, the general discussion, looks at the quality of care at the end of life. It points out how important it is that trusted people should be present for care at the end of life; people who know the ill person well and are well able to interpret their signals. ‘Being there’ requires good relationship skills that are related to a vision of care that is based on care ethics. The chapter also examines the fact that care staff are often strongly willing to take on the care at the end of life, as well as looking at the lack of knowledge and skills that they have in the palliative care domain. A lack of knowledge can for example result in signals pointing to pain or other symptoms not being (timely) recognized. The quality of care at the end of life can be improved inter alia through additional training for supervisors and better cooperation between ID care services and organizations that specialize in end-of-life care.

It is essential to find out the wishes of people with ID and increase their involvement in decisions about care at the end of life. Trust is a key factor in this: both confidence in the opportunities for the ill person to be involved and confidence in the skills of relatives and care staff for involving them. This trust is not always present, meaning that people with ID may feel they are not being understood and heard. This underlines the importance of a relational approach to respect autonomy.

Good cooperation is also essential for the recognition and alleviation of symptoms. The bonds between relatives and the ID case service – links that often go back years or even decades – make the care relationship between relatives and the care staff unique within the healthcare sector, as well as making it (potentially) very valuable for offering care jointly at the end of life. Nevertheless, the various challenges in the relationship between relatives and the care staff (such as conflicts about who decides or helps decide about what and the higher level of expectation that relatives have regarding the care staff members' knowledge) require greater openness in the communications and more support from the organization. This special bond that the care staff often have with their clients is discussed, along with what this means for end-of-life care. ‘Caring for carers’ is important, with plenty of scope for reflection among the care staff on their care relationships and their own emotions and values.

‘The challenge of caring together’ (the title of this thesis) is about the special attention that three care relationships require. Firstly, the relationship with the person with ID: involving the ill person in the end-of-life care and the decisions about it. Secondly, the relationships between professionals themselves, because good cooperation is extremely important in ensuring appropriate responses to symptoms (among other things). Thirdly, there is the relationship between professionals and relatives, because good relations between the parties involved are important for good care at the end of life.

Going on from what was described earlier, this concluding chapter contains practical recommendations for involving people with ID in their care and decisions about it, encouraging expertise and ‘care for carers’, cooperation between care staff in the ID care sector and professionals with specific expertise in end-of-life care, cooperation between relatives and professionals, and developing targeted policy for care at the end of life within the ID care sector.

This concluding chapter also looks at the methodological considerations. One limitation of this doctoral research is for instance that the opinions of ill people with ID who are already receiving end-of-life care were not studied. The viewpoints of co-residents of clients receiving end-of-life care were also not examined. However, the viewpoints of various other relevant parties were included: people with ID who are not yet ill, relatives of deceased people with ID, and various groups of professionals. This has made it possible to paint a rich and subtle picture of end-of-life care for people with ID.

Another strength is the combination of qualitative and quantitative methods, which allowed us both to obtain information from representative groups of professionals and to obtain insights into the personal stories of people with ID, their relatives and professionals.

A recommendation for further study is to obtain greater insights into (possible) limitations in the accessibility of care at the end of life for people with ID outside the ID care sector. Follow-up studies are also needed to obtain more insights into how to encourage good relationships between the ill person, relatives and care staff at the end of life. Research would also be desirable to obtain insights into the viewpoints of co-residents on end-of-life care. Other topics for follow-up research are caring for the relatives of people with ID, strategies for involving people with ID more in decisions about end-of-life care, and the possibilities for recognizing pain and other symptoms of people with severe ID at the end of life, as well as determining what their care wishes are.

# Samenvatting

Goede zorg aan het levenseinde van mensen met een verstandelijke beperking is niet vanzelfsprekend, bijvoorbeeld omdat de verstandelijke beperking mensen belemmert in het duidelijk overbrengen van hun wensen en zorgbehoeften. Verschillende mensen zijn betrokken bij deze zorg en zij hebben allemaal hun eigen perspectief. Het doel van dit promotieonderzoek is om meer inzicht te geven in de zorg aan het levenseinde van mensen met een verstandelijke beperking, vanuit de perspectieven van zowel mensen met een verstandelijke beperking als familie, begeleiders en artsen.

## Inleiding

In **Hoofdstuk 1** wordt het onderzoeksthema geïntroduceerd. Een verstandelijke beperking kenmerkt zich door significante beperkingen in zowel het intellectueel functioneren als het adaptief gedrag, ontstaan vóór de leeftijd van achttien jaar. Mensen ontvangen vooral zorg en ondersteuning vanuit organisaties voor verstandelijk gehandicaptenzorg, meestal van begeleiders met een agogische opleiding, maar ook van verpleegkundigen en verzorgenden. Medische zorg wordt voornamelijk gegeven door huisartsen en AVG's (artsen voor verstandelijk gehandicapten). Ouder wordende mensen met een verstandelijke beperking zijn een kwetsbare groep: zij lijden vaak aan chronische ziekten en hebben vaker dan mensen in de algemene bevolking te maken met multimorbiditeit. Doordat ook de groep van mensen met een verstandelijke beperking vergrijst en zij vaak over een langere periode lijden aan chronische, progressieve ziekten, hebben meer mensen met een verstandelijke beperking te maken met zorg aan het levenseinde. Zorg aan het levenseinde, ook wel aangeduid als 'palliatieve zorg', is de zorg die zich richt op de kwaliteit van leven van mensen met een ongeneeslijke en uiteindelijk fatale aandoening. Bekend is dat zorg aan het levenseinde van mensen met een verstandelijke beperking een uitdaging kan zijn, bijvoorbeeld doordat deze mensen hun wensen en behoeften vaak moeilijk kunnen uiten. Dit kan er toe leiden dat zij niet betrokken worden bij beslissingen in de laatste levensfase. Daarnaast is er een gebrek aan kennis onder begeleiders in de verstandelijk gehandicaptenzorg over somatische aandoeningen en zorg aan het levenseinde en wordt vaak laat vastgesteld dat een cliënt aan een ongeneeslijke ziekte lijdt. Ook vindt er vaak weinig samenwerking plaats tussen zorgverleners in de verstandelijk gehandicaptenzorg en zorgverleners met specifieke expertise over levenseindezorg.

De betrokkenheid van velen laat zien dat er aandacht nodig is voor de relationele aspecten in deze zorg. Daarbij kunnen voornoemde uitdagingen en knelpunten leiden tot ethische vragen, zoals: wat is nu goede zorg voor iemand met een verstandelijke beperking aan het eind van het leven en wie kan het beste beslissen over deze zorg? De zorgethiek, gebaseerd op een relationele mensvisie en uitgaande van de fundamentele kwetsbaarheid en afhankelijkheid van mensen, kan helpen om deze ethische vragen en relationele aspecten te doordenken. Belangrijke zorgethische waarden als aandachtigheid, verantwoordelijkheid en competentie, kunnen helpen om onderzoeksbevindingen te interpreteren.

## Deel 1: de zorg, behoeften en wensen van mensen met een verstandelijke beperking aan het eind van het leven

**Hoofdstuk 2** presenteert de resultaten van een retrospectief casusonderzoek. Vijfenveertig interviews met familie en zorgverleners rondom twaalf casussen van recent overleden mensen met een verstandelijke beperking vormden de basis voor de analyse. Dit hoofdstuk laat vijf belangrijke verschuivingen aan het levenseinde van mensen met een verstandelijke beperking zien:

1) Begeleiders en familie ontwikkelen een nieuwe zorgstrategie gericht op het bieden van comfort, symptoombestrijding en het overnemen van taken van de zieke. Veel begeleiders en familieleden worstelen met het loslaten van de zorgstrategie die zij gewend waren, namelijk het stimuleren van participatie en zelfstandigheid.

2) In de laatste levensfase wordt het vinden van een balans tussen professionele afstand en liefdevolle zorg steeds vaker een worsteling voor begeleiders (die vaak sterk emotioneel betrokken zijn).

3) De samenwerking tussen zorgverleners onderling en met familie om signalen van pijn of andere symptomen te herkennen en te kunnen interpreteren wordt steeds belangrijker.

4) Familie ervaart meer verantwoordelijkheid voor het nemen van medische beslissingen, wat gerelateerd lijkt aan de toenemende afhankelijkheid van de zieke.

5) Aan het levenseinde groeit het bewustzijn dat de zieke vaak twee ‘families’ heeft die voor hem/haar zorgen: de eigen familie en de ‘familie’ van begeleiders.

Zes waarden in het handelen van familie en zorgverleners blijken belangrijk binnen deze verschuivingen: ‘er zijn’ voor de zieke, responsief zijn voor zijn/haar behoeften, aandacht voor persoonlijke wensen en behoeften, reflectie op de eigen emoties en zorgrelaties, verantwoordelijkheid voor het nemen van gezamenlijke beslissingen in het belang van de zieke en de openheid om samen te werken en de zorg te delen met anderen.

**Hoofdstuk 3** gaat over kwalitatieve groepsinterviews onder mensen met een verstandelijke beperking. Onderzocht is wat mensen met een verstandelijke beperking zelf belangrijk vinden binnen zorgrelaties in de zorg aan het levenseinde. Zeven groepen mensen met een lichte verstandelijke beperking (33 mensen in totaal) die geen levenseindezorg kregen en niet ongeneeslijk ziek waren, namen twee maal deel aan de groepsinterviews, aan de hand van een nominale groepstechniek die aangepast was aan de doelgroep en het doel van deze studie. Mensen met een lichte verstandelijke beperking vinden twee elementen belangrijk binnen zorgrelaties aan het levenseinde: het omgaan met de wensen van de zieke en “er zijn”. Omgaan met wensen betreft zowel het achterhalen en vastleggen van wensen (zoals wensen over de zorg, laatste wensen over wat de zieke nog graag wil doen en wensen over de uitvaart), als ook het respecteren van deze wensen. Mensen met een verstandelijke beperking zijn soms bang dat hun wensen niet gerespecteerd worden, wat gebaseerd lijkt op eerdere ervaringen in de zorg. “Er zijn” gaat over het bieden van praktische steun (zoals bij het aankleden, eten en drinken, en medische hulp krijgen van een dokter), maar ook over emotionele steun (troosten, inleven, luisteren), sociale steun (zoals afleiding bieden) en spirituele steun (zoals praten over de naderende dood, het geloof of het leven). Mensen met een lichte verstandelijke beperking vinden het belangrijk om positieve ervaringen op te kunnen doen en comfortabel te kunnen leven tot het einde.
De aangepaste nominale groepstechniek blijkt geschikt voor het achterhalen van ideeën van mensen met een verstandelijke beperking over de zorg aan het levenseinde. Hierbij is het belangrijk om goede uitleg te geven aan de deelnemers over het onderzoek, ervoor te zorgen dat er een vertrouwd persoon aanwezig is bij de interviews, gebruik te maken van beeldend (ondersteunend) materiaal en twee opeenvolgende bijeenkomsten te houden zodat sessies niet te lang duren en deelnemers de tijd hebben zich voor te bereiden op de tweede bijeenkomst. Belangrijk is ook om te zorgen voor ondersteuning buiten de sessies mocht een deelnemer daar behoefte aan hebben.

**Hoofdstuk 4** is ook gebaseerd op het retrospectieve casusonderzoek en beschrijft hoe zorgverleners en familie respect voor autonomie in de zorg aan het levenseinde vormgeven. Respecteren van autonomie van iemand met een verstandelijke beperking komt vooral naar voren tijdens veranderingen, bijvoorbeeld bij het helpen van de zieke met het begrijpen van nieuwe informatie over de ziekte, het achterhalen van veranderende behoeften en wensen en bij belangrijke beslissingen. Zorgverleners en familieleden vinden het echter vaak moeilijk om te achterhalen aan hoeveel informatie over de ziekte de persoon behoefte heeft, zeker bij mensen met een ernstige verstandelijke beperking. Ook weten zorgverleners en familieleden vaak niet goed hoe ze moeten communiceren over ernstige ziekte en doodgaan en ervaren met name agogisch opgeleide begeleiders een tekort aan kennis over de zorg aan het levenseinde. Wensen achterhalen is ook lastig, vooral omdat wensen kunnen veranderen aan het einde van het leven en omdat wensen van mensen met ernstige verstandelijke beperkingen vaak 'verborgen' zijn. Daarbij kunnen de wensen van de zieke soms conflicteren met wensen van anderen. Daarnaast is de toenemende afhankelijkheid van de zieke met name voor familie van mensen met ernstige verstandelijke beperkingen lastig, vooral wanneer zij belangrijke beslissingen moeten nemen.

Verschillende ‘kwaliteiten’ van zorgverleners en familie blijken belangrijk om autonomie te respecteren: aandacht voor de informatiebehoefte van de zieke, in contact kunnen komen, het herkennen van zorgbehoeften, ruimte bieden om wensen en voorkeuren aan te geven en het kunnen bespreken van dilemma’s.

Respecteren van autonomie van mensen met een verstandelijke beperking aan het eind van het leven blijkt, in termen van de zorgethiek, een ‘relationeel construct’ dat in samenwerking met de zieke tot stand kan komen.

## Deel 2: besluitvorming aan het einde van het leven van mensen met een verstandelijke beperking

**Hoofdstuk 5 en hoofdstuk 6** gaan over een vragenlijstonderzoek onder zorgverleners naar beslissingen over de woonplek en potentieel belastende medische interventies, zoals extra onderzoek, chemotherapie, een levensverlengende operatie of pijnbestrijding met een infuuspomp. De vragenlijst is verstuurd aan 294 begeleiders (respons 67%), 273 artsen voor mensen met een verstandelijke beperking (respons 53%) en 1000 huisartsen (respons 38%).

De ondervraagde zorgverleners vinden dat er zoveel mogelijk in het werk gesteld moet worden om ervoor te zorgen dat de cliënt tot aan het eind van zijn leven in zijn eigen woning kan blijven (hoofdstuk 5). Ook vinden ze het belangrijk dat er 24-

uurszorg beschikbaar is. Bij het besluit om de cliënt *niet* te verhuizen is de vertrouwdheid van de eigen woonomgeving een belangrijke overweging, net zoals de expertise van het team in zorg aan het levenseinde. Overwegingen om een cliënt *wel* te verhuizen betreffen voornamelijk een gebrek aan expertise in het eigen team en een gebrek aan de juiste inrichting van de woning. Hoewel veel zorgverleners vinden dat de wens van de cliënt over waar hij/zij wil wonen altijd leidend moet zijn, worden de wensen van de cliënt weinig als overweging genoemd. Deze discrepantie vraagt om meer aandacht voor het betrekken van mensen met een verstandelijke beperking bij dergelijke besluiten.

Verder noemen zorgverleners de kwaliteit van leven en het welzijn van de zieke het vaakst als overwegingen bij besluiten over al dan niet starten of doorgaan met een potentieel belastende medische interventie (hoofdstuk 6). Zorgverleners hebben wisselende opvattingen over of mensen met een verstandelijke beperking wel of niet altijd geïnformeerd moeten worden over een medische interventie. Ook verschillen de meningen over of het altijd gerespecteerd moet worden als een persoon een medische interventie weigert. De wensen van familieleden worden vaker expliciet genoemd als overweging dan de wensen van de zieke zelf. Hoe mensen met een verstandelijke beperking het beste bij medische besluitvorming betrokken kunnen worden vergt nader onderzoek.

## Deel 3: opleiding en bijscholingswensen van begeleiders in de verstandelijk gehandicaptenzorg

**Hoofdstuk 7** presenteert de resultaten van een vragenlijstonderzoek onder begeleiders in de verstandelijk gehandicaptenzorg. Onderzocht is wat de begeleiders vinden van de kwaliteit van de zorg aan het levenseinde binnen hun team of afdeling, welke opleidingen zij hebben gehad over zorg aan het levenseinde en wat hun bijscholingswensen hierover zijn. De vragenlijst is verstuurd aan 181 begeleiders (respons 72%).

Begeleiders zijn veelal positief over de kwaliteit van de zorg aan het levenseinde in hun team of afdeling. Toch voelt 85% zich onvoldoende opgeleid om goede zorg aan het levenseinde te bieden. Bijvoorbeeld instrumenten om pijn te kunnen signaleren zijn in hun opleiding niet besproken. Begeleiders willen graag bijgeschoold worden, vooral in ondersteuning aan de cliënt bij het omgaan met het naderende overlijden en afscheid nemen. In vergelijking met begeleiders met een agogische opleiding hebben degenen met een verpleegkundige achtergrond meer opleiding gehad in zorg aan het levenseinde en hebben zij minder bijscholingswensen. Ook blijkt uit dit vragenlijstonderzoek dat begeleiders zich vaak niet bewust zijn van consultatiemogelijkheden over zorg aan het levenseinde.

## Discussie

**Hoofdstuk 8**, de algemene discussie, gaat in op de kwaliteit van de zorg aan het levenseinde. Daarbij wordt aangeduid hoe belangrijk het is dat er in de zorg aan het levenseinde vertrouwde mensen zijn, die de zieke goed kennen en zijn/haar signalen goed kunnen interpreteren. “Er zijn” vraagt om goede relationele vaardigheden, vaardigheden die gerelateerd zijn aan een zorgethische visie op zorg. Ook gaat het hoofdstuk in op de sterke bereidheid onder begeleiders om de zorg aan het levenseinde op zich te nemen, als ook het tekort aan kennis en vaardigheden dat zij hebben op het terrein van zorg aan het levenseinde. Een tekort aan kennis kan er bijvoorbeeld toe leiden dat signalen die duiden op pijn of andere klachten niet (tijdig) worden herkend. De kwaliteit van zorg aan het levenseinde kan vergroot worden door onder meer bijscholing van begeleiders en een betere samenwerking tussen de verstandelijk gehandicaptenzorg en organisaties die gespecialiseerd zijn in zorg aan het levenseinde.

Het is essentieel om de wensen van mensen met een verstandelijke beperking te achterhalen en hun betrokkenheid bij besluiten over de zorg aan het levenseinde te vergroten. Vertrouwen is daarbij een centrale waarde: zowel het vertrouwen in de mogelijkheden van de zieke om betrokken te kunnen zijn als in de vaardigheden van familie en zorgverleners om de zieke te betrekken. Dit vertrouwen ontbreekt soms, waardoor mensen met een verstandelijke beperking zich onbegrepen en niet 'gehoord' kunnen voelen. Dit onderstreept het belang van een relationele benadering van respect voor autonomie.

Goede samenwerking is ook essentieel voor adequate herkenning en verlichting van symptomen. De banden tussen familie en de zorgorganisatie die vaak (tientallen) jaren teruggaan, maken de zorgrelatie tussen familie en begeleiders uniek binnen de zorg en ook van grote (potentiële) waarde voor het bieden van gezamenlijke zorg aan het levenseinde. Toch vragen de verschillende uitdagingen in de relatie tussen familie en zorgverleners, zoals conflicten over wie waar over (mee) beslist en de hogere verwachting die familie heeft van het niveau van kennis onder begeleiders, om meer openheid in communicatie en meer ondersteuning vanuit de organisatie. De speciale band die begeleiders vaak hebben met hun cliënt wordt besproken evenals wat dit betekent voor de zorg aan het levenseinde. “Zorg voor zorgenden” is belangrijk, met veel aandacht voor reflectie onder begeleiders op hun zorgrelaties en eigen emoties en belangen.

“De uitdaging van samen zorgen” (de titel van het proefschrift) gaat over de speciale aandacht die drie zorgrelaties nodig hebben. Ten eerste de relatie met de persoon met verstandelijke beperkingen zelf: het betrekken van de zieke bij (besluiten over) de zorg aan het levenseinde. Ten tweede de relatie tussen zorgverleners onderling, omdat goede samenwerking erg belangrijk is voor, onder meer, adequate symptoombestrijding. Ten derde de relatie tussen zorgverleners en familie, omdat een goede verhouding tussen deze betrokkenen van belangrijke waarde is voor goede levenseindezorg.

In aansluiting bij wat hiervoor is beschreven, bevat dit afsluitende hoofdstuk praktische aanbevelingen voor het betrekken van mensen met een verstandelijke beperking bij (besluiten over) de zorg, deskundigheidsbevordering en 'zorg' voor begeleiders, samenwerking tussen zorgverleners uit de verstandelijk gehandicaptenzorg en zorgverleners met specifieke expertise over zorg aan het levenseinde, samenwerking tussen familie en zorgverleners en het ontwikkelen van gericht beleid over zorg aan het levenseinde binnen de verstandelijk gehandicaptenzorg.

In dit afsluitende hoofdstuk komen ook methodologische overwegingen aan bod. Een beperking van dit promotieonderzoek is bijvoorbeeld dat de meningen van zieke mensen met een verstandelijke beperking die zelf al levenseindezorg krijgen niet onderzocht zijn. Ook zijn de perspectieven van medebewoners van cliënten die levenseindezorg krijgen niet onderzocht. Dat neemt niet weg dat perspectieven van verschillende andere betrokkenen wel zijn meegenomen: mensen met een verstandelijke beperking die nog niet ziek zijn, familie van overleden mensen met een verstandelijke beperking en verschillende groepen professionals. Hierdoor kon een rijk en genuanceerd beeld van de zorg aan het levenseinde van mensen met een verstandelijke beperking geschetst worden.

Een andere sterke kant is de combinatie van kwalitatieve en kwantitatieve methoden, waardoor we zowel informatie kregen van representatieve groepen zorgverleners als inzicht verwierven in de persoonlijke verhalen van mensen met een verstandelijke beperking, familieleden en zorgverleners.

Een aanbeveling voor vervolgonderzoek is de behoefte aan meer inzicht in (mogelijke) belemmeringen in de toegankelijkheid van zorg aan het levenseinde voor mensen met een verstandelijke beperking buiten de verstandelijk gehandicaptenzorg. Ook is vervolgonderzoek nodig om meer inzicht te krijgen in hoe een goede relatie tussen "zieke-familie-begeleiders" in de zorg aan het levenseinde bevorderd kan worden. Tevens is toekomstig onderzoek gewenst om inzicht te krijgen in de perspectieven van medebewoners op zorg aan het levenseinde. Andere onderwerpen voor vervolgonderzoek zijn de zorg aan familie van mensen met een verstandelijke beperking, strategieën om mensen met een verstandelijke beperking meer te betrekken bij besluiten in de zorg aan het levenseinde en mogelijkheden om pijn en andere symptomen, evenals de zorgwensen van mensen met ernstige verstandelijke beperkingen aan het einde van het leven te herkennen.

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# Curriculum Vitae

Nienke Bekkema was born on March 10, 1979, in Almelo, the Netherlands. After finishing high school at the Erasmus in Almelo, she studied Pedagogical Sciences (Orthopedagogiek) at Groningen University. After her studies she worked as a researcher at Utrecht University (2003-2005) and TNO Quality of Life (2006-2008), with a focus on childcare and youth healthcare. She co-founded and served as a board member for EducAIDed for 5 years, an NGO aimed at improving education in developing countries. In 2008-2009 she worked abroad as an educational advisor in Honduras. In 2009 she started working at NIVEL, the Netherlands Institute for Health Services Research. Initially she studied the responsible use of guidelines among ID care staff. She subsequently worked on projects involving patient experiences in midwifery care and chronic disease management programs. In 2011 she started researching end-of-life care for people with ID. She managed four projects studying several aspects of end-of-life care and published scientific papers as well as practical tools. Her practical book for people with mild ID to assess and record their end-of-life care wishes (co-authored by Anke de Veer and Anneke Francke) won the 2015 Palliative Care Impact Prize. Under supervision of prof. Anneke Francke, prof. Cees Hertogh and dr. Anke de Veer she finished her PhD in 2016. At NIVEL Nienke was also coordinator of the ‘Panel Living Together’ (Panel Samen Leven), a panel of people with ID and their representatives.

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