

## Systematic Review

# Shared decision-making with people with intellectual disabilities in the last phase of life: A scoping review

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## Abstract

**Background** Shared decision-making (SDM) is the process in which healthcare professionals and patients jointly discuss and decide which care and treatment policy is to be followed. The importance of SDM is increasingly being recognised across health settings, including palliative care. Little is known about SDM with people with intellectual disabilities (IDs) in the last phase of life. This review aimed to explore to which extent and in which way people with ID in the last phase of life are involved in decision-making about their care and treatment.

**Method** In this scoping review, we systematically searched in the Embase, Medline and PsycINFO databases for empirical studies on decision-making with people with ID in the last phase of life.

**Results** Of a total of 281 identified titles and abstracts, 10 studies fulfilled the inclusion criteria. All focused on medical end-of-life decisions, such as foregoing life-sustaining treatment, do-not-attempt-resuscitation orders or palliative sedation. All studies emphasise the relevance of involving people with ID themselves, or at least their relatives, in making decisions at the end of life. Still, only two papers described processes of decision-making in which persons with ID actively participated. Furthermore, in only one paper, best practices and guidelines for decision-making in palliative care for people with ID were defined.

**Conclusion** Although the importance of involving people with ID in the decision-making process is emphasised, best practices or guidelines about what this should look like are lacking. We recommend developing aids that specifically support SDM with people with ID in the last phase of life.

**Keywords** decision-making, end-of-life, intellectual disabilities, palliative phase, scoping review, shared decision-making

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## Introduction

In palliative care, the emphasis on shared decision-making (SDM), i.e. decision-making by healthcare professionals and patients (Charles *et al.* 1997) is growing. Palliative care is ‘an approach that improves quality of life of people with life limiting conditions due to illness and frailty and their families’ (World Health Organisation n.d.). Timely recognition of the palliative phase is key to allow discussion of preferences and to apply these in provided care (Vrijmoeth *et al.* 2016a; Vrijmoeth *et al.* 2016b). A structured approach may provide support in these SDM processes.

The concept of SDM first appeared in literature in 1997 (Charles *et al.* 1997). Through SDM patients can be included in decision-making when multiple treatment options coexist (Stiggelbout *et al.* 2015). Ideally, healthcare professionals clearly explain relevant care or treatment options and support patients in weighing their preferences and values in the context of these options before a treatment decision is made (Stiggelbout *et al.* 2012). A systematic review showed that patients who participated in SDM tended to report positive outcomes, such as a higher degree of patient satisfaction and less decisional conflict (Shay and Lafata 2015). SDM is often mentioned in the context of medical treatment but is also applicable in non-medical care and support, such as adjusting daytime activities and hobbies (Stiggelbout *et al.* 2015). So far, SDM models have not specifically described their use by people with intellectual disabilities (IDs) (Charles *et al.* 1999; Towle and Godolphin 1999; Makoul and Clayman 2006; Elwyn *et al.* 2012).

The relevance of involvement of people with ID in SDM might be obvious. However, in practice, assessing preferences and values of people with ID can be hindered by their ID or co-morbid conditions. Involvement of close proxies may be required. Other barriers for SDM include negative attitudes and lack of knowledge and skills in healthcare professionals (Stiggelbout *et al.* 2015). In addition, it can be difficult for people with ID to weigh the different options and to oversee their consequences in the longer term. People with ID may not always comprehend the information about and implications of their illness,

which limits their decision-making capacity (Tuffrey-Wijne 2013; Szmukler 2019).

In spite of limited or absent decision-making capacity, people’s preferences have to be taken into account. Article 12 recognises the right of people with ID to (United Nations 2006) be recognised by law as a person equal to others (United Nations 2006). People with ID have the right to be supported in making choices even if they cannot make such decisions by themselves (Szmukler 2019). See Box 1 for an example.

Knowledge about how to engage people with ID in decision-making in the last phase of life is limited, and research is still scarce. This review aimed to explore in which way people with ID in the last phase of life are involved in decision-making about their care and treatment.

## Methods

### Design

Given the exploratory nature of this study we opted for a scoping review. This is defined as: ‘a form of knowledge synthesis that addresses an exploratory research question aimed at mapping key concepts, types of evidence and gaps in research related to a defined area or field by systematically searching, selecting and synthesizing existing knowledge’ (Colquhoun *et al.* 2014). We followed the PRISMA Scoping Review Guidelines (Tricco *et al.* 2018). (Appendix A). In collaboration with a biomedical information specialist, we systematically searched the electronic databases Embase, Medline and PsycINFO in October 2018 and updated the search in September 2019. We used relevant search terms and synonyms related to ID, SDM and palliative care (see Box 2 for the Embase search protocol).

We used the following inclusion criteria: (1) study with or about people with ID in the last phase of life; (2) occurrence of elements of decision-making in practice in experimental or observational studies; (3) peer-reviewed journal and (4) written in English. We applied no limits on date of publication. We excluded editorials, letters and conference reports and checked systematic reviews and meta-analyses for useful references.

Mary is 45 years old and has an intellectual disability. More than anything Mary likes to slide down her wheelchair and crawl towards her cup of coffee. Mary seems satisfied: she smiles every time she drinks her coffee on the floor. But one day Mary's caretakers discover a strange hump on her leg. Hospital tests reveal a malignant cancer growth. Doctors say they can operate the growth, but it would not cure her, and they cannot guarantee that Mary would still be able to glide down her wheelchair after surgery. Mary's family and caretakers consider various treatment options together with the doctors. Treating or operating Mary's malignant cancer growth will not be lifesaving and would cause her lots of agitation and discomfort. It is possible to purchase a customised pillow that makes it easier for Mary to slide down her wheelchair. Supportive medication will ensure that Mary will not experience pain. They talk all options through with Mary, even though she is not considered to be competent for these decisions. Together with Mary's doctors, family and caretakers they decide not to opt for surgery or any other of the proposed treatments.

**Box 1.** An example of decisions in the context of a person with ID.

## Procedure

Two researchers (HN and IK) independently screened titles and abstracts. Disagreements about study inclusion were resolved by discussion by HN, IK and ME. They developed, reviewed and approved a data extraction form containing title, country, study aim, study design, described decisions and involvement of people with ID in the process of decision-making. HN and ME pilot tested this form by independently extracting data from one study and

comparing their results. Changes to the data extraction form were not required. Using this form, HN and ME independently extracted data from the remaining studies.

Aiming at high inter-rater agreement, HN, IK and ME discussed the results to identify elements of decision-making and possible differences in the interpretation of these elements. To evaluate, structure and describe processes of decision-making as identified in this review, we looked for a suitable SDM framework. We opted for the SDM model of

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('mental deficiency'/de OR 'intellectual impairment'/de OR 'Down syndrome'/de OR 'mentally
disabled person'/de OR 'developmental disorder'/de OR (((intellectual* OR Mental* OR
developmental*) NEAR/3 (impair* OR deficien* OR disab* OR handicap* OR disorder* OR retard* OR
defect*)) OR (down* NEAR/3 syndrome*)):ab,ti) AND ('shared decision making'/de OR 'family
decision making'/de OR 'patient decision making'/de OR ('decision making'/de AND ('patient'/de OR
'family'/de OR 'parent'/exp)) OR (((shar* OR famil* OR patient* OR parent* OR support* OR selfOR
surrogate* OR substitut*) NEAR/6 decision*)):ab,ti) AND ('palliative therapy'/exp OR 'palliative
nursing'/de OR 'terminal care'/de OR 'death'/de OR dying/de OR 'life sustaining treatment'/exp OR
'hospice'/de OR 'hospice care'/de OR 'hospice nursing'/de OR 'hospice patient'/de OR 'treatment
withdrawal'/exp OR 'living will'/exp OR euthanasia/exp OR (palliative OR terminal* OR end-of-life OR
death OR dying OR (last NEAR/6 (day* OR week* OR month*) NEAR/6 life) OR deathbed OR life-
sustain* OR life-prolong* OR hospice* OR ((treatment* OR therap*) NEAR/3 (withdraw* OR cessat*
OR withhold* OR terminat*)) OR living-will* OR (advance NEAR/3 directive*) OR euthanas*)):ab,ti)
NOT ([Conference Abstract]/lim) AND [english]/lim
```

**Box 2.** Complete Embase search string.

van de Pol *et al.* (2016) because it is aimed at frail patients with multimorbidity, in the context of a continuous SDM process and takes into account the involvement of relatives. van de Pol's model distinguishes six steps (Box 3). In brief, these steps are (1) Preparation (history; problem analysis); (2) Goal talk (identify discussion partner; identify patient values and goals of care); (3) Choice talk (summarise and offer choice; patient formulates treatment aims); (4) Option talk (personalised treatment aims are discussed); (5) Decision talk (focus on preferences; connect to the patient's values; goals of care and treatment aims; decide) and (6) Evaluation (evaluate the SDM process; prepare a treatment plan).

## Results

We found 281 articles. After the selection process, as shown in Fig. 1, 10 studies were included for data extraction.

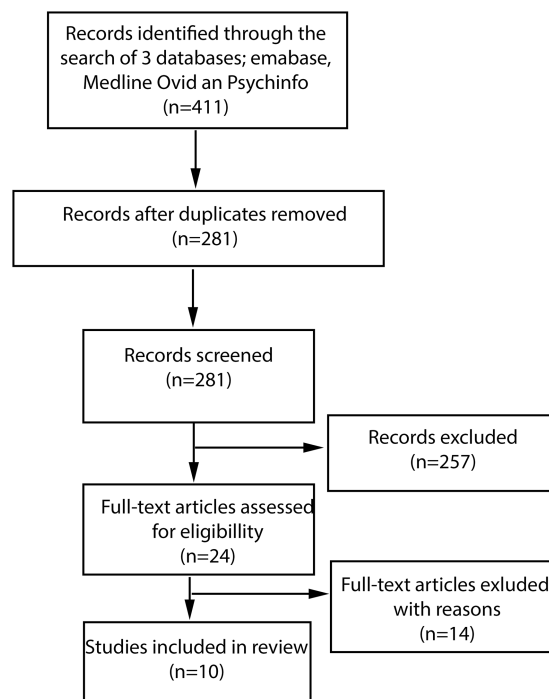


Figure 1. Study flow diagram.

### 1. Preparation:

- History: has the patient already documented anything with regard to advance care planning, treatment e.g.
- Problem analysis: what are the current problems of the patient.

### 2. Goal talk:

- Is the patient capable of making choices? Does the patient want to make these choices? If not, what has designated to make these choices for the patient? What are important values and care goals for the patient?

### 3. Choice talk:

- Summarise the earlier described steps. Explain that there are several treatment options and offer choice. Encourage the patient to express their treatment aims.

### 4. Option talk:

- The chosen treatment options will be discussed.

### 5. Decision talk:

- Focus on the patient's preferences. Connect to the values, care and treatment goals that are important to the patient. Make a decision.

### 6. Evaluation:

- Evaluate the SDM process. If everybody is satisfied a treatment plan can be prepared.

Box 3. The six steps of van de Pol *et al.* (2016) SDM model.

## Study characteristics

Of the 10 included studies, eight were conducted in the Netherlands, one in Australia, and one in the United States. Six studies were (semi-structured) interview studies (Van Thiel *et al.* 1997; Wagemans *et al.* 2013a; Wagemans *et al.* 2013b; Bekkema *et al.* 2015; Zaal-Schuller *et al.* 2016; Zaal-Schuller *et al.* 2018), two included multiple case studies (Watson *et al.* 2017; Tuffrey-Wijne *et al.* 2018), one was a single case study (Lohiya *et al.* 2003), and in one study, medical files were examined (Wagemans *et al.* 2010). All studies had an observational design.

The total number of participants across all studies was 317, ranging from 1 to 89 per study and included parents, physicians and people with ID. All studies involved interviews with and examination of the role of target groups. Five studies focused on healthcare professionals and relatives of people with an ID (Wagemans *et al.* 2010; Bekkema *et al.* 2015; Zaal-Schuller *et al.* 2016; Watson *et al.* 2017; Zaal-Schuller *et al.* 2018), four on healthcare professionals (Van Thiel *et al.* 1997; Lohiya *et al.* 2003; Wagemans *et al.* 2013a; Tuffrey-Wijne *et al.* 2018) and one on relatives of people with an ID (Wagemans *et al.* 2013b). Eight studies were published within the last 10 years; the other two were from 2003 and 1997.

All studies focused on end-of-life decision-making, e.g. deciding about life-prolonging treatments. All studies focused on adults with ID; four studies also focused on children with ID (Wagemans *et al.* 2013b; Bekkema *et al.* 2015; Zaal-Schuller *et al.* 2016; Zaal-Schuller *et al.* 2018); six studies included people with various levels of ID (Van Thiel *et al.* 1997; Wagemans *et al.* 2010; Wagemans *et al.* 2013a; Wagemans *et al.* 2013b; Bekkema *et al.* 2015; Watson *et al.* 2017), two studies focused on people with profound ID (Zaal-Schuller *et al.* 2016; Zaal-Schuller *et al.* 2018), one involved people with mild ID (Tuffrey-Wijne *et al.* 2018) and one people with moderate ID (Lohiya *et al.* 2003).

In four studies, people with ID received residential care (Van Thiel *et al.* 1997; Lohiya *et al.* 2003; Wagemans *et al.* 2010; Wagemans *et al.* 2013a), in four other studies, they received residential and community-based care (Wagemans *et al.* 2013b; Bekkema *et al.* 2015; Watson *et al.* 2017; Tuffrey-Wijne *et al.* 2018), and for two studies, this

was not specified (Zaal-Schuller *et al.* 2016; Zaal-Schuller *et al.* 2018), see Table 1 for an overview.

### *Preparation: history and problem analysis*

*Life history of the person with intellectual disability.* Five articles included a description of how the life history of the person with ID was taken into account in the decision-making process (Van Thiel *et al.* 1997; Wagemans *et al.* 2013b; Bekkema *et al.* 2015; Zaal-Schuller *et al.* 2016; Watson *et al.* 2017). In two studies, life stories helped to provide information about people's preferences considering health and treatment (Wagemans *et al.* 2013b; Watson *et al.* 2017). In one study, the views of parents and physicians on the quality of life of people with ID were reported to differ (Zaal-Schuller *et al.* 2018).

*Problem analysis.* In one study, the experiences of 17 parents of children with profound multiple and intellectual disabilities during end-of-life-decision-making were reported (Zaal-Schuller *et al.* 2016). These parents reported they had to explain to physicians how their child was feeling and, for example, when their child was in pain (Zaal-Schuller *et al.* 2016). Parents mentioned to prefer starting the end-of-life decision-making process with a physician who already had a relationship with their child and therefore could better analyse their child's problems. They believed that a physician who is aware of the history of their child can provide better treatment than a physician without that awareness (Zaal-Schuller *et al.* 2016).

### *Goal talk: identify discussion partner and identify patient values and goals of care*

All articles emphasised that the extent to which people with ID may be partners in the decision-making process is dependent upon their capacity to make difficult choices. In her description of the participation of people with ID in decision-making processes about euthanasia and assisted suicide requests, Tuffrey-Wijne *et al.* (2018) noted two aspects that are difficult for people with ID: appreciating the significance of the information and weighing of treatment options and their consequences. In eight identified studies, people with ID did not participate in the decision-making process.

Table 1 Key characteristics of included papers ( $n = 10$ )

First author, year of publication	Article title	Country	Degree of ID	Age of participants with ID	Residency of people with ID
Van Thiel <i>et al.</i> 1997	Retrospective study of doctors' 'end of life decisions' in caring for mentally handicapped people in institutions in the Netherlands	Netherlands	Mild ( $n = 4$ ), Moderate ( $n = 29$ ), Severe ( $n = 15$ ), profound ( $n = 16$ ).	0–49 year ( $n = 25$ ) 50–64 year ( $n = 26$ ) 65–79 year ( $n = 12$ ) > 80 year ( $n = 3$ )	Residential care
Wagemans <i>et al.</i> 2010	End-of-life decisions: an important theme in the care for people with intellectual disabilities	Netherlands	Mild, Moderate, Severe and Profound	<50 to >90 years	Residential care
Wagemans <i>et al.</i> , 2013a	End-of-life decisions for people with intellectual disabilities, an interview study with patient representatives	Netherlands	Mild ( $n = 4$ ), Moderate ( $n = 1$ ), Severe ( $n = 3$ ), Profound ( $n = 2$ )	40–78 years	Residential and community based care
Wagemans <i>et al.</i> , 2013b	The factors affecting end-of-life decision-making by physicians of patients with intellectual disabilities in the Netherlands: a qualitative study.	Netherlands	Mild ( $n = 4$ ), Moderate ( $n = 1$ ), Severe ( $n = 3$ ), Profound ( $n = 2$ )	40–78 years	Residential and community based care
Bekkema <i>et al.</i> 2015	'From activating towards caring': shifts in care approaches at the end of life of people with intellectual disabilities: a qualitative study of the perspectives of relatives, care-staff and physicians	Netherlands	Mild ( $n = 4$ ), Moderate ( $n = 4$ ), Severe/profound ( $n = 4$ ).	13–74 year	Residential and community based care
Zaal-Schuller <i>et al.</i> 2016	How parents and physicians experience end-of-life decision-making for children with profound intellectual and multiple disabilities	Netherlands	Profound ( $n = 14$ )	0–4 ( $n = 2$ ), 5–9 ( $n = 1$ ), 10–14 ( $n = 8$ ), 15–19 ( $n = 3$ )	Unclear
Zaal-Schuller <i>et al.</i> 2018	Considering quality of life in end-of-life decisions for severely disabled children	Netherlands	Profound ( $n = 14$ )	0–4 ( $n = 2$ ), 5–9 ( $n = 1$ ), 10–14 ( $n = 8$ ), 15–19 ( $n = 3$ )	Unclear
Tuffrey-Wijne <i>et al.</i> 2018	Euthanasia and assisted suicide for people with an intellectual disability and/or autism spectrum disorder: an examination of nine relevant euthanasia cases in the Netherlands (2012–2016)	Netherlands	Mild ( $n = 2$ ), Intellectual disability but degree of ID is not given ( $n = 2$ ), unknown ( $n = 3$ )	30–95 years	Residential and community based care
Lohiya <i>et al.</i> 2003	End-of-life care for a man with developmental disabilities	United States of America	Moderate(1)	30	Residential care
Watson <i>et al.</i> 2017		Australia		Unclear	



Table 1. (Continued)

First author, year of publication	Article title	Country	Degree of ID	Age of participants with ID	Residency of people with ID
	Supporting end of life decision making: case studies of relational closeness in supported decision making for people with severe or profound intellectual disability		Severe or Profound ID ( $n = 5$ )		Residential and community based care

Table 1. (Continued)

First author, year of publication	Aim of the study	Methodology	Kind of decisions	The way people with an ID were involved in the decision making process at the end of their lives
Van Thiel <i>et al.</i> 1997	To gain insight into the reasons behind and the prevalence of doctors' end of life decisions in institutions caring for people with ID in the Netherlands	67 doctors were interviewed about their most recent end-of-life decision with mentally handicapped people.	End of life decision making, euthanasia.	Unclear
Wagemans <i>et al.</i> 2010	To establish the prevalence and nature of end-of-life decisions in people with IDs in a Dutch residential care centre.	Medical files of the residents who died between January 2002 and July 2007 were systematically reviewed. One or more end-of-life decisions were made in 27 cases.	End of life decision making, medical decision making in general, treatment or no treatment.	Unclear
Wagemans <i>et al.</i> , 2013a				Unclear

Table 1. (Continued)

First author, year of publication	Aim of the study	Methodology	Kind of decisions	The way people with an ID were involved in the decision making process at the end of their lives
Wagemans <i>et al.</i> , 2013b	To clarify the process of end-of-life decision-making for people with ID from the perspective of patient representatives. To investigate how physicians make end-of-life decisions for patients with ID, and in particular, what factors affect the process of decision-making.	Sixteen patient representatives were interviewed after the deaths of 10 people with ID. Nine semi-structured interviews with ID physicians after the deaths of people with ID that involved end-of-life-decisions.	End of life decision making, medical decision making in general, treatment or no treatment End of life decision making, sedation, medical decision making in general, treatment or no treatment.	They are not involved.
Bekkema <i>et al.</i> , 2015	To explore relatives' and professionals' shifts in the approaches to end of life care in people with ID	Forty-five relatives and professionals who were closest to 12 recently deceased people with ID were interviewed.	End of life decision making, medical decision making in general, treatment or no treatment.	Unclear
Zaal-Schuller <i>et al.</i> , 2016	To investigate the experiences of the parents and the involved physician during the end-of-life decision making (EoLDM) process for children with PIMD.	Semi structured interviews with the physicians and parents of 14 children with PIMD for whom an EoLDM was made within the past 2 years.	End of life decision making, sedation, medical decision making in general, treatment or no treatment.	Unclear
Zaal-Schuller <i>et al.</i> , 2018	To determine which elements contribute to Quality of Life (QoL) according to parents and physicians, how QoL is incorporated into EoLDM and how parents and physicians discuss QoL considerations in the Netherlands.	Semi structured interviews with the physicians and parents of 14 children with PIMD for whom an EoLDM was made within the past 2 years.	End of life decision making, sedation, medical decision making in general, treatment or no treatment.	They are not involved.
Tuffrey-Wijne <i>et al.</i> , 2018	To examine and reflect on the way in which legal criteria are applied to patients with intellectual disabilities and/or autism spectrum disorder who request Euthanasia and Assisted Suicide (EAS) in the Netherlands.	Direct content analysis was used on 9 EAS cases for people with ID and autism disorder.	End of life decision making, euthanasia, medical decision making in general, treatment or no treatment.	Unclear
Lohiya <i>et al.</i> , 2003	To examine if the artificial maintenance of life functions prolongs life or dying.	Examines a single case study of a person with ID under guardianship of a state conservator.	End of life decision making, medical decision making in general, treatment or no treatment.	Unclear
Watson <i>et al.</i> , 2017	To characterise supported decision making for people with severe or profound intellectual disability. Secondly the researchers aimed to identify the processes, enablers and	Five case studies were examined as well as the role that paid and unpaid supporters played in supported decision making.	End of life decision making, medical decision making in general, treatment or no treatment.	The role of a person with ID is to express their own will and preference, intentionally and



Table 1. (Continued)

First author, year of publication	Aim of the study	Methodology	Kind of decisions	The way people with an ID were involved in the decision making process at the end of their lives
	barriers to supported decision making for people with ID to understand how it can be fostered in practice.			unintentionally, using a range of modalities.
D. intellectual disability.				

In one study, a case is described in which care professionals thought it was better to tell a woman with ID she had reached the last phase of life, whereas her family did not want to inform her, to protect her from distress (Wagemans *et al.* 2010).

Two studies described the involvement of people with ID in the decision-making process. In the described euthanasia and suicide requests study from Tuffrey-Wijne *et al.* (2018), each case report included the statement ‘the physician had sufficiently informed the patient about his/her situation and his/her prospects’. The authors noted that there is no information about how the people with ID were helped to understand this information (Tuffrey-Wijne *et al.* 2018). In another study, it was stated that two people with ID were involved in the decision-making process but it was not specified how (Van Thiel *et al.* 1997). In five studies, it was not clear how the signals, values and goals of care of people with ID were weighted in the decision-making process (Van Thiel *et al.* 1997; Lohiya *et al.* 2003; Wagemans *et al.* 2013a; Wagemans *et al.* 2013b; Tuffrey-Wijne *et al.* 2018).

Seven studies stated the importance of including the people who care for and about a person with ID in the decision-making process (Van Thiel *et al.* 1997; Wagemans *et al.* 2013a; Wagemans *et al.* 2013b; Bekkema *et al.* 2015; Watson *et al.* 2017; Tuffrey-Wijne *et al.* 2018; Zaal-Schuller *et al.* 2018). Judicial regulations around decision-making for people with ID who lack capacity vary. In the Netherlands (where most studies were based), parents or other legal representatives have to make decisions for persons with ID who lack decision-making capacity, although a physician remains ultimately responsible for the medical care as provided (Van Thiel *et al.* 1997; Lohiya *et al.* 2003; Wagemans *et al.* 2010; Wagemans *et al.* 2013a; Wagemans *et al.* 2013b; Zaal-Schuller *et al.* 2016; Zaal-Schuller *et al.* 2018). Watson *et al.* (2017) stated that to properly represent the interests of a person with severe or profound ID an emotionally involved support network is needed. They developed a continuum of so-called relational closeness tools that can be used to find out who is close to a person with severe or profound ID (Watson *et al.* 2017). To support such networks, they developed a supported decision-making framework (Watson *et al.* 2017). They characterise supported decision-making 'as a process of enhancing the

decision-making capability of people with severe or profound intellectual disability through collaborative support from a group of people in the relevant person's life who know them. An important component of this approach is the use of a circle of support, a group of key members of the concerned person's life who have a good understanding (or are committed to developing one) of the person's life history, personal characteristics and their preferences' (Watson 2016).

*Choice talk: summarise, offer choice and patient formulates treatment aims*

In two studies, people with ID were informed about their situation and their prospects (Van Thiel *et al.* 1997; Tuffrey-Wijne *et al.* 2018). In one study, this was done by 'sufficiently informing her at her own level' (Tuffrey-Wijne *et al.* 2018). In another study, this process remained unclear (Van Thiel *et al.* 1997). In the eight other studies, patient representatives and physicians formulated treatment aims without direct involvement of people with ID (Lohiya *et al.* 2003; Wagemans *et al.* 2010; Wagemans *et al.* 2013a; Wagemans *et al.* 2013b; Bekkema *et al.* 2015; Zaal-Schuller *et al.* 2016; Watson *et al.* 2017; Zaal-Schuller *et al.* 2018).

Watson *et al.* (2017) described how available treatment options for a person with a profound disability were explored by a group of people who knew him very well (Watson *et al.* 2017). Zaal-Schuller *et al.* (2016) described how 17 parents anticipated the recurrence of serious illness of their children. 'Almost half of them' believed that it would have been easier to discuss end-of-life decisions with the physicians earlier, when their child was still in a stable condition (Zaal-Schuller *et al.* 2016).

According to one study, parents and physicians agreed about three elements being key to quality of life of children with profound intellectual and multiple disabilities: (1) the ability to enjoy themselves, (2) the absence of physical problems and (3) comfort (Zaal-Schuller *et al.* 2018). Some parents thought that not all important aspects of quality of life had been fully explored in discussions with care professionals. Physicians, on the other hand, reported they already knew parents' views on quality of life and were therefore not discussing it (Zaal-Schuller *et al.* 2018). Another study described how instead of

parents, an ethics committee participated in choice talk (Lohiya *et al.* 2003).

*Option talk: personalised treatment aims are discussed*

According to one study, physicians and parents agreed that disagreements between physicians and parents could ultimately improve the end-of-life-decision-making process, because these enabled the exploration of alternative treatments (Zaal-Schuller *et al.* 2016). Another study, based on nine interviews with ID physicians, stated that wishes of relatives weighed heavily when discussing treatment options (Wagemans *et al.* 2013a). In eight out of nine cases presented by Wagemans *et al.* (2013a), physicians followed the relatives' wishes about treatment choice. Parents and physicians indicated that if an invasive treatment was not expected to lead to a significant improvement of the child's quality of life, they would rather withdraw or withhold that treatment (Wagemans *et al.* 2013b; Zaal-Schuller *et al.* 2018).

Lohiya *et al.* (2003) reported how difficult it is when people with ID have never been able to express their values or preferences (Lohiya *et al.* 2003). Watson *et al.* (2017) explained that, in terms of roles, people with ID express their preferences (e.g. by behaviour; eye movement; vocalisation; self-harm or facial expression), whereas the professionals or relatives need to respond to these preferences by interpreting and acknowledging them.

*Decision talk: focus on preferences, based on the patients' values, goals of care and treatment aims, decide*

If people with ID were involved in decision-making processes, it was not clear how the physician helped them to understand their situation (Van Thiel *et al.* 1997; Tuffrey-Wijne *et al.* 2018). In two studies, people with ID were not involved in the decision-making process because of their lack of decisional competence (Wagemans *et al.* 2013a; Zaal-Schuller *et al.* 2018).

In four studies, the difficulties patient representatives may experience when having to decide between various options, including feeling morally responsible for end-of-life decisions they have to make, were shown (Wagemans *et al.* 2010; Wagemans *et al.* 2013a; Wagemans *et al.* 2013b; Bekkema *et al.* 2015).

*Evaluate: evaluate the shared decision-making process and prepare a treatment plan*

In one study, when asked about the provision of information, parents stated they felt a lack of information during the end-of-life decision-making process (Zaal-Schuller *et al.* 2016). When they were provided with information they felt they lacked the necessary medical background to put that information in the right context. However, parents felt the physician took them seriously as being experts about their children and allowed them to influence the decision-making process (Zaal-Schuller *et al.* 2016). Based on 16 interviews with patient representatives, Wagemans *et al.* (2013b) indicated that patient representatives found support of a doctor very important in the decision-making process. According to Wagemans *et al.* (2013a) physicians sought consensus with relatives and paid care staff. In this process, physicians often gave greater weight to a good relationship with relatives and paid care staff than to their own assessment of the best interest of the person with ID.

## Discussion

This review is the first to provide an overview of how decision-making with people with IDs in the last phase of life is practiced. The results show us that making decisions together with people with ID is not common practice. We found that people with ID participated in the decision-making process in only 2 out of 10 studies (Van Thiel *et al.* 1997; Tuffrey-Wijne *et al.* 2018). Where people with ID participated, it was largely unclear how this process was enabled and what kind of support, if any, was provided to the people with ID.

Most authors stated that relatives and care staff know persons with ID best (Van Thiel *et al.* 1997; Wagemans *et al.* 2013a; Wagemans *et al.* 2013b; Bekkema *et al.* 2015; Watson *et al.* 2017; Tuffrey-Wijne *et al.* 2018; Zaal-Schuller *et al.* 2018). They know how to communicate with the person with ID and how to interpret the signals they are giving (Bekkema *et al.* 2015; Watson *et al.* 2017). By using this information, it is possible to act on the preferences of people with ID, even if they themselves cannot articulate them clearly, and to make decisions that match their wishes and preferences (Watson

*et al.* 2017). That is why good communication between relatives and professionals is important (Wagemans *et al.* 2013a; Bekkema *et al.* 2015; Zaal-Schuller *et al.* 2016; Watson *et al.* 2017). Using documentation such as videos and diaries can help to match decisions with the preferences of people with ID (Watson *et al.* 2017), even when they cannot participate actively in the decision-making process, or do not have sufficient decision-making capacity (Watson *et al.* 2017).

The existing literature offers few good examples of SDM models for people with ID. Based on Elwyn *et al.* (2012) SDM model, van de Pol *et al.* (2016) developed a model for SDM with frail older people. This takes into account co-morbid conditions and involvement of relatives and considers decision-making as a process rather than a one-off event. The model could provide a good basis for SDM for people with ID. Van de Pol's model does not pay attention to eliciting values that are important for the patient or how to address decision-making capacity.

Watson *et al.* (2017) developed a supported decision-making framework for people with ID. This framework ensures that if people with ID are not able to participate in the decision-making process themselves, a key group around the person with ID participates on behalf of them, keeping the values and preferences of the person with ID in mind. Research into best practices with regard to SDM in the last phase of life shows some examples about other vulnerable populations. One study focused on people with dementia and their caregivers and the extent to which housing decisions matched with an interprofessional SDM approach (Garvelink *et al.* 2018). This study indicated that honesty, timely communication and advance care planning helped to better align decisions with preferences of patients. This research can potentially be used as a basis for the development of an aid to support SDM in people with ID in the last phase of life. The supported decision-making model of (Watson *et al.* 2017) could also be used as foundation to develop an SDM model for people with an ID. New research could focus on making this model suitable for people with ID with different levels of participation abilities. For future research, we would recommend that people with an ID are involved in the development and implementation of new approaches considering

decision-making with people with an ID. The target group itself can provide valuable input about what does and does not work for them.

### Implications for practice and research

Adequate SDM processes are underpinned by good relationships between physicians and relatives and care staff of people with ID (Wagemans *et al.* 2013a; Bekkema *et al.* 2015; Zaal-Schuller *et al.* 2016; Watson *et al.* 2017). Defining clear roles and responsibilities for everybody involved could improve the process of developing these relationships (Wagemans *et al.* 2013b). When parents and care staff build a strong relationship well before there is a crisis or a need for important end-of-life decisions, they can better collaborate as a team when death is approaching (Bekkema *et al.* 2015). There is a need for cooperation in building up a shared understanding of the signals and needs of a person with ID. To make a good decision, professionals and relatives should be attentive to the expression and signals of need and distress of the person with ID (Bekkema *et al.* 2015; Watson *et al.* 2017). Support may be even more necessary since people with ID may have limited experience in making important decisions that will affect their lives and may lack the necessary skills to make end-of-life decisions (Tuffrey-Wijne *et al.* 2018).

### Strengths

This review sheds light on an important and timely concept. It also contributes to answering current questions around advance care planning and end-of-life-decision-making with people with ID (Wagemans and Van Wijmen 2014; Voss *et al.* 2017; Wagemans and van Bokhoven 2018). We systematically searched the electronic databases in collaboration with a biomedical information specialist. Thereby, two researchers (HN and IK) independently screened all abstracts for inclusion. Another strength is that we followed the PRISMA Scoping Review Guidelines (Tricco *et al.* 2018), which ensured complete and transparent reporting of our scoping review. The use of van de Pol's SDM model as a framework ensured that we used an inclusive conceptualisation of SDM.

### Gaps and deficiencies

All studies were conducted in high-income countries, with a high proportion of Dutch studies ( $n = 8$ ). This limits the generalisability of the combined study results; because of differences in healthcare systems and cultures, it is unknown to what extent the results of this review can be generalised to other countries. The legislation around capacity and decision-making varies across countries. In addition, most people with ID received residential care, the number of participants was limited in most studies and there was generally little variation in the level of ID.

Furthermore, the last phase of life was not clearly defined in the included studies, and its interpretation may therefore differ per study. In addition, none of the studies found included a definition of SDM. These results are indicative of decision-making in the palliative care context being still in its infancy.

### Conclusion

People with ID do not often actively participate in decision-making processes in their last phase of life, and their opinion about not being involved is unclear. Although it is emphasised in the literature that people with ID should be involved in decision-making in the last phase of life, a uniform best practice about what this should look like is lacking. On the basis of the results, we recommend developing an aid that specifically supports systematically taking preferences of people with ID in the last phase of life into account. As indicated in the literature, even if a person is not able to actually participate in the decision-making process, decisions can be aligned to the values and preferences of a person with ID (Watson *et al.* 2017). This can be achieved by involving the inner circle around the person with ID and by looking at the life history and earlier medical experiences of the person with ID. To make good decisions, professionals and relatives should be attentive to the expression and signals of needs and distress a person with ID is giving. A good relationship between relatives and professionals is essential to ensure good end-of-life care in the best interest of the person with ID. Further research should be conducted to investigate what role people with ID see for themselves in SDM around end-of-life decision-making in the last phase of life.

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## Conflict of interest

The manuscript, or part of it, has neither been published nor is currently under consideration for publication by any other journal. All authors (Hanna Noorlandt, Michael Echteld, Irene Tuffrey-Wijne Dederieke Festen, Cis Vrijmoeth, Agnes van der Heide and Ida Korfage) have read the manuscript and approved its submission to the Journal of Intellectual Disability Research. They all declare that they have no competing interests.

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## Supporting Information

Additional Supporting Information may be found online in the supporting information tab for this article.

## Data S1. Supporting Information