Advance Care Planning

in palliative care for people with intellectual disabilities

Hille Voss

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Advance Care Planning in palliative care for people with intellectual disabilities

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General introduction

General introduction

Bram is a 50-year-old man with a severe intellectual disability. He is not able to communicate verbally and is in need of daily supervision and support. Bram lives in a residential home, uses a wheelchair and has had complex health needs throughout his life. One evening, Bram's health goes into rapid decline: he has trouble breathing and has low oxygen levels in his blood. The physician on duty does not know Bram very well. In his medical record is a signed 'do not resuscitate' declaration. No other preferences regarding end-of-life care have been noted. When the physician hears about Bram's situation, he orders an ambulance and rushes to check on him. Bram's sister, who is visiting, becomes very upset when she hears an ambulance has been called. According to the sister, she has discussed with Bram's own physician, a specialist physician for people with intellectual disabilities, that she does not want Bram to go to the hospital if Bram's situation worsens. Bram's sister stresses, "We've talked about this. He's not going to the hospital!". The physician on duty understands where Bram's sister is coming from, but he also wants Bram to have a chance of treatment. The physician on duty feels there is no time to discuss this, as an acute decision has to be made. The physician feels insecure — what would Bram's wishes be? Have these wishes been discussed? And if so, why were those wishes not recorded?

Bram's case shows that proactive and timely communication about the wishes and preferences for future medical treatment and care and the documentation of these wishes is of the utmost importance for people with an intellectual disability (ID) and their relatives. This process is referred to as Advance Care Planning (ACP). Despite the importance of ACP, it is not yet common practice, and situations as described in Bram's case occur frequently. Decisions are taken on an ad hoc basis, without sufficient time to consider all the options or discuss wishes and preferences.

This thesis aims to gain insight into ACP in palliative care for people with ID. It also describes the development, implementation and evaluation of an ACP programme for professionals in ID care.

The present introduction to this thesis starts by providing information about people with ID and their lives in the Netherlands.

Next, it elaborates on the palliative care needs of people with ID and discusses the challenges of caring for a person with ID in need of palliative care. It further explains the definition and importance of ACP.

Also, this introduction describes the factors influencing the implementation of innovations in palliative care for people with ID. Finally, the aim and structure of this thesis are presented.

People with intellectual disabilities: the Dutch context

Intellectual disability is a disability which occurs before the age of eighteen and is characterised by a limitation in intellectual functioning and in adaptive behaviour, which covers many everyday social and practical skills [1-3]. Usually, an intelligence quotient (IQ) is used to indicate the level of ID. An IQ score between 70 and 55/50 is traditionally classified as 'mild ID', between 55/50 and 40/35 as 'moderate ID', between 40/35 and 25/20 as 'severe ID' and below 25/20 as 'profound ID' [2].

In the Netherlands, there are no exact numbers about the prevalence of people with ID. However, recent estimates suggest that there are approximately 142,000 people with an IQ score of less than 70, which is nearly 1% of the population. Of the approximately 142,000 people with ID in the Netherlands, 68,000 were classified as having an IQ score below 50 [4].

Most people with ID in the Netherlands receive support from a care organisation specialised in caring for people with ID ('ID care organisation'). People with ID often live in residential settings or group homes in the community, or independently with their family or others [5]. Various professionals with different backgrounds work within ID care organisations in the Netherlands [6]. Care staff or support workers who work in an ID care organisation provide the daily support for people with ID; they are trained social workers, nurses or nursing assistants. Medical care is provided by ID physicians or general practitioners. Dutch ID physicians have received three years of specialist training in caring for people with ID. The Netherlands is the only country in the world with 'ID physician' as an acknowledged medical specialism.

Vulnerability, ageing and palliative care needs of people with intellectual disabilities

Compared with the general population, people with ID have an increased risk of motor and sensory disabilities, chronic diseases, mental health problems and syndrome-specific complications. Also, comorbidity of chronic conditions is common among people with ID [7].

People with ID die on average at an earlier age than the general population [8, 9]. Some people with ID, especially people with profound intellectual and multiple disabilities, are susceptible from a young age to life-threatening somatic conditions [10] and may require lifelong care geared to improvement of their quality of life, in other words, palliative care [11].

According to the World Health Organization [12], palliative care is: “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”. The Dutch Palliative Care Quality Framework extended this definition to also refer to “patients and their families facing the problem associated with a life-threatening illness *or vulnerability*” [13]. The extension to include vulnerability in the palliative care definition is important for people with ID, since people with ID are vulnerable in many ways (physically, psychologically and socially) and often have to deal with complex and multiple health conditions (multimorbidity) during their entire life.

Despite the fact that people with ID die on average at an earlier age than the general population, the life expectancy of people with ID is increasing [14]. Old age (older than 50), which until 30 years ago was not often seen in this population, is now quite common in the life course of people with ID [15]. Although this is a positive development, it means that people with ID are increasingly experiencing life-threatening illnesses that are particularly likely to affect older people, such as chronic cardiovascular diseases, chronic lung diseases, cancer and dementia [8, 15]. Consequently, professionals increasingly have to provide support for ageing people with ID who are in need of palliative care.

Challenges of caring for a person with ID in need of palliative care

Identifying palliative care needs and ensuring the adequate provision of palliative care for people with ID can be challenging for several reasons. Firstly, people with ID often experience difficulties in understanding their illness and have limited understanding of dying and death and few opportunities to learn about this [16-18]. People with ID are not always able to communicate their feelings or needs to others verbally, which makes it hard for professionals and relatives to assess their wishes and needs [6].

Secondly, because of communication difficulties and because people with ID often have multiple and complex health needs, whereby the underlying diagnoses or causes of the problems are not always identified [19-21], palliative care needs are often only recognised at a late stage when death is already nearing. The study by Vrijmoeth and colleagues (2016) showed that 20% of physicians did not foresee death until the last week of life [22]. Moreover, professionals might feel barriers to communicating openly about the person's condition and often do not involve people with ID in decision-making about care at the end of life [23, 24]. Professionals fear that it would only harm the person with ID if they were informed about their illness and that people with ID would not understand their situation. Some professionals also avoid talking about illness, dying and death, either to protect themselves from possible distress or because they do not know how to talk about these topics with people with ID [18, 25].

A third challenge is that professionals who provide daily care for people with ID in the Netherlands are often social workers who have received limited or no training in palliative care. Relatively few people with ID in ID care organisations require palliative care compared to other health settings such as nursing homes or hospices, which makes it difficult for professionals to develop expertise in palliative care [26-28]. Although professionals in ID care often find providing care at the end of life to be rewarding and honourable [28], the large majority of professionals feel inadequately trained to provide palliative care, especially in terms of supporting people with ID in dealing with the impending death and the farewell process [23].

A fourth challenge is that in most ID care organisations, there is no clear policy regarding palliative care or end-of-life decisions, or if such a policy exists, then professionals, relatives and the people with ID are unaware of it. As a result, professionals are uncertain about their role and responsibilities in palliative care provision [29-31].

A fifth challenge is related to access to and cooperation with specialist palliative care services. Research shows that palliative care services, such as hospices or nursing homes, are under-used by people with ID [32, 33] and there is often little collaboration with ID care organisations [34, 35]. A related problem is that professionals working in specialist palliative care services might not feel competent to engage with and care for a person with ID [18].

A final challenge concerns decision-making in palliative care [28, 36-38]. At the end of life, big, important decisions often need to be taken, e.g. regarding abandoning or continuing potentially life-prolonging treatment or regarding the place of end-of-life care. People with ID, especially people with more severe ID, depend on others to articulate their wishes and needs regarding those decisions. Deciding what is in the best interests of a person who cannot decide for themselves can be a challenging and emotional process, which should be carefully discussed with everyone who is involved. In the Netherlands, there is a model for substitute decision-making. This means that representatives are appointed for people who are incompetent in the matter at hand, for example regarding decisions about medical interventions. These patient representatives are usually close relatives of the person with ID and may feel a great responsibility to make the right decisions on behalf of their loved one [39, 40]. Within the Dutch legal system, the physician (e.g. the ID physician or the general practitioner) is ultimately responsible for medical decisions. In cases where no consensus can be reached between physicians and relatives, however, physicians find it very difficult to go against the relatives' wishes [38].

Implementation of palliative care innovations

Although the scientific knowledge about palliative care for people with ID is increasing, there is a notable shortage of palliative care innovations and lack of knowledge about what factors influence implementation and the sustainment of such innovations [41, 42]. Examples of palliative care innovations include training for professionals or tools to identify palliative care needs and symptoms. Another example is a communication-based intervention such as 'Dying Your Own Way' (in Dutch: 'STEM') [43], which is an intervention designed to accelerate expertise, to create awareness of the diversity of patients' wishes and needs at the end of life, and to improve professionals' ability to communicate proactively with patients and relatives about wishes and needs at the end of life.

Implementing care innovations in daily practice is often difficult, especially when complex changes are needed, cooperation between disciplines is required, or behaviour needs to be changed [44]. Moreover, it is important to consider how an innovation can be adapted to make it more suitable for a particular population or fit better with the organisation [45, 46]. Implementing palliative care innovations for people with ID may be particularly challenging because, as previously mentioned, professionals in organisations for people with ID are often not trained in palliative care and have limited expertise in providing palliative care [23, 26, 47, 48].

Because palliative care innovations may help ensure the timely identification of palliative care needs, proactive communication about future medical treatment and care, and improvements in the quality of palliative care, it is necessary to gain more insight into which factors are important for the successful implementation and sustained use of palliative care innovations in ID care organisations.

Importance of Advance Care Planning for people with intellectual disabilities

ACP is defined as the process of enabling individuals to define goals and preferences for future medical treatment and care, to discuss these goals and preferences with family and health-care providers, and to record and review these preferences if appropriate [49]. ACP is a broad concept and not limited to medical decisions, but should also include discussions about social, psychological and spiritual care needs. Wishes and needs for future care should be discussed with everyone who is important to the individual in question, which in the case of people with ID often includes close relatives, care staff and/or spiritual caregivers [50-53].

ACP is important in putting people with ID more in control of their own care. If possible scenarios are considered at an early stage, unforeseen, complex situations are less likely to arise, which could reduce unwanted admission to hospitals and have a positive effect on the quality of care and quality of life of a person with ID [54-56].Research has indicated that people with ID find it important to be involved in decisions about their care [17, 51, 57]. However, because of the abovementioned challenges of caring for a person with ID in need of palliative care, communication about care needs is not always done proactively; rather, adjustments in care plans tend to be made in response to problems that have arisen [57, 58]. Therefore, it is important that professionals receive training in how to provide ACP as an integral, continuous process in the care for people with ID.

The aim and structure of this thesis

This thesis aims to give insight into ACP in palliative care for people with ID. The thesis also deals with the development, implementation and evaluation of an ACP programme for professionals caring for people with ID. The thesis is divided into two parts:

Part 1. State of the art of ACP in palliative care for people with ID

The first part of this thesis focusses on the existing knowledge about ACP for people with ID and the use of ACP. The research questions addressed in part 1 can be summarised as follows:

- What is already known in the scientific literature about the use, content and effects of ACP for people with ID in need of palliative care? (*Chapter 2*)
- How does ACP currently take place within organisations for people with ID requiring palliative care and how are the wishes of people with ID taken into account? (*Chapter 3*)
- What do people with ID, relatives and professionals perceive as important for ACP in palliative care for people with ID? (*Chapter 4*)

First, a systematic review is presented of the use, content and effects of ACP in palliative care for people with ID (*Chapter 2*). *Chapter 3* describes the actual use of ACP in six ID care organisations in the Netherlands, specifically focussing on how the wishes of people with ID are being considered. To this end, analyses were performed of medical files and additional structured interviews were held with ID care professionals and relatives. In *Chapter 4*, a qualitative interview study is described focussing on what people with ID, relatives and professionals think are important elements of ACP for people with ID.

Part 2. Development, implementation and evaluation of ACP in palliative care for people with ID

The main research questions addressed in part 2 are:

- What facilitators and barriers influence the implementation process and sustainment of innovations in palliative care for people with ID? (*Chapter 5*)
- How is an ACP programme for professionals caring for people with ID developed, implemented and evaluated? (*Chapter 6*)

In part 2, we first explore which barriers and facilitators are important for the implementation and sustained use of palliative care innovations, focussing on the timely identification of palliative care needs and proactive communication in organisations for people with ID. Therefore, data was collected on nine implementation projects in ID care organisations that participated in the National Quality Improvement Programme for Palliative Care (*Chapter 5*). Moreover, based on the existing knowledge about ACP and the important factors influencing implementation, we developed and implemented an ACP programme in six ID care organisations in the Netherlands. The experiences with the ACP programme were evaluated among participating professionals providing palliative care for people with ID (*Chapter 6*).

The thesis ends with a summary and general discussion (*Chapter 7*), in which the main findings are summarised and discussed, methodological considerations are described and recommendations are made for practice and future research.

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Chapter 2

Advance care planning in palliative care for people with intellectual disabilities: a systematic review

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Abstract

Context: Advance care planning (ACP) is defined as a person-centred, ongoing process of communication that facilitates patients' understanding, reflection and discussion of goals, values and preferences for future care. There is evidence for the general palliative care population that ACP increases compliance with patients’ end-of-life preferences and improves quality of care near the end of life.

Objectives: To gain insight into what is known about the use and effects of ACP in palliative care for people with intellectual disabilities (ID).

Methods: Four databases were searched systematically: PubMed, PsycINFO, Embase and CINAHL. A stepwise procedure was used to identify relevant studies based on the Preferred Reporting Items for Systematic Reviews and Meta-Analyses Statement. The review included empirical quantitative, qualitative and mixed methods studies concerning people with ID who receive palliative care or who died non-acutely, and describing ACP. Methodological quality was graded using a critical appraisal tool.

Results: Fourteen studies were included. Most studies examined the perspective of professionals and/or relatives. None of the studies focused on the perspective of patients with ID. The studies concerned different elements of ACP, mainly decision-making and organizational policies. No effect studies were found. Obstructing factors were difficulties in recognizing palliative needs and uncertainties among relatives and professionals about their roles and tasks in ACP. Conducive factors were good working relationships between professionals and relatives.

Conclusion: There are some indications that ACP could be useful for people with ID, but more knowledge is needed about whether and how ACP should be used.

Key words: advance care planning, palliative care, intellectual disability, end-of-life decisions, organizational policies

Introduction

Background

According to the World Health Organization (WHO), 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” [1]. Intellectual disability (ID) is defined as a disability characterized by significant limitations both in intellectual functioning and in adaptive behaviour, which covers many everyday social and practical skills. The disability develops before the age of eighteen and is often expressed using the IQ scale (with profound to mild intellectual disability covering the IQ range 0-70) [2].

People with ID have twice as many health problems as the general population [3]. Moreover, the reported prevalence of chronic health conditions in children with ID is much higher than in the general population [4]. Among them, there are individuals who are extremely fragile in terms of their health from birth on. Therefore, people with ID could have palliative care needs at an early stage of life. On the other hand, the life expectancy of people with ID has increased in line with that of the general population because of social and medical advances [5]. This epidemiological development is linked to growing incidences of life-threatening illnesses at an older age, such as progressive cancer, chronic cardiovascular diseases, chronic lung diseases and dementia [6-8]. Consequently, relatives and professionals increasingly have to cope with people with ID who are in need of palliative care.

Advance care planning (ACP) can be seen as an integral process of palliative care [9] and is defined as a person-centred, ongoing process of communication that facilitates patients' understanding, reflection and discussion of goals, values and preferences for future care [10]. ACP has already been studied in various settings and populations and there is evidence that ACP increases compliance with patients’ end-of-life wishes and satisfaction with palliative care [11]. In elderly patients, ACP has been shown to improve the quality of palliative care and patients' and families’ satisfaction [12]. A recent systematic review among nursing-home residents showed that ACP had beneficial effects in this population and led to more actions consistent with residents' wishes and a reduction in unwanted medical interventions at the end of life [13].

The ACP framework and the way in which ACP highlights the wishes and preferences of patients may also be applicable and important for people with ID and their relatives. Due to their limited capacity for understanding and communication, people with ID often do not understand their own health condition, can experience difficulty expressing pain and other symptoms and feelings, and have difficulties with medical examinations or interventions. End-of-life decisions should therefore always be carefully weighed against the benefits for the patient’s quality of life. In ACP for people with ID, all stakeholders, including professionals, relatives and the patient if capable, should be involved in a timely discussion about wishes for future care. As ACP is a broad concept, problems with medical examinations and treatment options now and in the future should be part of the discussions, but ACP also encompasses psychological, social and spiritual matters in palliative care. For example, discussions about the place of palliative care (whether people prefer to stay in their own home environment or move to a hospital or nursing home) [14], or what kind of activities a person would still want to do, can also be elements of ACP. It is important that everybody who is involved in the palliative care for a person with ID joins in discussing the possibilities and restrictions with regard to future care.

Professionals often find it challenging to initiate the process of ACP [15]. In patients with cancer, physicians wrongly avoid ACP discussions because they are afraid such conversations will cause psychological suffering [16]. Research in ID care showed that if relatives and professionals were uncertain whether a person with ID could understand the information, they tended to withhold potentially upsetting news to spare the individual from distress [17]. Moreover, a retrospective study of the medical files of people who died in a Dutch healthcare centre showed that patients with ID were not actively involved in any of the end-of-life decisions [18]. However, research also shows that most individuals with mild or moderate ID want to be involved. Moreover, they are able to express their views on end-of-life care provision, understand treatment information and make treatment choices, although that ability decreases with the increasing complexity of decision-making regarding care [19-21].

Objectives

Because of the limitations of people with ID in understanding and communicating information, the growing number of people with ID in need of palliative care, and the importance of advance planning in making sure that palliative care is in accordance with a persons' wishes, we want to gain more insight into what is known about the use and effects of ACP in palliative care for people with ID.

The questions addressed in this systematic literature review are:

1. What is known about the use and content of ACP or elements of ACP in palliative care for people with ID?
2. Is there evidence that ACP leads to (a) a higher quality of palliative care and (b) a higher quality of life for people with ID?
3. When is ACP initiated and what are the conducive and obstructing factors when introducing ACP in palliative care for people with ID?

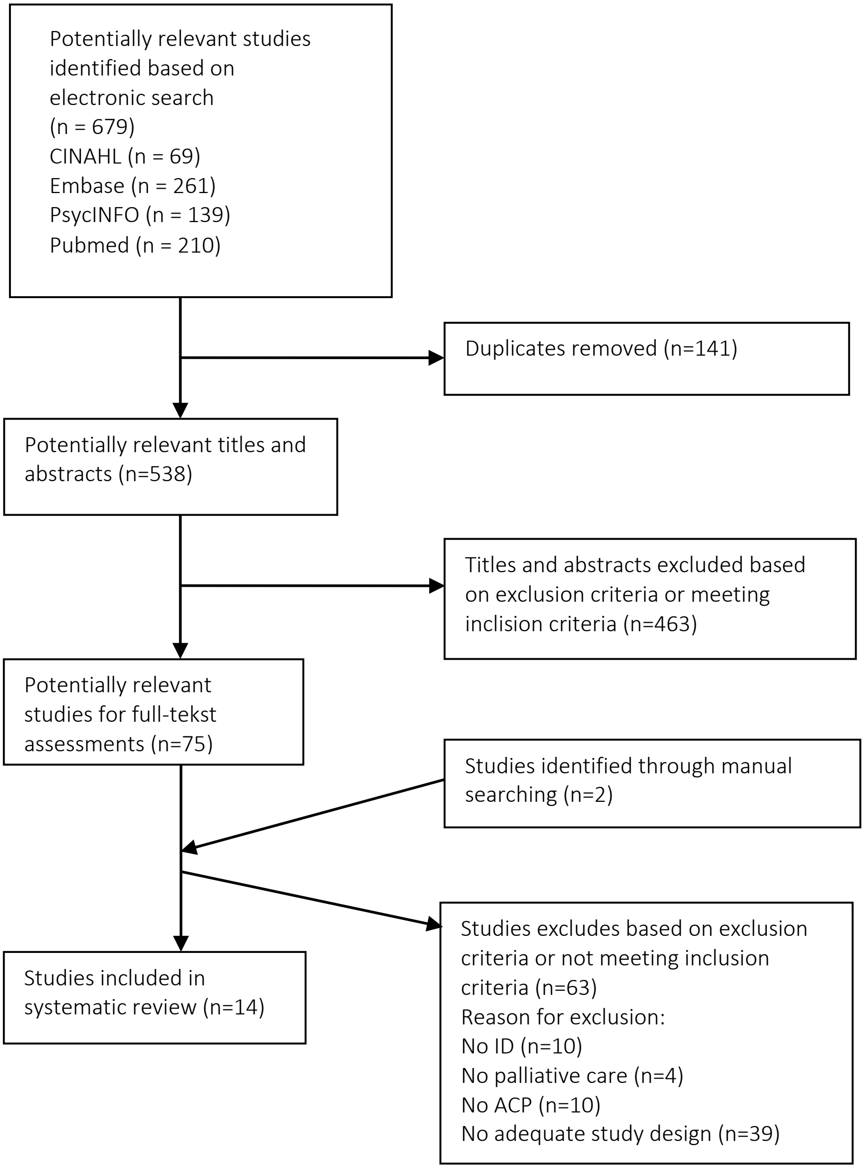
Methods

Design

A stepwise procedure was used to identify relevant papers based on the Preferred Reporting Items for Systematic Reviews and Meta-Analyses Statement (PRISMA, see Figure 2.1) [22]. In the first step, all the titles and abstracts of the references identified in the searches were read and an assessment made as to whether they appeared to meet the inclusion or exclusion criteria (see Box 2.1). In a second step, the first two authors (HV, AV) read the full texts of all the references that potentially met the inclusion criteria to see whether they indeed met the inclusion criteria. The inclusion process was carried out by the two researchers independently (HV, AV). Disagreements were resolved through discussion or, if there were still doubts, by consulting a third researcher (AdV).

The methodological quality of the studies was graded using a critical appraisal tool [23]. This tool was chosen because it was specifically developed to assess multiple kinds of study designs. It consists of nine items (abstract and title, introduction and aims, method and data, sampling, data analysis, ethics and bias, results, transferability or generalizability, and implications and usefulness). Each item can be scored on a 4-point scale ranging from very poor (a score of 1) to good (a score of 4). The total scores can range from 9 to 36. Scores of 18 or less were labelled ‘poor’ methodological quality, from 19 to 27 ‘moderate’ quality and above 27 ‘good’ quality. The methodological assessment of each study was done by two researchers independently (HV and AdV or HV and AV). If the maximum difference in the scores of the two researchers was 5, the methodological quality was the average of the two scores. Disagreement between the two researchers (i.e. a difference of more than 5 points) did not occur. To prevent bias, the methodological assessment of a study was never performed by a researcher who had co-authored that publication.

Figure 2.1 Flow diagram based on the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) [22]



Information was extracted by one researcher (HV) and checked by a second researcher (AdV) using a standardized data extraction form (see Appendix A). The extracted data included study identification, study design and data collection, background information on the people with ID, results of the study, and strengths and limitations of the study design.

Data sources

The following international literature databases were searched: PubMed, PsycINFO, Embase and CINAHL. The NIVEL library and Google Scholar were also used to search for relevant studies meeting the inclusion criteria. The search string presented in Box 2.2 was used for PubMed and checked by an experienced librarian. For other databases, the search was based on the string used for PubMed with adjustments where necessary. The databases were searched in June 2016. No language or period restrictions were applied.

Box 2.1 Inclusion and exclusion criteria

Inclusion criteria:

- describes empirical qualitative, quantitative or mixed methods research
- concerns people with ID who receive palliative care and/or their relatives/professionals OR concerns people with ID who died non-acutely (after an identifiable period of illness) and/or their relatives/professionals
- describes the use of ACP or elements of ACP such as physical, psychological, social or spiritual matters in palliative care, AND/OR effects of ACP or elements of ACP on the quality of palliative care/quality of life

Exclusion criteria:

- letters, editorials, comments or congress abstracts
- case stories that are not analysed systematically
- literature studies (although their reference lists were studied to identify relevant empirical studies)

Box 2.2 Search string used for PubMed

("Intellectual Disability"[MeSH Terms] OR (mental*[tiab] or developmental*[tiab] OR intellectual*[tiab] OR cognitive[tiab] OR learning[tiab]) AND (retard*[tiab] OR disab*[tiab] OR deficien*[tiab] OR delay*[tiab])) AND ("palliative care"[MeSH Terms] OR "hospice and palliative care nursing"[MeSH Terms] OR "terminal care"[MeSH Terms] OR "life support care"[MeSH Terms] OR "hospice care"[MeSH Terms] OR "emergency treatment"[MeSH Terms] OR "resuscitation orders"[MeSH Terms] OR hospice and palliative care nursing[tiab] OR (palliative[tiab] OR terminal[tiab] OR hospice*[tiab] OR life support*[tiab] OR end-of-life[tiab] OR emergenc*[tiab]) AND (care[tiab] OR caring[tiab] OR nurs*[tiab] OR therapy[tiab] OR sedation[tiab]) OR dying[tiab] OR death[tiab] OR end-of-life[tiab] OR terminally ill[tiab] OR critically ill[tiab] OR do-not- resuscitate order[tiab] OR DNR[tiab] OR resuscitat*[tiab] OR do-not-hospitalize order[tiab] OR DNH[tiab] OR hospitalize order[tiab] OR euthanasia[tiab] OR assisted suicide[tiab] OR treatment withholding[tiab] OR life sustaining treatment[tiab] OR emergency treatment[tiab] OR limited life*) AND ("advance care planning"[MeSH Terms] OR "advance directives"[MeSH Terms] OR "patient care planning"[MeSH Terms] OR "living wills"[MeSH Terms] OR (advance*[tiab] OR medical*[tiab] OR end-of- life[tiab]) AND (directive*[tiab] OR care plan[tiab] OR care planning[tiab] OR decision*[tiab] OR decision-making[tiab]) OR decision-making[tiab] OR supported decision-making[tiab] OR truth telling[tiab] OR disclos*[tiab] OR living will*[tiab]) AND hasabstract[text]

Results

The database search resulted in 538 studies after excluding 141 duplicates. Based on the title and/or abstract, 463 studies were excluded due to meeting exclusion criteria and/or not meeting inclusion criteria. Two studies were added after manually searching Google Scholar. Based on the full text assessment, 14 studies met the inclusion criteria and were included in the systematic review (see Figure 2.1). Table 2.1 shows the main characteristics of the studies included in this review. Eleven studies focused on one perspective: that of professionals, relatives, managers or medical records. Three studies reported on multiple perspectives. Of these, two reported on professionals and relatives [24, 25] and one on professionals, relatives and medical records [26]. No studies focused on the perspective of the patient with ID. Most studies focused on one element of ACP; four studies focused on two or more elements. The majority of the studies included made use of retrospective data (n=10). The quality of the studies varied from moderate to good. Table 2.2 shows the data extracted from the studies.

Table 2.1: Characteristics of the studies included (n = 14)

Study characteristics		**N**
Type of study	Quantitative	7
	Qualitative	3
	Mixed methods	4
Country	The Netherlands	5
	USA	4
	Switzerland	2
	UK	2
	Belgium	1
Population studied	Adults with intellectual disability	
	Mild to profound	7
	Degree of disability unknown/not reported	4
	Children and young adults with severe intellectual disability and complex medical problems	3
Perspective of*	Professionals	7
	Relatives	5
	Managers	3
	Medical records	3
Element of ACP*	Decision-making	
	End-of-life decisions	5
	Decision-making about medical interventions	1
	Decisions about place of palliative care	1
	Decision-making in palliative care	1
	Organizational policies	5
	Collaboration in palliative care	3
	Communication around advance directives	1
	Inclusion of family	1
	Documentation of wishes for future care	1
	Start of ACP	1

*Note. Categories are not mutually exclusive

Table 2.2: Extracted data for the studies that were included (n = 14)

	1. Study ID	2. Study design and data collection	3. Background information on people with ID	4. Results	5. Strengths of study design and limitations	6.Methodological quality
	• Reference authors/ date/title • Country • Aim and/or research questions	• Quantitative and/or qualitative • Total number of participants (n) and response rate • Type of analyses • Perspective of professionals, patients, relatives, others	• Age • Severity of disability • Medical condition/ cause of death • Residence	• Content of ACP • Answers to research questions • Main conclusion by authors		
1	• Bekkema, N. et al. (2014). Decision making about medical interventions in the end-of-life care of people withintellectual disabilities: A national survey of the considerations and beliefs of GPs, ID physicians and care staff. (29) • The Netherlands • Research questions: 1. What considerations do professionals take into account in decision-making about the use of potentially burdensome medical interventions? 2. What beliefs do professionals have about the use of potentially burdensome medical interventions in the end-of-life care for people with ID?	• Quantitative; pre-structured question-naires • 718 questionnaires completed, overall response rate: 46%. 248 ID physicians, GPs and care staff completed questions about the last patient with ID for whom they provided end-of-life care. Considerations in decisions about the use of potentially burdensome medical interventions were described for 140 people with ID. • Descriptive analyses • Perspective of professionals	• Age not reported/ unknown • Mild/ moderate ID (52%) and severe/ profound ID (48%) • Causes of death: cancer (37%), overall decline due to old age (17%), dementia (14%), heart failure (6%), stroke (5%), asthma/COPD (2%), other/ unknown (19%) • Residence not reported/ unknown	• Content of ACP: end of life decision-making about medical interventions • Answers to research questions: - Considerations concerning the quality of life and wellbeing (53%), the wishes of family members (23%) and the wishes of patient with ID (21%) were the most common considerations in decisions about the start or continuation of interventions. - The decision to forgo or withdraw an intervention was based on futility of the intervention (38%), quality of life and wellbeing (37%), the direct physical strain on the patient from the intervention (28%), the wishes/preferences of family members (28%), and the direct emotional strain on the patient from the intervention (25%). - The wishes of people with mild/moderate ID were considered more often (28%) than the wishes of people with severe/profound ID (3%). 93% of professionals believe that considerations concerning the quality of life are most important, 72% believe that the patients' possible discomfort outweighs prolongation of life. 70% believe that people with ID should always be informed about intervention options, even when the ability to	• Strengths: three different groups of professionals were incorporated • Limitations: - Questionnaire with multiple choice list which may influenced respondents' answers - Recall bias: respondents may be more likely to recall patients who made agreat impact on them - No knowledge about personal input and role of respondent in decision-making process.	Good

				communicate is limited. Only 43% believe the wishes of the patient with ID should be the deciding factor. If a patient with ID is unable to decide for themselves, 52% of professionals believe that the opinion of the relatives should be followed, 40% believe the opinion of the physician is decisive. • Main conclusion by authors: Professionals believe the quality of life is most important. The wishes of people with ID were often not considered in decision-making about medical interventions.		
2	• Bekkema, N. et al. (2015). '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. (32) • The Netherlands • Aim: study general beliefs and considerations in decision-making about the best place for end-of-life care Research questions: 1. To what extent do people with ID move to another care residence to receive end-of-life care? 2. What considerations do professionals take into account when deciding on the	• Quantitative; pre-structured questionnaires • 255 care staff and ID physicians responded about the last patient with ID for whom they provided palliative care; response rate for care staff: 67%; rate for ID physicians: 53%. Considerations underlying decisions about the place of palliative care were described for a total of 237 people with ID. • Descriptive analyses • Perspective of professionals	• Age, severity of disability and medical condition not reported/ unknown • Residential setting (56%), community residence (34%), living independently or with family (7%), other/ unknown (3%)	• Content of ACP: end of life decisions about the place of palliative care • Answers to research questions: - 79% of people with ID received end-of-life care in their own home environment. - Familiarity with the environment (59%), teams' expertise in end-of-life care (45%) and the wishes/preferences of family members (16%) Were the most frequently mentioned considerations underlying the decision to provide end-of-life care in the patient's original home environment. The most frequently mentioned considerations underlying the decision to move the patient to another place were: teams' lack of expertise (57%), the lack of equipment in the residence (28%) and the unavailability of 24/7 care (17%). The wishes of the patient with ID were mentioned by 8% of professionals as a consideration in whether or not to move the patient. - 89% of professionals believe that every effort should be made to ensure	• Strengths: broad group of professionals • Limitations: - Overrepres-entation of people living in a residential residence - Retrospective study	Good

	place of end-of-life care for people with ID? 3. What beliefs do professionals have with regard to an appropriate environment for end-of-life care for people with ID?			the patient can stay in their own home environment. 82% believe care should be offered 24/7. 72% believe the wishes of the patient with ID should be the deciding factor, 41% believe this preference outweighs the quality of care that can be given at that place. • Main conclusion by authors: Despite the belief of professionals that the wishes of the patient with ID should always be given top priority in deciding on the place of care, only 8% of the professionals mentioned that the wishes of the patient were taken into account in this decision.		
	1. Study ID	**2. Study design and data collection**	**3. Background information on people with ID**	**4. Results**	**5. Strengths of study design and limitations**	**6.Methodological quality**
3	• Brown H. et al. (2003). ‘Please don’t let it happen on my shift!’ Supporting staff who are caring for people with learning disabilities who are dying. (24) • UK • Aim: document how agencies mobilized services and made decisions, how agencies worked together and what support staff needed in the patient’s last months and weeks.	• Qualitative • Interviews with care staff (n=31), service managers (n=16), health/learning disability professionals (n=18), service users (n=3), family members (n=6), local community representatives (n=8), and external representative (n=1) about 21 patients with learning disabilities who had died. • Five meetings with staff/ team and one with a group of older people with learning disabilities (not reported). • Data were analysed by making a graph of the trajectory of illness and key transition points and a network map showing	• Age range at time of death: 22 – 80 years • Down syndrome (5), multiple physical and cognitive impairments (2), cerebralpalsy (2), mild learning disability (3), moderate learning disability (6), severe learning disability (1), unknown (2) • Causes of death: cancer (n=8), dementia (n=3), respiratory disease/ difficulties (n=3), pneumonia (n=2),congenital impairments (n=1), brain tumour (n=1), emphysema (n=1), epilepsy (n=1), unknown (n=1) • Different community-	• Content of ACP: decision-making in palliative care, organizational policies, collaboration in palliative care • Answers to research questions: - Diagnosis of the illness was often late because professionals saw symptoms as part of the learning disability and patients were not able to monitor changes in their own health or express concerns about symptoms. - Services were well supported by primary care teams and by oncology services but less by specialist dementia services. Only one patient had access to a hospice, but the hospice staff were unable to meet his additional needs. - There were no formal protocols in hospitals on care for people with learning disabilities. There was no agreed format for decision-making in any of the cases. - Professionals report	• Strengths and limitations of the study: not mentioned	Moderate

		the involvement of various agencies for each patient separately • Perspective of professionals and relatives	based services (e.g. private residential home, staffed housing service, long-stay hospital and community learning disability service).	confusion about who should be involved in decision-making and especially about the role of relatives. - Decision-making issues were most acute at the point where a shift of treatment goals was most explicit. - Only in the case of one patient were professionals open to the patient about his impending death. • Main conclusion by authors: Professionals were not prepared for disclosure to the patient about their impending death because they had the notion that the patient would not understand and they did not know who had the authority to tell the patient directly.		
4	• D'Haene, I. et al. (2010). End-of-life care policies in Flemish residential care facilities accommo-dating persons with intellectual disabilities. (33) • Belgium • Aim: describe presence, content and implement-tation strategies of written policies on medical end-of-life decisions (ELDs)	• Quantitative and qualitative study • Postal survey among directors of residential care facilities (n=84, response rate 60%). Descriptive analyses. • Systematic content analysis of policy documents (from 25 institutions). Two researchers performed the analysis independently. • 'Other' perspective: Management	• Age not reported/un-known • Occupation-al type and nursing type (high care needs) for residents • Residence: all residential care facilities (RCF) for people with ID, no selection for type of facility.	• Content of ACP: organizational policies • Answers to research questions: - 35% of the institutions have a policy on ELD - Policy documents contained statements about the policy on one or more end-of-life decisions, bereavement care, communication with the family, special needs of people with ID. - Communication of ELD policies to professionals was standard practice in 67-88% of the institutions. Communication of ELD policies to residents and families varied between being standard practice (37-56%) and occurring on request (39%-62%) in institutions. • Main conclusion: End of life care policies are often not present in Flemish RCFs and are not specifically designed for people with ID.	• Strengths: not mentioned • Limitations: - Self-reported data: potentially recall and social desirability bias. - Multiple disabilities of people with ID in RCF unknown: hard to generalize.	Good

	1. Study ID	2. Study design and data collection	3. Background information on people with ID	4. Results	5. Strengths of study design and limitations	6.Method-ological quality
5	• Friedman, S. L. (2006). Parent resuscitation preferences for young people with severe developmental disabilities. (34) • USA • Aim: Assess the effects of providing information regarding resuscitation to parents and/or guardians of children with severe developmental disabilities	• Quantitative; effect study, chart review, before and after intervention. The intervention consisted of written information for parents about resuscitation policy in the case of cardio-pulmonary arrest. Parents requesting resuscitation returned a form that specified their wishes. The other parents were contacted by phone and an appointment was made with the child's physician. • Charts of 60 patients were reviewed. After two years, all parents responded (response rate 100%) • Descriptive, univariate and multivariate analyses • 'Other' perspective: Medical records	• Age: 1 to 32 years (mean 21) • Residence: A paediatric nursing home for children with severe developmental disabilities and complex medical problems requiring 24-hour skilled nursing care.	• Content of ACP: organizational policies • Answers to research questions: The preference for do not resuscitate (DNR) increased from 18% to 43%. There were no parents who wished to change their child's status from DNR to resuscitation. • Main conclusion by authors: Provision of information about resuscitation resulted in a significant increase in those who chose DNR rather than full resuscitation.	• Strengths: not mentioned • Limitations: - Small population - Variables that impact outcome were not available (quality of life) - Data were collected retrospectively via chart review; issues in accuracy and detail of information - Relatively old data; social and political climate could be different regarding end-of-life considerations - No generalizability to children who reside in their own home or children with other chronic medical conditions	Good
6	• Friedman, S. L. & Gilmore, D. (2007). Factors that impact resuscitation preferences for young people with severe developmental disabilities. (35) • USA • Aim: to identify factors that influence parents' and guardians' resuscitation decisions	• Quantitative; pre-structured questionnaire. The survey was part of the implementation of a so-called Comfort Care programme for residents with a DNR order. • 30 parents/ guardians (Response rate 46%) • Descriptive analysis • Perspective of relatives	• Age: 2 to 36 years (mean 19) • Residence: A paediatric nursing home that provides care for young people with severe developmental disabilities and complex medical problems requiring 24-hour skilled nursing care.	• Content of ACP: organizational policies • Answers to research questions: - Provision of explanatory information about resuscitation resulted in a significant increase in those who chose DNR rather than full resuscitation. There were no parents who wished to change their child's status from DNR to resuscitation. - Perceptions of the quality of life and medical condition of patient with ID were not significantly different between groups who chose resus-citation and those with a preference for DNR.	• Strengths: not mentioned • Limitations: - Survey sample did not reflect population of facility and findings cannot be generalized to other groups of individuals - Small number of partici-pants - No use of standardized measures of quality of life - Other potential contributing factors (parental emotional health and marital relationships) not	Good

				- Parents who chose resuscitation were more influenced by family members (29%), religious leaders (22%) and discussions with the physician (44%) compared to those with a preference for DNR (resp. 0%, 0% and 11%). - Parents who opted for DNR were less likely to discuss comfort management with the physician at the end of life. • Main conclusion by authors: - Parents changed their resuscitation preference to DNR after explanations were provided. Interpersonal relationships were more influential for parents who chose full resuscitation.	evaluated.	
7	• Grossberg, R. I. et al. (2013). Direct care staff and parents'/ legal guardians' perspective on end-of-life care in a long-term care facility for medically fragile and intellectually disabled pediatric and young adult residents. (25) • USA • Aim: determine the impact of residents' end-of-life experience on their primary care staff members and their parents/ guardians	• Quantitative and qualitative 1. Questionnaires about end-of-life experience; 11 bereaved parents/ guardians (response rate: 23%) and 18 care staff members who cared for the resident for at least 5 days during the last 30 days prior to death (response rate: 29%) responded. Descriptive analyses. 2. Thematic analysis by two independent statisticians of additional comments on the perspectives of end-of-life care survey. • Perspective of professionals and relatives	• Age: 10-29 years at death • Cause of death included respiratory failure/pneumonia, sepsis, intractable seizures, and complications in the underlying disease state/ syndrome • Residence: A specialized long-term care facility for children and young adults with neurologic impairments and severe to profound intellectual disability who are considered medically fragile	• Content of ACP: communication around advance directives, inclusion of family • Answers to research questions: - Parents gave the highest ratings (mean of 4.6 with 1 being very dissatisfied and 5 very satisfied) for doctor's attention to patients' description of symptoms, nursing availability, and family inclusion in treatment and care decisions. Care staff gave the highest rating for nursing availability (mean 4.2). Parents were more satisfied (median 4.2) with the care provided in the palliative phase than were care staff (median 3.8) - Parents reported good communication around advanced directives - Care staff noted the importance of nurturing relationships with the resident's family • Main conclusion by authors: Overall satisfaction with the care provided by parents and care staff. Both parents and care staff	• Strengths: not mentioned • Limitations: - Low response rates - Retrospective nature (recall bias) - Nature of a survey	Good

				felt the patients’ needs in terms of pain control, respect and decision-making by parents were adequately met.		

	1. Study ID	2. Study design and data collection	3. Background information on people with ID	4. Results	5. Strengths of study design and limitations	6.Method-ological quality
8	• Heslop, P. et al. (2013). Confidential Inquiry into premature deaths of people with learning disabilities (CIPOLD). Final Report. (26) • UK • Aim: to review the patterns of care that people received in the period leading up to their deaths, to identify errors or omissions contributing to these deaths, to illustrate evidence of good practice, and to provide improved evidence on avoiding premature death.	• Quantitative and qualitative 1. Retrospective study of the deaths of 247 people with learning disabilities in 2010–2012 2. Interviews with professionals, family members (response rate 34%) and friends and panel meetings with everyone involved in supporting the person • Medical records, perspective of professionals and relatives	• The median age at death was 65 for men and 63 for women • 40% had mild, 31% moderate, 21% severe, and 8% profound and multiple learning disabilities • Frequent causes of death were disorders relating to the heart and circulatory disorders (22%) and cancer (20%). • Residence: residential home (46%), nursing home (18%), housing provider (13%), parental home (13%), own home (8%), other private residence (2%)	• Content of ACP: documentation of wishes for future care, start of ACP, collaboration in palliative care • Answers to research questions - End-of-life care planning took place for two-fifths (43%) of people with learning disabilities. Mostly in the form of a ‘When I die’ booklet that documented a person’s preferences for palliative care. A ‘Child and Family Wishes Advance Care Plan’ had been used for some children. - There was evidence that ACP contributed to effective care and that professionals acted according to the plans. After ACP discussions, professionals felt more confident. - Planning often appeared to be responsive to problems that arose, rather than anticipating problems that could arise in the future. Unplanned end-of-life care increased the risk for not having a ‘good death’. - 20% had received support from a specialist palliative care team, 10% from a hospice, and 6 persons had died in a hospice. • Main conclusion by authors: - Inattention to predicting potential problems, recognizing changing needs and adjusting the provision of care as needs changed made people with learning disabilities particularly vulnerable to premature death.	• Strengths and limitations of the study design: not mentioned	Good

				- People with learning disabilities were less likely than the comparator group of people without learning disabilities to have access to specialist palliative care services.		
9	• Ronneberg C. R. et al. (2015). Promoting collaboration between hospice and palliative care providers and adult day services (ADS) for individuals with intellec-tual and develop-mental disabilities. (36) • USA • Aim: -Determine whether collaborations exist between ADS and hospice/palliative care organizations -Explore existing barriers to collaboration	• Quantitative and qualitative 1. Two online surveys: One for ADS providers with 103 respondents (response rate: 15%) and one for hospice/palliative care providers with 87 respondents (response rate: 0.05%). 2. Four focus groups, two with 10 ADS professionals, and two with 7 hospice/palliative care providers. • Perspective of professionals	• Age, severity of disability, and medical condition of people with ID not reported/ unknown • Residence: hospices and adult day services (48% of ADS with patients with acquired/developmental disabilities as primary population) across the USA and organized in an umbrella organisation	• Content of ACP: collaboration in palliative care, organizational policies • Answer to research questions: - 86% of ADS providers were very or somewhat familiar with hospices. 74% of hospice providers reported being very or somewhat familiar with ADS. 55% of ADS providers had an ongoing relationship with one or more hospice programmes. 36% of hospice providers reported engagement in activities with one or more ADS program-mes. Familiarity with hospices among the ADS providers in the focus group varied widely. To some extent, end-of-life program-ming takes place in ADS programmes: bereavement and memorial services were provided for staff, patients and their families. Almost half of the hospice and palliative care providers in the focus group referred their patients to ADS. - ADS providers in the focus group noted a lack of clear DNR policies within ADS centres. Recognition of nonverbal symptoms of pain was a challenge in ADS centres. Hospice service providers reported limited experience with people with ID. • Main conclusion by authors: - A small number of patients are receiving dual services (hospice and ADS). Both services recognize the potential benefits of collaboration: increasing choice, improving quality of care and quality of life for patients	• Strengths: not mentioned • Limitations: - Small sample size and limited by including only organizations that were members of NADSA, Easter Seals and NHPCO. - Nonprobability sampling - Self-selection of sample - Grounded study	Moderate

	1. Study ID	2. Study design and data collection	3. Background information on people with ID	4. Results	5. Strengths of study design and limitations	6.Method-ological quality
10	• Wagemans A. et al (2010). End-of-life decisions: an important theme in the care for people with intellectual disabilities. (18) • The Netherlands • Aim: investigate prevalence and nature of end-of-life decisions in a residential care centre	• Quantitative; retrospective study of medical files of people who died between January 2002 and July 2007 (n=47), systematically reviewed using a checklist • ‘Other’ perspective: Medical records	• 19 residents died in their fifties • Severity of disability: mild to profound • Causes of death: pneumonia (n=9), exhaustion (n=6), heart failure (n=3), peritonitis (n=3), epileptic seizure (n=2), cerebro-vascular accident (CVA, n=2), choking (n=1), unknown (n=21) • Residence: Dutch residential care facility providing care to 335 children and adults with ID living in wards or group homes of 4 to 12 patients	• Content of ACP: end-of-life decisions • Answer to research questions: -One or more end-of-life decisions (withholding nasal feeding tube, withholding therapy, DNR, pain relief/symptom control, second opinion) were made in 27 out of 47 cases -The family was involved in half of the cases; nurses were involved in all of the cases. Parents, if involved, had a very important voice in taking end-of-life decisions -No information was noted in the files about the views of the residents themselves or about their capacity to take an end-of-life decision. • Main conclusion by authors: Medical end-of-life decisions were made for significant numbers of people with ID	• Strengths: not mentioned • Limitations; - Only one service included (small sample and not representative) - Retrospective study	Good
11	• Wagemans A. et al. (2013). End-of-life decisions for people with intellectual disabilities, an interview study with patient represen-tatives. (30) • The Netherlands • Aim: investigate the role and influence of patient representatives in the process of making end-of-life decisions	• Qualitative study based on semi-structured interviews with patient representatives about deceased patients with ID who died in the past year (n=10). The interviews were analysed following the procedures of Grounded Theory. • Perspective of relatives	• Age range: 40 – 78 years • Mild ID (n=4), moderate ID (n=1), severe ID (n=3), profound ID (n=2) • Causes of death: cancer (n=3), feeding problems (n=2), cognitive or neurological decline (n=2), Parkinson’s disease(n=1), COPD (n=1), dementia (n=1) • Different residential facilities for people with ID	• Content of ACP: end-of-life decisions • Answers to research questions: - The patient representatives felt highly responsible for the end-of-life decisions and were passionately involved with the patient. - The patient representatives were not sure about the boundaries and limitations of deciding for someone else. They were also not sure about the relevant legislation. - Patient representatives were unsure if patients with ID were able to make choices by themselves and could understand the burden of interventions. - Apart from the doctors, no other professional care providers, such as nurses, social workers or priests, were involved in the decision-making process.	• Strengths: not mentioned • Limitation: only included deceased patients whom the patient representatives had known intimately throughout their lives.	Good

				- Quality of life and prevention from (further) suffering were the most important considerations in the decision-making process. - Support from doctors was important for the representatives. Patient representatives were unaware that doctors are ultimately responsible for end-of-life decisions • Main conclusion by authors: According to patient representatives, the process of end-of-life decision making can be improved by ensuring clear roles and an explicit description of the tasks and responsibilities of all participants.		
12	• Wagemans A. et al. (2013). The factors affecting end-of-life decision-making by physicians of people with intellectual disabilities in the Netherlands: a qualitative study. (31) • The Netherlands • Aim: investigate the process of end-of-life decision-making from the perspective of physicians	• Qualitative study • Based on semi-structured interviews with 7 ID physicians about deceased patients with ID who died in the past year after a process of end-of-life decision-making (n=10). • The interviews were analysed following the procedures of Grounded Theory. • Perspective of professionals	• Age range: 40 – 78 years • Mild ID (n=4), moderate ID (n=1), severe ID (n=3), profound ID (n=2) • Causes of death: cancer (n=3), feeding problems (n=2), cognitive or neurological decline (n=2), Parkinson's disease(n=1), COPD (n=1), dementia (n=1) • Small facilities in the community or on a larger campus for people with ID	• Content of ACP: end-of-life decisions • Answers to research questions: - No patients were involved in the decision-making process. According to physicians, patients were unable to understand the consequences of a choice and could not comprehend the process of decision-making. - In all but one case, the physicians followed the wishes of the relatives. - Physicians preferred to discuss end-of-life decisions in a stable and calm situation, when emotions were under control and enough time could be spent on a sensible discussion. - According to physicians, good working relations with relatives and paid care staff was the most important contributory factor in ensuring an effective process of decision-making. • Main conclusion by authors: Physicians gave a lot of weight to the opinions and wishes of relatives in the end-of-life decision process.	• Strengths: - ID physicians were open about shortcomings and doubts - Representative sample of ID physicians • Limitations: - Not representative for GPs or medical specialists - Retrospective study, recall bias	Good

	1. Study ID	**2. Study design and data collection**	**3. Background information on people with ID**	**4. Results**	**5. Strengths of study design and limitations**	**6.Method-ological quality**
13	• Wicki, M.T. & Hattich,A. (2016). End of life decisions for people with intellectual disability – a Swiss survey. (27) • Switzerland • Aim: explore the prevalence and nature of end-of-life decisions and whether residents are involved in making these end-of-life decisions. Research question: - Is there a difference in end-of-life decisions and involvement between people with ID and people with other disabilities (sensory, physical and psychosocial)?	• Quantitative study • Based on cross-sectional survey with written questionnaires filled in by 78 directors of residential homes (response rate 58%) about people who died (n =233), 97 patients with ID, 59 patients with ID and another disability, 77 patients without ID but with one or more other disabilities (sensory, physical, and/or psychological). • Descriptive statistics • 'Other' perspective: Management	• Mean age at death 56.3 years (range 17-98) • 5.8% had a very low, 5.8% a low, 26.3% a middle, and 57.7% a high degree of disability. The degree of disability was unknown for 4.4% • Causes of death were coronary diseases (30.7%), respiratory diseases (16.1%), and cancer (13.5%). 39.7% died due to other or unknown causes • All residential homes for adults with disabilities (6 – 300 people)	• Content of ACP: end-of-life decisions • Answers to research questions: - End-of-life decisions were made for 164 residents (70.4%). These decisions were made significantly more often for residents with ID (74.4%) than for residents without ID (62.3%). - When end-of-life decisions were made, family members were involved in the decision-making process in 120 out of 164 cases (73.2%). Legal representatives were involved in 142 cases (86.6%). Nurses were involved in 136 cases (82.9%). - People with ID were less involved in end-of-life decisions than people with other disabilities. 30.1% of people with ID were not involved at all in end-of-life decisions versus 6.5% of people without ID. -The decision to withhold life-prolonging treatment was more likely to be made if advanced directives were present, if the degree of disability was higher and if residents had an ID. • Main conclusion by authors: Decisions concerning withholding life-prolonging treatment are made more often for people with ID than for people with other disabilities, and people with ID are less likely to be involved in these decisions.	• Strengths: not mentioned • Limitations: - Retrospective design and recall bias - End-of-life decisions reported not by physicians but by directors - Because of cross-sectional nature, no definite conclusions can be drawn - No possibility to analyse further factors that could determine the prevalence of end-of-life decisions (such as health condition and prognosis) - Directors described a higher degree of disability for people with ID than for people with other disabilities	Good
14	• Wicki M. T. (2016). Withholding treatment and intellectual disability: second survey on end-of-life decisions in Switzerland. (28) • Switzerland	• Quantitative study • Based on cross-sectional survey with written questionnaires. Of 156 residential homes for people with disabilities in the three biggest German-speaking	• Mean age of death 57.2 years • Severity of disability not reported/ unknown • Causes of death were heart and circulatory dis-	• Content of ACP: end-of-life decisions • Answers to research questions: - An end-of-life decision was made in total for 44 people (53.7%) of whom 28 people with ID (62.2%) and 16 people without ID (43.2%). This difference was	• Strengths: not mentioned • Limitations: - Retrospective design and recall bias - End-of-life decisions reported not by physicians but by	Good

	• Aim: explore the preva-lence and nature of end-of-life decisions for people with disabilities. • Research question: - Is there a difference in end-of-life decisions and involvement between people with ID and people with other disabilities?	regions (response rate: 76.6%), 43 reported on people who died (n=82, 45 patients with ID, 37 patients without ID) • Descriptive statistics • ‘Other’ perspective: Management	eases (26.7%), respiratory diseases (15.6%), cancer (6.7%), and nervous system diseases (17.8%). 33.3% died due to other or unknown causes. • Residential homes for people with disabilities	not significant. - The decision to withhold treatment was made significantly more often for people with ID (28.9%) than for people without ID (8.1%). • Main conclusion by authors: In this study the preva-lence of end-of-life deci-sions (53.7%) is lower than in the first study (70.4%; Wicki & Hattich, 2016)	directors - Because of cross-sectional nature, no definite conclusions can be drawn - Only people living in residential homes included	

Use and content of ACP

No studies have been found that focused on the broad concept of ACP or studied an ACP programme within palliative care for people with ID. The most commonly studied element of ACP was decision-making, specifically end-of-life decisions (5 of the 14 studies), decisions about medical interventions (n=1), the place of palliative care (n=1), or decision-making in palliative care (n=1). Studies of decision-making show that in the Netherlands, one or more end-of-life decisions were taken for 57% of patients with ID [18]. In a comparable study in Switzerland, a percentage of 54% was found [27]. Decisions concerning the withholding of life-prolonging treatment are made more often for patients with ID than for patients with other disabilities [27, 28]. In end-of-life decisions regarding medical interventions, professionals and relatives believe quality of life and the prevention of suffering to be most important [14, 29].

Seven studies regarding decision-making reported on the involvement of patients with ID in the decision-making process. The wishes of patients with ID, especially of patients with severe to profound ID, are rarely if ever taken into account in decisions about medical interventions in the Netherlands [14, 18, 30]. Regarding decisions about the place of palliative care, 72% of professionals believe that the wishes of the patient with ID should be followed but only 8% stated that the patient's wishes were actually taken into account in the decision [31]. Wicki & Hättich (2016) showed that patients with ID were less involved in end-of-life decisions than patients with other disabilities. Nevertheless, the involvement of patients with ID was higher in their study than in other studies: they found that almost 70% of patients with ID were involved somewhat to very strongly in end-of-life decisions [27].

Five studies reported on organizational policies regarding ACP [24, 32-35]. Three of the five studies looked at 'do not resuscitate' (DNR) policies, in paediatric nursing homes for children with severe developmental disabilities and complex medical problems [33, 34] and in adult day services (ADS) [35]. The other two studies focused on policies regarding medical end-of-life decisions (ELDs) [24, 32]. The studies showed that no or only a few facilities for people with ID have policy documents on ACP. Moreover, there was often a lack of communication about these policies to professionals, family and/or residents [32].

Three studies reported on collaboration in palliative care [24, 26, 35]. People with ID were less likely than people without ID to have access to specialist palliative care services [26]. Moreover, hospice service providers had limited experience with people with ID [24, 35]. Other elements of ACP that studies reported on were: communication about advance directives (n=1), the inclusion of family in ACP (n=1), the documentation of wishes for future care (n=1) and the start of ACP (n=1).

Effects of ACP

None of the studies included in this review fitted the description of an effect study measuring the outcomes of an ACP programme or intervention. One study did report on the effects of ACP [26]. According to the case reviews in this study, ACP contributed to effective care for the patient's illness and conditions, and professionals acted in accordance with the plans that had been discussed. Moreover, professionals felt more confident after ACP discussions in their dealings with the patient.

Start of ACP and conducive and obstructing factors

One study looked at the start of ACP [26] and two studies reported on the start of decision-making [24, 30]. Obstructing factors were mentioned in three studies regarding different elements of ACP [24, 26, 35], and conducive factors were mentioned in two studies [25, 30]. Physicians preferred to discuss end-of-life decisions in a stable and calm situation, when emotions were under control and enough time could be spent on a sensible discussion [30]. However, this was often not possible because of a late diagnosis of the illness [24]. Professionals did not always recognize non-verbal symptoms or saw symptoms as part of the disability [24, 35]. Moreover, professionals were not prepared to discuss end-of-life issues with the patient because they believed the patient would not understand and they did not know who had the authority to tell the patient directly [24]. Therefore, if ACP occurred, it was often acute as a consequence of problems that had arisen, instead of anticipating possible problems that could appear in the future [26]. Regarding conducive factors in ACP, professionals noted the importance of nurturing good relationships with the patient’s family [25]. Physicians believed that a good working relationship with relatives and other professionals was the most important factor contributing to an effective decision-making process [30].

Conclusion

Our first research question focused on the use and content of ACP in palliative care for people with ID. Studies included in this review mostly focused on only one element within the broad concept of ACP, which embraces physical, psychological, social and spiritual matters in palliative care. Decision-making, specifically end-of- life decisions, and organizational policies regarding medical issues were the most commonly studied elements of ACP. Other elements that studies reported on were: collaboration in palliative care, communication about advance directives, the inclusion of family in ACP, the documentation of wishes for future care, and the start of ACP. Therefore, we can conclude that some elements of ACP have been studied, but more research is needed to investigate whether ACP should be used and what this process should look like within palliative care for people with ID. For example, it is still unclear when the process of ACP should be initiated, who should be involved, what the roles and tasks of the people involved should be, and what should be discussed.

The lack of reported evidence means we cannot answer our second research question about the effectiveness of ACP regarding the quality of palliative care and quality of life of people with ID. This is striking because the effects of ACP are well studied in various other patient groups for several types of interventions and programmes [11]. Furthermore, a review of ACP programmes in long-term care homes studied whether the programmes took the needs of patients with dementia into account [36]. For example, a well-studied ACP programme called 'Let Me Decide' [37] focusing on understanding treatment options in palliative care was considered dementia-friendly because it could be used for both patients with or without mental capacity, and patients and/or relatives were encouraged to re-evaluate their wishes and preferences when the patient’s health status changed [36]. Although dementia differs from ID in many respects, it would also be interesting to investigate the needs of people with ID regarding ACP and whether programmes like 'Let Me Decide' could effectively fulfil these needs.

The third research question focused on the start of ACP and conducive and obstructing factors in introducing ACP in palliative care for people with ID. Based on this review, there is no evidence that ACP discussions occur frequently. Where ACP is used, this is often in acute circumstances and only after problems have arisen [24, 26]. As studies included in this review indicate, there are no clear organizational policies on ACP, which can cause confusion and uncertainty among professionals, as well as among relatives, about tasks and responsibilities [24, 29]. Therefore, professionals should be informed about their role and tasks in discussing end-of-life issues and trained in talking about this with people with ID and/or relatives. In that way, professionals will be better prepared for discussing end-of-life issues with people with ID and/or their relatives and be more comfortable with this [15].

Advance planning for future care may be important not only for those who become incurably ill and are in need for palliative care, but also for those who are medically fragile from birth on [25, 33, 34]. In these patients, the need for ACP discussions and end-of-life decisions may manifest already from birth and will therefore be mainly influenced by the wishes of relatives. However, the need for ACP is not so easy to detect in people with ID with milder disabilities [26]. A recent study in the Netherlands showed that only 44% of ID physicians foresaw the death of patients with ID before the last month of life [38]. Multiple signals from different information sources and interactions between the patient, professionals and family are needed to be able to identify people with ID who are in need of palliative care [39]. This shows that ACP for people with ID is not a uniform process but will take different forms depending on the degree and complexity of the disability and vulnerability of the person with ID. Professionals should therefore use ACP as a flexible process depending on the needs and preferences of the individual and their relatives.

The studies included in this review showed that people with ID are often not involved in decision-making about medical issues. Moreover, no study included in this review used people with ID as participants in the study. As a consequence, not much is known about how people with ID in palliative care can be involved in ACP. Although including people with ID in healthcare research can be challenging [40], studies show that it is important to talk to people with ID about their illness and preferences in the palliative phase [41, 42]. This enables clarification about what people with ID find important and how people with ID see their own role in discussions about future care. The same applies to including people with ID in ACP discussions in practice. ACP focuses on person-centred care. Without asking the patients themselves about their views and preferences, it remains uncertain whether their wishes will be respected and whether the care provided will satisfy their needs [19, 41, 42]. However, allowances need to be made for the fact that people with ID find it more difficult to understand the concept of death, and self-determination in end-of-life planning is less developed [43].

A strength of this study is that it is the first to provide an overview of the use and content of ACP in palliative care for people with ID. Moreover, most of the studies included in this review are of a good methodological quality (12 out of 14). However, the majority of the studies were retrospective or made use of self-reported data and as a consequence were susceptible to recall bias. A limitation of the review is that we only included articles found in PubMed, PsycINFO, Embase, and CINAHL. We are aware that there could be more relevant studies that are not included in the databases we searched. Two additional studies were detected by manually searching for relevant references.

In conclusion, there are some indications that ACP in palliative care could be useful for people with ID, but more knowledge is needed about the use and effects of ACP as a broad process. Specifically, there is a lack of knowledge about the views and preferences of people with ID themselves about what is important in end-of-life care planning. As a consequence, it is not clear whether and how ACP should be used within palliative care for people with ID and how to involve their needs and preferences in this process. In future research, it is therefore important to investigate the perspective of the patients with ID and ask them about their experiences with and ideas about ACP. In healthcare practice, professionals should be trained in communicating with people with ID and/or their relatives about end-of-life issues.

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Appendix A Data extraction form

1. Study ID:
    - Reference: authors/date/title
    - Country
    - Aim and/or research questions

2. Study design and data collection:
    - Quantitative and/or qualitative
    - Total number of participants (n) and response rate
    - Type of analyses
    - Perspective of professionals, patients, relatives, others?

3. Background information of people with ID:
    - Age
    - Severity of disability
    - Medical condition/cause of death
    - Residence

4. Results:
    - Content of ACP
    - Answers to research questions
    - Main conclusion of authors

5. Strengths of study design and limitations

6. Methodological quality

Chapter 3

Advance care planning in the palliative phase of people with intellectual disabilities: analysis of medical files and interviews

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Abstract

Background: Advance care planning (ACP) is a process in which professionals, patients and their relatives discuss wishes and options for future care. ACP in the palliative phase reduces the chance that decisions have to be taken suddenly and can therefore improve the quality of life and death. The primary aim of this study is to explore how ACP takes place in cases of people with intellectual disabilities (ID).

Method: Medical files were analysed, and interviews were held in six care organisations for people with mild to severe ID. The data concerned people with ID (*n* = 30), 15 in the palliative phase, identified using the ‘surprise question’, and 15 who had died after an identifiable period of illness. Additional pre-structured telephone interviews were conducted with their relatives (*n* = 30) and professionals *(n* = 33).

Results: For half of the people with ID who had died, the first report in their file about palliative care (needs) was less than 1 month before their death. Professionals stated that ACP was started in response to the person's deteriorating health situation. A do-not-attempt-resuscitation order was recorded for nearly all people with ID (93%). A smaller group also had other agreements between professionals and relatives documented in their files, mainly about potentially life-sustaining treatments (43%) and/or hospitalisation admissions (47%). Relatives and professionals are satisfied with the mutual cooperation in ACP in the palliative phase. Cognitive and communication disabilities were most frequently mentioned by relatives and professionals as reasons for not involving people with ID in ACP.

Conclusions: Advance care planning in the palliative phase of people with ID focuses mainly on medical issues at the end of life. Specific challenges concern a proactive identification of changing needs, fear to initiate ACP discussions, documentation of ACP in medical files and the involvement of people with ID in ACP. It is recommended that relatives and professionals should be informed about the content of ACP and professionals should be trained in communicating in advance about wishes for future care.

Key words: advance care planning, decision-making, end of life, intellectual disabilities, medical files, palliative care

Introduction

People with intellectual disabilities (ID) have 2.5 times as many health problems as people without ID [1]. Respiratory disease and circulatory diseases are the main causes of death of people with ID [2]. The life expectancy of people with ID has increased over the past few decades, with a shift from sudden deaths and brief illnesses to more lengthy illness processes. As a result, an increasing number of people with ID require palliative care [3-6]. The World Health Organization defines palliative care 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".

It is known from previous research that health care professionals find it difficult to recognise a need for palliative care in people with ID [7]. This difficulty often stems from the fact that people with limited cognitive and communicative capabilities are not always able to clearly express their needs and wishes [4, 8-13]. Professionals who care for people with ID do not feel sufficiently trained in providing palliative care, and they often find it difficult to discuss vulnerability and future death [10, 14, 15].

The process in which professionals, patients and their relatives discuss wishes and options for future care is called advance care planning (ACP). Ongoing processes of ACP address all kinds of concerns across the physical, psychological, social and spiritual domains. Documentation of wishes and decisions is crucial to enable them to be taken into account in future decision-making [16]. Identifying wishes for future care at an early stage puts patients in more control of their own care and leads to more effective care [17-19].

ACP also reduces the risk that patients, relatives and professionals are faced with unforeseen situations and lets professionals act with greater self-confidence [4, 20]. ACP can be important at any stage in life but is particularly important in the palliative phase because it helps to ensure that patients receive treatment and care in accordance with their wishes and expectations at the end of life [4, 21].

To date, research about the use and content of ACP in the palliative phase has mainly concentrated on the general population [15, 17, 22, 23]. The aim of this study is to explore how ACP is performed for people with ID and how this is experienced by relatives and professionals. The key questions in this study are as follows: How does ACP take place in the palliative phase of people with ID and how is this perceived? The associated sub-questions are as follows: (1) What is the reason for starting ACP? (2) What agreements are documented and/or discussed in ACP? (3) Who is involved in ACP? (4) How are the wishes of people with ID taken into account in ACP?

Method

Procedure

This study consisted of (1) quantitative analysis of medical files of people with ID in the palliative phase and people with ID who recently died after an identifiable period of illness and (2) supplementary pre-structured interviews with care professionals and relatives of the people with ID whose files had been studied.

In the Netherlands, people with ID generally live with their family or independently with support, in a (small-scale) group home in the community or in a large residential setting. Data in this study were collected in six organisations in the Netherlands caring for people with mild to severe ID living in group homes with 24-h care or in residential settings. On a day-to-day basis, they are supported by care-staff, and they receive medical care from a physician employed by the organisation. Some of these physicians are medical ID specialists. Medical ID specialists receive 3 years of specialist training in the care for people with ID.

Physicians reviewed all their current patients and asked the surprise question to identify people with ID who were in the palliative phase during the study period of May 2016 to March 2017. The surprise question is as follows: ‘Would you be surprised if this patient died within a year?’ If the physician's answer to this question was negative, the person with ID was seen as in the palliative phase and therefore nominated for the study. The surprise question features in many European tools for identifying patients in the palliative phase [24]. Physicians also nominated all people with ID who had died no more than 6 months prior to the study period after an identifiable period of illness.

This procedure resulted in 117 nominated persons in the palliative phase and 47 recently deceased persons. The following background characteristics were recorded for each person: name of organisation, age, gender, cause of ID, severity of ID, living situation, life-threatening illness or cause of death, and relatives involved. A selection was made based on age, severity of ID, living situation, and life-threatening illness or cause of death. A spread in these variables was required to do justice to the diversity among people with ID, which was also expected to be related to how ACP is performed.

Eventually, 32 people with ID in the palliative phase and 34 deceased people with ID were selected for the study, regardless of the availability of relatives. Of this group, relatives of 15 people with ID in the palliative phase and relatives of 15 deceased approved to check medical files and agreed to be interviewed.

Physicians (*n* = 10) and care-staff (*n* = 23) of the people with ID were also interviewed. In the case of three people with ID, two professionals were interviewed because the first interview gave insufficient additional information.

Medical files

The extraction form for the files was developed by the authors of this article in collaboration with experts in palliative care for people with ID, based on practical experience and the existing literature on this subject (see Appendix A). The analysis of the medical files *(n* = 30) provided factual information about the documentation of agreements and communication regarding ACP. To determine the reliability of the data extracted, 19 files were studied by two authors independently (A. W. and H. V. or A. V. and H. V.). After it had been established that the extracted data were in agreement (100%), the remaining files were studied by one author (A. W., A. V. or H. V.).

Telephone interviews

The primary aim of these pre-structured interviews was to validate and refine the information about the use of ACP that had not been found in the files. An additional aim was to gain some information about experiences of relatives and professionals with ACP. The interview items were directly related to the items of the extraction form for the medical files.

The interview started with closed, factual questions (e.g. ‘Did you discuss care as the person's illness progressed?’), followed by an open question about their experiences (e.g. ‘How did you experience these discussions?’). These items were addressed in the interviews of both the relatives and the professionals concerning people with ID in the palliative phase or who had died, although the questions were adapted to fit the interviewee's situation. All interviews were conducted between January and October 2017 and were recorded. Interviews with relatives took an average of 15 min (range 7–31 min); interviews with professionals lasted 10 min on average (range 4–20 min).

Table 3.1 provides characteristics of the relatives and professionals who were interviewed. The largest group of relatives (43%) saw the person with ID at least once a week, 27% saw the person with ID once to twice a month and 30% saw him or her several times a year. Professionals had been working with people with ID for 14 years on average (range 2–31 years) and had been caring for the person in question for an average of 5 years (range 0.2–16).

Table 3.1 Interviewed relatives (*n* = 30) and professionals *(n* = 33)

	People with ID in the palliative phase (*n* = 15)	**Deceased people with ID (*n* = 15)**
Interviewed relatives	*n*	*n*
Parents	2	1
Brother or sister	7	8
Other family members	3	1
Non-family (legal representative/mentor)	3	5
Interviewed professionals	*n (%)*	*n (%)*
ID physician/General practitioner	4 (24%)*	6 (37%)*
Care-staff (social worker/nurse)	13 (76%)*	10 (63%)*

** n > 15; in the case of three people with intellectual disabilities (ID), two professionals were interviewed.*

Analysis

Descriptive analysis using STATA (version 14) was performed on the data from the medical files and the closed questions from the interviews. Open questions in the interviews were analysed as follows: The recorded interviews were listened to, and an extensive summary was made, also containing the actual quotations about the personal experiences with ACP. The quotations were independently coded by two authors (H. V. and A. V.), using keywords that were taken from the own words of the interviewees. The codes were compared by these two authors and clustered into significant themes. A selection of 10% of the interviews were replayed and analysed by another co-author (A. F. or A. W.) to validate the summaries and the identified themes.

Ethical approvals

The research protocol was assessed and approved by the Medical Ethical Committee of the University Medical Centre Amsterdam. Participation in the study was voluntary, and all relatives and professionals gave their informed consent. Research data were anonymised for analysis.

Results

Background characteristics of the people with intellectual disabilities

Table 3.2 provides background characteristics of the people with ID. Cause of the ID was unknown in the majority of cases (60%), 20% had Down's syndrome and the cause for the remaining 20% was another chromosomal abnormality, a metabolic disorder or another reason.

Table 3.2 Background characteristics of people with intellectual disabilities (ID)

	People with ID in the palliative phase (*n* = 15)	**Deceased people with ID (*n* = 15)**
Average age (years)	69 Min. 40 Max. 96	67 Min. 25 Max. 86
Sex, n		
Male	6	4
Female	9	11
Degree of ID, n		
Mild	4	4
Moderate	6	6
Severe	5	5
Place of care, n		
Residential setting	12	13
Community setting – 24-h supervision	3	2
Life-threatening illness, n		
General physical deterioration	6	5
Dementia	6	4
Malignancy	2	3
Other	1	3

Start of advance care planning

The medical files of 7 of the 30 people with ID contained no report of (recognition of) palliative care needs or delivery of palliative care, even though they had been selected by their physician as a person in the palliative phase or who had died. The other files of people with ID in the palliative phase (*n* = 11) or who had died (*n* = 12) did contain reports of palliative care. In half of the files of the 12 people with ID who had died, the first report of a need for palliative care was less than 1 month before death; in four cases, it was between 1 month and 6 months before death; and in two cases, it was more than 6 months before death (with a maximum of 4.5 years).

In interviews, professionals and relatives explained that they experienced difficulty in recognising care needs due to the communicational and cognitive disabilities of the person with ID; see Quote 1. In 19 of the 30 cases, professionals said that there was a clear start of palliative care marked by a substantial deterioration in the health of the person with ID or the diagnosis of a life-threatening illness. The need to start ACP was evident from the person's behaviour; see Quote 2. In the remaining cases, professionals said that there was no palliative phase (*n* = 2), were unsure whether there was a palliative phase (*n* = 2) or said that there was no clear lead-up and/or identification of the palliative phase (*n* = 7), for example, because it was a lengthy process of gradual deterioration. Professionals sometimes found it difficult to initiate discussions about wishes and options for future care, because they feared resistance from relatives to engage in such discussions and it was an emotional topic for them.

Quote 1: Brother of person with ID in the palliative phase (male, moderate ID, general physical deterioration, aged 71) “His pain threshold is very high, he never complains, so it is incredibly difficult to determine if something's wrong. He doesn't communicate anything. That makes it so difficult.”

Quote 2: Professional who cared for a person with ID (female, mild ID, dementia, aged 86) “She began to refuse her food: ‘I don't want any more, I'm tired’; that's when the turning point came. And then there were discussions, and agreements were made.”

Advance care planning agreements

Although palliative care needs or delivery of palliative care was not documented in all medical files, ACP agreements were recorded for 29 of the 30 people with ID. These were mainly medical decisions (Table 3.3). Six of the seven people with ID who had no record of palliative care according to their medical file, did have a do-not-attempt-resuscitation order. For two-thirds of the people with ID (*n* = 20), there were multiple reports in the medical files of discussions and/or agreements about future care. In the cases of 11 people with ID, agreements in the file were extended or modified because the person's situation became more fragile or, conversely, stabilised.

Table 3.3 Documentation of advance care planning agreements in medical files

	People with ID in the palliative phase (*n* = 15)	**Deceased people with ID (*n* = 15)**
Agreement about	*n*	*n*
DNAR	14	14
Diagnostic tests	3	1
Medical treatments	5	8
Hospitalisation admissions	7	7
Intensive care treatment	6	4
Antibiotics	1	4
Artificial nutrition or hydration	3	2
Place of care at the end of life	1	1

DNAR, do-not-attempt resuscitation; ID, intellectual disabilities.

While often not documented in the medical files, it transpired in the interviews that relatives and professionals also discussed issues concerning the (social) environment and daily activities and personal care. Occasionally professionals made use of an aid for discussing wishes and making agreements, such as a checklist with topics that are important in the palliative phase. This was found to be helpful for ACP. However, no clear organisational procedures were mentioned about how or when to have ACP discussions or make agreements.

The interviews showed that, because it was often unclear how the deterioration in the person with ID would progress, relatives frequently found it difficult to talk and take decisions about the future; see Quote 3. Professionals thought it was important to take time and give relatives a chance to get familiar with the idea that their loved one was deteriorating. As it became clearer as the illness progressed what was possible and what not, agreements could be made about future care. This brought peace in the final phase of life; see Quote 4.

Quote 3: Father of a person with ID in the palliative phase (male, moderate ID, progressive illness, aged 40) “No, we don't have discussions about that [ACP]. It would be different if we expected him to have to go to hospital next week. Then, of course, we'd be preparing for something that is definitely going to happen. But we don't discuss anything that is possibly going to happen in the future.”

Quote 4: Brother of a deceased person with ID (male, moderate ID, neurological condition, aged 66) “The nice thing for us as a family was really that as soon as he got the announcement that he was not going to be operated on, a kind of peace came over him. He never became angry again then or escalated things or whatever. He passed away very peacefully, actually.”

Involvement in advance care planning

The medical files indicated that 90% of people with ID for whom ACP agreements had been made (*n* = 29), there had been discussions between the physician and relatives about these agreements and/or that relatives agreed with decisions. In four cases, it was reported that the person with ID attended ACP discussions. In most files (*n* = 18) was reported that besides the physician, other professionals were involved in ACP, such as daily care-staff, behavioural experts and/or spiritual caregivers. Medical specialists at the hospital were involved in ACP in four cases. An internal supporting palliative team was involved in one case.

In the interviews, professionals acknowledged the importance of relatives' involvement; see Quote 5. Relatives said that they received sufficient, clear information about how the illness could be expected to progress and the pros and cons of possible decisions. They also perceived they were given sufficient room to express their own wishes for future care. However, some relatives felt it was unclear what role and responsibilities they had in decisions about future care and not all relatives were informed about their legal role as representative of the person. In most cases, there was agreement between relatives and professionals concerning views on the future care for the person with ID or relatives trusted professionals to make a good decision in the interests of the person with ID; see Quote 6.

Quote 5: Professional caring for a person with ID in the palliative phase (female, mild ID, general physical deterioration, aged 85) “I try to involve them [family] as much as possible so they know how she's doing. And about the deterioration, how it's progressing and what they [family] think about that.”

Quote 6: Aunt of a deceased person with ID (female, moderate ID, malignancy, aged 83) “I have to say that I'm very satisfied with how it went. We only disagreed when they [professionals] still wanted to arrange the examination – or at any rate wanted to consult a physician – and I didn't like the sound of it, but we talked about all of this very openly and clearly. I didn't feel I was being pressured either. And eventually I thought: they spend so much time with her, they'll be better able than me to estimate whether she can cope with this.”

Wishes of people with intellectual disabilities in advance care planning

In the interviews, professionals in 22 of the 30 cases said that it was not possible to involve the person with ID in ACP because the person did not have the intellectual capacity, could not oversee the situation and/or was too fearful to discuss the matter; see Quote 7. In eight cases, it was possible to involve the person with ID to some extent, according to the professionals, by informing them of the illness and/or deterioration in a simple manner adapted to their cognitive level and capabilities. The persons' own wishes about future care were mentioned in the medical file in only four out of 30 cases. The interviews also revealed that relatives and professionals did believe they had acted on the basis of the persons' wishes wherever possible. In half of the cases, relatives and professionals said that they had discussed with the person in question what they found important in their life. They looked at what the person with ID would want and at ways of taking account of their wishes; see Quote 8. Sometimes, this discussion was conducted with the help of a spiritual caregiver. In the case of people with ID who could not express themselves verbally, their wishes were deduced from their behaviour and agreements made on this basis. In this way, the person with ID remained in control.

Quote 7: Physician caring for a person with ID in the palliative phase (female, mild ID, general physical deterioration, aged 82) “Getting older .... I don't know what I should say to her about that. That she is getting older and she will eventually die? I think that would only make her anxious.”

Quote 8: Mentor of a deceased person with ID (female, mild ID, malignancy, aged 53) "…. And that was what I already said at her level, right. When I was talking to her about it or when the daily care-staff was, because of course they were much closer to her in fact than I was because I only come every now and then of course. But we've built that up, and when talking to people she really trusted she would say, listen, if I can't smoke any more cigarettes and can't drink any more coffee then I'm done. Then I don't really see the point any more. You can say that's oversimplifying things but if you put it in the context of that lady's life and what was important for her …. That was her life. And then I think you have to take that into account."

Discussion

This study shows that ACP mainly takes place in the final phase of the life of people with ID. Agreements documented in the medical files mainly focus on medical end-of-life decisions made by relatives and professionals. No palliative care needs were reported in the files of 20% of the deceased persons with ID, while such needs were only reported less than 1 month before death for a further 40% of the deceased persons with ID. Hence, identification of changing needs and adjusting the provision of care as needs change is not always done proactively but rather seems responsive to problems that arise. Moreover, the findings in our study are in accordance with previous research showing that professionals find the end of life a difficult and emotional topic, which could lead to postponing or avoiding conversations about future care [14, 21, 25, 26].

The interviews in this study revealed that wishes and options for future care were discussed with regard to the persons' social environment, activities and personal care, but documentation about discussions and/or agreements concerning psychosocial and spiritual aspects of palliative care were not found in the medical files. Earlier research also showed that there was little documentation about the process of decision-making and the roles of the people involved in the case of end-of-life decisions for people with ID [27].

In addition, the interviews indicated that relatives were often involved in ACP and that collaboration between professionals, relatives and – where possible – the person with ID often went well. Previous research showed that it is important for professionals to develop a bond with relatives and inform and involve them in the care of the person with ID [28]. Physicians attached considerable importance to relatives' opinions, and they often leave the assessment of a persons' quality of life to the relatives [29]. In the current study, some relatives felt that decisions about future care rested on their shoulders. Although the opinion of relatives can be very valuable and in some cases, the deciding factor, it is important that professionals inform relatives of the legal situation of representatives and the associated responsibilities.

This study showed that people with ID were often not involved directly in ACP. Relatives and professionals explained that they looked carefully at the person in question and from their perspective, thereby acting on behalf of the persons' interests. However, it is known from previous research that people with mild ID want to be informed about their health status, to be asked more about their care wishes and to be listened to [21, 30, 31]. In line with previous research, we also recommend that relatives and professionals consider ways of involving people with ID in ACP rather than assuming, possibly incorrectly, that it would be too burdensome for them to become involved [9, 11, 21, 27, 32, 33]. Further research that includes the perspective of people with ID themselves would be valuable to obtain more in-depth information about the wishes and needs for ACP.

Strengths and limitations

In this study, medical files of people with ID were examined rather than their daily care files. This means that reports by care-staff were not examined. However, in the participating organisations, the physician is responsible for the medical policy on palliative care, and if any agreements had been made, these should have been documented in the medical files. A second limitation concerns the retrospective character of part of this study. Professionals of deceased people with ID sometimes found it difficult to answer the questions because they could no longer recall the process of ACP. However, if this meant that an interviewee gave very limited information, other professionals were approached for an additional interview. A final limitation concerns a possible selection bias. Relatives who were satisfied with the care provided or who are more closely involved might be more likely to participate than relatives who were less satisfied or less involved in the care.

Also, the findings may not apply to individuals who live at home independently or with their family.

A strength of this study is that insights were obtained from multiple sources of information. Furthermore, the selection based on a spread in the background characteristics of people with ID meant that the diversity of the group was taken into account, giving as comprehensive a picture as possible of the use of ACP for people with ID.

Conclusions

This study shows that ACP in the palliative phase of people with ID is not often performed to discuss possible issues and wishes for care in the future but rather takes place in response to acute medical issues at the end of life. Despite the fact that relatives and professionals were involved with the person with ID and worked well together, professionals found it difficult to recognise palliative care needs, initiate ACP discussions, document wishes and agreements in advance and find ways to involve the person with ID. It is important that professionals learn to proactively identify and discuss changes in care needs with the person with ID and/or relatives. To achieve this, relatives and professionals should be informed about the essence of palliative care and ACP, and professionals should be trained in communication and documentation of wishes and agreements for future care. Furthermore, it is important to hear from people with ID themselves how they wish to be involved in ACP. In this way, palliative care can be provided in line with what the person wants and needs.

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Conflict of interest

The authors declare that there is no conflict of interest.

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Appendix A Extraction Form for File Investigations

General	Researcher name:	
	Date of file investigation:	
Client background	Client number:	Other relevant information?
	Age:	
	Sex:	
	Cause of intellectual disability:	
	Level of intellectual disability:	
	Current accommodation situation:	
	Medical reason for limited life expectancy:	
	- Dementia	
	- Malignancy	
	- CVA	
	- Heart failure	
	- COPD	
	- Metabolic disorder	
	- General physical deterioration	
	- Other	
	The relatives involved: yes/no?	
	Who?	
	Estimated life expectancy:	
	- >12 months	
	- ≤12 months	
	- Unknown	
	Deceased:	
	- Cause of death?	
	- Place of death?	
Palliative care	Clear marking of the palliative phase? (Professional expects client to live one more year at most/surprise question is answered in the negative)	Other relevant information?
	Yes/No	
	If yes, when did palliative care start?	

-continued Appendix A-

ACP agreements	Have ACP agreements been made?	Other relevant information?
	Yes/No	
	What has been agreed?	
	- Resuscitation policy/DNAR order?	
	In case of DNAR: date of order?	
	- Hospitalisation: yes/no?	
	- Intensive care: yes/no?	
	- Agreements on diagnostic tests: yes/no?	
	- Agreements on treatment: yes/no?	
	- Antibiotics: yes/no?	
	- Agreements on artificial nutrition or hydration: yes/no?	
	- Policy geared to comfort/dealing with symptoms: yes/no?	
	o Pain control	
	o Reducing anxiety	
	o Reducing unrest	
	o Other?	
	- Place of death discussed? Is this the wish of the client/relatives/supervisors?	
	Yes/No	
	o Client's wishes concerning ACP discussed?	
	Yes/No	
	If yes, have they been adhered to?	
	Yes/No	
	- Relative's wishes concerning ACP discussed?	
	Yes/No	
	If yes, have they been adhered to?	
	Yes/No	

Who was involved in ACP?	- Physician	Other relevant information?
	o ID physician	
	o General practitioner	
	o Basic physician	
	- Client	
	- Relative	
	o Parent	
	o Brother/sister	
	o Cousin/niece/nephew	
	o Friend	
	o Other	
	- Legal representative/Mentor (non-family)	
	- Daily care-staff	
	- Other care professional	
	- Other	
	o Who?	
	Were there factors that made communication difficult?	
	Yes/No	
	If so, what?	
Frequency?	How often was ACP discussed?	
	- Not applicable	
	- 1×	
	- ≥2×	
Changes to agreements?	Were changes made to the ACP agreements?	
	Yes/No	
	- What was changed?	
	- Why was this changed?	

Chapter 4

What is important for advance care planning in the palliative phase of people with intellectual disabilities? A multi-perspective interview study

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Abstract

Background: Advance care planning (ACP) is the process of discussing and documenting wishes and preferences for future care. Research about ACP for people with intellectual disabilities is limited. This study describes what is important for ACP in the palliative phase of people with intellectual disabilities.

Method: In-depth interviews were conducted with people with intellectual disabilities (n = 5), relatives (n = 7) and professional caregivers (n = 8). Qualitative data were analysed inductively, using the principles of thematic analysis.

Results: Important themes in ACP were as follows: tailoring care, working as a team and taking and giving time. The perceived role of people with intellectual disabilities in ACP was to express their wishes. Relatives had a signalling, representing and contributing role. Professionals felt their role was to inform, collaborate and coordinate.

Conclusions: A staff training programme about ACP should cover how to build and maintain close relationships, provide a safe environment and address ACP as an integral part of care.

Key words: advance care planning, decision-making, end of life, intellectual disability, palliative care, staff training

Introduction

People with intellectual disabilities are an ageing population and, in line with the general population, are living longer due to social and medical advances [1]. This is linked to growing prevalence of life-threatening illnesses at an older age, such as progressive cancer, chronic cardiovascular diseases, chronic lung diseases and dementia [2, 3]. Therefore, more attention is currently being given to how to support older people with intellectual disabilities who are in need of palliative care [4-7]. The World Health Organization defines palliative care 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.” [8].

Despite the growing attention paid to palliative care for people with intellectual disabilities, very little work has been done on the views of people with intellectual disabilities regarding their preferences for end-of-life care [9, 10]. To increase the quality of palliative care, it is important to identify the end-of-life wishes and preferences of people with intellectual disabilities [5, 7, 11, 12]. The process of discussing and documenting wishes and preferences is referred to as advance care planning (ACP). ACP is a person-centred, ongoing process of communication between the patient, relatives and professionals that facilitates patients' understanding, reflection on and discussion of goals, values and preferences for future care [13]. As ACP is a broad concept, medical decisions should be part of discussions, but ACP also focuses on psychosocial and spiritual matters in the palliative phase. Moreover, ACP takes place not just within the context of the physician–patient relationship but also within relationships with relatives and other professionals who are important for the support and experiences of the person with intellectual disabilities [7, 14, 15].

Research has demonstrated that people with mild or moderate intellectual disabilities are often able to express their preferences about end-of-life issues [5, 16-19]. Therefore, ACP has the potential to put people with intellectual disabilities more in control of their own care [5, 9]. Nevertheless, limited cognitive and communicative capabilities of people with intellectual disabilities mean that professionals experience difficulties in recognizing palliative care needs and initiating discussions about future care [15, 20-26]. Moreover, the presence of co-morbid medical conditions in this population and complex social circumstances adds complexity to the ACP process [27, 28]. The challenges of communicating and interpreting the future care needs of people with intellectual disabilities mean that care is not always adjusted proactively; rather, adjustments tend to be made in response to problems that have arisen [2, 5]. Therefore, people with intellectual disabilities are at risk of having their wishes overlooked or misunderstood at the end of life [7, 29].

If palliative care is to be in line with what individuals with intellectual disabilities want and need, it is important to study what is needed to implement ACP in the palliative phase. Previous research has given useful information on how to provide tailored palliative care for people with intellectual disabilities [30]. Moreover, incorporating the views of people with intellectual disabilities themselves about end-of-life care is critical to working out how best to support them [31]. However, to our knowledge, there has not yet been an empirical exploration of what is important for ACP from multiple perspectives, namely the perspectives of people with intellectual disabilities themselves and their relatives and professionals. Therefore, two main research questions were addressed in this multi-perspective interview study: (a) 'What should ACP look like?' and (b) 'What roles should people with intellectual disabilities, relatives and professionals have in ACP?' The results of this study will be used for the development of a staff training programme about ACP for people with intellectual disabilities.

Method

Design

A qualitative design was used, involving individual in-depth interviews with a total of eight professionals, seven relatives and five people with moderate intellectual disabilities.

Ethical consideration

The protocol and topic list for the interviews were approved by the Medical Ethical Committee of the VU University Medical Centre. Study participation was voluntary. The responses were anonymous and non-traceable to individuals.

Participants

Professionals (n = 8) and relatives (n = 7) who were close to a person with intellectual disabilities in the palliative phase or to an individual with intellectual disabilities who had recently died after a period of illness, were identified by physicians who were employed at five intellectual disability residential care organizations in the Netherlands. Via the physicians who functioned as recruiters in our study, potential participants received a letter from the relevant intellectual disability care organization informing them about the aim and content of the study and asking them to sign an informed consent form if they wished to participate. Other than having prior experience with caring for a person with intellectual disabilities at the end of life, there were no inclusion criteria for the professionals or relatives.

People with intellectual disabilities (n = 5) were recruited from one of the intellectual disability residential care organizations by their intellectual disability physician in consultation with care staff. Potential participants with intellectual disabilities were informed about the subject of the interview, and information was provided verbally; they could then verbally provide informed consent. The information was provided by their care staff, who also emphasized that participation was voluntary. The inclusion criteria for people with intellectual disabilities were as follows: (a) having an understanding of the concept of death and dying; (b) having previous experience with being seriously ill and/or being hospitalized themselves; and (c) being at least 18 years old. We asked care staff to assess these criteria in their clients. The exclusion criterion was (for ethical reasons) having a life- limiting illness or receiving palliative care themselves. However, most participants had a vulnerable health situation due to their age (three of the five participants were over 80 years old). Table 4.1 shows the background characteristics of the participants.

Data collection

A topic list was used to guide the interviews (see Box 4.1). The topic list was developed together with all the co-authors, who are researchers and practitioners working in the field of either family medicine or palliative care. Open questions were used, encouraging interviewees to describe personal wishes for and views about ACP in their own words.

Box 4.1 Topic list for in-depth interviews (n=15)

- Importance of ACP*

Why is it important to talk about ACP?

- Start of ACP

When should ACP be discussed?

- Content of ACP

What should be discussed in ACP?

- Involvement and roles in ACP

Who should be involved in ACP?

- Facilitators of ACP

What could help discussion of ACP?

- Barriers to ACP

What could be disadvantageous for ACP?

*ACP was explained as a process of discussing and recording wishes and preferences for future care.

For participants with intellectual disabilities, the number and sort of questions were adjusted to suit their cognitive and communicative abilities. In interviews with people with intellectual disabilities, the subject of the interview was introduced with a short picture story about a woman with intellectual disabilities who heard from her doctor that she was incurably ill. In two interviews with people with intellectual disabilities, the introduction story was skipped because the participants had been prepared for the interview by care staff and shared their ideas about ACP as soon as the interviewer arrived. Interviews were held at a place preferred by the interviewees and were conducted by the first and/or second author (HV and AV). All participants with intellectual disabilities were familiar with the second author, who is involved in their care as a specialized intellectual disability physician. Aftercare was discussed with participants' care staff to ensure that support was available if needed. The interviews with professionals and relatives lasted 1 hour. The interviews with people with intellectual disabilities lasted approximately 15-30 minutes. The interviews were audio- or video-taped and transcribed verbatim.

Data collection and analysis took the form of a cyclical process of data collection —analysis —more data collection. The number of interviews in this study was not pre-determined but was chosen on the basis of the principle of data saturation [32]. After fifteen interviews, no new information or themes were observed and data saturation was reached.

Table 4.1 Background characteristics of participants

	Interview	Participants	
People with intellectual disability			*Sex, age; degree of ID*
	1.	(1) J.	Male; 71 years old; moderate ID
	2.	(2) L.	Female; 83 years old; moderate ID
	3.	(3) T.	Female; 83 years old; moderate ID
	4.	(4) N.	Female; 81 years old; moderate ID
	5.	(5) R.	Male; 40 years old; moderate ID
Relatives			*Initial of their relative with ID: sex; age; illness or cause of death; degree of ID*
	6.	(1) Mother	P: male; 47 years old; malignancy; severe ID
		(2) Father	
	7.	(3) Sister	T.: female; 62 years old at death ; died in 2017 from frailty in old age; moderate ID
	8.	(4) Sister	G.: female; 61 years old; dementia; moderate ID
	9.	(5) Mother	G.: male; 20 years old; co-morbid somatic illness; profound ID
		(6) Father	
	10.	(7) Mother	J.: male; 14 years old; co-morbid somatic illness; severe ID

- Table 4.1 continues -

- Table 4.1 continued -

	Interview	**Participants**	
Professionals			*Work experience as a professional in ID care*
	11.	(1) Intellectual disability physician	30 years
	12.	(2) Nurse specialized in palliative care	16 years
		(3) Nurse specialzed in palliative care	41 years
	13.	(4) Nurse specialzed in care for people with ID	30 years
	14.	(5) Behavioural specialist	8 years
		(6) Behavioural specialist	30 years
	15.	(7) Nurse	22 years
		(8) Social worker	10 years

Data analysis

The data were analysed inductively, using principles of thematic analysis [33]. Thematic analysis can be considered a pragmatic, realist qualitative method, aimed at reporting experiences, meanings and the reality of participants. The analysis process is stepwise. It starts with noticing and looking for issues of potential interest in the data, which begins during data collection.

As a first step, interviews were reread, important text fragments regarding the research questions were noted, and ideas were marked for coding by two authors (HV and AdV). A qualitative software analysis programme was used to facilitate the coding process (MAXQDA). Descriptive codes were used, based directly on words or sentences that the interviewees used (e.g. 'Stronger than I am able to be'), as well as interpretative codes (e.g. 'Deciding together'). Interview fragments with the same codes were continuously compared and discussed by HV and AdV. In the next step, codes were sorted into potential themes. Potential main themes and corresponding subthemes were then formulated. Themes and subthemes were also discussed with the other co-authors (AV, AW, AC, JM and AF), who individually reviewed the themes and subthemes by reading and analysing at least one interview. After slight adjustments in formulations of themes and subthemes and their interpretations, consensus was reached with all authors about the themes. In the next step, the themes were reviewed in relation to the entire data set and refined in a discussion. Subsequently, the researchers further identified the essence of the themes and determined what each theme was about and what aspect of the data it captures. As a next step, definitive themes and subthemes were formulated in consultation with all authors. Finally, all co-authors as well as peer researchers reflected on the draft paper and the main themes presented (the 'peer debriefing'). In the draft and final papers, pseudonyms were given to participants to maintain anonymity.

Results

Thematic analysis of the interviews (n = 15) resulted in the identification of three themes and six subthemes, addressing research question (a) 'What should the ACP process look like?' (see Figure 4.1). A description of the roles is depicted for research question (b) 'What roles should people with intellectual disabilities, their relatives and professionals have in ACP?' (see Figure 4.2). The results are described in detail below.

Figure 4.1 Representation of themes indicating what people with ID, their relatives and professionals perceived as important for ACP in relation to the research question: "What should the ACP process look like?"

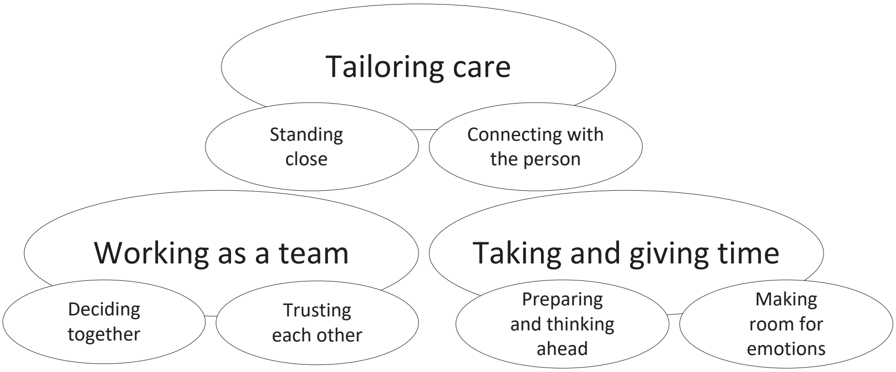
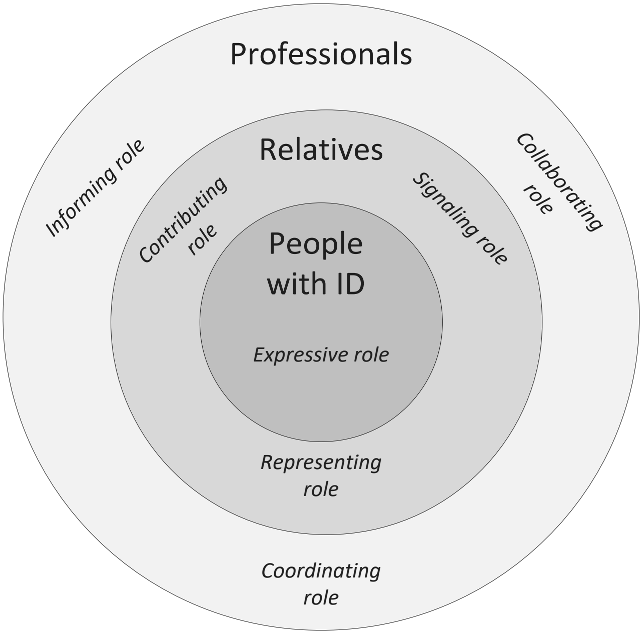


Figure 4.2 Representation of themes indicating what people with ID, their relatives and professionals perceived as important for ACP in relation to the research question "What roles should people with ID, their relatives, and professionals have in ACP?"



What should the ACP process like?

Tailoring care

Advance care planning is perceived as a tailor-made process. Each individual is different and has various care needs that needed to be taken into account. According to the participants, there is no single right way to provide ACP. Two subthemes emerged in relation to what is needed to provide tailored care.

Standing close

Good knowledge of the person with intellectual disabilities was felt to be necessary in order to shape the ACP process to suit the individual with intellectual disabilities and their relatives. For example, to involve the person with intellectual disabilities in the ACP process appropriately, information is needed about that person's life history and previous experiences with illness and losses.

Participants emphasized the importance of close relationships with everyone directly involved in the care for the person with intellectual disabilities, in which open communication can take place.

One parent clarified:

> No, but they just know him. …He is also placed in care with a fixed group. He is lucky with that, because these people have all known each other for like 30 years. And well, yes, that is a very good team. Yes, they just do that [communicate] very well, with only half a word you know, you know what it is. Yes, they are, it is a very good place for him.— Interview 6, relative (2)

One of the participants with intellectual disabilities also spoke about the need for familiar people caring for and about her:

> Because a stranger… I can’t handle people I don’t know. Like here, these care professionals, and this girl who works with us now, she is a student. I need to see who it is. Because I really can’t deal with them otherwise. I immediately get in a conflict [with them].— Interview 3, participant with intellectual disabilities (3)

Connecting with the person with intellectual disabilities

To be able to truly simulate what the person with intellectual disabilities wishes, it is import to set aside own preferences and to model what that person stands for in their life, and adjust care according to their goals and needs. One professional elucidated the need to connect to the individual in order to be able to reflect the wishes of the person with intellectual disabilities:

> It is of course essential to act and think in the interest of the client [person with ID]… that should be a requirement. … It is important to know who you are talking about, [especially] if people cannot talk about it any more [themselves]. In what other way could you make a decision? Then the basis on which you make decisions would be very medical or mechanically.— Interview 14, professional (5)

Additionally, professionals found it important to adapt the ACP process to suit the level of understanding and experiences of the person with intellectual disabilities. One professional explained how she responded to fears from an individual with intellectual disabilities about what happens after death:

> For example, there was a client [with ID] who wanted to take her television with her when going to heaven, [that was] very important [for her]. Or her electric wheelchair needed to go with her because how else could she move there?… It was very important for her; 'what is heaven like?'. You [as a professional] have to deal with that very seriously, like; 'Yes, of course it can come to heaven with you', or; 'No that is not possible, but perhaps they have a very large cinema [in heaven]. Or they have televisions and you do not have to bring your own.' So these things are important for clients.— Interview 12, professional (2)

Working as a team

The ACP process requires teamwork. Participants described various people involved in the care for the person with intellectual disabilities as participating in ACP discussions and decisions. Two subthemes were identified.

Deciding together

According to participants, it is important that everyone who is involved in the care for persons with intellectual disabilities who need palliative care should work together to inform one another and identify whether things are going well or not. Participants with intellectual disabilities said that their relatives should be informed about their health situation, because: ‘Then they [relatives] know what is wrong with me’— Interview 2, participant with intellectual disabilities (2), and ‘Otherwise they [relatives] would worry— Interview 5, participant with intellectual disabilities (5)

In decision making about future care, different beliefs played a role. The participants felt it was important to discuss with everyone who is involved how best to act in the interests of an individual with intellectual disabilities. In this way, decisions can be made that improve the quality of life, and care can be adjusted to changing needs. Moreover, for relatives as well as for professionals, there is a need for consensus on the decisions for future care. One parent clarified why he felt it was important to have care staff involved in discussions about the future care for his son with intellectual disabilities:

> But in all these [ACP] discussions, there is also just, his direct caregivers are there. We wouldn't want it without them being there, because, yes, they are there every day, they just need to deal with it. And they [daily care professionals] also needed to know what we [parents] thought.—Interview 6, relative (2)

Trusting each other

All participants indicated that trust was essential in discussing insecurities and fears, and in making decisions for future care. Relatives explained that it is important to be able to rely on the expertise of professionals in providing and adjusting care according to the needs of the individual. A sister explained:

> .. if a decision needed to be taken, they would let me know and asked if it was fine. Well yes, I thought they [professionals] will know best how to arrange it. … We [as a family] just knew, they are caring for T. and for the other residents every day. So they will know better than us what to do.—Interview 7, relative (3)

Professionals explained that honesty and openness within a team of people caring for and about a person with intellectual disabilities contributed to a more positive and calmer ACP process.

Moreover, the professional and personal experience of professionals with palliative care and losses plays a role in their ability to openly discuss ACP. One professional said that it takes time to feel comfortable taking care of people who need palliative care. Her experience is that it is helpful to be honest about this, to be able to share this with the team and to feel free to ask for help if needed:

> I notice with us, also within the team, it is very discussable. We don't avoid that subject at all and that is pleasant, because I have worked in a team where it was harder. That was with children and then you sometimes have colleagues who can't deal with that for themselves… they really have something like: 'I don't want to provide end-of-life care and I think that's scary'. Here as well, I also have colleagues here who are younger, who have not yet had clients who have died. The first time is very difficult, [not just as a professional,] but also as a person. Here [in our current team], I notice that it is discussable. — Interview 15, professional (7)

Taking and giving time

ACP is characterized by the participants as a dynamic, continuous process with ongoing discussions. Proactive care and making decisions about future care required time and attention. Two important subthemes are as follows: (a) preparing and thinking ahead and (b) making room for emotions.

Preparing and thinking ahead

Professionals said that in some cases the ACP process had a noticeable beginning, for example because of a diagnosis of a life-limiting illness, when it is clear for everyone involved that wishes and treatment options for future care need to be discussed.

However, for most people with intellectual disabilities, ACP was experienced as a slow process, in which time is needed and should be used to adjust to the changing situation and decisions should be made step by step, with ongoing adjustment for the current situation. A professional described her experience with adjusting ACP decisions in the palliative phase:

> Yes, and decisions can also be reversed because we had that once with a client [person with ID], that a decision [to not force her to drink] was made because she was asphyxiating more and more, with every drop [she drank], she asphyxiated. And that was really when we thought the end had come and then she, actually after more than a week, she started drinking again. Then we changed that again too, because you could notice that she had the need to, she wanted that again. It is a process: adjusting, tuning, resetting and then thinking ahead again.— Interview 12, professional (2)

Some participating professionals and relatives described that they experienced talking with or about someone with intellectual disabilities at the end of life to be difficult for them as well as for the individual with intellectual disabilities. One participant with intellectual disabilities (interview 1) answered 'I would be shocked' in relation to the question what he would do if the doctor disclosed unfavourable information about his health. All participants with intellectual disabilities knew that if they had a disease that could not be cured, they would die. However, professionals indicated that people with intellectual disabilities often found it scary to think about the consequences of their situation. In response to 'dying', one participant with intellectual disabilities said:

> I do not think about that yet. That will take a long time.— Interview 2, participant with intellectual disabilities (2)

Acceptance of decline was described by professionals as a process that requires time. It was therefore felt to be important that professionals proactively inform everyone who is involved about the current situation and are realistic when discussing ACP, for example, about treatment options for future care. In this way, the person with intellectual disabilities and their relatives could be better prepared and have a better understanding of what can be expected in the future.

ACP was also found to be helpful for professionals so that they know how to best support an individual in changing situations. A professional explained that it might be hard to start discussions in advance, but it helps to ensure peace in the final phase of life, which is important for a good farewell:

> It is just hard when it [the need for ACP] is spoken out loud. And if you can substantiate it very well, then the family understands. Then they see, then they say, ‘actually yes, you are telling now what I actually already have seen’. And then you also see that the family is very happy with that afterwards. That everything is just addressed in a timely manner. That not everything needs to be arranged at the last minute, that a lot of things are just fixed, that they really calmly, can be present in the phase of dying. Without having all stuff on your mind like: ‘this needs to be done, that needs to be done’. That is not necessary anymore.— Interview 12, professional (2)

Making room for emotions

Having discussions about future care for a person with intellectual disabilities and making decisions for or together with that person can have a big impact and can be accompanied by strong emotions according to participants. Therefore, relatives and professionals recommended taking the impact that ACP in the palliative phase can have into consideration. In relation to personal experiences with discussions about future care for her son with intellectual disabilities, one mother notes the importance of approaching ACP gently and sensitively, with attention to personal and emotional support if needed:

> … But always pay attention to the feelings. You can bring it up very technically, but don't just keep it there. Also ask what they think is important for their child [with ID]. And don't just say: ‘I decide’.— Interview 10, relative (7)

She also talked about the difficulty she experienced signing a do-not-resuscitate order for her son with intellectual disabilities, knowing that such decisions can have far-reaching consequences:

> …as if you are Lord and God over someone’s life. Something like that. You have to know with every fibre [of your being] that you do it for them, that you do something for him, that I do something for him what I [would] do for myself also [in the same situation]. That is, that was very important to me, to get that feeling very clear. So it took a while, before I put my signature [on the do-not-resuscitate order]… I could talk considerably to the doctor about it when she called, like ‘Did you already send that back? Because I have nothing yet.’ Then we also talked about it. I mean, she also had the time for that. In the end it remains my signature, but it is nice if you have someone on the other side who also has time to talk about something like this.— Interview 10, relative (7)

One social worker (interview 15) explained ACP should be considered as a ‘human approach’ rather than a more technical or procedural approach. Moreover, one intellectual disability physician described that there should be room to open up about feelings that arise, including as a professional:

> I think it would beneficial to also name your own feelings, you are allowed to have feelings too [as a professional]. It might be better if you name them [your own feelings]. Without any consequences, but just that it, well it is also difficult for you as a physician. Maybe I have been too distant in the past too … Yes, so when I look back then I think it is important to be able to talk about emotions.— Interview 11, professional (1)

What roles should people with intellectual disabilities, their relatives and professionals have in ACP?

Roles of people with intellectual disabilities in ACP.
The interviews also addressed the roles of the individuals with intellectual disabilities in their own ACP process. Participants with intellectual disabilities wanted to be informed about their health situation because, as one participant explained simply:

> If I know what's wrong with me, then I can do something about it.— Interview 5, participant with intellectual disabilities (5)

According to the participants, people with intellectual disabilities have an expressive role in ACP. Based on their own previous experiences and wishes, people with intellectual disabilities can communicate their preference, sometimes verbally but more often in non-conventional ways. When asked by the interviewer why she wanted to be involved in ACP, one participant with intellectual disabilities replied:

> It is my life!— Interview 4, participant with intellectual disabilities (4)

However, people with intellectual disabilities often find direct participation in decisions about future care difficult. One participant with intellectual disabilities (P) told the interviewer (I) that she wanted the people close to her to make decisions regarding future care on her behalf:

I: And if decisions need to be taken?
P: They [doctor, staff, relatives] need to do that too.
I: Why all those people together?
P: They are stronger than I am able to be.
I: Do you think it's hard [to make decisions]?
P: Yes.

— Interview 3, participant wih intellectual disabilities (3)

Roles of relatives in ACP
According to professionals, in cases where relatives have a lifelong, close relationship with the person with intellectual disabilities, the relatives can have an

important role in interpreting expressions of the person with intellectual disability. Therefore, relatives may have a signalling role, since they can recognize dissimilar behaviour. Also, relatives often want to and should have a contributing role in ACP discussions about future care and decision making, depending on their own involvement and desire to be involved in the care for the individual with intellectual disabilities, as a professional explained:

> Well, it also depends in what way relatives themselves fulfil that role. Then of course, that is an interaction. Sometimes relatives are very closely involved, they want something very strongly. Then, yes, I explicitly take that into account. There are still also relatives, not that much now anymore, but they say: 'you decide, what you decide is fine'. And yes, many people are also in between, they want to think together, but also realize that their scope is limited.— Interview 11, professional (1)

Moreover, relatives have a representing role, as they can participate in ACP as a representative of the person with intellectual disabilities and articulate wishes on their behalf if the individual is not able to do so themselves. In the interviews, relatives expressed their wish to have a say in how best to act in the interest of a person with intellectual disabilities. One parent explained his personal involvement and sense of responsibility about his son with intellectual disabilities:

> That is always nice for us like, 'well, he feels good'. And we are doing well as parents; we arranged it [care] well for him.— Interview 9, relative (6)

Roles of professionals in ACP

According to relatives and people with intellectual disabilities, professionals should first of all have an informing role. Professionals should clearly and realistically explain possible future scenarios for care and the treatment options to relatives and/or the person with intellectual disabilities, taking the latter's level of cognitive functioning into account. One parent responded to the question of why it was important to receive information from professionals:

> Yes, because we wanted to know how his disease would progress and that he wouldn't… wouldn't suffer a lot.— Interview 6, relative (6).

Secondly, professionals should have a collaborating role. Since ACP requires teamwork, professionals should maintain harmonious relationships and provide a safe environment where all involved are able to openly discuss wishes and options for future care. In the interviews, professionals explained that it is important to take the initiative to discuss wishes and options for future care with relatives and/or the person with intellectual disabilities. One parent described how professionals should start ACP discussions gently:

> It is very important to keep discussing the situation continuously. But you [as a professional] should also get a sense of the parents [of people with ID]. Some are really not ready yet to think about the future and death. But you can bring it up very openly. And then parents can always say ‘no’.— Interview 10, relative (7)

Lastly, professionals should have a coordinating role as they explained that they monitor the changing behaviour of the person with intellectual disabilities; in the end, they are and feel responsible for providing care in line with what the person with intellectual disabilities wants and needs. Professionals should therefore explore and collate the views and wishes of everybody who is involved in the care for a person with intellectual disabilities, as one behavioural specialist explained:

> Well, what do we see? Then it's really our role. What do we see as the quality of life? What do we feel is important to a client in their life? You can make a good guess. Yes, some people find it so difficult to give up any of that being in control and independence, right? So you expect that when it gets to them having a feeding tube, for example, they would definitely not be happy about that. You know. And we look at it together like that, you know, what kind of person do we think the client is? Definitely if the level is really low. And what are the thought processes for getting it right here? We really try to do that together, as a multidisciplinary group, preferably actually with relatives or the representative.— Interview 14, professional

(6)

Discussion

This study shows that tailoring care, working together and taking and giving time are important themes in the ACP process. To be able to provide ACP in the palliative phase in accordance with the wishes and needs of the individual, it is essential to have close and harmonious relationships between individuals with intellectual disabilities, their relatives and professionals. This may be especially important in the palliative phase, where the proximity of death often leads to increasing dependency of the person with intellectual disabilities and requires an intensified caring relationship between the individual with intellectual disabilities, their relatives and professionals [34]. Earlier research also showed that if relational closeness is enhanced, responsiveness is likely to be increased [7]. Moreover, evidence from studies of surrogate or supported decision making suggest that the ability to ‘simulate’ another person's wishes is linked to the ability to empathize with that person [35, 36].

In addition, this study showed that ACP in the palliative phase requires significant teamwork, and teamwork is essential in establishing honest, realistic and open communication about possibilities for future care and in making decisions together. Trust was mentioned by the participants in this study as a key factor in being able to work together constructively. This finding is consistent with previous research on palliative care stating that ‘good working relationships’ and ‘openness to cooperation and sharing’ were important when aiming for the shared goal of providing the best possible warm, comforting care for the person with intellectual disabilities in the palliative phase [34, 37, 38]. Open, continuous communication between the individual with intellectual disabilities, their relatives and professionals should be a hallmark of ACP programmes.

Participants with moderate intellectual disabilities highlighted their wish to be informed about their health situation and to have familiar people supporting them in their decision making. However, some participating professionals and relatives said that they found talking with or about someone with intellectual disabilities at the end of life to be difficult for them as well as for the individual with intellectual disabilities. This fear could be a barrier for ACP if it leads to the postponement or avoidance of discussions about needs for future care and the impending death [20, 26, 39, 40]. A model for breaking bad news to people with intellectual disabilities was developed in previous research; it describes how people with intellectual disabilities process and understand unfavourable information, and how professionals can use this when they are faced with bad news situations [41]. Moreover, training must be developed in partnership with staff as that increases the likelihood that it will continue to be used in the long term [12, 42].

The present study also explored the various roles of people with intellectual disabilities, their relatives and professionals in ACP. Participants with intellectual disabilities were considered to have and expressive role, as they communicate their wishes and preferences in their own way, often represented by their relatives. Relatives explained they have an important signalling role in interpreting the person with intellectual disability expressions and contributed to decision making for future care. Professionals saw their role as coordinating ACP, as in the end, they are responsible for providing care in line with what the person with intellectual disabilities wants and needs.

According to the participating professionals, experience is required to become comfortable and confident in leading ACP discussions. Therefore, it is recommended that professionals receive training in providing ACP as an integral part of care, and in how to do so in a timely manner. Professionals should take the initiative to start discussions about ACP, which should preferably be held before acute decisions need to be made in the last phase of life. This is in line with an earlier study about ACP for people with intellectual disabilities that recommended starting discussions about future care 'as early as possible' and also suggested investigating the ACP process while people with intellectual disabilities are still feeling well [5].

Although the themes concerning ACP in the palliative phase that were found in this study may also be relevant for other patient groups or care settings, several aspects appear particularly important for people with intellectual disabilities and their relatives. Firstly, people with intellectual disabilities have lifelong disabilities and are often dependent on others throughout their lives. Various specialized professionals have an extensive caring relationship with the individual with intellectual disabilities and their relatives, and professionals are often involved in their care for years or even decades, which is unique in the healthcare sector. Conversely, the close involvement of numerous professionals and relatives can make ACP in the palliative phase challenging, especially in the case of conflicting beliefs and interests [29, 34]. Secondly, people with intellectual disabilities are not always able to clearly communicate their needs or understand the consequences of their situation. Professionals and relatives frequently need to make medical and other decisions on behalf of the person with intellectual disabilities but rely on interpretations of their signals and expressions, which can be a heavy responsibility

[7, 38, 43]. Thirdly, ACP in the palliative phase may be complex since people with intellectual disabilities often have co-morbidities and are more vulnerable concerning their health [2]. Among them, there is a group of people with intellectual disabilities who need palliative care from birth on [44]. Therefore, it is even more important to talk about the future at an early stage.

Strengths and limitations

The inclusion of multiple perspectives, including first-person information provided by people with intellectual disabilities, is a strength of this study, capturing a range of thoughts and opinions about the important aspects of ACP. The views of people with intellectual disabilities were generally in agreement with the views of relatives and professionals, which further attests to the validity of the findings.

One limitation that should be noted is that the study participants (n = 20) were recruited via intellectual disability care organizations. As a result, the findings may not apply to individuals with milder disabilities who receive care outside the intellectual disability care sector. Moreover, all the participants with moderate intellectual disabilities lived in one residential care organization and were familiar with the second author, who is involved in their care as a specialized intellectual disability physician. This could possibly lead to a selection bias. However, having a familiar professional conducting the interview or being in the same room helped to provide a safe environment for the participants with intellectual disabilities.

Future research could focus on the extent to which the important aspects of ACP found in this study match the views and roles of people with milder intellectual disabilities who live independently and/or receive non-specialized care. While their ACP process might look different, the need for the support and involvement of professionals as well as relatives will conceivably lead to comparable challenges.

Conclusion

People with intellectual disabilities often have numerous professionals and relatives involved in their care, limited cognitive and communicative capabilities and high levels of co-morbidity, which presents challenges for the provision of ACP. Important ACP themes in the palliative phase for people with intellectual disabilities found in this study concern tailoring care to adjust the ACP process to the needs and wishes of the individual, working together as a team so that open communication can take place, and taking and giving time to prepare for future scenarios. Hence, based on these findings, an ACP training programme might cover such aspects as building and maintaining close relationships, providing a safe environment and addressing ACP as an integral part of care. People with intellectual disabilities must have an explicit role in ACP by expressing their wishes, which makes it important for all people involved in their care to learn how best to respond to these expressions. Professionals have a coordinating role in this process, which requires experience and training. Therefore, it is recommended that professionals receive training in how to provide ACP as an integral, continuous process in the care for people with intellectual disabilities.

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Chapter 5

Implementation and sustainment of palliative care innovations within organizations for people with intellectual disabilities: a multi-method evaluation

Reference:
Voss, H., Francke, A. L., de Veer, A. J. E. Implementation and sustainment of palliative care innovations within organizations for people with intellectual disabilities: a multimethod evaluation. (under review)

Abstract

Background: Providing palliative care for people with intellectual disabilities (ID) is challenging and professionals caring for people with ID often received limited training in palliative care.

Objective: To gain insight into the implementation and sustainment of palliative care innovations, originally developed for the general palliative care population, in organizations for people with ID.

Methods: A multi-method evaluation was performed of nine implementation projects concerning three palliative care innovations. Methods included document analyses of project proposals and reports, group interviews with project managers and professionals, and a questionnaire completed by projects managers. Factors influencing implementation were categorized using the Consolidated Framework for Implementation Research.

Results: The three innovations were applicable in organizations for people with ID, although some adaptations had been made. Implementation activities were focussed on training, cooperation and dissemination of the innovation. Influencing factors were mostly related to the inner setting of the organization, including management support and available resources. Five of the nine project managers were not sure if the innovation was sustained properly within their organization.

Conclusions: Innovations originally developed for use in the general palliative care population can be successfully implemented in organizations for people with ID, although adaptation to the specific care setting might be necessary.

Key words: intellectual disability, palliative care, innovations, implementation, sustainment

Introduction

The life expectancy of people with intellectual disabilities (ID) is increasing [1-3]. As a result, people with ID are increasingly experiencing life-threatening conditions that are particularly likely to affect older people, such as chronic cardiovascular diseases, chronic lung diseases, cancer and dementia [3]. The World Health Organization [4] defines palliative care 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”.

According to the Dutch Palliative Care Quality Framework[5], palliative care should be provided for “Patients and their families facing the problems associated with a life-threatening illness or *vulnerability*”. People with ID are vulnerable in many ways (both physically and psychosocially) and often have to deal with multiple and complex health problems throughout their lives. People with profound intellectual disabilities and multiple disabilities in particular are susceptible from a young age to life-threatening somatic conditions and therefore may require palliative care from birth on [6].

Identifying palliative care needs and adequate provision of palliative care for people with ID can be challenging, because people with ID often experience difficulties in understanding their illness and are not always able to verbally communicate their feelings and symptoms to others [7-9]. This makes it hard for professionals to assess and discuss the wishes and needs of people with ID [10].

Moreover, professionals caring for people with ID are often social workers, rather than nurses or physicians, who have no or limited training in palliative care [11, 12]. In addition, training material and innovations on palliative care geared to professionals providing care for people with ID are scarce [13-15].

A final factor concerns the joint provision of palliative care by relatives and care staff. This can be challenging if perceptions of what ensures good end-of-life care differ between staff and relatives. On the other hand, if professionals and relatives agree about care goals and cooperate, this can greatly enhance the quality of care and joint interpretation of symptoms [12].

The above mentioned challenges make palliative care for people with ID different from palliative care for patients without ID. However, palliative care innovations that were originally developed for use in the general population, may also be helpful for improvement of palliative care and pro-active communication about needs for future treatments and care (advance care planning) for people with ID.

In the Netherlands, a variety of palliative care innovations were implemented in the National Quality Improvement Programme for Palliative Care between 2012 and 2016 [16]. Nine of the 76 implementation projects within the framework of this national programme were carried out in organizations providing residential care for people with ID.

In general, it is important for successful implementation of care innovations to consider the innovation's fit with the specific patient population and context [17, 18]. In addition, successful implementation is influenced by factors described in the Consolidated Framework for Implementation Research (CFIR): the characteristics of the innovation, the outer setting, the inner setting, the individuals involved and the process of implementation [19, 20].

To our knowledge, no research has been done regarding factors influencing the implementation and sustainment of palliative care innovations in organizations providing care for people with ID. This paper aims to give insight into how palliative care innovations, originally developed for the general population of palliative care patients, can be implemented and sustained in care organizations for people with ID. The research questions are:

1. How were the palliative care innovations implemented in organizations providing care for people with ID, within the framework of the Dutch National Quality Improvement Programme for Palliative Care?
    a. What were the planned implementation activities and to what extent were these plans realized in the project period of one year?
    b. What adjustments to the innovations were needed to enhance implementation specifically in care organizations for people with ID?
2. What facilitators and barriers influenced implementation of the palliative care innovations in care organizations for people with ID?
3. To what extent were the innovations sustained within the organizations one to three years after the start of the implementation, and which factors played a role in the sustainment?

Materials and methods

Design

A multi-method evaluation was conducted, involving document analyses of the project proposals and final reports, qualitative group interviews and a structured questionnaire completed by project managers. These project managers worked for the participating organizations and coordinated the implementation projects in question. The various methods complemented each other to provide a more in-depth understanding of the implementation and sustainment process of palliative care innovations [21-23].

Context

This evaluation study was a sub study within the broader evaluation of the National Quality Improvement Programme in the Netherlands [16]. During the programme period (2012–2016), health care organizations were invited to choose a specific innovation and to submit a project proposal to the Netherlands Organization for Health Research and Development. Nine project proposals of care organizations for people with ID were accepted and received a grant to implement the innovation. The formal project period lasted one year, all nine projects started between 2014 and 2016. The questionnaire was completed by project managers in 2018; this was three years (n=1), two years (n=2) or one year (n=6) after the end of the formal project period.

The participating organizations provided residential care for people with ID living in group homes in the community or in larger-scale supported accommodation facilities. Professional staff working onsite in these accommodations were mostly educated as social workers or nurses. The palliative care innovations chosen in the projects concerned:

- a tool (‘Signal Box’) to identify palliative care needs, or
- the introduction of a specialist palliative care consultant, or
- a communication intervention (‘Dying Your Own Way’) to support the communication between patients, their families and professionals about the patient’s wishes and needs at the end of life.

The three above-mentioned care innovations were implemented in respectively three, four and two implementation projects in organizations for people with ID. For more details about the palliative care innovations, see Table 5.1.

Table 5.1. Description of the palliative care innovations

Signal Box: a tool to identify palliative care needs and symptoms

The Signal Box consists of an instruction and action plan, worksheets and information for care professionals about assessing palliative care needs and symptoms and about implementation of the tool. The aim of the Signal Box is to help the professional identify palliative care needs and articulate those needs, to strengthen cooperation with colleagues in this regard, to pro-actively communicate with others about palliative care needs and symptoms (advance care planning), and to meet the wishes and needs of the patients. Professionals learn how to use the Signal Box in workshops and a train-the-trainer course.

Specialist palliative care consultant

A specialist palliative care consultant spreads knowledge, methodologies and facilities related to palliative care within an organization. Professionals receive training to work as a specialist palliative care consultant within their organization. The training to become a specialist palliative care consultant consists of nine training days, with a total study time of 200 hours. The training focusses on knowledge about palliative care, communication, palliative symptoms, coaching, spiritual care and ethical dilemmas.

Dying Your Own Way (in Dutch: STEM): a communication-based intervention

Dying Your Own Way is a communication intervention to accelerate expertise, to create awareness of the diversity of patients' wishes and needs at the end of life, and to improve professionals' ability to communicate pro-actively with patients and relatives about wishes and needs at the end of life. Dying Your Own Way involves information books and a card game for professionals, and a workbook for patients and relatives about wishes and needs related to the end of life. The innovation is supported by training meetings, workshops, and an e-learning course for professionals.

Data collection and analyses

Multiple sources of data were used to answer the research questions (see Table 5.2):

(a) Document analysis of project proposals and final reports on the implementation projects written by the project manager was conducted. A structured extraction form developed by the authors of this study was used to extract relevant data from the documents, with topics concerning general characteristics of the organization, implementation activities and influencing factors. The project proposals described the aim of the project and planned implementation activities. The final reports described to what extend the aim had been achieved and the implementation activities were realized, and which facilitators and barriers played a role.

(b) Qualitative group interviews (n=9) with the project managers and other professionals involved in the innovation, such as nurses, social workers, palliative care specialists and managers, were conducted for each project separately one year after the start of that specific project. A topic list was used to guide the interviews, concerning questions about whether the innovation was implemented as planned, what the facilitators and barriers were for the implementation, which implementation activities were carried out and whether the project managers and other stakeholders felt implementation had been successful and effective. The group interviews took around one hour, were audio-recorded and transcribed verbatim. The group interviews were analysed qualitatively with the aid of the software package MAXQDA, using the principles of thematic analysis [24]. In a first step, the three authors (AF, AdV, and HV) read all documents to become familiar with the data. Then, in a second step, the interview transcripts were inductively coded by two authors independently (AF and HV or AdV and HV). Subsequently, these codes were grouped and categorized with the aid of the five main constructs of the CFIR [19]: characteristics of the innovation (the core components and adaptable elements, structures and systems related to the innovation), the outer setting (the context outside the organization), the inner setting (the context within the organization), the characteristics of the individuals who implemented the innovation, and the implementation process. The codes were linked directly to the content of the interview fragments. In the third step of our thematic analysis, the codes and their assignment to the constructs of the CFIR were discussed by all authors in order to reach consensus. After that, the main themes related to the codes were reviewed and compared, taking account of the coded material in the interview transcripts and comparing transcripts. In step five, the names and specifics of each theme were further refined and defined in a discussion with all the authors. In the last step, the themes, content and associated quotations were reported in this paper.

(c) A questionnaire was sent to the project managers (n=9) one to three years after the implementation project. Seven project managers completed the questionnaire online, while the two other project managers preferred to complete the questionnaire in a telephone interview. The first part of the questionnaire included closed-ended questions about the sustainment of the innovation: “Is the innovation still being applied?” (“yes”, “partly”, “no”), and “Is the innovation well implemented and sustained?" ("yes", "I'm not sure, but I do think so", "I'm not sure, but I don't think so", "no"). Project managers also indicated if they thought the innovation currently had added value for patients, family or professionals ("yes", "no"). At the end of the questionnaire, two open-ended questions were included about the barriers and facilitators for sustaining the innovation. The answers were coded according to the codes developed from the group interviews. The information from the questionnaire was analysed descriptively using STATA version 15.

Table 5.2. Overview of information sources for each research question

Research question	Information source
1. How were the palliative care innovations implemented in organizations providing care for people with ID, within the framework of the Dutch National Quality Improvement Programme for Palliative Care?	- implementation plans written by the project manager - final reports of the implementation projects written by the project manager - group interviews with project managers and other professionals
2. What facilitators and barriers influenced implementation of the palliative care innovations in care organizations for people with ID?	- final reports of the implementation projects written by the project manager - group interviews with project managers and other professionals
3. To what extent were the innovations sustained within the organizations one to three years after the start of the implementation, and which factors played a role in the sustainment?	- final reports of the implementation projects written by the project manager - group interviews with project managers and other professionals - online questionnaire completed by project managers

Results

Implementation process of palliative care innovations

Planned and realized implementation activities

The first research question concerned planned and realized implementation activities and the adjustments made to the innovation. The planned and realized implementation activities consisted of training for professionals, cooperation with other organizations and other implementation activities, mainly focussing on dissemination of the innovation (see Table 5.3).

All nine projects planned and realized training as part of the implementation process. The number of professionals who received training varied between the implementation projects, depending in part on the kind of innovation. In the projects implementing the Signal Box, between seventeen and 49 professionals attended workshops and a train-the-trainer course; in the projects implementing the Dying Your Own Way communication intervention ten professionals were trained; and in the projects introducing a specialist palliative care consultant, between two and six professionals completed nine training days to become a palliative care consultant.

In addition, all nine projects planned activities aimed at cooperation with other organizations, such as other care organizations for people with ID or organizations within a palliative care network. In most projects, cooperation was successfully realized. However, two organizations (implementation projects 2 and 3) that planned to work together did not succeed in doing so. In implementation project 6, contact was made with a hospital, but this had not yet resulted in actual cooperation.

All projects also planned for and realized other implementation activities, in full or in part. These activities mainly focussed on dissemination of the innovation within their organization by making use of internal communication networks and social media, distributing flyers, and organizing meetings to inform professionals or other stakeholders about the innovation. Other implementation activities concerned embedding the innovation within existing workflows or systems, appointing opinion leaders who were responsible for the use of the innovation, and reflecting on or evaluating the use of the innovation.

Table 5.3. Overview of the implementation projects, setting, and planned and realized implementation activities

Implementation projects		
Project number, innovation and setting	*Planned implementation activities*	*Realized: yes, partly, no*
1. Signal Box: a tool to identify palliative care needs *Implementation project in one location of an ID care organization which provides care for ageing people with ID*	*Training*: Provide training for 58 professionals with different backgrounds to work with the innovation.	*Partly:* 49 professionals were trained.
	Cooperation activities: Results will be shared with various palliative care networks.	*Yes*
	Other implementation activities: - A protocol for using the innovation will be made. - The innovation will be part of regular team discussions.	*Yes*

Implementation projects

Project number, innovation and setting	*Planned implementation activities*	*Realized: yes, partly, no*
2. Signal Box: a tool to identify palliative care needs *Implementation project in a care organization providing residential care and support for people with ID*	*Training:* Offer training for 20 care staff who provide palliative care for people with ID throughout the organization.	*Partly:* 17 professionals were trained.
	Cooperation activities: Working together with another ID care organization and the organization that developed the Signal Box.	*Partly:* Cooperation with another ID care organization was not realized.
	Other implementation activities: - An instrument to signal palliative care needs for people with ID will be added to the Signal Box. - A process evaluation will be conducted among participating professionals to check if changes are required.	*Yes*

Implementation projects

Project number, innovation and setting	*Planned implementation activities*	*Realized: yes, partly, no*
3. Signal Box: a tool to identify palliative care needs *Implementation project in three teams who provide care for ageing people with ID (aged ≥50) within an ID care organization*	*Training*: Plan to train 38 professionals to work with the innovation.	*Yes*
	Cooperation activities: Working together with another organization to develop an addition to the Signal Box focussing on palliative care needs of people with ID.	*No:* Cooperation with another ID care organization was not realized.
	Other implementation activities: - A kick-off meeting will be organized three months after the start of the project. - Knowledge about signalling palliative care needs will be spread throughout the organization.	*Yes*

Implementation projects

Project number, innovation and setting	*Planned implementation activities*	*Realized: yes, partly, no*
4. Introduction of a specialist palliative care consultant *Implementation project in an organization providing care for people with ID, people with psychiatric disabilities and nursing-home residents*	*Training*: Three professionals working within the organization will be trained to become a palliative care consultant.	*Yes*
	Cooperation activities: Plan to work together with other organizations for people with ID.	*Yes*
	Other implementation activities: - A guide will be developed specifically for providing palliative care for people with ID. - Training will be developed for professionals and volunteers.	*Yes*

Implementation projects

Project number, innovation and setting	*Planned implementation activities*	*Realized: yes, partly, no*
5. Introduction of a specialist palliative care consultant *Implementation project in two locations of an ID care organization providing residential care and support*	*Training*: Four professionals will be trained to become a palliative care consultant.	*Partly:* One trained palliative care consultant resigned.
	Cooperation activities: Cooperation between different locations of the organization, with the palliative care network, and with external experts.	*Yes*
	Other implementation activities: - A kick-off meeting will be organized in both locations and an advisory committee will meet five times during the implementation project. - Palliative care consultants will be working throughout the organization.	*Yes*

Implementation projects

Project number, innovation and setting	*Planned implementation activities*	*Realized: yes, partly, no*
6. Introduction of a specialist palliative care consultant *Implementation project in three locations of an organization providing care for people with ID and people with psychiatric disabilities*	*Training*: Six nurses will be trained as palliative care consultants.	*Yes*
	Cooperation activities: - The organization will work together with a foundation for palliative care and the palliative care network. - Cooperation will be established between the organization and palliative care teams in hospitals in the north of the Netherlands.	*Partly:* - Cooperation with the palliative care foundation and the palliative network was realized. - Contact with a hospital was made, but there was no cooperation as yet.
	Other implementation activities: - Palliative care policies and instruments used within the organization will be further developed. - A guide will be developed specifically for providing palliative care for people with ID.	*Partly:* - Palliative care was embedded in the organizational policy; the ‘Palliative care’ handbook was updated. - The guide for providing palliative care for people with ID was not developed.

Implementation projects		
Project number, innovation and setting	*Planned implementation activities*	*Realized: yes, partly, no*
7. Introduction of a specialist palliative care consultant *Implementation project in one location of an ID care organization providing residential care and support*	*Training*: Two nurses will be trained as palliative care consultants.	*Yes*
	Cooperation activities: The organization will work together with palliative care networks.	*Yes*
	Other implementation activities: - Materials will be developed to introduce the palliative care consultants. - Training will be aligned with palliative care needs of people with ID in cooperation with other organizations.	*Partly:* - The palliative care consultants introduced themselves to the organization and made flyers. - Alignment of the training with palliative care needs of people with ID had not yet been realized.

Implementation projects

Project number, innovation and setting	*Planned implementation activities*	*Realized: yes, partly, no*
8. Dying Your Own Way (in Dutch: STEM): a communication intervention *Implementation project in three locations of a care organization providing residential care and support for people with ID, people with non-congenital brain injury, people with physical disabilities and people with psychiatric disabilities*	*Training*: Ten professionals will be trained to work with the innovation.	*Yes*
	Cooperation activities: - The organization will work together with a palliative care network in the region and with the organization that developed the innovation.	*Yes*
	Other implementation activities: - The material will be modified to align it with the target group of people with ID. - A kick-off meeting, feedback and multidisciplinary meetings and an evaluation meeting with trained professionals will be organized.	*Yes*

Implementation projects

Project number, innovation and setting	*Planned implementation activities*	*Realized: yes, partly, no*
9. Dying Your Own Way (in Dutch: STEM): a communication intervention *Implementation project in three teams within an ID care organization: a team of professionals who work with children with ID, a team of professionals who work with adults with ID, and a team of professionals who work with ageing people with ID*	*Training*: Three teams of professionals will be trained.	*Partly:* Ten professionals participated in the training.
	Cooperation activities: - Cooperation will be established with other ID care organizations that implemented Dying Your Own Way. - Knowledge and experience of the implementation project will be shared with a foundation for palliative care and other relevant organizations.	*Yes*
	Other implementation activities: - A kick-off meeting, multidisciplinary meetings, and a final meeting will be organized. - Two opinion leaders will be appointed in each team. - Where necessary, the innovation materials will be adapted for people with ID.	*Partly:* - Three meetings were organized: a kick-off meeting, an intermediate meeting, and a final meeting. - Opinion leaders were appointed. - The innovation materials were not adapted for people with ID.

Adjustments to the innovation

In five of the nine implementation projects, there were plans to further develop the innovation to align it with the specific needs of people with ID. However, according to the final reports, only two implementation projects managed to do so in practice, see also Table 5.3. In implementation project 2, in which the Signal Box was implemented, an instrument for recognizing palliative care needs specifically in people with ID (PALLI [25, 26]) was incorporated in the training for professionals. In implementation project 8 regarding the Dying Your Own Way communication intervention, short movies about palliative care specifically for people with ID were developed and used as a visual tool in training.

Facilitators and barriers for implementation

The second research question focussed on facilitators and barriers for implementation. Various influencing factors for implementation were identified, see Table 5.4. In the text, the influencing factors are marked in italics and further explained.

Table 5.4. Overview of factors influencing implementation within the Consolidated Framework for Implementation Research constructs

Characteristics of the intervention	Outer setting	Inner setting	Characteristics of professionals or teams	Process of implementation
- Applicability - Quality of materials and training - Fit with the target group - Complexity - Costs	- Cooperation with other organizations - National policy and incentives	- Involvement of board and management - Priority given to palliative care within organization - Integration of innovation in existing workflows or systems - Self-directing versus a hierarchical structure - Multidisciplinary collaboration - Commotion within organization -Available resources - Needs and characteristics of clients/relatives	- Motivation for participation - Awareness of importance of palliative care - Knowledge and skills	- Planning and clarity of goals - Announcing and stimulating use of innovation - Involving the right people - Having an 'opinion leader' or professional who is responsible for implementation - Reflecting on and evaluating the use of the innovation

Influencing factors: characteristics of the intervention

Five characteristics of the innovation influence the implementation process: the applicability of the innovation, the quality of the materials and training, the fit of the innovation with the patient group, the complexity, and the costs of the innovation. The *applicability*, which is regarded as the degree to which the innovation has been found usable and valuable in practice, was considered to be high in all implementation projects. As an example, a participant talking about the Signal Box said that the instrument could be widely used within the organization and was found to be valuable in practice by different professionals:

There's an awful lot of enthusiasm, in particular about the practical applicability of the Signal Box. There's not really anyone — at least from the feedback we've had — who has said they can't do anything with this or that it isn't remotely appropriate. The daily care professionals themselves, the supervisors, also find it really easy to use — of course it is very concrete. You just ask questions and you're able to give an answer. But we also see that the group around them — the doctors and behavioural specialists — also find it very valuable. (Source: interview, implementation project 3, Signal Box).

Secondly, the high *quality of the materials and training* facilitated implementation. For example, project managers and professionals perceived the design, content and presentation of the materials in the Signal Box to be of high quality. The communication intervention Dying Your Own Way was found to be of high quality because of its constructive content with an obvious start and end.

Nevertheless, barriers regarding the characteristics of the innovation were also mentioned. Regarding Dying Your Own Way, the focus on 'Dying' was mentioned as a factor obstructing implementation. Talking with people with ID and/or their relatives about ways of dealing with dying was perceived as too challenging and raw for some professionals, who rather preferred to talk about styles in how to live their remaining life. As one participant said:

That's why I do wonder sometimes whether it might help if you didn't call it 'dying' but 'styles of living' instead. Don't see it as palliative care but see it as... Because that's very challenging for supervisors. Whereas I know for sure that they could use it, shall we say. And that's difficult with the Dying Your Own Way method. To say, 'Right, we're going to spend a morning talking about styles of dying'. (Source: interview, implementation project 8, Dying Your Own Way)

A third influencing factor concerned the *fit with the target group*. The lack of fit was experienced as a barrier because the innovations and corresponding training were not specifically developed for professionals caring for people with ID. Concerning the palliative care consultant training, some of the professionals involved said that the training focused too much on the medical dimension of palliative care. They missed information about how to deal with psychosocial and spiritual needs, and about their changing roles and responsibilities when providing palliative care for people with ID. Regarding the Signal Box, participants said that they missed specific information in the instrument about relevant symptoms for people with ID in the palliative phase such as epilepsy and auto-mutilation.

The *complexity* of the innovation was a fourth influencing factor for implementation. Professionals said they found it complex to determine when the innovation could be used, for example when the palliative care specialist can be consulted or at what moment in the illness trajectory or life course of an individual the Signal Box can be helpful, which was considered as a barrier for implementation. Also, the professionals who completed the training to become a palliative care consultant said it was challenging because they had little time available for their work as a consultant in addition to their regular work tasks. Furthermore, the Dying Your Own Way innovation was perceived as intensive due to the frequent meetings and workshops included in the training programme.

The last influencing factor regarding the characteristics of the innovation was *costs.* High costs, for example associated with training for professionals as part of the implementation, hindered further implementation because not all professionals within an organization could be included due to financial limitations. A project manager said: *We would like to continue [the innovation]. But the costs of hiring the trainer are so high. (Source: interview, implementation project 9, Dying Your Own Way)*

Influencing factors: the outer setting

Regarding the outer setting, two influencing factors were mentioned: cooperation with other organizations and the national policy and incentives. *Cooperation with other organizations* was mentioned in all projects as a facilitator for implementation. Project managers found it important to learn from other organizations, and to share their own knowledge and expertise. They cooperated with other care organizations for people with ID, but also with other organizations or care settings, such as hospitals, and with regional palliative care networks. One participant said that, because the importance of palliative care for people with ID was widely recognized, she found organizations to be very willing to cooperate and share information:

Once I got involved with this and started searching the Internet and phoning people and sending them e-mails, saying ‘Hey, I’m working on this and I hear you’re doing whatever’... I e-mailed Person M because she’s got a palliative unit in her organization, of course. I found there was an incredible willingness to share information. I have yet to speak to an organization that says, ‘Well, we’ve developed this manual but you can’t have it’. It’s often a question of: ‘Here it is, see what you can get out of it’. Precisely because everyone recognizes the importance of setting that care up well. (Source: interview, implementation project 4, specialist palliative care consultant)

The need for cooperation also arose because caring for incurably ill people in the final stage of life was not everyday practice in the organizations involved. The organizations had relatively few deaths among people with ID and the participants explained they had little experience in supporting people with ID at their end of life. In order to increase the use of the innovation and develop expertise, organizations found it helpful to be able to work together with other organizations for people with ID.

Other facilitating factors for the implementation that were mentioned concerned *national policy and incentives*. The implementation projects’ organizers submitted an application to participate in the National Quality Improvement Programme for Palliative Care and, if accepted, were granted money to implement the innovation. One participant said that the participation in the National Programme and the associated obligations, for example writing a project proposal, helped in starting up the implementation and boosted implementation within the organization:

The National Quality Improvement Programme for Palliative Care gives a really good basis for initiating genuine change and improvement. It gives you not only a temporary financial boost, but also a real plan, not just some optional project proposal. (Source: interview, implementation project 6, specialist palliative care consultant)

Influencing factors: the inner setting

Most influencing factors mentioned by participants concerned the inner setting of the organization. First of all, according to the participants, the *involvement of the board and management* in the project was necessary to start and facilitate the implementation in the organization. Project managers mentioned that the board or management endorsed the importance of implementation, which was seen as a facilitating factor for implementation. Also the high *priority given to palliative care within an organization*, often due to a growing number of ageing or frail people with ID, was a facilitating factor for implementation. In some of the organizations, palliative care was explicitly mentioned in their vision and policy documents and implementation was therefore supported.

A third influencing factor regarding the inner setting which came up in most implementation projects was the *integration of the innovation in existing workflows or systems*. This was seen as another important facilitator. Integrating the innovation was done in several ways, for example by inserting the Signal Box instrument in the organization's reporting system, inviting the palliative care specialists to attend regular team meetings, or including Dying Your Own Way in existing training opportunities.

A fourth influencing factor was the degree to which the organization had *self-directing teams rather than a hierarchical structure*. This was mentioned as a barrier for implementation. If professionals in self-directing teams were not motivated or set other priorities, it was found to be difficult to stimulate the use of the innovation. One participant said that the absence of a clear leader who tells others what to do hindered implementation:

What we do see… Because we also switched to self-directing teams, we lost an entire stratum of team leaders. And then you do see that sometimes you simply, well, miss someone you can speak to in some cases. And sometimes that resolute person who says, 'Hey, this is what we agreed, so we should stick to that'. (Source: interview implementation project 6, specialist palliative care consultant)

A fifth influencing factor related to the organization was *multidisciplinary collaboration*. Participants explained that in palliative care for people with ID, different professionals were involved, such as daily caregivers, physicians, specialized physicians for people with ID, behavioural specialists and spiritual caregivers. Involvement of a multidisciplinary group of professionals, and good and frequent communication and collaboration between these professionals, boosted the use of the innovation and was considered as a facilitating factor for implementation.

Other influencing factors were *commotion within the organization*, for example because of personnel changes or reorganizations, and insufficient *available resources* such as time, money and materials .These factors were mentioned in all projects as barriers for implementation. Project managers explained that it was important to choose the right moment to start the implementation, a moment when there is sufficient time available.

A final influencing factor concerning the inner setting was *characteristics of the client group*. Participants explained that for some people with ID and close family members, it was difficult or too challenging to talk about palliative care and death, because the need for palliative care was not always understood or accepted. This was mentioned as a barrier for the use of innovations, which focussed on pro-active communication about palliative care needs and discussing wishes around the end of life. However, participants also said that some relatives of people with ID did see the importance of talking about wishes and needs for future care, which boosted the use of the innovation:

I sat in on a meeting and relatives were there too. The care professionals had the attitude of 'Right, we'll tackle this topic'. So at some point they started talking about death, whereas the client wasn't anywhere near that stage. At first, it was a bit like, 'OK, what's happening now?' But there came a point where the relatives saw the benefit of reflecting on this already. Then they asked us about the wishes booklet: had we actually filled it in, well we really should do that. So they came up with it themselves and starting discussing things with us. (Source: interview, implementation project 7, specialist palliative care consultant)

Influencing factors: characteristics of professionals or teams

Three influencing factors were found regarding the characteristics of the professionals or teams. *Motivation for participation* among professionals and teams was an important facilitating factor for implementation. The project managers mentioned in the interviews that some of the teams involved had already seen palliative care as important before the implementation project, and therefore those professionals were interested in becoming involved and motivated to participate in the training and use the innovation. At the start of the project, *the awareness of the importance of palliative care* was less clear in other teams, which was seen as a barrier for implementation, for example because professionals held the opinion that the people they cared for did not (yet) require palliative care. One team leader said that, while they cared for ageing people with ID, investing time in palliative care was not seen as a priority:

Right, I can only speak for my own team but it wasn't feasible for me to tackle it with the entire team in one go because it simply costs too much time. If we already need three half-days for training in it, let alone how you train a team that doesn't have much affinity with the subject… the target group isn't yet big enough, shall we say. Our group is getting older but not yet to the extent that you think, 'Well, we really need this'. (Source: interview, implementation project 1, Signal Box)

Moreover, professionals in care organizations for people with ID were not always experienced in palliative care, and therefore felt they lacked *knowledge and skills* regarding palliative care or felt unsure about using the innovation, which was another barrier for implementation. Participants explained that professionals found it difficult to discuss end-of-life issues, and they required experience to feel comfortable and secure.

Influencing factors: the implementation process

Five influencing factors were mentioned regarding the implementation process. First of all, *planning and clarity of goals* were often mentioned as facilitators. Project managers said that it was important to make a clear implementation plan before the start of the implementation, together with the professionals involved, which the management or board also approves and which clearly describes the aim and goal of implementation within the organization.

During the implementation process, another important facilitator was *announcing and stimulating the use of the innovation.* Various dissemination strategies and materials were used to promote the implementation, such as distributing flyers, organizing meetings, mentioning the innovation in newsletters and making use of social media. A project manager explained the importance of spreading information about the innovation:

I notice that it helps. Even a small piece of information on our webpage. Not everybody will read it, but at least the people who do are informed. The same text is spread in the newsletter or send to the medical consultation board. So we use existing channels to make people enthusiastic. (Source: interview, implementation project 1, Signal Box).

A third influencing factor was *involving the right people* in the implementation process. This was done in implementation projects in different ways. One project leader of an organization that implemented palliative care consultants said that professionals had to apply to be included in the training and they made use of strict selection criteria. In another implementation project concerning the Signal Box, the team manager decided which professionals they wanted to involve and how. Moreover, *having an 'opinion leader' or professional who was responsible for implementation* was a fourth influencing factor, seen as an important facilitating factor in boosting the use of the innovation. One participant even said that the success of the implementation depended on the opinion leader:

That has been a pitfall in the sense that, well, the whole process of implementing it in the home depends crucially on the opinion leader who is in charge of doing it. Well, some manage just fine and others find it more difficult. (Source: interview, implementation project 1, Signal Box)

The last influencing factor related to the implementation process, *reflecting on and evaluating the use of the innovation* with the professionals involved, facilitated implementation. Participants found it helpful to know if and how professionals made use of the innovation in practice, in order to find out if changes were required to the implementation process.

Sustainment of innovations

The third research question focused on the sustainment of innovations within the organizations. According to the questionnaire completed by the project managers (n=9), all organizations were still using the innovation one to three years after the start of the implementation. Also, they all believed that the innovation currently had added value for the people with ID, their families and professionals. On the other hand, when the project managers were asked if they felt the innovation was well implemented and sustained within their organization, five of the nine were hesitant. One project leader responded "I'm not sure, but I don't think so", four project managers answered "I'm not sure, but I think so", and four project managers answered "Yes".

Barriers for long-term sustainment mentioned by project managers were lack of *involvement of the board and management* and insufficient *available resources* (inner setting), and not *having an 'opinion leader' or professional who is responsible for implementation* (process). According to the project managers, everyone was still enthusiastic about the innovation after one to three years. Professionals were convinced of the innovation's *applicability* and its value in practice (innovation characteristics), which was an important facilitator for sustainment. Other facilitators for sustainment mentioned in the questionnaires were: *cooperation with other organizations* (outer setting), the *priority given to palliative care within the organization* (inner setting), and the *professionals' motivation* to use the innovation (characteristics of individuals or teams).

Discussion

This study described how palliative care innovations were implemented in organizations for people with ID. Training of professionals, cooperation with other organizations and dissemination of the innovation were important activities when implementing palliative care innovations. The innovations, originally developed for use in the general population of palliative care patients, were found to be applicable in care organizations for people with ID, although some adaptations had to be made. When making use of an existing innovation, it is important to consider how an innovation can be adapted to make it more suitable for a particular population or a better fit with the organization [17, 27]. Two of the nine projects did indeed adapt the innovation, and in almost all projects there were plans for adaptations. The one-year implementation period might have been too short to adapt the innovation to fit the specific needs of people with ID, which requires expertise and practice [28].

This paper also described facilitators and barriers for implementation. Categorized according to the Consolidated Framework for Implementation Research, most influencing factors concerned the inner setting of the organizations. As is the case in the implementation of palliative care innovations in other settings [29, 30], it was found to be important to have support from management, to have sufficient resources available and to integrate the innovation in existing workflows or systems. Moreover, the quality and applicability of the innovation and having an 'opinion leader' or professional who is responsible for implementation are important influencing factors.

Some influencing factors for implementation were related to the specific care setting of people with ID. The limited palliative care knowledge and skills of professionals working with people with ID, which has been highlighted in previous research [9, 11, 28, 31-33], was found to be a barrier for the use of the innovation. It is also known that professionals avoid the end of life as a topic and do not know how to talk about it. Therefore, it is very important to include training for professionals providing care for people with ID as part of the implementation of palliative care innovations. For example, professionals should be trained in how to inform people with ID about dying and death and in proactively discussing wishes and needs (advance care planning)[34, 35]. Moreover, cooperation with other organizations for people with ID was a facilitating factor for implementation and sustainment. Despite growing incidences of the life-threatening illnesses that affect older people, it is still the case that relatively few people in care organizations for people with ID need palliative care [36]. The European Association of Palliative Care (EAPC) White Paper about consensus norms for palliative care for people with ID also states that the on-going exchange of experiences and expertise and sharing best practices is necessary to ensure that people with ID have their palliative care needs met [37].

Lastly, this paper focussed on the long-term sustainment of the palliative care innovations. After one to three years, the innovations were still used in all the ID organizations involved. Although project managers were convinced of the applicability of the innovation and its value for practice, five of the nine project managers were unsure whether the innovation was *well* implemented and sustained within their organization. Previous research also shows that effective implementation is challenging [38] and even when innovations are well designed, contextual factors may hinder implementation and sustainment of an innovation [17, 39]. Barriers for long-term sustainment found in our study were lack of support from management, insufficient resources and the absence of an 'opinion leader'. It is recommended that organizations have resources available to sustain the future use of the innovation and appoint professionals for a long term who can lead the implementation and further use of the innovation.

Strengths and limitations

This study has a unique focus on implementing and sustaining palliative care innovations in care organizations for people with ID and offers important insights for practice. The inclusion of multiple care organizations, different palliative care innovations and multiple data sources (document analysis, interviews, and a questionnaire) leads to a comprehensive picture of influencing factors for implementation and sustainment and improves the generalizability of the results. Moreover, this study contributes to the implementation of innovations concerning timely identification of palliative care needs and pro-active communication about wishes for future care (advance care planning), which is important for high-quality palliative care. However, some limitations of this study should be noted.

One limitation concerns that the project reports were written by the project managers, and therefore these documents may represent a more positive image of implementation plans and achieved activities than was actually realized. In future research, additional observations on site during the formal project period could be conducted to gain more insight into the actual implementation process and its influencing factors. A second limitation concerns the fact that the questionnaire for project managers was sent one to three years after the formal project period. This difference in timing of data collection may have caused variability in sustainment between projects. A final limitation is that it remains unknown how people with ID and their relatives experience the palliative care innovations and whether they think that the implementation of the innovation in question has improved palliative care.

Conclusions

This study shows that palliative care innovations originally developed for use in the general palliative care population can be successfully implemented in care organizations for people with ID. The three palliative care innovations were found to be applicable, although adaptation to the specific setting might be necessary. Training for professionals, collaboration with other organizations and dissemination activities are important elements when implementing palliative care innovations. Comparable to other healthcare settings, most factors influencing implementation concern the inner setting of the organization; they include having support from management and integrating the innovation in existing workflows. Organizations should have resources available to sustain the future use of the innovation and they should appoint ‘opinion leaders’ who are responsible for the implementation.

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Chapter 6

Development, implementation, and evaluation of an Advance Care Planning program for professionals in palliative care of people with intellectual disability

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Abstract

In this paper we describe how an advance care planning (ACP) program was developed, implemented, and evaluated. Our aim was to improve ACP in palliative care for people with intellectual disability (ID). The program was based on ten competencies needed for ACP and was developed in a co-design process with people with ID, relatives and professionals. The program was implemented in six ID care organizations in the Netherlands and consisted of an information pack, a training course, and an implementation interview about implementing ACP. Professionals indicated that their competencies had improved, particularly regarding communication and the application of ACP as a standard element in palliative care practice. This program therefore seems helpful in training ID care professionals in the competencies needed for ACP.

Key words: Advance Care Planning, Palliative Care, Intellectual Disability, End of Life

Introduction

According to the World Health Organization [1], palliative care is “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”. There has been an increase in awareness of end-of-life and palliative care issues in recent years for people with intellectual disability (ID) [2, 3].

The WHO definition on palliative care was extended in the Dutch Palliative Care Quality Framework to refer to “Patients and their families facing the problem associated with a life-threatening illness or vulnerability” [4]. This extension means the definition also applies well to palliative care for people with ID. Previous studies have shown that a higher percentage of people with ID are susceptible to life-threatening somatic conditions (such as serious respiratory or cardiovascular conditions) — often from a relatively young age — than of people without ID [5]. People with profound intellectual and multiple disabilities are particularly susceptible [6, 7]. They need care from birth that is geared to the quality of life, comfort and pain relief — in other words, palliative care. At the same time, people with ID are living longer on average because of improvements in medicine and their deaths are now often preceded by a period of illness and increasing vulnerability [8-10].

Good palliative care requires Advance Care Planning (ACP) [11]. ACP lets people define objectives and preferences for future medical treatments and care, discuss these objectives and preferences with relatives and care professionals, document them and revise them if necessary [12]. The patient’s wishes and needs are given priority and are recorded in their care plan. The care plan can also include advance care directives, which are legal documents recording preferences for a surrogate decision-maker and regarding life-prolonging procedures [13]. However, ACP is about more than just using an Advance Care Directive. The primary focus in ACP is on the process of communication about future physical, psychological, social and spiritual care needs, which goes beyond the completion of advance directives.

ACP helps put the person with ID in control and improves the quality of the palliative care [14-16]. It also increases the likelihood that the person with ID will receive care commensurate with their wishes and expectations [10, 17]. Relatives can provide support for the person with ID and they play an important role in ACP conversations and decisions [18, 19]. Because possible scenarios are considered at an early stage, there are fewer unforeseen situations, which has a positive effect on the quality of life of the person with ID [10, 20].

For ACP to work, the person with ID's wishes and needs have to be identified at an early stage. However, professionals often find it difficult to recognize the needs of individuals with ID, especially the need for palliative care [9, 21-23]. This is partly because people with limited cognitive and communicative capabilities have difficulty expressing their wishes and needs [10, 23-27].

Uncertainty among professionals about various aspects also serves as a barrier to ACP. It is not always clear who is responsible for taking the initiative for ACP as several different professionals may be involved in the care delivery, for example physicians, nursing staff, caregivers, social workers and behavioural experts [21]. Furthermore, not all professionals know when to start ACP. What the medical condition means for an individual's future may also be unclear. Moreover, some professionals believe that ACP causes stress and anxiety among people with ID and/or their relatives [28, 29]. They may feel uncomfortable with the emotions that ACP conversations can evoke and some professionals lack the communication skills to discuss emotionally charged subjects [25, 30] and feel insufficiently equipped to provide good palliative care [31-33].

As far as is known, no ACP program had been developed yet for professionals caring for people with ID. It is important to have a program geared to this specific group as people with ID and their relatives vary considerably in how they want (and are able) to consider the future [34, 35]. It is particularly difficult to determine what constitutes good quality of life for people with ID who have difficulty expressing themselves verbally [19]. Consequently, care professionals need to use a sensitive, tailored approach in ACP for people with ID.

In the Netherlands, medical care is usually provided by general practitioners (GP) or specialized ID physicians. Specialized physicians who work with people with ID have three years of specialist training in the care for people with ID and are generally employed by an ID care service. Dutch GPs mainly care for people with mild ID living in the community, while specialized ID physicians largely care for people with moderate or severe ID, often in residential care. ID care staff in residential settings have varying backgrounds, but most are social workers or nursing staff (registered nurses or certified nursing assistants). The physician conducts the ACP conversations about the policy for medical care. However, social workers and nursing staff involved in the daily care of the person with ID also have an important role as they often conduct ACP conversations concerning the wishes and needs for future care.

Aim

The aim of this article is to provide information about the development, implementation and evaluation of an ACP program, defined as an intervention program aimed at improving ACP for palliative care among people with ID.

We answer the following research questions:

1. How did the development process inform the ACP program content?
2. What were the perceived effects of the ACP program on professional knowledge, skills, and application of ACP competencies?

Method

Participants

People with ID, relatives, and professionals who participated in the development, implementation and/or evaluation of the ACP program, were recruited from six care organizations in the Netherlands, supporting people with mild to severe ID living in group homes with 24-hour care or in residential settings. Physicians who were employed at the ID care organizations functioned as recruiters in our study. Relatives and professionals involved in the development of the ACP program received a letter from the relevant ID care organization informing them about the aim and content of the project and asking them to sign an informed consent form if they agreed to participate in the file analyses and interviews (n=63), in-depth interviews (n=15) or co-design sessions (n=12). Other than being close to a person with ID in the palliative phase or to an individual with ID who had recently died after a period of illness, there were no inclusion criteria.

People with ID involved in the development of the ACP program were informed by their care staff about the project, and could then verbally provide informed consent if they wished to participate in the in-depth interviews (n=5) or co-design sessions (n=3). The inclusion criteria were:

(a) having an understanding of the concept of death and dying; (b) having previous experience with being seriously ill and/or being hospitalized themselves; and (c) being at least 18 years old. We asked care staff to assess these criteria in their clients with ID. Because of ethical reasons, people with ID who had a life-limiting illness or received palliative care themselves were excluded from participation.

A divers group of 58 professionals who work in ID care took part in the ACP program, with between 7 and 13 from each care organization. Each organization was free to select and invite professionals for whom they felt it was relevant to participate. To evaluate the program, 53 of the 58 participants were sent a digital questionnaire (no e-mail address was available for five participants). The participants who evaluated the program (79%) consisted of physicians (n=19), social workers (n=8), nurses (n=6), and other professionals (n=9).

Procedure

The development, implementation, and evaluation of the ACP program consisted of five successive phases (Table 6.1). In Phase 1 of the development process, the project group (the co- authors of this article) studied three sources (see also Table 6.1). The first was a systematic international review of what is known about ACP in the palliative phase for people with ID [21]. Secondly, the project group used the results of patient file analyses and structured interviews with the associated relatives and professionals [29]. Thirdly, results from in-depth interviews with people with moderate ID, relatives and professionals [18] were used as the basis for choices about what the ACP program should cover.

Phase 2 of the development process consisted of co-design sessions with people with ID, relatives and professionals. The aim of the sessions was to develop a program that both fits the needs of people with ID and their relatives, and fits with what professionals require and are able to do. We followed the principles of Experience-Based Co-Design (EBCD) [36, 37]. In EBCD, various groups of stakeholders collaborate closely for a relatively long period in developing an intervention. The participants' experiences and the discussion of these experiences constitute important input for the intervention. In this instance, the stakeholders were people with ID, relatives and care professionals.

Table 6.1 Phases in the development, implementation, and evaluation of the ACP program

	Development
Phase 1:	Description of competencies that the ACP program should to focus on based on the outcomes of: a systematic international review of ACP for people with ID [21]; an analysis of the medical files of people with ID in the palliative phase (n=15) and people with ID who had died less than six months previously (n=15). The analysis of the files was validated and refined using structured interviews with relatives (n=30) and professionals (n=33) involved in the care of the individuals in question [29] in-depth interviews with five people with moderate ID, seven relatives and eight professionals [18].
Phase 2:	Development of a draft ACP program in co-design sessions with people with ID (n=3), relatives (n=2) and professionals (n=10).
Phase 3:	Development of the training course for the ACP program in consultation with two education experts.
	Implementation
Phase 4:	Implementation of the ACP program in six ID care organizations. The ACP program was aimed at care professionals, a total of 58 professionals took part in the program. The ACP program consisted of three elements: an information pack, two training sessions and an implementation interview.
	Evaluation
Phase 5:	Participants were sent a digital pre-structured questionnaire to evaluate the ACP program. Participants were asked (1) how they felt about their command of the ACP competencies, (2) if they felt their ACP competencies had improved after the ACP program, and (3) if they felt they had become better at applying their ACP competencies in their work.

Two co-design teams were formed. The first team consisted of two relatives of people with ID, two physicians, seven social workers and a pastoral worker from an organization that provides support and care for people with ID. This team met four times, with each session lasting 1.5 hours. The sessions with relatives and professionals were led by two project group members (AW and AV). The first co-design session focussed on the importance of ACP, what the right moment to discuss ACP is, who should take the initiative for ACP, and how the person with ID should be involved. In the second session, participants discussed the content of the ACP program. They also came up with the content of and approach for a conversation about ACP. In the third session, the broad outline of an ACP conversation as formulated in session two was further refined in the form of guidelines for such a conversation. Prior to the session, each participant had noted down the desired elements for such ACP conversation guidelines. They also discussed and gave feedback on an information folder about ACP for relatives. The guidelines for an ACP conversation were finalized in the fourth and last session. Participants also reflected on the topics that had arisen during the four sessions and used this to produce a summary of the components that the ACP program should contain.

The second co-design team consisted of three people with moderate ID from one ID care organization. This co-design team met three times. The aim of the sessions with this team was to highlight the views of people with ID on their future care and on talking about this so that their wishes could be taken into account in the ACP program. In the co-design sessions with people with ID, a fictional person (Anne) was introduced who was seriously ill. Illustrations forming a narrative were used. The participants with ID could use them to create their own story. Previous studies had shown this to be an approach that works well in allowing people with ID to talk about illness and death [38, 39]. In the first session, the participants discussed what they would find important if they were ill, what kind of assistance they would want and who they would want to deliver that assistance. In the second session, the participants discussed what people with ID need when they are ill and who they would want and be able to discuss this with. The third session covered decisions about treatment. Some examples were given, such as whether or not to resuscitate someone or send them to the hospital.

In Phase 3 of the development process, two project team members (AW and AC) worked with two external educational experts (specialized in communication training for medical professionals) to develop the training course within the ACP program, building on phases 1 and 2. The project team members and educational specialists met six times to discuss and design the training course.

In Phase 4 of the project, the ACP program was implemented in six ID care organizations in the Netherlands. All these organizations had also been involved in the earlier project phases. The ACP program consisted of three elements: an information pack, two training sessions, and an implementation interview. The information pack included an article in Dutch about ACP [40] and various links to research publications about decision-making in palliative care for people with ID [41-43] and to the Dutch Palliative Care Quality Framework [4], which describes what is important for high-quality palliative care. The information pack also contained practical tools: guidelines for ACP conversations, a palliative care checklist and a method for discussing the wishes of people with ID [44].

The two training sessions formed the central component in the ACP program. Each session lasted two hours, with four weeks between the first and second session. The training was given by two ID physicians (project group members AV and AW) who have extensive experience of ACP among people with ID. The training sessions were held in the participants' own organization and were aimed at professionals with a range of backgrounds and tasks in the delivery of palliative care within a given organization. The idea behind this was to encourage closer cooperation between the different professionals within the organization. The training course also covered awareness of the different ways in which people deal with dying and what that means for communication, how to deal with ethical dilemmas and how to conduct discussions of wishes and needs. There was room for participants to air their own dilemmas and discuss cases. The participants were given homework assignments for the period between the two sessions.

The third element of the ACP program, the implementation interview, took place one month to three months after the second training session in the organization in question. One of the trainers (AW) travelled to the organization for the implementation interview. The interviews were held with a number of core members from the group that had attended the training sessions, along with other stakeholders in the organization such as managers. The aim of the interviews was to discuss progress in implementing and continuing ACP within the organization. Other issues discussed in the interview were whether and how ACP was integrated in palliative care and whether further work was needed on improving the competencies for ACP.

In Phase 5, an evaluation was performed to determine whether the ACP program had led to an improvement in the competencies of the professionals who had taken part and in their use of ACP in their daily practice. To this end, the participants completed a digital questionnaire six months after the second training session.

Materials

Professionals who took part in the ACP program were sent a digital pre-structured questionnaire to evaluate the program. Participants were asked (1) how they felt about their command of the ACP competencies (rated on a 3-point Likert scale: ‘insufficient’, ‘sufficient’ or ‘good’), (2) if they felt their ACP competencies had improved after the ACP program (response options: ‘yes’ or ‘no’), and (3) if they felt they had become better at applying their ACP competencies in their work (response options: ‘yes’, ‘no’ or ‘don’t know’). A draft version of the questionnaire had previously been tested within the project group and on two social workers to check the comprehensibility and validity of the content. Differences between physicians and other professionals were tested for statistical significance using Fisher’s exact tests (in STATA, version 15). For the purpose of the analysis, the responses to questions 1 and 3 were dichotomized by merging the responses ‘sufficient’ and ‘good’ (question 1) and ‘no’ and ‘don’t know’ (question 3).

Ethical Approvals

The research protocol was assessed and approved by the Medical Ethical Committee of Amsterdam University Medical Centre. Participation in the study was voluntary and all participants gave their informed consent. Research data were anonymized before analysis.

Results

Development of the ACP program

Based on the comprehensive study of the results from three sources (an international review, an analysis of patients’ medical files, and interviews in project phase 1), followed by discussions and reflections in the project group, the professional competencies (knowledge, attitudes and skills) that should be the focus of the ACP program were formulated. The co-design sessions (phase 2) with people with ID, relatives and professionals and consultations with educational experts (phase 3) were used to further develop the ACP program centred on these competencies. Table 6.2 provides a summary of the ten resulting competencies, the associated aspects and the specific sources showing the need for that competency. The form, content and competencies that each element of the ACP program covers is described in Table 6.3.

Table 6.2 Ten competencies and sources addressed by the ACP program

Competency	Knowledge (K), Attitude (A), Skill (S)	Source
1. Timely identification of the palliative phase	- Knows about palliative care (K) - Knows how to recognize the palliative phase (K) - Regularly assesses care needs (S) - Can recognize that someone with ID is in the palliative phase (S)	B
2. Spotting changes in symptoms, problems and care needs in the palliative phase at an early stage	- Knows about care needs and how they change in the palliative phase in people with ID (K) - Knows about tools for identifying symptoms, problems and care needs (K) - Recognizes when there is a reason to assess or reassess this (K) - Knows what follow-up steps should be taken in the event of a change in symptoms, problems or care needs (K) - Can assess symptoms, problems and care needs (S) - Makes changes to the care in the event of a change in symptoms, problems or care needs (S)	B
3. Communicating about wishes, agreements and care needs	- Knows that different people have different ways of communicating (K) - Is familiar with important communication techniques for ACP (K) - Can adjust own communication to allow for this (K) - Finds it normal to discuss future care (A) - Takes the initiative for ACP conversations in a timely manner with the people involved (S) - Regularly holds conversations about wishes, agreements and care needs, and any changes in these (S) - Applies conversational skills correctly; allows room for emotions (S)	A, B, C

- Table 6.2 continues –

- Table 6.2 continued -

Competency	Knowledge (K), Attitude (A), Skill (S)	Source
4. Reporting on wishes, agreements and care needs	- Knows how, where and what to report regarding ACP (K) - Finds it important to report on ACP (A) - Reports on wishes, agreements and care needs in the patient's - (medical) file (S)	B
5. Integrating ACP as a process in palliative care	- Knows the purpose and importance of ACP (K) - Knows how ACP should be deployed in the palliative phase (K) - Knows what tasks and responsibilities he/she has (K) - Finds ACP important in palliative care (A) - Applies ACP in palliative care as a process (S) - Takes the appropriate responsibility for his/her role in ACP (S)	A, C
6. Taking the wishes of the person with ID into account	- Knows how to involve people with ID in ACP (K) - Is aware of the person with ID's wishes (K) - Gives priority to the interests of the person with ID (A) - Consults with the people involved about whether/how to involve the person with ID in ACP (S) - Acts in accordance with the wishes of the person with ID (S)	A, B, C
7. Collaborating with the person with ID, relatives and other care professionals	- Knows who is involved with the person with ID (K) - Knows who he/she can go to for help or expertise (K) - Is in contact with the people involved and liaises with them (S) - Listens to and respects others (S) - Ensures or maintains a good relationship with the people involved (S) - Calls on the expertise of others where necessary (S)	B, C
8. Paying attention to ethical dilemmas in ACP for people with ID	- Knows what dilemmas play a role in the target group of people with ID (K) - Knows the legal rules (K) - Knows who can be approached for assistance if necessary (K) - Creates an open, calm, respectful environment in which any dilemmas can be discussed (S) - Places himself/herself in another's position (S) - Explains the situation clearly (S)	B, C

- Table 6.2 continues –

- Table 6.2 continued -

Competency	Knowledge (K), Attitude (A), Skill (S)	Source
9. Fostering expertise in ACP within palliative care	- Knows what he/she finds difficult about palliative care and about discussing it (K) - Knows where there are points for improvement in himself/herself and in the team (K) - Is keen to learn from others (A) - Evaluates own capabilities, collaboration and how ACP process went (S) - Adapts or improves own actions based on new knowledge or learning points (S)	C
10. Continuing ACP as a component of palliative care	- Communicates the importance of ACP to others involved in the care delivery (S) - Adopts and promotes a structured approach to ACP (S) - Brings in expertise, room and opportunity for training (S)	A

A = systematic review [21], B= analysis of patient’s files and interviews [29], C= in-depth interviews [18]

K = knowledge, A = attitudes, S = skills

Table 6.3 The form and content of the ACP program and the competencies that were covered

Program element	Content	Competency*
Information pack	Scientific literature about ACP, tools for identifying the palliative phase, guidelines for discussing ACP and a list of links to guides and tools	1,2,3,4,5,6,7,8
Training session 1	Participants learn that people have different ways of communicating; this helps them apply conversational skills appropriately. Participants learn to listen to others and respect them. Each participant evaluates their own capacities in the ACP process by hearing from someone else what they found good about a conversation; they can then adjust their own behaviour accordingly if necessary.	3,7,9
	Case study 1, motives: Paying attention to ethical dilemmas in ACP for people with ID. This case study is an exercise in experiencing how difficult it is to have to take decisions for someone else.	8
	Case study 1, conversation practice: Participants learn conversational skills and adjust their own communication accordingly. In this case study, participants practice cooperating with the person with ID, relatives and other professionals. As ACP is teamwork, it is crucial that everyone has a say, including the person with ID (if possible). The participants gain an understanding of the different interests and learn that the interests of the person with ID must be given priority.	3, 6, 7

- Table 6.3 continues -

- Table 6.3 continued -

	Reflection report as homework This report can deal with: - a conversation held by the participant about ACP or one aspect of ACP; - a case in which ACP was important. Depending on the topic, the participants learn to reflect on the different competencies that are required for ACP. They become aware of the competencies that they have and those they still need to acquire.	1,2,3,5,6,7,8,9
Training session 2	Discussion of reflection reports: The participants experience recognition and acknowledgement from the other participants. They reflect on their own actions and receive feedback from the other participants.	9
	Theory: How to compile files, tools, the importance of the person with ID and their involvement, conversation topics, collaboration, ethics and legislation.	1,2,3,4,6,7,8
	Conversation practice 1: Talking about wishes, agreements and care needs. Participants practice applying the key conversational skills for ACP in the correct manner. Given the capabilities of the person with ID in this case, the relatives represent the interests of the person with ID. They experience ethical dilemmas in taking decisions. In this case study, they work with the social workers and the physician to come up with a shared ACP goal. They listen to one another and respect one another's opinions and views.	3,6,7,8

- Table 6.3 continues –

- Table 6.3 continued -

Program element	Content	Competency*
	Conversation practice 2: Talking about wishes, agreements and care needs, in which the interests of the person with ID take priority. In this case study, participants work with all the parties involved, with due regard for the ethical dilemmas.	3,6,7,8
Implementation interview	Integration of ACP in palliative care and enhancing expertise	5, 9, 10

* See Table 6.2 for the competency associated with a given number

The sources show that it is important for professionals to recognize a need for palliative care in a person with ID (competency 1). They should also be aware that palliative care needs can change over time (competency 2). Not all professionals know what palliative care is and how to recognize this care need in people with ID. Professionals often do not act proactively or systematically in assessing changes in symptoms, problems and care needs. The various sources also suggest that professionals sometimes feel a reluctance to discuss the end of life and the care that will then be needed. Discussions about future care are often postponed until urgent problems arise or someone becomes terminally ill. It is important for professionals to be prepared to talk about wishes, agreements and care needs (competency 3).

Professionals also appeared to keep few records on ACP decisions in medical files; if anything was documented at all, it was usually only medical end-of-life decisions, such as a non-resuscitation policy. Conversations about wishes and agreements about psychosocial or spiritual care were not documented. It therefore seemed necessary to include the reporting of needs and wishes concerning future care and decision-making (competency 4) in the ACP program.

Professionals said they did not always know who was responsible for ACP, what their own role or task was and whether or when they should initiate ACP. ACP should be applied as a process, in which it is important to know which tasks and responsibilities belong to whom (competency 5).

It also became apparent that people with ID often were not involved in ACP. Professionals found it difficult to involve people with ID. In the in-depth interviews, people with ID said they would like to talk with people they know well about their illness and future care, and they would like to receive personal attention and time when ill. In order to align the delivery of care with the needs and wishes of people with ID, they must be involved in important decisions, depending on their capabilities and needs (competency 6).

Another finding was that a range of different professionals are involved in ACP. ACP turns out to be teamwork in which it is important to maintain close contact with all the relevant individuals. If the interests of the person with ID are to be served, everyone should be heard. The interviewed professionals said that is why it is important for professionals to communicate openly with one another, relatives and the person with ID, and to take time to prepare for future scenarios (competency 7).

It transpired that ethical dilemmas often play a role in decisions on what constitutes good care and quality of life for people with ID; this was something that therefore required attention (competency 8). It also became apparent that panic situations can arise if there is no one with palliative care experience in a team. There is therefore a need to evaluate ACP processes and learn from one another (competency 9).

Finally, it is important that professionals integrate ACP in day-to-day care. However, that is not yet standard practice. The lessons the professionals learn in the program need to become embedded in their daily practice (competency 10).

Evaluation of the ACP program

After the implementation, the ACP program was evaluated (phase 5). Most respondents (88%) said they had provided palliative care to one or more people with ID since the training course. When asked about the extent to which professionals now mastered the competencies required for ACP, most (>85%) replied that they mastered the competency in question sufficiently or good (see Table 6.4). Two competencies were estimated as less strong: paying attention to ethical dilemmas (competency 8; 21% felt they were insufficiently competent) and ability to reflect on ACP (competency 9; 19% felt they were insufficiently competent).

Table 6.4 Ratings of perceived effects of the ACP program on competencies for palliative care

	Professionals (n=42) who felt their command of this competency was sufficient or good	Professionals who felt their command of this competency had improved as a result of the ACP program		Professionals who said they were better able to apply this competency in daily practice after following the ACP program	
Competencies		Physicians (n=19)	Non-physicians (n=23)	Physicians (n=19)	Non-physicians (n=23)
1. Timely identification of the palliative phase	93%	47%	74%	53%	70%
2. Spotting changes in symptoms, problems and care needs in the palliative phase at an early stage	98%	26%*	83%*	37%*	83%*
3. Communicating about wishes, agreements and care needs	93%	74%	83%	58%	83%
4. Reporting on wishes, agreements and care needs	88%	37%*	74%*	26%*	74%*
5. Ensuring that ACP is part of the palliative care for the person with ID#	88%	74%	74%	74%	83%

- Table 6.4 continues –

- Table 6.4 continued -

	Professionals (n=42) who felt their command of this competency was sufficient or good	Professionals who felt their command of this competency had improved as a result of the ACP program		Professionals who said they were better able to apply this competency in daily practice after following the ACP program	
Competencies		Physicians (n=19)	Non-physicians (n=23)	Physicians (n=19)	Non-physicians (n=23)
6. Taking the wishes of the person with ID into account	93%	58%	70%	53%	78%
7. Collaborating with the person with ID, relatives and other care professionals	95%	47%	65%	53%	74%
8. Paying attention to ethical dilemmas in ACP for people with ID	79%	63%	78%	42%*	87%*
9. Reflecting on the use of ACP in palliative care and considering what went well and what could be improved[#]	81%	58%	87%	42%*	74%*
10. Communicating the importance of ACP in your team or organization[#]	86%	58%	87%	58%	70%

Significant difference between physicians (n=19) and non-physicians (n=23), Fisher's exact test, *p<0.05.

[#] The wording for this competency differs from that of the competency as formulated in the ACP program (see Table 6.2).

For all competencies, at least half of all participants (range 57–79%) felt their level of competence had improved thanks to the ACP program. At least 70% of the participants thought they were better able to talk about wishes, agreements and care needs (competency 3), applied ACP more often in the delivery of care (competency 5), paid more attention to dilemmas (competency 8), were better able to reflect on ACP (competency 9) and were better at communicating the importance of ACP (competency 10). The participants saw least improvement in the identification of symptoms, problems and care needs (competency 2), reporting in writing (competency 4) and collaboration with people with ID, relatives and the other professionals involved (competency 7). In general, an improvement in a competency also meant that the professionals felt they were using it more often and more effectively in their daily care of the people with ID.

Physicians saw less improvement in two competencies than non-physicians (social workers, nursing staff and other professionals). The physicians also saw less change in the application of the competencies in their daily care delivery than the non-physicians (see Table 6.4).

Discussion

This article described the development, implementation and evaluation of an ACP program in palliative care for people with ID. Results from various sources (an international review, an analysis of patients' medical files, and interviews) were used in the development of the ACP program; the project group drew on them to come up with ten important competencies that professionals should have for ACP with people with ID. These sources and other previous research [22, 45] showed that professionals have difficulty recognizing a need for palliative care, they feel they lack the necessary expertise to use ACP in the palliative phase for people with ID, and they find it difficult to talk about the end of life. The competencies addressed in the ACP program were geared to resolving these problems. Co-design sessions with people with ID, relatives and professionals and consultations with educational experts were used to develop an ACP program centred on the competencies. The program had three main elements: an information pack, two 2-hour training sessions, and an implementation interview.

This ACP program is unique. In our international review, we did not encounter a single comparable program in palliative care for people with ID [21]. It is important to have a dedicated ACP program for professionals caring for people with ID, as assessing the wishes and needs of this group is often a challenge. People with ID often have complex, multimorbid and/or psychosocial care issues and limited communicative capacities, which makes it particularly difficult to determine what would constitute good care [10, 22, 25-27].

Many professionals do not feel competent and confident enough to provide this group with palliative care [31-33], which can make it difficult to discuss ACP. This ACP program is a response to the specific questions of professionals concerning communication with incurably ill people with ID and their relatives, and how best to assist them. Given that the content of the ACP program is based in part on our international systematic review, the results of the current study are relevant not just for Dutch care professionals. The professional competencies, and the knowledge, attitudes and skills that were identified apply in a broader international context and could offer a way to improve ACP in palliative care for people with ID.

The in-depth interviews and co-design sessions with people with moderate ID show that they are willing to be involved in ACP, in a manner appropriate to their capacities and needs. Previous studies had already shown that people with mild ID want to be informed about their state of health, to be listened to and to be asked more about their care wishes [17, 38, 46], and that now seems to apply to people with moderate ID as well.

The ACP program was evaluated using a digital questionnaire that participants completed six months after the training ended. The response rate of 79% suggests that the questionnaire gives meaningful information about the results of the program [47]. The training sessions focused mainly on communication and paying attention to ACP. The professionals felt their competencies had improved in those areas. After the ACP program, professionals saw most improvement in their communication with people with ID about their wishes, agreements and care needs, and in making sure that ACP is a standard element in the palliative care for people with ID.

A relatively high proportion of participants said they were competent in collaborating with the person with ID, relatives and other care professionals and in the early identification of symptoms, problems and care needs. These were competencies where improvement was seen least often after the ACP program; this could be because of the relatively high level of (perceived) competence at the outset. It is striking that previous research had shown that professionals often find it difficult to identify care needs [22, 45]. One explanation for this discrepancy could be that precisely those professionals who were already skilled in identifying changes to care needs in people with ID, felt the need for training in how to deal with this and therefore opted to take part in the ACP program.

There was also relatively little improvement in the competency of reporting on wishes, agreements and care needs after following the program. Non-physicians, such as social workers and nursing staff, were more likely to see an improvement in the reporting competency than physicians. This could be because physicians are responsible for the medical policy and are already accustomed to reporting on this in the patient's file. Physicians conduct the conversations in which the medical policy and end-of-life decisions are discussed in contrast to social workers and nursing staff who may have informal conversations about future care needs. The policy is less clear on such conversations and when any decisions should be documented. We recommend that informal conversations should be documented if they cover wishes and needs for future care. At present, there is a great deal of variation between organizations in their electronic or paper reporting systems. Preferably, reporting systems would have a section (tab) on ACP as this would remind professionals of the need to document wishes, agreements and care needs [13].

Strengths and weaknesses

A strength of the ACP program is that it was developed in part through a co-design process with people with ID, relatives and professionals. The ACP program was implemented in six different ID care organizations. The evaluation suggests that the program boosts professionals’ perceived competencies and promotes the use of ACP in practice.

One weakness is that no baseline measurement was carried out for the evaluation of the ACP program. It is therefore not known how the professionals assessed their competencies prior to the ACP program. The evaluation was also subjective as the participants assessed their own level of competence and may have given socially desirable answers. Consequently, while the evaluation provides indications, it does not yet give ‘hard’ evidence that this ACP program actually leads to ACP being used more often and more effectively in palliative care for people with ID.

Conclusions

As far as is known, the ACP program that was developed is the first ACP program for professionals delivering palliative care to people with ID. Ten competencies were identified as relevant for professionals based on a systematic review, examinations of patients’ files and interviews. These professional competencies were incorporated in an ACP program developed in a co-design process with people with ID, relatives and professionals and in collaboration with educational experts. The program was successfully implemented in six ID care organizations.

Professional who took part in the ACP program experienced an improvement in the competencies that are required for using ACP in palliative care for people with ID, in particular in the competencies focusing on communication and using ACP as a standard element in palliative care. Reporting on wishes, agreements and care needs remains a point for concern, particularly among social workers and nursing staff. The differences between physicians and non-physicians regarding ACP competencies and improvements in those competencies might indicate a need to tailor aspects of the training sessions further to suit the different disciplines and roles of the participants. The program should be developed further and evaluated among people with ID and relatives to measure the effects in practice.

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Summary and general discussion

Advance Care Planning

in palliative care for people with intellectual disabilities

Advance Care Planning (ACP) is defined as: “the ability to enable individuals to define goals and preferences for future medical treatment and care, to discuss these goals and preferences with family and health-care providers, and to record and review these preferences if appropriate” [1]. The idea is that this proactive approach will ensure that the care provided is in line with a person’s wishes and needs, resulting in a better quality of life.

Despite the importance of ACP, little is known as yet about ACP in the care for people with ID. This thesis aims to obtain a better understanding of ACP in palliative care for people with ID. In this final chapter the main findings are summarised and reflections on important findings are discussed. This is followed by methodological considerations, in which the strengths and limitations of this thesis are examined. At the end of this final chapter, implications for practice and future research are discussed.

The thesis introduction (*Chapter 1*) gives information about the care needs of people with ID in the Netherlands. Furthermore, a definition is given of palliative care and the importance of ACP is discussed.

An intellectual disability is a disability that occurs before the age of eighteen and is characterised by limitation in cognitive functioning, often defined as an intelligence quotient (IQ) score of less than 70, and limitation in adaptive behaviour (conceptual, social and practical skills). People with ID often have complex care needs throughout their lives. On average, they experience more chronic illness than the general population and they are more likely to have multiple conditions (multimorbidity). Even so, their life expectancy has increased in recent decades. As a result they, in line with the general population, are at increased risk of developing life-threatening conditions that are relatively common among older people, such as cardiovascular diseases, dementia and cancer. This new development means that there are now more people with ID who live to a greater age and experience a longer period of chronic disease. There is therefore a growing need for palliative care for people with ID.

Palliative care is care that that improves the quality of life of patients and their families facing the problem associated with life-threatening illness or vulnerability, 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 (the Dutch Palliative Care Quality Framework, 2017). There are a number of challenges in providing palliative care for people with ID:

- Firstly, people with ID often have only limited understanding of their state of health and they are not always able to find the words to describe their problems, wishes and needs.
- A second challenge is related to the first and concerns the recognition of the need for palliative care. Because of limitations in the communication abilities of people with ID and the limited understanding of their illness, it is difficult for professionals to proactively recognise a need for palliative care.
- A third challenge is that important decisions are particularly likely to have to be taken for people with ID because they are a group with complex care needs. For example, decisions may need to be taken on whether or not to provide life-sustaining treatment, whether or not to admit them to hospital and whether or not to move them to a different care setting. People with ID are often unable to take these decisions on their own and they therefore depend on their relatives and care professionals to make their wishes known.
- A fourth challenge is that professionals working in care organisations for people with ID often do not have medical or nursing backgrounds and they were taught little or nothing about palliative care in their basic training. As a result, they know little about palliative care.
- A fifth challenge is the lack of a palliative care policy in many care organisations for people with ID. For example, organisations often have no guidelines or protocols for how to take decisions about the end of life, and care professionals often do not know what role they should play in palliative care.
- The lack of proper collaboration with care organisations specialised in palliative care, such as hospices or nursing homes, constitutes a sixth and final challenge.

These challenges make it especially important to incorporate palliative care in general, and ACP in particular, into the care for people with ID. The first part of this thesis describes the state of the art concerning ACP in the care for people with ID. The second part describes the development, implementation and evaluation of an ACP programme for people with ID that is based on this. The programme draws on five sub-studies. The main findings from these sub-studies are set out below, linking them to the key research questions for this thesis.

The first key research question was: **What is already known in the scientific literature about the use, content and effects of ACP for people with ID in need of palliative care?**
Previous research into ACP has focussed on elements of ACP but not on the broad ACP process in palliative care for people with ID. The studies that were found also failed to consider the use of ACP in practice and the effects of ACP for this specific group. These findings come from the systematic literature study presented in *Chapter 2*.

A total of fourteen relevant studies were found in four different literature databases (PubMed, PsycINFO, Embase and CINAHL). A majority of the studies (n=8) focussed on decision-making, specifically medical end-of-life decisions such as Do-Not-Attempt-Resuscitation (DNAR) orders, agreements about hospitalisation admissions and intensive care treatments that are potentially life shortening. These studies indicated that such decisions were taken more often for people with ID compared to people with other disabilities (sensory, physical and psychological) or people in the general population. It also transpired that the people with ID themselves were often not involved in these decisions.

Five of the fourteen studies looked at the organisational policy on palliative care and ACP. The studies showed that organisations for people with ID often did not have a written policy on ACP. Other aspects of ACP that were covered in the studies that were found were communication around advance directives (n=1), inclusion of family (n=1), documentation of wishes for future care (n=1) and the start of ACP (n=1).

The second key research question was: **How does ACP currently take place within organisations for people with ID requiring palliative care and how are the wishes of people with ID taken into account?**
Chapter 3 shows that ACP is not common within care organisations for people with ID. Any agreements on future care are mainly concerned with medical decisions, and the people with ID themselves are often not involved. These results are based on the analysis of the medical files of fifteen people with ID in the final phase of life and fifteen people with ID who had recently died after a period of illness. Furthermore, interviews were held with professionals who were involved in their care (physicians or daily caregivers) and relatives.

Although the term ‘palliative care’ was not mentioned in seven files, 29 of the 30 files did contain agreements concerning future care. These agreements mainly concerned medical decisions such as resuscitation, future treatments or admission to hospital. Two of the 30 files noted an agreement about the preferred location for care at the end of life. The agreements were often noted down only a short time before the death of the person with ID: in half the cases of people with ID who had died, decisions were recorded less than one month before their death.

The medical files showed that decisions about future care were made jointly by the physician and relatives. Other professionals were also often involved, such as daily caregivers, behavioural specialists, spiritual caregivers or medical specialists from the hospital. Only four of the 30 files contained evidence that the person with ID had also taken part in the discussions about these decisions. In the interviews, 22 of the 30 professionals stated that it was not possible to involve the person with ID in ACP. The reasons they gave for this were that the person with ID did not have sufficient cognitive capabilities or was unable to oversee the consequences of decisions that had to be made, or that it would be too much of a burden for them to be involved.

The third key research question was: **What do people with ID, relatives and professionals perceive as important for ACP in palliative care for people with ID?** Offering care tailored to needs of the individual with ID, good teamwork, and taking and giving sufficient time turn out to be important elements for ACP in the palliative care for people with ID, as is described in *Chapter 4*. The analysis of fifteen in-depth interviews with people with ID (n=5), their relatives (n=8) and professionals (n=7) revealed the following three themes and six subthemes:

Theme 1) Tailored care. The subthemes that were revealed were ‘standing close’ and ‘connecting with the person’. The form an ACP process should take depended on the individual concerned. It was essential to have a good relationship with the person with ID and to know them well.

Theme 2) Teamwork. ‘Deciding together’ and ‘trusting each other’ were important subthemes here. Everyone who is important to the person with ID should be involved in ACP. Trusting one another and being open and honest with one another were also considered to be important.

Theme 3) Taking and giving time. ‘Preparing and thinking ahead’ and ‘making room for emotions’ were mentioned here as subthemes. Wishes and needs concerning possible future situations should proactively be discussed in order to create a calm situation at the end of life. There should be room for expressing emotions because the topics being discussed in ACP can be difficult for people with ID and their relatives.

The people with ID who were interviewed said that they found it difficult to take decisions themselves and they want to be helped by people who know them well. The interviewed relatives saw a role for themselves as the representative of the person with ID in ACP, in contributing to decisions and in signalling needs and preferences. The professionals emphasised that their role in ACP should be to coordinate, provide information and ensure collaboration.

The fourth key research question was: **What facilitators and barriers influence the implementation process and sustainment of innovations in palliative care for people with ID?**

Support from the organisation and the presence of an opinion leader who is responsible for using the innovation are important facilitators for implementing and sustaining palliative care innovations for people with ID. These findings come from an evaluation study of nine implementation projects in care organisations for people with ID, implemented as part of the National Quality Improvement Programme for Palliative Care.

The study, which is described in *Chapter 5*, combined different research methods such as analyses of the project plans and final reports, interviews with project managers and other relevant professionals, and a questionnaire for the project managers. The following palliative care innovations were implemented in these projects:

1) The introduction of a specialist palliative care consultant whose task was to support professionals during palliative care (four projects).
2) The ‘Signal Box’, a tool for identifying and proactively discussing palliative care needs (three projects).
3) ‘Dying Your Own Way’ (‘STEM’ in Dutch: two projects), an innovation designed to accelerate expertise, to create awareness of the diversity of patients’ wishes and needs at the end of life, and to improve professionals’ ability to communicate proactively with patients and relatives about wishes and needs at the end of life.

Training for professionals, collaboration with other organisations and activities aimed at encouraging the use of the innovation and spreading it across the organisation turned out to be important implementation strategies. Factors that facilitated the implementation were often related to the characteristics of the organisation, such as commitment from management or sufficient financial means and the availability of materials. The innovations were not originally developed for the specific target group of people with ID. Consequently, in some of the implementation projects, the innovation was developed further so that it would be better suit to this target group.

When asked one to three years after the implementation project had finished, the project managers said the innovation was still being used within the care organisations. However, five of the nine project managers did have doubts whether the innovation was sufficiently sustained for the longer term. They said the factors affecting this were organisational characteristics such as the support from management, access to resources such as money, time and materials, and the integration of the innovation in existing workflows.

The fifth and final key research question was: **How is an ACP programme for professionals caring for people with ID developed, implemented and evaluated?** The ACP programme, which was based on ten important ACP competencies for professionals, was developed using a co-design process with people with ID, their relatives and professionals. When the ACP programme was implemented in the care organisations participating in the project, it led to an improvement in the competencies of the professionals (based on their own assessment). This is described in *Chapter 6*.

Based on the results of the systematic review, the analyses of the medical files and the interviews (see the descriptions above of *Chapters 2 and 3*) and the important elements of ACP (*Chapter 4*), ten competencies were formulated that professionals should have in order to be able to use ACP in palliative care. These ten competencies were: identifying palliative care needs and symptoms, communicating, reporting wishes and decisions, integrating ACP in palliative care, taking the wishes of the person with ID into account, collaborating, paying attention to ethical dilemmas, fostering expertise within ACP and sustaining the use of ACP in palliative care.

In a co-design process with people with ID, relatives, professionals and education experts, the competencies were turned into an ACP programme for professionals who provide palliative care to people with ID. Two co-design teams were formed to develop the programme, one consisting of relatives and professionals and the other consisting of people with moderate ID. The ACP programme that was developed was then implemented in six care organisations for people with ID. A total of 58 professionals (including physicians, daily caregivers, nurses, behavioural specialists, and spiritual caregivers) took part in the programme, which had three components: an information pack, two 2-hour training sessions and an implementation interview focusing on the integration and sustainment of the ACP programme.

The programme was evaluated by asking the professionals who took part to complete an online questionnaire six months after the training. The evaluation showed that the professionals felt that taking part in the programme had improved their ACP competencies. At least 70% of the participants said that their communication skills had improved, that they used ACP more often when providing care, that they paid more attention to dilemmas, that they were better able to reflect on ACP and that they had become better in conveying the importance of ACP. The professionals also said that they used these competencies more often and more effectively in the daily care for people with ID. There was less of an improvement in signalling symptoms and care needs, reporting wishes and agreements, and collaborating with the person with ID, their relatives and other professionals.

Reflections on the findings

The use, content and effects of ACP for people with ID

As became apparent from the systematic review presented in *Chapter* 2 of this thesis, not much was known previously about the use, content and effects of ACP for people with ID. There were no indications from previous research that ACP occurs frequently in palliative care for people with ID. In addition, no previous studies were found that focussed on the use of ACP as a broad and complex process. Instead, each of the studies that were found only focussed on one element of ACP. The scarcity of relevant studies is striking since it is well known from research in other populations that ACP has clear benefits, such as improvements in the quality of communication, quality of care and quality of life, and reductions in unwanted hospitals admissions [2, 3].

In line with findings from the systematic review *(Chapter 2),* the study presented in *Chapter* 3 indicated that ACP in its broadest sense was not common practice as yet in Dutch ID care organisations. Often only the specific aspect of medical end-of-life decisions, in most cases do-not-attempt resuscitation (DNAR) orders, was addressed in the medical files of people with ID (*Chapter 3*). However, ACP should go beyond discussing DNAR orders. For example, discussions about the place where palliative care is delivered (whether people prefer to stay in their own home environment or are open to move to a hospital or hospice), or what kind of pleasant activities a person would still want to do at the end of life, are also important [4].

Difficulties with identifying and interpreting palliative care needs and initiating ACP

The analysis of medical records of people with ID described in *Chapter 3* showed that when end-of-life decisions were made, this was mostly in response to acute situations or for someone who was already terminally ill. This has also been found in other research, and shows that proactively discussing wishes and needs with people with ID and/or their relatives was often hampered by a late diagnosis of the life-limiting illness [5, 6].

Late diagnosis might be due to the complexity of identifying and interpreting symptoms and problems in people with ID. More often than in the general population, the nature of the underlying disease or physical decline of people with ID is unknown, and palliative care needs are difficult to recognise [7]. Also, professionals do not always recognise non-verbal signs or the needs of people with ID or they see symptoms as part of the disability [5]. Especially when there is a trajectory with a prolonged gradual decline rather than a diagnosis of a specific life-limiting illness (a situation that is common in frail elderly people), identifying the need for palliative care and deciding when to start ACP for people with ID can be particularly challenging [8].

Taking wishes of people with ID into account in ACP

This thesis showed that the wishes of people with ID about their end-of-life care are often not discussed (*Chapters 2, 3 and 4*). Other studies performed in the United Kingdom, Ireland and Australia also showed that professionals found it difficult to talk to people with ID about death and dying and give them support [9, 10]. The study by Tuffrey-Wijne et al. (2020) showed that only half of people with ID who were terminally ill were told about their illness, and less than 20% were told they would die [11]. If an attempt to talk about end-of-life care was made, professionals only discussed funeral wishes; wishes concerning preferred carers, place of care, spiritual needs or will-making were often not discussed [12]. The findings of this thesis confirm that some professionals in ID care do not know how to initiate discussions about wishes and options for future care, and sometimes avoid talking about fatal illnesses and the impending death of a person with ID (*Chapter 3*).

This thesis also showed that, especially of people with more severe ID, decisions for future care are often taken by their relatives and professionals. Previous studies indicated that those who are closest to the person, such as family members or trusted professionals or friends, might be able to identify the person’s preferences if that person is unable to express them [13, 14]. Deciding what is in the best interests of a person who cannot decide for themselves is, however, a challenge. Especially because there is often a relatively large group of professionals and relatives involved in the care for a person with ID who may have different or even conflicting views [15]. Moreover, not discussing the wishes of a person with ID in ACP is problematic because decisions will then be based on the values of others and may not reflect the values of the individual with ID [16]. This puts people with ID at risk of having their wishes dismissed or misunderstood at the end of their lives [13].

Importance of involving people with ID in ACP

Although relatives and professionals were often of the opinion that people with ID were not able to be involved in ACP (*Chapters 3 and 4*), participants with moderate ID who took part in the study (*Chapter 4 and 6)* did stress their wish to be involved. They wanted to be informed about their health situation and were able to talk about their needs and preferences, as has also been shown in studies about end-of-life care involving people with mild intellectual disabilities [4, 15, 17]. Regarding decision-making, people with ID would like to be informed and make decisions together with the people who know them best. This is known as supported decision-making, which refers to the process of supporting people in making decisions whose decision-making ability may be impaired, by taking the time to listen and communicate, and in this way promote autonomy and avoid the need for substitute decision-making [18, 19].

When one participant with ID was asked why they wanted to be involved in ACP, they gave a very clear response: *“It’s my life”.* This wish of people with ID to be involved in ACP is in accordance with the European White Paper on end-of-life care for people with ID [20]. The White Paper states that an incapacity to make decisions about end-of-life care, or certain aspects of this, should not be assumed beforehand. On the contrary, professionals should assume that a person with ID has the capacity to make decisions concerning care unless this is demonstrated not to be the case. The United Nations Convention on the Rights of Persons with Disabilities also stresses the importance of respecting the will and preferences of the person who needs support [21]. In addition, the Caring at the End of Life position statement of the American Association on Intellectual and Developmental Disabilities (AAIDD) mentions respect for autonomy as one of four major principles and notes that professionals should always attempt, as much as possible, to discover the wishes and needs of the person with ID and honour those wishes [22].

Important elements for ACP in palliative care according to people with ID, relatives and professionals

Several elements are important when providing ACP for people with ID, as is shown in the studies presented in *Chapters 4 and 6*. It was found to be important for people with ID to be supported by familiar people in ACP who are close to them and know them well. Professionals and relatives should connect with the person with ID and focus on what is of importance to the person as they approach their end of life. In the literature, this is also referred to as 'person-centred care'. The study by McNamara et al. depicts a model of person-centred care for people with ID at the end of life which places the person with ID at the centre, includes relatives as partners in planning and aims to improve the person's quality of life [23]. ACP should also be considered as a person-centred process placing the wishes of people with ID front and centre when providing palliative care.

A second important element for ACP that has been revealed in this thesis is working together with everyone who is involved in the care of a person with ID. The results of this thesis indicated that collaboration between professionals, relatives and the person with ID often went well (Chapters 3 and 4) and relatives appreciated the warm and intimate care professionals provided for their loved one with ID. Other studies have indicated that as death nears, the person with ID becomes increasingly dependent on others and this requires an intensified caring relationship between the individual with ID, their relatives and professionals [24, 25]. Previous research showed that it is especially important for professionals providing palliative care to develop a bond with relatives and inform and involve them in the care of the person with ID [4, 13, 23, 24]. This bond and the notion of 'being there' helps in establishing honest, realistic and open communication about possibilities for future care, in making decisions together, and finally in ensuring a good death [26].

Taking and giving time is a third element that was found to be important for ACP (*Chapter 4*). In accordance with the literature, it is preferable to discuss ACP for people with ID as early as possible, once the individual's state of health worsens or becomes more fragile [1, 4]. In this way, decisions can be made in a stable and calm situation and enough time can be spent on a sensible discussion with everyone involved. Research focussing on end-of-life decision-making for children with severe developmental disabilities also showed that parents underlined the importance of having sufficient time and obtaining sufficient information during the decision-making process [27]. Therefore, professionals should proactively inform people with ID and/or their relatives about changing needs and symptoms, discuss wishes and options for future care and treatment and make decisions together with them.

Influencing factors for implementation and sustainment in ID care

The results of this thesis showed that factors related to the organisation, such as the involvement and support of managers, the availability of sufficient financial means and materials and the integration of the innovation in existing workflows, are important for the implementation and sustainment of palliative care innovations such as tools to identify palliative care needs or ACP innovations and related training for professionals (*Chapter 5*). Other important influencing factors for the implementation and sustainment of innovations in ID care identified in this study that were also found in implementation studies in other health-care settings were the quality and applicability of the innovation and the presence of an opinion leader who is responsible for the use of the innovation [28, 29].

Furthermore, some influencing factors for the implementation of palliative care innovations were found that were related to the specific setting of ID care organisations (*Chapter 5*). Firstly, it is important to include training for ID care professionals as part of the implementation process of palliative care innovations. This is especially important because the majority of nurses and social workers who work in ID care organisations feel they have not had sufficient training to provide palliative care. The need to improve expertise and to develop appropriate end-of-life care training for professionals in ID care has also been highlighted in other research [30-34]. Without training or experience in providing palliative care, professionals might feel insecure or not competent to take care of a person with ID approaching the end of life, which requires a shift from a focus on activating the person with ID towards a focus on comfort care and symptom relief [24, 34]. It is therefore important to increase their knowledge and skills with regard to palliative care and ACP.

Cooperation with professionals and experts working in other ID care organisations or other kinds of organisation, such as hospices, hospitals or nursing homes, is another facilitating factor for the implementation of palliative care innovations in ID care (*Chapter 5*). Previous studies also showed that cooperation, for example in exchanging research and expertise, adapting innovations and developing policies, is important in facilitating high-quality palliative care for people with ID [35, 36]. In the study by Grindrod & Rumbold (2017), an organisational model is developed for collaboration between disability services and palliative care services; it gives structural, cultural and practical solutions for meeting the consensus norms of palliative care for people with ID [20, 37].

The development of an ACP programme for professionals in ID care

Based on the systematic review *(Chapter 2)*, examinations of patients' files and interviews *(Chapters 3)* and the important elements of ACP *(Chapter 4),* ten competencies for professionals involved in ACP for people with ID were identified in this thesis and include taking the wishes of the person with ID into account, and communicating and reporting about wishes, needs and decisions (*Chapter 6*). These competencies align closely with the recommended roles and tasks for ACP found in an international Delphi study of the definition and recommendations for ACP. This Delphi study indicated that health-care professionals should adopt a person-centred approach when engaging in ACP discussions, have the necessary communication skills and document discussions in the patient's file [1].

The ACP programme described in *Chapter 6* is, as far as is known, the first ACP programme in the Netherlands for professionals providing palliative care to people with ID. The programme consists of an information pack, two 2-hour training sessions and an implementation interview. To increase the competencies of the participating professionals, the training sessions provided information about ACP in palliative care and included role play in which participating professionals practiced how to discuss ACP and initiate conversations regarding wishes for end-of-life care. In successful ACP innovations in other care settings, education, training and discussion are often combined [3]. A systematic review of ACP innovations in nursing homes also showed that the provision of information regarding ACP, training for professionals and incorporation of ACP as a standard element in palliative care were important in encouraging the use of the innovation [38].

Methodological considerations

Strengths and limitations of this thesis

One strength of this thesis is that research data was gained from a variety of sources, such as an international systematic review *(Chapter 2)*, medical files of people with ID *(Chapter 3)*, interviews with people with ID, relatives and professionals *(Chapters 3 and 4)*, data from the evaluation of the National Quality Improvement Programme for Palliative Care *(Chapter 5),* and co-design sessions with people with ID, relatives and professionals *(Chapter 6)*. The combination of multiple data sources and methods yields rich information about the state of the art of ACP in palliative care for people with ID and the development, implementation and evaluation of ACP in palliative care for people with ID. Moreover, based on these sources, ten professional competencies needed for ACP were formulated and incorporated in an ACP programme. Therefore, the ACP programme offers a way to improve ACP competencies of professionals providing palliative care for people with ID.

A second strength of this thesis is the involvement of people with ID in the development of the ACP programme. Including people with ID as participants in research is increasingly popular, but there is still limited incorporation of the perspectives of people with ID in studies about the end of life [17, 39]. In this thesis, people with ID were asked about their views on and experiences with ACP and they were directly involved in the development of the ACP programme through co-design sessions. Other studies have also considered it useful to hear the views of people with ID and have given them an opportunity to inform research while supporting one another by sharing their experiences [15, 17, 39].

Thirdly, the incorporation of multiple perspectives in the studies can be considered as a strength of this thesis. Besides people with ID, relatives and professionals were also involved in interviews about ACP *(Chapters 3 and 4)* and in the co-design sessions to develop the ACP programme *(Chapter 6)*. The co-design process followed the principles of Experience-Based Co-Design (EBCD) [40, 41] and was spread over a period of three months in which the team met four times. Although partnership with professionals who make use of the innovation is not uncommon [41-43], patient and family involvement in developing palliative care innovations is still rare [44]. This thesis showed that when people with ID, relatives and professionals are involved in the development of an innovation, different views can be taken into account and the innovation may better fit their wishes and needs.

A fourth and last strength is that the ACP programme also took account of facilitating factors and barriers for the implementation and sustainment of palliative care innovations in ID care organisations, as presented in *Chapter 5*. The previous experiences with the National Quality Improvement Programme for Palliative Care [45, 46] offered important insights that were used in the ACP programme; this increases the probability that what has been learned will actually be applied in practice. Moreover, because of the importance of teamwork in providing palliative care in line with the wishes and needs of the individual with ID, the ACP programme was aimed at professionals with a variety of backgrounds (physicians, nurses, social workers and behavioural experts). In this way, cooperation between different professionals was encouraged within the organisations.

Some limitations of the studies described in this thesis should also be noted. First of all, there are limitations regarding the generalisability of the study findings. The research was performed within residential care facilities specialised in care for people with ID *(Chapters 3, 4, 5 and 6)*. The findings may therefore not apply to individuals with ID who live at home independently or with their family. Also, although participants with ID included in the studies did have an understanding of life-limiting illnesses and the dying process and had previous experience of being seriously ill or hospitalised, they were not at the end of life themselves (*Chapters 4 and 6*). As such, their views may differ from people with ID who are receiving end-of-life care.

A second limitation concerns the fact that in the interviews described in *Chapters 3 and 4,* the relatives of people who were satisfied with the care might be over-represented. Relatives who were not satisfied or less involved in end-of-life care might be less open to participation in such research, which possibly caused a selection bias. Moreover, physicians from the relevant intellectual disability care organisation functioned as recruiters in our study. The physicians informed potential participants about the aim and content of the study and asked them to sign an informed consent form if they wished to participate. As a consequence, in some cases, the number of people approached and what proportion agreed to participate, remained unknown to the researcher. This presents an additional limitation for judging the generalisability of the samples included in this thesis.

The retrospective character of some of the questions in the interviews presented in *Chapter 3* is a third limitation of this thesis. Professionals who had cared for people with ID who had since died sometimes found it difficult to answer the questions about agreements or decisions that were made concerning future care, because they could no longer recall all aspects of the ACP process in the cases addressed.

An additional limitation concerns the use of written data sources in this study. In *Chapter 3*, the medical files of people with ID were examined rather than their daily care files. It is known that the process of end-of-life decisions and the people involved are often not recorded or only briefly noted down in patients' medical files [47]. It therefore could be the case that ACP discussions occurred more frequently than appeared from the analysis of patients' medical files. In *Chapter 5*, a document analysis was conducted of project plans and final reports on implementation projects concerning palliative care innovations in ID care organisations. The project reports were written by the project managers, and therefore these documents may present a more positive image of plans and achieved activities regarding implementation and sustainment than was the case in reality.

A fifth and final limitation is that no baseline measurement was carried out for the evaluation of the developed ACP programme *(Chapter 6)*. Therefore the professionals' assessment of their competencies prior to the ACP programme is not known. The evaluation also did not include patient outcomes; it only considered the subjective evaluations and outcomes of the professionals. After taking part in the ACP programme, the participating professionals assessed their own level of competence and they may have given socially desirable answers or have overestimated their competencies. Consequently, further study is needed to determine whether the ACP programme actually leads to ACP being used more often and more effectively in palliative care for people with ID.

Implications for practice

Embed palliative care within organisational policies and integrate ACP in workflows and systems

As the population of ageing people with ID continues to grow, professionals working in ID care organisations will be more likely to have to deal with people with ID who need care or support at their end of life. ID care organisations should therefore acknowledge the importance of palliative care and should develop a vision and policy around palliative care, informing professionals about palliative care and ACP, and their role in this process. This policy should also be clearly communicated within the organisation and acted upon.

Moreover, since ACP is important in improving the quality of palliative care, professionals should proactively discuss needs and preferences for future care with people with ID and/or their relatives. As this thesis showed, ACP should be regarded as an inherent component of palliative care. This can be established by integrating ACP in the organisation’s existing workflows and systems, for example by discussing ACP as a standard agenda item in multidisciplinary team meetings, by incorporating ACP in electronic or paper reporting systems, or by embedding ACP training within the educational options offered by the organisation.

Train professionals in palliative care and ACP for people with ID

This thesis confirmed the findings from other studies that professionals in ID care often feel insecure in providing palliative care and talking about the end of life. Professionals who provide daily care for people with ID in the Netherlands were often educated as social workers or nurses. Generally, they have had little or no training in delivering and talking about palliative care to people with ID. Since ID care professionals have an important and coordinating role in providing palliative care and should take the initiative for ACP discussions, it is necessary that training and tools are provided for professionals in ID care in order to increase their expertise and experience in palliative care and ACP. Training should cover physical aspects, such as assessing pain and other symptoms, but should also focus on the psychosocial and spiritual aspects of palliative care and ACP, such as talking about wishes regarding preferred carers and place of care, and it should pay attention to ethical dilemmas.

The ACP programme developed in this thesis focussed on ten important competencies for ACP in order to improve the knowledge, attitude and skills of professionals caring for people with ID. The central component of the ACP programme consists of two 2-hour training sessions, given by two experienced ID physicians [48]. Three months after the training, an implementation interview about the use and sustainment of ACP is held within the ID care organisation. Participants also receive an information pack, which lists extensive information, tools and resources that can be helpful for ACP in palliative care for people with ID. For example, the information pack provides a link to a wish book for people with ID, “Wat wil ik? Als ik niet meer beter word…”[in English: ‘What do I want? If I don’t ever get better...’], which can be used to assess and record the wishes of people with (mild) ID regarding their end-of-life care [49] . Also, a tool (PALLI) is described that has questions regarding physical decline, decline in activities, changes in behaviour, symptom burden, frailty, illness and prognosis that may help professionals to proactively identify palliative care needs in people with ID [50, 51]. More information about the ACP programme (in Dutch) can be found at: https://www.koraal.nl/gehandicaptenzorg/wat-maakt-ons-bijzonder/zorg-dichtbij/advance-care-planning.

Carefully consider how people with ID can be involved in ACP

An important finding of this thesis is that people with ID were often not involved in ACP. Decisions that had to be made regarding the end of life were often made *about* a person with ID (substitute decision-making) instead of *together with* the individual (supported decision-making).

There is no question that including people with ID in ACP can be complex and challenging. As was mentioned by relatives and professionals who took part in the studies, some people with ID are not able to understand their own health situation and cannot oversee the consequences of the difficult decisions that had to be made. It is also known from previous research that people with ID do not always understand the concept of death, are less aware about the decisions that have to be made regarding their end of life and report a greater fear of death [52]. Hence, in some cases, it could be in a person’s best interests to decide not to include an individual with ID in difficult end-of-life decisions or not to disclose bad news around life-limiting illness and death. However, people with ID have a right to know about and be involved in their own care. The decision to include or not include a person with ID in ACP should, therefore, not be made beforehand but requires careful assessment [53].

This can for example be done by making use of a model for breaking bad news around life-limiting illness and death [15]. In the ‘process model’, which has been specifically developed for people with ID, an important thing to consider is a person’s capacity to make their own decisions. To asses a person’s capacity with regard to a particular issue at a particular time, it is important to provide them — piece by piece — with the information needed and help them to understand the information. Other important things to consider are if inclusion in or exclusion from ACP could be harmful for the person with ID and if the person with ID wants to be involved.

Foster close relationships between people with ID, relatives and professionals

If palliative care is to be provided in accordance with the wishes and needs of the person with ID, it is important to work together with everyone who is involved in the care of that individual. Involving key people such as close relatives in ACP is important for making decisions about future care that improve the quality of life of the individual with ID [30, 54].

This thesis showed that relatives found it important to be involved in ACP and wanted to be informed about the changing needs and symptoms of the person with ID. It is therefore important that professionals actively encourage their involvement, take the initiative to discuss and share views about changing care needs with relatives and have a collective understanding of what is in the best interests of the individual with ID based on their needs, preferences and quality of life. Participants with ID also said they preferred to have familiar people taking care of them when they are ill. This emphasises the importance of building and fostering close relationships between people with ID, relatives and professionals. Especially for people with more severe ID who do not have the capacity to make their own decisions, knowing a person and their life story is very important in articulating their wishes and needs and making decisions on their behalf [55].

Cooperate with other ID care organisations and organisations specialised in palliative care

As mentioned before, people with ID are nowadays an ageing population. However, compared to other care facilities for the aged, relatively few people in ID care organisations need palliative care. Recent results from a study by Todd et al. (2020) suggest that a large ID care organisation may only experience two to three deaths per year, showing that death is not a common event within ID services [56]. To ensure that people with ID have their palliative care needs met and experience a good death, it is important to have cooperation in place with other ID care organisations and organisations specialised in palliative care. In this way, organisations can learn from each other and share experiences and best practices.

In the Netherlands, ID care organisations can join a Regional Palliative Care Network, which is a collaborative network of organisations providing palliative care. It would be beneficial for ID care organisations to be actively involved in these networks as they can benefit from the expertise of organisations specialised in palliative care, for example regarding the provision of good housing and activities, the provision of mental and physical health care, and dealing with dementia.

The regional networks across the country are clustered into seven palliative care consortia with academic centres specialised in palliative care and educational institutions. In this way research, care and education are developed collaboratively, which is also important for palliative care provision in ID care organisations.

Implications for future research

Involve people with ID in palliative care research

Inclusive research has developed over the past decades and is increasingly common in ID research [57]. However, the active involvement of people with ID is less common in palliative care research and the perspectives of people with ID are often not included in study designs [58]. To allow all voices to be heard, it is important to consider alternative research approaches and facilitate the inclusion of people with ID.

Palliative care research should be conducted *with* people with ID instead of being *about* them. Inclusive or participatory research in this field can for example be achieved by involving people with ID as co-researchers in research. As a co-researcher, a person with ID can perform various tasks such as formulating the research topics, recruiting participants, conducting interviews or being involved in analysing and reporting the data. Researchers and co-researchers should decide together what roles and tasks co-researchers can take on, taking the qualities and preferences of co-researchers into account [59, 60]. In this way, research will better reflect the ideas, needs and preferences of people with ID regarding palliative care and can potentially lead to more effective quality improvement and person-centred care around the end of life.

Explore the wishes and needs of people with ID who do not use speech to communicate

There is a need for research that describes decision-making support for people with severe or profound ID and focusses on exploring the wishes and needs around the end of life of people with ID who do not use speech to communicate. While there is an increased focus on including people with mild ID in decision-making in palliative care and ACP by developing tools such as wish books to discuss wishes for care around the end of life and models of supported decision-making, only a few studies, models and tools are available focussing on the inclusion of people with severe or profound ID [33].

As mentioned before, exploring the wishes and needs of people with ID can be complex. This is especially true for people with ID who do not use speech to communicate. People with severe or profound ID often communicate non-verbally, whether intentionally or unintentionally, using vocalisations, facial expressions, body movements, eye gaze and touch. Because of these communicational challenges, professional and relatives often have the perception that people with severe or profound ID are unable to express their preferences in relation to decisions about their life and the end of life [54, 55, 61]. Hence, research should focus on ways of acknowledging the non-symbolic nature of their communication, and recognising their expressions of preferences, for example, how to recognise pain or distress, and what could help to make people with severe or profound ID comfortable or feel better. Moreover, training and tools for professionals and relatives should be developed to assist them in this process.

Study the quality of care relationships in palliative care for people with ID

It is important to gain more insight into what ensures good working relationships during the end of life of people with ID. Because of their lifelong disability, professional caring relationships can be very important for people with ID [62]. Especially during palliative care, when relationships often become even more intensified [24, 63]. However, studies focussing on the determinants of the quality of care relationships with people with ID are scarce [64]. It would therefore be interesting to study what can positively or negatively influence the relationships between people with ID, their relatives and professionals.

Moreover, it would be valuable to know which competencies and skills displayed by professionals are important in developing good working relationships and to study how these relationships change during end-of-life care for people with ID.

Gain insight into ACP for people with ID who live independently

This thesis focusses on ACP for people with ID who receive residential care or support from a specialised ID care organisation. Future research could gain more insight into the extent to which the important aspects of ACP found in this study match the views and roles of people with milder intellectual disabilities who live independently and/or receive non-specialised care. While their ACP process might look different, the need for the support and involvement of professionals as well as relatives will conceivably lead to comparable challenges [62, 65]. Also, it would be interesting to study if and how general practitioners (GPs) who care for these individuals discuss ACP or initiate conversations regarding wishes for end-of-life care with the individual and/or their relatives, collaborate with ID physicians concerning the provision of palliative care, or refer to hospices or other organisations specialised in palliative care. In this way, more knowledge can be gained of the perceived roles and responsibilities of GPs in ACP in palliative care for people with ID.

Translate knowledge into palliative care practices

In recent decades, more knowledge has become available about palliative care for people with ID. However, to a large extent, it remains unclear whether this knowledge is being translated into palliative care practices for people with ID and how it can potentially improve the quality of palliative care provision for people with ID and their relatives [33, 58]. The few palliative care innovations for people with ID that have been developed based on scientific knowledge often lack information about the process and effects of implementation. For example, not much is known about implementation strategies used within palliative care for people with ID, how changes in palliative care practice can be and are being evaluated, and how the use of potentially effective innovations can be sustained. It is therefore important to gain more insights into the effects of changed palliative care practice for people with ID, their relatives and professionals.

Further investigate the effects of the ACP programme

As described above, the evaluation of the ACP programme for people with ID yielded positive outcomes and a perceived increase in the competencies of professionals [66]. However, no baseline measurements were carried out, and the evaluation did not include patient outcomes. Whether the ACP programme actually leads to more effective ACP and improved outcomes for people with ID and their relatives remains unclear. Future research is therefore recommended with a pretest-posttest design and focussing on exploring the effects on the actual care provided and on patient outcomes.

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Nederlandse samenvatting

Advance Care Planning

in de palliatieve zorg voor mensen met een verstandelijke beperking

Advance Care Planning (ACP) stelt mensen in staat om doelen en voorkeuren te formuleren voor toekomstige medische behandelingen en zorg, deze doelen en voorkeuren te bespreken met familie en zorgverleners, en om eventuele voorkeuren vast te leggen en indien nodig te herzien. Deze proactieve benadering moet ertoe leiden dat zorg wordt geboden volgens de wensen en behoeften van de cliënt, wat de kwaliteit van leven bevordert. Ondanks het belang van ACP, is er nog weinig over bekend in de zorg voor mensen met een verstandelijke beperking.

Het doel van dit proefschrift is om meer inzicht te krijgen in ACP in de palliatieve zorg voor mensen met een verstandelijke beperking. Ook beschrijft het proefschrift de ontwikkeling, implementatie en evaluatie van een ACP-programma voor professionals.

De inleiding van dit proefschrift (*Hoofdstuk 1*) geeft informatie over de zorgbehoeften van mensen met een verstandelijke beperking in Nederland. Daarnaast stellen we de definitie van palliatieve zorg vast en het belang van ACP.

Een verstandelijke beperking ontstaat voor het achttiende levensjaar en kenmerkt zich door een beperking in het cognitief functioneren, die vaak wordt uitgedrukt door een intelligentiequotiënt (IQ) score van lager dan 70, en een beperking in het adaptieve gedrag (conceptuele, sociale en praktische vaardigheden).

Mensen met een verstandelijke beperking hebben vaak een leven lang complexe zorgvragen. Naast een verstandelijke beperking, hebben zij gemiddeld meer chronische, lichamelijke aandoeningen dan de algemene bevolking en ook is er vaker sprake van meerdere aandoeningen tegelijkertijd (multimorbiditeit). Desondanks is hun levensverwachting in de laatste decennia toegenomen. Hierdoor hebben zij, net als mensen zonder verstandelijke beperking, een grotere kans op het ontwikkelen van levensbedreigende aandoeningen die relatief vaker voorkomen op hoge leeftijd, zoals hart- en vaatziekten, dementie en kanker. Door deze nieuwe ontwikkeling, zijn er meer mensen met een verstandelijke beperking die ouder worden en te maken hebben met een langere periode van chronische ziekten. Hierdoor is er een groeiende behoefte aan palliatieve zorg voor mensen met een beperking.

De definitie van palliatieve zorg is: “Zorg die de kwaliteit van het leven verbetert van patiënten en hun naasten die te maken hebben met een levensbedreigende aandoening of kwetsbaarheid, door het voorkomen en verlichten van lijden, door middel van vroegtijdige signalering en zorgvuldige beoordeling en behandeling van problemen van fysieke, psychische, sociale en spirituele aard.” (Kwaliteitskader palliatieve zorg, 2017). Palliatieve zorg voor mensen met een verstandelijke beperking kent een aantal uitdagingen:

- Ten eerste hebben mensen met een verstandelijke beperking vaak een beperkt begrip van hun gezondheidssituatie en kunnen zij hun problemen, wensen en behoeften niet altijd goed onder woorden brengen.
- Een tweede uitdaging hangt daarmee samen en betreft de herkenning van de behoefte aan palliatieve zorg. Mede door de beperkingen in verbale communicatie en door beperkt ziektebegrip van mensen met een verstandelijke beperking, is het voor zorgverleners moeilijk om een behoefte aan palliatieve zorg tijdig te herkennen en om mensen met een verstandelijk beperking te informeren over en te betrekken bij de zorg.
- Een derde uitdaging is dat er juist bij deze cliëntengroep, vanwege de complexe zorgbehoeften, vaak belangrijke beslissingen genomen moeten worden, zoals over het inzetten van levensverlengende behandelingen, opname in een ziekenhuis of over het overplaatsen naar een andere zorgsetting.
- Een vierde uitdaging is dat professionals in de verstandelijk gehandicaptenzorg vaak geen medische of verpleegkundige achtergrond hebben en in hun basisopleiding niet of nauwelijks geschoold zijn in palliatieve zorg. Hierdoor hebben zij weinig kennis van palliatieve zorg.
- Een vijfde uitdaging is de afwezigheid van een palliatief zorgbeleid in veel zorgorganisaties voor mensen met een verstandelijke beperking. Zo ontbreken er bijvoorbeeld vaak richtlijnen of protocollen rondom het nemen van einde-levensbeslissingen en weten zorgverleners niet goed welke rol zij hebben in de palliatieve zorg.
- Het gebrek aan goede samenwerking met zorgaanbieders die meer ervaring hebben met het bieden van palliatieve zorg, zoals hospices of organisaties binnen regionale netwerken palliatieve zorg, is een zesde en laatste uitdaging.

Deze uitdagingen maken het extra belangrijk om palliatieve zorg in het algemeen, en ACP in het bijzonder, onderdeel te maken van de zorg voor mensen met een verstandelijke beperking. Dit proefschrift beschrijft de stand van zaken aangaande ACP in de zorg voor mensen met een verstandelijke beperking. Op basis daarvan wordt de ontwikkeling, implementatie en evaluatie van een ACP-programma voor mensen met een verstandelijke beperking beschreven. Het programma is ontstaan op basis van vijf deelstudies. Hierna volgen de belangrijkste inzichten uit deze deelstudies, aan de hand van de hoofdvragen uit dit proefschrift.

Een eerste hoofdvraag luidde: **Wat is er bekend over gebruik, inhoud en effecten van ACP in de palliatieve zorg voor mensen met een verstandelijke beperking?**
Onderzoek naar ACP is gericht op elementen van ACP, maar niet op het brede proces van ACP in de palliatieve zorg voor mensen met een verstandelijke beperking. Ook het gebruik van ACP in de praktijk en de effecten van ACP voor deze doelgroep bleven in de gevonden studies buiten beschouwing. Dit blijkt uit de systematische literatuurstudie beschreven in *Hoofdstuk 2*.

In vier verschillende literatuur databanken (PubMed, PsycINFO, Embase en CINAHL) zijn veertien studies gevonden die voldeden aan de inclusie criteria. Het merendeel (n=8) had een focus op besluitvorming, specifiek over medische einde-levensbeslissingen en lieten onder andere zien dat bij veel mensen met een verstandelijke beperking besluiten, onder andere over reanimatie, werden genomen. Ook bleek dat mensen met een verstandelijke beperking zelf vaak niet worden betrokken bij deze beslissingen.

Vijf van de veertien studies richtten zich op het organisatorisch beleid rondom palliatieve zorg en ACP. De studies toonden aan dat organisaties voor mensen met een verstandelijke beperking vaak geen op schrift gesteld beleid hebben ten aanzien van ACP. Andere elementen van ACP, waar de gevonden studies zich (ook) op richtten, waren communicatie over wilsverklaringen (n=1), het betrekken van familie bij ACP (n=1), documentatie over ACP (n=1) en de start van ACP (n=1).

Een tweede hoofdvraag luidde: **Hoe wordt ACP binnen zorgorganisaties voor mensen met een verstandelijke beperking toegepast en hoe wordt daarbij rekening gehouden met de wensen van mensen met een verstandelijke beperking?**
Uit *Hoofdstuk 3* blijkt dat ACP weinig wordt toegepast binnen zorgorganisaties voor mensen met een verstandelijke beperking. Afspraken voor toekomstige zorg richtten zich vooral op medische interventies en mensen met een verstandelijke beperking worden daar vaak niet in betrokken. Deze resultaten volgen uit analyses van medische dossiers van vijftien cliënten in de laatste levensfase en van vijftien cliënten die recent overleden waren na een ziekbed. Daarnaast zijn aanvullende interviews gehouden met betrokken professionals (artsen of begeleiders) en naasten.

Ondanks dat in zeven dossiers het woord palliatieve zorg niet viel, waren in 29 van de 30 dossiers wel afspraken over toekomstige zorg genoteerd. Dit waren voornamelijk afspraken over medische interventies, zoals reanimatie, toekomstige behandelingen of ziekenhuisopnamen. In twee van de 30 dossiers was ook een afspraak over de voorkeursplaats van zorg aan het levenseinde gerapporteerd. De afspraken waren vaak relatief kort voor het overlijden genoteerd, bij de helft van de overleden cliënten minder dan één maand voor het overlijden.

Afspraken over toekomstige zorg waren gezamenlijk door de betrokken arts en naasten genomen, zo bleek uit de dossiers. Ook andere professionals waren veelal betrokken, bijvoorbeeld de begeleiders, gedragsdeskundigen, geestelijk verzorgers of medisch specialisten uit het ziekenhuis. Slechts uit vier dossiers bleek dat ook de persoon met een verstandelijke beperking aan de gesprekken over deze beslissingen had deelgenomen. In de aanvullende interviews gaven 22 van de 30 professionals aan dat de persoon met een verstandelijke beperking niet kon worden betrokken bij ACP. Als redenen hiervoor gaven zij dat de persoon hiervoor onvoldoende cognitieve vermogens had, de situatie niet kon overzien of het te belastend zou zijn voor hem of haar om daarbij betrokken te zijn.

Een derde hoofdvraag luidde: **Wat is volgens mensen met een verstandelijke beperking, naasten en zorgverleners belangrijk voor ACP in de palliatieve zorg voor mensen met een verstandelijke beperking?**

Het bieden van zorg op maat, goed teamwerk en voldoende tijd nemen en geven blijken belangrijke elementen voor ACP in de palliatieve zorg voor mensen met een verstandelijke beperking beschreven in *Hoofdstuk 4*. Uit de analyse van vijftien diepte-interviews met mensen met een verstandelijke beperking (n=5), naasten (n=8) en professionals (n=7), kwamen de volgende drie thema's en zes subthema's naar voren:

Thema 1) Zorg op maat. De subthema's die hierbij naar voren kwamen waren: 'dichtbij staan' en 'verbinden met de persoon'. Hoe een ACP-proces eruit moet zien, was afhankelijk van de individuele persoon waar het om gaat. Een goede relatie met de persoon met een verstandelijke beperking en hem of haar goed kennen was cruciaal.

Thema 2) Teamwerk. Hierbij waren 'samen beslissen' en 'elkaar vertrouwen' subthema's. Iedereen die belangrijk is voor een persoon met een verstandelijke beperking zou betrokken moeten zijn bij ACP. Ook elkaar vertrouwen en open en eerlijk naar elkaar zijn, werd belangrijk gevonden.

Thema 3) Tijd nemen en geven. 'Voorbereiden en vooruitdenken' en 'ruimte maken voor emoties' werden hierbij als subthema's genoemd. Wensen en behoeften over mogelijke toekomstige situaties zouden tijdig besproken moeten worden, zodat er rust is aan het einde van het leven. Tijdens ACP-gesprekken moet ruimte zijn voor het uiten van emoties, omdat het voor cliënten en naasten mogelijk moeilijke onderwerpen zijn die worden besproken.

De geïnterviewde personen met een verstandelijke beperking gaven aan dat het moeilijk was om zelf beslissingen te nemen, hierbij zouden ze graag hulp krijgen van mensen die hen goed kennen. De geïnterviewde naasten zagen een rol voor zichzelf als vertegenwoordiger van de persoon met de verstandelijke beperking tijdens ACP-gesprekken, droegen bij aan beslissingen en konden daarnaast een signalerende rol innemen. De professionals benadrukten vooral dat zij een coördinerende, informerende en samenwerkende rol zouden moeten innemen in ACP.

Een vierde hoofdvraag was: **Welke bevorderende en belemmerende factoren beïnvloeden de implementatie en borging van innovaties in de palliatieve zorg voor mensen met een verstandelijke beperking?**
Ondersteuning vanuit de organisatie en de aanwezigheid van een kartrekker die verantwoordelijk is voor het gebruik van de innovatie zijn belangrijke beïnvloedende factoren voor implementatie en borging van palliatieve zorginnovaties voor mensen met een verstandelijke beperking. Dit blijkt uit een evaluatieonderzoek van negen implementatieprojecten in zorgorganisaties voor mensen met een verstandelijke beperking die zijn uitgevoerd in het kader van het landelijke Verbeterprogramma Palliatieve Zorg, waarbij bestaande innovaties werden geïmplementeerd om de palliatieve zorg te verbeteren.

Het onderzoek, beschreven in *Hoofdstuk 5*, combineerde verschillende onderzoeksmethoden zoals analyses van projectplannen en eindverslagen van de projecten, interviews met projectleiders en andere betrokken professionals, en vragenlijstonderzoek onder projectleiders. De palliatieve zorginnovaties, ingevoerd in de implementatieprojecten, betroffen:

1) De introductie van specialistische zorgconsulenten palliatieve zorg (vier projecten), bedoeld om professionals te ondersteunen tijdens een palliatief zorgtraject.
2) De ‘Signaleringsbox’ (drie projecten), een instrument om tijdig palliatieve zorgbehoeften in kaart te brengen en te bespreken.
3) ‘Sterven op je Eigen Manier’ (STEM; twee projecten), een innovatie gericht op communicatie over wensen en behoeften rondom het levenseinde in aansluiting bij de communicatiestijlen en –mogelijkheden van mensen in de laatste levensfase.

Trainingen voor professionals, samenwerking met andere organisaties en activiteiten gericht op het stimuleren en verspreiden van de innovatie binnen de organisatie, bleken belangrijke implementatiestrategieën. Factoren die de implementatie bevorderden, betroffen vaak organisatorische kenmerken, zoals betrokkenheid van management en voldoende financiële middelen en beschikbare materialen. De innovaties waren oorspronkelijk niet ontwikkeld voor de doelgroep van mensen met een verstandelijke beperking. Binnen een aantal implementatieprojecten werd de innovatie daarom nog doorontwikkeld om deze beter aan te sluiten bij de doelgroep.

Eén tot drie jaar na afronding van de projectfase gaven de projectleiders aan dat de innovatie nog steeds werd gebruikt binnen de zorgorganisaties. Wel twijfelden vijf van de negen projectleiders of de innovatie voor de langere termijn goed geborgd was. Dit werd volgens hen beïnvloed door organisatorische kenmerken, zoals de ondersteuning vanuit het management, de beschikking over middelen als geld, tijd en materialen, en de integratie van de innovatie in bestaande werkstructuren.

Een vijfde en laatste hoofdvraag luidde: **Hoe is een ACP-programma voor professionals die zorgen voor mensen met een verstandelijke beperking ontwikkeld, geïmplementeerd en geëvalueerd?**
Op basis van de resultaten van de systematische review, dossieronderzoek en interviews (zie de hierboven beschreven *Hoofdstukken 2 en 3*) en de belangrijke elementen van ACP (*Hoofdstuk 4*), werden tien competenties geformuleerd die professionals zouden moeten hebben om ACP in de palliatieve zorg toe te passen (*Hoofdstuk 6*). Deze tien competenties waren: identificeren van de palliatieve fase, signaleren van symptomen en zorgbehoeften, communiceren over wensen en behoeften, rapporteren van wensen en afspraken, integreren van ACP als proces in de palliatieve zorg, rekening houden met de wensen van de cliënt, samenwerken met de cliënt, naasten en professionals, aandacht hebben voor ethische dilemma’s, bevorderen van deskundigheid en borgen dat ACP een onderdeel wordt van palliatieve zorg.

In co-creatie met mensen met een verstandelijke beperking, naasten, professionals, en onderwijsexperts zijn deze tien competenties vertaald naar een ACP-programma voor professionals die palliatieve zorg bieden aan mensen met een verstandelijke beperking. Er werden twee co-creatie teams gevormd om het programma te ontwikkelen: één team bestaande uit naasten en professionals en één team bestaande uit mensen met een matige verstandelijke beperking.

Het ontwikkelde ACP-programma werd vervolgens geïmplementeerd in zes zorgorganisaties voor mensen met een verstandelijke beperking. In totaal namen 58 professionals (o.a. artsen, begeleiders, verpleegkundigen, gedragsdeskundigen en geestelijk verzorgers) deel aan het programma dat bestond uit drie elementen: een informatiepakket, twee 2-uur durende trainingsbijeenkomsten en een implementatie gesprek gericht op de toepassing van het ACP-programma.

Het programma werd geëvalueerd door deelnemende professionals een online vragenlijst voor te leggen zes maanden na de training. Uit de evaluatie bleek dat professionals hun ACP-competenties vonden verbeterd door deelname aan het programma. Minimaal 70 procent van de deelnemers vond dat hun communicatievaardigheden waren verbeterd, dat ze ACP vaker toepasten in de zorg, meer aandacht hadden voor dilemma's, beter konden reflecteren over ACP en het belang van ACP beter uitdroegen. Ook gaven de professionals aan dat zij deze competenties vaker en beter inzetten in de dagelijkse zorg voor mensen met een verstandelijke beperking. Het signaleren van symptomen en zorgbehoeften was minder verbeterd, evenals het rapporteren wensen en afspraken en het samenwerken met de cliënt, naasten en professionals.

Het afsluitende hoofdstuk (*Hoofdstuk 7*) presenteert de belangrijkste onderzoeksbevindingen, en inhoudelijke en methodologische reflecties. Ook worden aanbevelingen gedaan voor de praktijk en toekomstig onderzoek.

Het onderzoek beschreven in dit proefschrift is met name kwalitatief van aard. Een sterk punt hierbij is dat mensen met een verstandelijke beperking zelf aan het woord zijn gekomen. Zo zijn er interviews met hen gehouden en hebben mensen met een verstandelijke beperking in een co-creatie groep meegewerkt aan de opzet van het ACP-programma. Daarnaast zijn de perspectieven van naasten en professionals meegenomen. Hierdoor sluit het ontwikkelde ACP-programma aan bij de wensen en behoeften van mensen met een verstandelijke beperking, naasten en professionals. Beperkingen van het onderzoek zijn dat 1) het ACP-programma is geëvalueerd door middel van een zelfevaluatie onder professionals en 2) de effecten op en de zorg of ervaringen van mensen met een verstandelijke beperking en naasten niet zijn gemeten.

Op basis van de resultaten wordt aanbevolen dat naasten en professionals nagaan op welke manieren mensen met een verstandelijke beperking kunnen worden betrokken bij ACP. Ook is training voor professionals en samenwerking met andere organisaties belangrijk om de kwaliteit van de palliatieve zorg voor mensen met een verstandelijke beperking te verbeteren en zorg beter aan te sluiten bij de wensen en behoeften van mensen met een verstandelijke beperking. Voor toekomstige onderzoek is het relevant om na te gaan hoe mensen met een verstandelijke beperking, waaronder ook mensen met een ernstige verstandelijke beperking die zich verbaal niet goed kunnen uiten, betrokken kunnen worden bij ACP en hiervoor handvatten te bieden aan professionals.

List of publications

List of publications

Included in this thesis:

Voss, H., Vogel, A., Wagemans, A. M., Francke, A. L., Metsemakers, J. F., Courtens, A. M., & de Veer, A. J. (2017). Advance Care Planning in Palliative Care for People With Intellectual Disabilities: A Systematic Review. Journal of pain and symptom management, **54**(6), 938-960. https://doi.org/10.1016/j.jpainsymman.2017.04.016

Voss, H., Vogel, A., Wagemans, A.M.A., Francke, A.L., Metsemakers, J.F.M., Courtens, A.M.C., Veer, A.J.E. de. (2018). Advance care planning in the palliative phase of people with intellectual disabilities: analysis of medical files and interviews. Journal of Intellectual Disability Research, **63**(10), 1262-1272. https://doi.org/10.1111/jir.12664

Voss, H., Vogel, A., Wagemans, A.M.A., Francke, A.L., Metsemakers, J.F.M., Courtens, A.M., Veer, A.J.E. de. (2020). What is important for advance care planning in the palliative phase of people with intellectual disabilities? A multi-perspective interview study, **33**(2), 160-171. Journal of Applied Research in Intellectual Disabilities. https://doi.org/10.1111/jar.12653

Voss, H., Vogel, A., Wagemans, A., Francke, A., Metsemakers, J., Courtens, A., & de Veer, A. (2020). Development, implementation, and evaluation of an Advance Care Planning program for professionals in palliative care of people with intellectual disability. Intellectual and Developmental Disabilities. *Advance online publication, available at:* http://aaidd.org/publications/journals/articles-accepted-for-publication

Voss, H., Francke, A., Veer, A. de. Implementation and sustainment of palliative care innovations within organizations for people with intellectual disabilities: a multimethod evaluation. *Under review*

Other publications:

Watson, J., **Voss, H.**, & Bloomer, M. J. (2019). Placing the preferences of people with profound intellectual and multiple disabilities at the center of end-of-life decision making through storytelling. Research and Practice for Persons with Severe Disabilities, **44**(4), 267-279. https://doi.org/10.1177/1540796919879701

de Veer, A. J., Fleuren, M. A., **Voss, H.**, & Francke, A. L. (2020). Sustainment of innovations in palliative care: a survey on lessons learned from a nationwide quality improvement programme. Journal of Pain and Symptom Management. https://doi.org/10.1016/j.jpainsymman.2020.07.035

Braamse, A., **Voss, H.**, Nikolaus, S., Wearden, A., & Knoop, H. (2020). The role of partners' fatigue and the patient-partner relationship in the outcome of cognitive behavioural therapy for chronic fatigue syndrome. Journal of Psychosomatic Research. https://doi.org/10.1016/j.jpsychores.2020.110133

Publications in Dutch:

Voss, H., Meulenkamp, T.M., Marangos, A.M., Boeije, H.R. Veel onduidelijkheid bij familieleden van mensen met een verstandelijke beperking over veranderingen in de langdurige zorg. Utrecht: Nivel, 2017

Voss, H., Boeije, H., Hees, S. van, Meulenkamp, T., Oldenkamp, M. Mijn eigen stekkie: woonbeleving van mensen met een verstandelijke beperking. Utrecht: Nivel, 2017.

Veer, A. de, **Voss, H.**, Fleuren, M., Francke, A. Van implementeren naar borgen: leerpunten uit het ZonMw Verbeterprogramma Palliatieve Zorg. Utrecht: Nivel, 2018.

Haarmann, A., **Voss, H.**, Boeije, H. Sporten en bewegen door mensen met een lichte verstandelijke beperking: belemmeringen en mogelijkheden. Utrecht: Nivel, 2019.

Veer, A. de, **Voss, H.**, Ramerman, L., Verkaik, R. Beeldvorming & Diagnostiek bij probleemgedrag in de praktijk: De aanpak bij volwassenen met een verstandelijke beperking met 24-uurszorg. Utrecht: Nivel, 2020

Francke, A., Meurs, M., Plas, A. van der, **Voss, H.** Inventarisatie van Advance Care Planning. ZonMw-projecten, methoden, uitkomsten en geleerde lessen over toepassing, implementatie en borging. Utrecht: Nivel, 2020.

Voss, H., Scheffer, M., Boeije, H. Voeding van mensen met een verstandelijke beperking: lekker en gezond, maar niet altijd een eigen keuze. Utrecht: Nivel, 2020.

Other published material:

Training Advance Care Planning. Optimaal ondersteunen in de palliatieve zorg voor mensen met een verstandelijke beperking (Koraal). Available at: https://www.koraal.nl/gehandicaptenzorg/wat-maakt-ons-bijzonder/zorg-dichtbij/advance-care-planning

Tips bij gebruik format advance care planning bij mensen met een verstandelijke beperking (Koraal). Available at: https://www.koraal.nl/gehandicaptenzorg/wat-maakt-ons-bijzonder/zorg-dichtbij/advance-care-planning

Portfolio

NAME COURSE, TRAINING, ACTIVITY	ORGANISER	EC	PLANNED	COMPLETED
MANDATORY COURSES (VU/VUmc specific)				
Scientific integrity course or research ethics - Online Course 2020	Amsterdam UMC	2		23-10-2020
Congress participation 15th World Congress of the European Association for Palliative Care, Madrid (oral presentation)	EAPC	2		18-20 May 2017
		4		
ELECTIVE MANDATORY COURSES (APH specific)				
Course qualitative research	Nivel	0.5		23 & 30 June 2016
Beginners course STATA	Nivel	0.5		13 & 16 June 2016
Second course STATA	Nivel	0.5		26 & 30 January 2017
Course ‘Epidemiologisch onderzoek: basisprincipes’	EpidM	1.4		13-01 – 17-01 2020
Qualitative Research Methods – *online course*	University of Amsterdam	1		13 May – 1 July 2020
Participatief actieonderzoek training (PAR) – *online course*	School of Participation, Amsterdam UMC	3	6-10-2020 – 11-12-2020	
Course ‘Kwaliteitssyteem Nivel en het verrichten van interne audits’	Nivel	0.5		12-10-2016
Participation in ‘Talent Ontwikkel Programma (TOP)’ for junior researchers	Nivel	0.5		October 2017 – March 2019
Workshop ‘Professioneel netwerken’	Nivel	0.1		24-10-2017
Workshop ‘Persoonlijke effectiviteit’	BoertienVergouw en Overduin	0.4		26-11-2018
Meeting research platform Disability Studies ‘Inclusie & Belonging’	Disability studies VU, Amsterdam	0.1		07-11-2019
Expert meeting palliative care for people with intellectual disabilities	Agora	0.3		08-06-2017
Wetenschapsmiddag Nivel (presentation)	Nivel	1		14-10-2019
Quest Meeting end-of-life research (invited presentation)	VUmc	1		11-02-2020
		10.8		

ELECTIVE COURSES				
Courses & workshops:				
Workshop ‘Online personal branding via LinkedIn’	Nivel	0.1		28-09-2016
Workshop ‘Omgaan met stakeholders’	Boertien Vergouwen Overduin	0.3		28-05-2018
Course ‘Academic Presenting in English’	BABEL	1.4		4 April - 16 May 2017
Course ‘Academic Writing in English’	BABEL	1.4		10 April - 22 May 2018
Workshop ‘Time Management’	Leidsvrouw	0.1		09-09-2019
		3.3		
Conference and symposia attendances:				
Conference attendance ‘Meer kracht en meer macht. Waarom empowerment juist nu nodig is’	Movisie	0.3		14-06-2016
Symposium attendance ‘7th Amsterdam Symposium on Palliative Care’	Amsterdam UMC	0.3		19-10-2017
Symposium attendance ‘The next big thing, will be a lot of small things’	Academische Werkplaats Leven met een verstandelijke beperking, Tranzo	0.3		20-04-2017
Conference attendance ‘Nederlands-Vlaamse Wetenschapsdagen Palliatieve Zorg’ (oral presentation)	Palliactief	1		30-11-2017 & 01-12-2017
Conference attendance ‘Samen Beslissen met kwetsbare mensen: een uitdaging!’	Het Landelijk Platform Gedeelde Besluitvorming	0.3		26-01-2018
Conference attendance ‘CaRe Days 2018 – Research integrity’	The Netherlands School of Primary Care Research	1		16 & 17-05-2018
Conference attendance ‘Focus op onderzoek - Partnerschap in onderzoek’ (oral presentation)	Kennisplein gehandicaptensector & ZonMw	1		01-06-2018
Conference attendance ‘Participatief onderzoek out of the box’	Disability studies VU, Amsterdam	0.3		27-09-2018

International seminar on Public Health Research in Palliative Care: Shifting the Paradigm (poster presentation)	EAPC Reference Group on Public Health and Palliative Care, Brussel	2		25 & 26-10-2018
Conference attendance ‘7e Nationaal Congres Palliatieve Zorg. Palliatieve zorg voor iedereen! Van kwaliteitskader naar unieke patiënt’	Netwerken palliatieve zorg	0.6		8 & 9 November 2018
Conference attendance ‘International Conference Upholding rights, preventing abuse and promoting autonomy’	Australian Guardianship and Administration Committee, Canberra	0.6		14-15 March 2019
Conference attendance ‘9^e Amsterdam Symposium Palliatieve Zorg: Palliatieve zorg en de ouder wordende mens’	Amsterdam UMC	0.1		10-10-2019
Masterclass 9^e Amsterdam Symposium Palliatieve Zorg Prof.dr. Sheila Payne	Amsterdam UMC	0.1		10-10-2019
Conference attendance ‘Zoek het uit! Het landelijke congres over praktijk en onderzoek in de ouderen- en gehandicaptenzorg’	Kennisinfrastructuur Langdurige Zorg	0.3		10-02-2020
		8.2		
Transferrable skills:				
Academic meetings ‘Wetenschappelijk Overleg (WO)’	Nivel	2		2016 - 2020
Visiting Scholar Deakin University (Melbourne, Australia) School of Health & Soc. Dev. – Disability and inclusion	Scholarship awarded from The Netherlands Organisation for Health Research and Development (ZonMw) & Stichting de Drie Lichten	6		March - June 2019
		8		
	Total:	**34,3**		

Dankwoord

Dankwoord

Veel mensen hebben mij de afgelopen vier jaar geholpen met het schrijven van dit proefschrift, hen wil ik heel graag bedanken.

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Ik bedank graag de leescommissie: prof.dr. Bregje Onwuteaka-Philipsen, prof.dr. Carlo Schuengel, prof.dr. Geraline Leusink, prof.dr. Yvonne Engels en dr. Judith Rietjens, voor de tijd die ze hebben genomen om mijn proefschrift te lezen en te beoordelen. Dank aan dr. Jeroen Hasselaar voor zijn deelname aan de oppositiecommissie.

Jo, thank you so much for having me as a visiting scholar at Deakin University and making me feel so welcome “Down Under”. It was a great experience! Thank you for the productive traffic jams, borrowing your bike (I survived!), introducing me to inspiring colleagues, new friends, interesting research projects, and Melbourne’s best coffee. It would never have been possible without you.
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Laar, al sinds de middelbare school zijn we vriendinnen, sinds kort ook huisgenootjes en nu ben je mijn paranimf! Tegen jou kan ik alles vertellen en je bent altijd overal voor in. Carnaval, Oktoberfest, wintersport, niks is te gek. Op naar nog veel meer avonturen en feestjes in ons fijne appartementje.

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About the author

About the author

Hille Voss was born on 5 August 1993 in Gennep (Netherlands). After finishing secondary school, she studied Pedagogical Sciences at the Radboud University in Nijmegen and participated in the Radboud Honours Programme. As part of the Honours Programme, Hille visited the University of Texas in Austin (US) for three months to participate in research activities. Following her Bachelor's degree, Hille obtained her Master's degree in Pedagogical Sciences at the Radboud University in Nijmegen in 2015. During her studies, Hille worked as a social worker in a sheltered workplace for people with disabilities (Bakkerscafé Brood op de Plank) in Nijmegen. After finishing her studies, Hille gained work experience as a starting researcher and psychologist at the Radboud University Medical Center.

In March 2016, Hille started as a junior researcher at the Netherlands Institute for Health Services Research (Nivel) in Utrecht. She also coordinated the Living Together Panel (Panel Samen Leven), a panel of people with mild or moderate intellectual disabilities and their relatives. Hille received a scholarship to work as a visiting scholar at Deakin University in Melbourne (Australia) from March to July 2019. In September 2019, Hille returned to Nivel to work as a researcher. Hille completed her PhD thesis in 2020 under the supervision of Prof.dr. Anneke Francke, Prof.dr. Job Metsemakers, dr. Anke de Veer and dr. Annemieke Wagemans.

NIVEL