Self-Management and Advance Care Planning at the End of Life

Rik Stoevelaar

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Zelfmanagement en advance care planning in de laatste levensfase

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PART II: ADVANCE CARE PLANNING IN PATIENTS WITH AN IMPLANTABLE CARDIOVERTER DEFIBRILLATOR


1

CHAPTER 1

General introduction

Until recently, healthcare was predominantly based on the biomedical model(1, 2). This model focused on illness and treatment from a biomedical perspective, and therefore placed a central role on the knowledge of the healthcare professional(3). Healthcare professionals tended to make decisions about treatments in the best medical interest of the patient, but made these decisions mostly unilaterally and based on their own knowledge, often without consulting the patient, even if the patient was mentally competent(2, 4). This resulted in a sometimes asymmetrical relationship between the patient and the healthcare professional(5, 6). Nowadays, there is a larger emphasis on the individual choice and autonomy of the patient (5). A patient is no longer only considered to be a mere collection of biological symptoms and a passive recipient of care, but is approached based on the biopsychosocial model of health(7). This biopsychosocial approach entails that an illness exists on interacting biological, psychological and social levels, making the illness experience unique for each patient(8-10). Many organisations, including the World Health Organization (WHO), endorse this patient-centred approach, and recommend to place more emphasis on the needs and preferences of the individual patient(11, 12). A more equal relationship between the patient and the healthcare professional is encouraged, and shared decision-making, defined as "an approach were clinicians and patients make decision together using the best available evidence, and where patients are encouraged to consider options and the likely benefits and harms of each, to achieve informed preferences and decisions"(13, 14), is viewed as an important component in many decisions made about treatment and care(1). In this approach, a major role and responsibility is placed on the patient. The patient is expected to be an expert on his or her health and an active user of care. However, this delegated proactive role of the patient can place high demands on the patient: The patient has to organise and coordinate care in an increasingly fragmented, specialised and complex healthcare system, and has to do this alongside the demands of daily life(15, 16). It is not known to what extent patients are willing and able to take this active role in their care(17, 18).

Self-management

In 2011, a group of international health experts proposed a new definition of 'health', in which health is described as "*the ability to adapt and to self-manage in the face of social, physical, and emotional challenges*" (19). Self-management in this sense can be defined as "*the individual's ability to manage the symptoms, treatment, physical and psychosocial consequences and lifestyle changes inherent in living with a chronic condition*"(20). One cannot not self-manage. If a patient decides not to engage in some sort of health behaviour, this can be considered 'self-management' as well(21). The term 'self-management' was first used in 1976 by the American psychologist Thomas Creer and colleagues in an article about rehabilitation and childhood asthma(21, 22). They based this term on the early writings of the Canadian psychologist Albert Bandura, and used the term self-management to indicate that the patient has an active role in his treatment(21). Corbin and Strauss built on this work, and described how a patient has to self-manage in three different areas (21, 23, 24): 1) the illness, or 'medical management', such as taking medication and symptom management; 2) everyday life, or 'maintaining, changing, and creating new meaningful behaviours or life roles', such as household activities, work and relationships; and 3) their 'biography'. Biography relates to identifying and maintaining an identity over a life course. An illness might potentially disrupt the patient's concept of the self, and the patient has to make the illness part of his or her identity, by coming to terms with one's own mortality and creating a new meaning in life(25). Self-management is thus a comprehensive concept, which is incorporated in the new proposed definition of health. A study of 2078 Dutch healthcare professionals from different professions (physicians, nurses and physiotherapists), patients, policymakers and researchers showed that the majority of respondents have positive associations with the new definition of health and think it is positive that it refers to self-management(26). However, 42% of the respondents are in doubt whether all patients are capable of self-management(18, 26).

Self-management gained increasing attention in research and practice(20). Studies on self-management largely focus on patients with chronic illness, such as arthritis, diabetes or asthma(20, 21). These studies showed that learning patients to effectively self-manage their illness led to a better self-reported health, less distress and fatigue, higher energy levels, more illness-related knowledge, better symptom management and less experienced limitations in social participation(20). Also, healthcare costs dropped(27). Less is known about the self-management of patients with advanced illness. Patients with advanced illness might face problems that are unique for this patient population, such as a large emotional, social and spiritual impact due to the knowledge that death is nearby (28, 29), and an uncertain illness trajectory with periods of experiencing multiple symptoms that could result in suffering (30, 31).

Advance care planning

An expression of self-management which may be relevant for patients with advanced illness is advance care planning (ACP). ACP can be defined as "*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*"(32). It consists of three key elements: 1) reflection on wishes and preferences for future medical treatment and care; 2) discussing these with healthcare professionals and relatives; and 3) recording and, if necessary, reviewing these preferences. ACP is relevant for patients with advanced illness, since patients in the last phase of life often have to make numerous decisions on treatment and care.

The term ACP was first used in the late 1980s and early 1990s(33-36) . Originally, it focused on the patient's completion of written documents, such as advance care directives or do-not-resuscitate orders, to be used in case the patient lost mental capacity in order to make decisions for him or herself(32). Nowadays, ACP is no longer seen as the mere completion of these documents, but as a process of thinking and talking about wishes and preferences at the end of life, with the option of drawing these up in an advance care directive(37, 38). ACP empowers patients to formulate their preferences about treatment and care, based on their values and beliefs. The process of ACP can both be used to extend the decisional capacity of the patient when he or she becomes mentally incompetent, but can also serve as a basis for patients who retain their mental competence(34). ACP helps patients thinking about and preparing for death (37, 39).

A systematic review on the effects of ACP on end-of-life care by Brinkman-Stoppelenburg and colleagues(34), showed that ACP has a positive impact on end-of-life care and quality of life. Research on ACP often focusses on patients with cancer(34). ACP could be very beneficial to other groups as well, but some research has shown that other groups of patients, such as those with cardiovascular disease, engage in ACP less often than patients with cancer(40-44). However, little research has been done on ACP in non-cancer populations(45, 46).

Cancer and cardiovascular disease

In this PhD thesis, I will focus on two populations: patients with cancer in an advanced stage, and patients with cardiovascular disease, in particular those with an implantable cardioverter defibrillator (ICD). Cancer and cardiovascular disease are currently the two main causes of death worldwide(47). For both patient groups, self-management and ACP might be very important and relevant, due to the far-reaching implications of the illness on everyday life and due to the extended illness trajectory(48). In the following section, I will briefly describe both patient groups.

Advanced cancer

Cancer is a common diagnosed illness(47). On January 1st, 2018, more than 600.000 people in the Netherlands had some sort of cancer, with the most common types being breast cancer (136.100 patients), skin cancer (109.500 patients), prostate cancer (83.800 patients) and colon cancer (80.500 patients)(49). In 2018, 47.000 people died of cancer, which makes it the most common cause of death in the Netherlands, accounting for 30% of all deaths(50). The illness trajectory of advanced cancer is usually rather predictable, and is characterised by a gradual and stable phase with little deterioration in health which can take several years, after which a sudden and rapid decline in health will take place, which will eventually end with death (Figure 1)(48).

Advanced cancer is cancer that cannot be cured anymore(51). This does not directly mean that the cancer is not treated. Rather, treatments no longer focus on curing the illness, but have a palliative approach: slowing down the illness progression and prolonging life, controlling symptoms and maintaining quality of life(52). Advanced cancer greatly disrupts the patient's life, with many symptoms in physical, psychological, social and spiritual domains, such as pain, fatigue, lack of energy, depression, anxiety, and spiritual distress, which can range from mild to very severe(53-55).

Self-management is important for patients with advanced cancer to deal with the consequences the illness has on daily life(52, 56). However, research on self-management of patients with cancer usually focusses on cancer in non-advanced stages. Living with advanced cancer may be very different, since the patient is living with the knowledge of having a shorter and uncertain life expectancy, an inevitable deterioration in health and often a high symptom burden(29, 31). Also, due to new treatments and technology, patients with advanced cancer live longer than before, and as a consequence need to deal with uncertainty and the consequences of the illness on a longer term(57, 58). Patients often spend the largest part of their time at home and not in contact with their healthcare professionals, trying to live with the consequences of their illness and treatment. However, this part is relatively unknown to clinicians and researchers. Better insight in the self-management of patients with advanced cancer and the difficulties they experience will shed more light on the complexity and on the necessity and importance of self-management. Also, better insight is important to be able to provide the best support for these patients(56). Therefore, I will examine self-management of patients with advanced cancer in this thesis.

Cardiovascular disease – the implantable cardioverter defibrillator

Cardiovascular disease is, after cancer, the second leading cause of death in the Netherlands. In 2018, 38.000 patients died of the consequences of cardiovascular disease, accounting for 25% of the deaths in the Netherlands(50). Chronic cardiovascular disease is characterised by episodes of acute illness, from which patients may recover (Figure 1)(48). In these episodes, patients are often hospitalised and receive intensive treatment. Patients usually survive numerous episodes of acute illness, although a decline in health and functioning is apparent in the long term. Although a patient could die in any of these episodes, the eventual time of death is uncertain, which can give the feeling of a sudden and unexpected death once the patient dies.

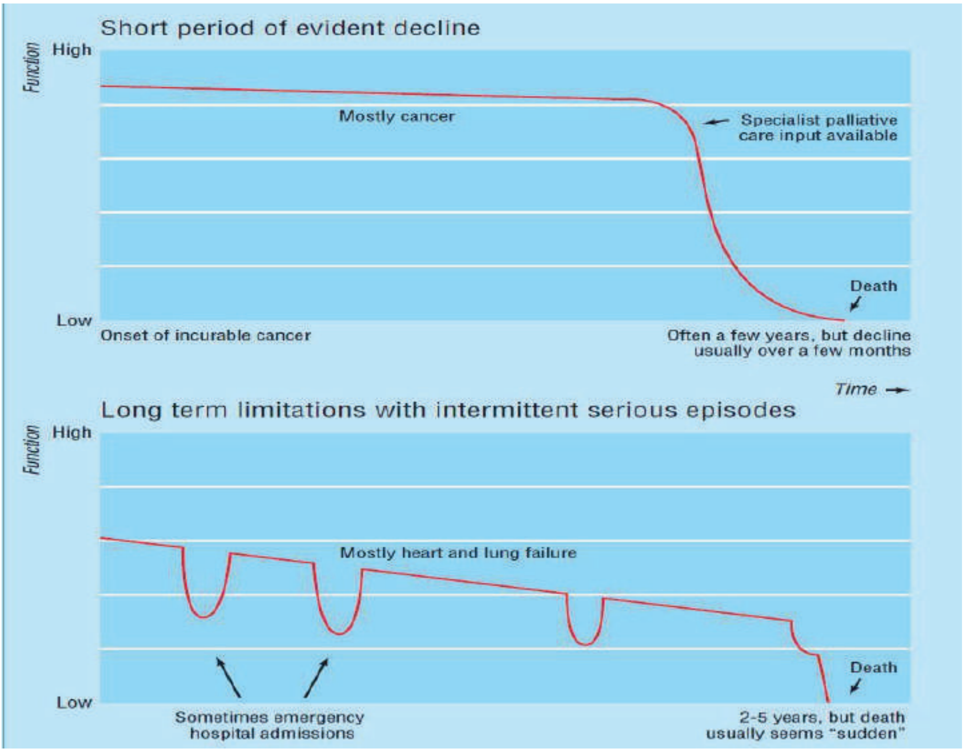

Figure 1. Illness trajectories of cancer (first graph) and heart and lung failure (second graph), adapted from Murray et al. 2005(48)

A proportion of patients with cardiovascular disease have an ICD implanted. The ICD is a small electronic device, developed by the cardiologists dr. Michel Mirowski and dr. Morton Mower in the late 1960s (Figure 2) (59, 60). It is implanted under the skin of the chest or abdomen of the patient, and has up to three electrodes or 'leads' running into the heart(61). It constantly analyses the hearth rhythm and intervenes when a ventricular arrhythmia arises, by giving rapid electronic pulses (anti-tachycardia pacing; ATP), or a strong electrical shock, thereby preventing death(62). The ICD also has a pacemaker function. The ICD is implanted for secondary prevention in patients who have experienced a cardiac arrest prior to receiving the ICD, or, more often, for primary prevention in patients who have a higher risk of a cardiac arrest, but not yet experienced one(63). It is estimated that in the Netherlands, 55.000 people have an ICD, and this number is rising(64, 65). In 2018, 6413 new ICDs were implanted(66).

Although the ICD is aimed at prolonging life, patients will eventually die, either due to a deterioration of their underlying heart disease, other illnesses such as cancer, or old age(67). For ICD patients who are in the last phase of life, the ICD could potentially complicate the dying process, by delivering inappropriate shocks. A landmark study from the USA, performed by Goldstein and colleagues in 2004, showed that up to 27% of patients experienced a shock in the last month before death, and 8% of patients experienced shocks in the last minutes(68). Another study from Sweden from 2014 showed that this percentage was even higher: up to 31% of patients received an ICD shock in the last 24 hours of life(69).

ICD patients in the last phase of life may be faced with the decision whether or not to deactivate the shock function of their ICD at some point, to avoid inappropriate shocks. However, this is not a conversation easily started or a decision easily made. At the same time, having conversations with patients is crucial to make them aware of the implications of having an active ICD at the end of life. Multiple guidelines have elaborated on this, and recommend to timely and frequently discuss ICD deactivation throughout the illness trajectory(67, 70, 71). It is however unknown to what extend these conversations are held, and whether these lead to an increase in deactivation. ACP could potentially be helpful for patients with an ICD to think, talk, and record their preferences about ICD deactivation. ACP has rarely been studied in cardiology, and specifically in patients with an ICD, while ACP could be very relevant for these patients(45). Therefore, in this thesis, I will examine the last phase of life and the practice of ACP in a population of patients with an ICD.

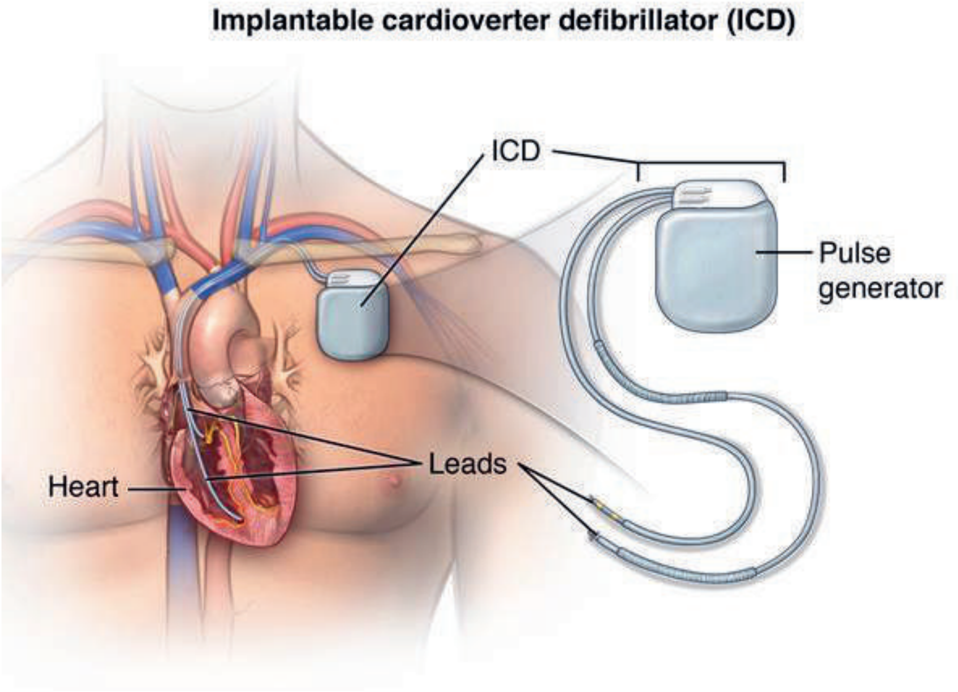

Figure 2. A dual lead ICD in place(72)

Aims and research questions

The overall aim of this thesis is to provide insight into the self-management behaviours and ACP practices of patients living with advanced illness. This thesis includes studies on self-management in patients with advanced cancer and studies on ACP in patients with cardiovascular disease, specifically those with an ICD.

Part I of this thesis addresses self-management in a population of patients with advanced cancer. The aim of this part is to better understand the consequences of having advanced cancer and patients' self-management to deal with these consequences. Further, the aim was to examine healthcare professionals' experiences and attitudes towards patients' self-management, and their perceived roles in self-management support.

The research questions of part I of this thesis are:

1. How do patients with advanced cancer deal with the consequences of their illness and its treatment?
2. What factors influence self-management of patients with advanced cancer?
3. What are the experiences and attitudes of healthcare professionals towards self-management of patients with advanced cancer, and what self-management support roles can be distinguished?

Part II of this thesis focusses on patients with an ICD. The aim of this part is to examine the last phase of life of patients with an ICD, and to examine the experiences of patients, relatives, and healthcare professionals with ACP conversations about whether or not to deactivate the shock function of the ICD.

The research questions of part II of this thesis are:

4. What is the impact of having an active ICD in the last phase of life?
5. How and when are decisions about ICD deactivation made, and what factors influence these decisions?
6. What barriers do patients and relatives experience towards ACP conversations about ICD deactivation?
7. What barriers do healthcare professionals experience towards ACP conversations about ICD deactivation?

Outline of this thesis

Part I of this thesis (chapter 2 to 4) aims at describing self-management in a population of patients with advanced cancer. **Chapter 2** gives an extensive overview of the literature on this topic and covers the self-management strategies patients with advanced cancer use on which domains. **Chapter 3** covers a large qualitative study in Dutch patients with advanced cancer, to explore the consequences of living with advanced cancer, to explore their self-management, and to examine the factors that influence self-management. **Chapter 4** covers the experiences of healthcare professionals with self-management support of patients with advance cancer.

Part II of this thesis (chapter 5 to 9) aims at describing ACP in a population of patients with an ICD. **Chapter 5** presents a systematic review on ICD shock incidence and the impact of these shocks at the end of life. **Chapter 6** covers a retrospective case study on a Dutch population of deceased ICD patients, and describes the ICD shock incidence, the occurrence of deactivation conversations and deactivation rates. **Chapter 7** and **chapter 8** describe the experiences of ICD patients and their relatives with the last phase of life, their experiences with ACP, and the barriers towards ACP. **Chapter 9** covers a viewpoint which elaborates on the barriers healthcare professionals experience concerning ACP in patients with an ICD, and how these could be overcome. The main findings of chapter 2 through 9 are discussed and reflected on in **chapter 10** and are summarised in **chapter 11**.

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2

CHAPTER 2

Self-management of patients with advanced cancer: A systematic review of experiences and attitudes

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ABSTRACT

Background

Patients with advanced cancer are increasingly expected to self-manage. Thus far, this topic has received little systematic attention.

Aim

To summarise studies describing self-management strategies of patients with advanced cancer and associated experiences and personal characteristics. Also, to summarise attitudes of relatives and healthcare professionals towards patient self-management.

Design

A systematic review including non-experimental quantitative and qualitative studies. Data were analysed using critical interpretive synthesis. Included studies were appraised on methodological quality and quality of reporting.

Data sources

MEDLINE, Embase, Cochrane Central, PsycINFO, CINAHL, Web of Science and Google Scholar (until 11 June 2019).

Results

Of 1742 identified articles, 31 moderate-quality articles describing 8 quantitative and 23 qualitative studies were included. Patients with advanced cancer used self-management strategies in seven domains: medicine and pharmacology, lifestyle, mental health, social support, knowledge and information, navigation and coordination and medical decision-making (29 articles). Strategies were highly individual, sometimes ambivalent and dependent on social interactions. Older patients and patients with more depressive symptoms and lower levels of physical functioning, education and self-efficacy might have more difficulties with certain self-management strategies (six articles). Healthcare professionals perceived self-management as desirable and achievable if based on sufficient skills and knowledge and solid patient–professional partnerships (three articles).

Conclusion

Self-management of patients with advanced cancer is highly personal and multifaceted. Strategies may be substitutional, additional or even conflicting compared to care provided by healthcare professionals. Self-management support can benefit from an individualised approach embedded in solid partnerships with relatives and healthcare professionals.

INTRODUCTION

Learning that one's cancer has progressed to an advanced and incurable stage is for most patients and their relatives an overwhelming experience that often includes shifting from curative treatment to focusing on life extension and/ or quality of life. Because patients have to live with the prospect of impending death and are increasingly unable to continue their daily activities and fulfil their usual social roles, they need to deal with considerable emotional, psychosocial and lifestyle consequences(1). They do this in the face of increasing multidimensional symptoms, such as fatigue, pain, anxiety and depression(1–4). Although usually, many healthcare providers are involved in medical care(5,6), most of the time, patients, together with their relatives, need to manage a huge part of their care and lives them- selves. This can be highly complex.

In recent years, patients have been increasingly stimulated to actively manage their health: healthcare policies have shifted towards out-of-hospital delivery of care, partly driven by increasing numbers of patients(7), workforce challenges(8) and a tendency to reduce costs(9). Self-management has frequently been studied in the context of chronic diseases, such as diabetes. In this context, self-management has been defined as '*the person's ability to manage the symptoms, treatment, physical and psycho-social consequences and lifestyle changes inherent in living with a chronic condition*'(10). Key to this definition is that self-management involves more than management of problems in the medical domain. A growing body of evidence demonstrates that people living with chronic conditions might use a wide range of self-management strategies, such as adhering to a special diet and dealing with emotions(10–18).

Because advanced cancer is generally characterised by a shorter prognosis compared to chronic diseases, such as diabetes, and increased complexity of healthcare (including rapid medical-technological developments, such as immunotherapy), self-management domains and strategies observed among patients with chronic diseases can- not self-evidently be extrapolated to patients with advanced cancer. Yet, self-management of patients with advanced cancer has received surprisingly little systematic attention. Several studies have assessed the topic, but among these studies, there is a lack of conceptual clarity (e.g. inconsistent use of terms like self-management, self-care and self-help; either a broad focus or a focus on certain domains, such as symptom self-management) and a large degree of heterogeneity regarding study designs and patient populations (e.g. all cancer types vs specific cancer types). Previous reviews have shown that patients with advanced cancer engage in multiple different self- management behaviours, including psychosocial and emotional strategies, to manage the end of life, such as preparing for death(19,20). However, one systematic review was published in 2009(20), whereas especially in the past decade, healthcare policies have increasingly steered towards out-of-hospital care and patient self-management, presumably affecting self-management experiences. The other review concerns a scoping review that predominantly focuses on self-management support interventions and lacks quality appraisal of the included studies(19) In addition, both reviews only assess patient perspectives, while it is also relevant to include perspectives of relatives and healthcare professionals(21–25). Comprehensive insight in self-management of this vulnerable patient population is needed to develop healthcare policies and self-management support programmes that are tailored to the needs and abilities of patients and their relatives, while also fitting in the healthcare system. Hence, this integrated systematic review of empirical studies aimed to obtain this insight by thoroughly analysing the non-experimental evidence currently available. For the population of patients with advanced cancer, we therefore examined the following:

1. The concept of self-management and its domains;
2. Patients' self-management strategies and corresponding experiences;
3. Patient characteristics that might be associated with the use of self-management strategies;
4. The attitudes of relatives and healthcare professionals towards patient self-management.

METHODS

Design

Following the principles of a mixed research synthesis(26), we conducted a systematic review(27) of published non- experimental quantitative and qualitative research. We used the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines for reporting and presentation of the flow of information through the different phases of the review(28).

Data collection

With help of biomedical information specialists (G.d.J. and W.B.), systematic electronic searches were per- formed in MEDLINE (Ovid), Embase, Cochrane Central, PsycINFO (Ovid), CINAHL (EBSCO), Web of Science and Google Scholar from inception until 11 June 2019. Supplementary Table 1 lists the search terms. Search components consisted of Medical Subject Headings (MeSH terms) or equivalent and free text words related to (1) self-management (identified using MeSH trees and search strings of previous research) and (2) advanced cancer. No automatic restrictions were placed on study type and year and language of publication. Articles were entered in Endnote, and duplicates were removed.

Study selection

Articles were included when they met the inclusion criteria (see Box 1). To determine eligibility of articles that purported to be on self-management (or a related search term), we adapted Barlow's self-management definition to the context of advanced cancer(10). The advanced cancer criterion was considered met when articles specifically addressed a population of patients with cancer that was 'unlikely to be cured'.29 Healthcare professionals may also use the terms 'secondary', 'metastatic', 'terminal' or 'progressive' cancer to describe it. Studies were only included if they reported results specifically for this patient group (i.e. studies addressing a mixed population with various cancer stages were excluded if they did not stratify results according to cancer stage). We selected articles with non-experimental quantitative, qualitative and mixed-method study designs(26).

Two reviewers (K.d.N. and S.I.v.D.) independently used a stepwise procedure to identify relevant articles. In case of disagreement, consensus was reached through discussion with a third reviewer (J.A.C.R.). Study selection was carried out with the online software Covidence(30). Articles were first screened based on title and abstract. Subsequently, remaining articles were screened based on full text. Articles of references selected for full text evaluation were downloaded, or, if not electronically available, requested from the first author. If full text articles were excluded, the first of the hierarchical inclusion criteria not satisfied (see Box 1) was considered the main reason for exclusion.

Box 1. Inclusion criteria for study selection.

1. Articles concerning self-management (i.e. 'the strategies used by persons to manage the symptoms, treatment, physical and psychosocial consequences and lifestyle changes inherent in living with advanced cancer')(10).
2. Articles concerning patients with advanced cancer (i.e. 'cancer that is unlikely to be cured')(29).
3. Articles concerning perspectives of patients with advanced cancer, their relatives (not necessarily restricted to family members, but could also include significant others, for example, close friends) and/or healthcare professionals.
4. Articles concerning empirical research with a non- experimental quantitative, qualitative or mixed-methods study design and a sample size of ⩾1 (i.e. no case studies, case reports, reviews and intervention studies)(26).
5. Articles published in the English or Dutch language in peer-reviewed scientific journals (i.e. no conference proceedings, abstracts and posters).
6. Articles concerning adults (⩾18 years) only.

Quality appraisal

Two reviewers (K.d.N. and R.S.) independently appraised the quality of included studies. Methodological quality of quantitative studies was assessed using a modified version of the guidelines of Cochrane Netherlands(31,32). This form consists of the following seven items: (1) research hypothesis, (2) study population, (3) selection bias, (4)exposure, (5) outcome, (6) confounding and (7) general opinion. Each criterion was assigned a score of 1 when it was sufficiently met, a score of 0 when it was insufficiently met or a question mark when it could not be rated due to lacking information. For each of the studies, a total score was calculated (ranging from 0 to 7). The quality of reporting of the qualitative studies was assessed with the consolidated criteria for reporting qualitative research (COREQ) checklist, which is recommended by Cochrane Netherlands(33). The COREQ checklist also includes aspects of methodological quality and is applicable to various types of qualitative research(33,34). It consists of 32 items that evaluate three domains, that is, (1) research team and reflexivity, (2) study design and (3) analysis and findings. Each criterion was assigned a score of 1.0 when it was properly described, a score of 0.5 when it was partially described or a score of 0.0 when it was not or unclearly described. For each of the studies, a total score was calculated (ranging from 0.0 to 32.0). Disagreements were discussed and resolved.

Data extraction and data analyses

To systematically extract data from included articles, we developed a data extraction form. This form included items on general study characteristics and characteristics of the study population. It also included open items about the definition of self-management and the self-management strategies addressed. We also extracted information regarding patients' experiences and characteristics associated with these self-management strategies. In addition, we extracted information concerning the attitudes of relatives and healthcare professionals towards patient self- management. The extraction form was completed by two reviewers (K.d.N. and S.I.v.D.); disagreements were solved by discussion.

Data were analysed using critical interpretive synthesis(35). Departing from the framework of self-management support components developed by Barlow and colleagues in the context of chronic diseases(10), we categorised self-management strategies into domains. Subsequently, we integrated data on self-management strategies with data on corresponding experiences, and compared these findings within and across self-management domains. In addition, we interpreted findings in light of self-management definitions provided by the included studies and com- pared them with findings from the context of chronic diseases.

RESULTS

Study selection

The database search yielded 2935 articles (see PRISMA flowchart in Figure 1). After removal of duplicates, 1742 articles were screened based on title and abstract. The remaining 187 articles were screened based on full text. Finally, 31 articles about 31 unique studies were included for analysis.

Eight of these articles had a quantitative study design and 23 of them had a qualitative study design (see Table 1).

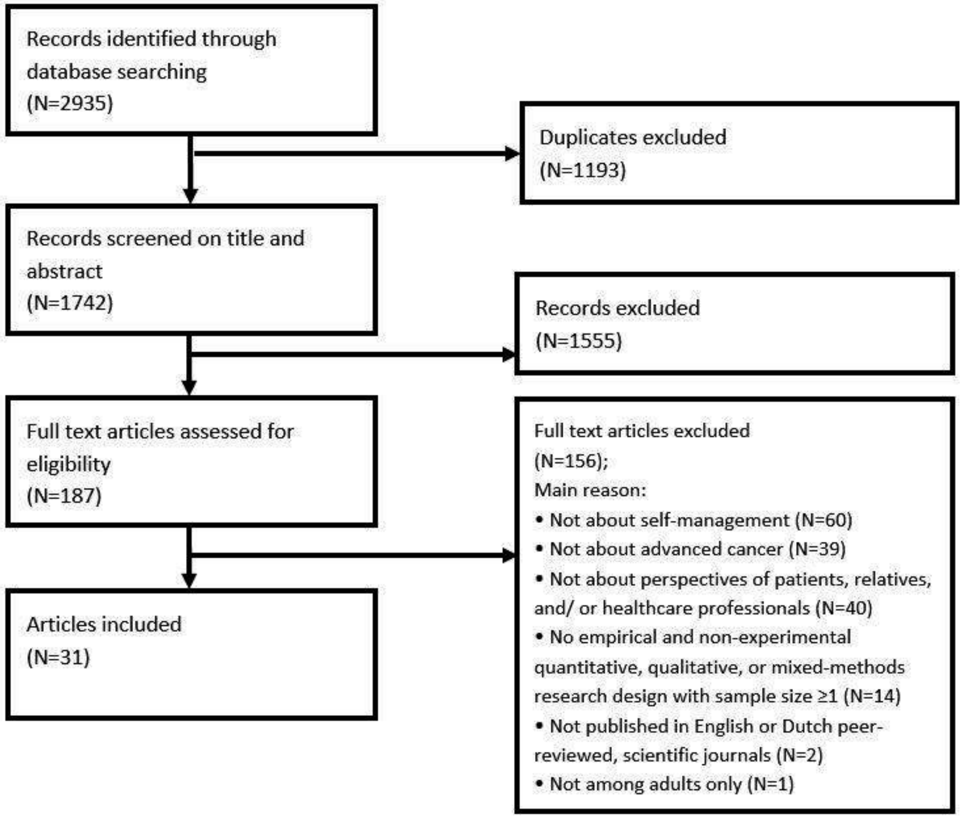

Figure 1. Process of study selection (PRISMA flowchart).

Quality of included studies

For non-experimental quantitative studies, the mean total methodological quality score was 5 out of 7 (range: 4–7; see Table 1). The studies had good ratings on the criteria for ‘Research hypothesis’, ‘Study population’ and ‘Outcome’, but nearly all of them scored poorly on the criteria for ‘Selection bias’ and ‘Confounding’. For qualitative studies, the mean total score for quality of reporting was 19.0 out of 32.0 (range: 12.5–25.0). Almost all qualitative studies had poor ratings on the first domain, ‘Research team and reflexivity’. Generally, studies with the highest scores on the first domain also provided sufficient information on the domains ‘Study design’ and ‘Analysis and findings’, thus resulting in a higher overall quality score.

Study characteristics

Study populations of 28 studies consisted of patients (see Table 1), one study was conducted among healthcare professionals only, and two studies contained both groups. None of the studies assessed the perspective of relatives. All but seven articles described assessment of self-management of patients residing primarily at home(36–42). Except for five studies that were performed in Brazil, China, Thailand and Turkey, respectively(36–38,41,42), studies were conducted in high-income countries, such as Australia (seven studies), Canada (one study), the United Kingdom (eight studies), the United States (five studies) and Scandinavia (five studies). Eighteen studies included various cancer diagnosis groups; the other studies specifically focused on lung cancer(38), breast cancer(36,43–47), prostate cancer(48), hepatocellular cancer(49) or myeloma(43). Some studies focussed on assessing specific self-management strategies, such as medication management(50), or the use of complementary and alternative medicine(36,51)

Table 1. Characteristics of the included articles (31 studies).

First author, year	Country	Study design	Patient population: setting	Patient population: type(s) of advanced cancer	Patient population: sex	Study population(s) (N)	Quality Appraisal [a]
Non-experimental quantitative study design (N=8)							
Alfano, 2014[36]	Brazil	Cross-sectional and prospective study (interviewer-administered surveys)	Cancer hospital	Breast	Female	Patients (N=126)	6
Baile, 2011[67]	United States	Cross-sectional study (self-report surveys)	Home	Various types, i.e.: breast, gastrointestinal, head and neck, lung, lymphoma, melanoma/ skin, urogenital, and other	Mixed	Patients (N=137)	4
Bennett, 2009[56]	United Kingdom	Cross-sectional study (interviewer-administered surveys)	Home	Various types, i.e.: breast, colorectal, gastrointestinal, lung, and urogenital	Mixed	Patients (N=90)	5
Chan, 2016[57]	Australia	Prospective study (interviewer-administered surveys)	Home	Various types, i.e.: breast, colorectal, lung, and prostate	Mixed	Patients (N=152)	4
Correa-Velez, 2003[58]	Australia	Prospective study (interviewer-administered surveys)	Home	Various types, i.e.: bladder, breast, gastrointestinal, lung, melanoma, ovarian, and prostate	Mixed	Patients (N=111)	6
Get-Kong, 2010[37]	Thailand	Cross-sectional study (self-report surveys)	Home and inpatient departments of two cancer centres and one university hospital	Various types; not specified	Mixed	Patients (N=240)	5

He, 2019[38]	China	Cross-sectional study (self-report surveys)	Chest-oncology department of public hospital	Lung	Mixed	Patients (N=261)	6
Norris, 2009[43]	United States	Cross-sectional study (self-report surveys)	Home	Breast	Female	Patients (N=173)	5
Non-experimental qualitative study design (N=23)							
Bennett, 2006[44]	United Kingdom	Semi-structured interviews	Home	Breast	Female	Patients (N=8)	14.5
Campling, 2017 [39]	United Kingdom	Focus groups and interviews	Home and local hospices	Various types, i.e.: bile duct, breast, colon, lung, and other	Mixed	Patients (N=15), Healthcare professionals (N=19)	17
Correa-Velez, 2005[51]	Australia	Semi-structured interviews	Home	Various types, i.e.: bladder, breast, gastrointestinal, lung, melanoma, ovarian, and prostate	Mixed	Patients (N=39)	14.0
Devik, 2013[60]	Norway	Narrative interviews	Home	Various types; not specified	Mixed	Patients (N=5)	17.0
Eliott, 2008[40]	Australia	Semi-structured interviews	Not specified	Various types, i.e.: breast, colorectal, kidney, lung, lymphoma, and other	Mixed	Patients (N=28)	18.0
Erol, 2018[41]	Turkey	Semi-structured interviews	Oncology clinic of university hospital	Various types, i.e.: colorectal, gastric, and lung	Mixed	Patients (N=16)	20
Gibbins, 2014[61]	United Kingdom	Semi-structured interviews	Home	Various types, i.e.: breast, lung, melanoma, mesothelioma, ovary, prostate, and sarcoma	Mixed	Patients (N=12)	17.5

Hansen, 2015[49]	United States	Semi-structured in-depth interviews	Home	Hepatocellular carcinoma	Mixed	Patients (N=14)	23.0
Hopkinson, 2007[54]	United Kingdom	Semi-structured interviews	Home	Various types, i.e.: breast, gastrointestinal, head and neck, lung, prostate, and other	Mixed	Patients (N=30)	12.5
Hughes, 2016[53]	United Kingdom	Focus group interviews	Home	Various types; not specified	Mixed	Healthcare professionals (N=17)	21.0
Johansson, 2006[62]	Sweden	Focus group interviews	Home	Various types, i.e.: bile duct, bladder, breast, and prostate	Mixed	Patients (N=5)	22.5
Johnston, 2012[55]	United Kingdom	Unstructured in-depth interviews	Home	Various types, i.e.: breast, colorectal, gastrointestinal, hematological, lung, and urogenital	Mixed	Patients (N=20), Healthcare professionals (N=20)	18.0
la Cour, 2009[59]	Denmark	Diaries, short-structured interviews, semi-structured interviews (with one third of study sample; face-to-face or by telephone)	Home	Various types, i.e.: breast, colon, lung	Mixed	Patients (N=45)	21.5
Levy, 2015[48]	United Kingdom	Semi-structured in-depth interviews	Home	Prostate	Male	Patients (N=5)	22.0
Lewis, 2016[45]	Australia	Semi-structured in-depth interviews	Home	Breast	Female	Patients (N=18)	18.0
Liu, 2017[42]	China	Semi-structured interviews	Medical oncology department of general hospital	Various types, i.e.: breast, lung, pancreatic, prostate, and other	Mixed	Patients (N=9)	21

McPherson, 2014[63]	Canada	Semi-structured interviews	Home	Various types, i.e.: breast, gastrointestinal, respiratory/thoracic, urogenital, and other	Mixed	Patients (N=18)	21.5
Pacsi, 2015[46]	United States	Conversations	Home	Breast	Female	Patients (N=6)	19.0
Peoples, 2017[66]	Denmark	Structured nterviews	Home	Various types, i.e.: breast, cervix, colon, lung, prostate, and other	Mixed	Patients (N=73)	19.0
Sand, 2009[51]	Norway	Semi-structured interviews	Home	Various types, i.e.: breast, gastrointestinal, lung, melanoma, and urogenital	Mixed	Patients (N=15)	16.0
Schulman-Green, 2011[47]	United States	Semi-structured interviews	Home	Breast	Female	Patients (N=15)	17.5
Stephens, 2014[50]	Australia	Semi-structured in-depth interviews	Home	Myeloma	Mixed	Patients (N=11)	25.0
Thomas, 1999[65]	Australia	Semi-structured in-depth interviews	Home	Various types; not specified	Mixed	Patients (N=19)	17.5

N: Number

[a] Quality scores of non-experimental quantitative studies ranged from 0 to 7, with higher scores indicating higher methodological quality. Quality scores of non-experimental qualitative studies ranged from 0.0 to 32.0, with higher scores indicating better quality of reporting

The concept of self-management and its domains in patients with advanced cancer

Self-management definitions. Twenty-one of the 31 included articles did not explicitly define self-management or any of the related terms. Three of the included articles provided a definition of 'self-management'(47,52,53). Three other articles defined 'self-care'(43,54,55). These definitions are described in Box 2. One of the definitions relates self- management to self-care, describing self-management as 'maintaining ones usual practices of self-care'(55). While some definitions conceptualise self-management as behaviours (or strategies, actions, activities, practices)(43,53–55), others also include patients' self-management skills, such as action-planning and using resources(47,52). Most definitions focus predominantly on the patient, with one of them explicitly emphasising the aim of self-mastering problems rather than relinquishing these to others. Yet, two definitions assign a role for relatives and healthcare professionals as well, describing self-management as a participatory process that may even be undertaken to serve (well-being of) others(54). Finally, the self-management definitions cover various domains of health and functioning and include different self-management outcomes, for example, enhanced quality of life(43,54), maintenance of physical and mental health and/or daily functioning(43,54), reduced symptom burden(53) and increased coping(53).

Self-management domains. Self-management strategies used by patients in the 29 non-experimental quantitative and qualitative studies could be categorised into seven overarching domains: medicine and pharmacology, lifestyle, psychology/mental health, social support, knowledge and information, navigation and coordination and medical decision-making (see Table 2). Five studies pro- vided information about one self-management domain(36,40,51,54,55), five studies reported on two self-management domains(44,56–59) and the remaining 19 studies covered three or more self-management domains.

Box 2. Definitions of self-management or related terms in the included articles.

'Self-management' (three articles):

- 'Self-management has been defined as any behaviour which an individual engages in specifically to try and relieve, minimise or prevent pain or more broadly to cope with their illness'(53).
- 'Self-management involves daily behaviours that individuals perform to handle a health condition, it includes the skills of problem solving, goal setting, decision making, using resources, forming patient- provider partnerships, action planning, and self- tailoring'.47
- 'Self-management has been described as a participatory process where patients and clinicians develop strategies together to equip patients with the skills and knowledge to manage the

impact of the condition, monitor their disease and make effective use of support services outside of the clinical setting'(52).

'Self-care' (three articles):

- 'Self care includes the actions individuals and carers take for themselves, their children and their families to stay fit and maintain good physical and mental health; meet social and psychological needs; prevent illness or accidents; care for minor ailments and long-term conditions; and maintain health and well-being after an acute illness or discharge from hospital'(54).
- 'Self-care can be used as an umbrella term to refer to all activities of self-management. [. . .]. It is defined as "maintaining ones usual practices of self-care – those things that are important and unique to oneself in maintaining ones sense of self. . . being given the means to master or deal with problems, rather than relinquish them to others"(55).
- 'Self-care involves the voluntary use of activities to promote one's own well-being. Self-care has been defined as the range of voluntary activities that an individual uses to maintain life, health and well-being'(43).

Self-management strategies and corresponding experiences of patients with advanced cancer

Non-experimental quantitative results concerning self- management strategies and corresponding experiences of patients with advanced cancer are displayed in Table 3. The results of the qualitative studies are presented in Supplementary Table 2.

Medicine and pharmacology. Medical and pharmacological strategies varied from self-monitoring(49) and self-administering medications and following prescribed treatment regimens(37,47,58,60) to adjusting or discontinuing treatment schedules and taking extra doses of additional (pain) medications(39,41,42,47,50,52,61–63). Patients used self- monitoring to better understand and describe their disease(49). Reasons for not taking medications as prescribed included a desire to alleviate suffering(62), attempts to find the optimal balance between beneficial and adverse (side) effects(39,50,63), fears of tolerance and addiction(63), non-pharmacological alternatives to manage pain(63), preferring 'grip on the pain' over 'becoming pain free'(61), sedative side effects that were experienced to be a 'threat to usual self'(61) and uncertainty about treatment efficacy(49).

Table 2. Self-management domains and self-management strategies used by patients with advanced cancer (29 studies)

Self-management domains Studies addressing the respective domain: (N)	Self-management strategies
Medicine and pharmacology (N=14)[37,39,41,42,47,49-50,52,58,60,61-63,67]	• Monitoring symptoms, bodily changes, treatment effects, and/ or disease risks

	• Self-administering medication • Adhering to prescribed treatment schedules • Adjusting or discontinuing treatment schedules (e.g. taking extra drug doses during breakthrough pain, replacing conventional treatment with alternative therapies, omitting use of medications)
Lifestyle (N=26)[36,37,40-48,51, 52, 54-63, 65 - 67]	• Adjusting nutrition and diet • Adjusting exercise (e.g. exercising more, balancing rest and physical activity) • Practising complementary and alternative medicine (e.g. taking medicinal herbs, practising meditation, Reiki, or homeopathy) • Practising religion • Using relaxation • Performing leisure activities (e.g. doing sports or creative activities) • Maintaining daily routine by adjusting activities (e.g. by taking breaks/ naps during the day, dividing activities into smaller parts, using assistive devices) • Not making any lifestyle changes
Psychology/ Mental health (N=9)[42, 43,45,46,48,55,60-62]	• Keeping a diary • Using mindful self-help strategies (e.g. practising assertive self-talk, focusing on feelings and thought of control, acceptance, and/ or positivity, readjusting purpose, expectations and meaning, channelling thoughts of own death towards future well-being of loved ones) • Doing meaningful (charity) activities (e.g. volunteering, promoting cancer awareness)
Social support (N=22)[37,39, 41, 43-48, 50,52,55-57,59-63,65-67]	• Seeking support from relatives and friends • Seeking support from healthcare professionals • Seeking support from other cancer patients; engaging in (online) support groups • Providing and/ or arranging social support to friends and relatives • Limiting social interactions to certain people or moments (e.g. selective communication, social isolation)
Knowledge and information (N=12)[39,42,45,47-50,52,60,61,65,67]	• Seeking information about disease and/ or treatments • Seeking information about self-care • Avoiding or neglecting information
Navigation and coordination (N=9)[39,42,47,48,50,55,60,62,63]	• Coordinating medical services (e.g. obtaining and exchanging health-related documents)

	• Delegating aspects of care (e.g. obtaining medications, deciding on pain treatment approach) to others • Coordinating and staying in charge of information dissemination to relatives • Making financial and practical plans (e.g. arranging funeral, arranging practical support at home)
Medical decision-making (N=8)[39,46-49,62,65,66]	• Making informed decisions about treatment • Engaging in advance care planning • Short-term goal-setting

N: Number

Lifestyle. Within the lifestyle domain, we identified different self-management strategies. Three non-experimental quantitative studies showed that 24%–56% of the patients used physical exercise to manage symptoms, for instance, fatigue(37, 43,56). Two quantitative studies(37,57) and three qualitative studies(46,52,54) showed that many patients changed their diet (i.e. 70%–82% adopted a balanced diet). The two quantitative studies also indicated that 92% and 89%, respectively, used relaxation to relieve cancer symptoms(37,57). In addition, six quantitative studies showed that 26%–72% of the patients used at least one complementary and alternative medicine modality (classified according to the criteria provided by the National Centre for Complementary and Alternative Medicine)(64), such as acupuncture or homoeopathy(36,37,43,56–58). Both quantitative and qualitative studies provided evidence that patients experienced benefits from using complementary and alternative therapies, such as an improved quality of life(40,51,55,65). Generally, health behaviours were adopted or changed in order to prolong life(40,58), boost strength, energy and immunity(52,58), improve quality of life(40,51,62), increase feelings of control and independence(40,54) and please relatives(40,54). Some patients decided to spend more time on their hobbies or engaged in new hobbies to distract them from disease-related symptoms and concerns(41,42,48,61–63), maintain physical ability48 or get a sense of enrichment(48,59). At the same time, patients indicated to strive for normality and maintenance of usual daily activities: 10 qualitative studies(45–48,55,59–62,66) and 1 quantitative study(67) showed that this was of great importance to most of them among others because it made them feel functional and helped them to maintain role, self and independence. For some patients, continuing life as it used to be was so important that they deliberately chose not to make any major lifestyle changes at all(45). Other patients maintained their daily routine using assistive devices, adjusting activities, dividing tasks into smaller parts or taking more breaks during the day(61–63,66).

Table 3. Non-experimental quantitative study results on self-management strategies and associated experiences and personal characteristics of patients with advanced cancer (eight studies)

First author, year Patient population: type(s) of advanced cancer (N)	Self-management strategies and corresponding experiences	Self-management domains	Patient characteristics associated with self-management strategies
Alfano, 2014[36] Breast cancer (N=126)	At least 1 CAM modality used after diagnosis: 50% • Biologically based practices (e.g. food supplements, fruit juices and homemade milkshakes): 33% • Mind-body interventions (e.g. individual or group prayer and art therapy): 27% • Body-based practices (e.g. massage): 3% • Whole medical systems (e.g. homeopathy): 2% • Energy therapies (e.g. Reiki): 2%	• Lifestyle	
Baile, 2011[67] Various cancer types, i.e.: breast, gastrointestinal, head and neck, lung, lymphoma, melanoma/ skin, urogenital, and other (N=137)	Most concerns reported about (mean score (SD) [a]): • Not being able to do usual activities: 1.83 (1.02) • Caring for themselves: 1.41 (1.13) • Managing treatment issues: 1.37 (1.06) • Getting insufficient information about disease or treatment: 1.04 (1.06) • Maintaining intimate/ sexual relationships: 0.94 (1.09) • Communicating with healthcare professional(s): 1.00 (1.15) • Maintaining relationships with significant others: 0.76 (1.09) Least concerns reported about (mean score (SD) [a]): • Getting insufficient support from others: 0.54 (0.93) • Doing spiritual or religious activities: 0.57 (0.94)	• Medicine and pharmacology • Lifestyle • Social support • Knowledge and information	More self-management concerns among patients with: • more anxiety symptoms: r=0.52 [b] • more depressive symptoms: r=0.40 [b] • lower self-efficacy: r=0.37 [b] (Univariate correlations; not adjusted for covariates)
Bennett, 2009[56] Various cancer types, i.e.: breast, colorectal,	Pain management strategies used: • Seeking help from relatives: 61% • Practising CAM: 26%	• Lifestyle • Social support	• No differences in pain management strategies between older (≥75 years; N=58) and younger (<61 years; N=32) patients: P=0.420

gastrointestinal, lung, and urogenital (N=90)	• Exercising or practising different postures: 24%		(Univariate Chi-square association; not adjusted for covariates)
Chan, 2016[57] Various cancer types, i.e.: breast, colorectal, lung, and prostate (N=152)	Fatigue management strategies weekly used: • Doing relaxing things: 92% • Drinking caffeinated beverages: 92% • Adopting a balanced diet: 82% • Doing things that distract from fatigue: 63% • Planning activities: 52% • Drinking nutritional supplements: 29% • Practising complementary and alternative medicine: 28% • Talking to someone about fears and concerns: 28% Most effective fatigue management strategies were (mean score (SD ^{c})): • Doing things that distract from fatigue: score 5.87 (2.98) • Planning activities: 5.83 (3.13) • Adopting a balanced diet: 4.74 (3.13) • Doing relaxing things: 4.72 (3.85) • Drinking nutritional supplements: 4.26 (3.92) • Practising complementary and alternative medicine: 4.13 (3.69) • Talking to someone about fears and concerns: 2.41 (3.72) • Drinking caffeinated beverages: 2.03 (3.16)	• Lifestyle • Social support	Predictors of total perceived effectiveness of self-management strategies: [d] • higher education scores: P=0.020 • higher total self-efficacy scores: P=0.001 • global self-efficacy scores: P<0.001 Predictors of global perceived effectiveness of self-management strategies: [d] • lower levels of depressive symptoms: P=0.040 • higher levels of global self-efficacy: P<0.001 (Multivariate generalised estimated equations (GEE) models; covariates not specified)
Correa-Velez, 2003[58] Various cancer types, i.e.: bladder, breast, gastrointestinal, lung, melanoma, ovarian, and prostate (N=111)	• Undergoing surgery: 66% • Undergoing radiotherapy: 63% • Receiving chemotherapy: 76% • Taking hormone treatment: 80% • At least 1 CAM modality used during study period (i.e. interviews every 4-6 weeks; on average 5.4 interviews per participant): 48%	• Medicine and pharmacology • Lifestyle	Compared to non-users, CAM users more often had: • a higher income (≥$20 001): 69% vs. 41%; P=0.007 • no religious affiliation: 15% vs. 33%; P=0.011 • a larger household size (≥4 people): 47% vs. 28%; P=0.045

	• CAM used in the week prior to the first interview: 32% • CAM used on a regular basis during the last year of life: 31%		(Univariate Chi-square associations) • Compared to non-users, CAM users more often had lower self-reported need for control over treatment decisions: P<0.001 (Multivariate analyses (not specified); adjusted for income, gender, cancer type and conventional medicine)
Get-Kong, 2010[37] Various cancer types; not specified (N=240)	Self-management strategies weekly used: • Both non-pharmacological and pharmacological approaches: 63% • Non-pharmacological approaches: 30% • Pharmacological approaches: 7% Non-pharmacological strategies perceived to be most effective: • Talking to someone who gave support: 100% • Practising mind-body interventions: 96% • Changing dietary habits: 89% • Taking vitamins or dietary supplements: 79% • Meditating: 61% • Exercising: 61%	• Medicine and pharmacology • Lifestyle • Social support	
He, 2019 [38] Lung cancer (N=261)			• Resigned acceptance was associated with more difficulty in self-care: P=0.019 • Negative mood was associated with more difficulty in self-care: P=0.006 (Hierarchical multiple regression analyses; adjusted for age, gender, cancer stage and type, cancer-related financial burden, and history of radiotherapy)
Norris, 2009[43] Breast cancer (N=173)	Self-management strategies used: • Confiding in someone when feeling upset: 94% • Engaging in spiritual practices on a regular basis: 72% • Exercising ≥ 2 times a week: 56%	• Lifestyle • Psychology/ Mental health • Social support	• More physical exercise among patients with higher levels of physical functioning: OR [95% CI] = 2.18 [1.07–4.42]; P<0.0001

• Practising yoga or meditation ≥ 1 time a week: 32% • Keeping a diary: 25% • Participating in a cancer support group: 45% • Participating in another support group: 18% • Participating in an internet chat group: 8%	• More diary writing among patients with lower levels of physical functioning: OR [95% CI] = 0.29 [0.16–0.50]; P=0.032 • Less yoga or meditation among older patients: OR [95% CI] = 0.94 [0.89–0.98]; P=0.005 • Less participation in other cancer support activities among older patients: OR [95% CI] = 0.92 [0.87–0.98]; P=0.008 (Multivariate logistical regression analyses; adjusted for age, race, marital status, education, income, employment status, physical functioning, time since first diagnosis, and time since metastatic disease)

N: Number; CAM: Complementary and alternative medicine; P = P value; 95% CI = 95% confidence interval.

[a] Scores on the Concerns Checklist (CCL) ranged from 0 (not at all worried) to 3 (very much worried).

[b] *r*= Spearman correlation coefficient.

[c] Effectiveness scores ranged from: 0 to 10, with higher scores representing greater effectiveness.

[d] Total summary scores reflected effectiveness of each behaviour used with equal weight for each of the behaviours. Global scores reflected a total rating of the effectiveness of each behaviour used, in which the weight of each behaviour may not be equal.

Psychology/mental health. Several patients managed their psychological well-being by keeping a diary(43) and participating in meaningful charity activities, such as volunteering and promoting cancer awareness(46,48). Furthermore, eight qualitative studies showed that patients used assertive self-talk or tried to control or accept their situation, readjust purpose, expectations and meaning in life, think of their family's future well-being rather than of their own death and focus on positive thoughts (e.g. good memories) and feelings(42,45,46,48,55,60–62). This often enhanced coping with the disease, but could also lead to psychological turmoil when motives were incompatible(48).

Social support. Four quantitative(37,43,56,57) and 15 qualitative studies(39,45–48,50,52,55,59–63,65,66) indicated that many patients sought support from relatives and friends (28%– 94% in the quantitative studies). This was mentioned as an effective strategy by 61% and 100% of the patients in two quantitative studies on pain management and general symptom management, respectively(37,57). Besides seeking social support, providing or arranging social sup- port for relatives was also mentioned as a self-management strategy(45–48,62). Patients considered maintaining or intensifying relationships with relatives important, because it gave them emotional strength(45,57,60,65), provided distraction from their cancer symptoms(62), made them feel important and helpful to others(62), protected their identities and usual social roles(48) and enabled them to rely on relatives in case their condition would worsen(39). In several qualitative studies, however, patients also mentioned that they found it difficult to accept or ask for support from relatives and friends, because they did not want to be a burden(55,59–63,65). Some of them only asked for help when they could no longer perform their usual activities(50,60,61,63,66) or restricted their social contacts to close relatives or moments when they felt good(57,66). Some patients mentioned selective communication of their thoughts and emotions or self-isolation as strategies to protect themselves and their loved ones from mental and emotional distress(48,52,57,63). A quantitative study among women with metastatic breast cancer demonstrated participation in cancer support groups (45%), Internet chat groups (8%) and other cancer support programmes (18%)(43). Compared to support from relatives and friends, support from fellow patients was sometimes perceived to be more effective(55,59,65). Some patients experienced professional support as positive when the severity of their pain increased(41), others reported that this depended on the person providing it(55,60,62,65). According to other patients, healthcare professionals listened insufficiently or paid too little attention to their emotional needs(50,61,65).

Knowledge and information. Several patients searched for more information about their disease, care and treatment(37,39,42,47–49,52,61,65,67). Most of them perceived this as an effective strategy to manage symptoms and other disease consequences, as it empowered them to optimise both their physical and psychological well-being(48,52). Also, lack of understanding of, for example, the meaning of palliative care or cancer symptoms could lead to confusion and concerns and, subsequently, hamper effective self-management(47,60). However, when obtaining useful information was complicated, this could also become a burden(47). Some patients preferred not to think, talk or read about cancer over obtaining knowledge and information, as this made them feel able to exert control over their disease experiences and protected them from sad feelings(45).

Navigation and coordination. Three studies described how patients prepared for their death by making financial and practical plans (e.g. planning funeral, sorting out affairs)(48,55,62). This was considered beneficial, because it enhanced patients' feelings of control over their death and the future well-being of their families, thus also protecting their own identities and family roles(48). In another study, patients with advanced breast cancer coordinated medical services between different healthcare providers, for example, by collecting and exchanging health-related documents(47). Patients also coordinated their care by delegating some of its aspects (e.g. responsibility for making and attending medical appointments, decisions on pain treatment approach) to relatives and friends(47,50,63), and by delegating or staying in charge of information dissemination from healthcare professionals to relatives or vice versa(39,63). A study among older patients who received outpatient cancer treatment and lived alone in rural areas showed that these patients perceived limited control over practical arrangements (e.g. arranging treatment schedules and public transport back home) and experienced navigating through the healthcare system (e.g. ordering tests, making appointments) as very energy-consuming(60).

Medical decision-making. Several patients participated in advance care planning or made together with their health-care professionals shared decisions regarding future medical care and treatments(47). Others used short-term goal setting as a strategy to reach long-term goals(39,48,65,66). Over time, several patients shifted their focus on quality of life, and for this reason, some of them considered foregoing treatment(49).

Personal characteristics associated with the use of self-management strategies among patients with advanced cancer

Two studies explored the cross-sectional association between age and self-management strategies (see Table 3). Older and younger patients did not differ in pain management strategies(56), but younger patients were shown to be more likely than older patients to practise yoga/ meditation and to participate in other cancer patient support activities (e.g. cancer retreats)(43). Patients with higher levels of physical functioning were more likely to practise physical exercise, while patients with lower levels of physical functioning were more likely to keep a diary. Use of complementary and alternative medicine seemed to be more prevalent among patients with higher income levels, larger household sizes, no religious affiliation and less need for control over treatment decisions(58). Higher education and self-efficacy levels and lower depressive symptom levels were positively associated with perceived effectiveness of self-management behaviours(57). Resigned acceptance and negative mood were associated with more difficulty(38). Furthermore, patients with lower self-efficacy and more anxiety and/or depressive symptoms reported more self-management concerns(67).

Healthcare professionals' attitudes towards self-management of patients with advanced cancer

Table 4 summarises the three articles about attitudes of healthcare professionals towards self-management of patients with advanced cancer. In one study, primary healthcare professionals concurred with patients in their view that it is important to view a patient holistically, that is, as an entire person rather than an illness, in terms of both self-management and care and treatment by others(55). Another study showed that healthcare professionals working in a specialist palliative care service perceived patient self-management as both desirable and achievable, but only if undertaken in partnership with them(53). Self-management could also be perceived as problematic, for example, when patients acted on limited knowledge(53). Some clinical nurse specialists were worried about supporting non-pharmacological methods because of the 'medical model' of training they had received. Nurses suggested more self-management education for patients and their relatives, for example, by means of a smart phone app and a webpage that stimulated active participation in obtaining information and medical decision-making(53). Community-based palliative care nurses also indicated that patients required their instruction and information to adequately educate their relatives regarding medication management(39). Communication between patients and healthcare professionals was perceived as vital in medication self-management(39).

Table 4. Non-experimental qualitative study results on healthcare professionals' attitudes towards self-management of patients with advanced cancer (three studies).

First author Healthcare professionals (*N*)	Attitudes towards patients self-management
Campling et al.[39] Community-based palliative care professionals (*N* = 19): clinical nurse specialist (*n* = 13), consultant (*n* = 1), inpatient unit nurse (*n* = 2), lecturer/practitioner (*n* = 1), lead nurse/commissioner (*n* = 1)	• With regard to patients who adopted an advocacy role in medication management, nurses emphasised the importance of getting the right drug, via the right route. • Nurses found that the educator role, in which patients educate their relatives regarding their medication management, required their professional instruction and information. They indicated the need to refine knowledge and information that patients and relatives had found on the internet.

	• Nurses perceived the communicator role, in which patients communicate relevant information to healthcare professionals, as vital in medication self-management
Hughes et al.[53] Professionals working in specialist palliative care centres (*N* = 17): community clinical nurse specialist (*n* = 6), complementary therapist (*n* = 3), hospice nurse (*n* = 5), hospice social worker (*n* = 1), hospice spiritual care coordinator (*n* = 1), palliative care consultant physician (*n* = 1)	• Desirable: - If patients act autonomously but in partnership with healthcare professionals and base decisions on information, dialogue and reflections on prior experiences. - Because it is unrealistic for professionals to provide comprehensive and complete solutions to patients' pain problems and enables patients to exert control over what is happening to them. • Achievable: - If patients take responsibility and are motivated to try to self-manage their pain. - If nurses provide education for patients and relatives and introduce self-management options early in the disease trajectory (when patients are still well enough to learn about self-management and act upon this knowledge). - Nurses suggested more self-management education for patients and relatives, provided in a range of formats and introduced early in the disease trajectory. Hospice nurses suggested the use of a smartphone app and a webpage to provide additional information and support decision-making among patients and relatives. - Non-pharmacological methods of pain relief were viewed as active and productive means of self-management; religious and spiritual practices were considered relevant strategies as well • Problematic: - If patients act on limited knowledge, exercise complete autonomy and reject professional advice, adopt strategies not supported by the medical model of professionals training or fully delegate control and responsibility to healthcare professionals. - If healthcare professionals take away too much control and responsibility (resulting in lack of understanding among patients and relatives). - Clinical nurse specialists were hesitant to support non-pharmacological self-management strategies because of the 'medical model' of training they had received.
Johnston et al.[55] Key professionals referred by patients with advanced cancer (*N* = 20): clinical nurse specialist (*n* = 9), nurse (hospice day care) (*n* = 3), oncologist (*n* = 1), general practitioner (*n* = 5), clinical	• Healthcare professionals found it important to view patients from a holistic rather than an illness perspective in terms of both self-management and care and treatment by others.

nurse specialist/hospice doctor (*n* = 1), community nurse (*n* = 1)

N: Number.

DISCUSSION

Main findings

This review summarised results from 31 non-experimental quantitative and qualitative studies that examined self- management of patients with advanced cancer. The vast majority of these studies were of acceptable quality and examined self-management from the perspective of patients. Most studies included mixed sex and diagnosis groups and had been conducted in Western high-income countries. Overall, the reviewed literature clearly shows that self-management of patients with advanced cancer covers numerous domains: medicine and pharmacology (e.g. self-administering pain medications), lifestyle (e.g. practising complementary and alternative medicine, taking breaks when doing daily chores), psychology (e.g. keeping a diary), social support (e.g. engaging in support groups), knowledge and information (e.g. seeking treatment information), navigation and coordination (e.g. making financial plans) and medical decision-making (e.g. participating in advance care planning). Within each of these domains, patients might use multiple strategies that are highly individual, sometimes ambivalent and generally aimed at optimising their own well-being and/or (future) well-being of loved ones. Healthcare professionals perceive self-management as both desirable and achievable if based on sufficient skills, knowledge, information and solid patient–professional partnerships.

That strategies used by patients with advanced cancer vary widely and sometimes even comprise contradictory behaviours is well illustrated by our data on lifestyle self-management strategies, which was the domain most often addressed by the included studies. On the one hand, patients frequently mentioned that the diagnosis of advanced cancer had caused them to change lifestyle habits and start complementary and alternative medicine or new hobbies. On the other hand, however, many of them also indicated that they preferred to maintain their usual daily routines as much as possible. In the social support domain, self-management strategies also varied strongly, from seeking and providing social support to limiting and even avoiding social support. Although most patients experienced social support as essential, for some of them, seeking and accepting it was hampered by fears of becoming a burden to loved ones and losing their established roles and identity. A large diversity of strategies and experiences with these strategies was observed in the other self-management domains as well. Medical and pharmacological strategies ranged from adhering to prescribed treatment to adjusting and discontinuing treatment. Whereas some patients actively gathered information about treatment and disease, others avoided obtaining more knowledge, because it was too energy-consuming or made them feel sad. More generally, ambiguity and inconsistency in self-management strategies often seemed to reflect a conflict between two seemingly opposing attitudes: appreciating life in the present versus planning for the future; readjusting purpose and expectations versus maintaining normality and a sense of established identity; and taking control versus letting things happen.

By including a larger number of studies as well as more recent studies, and by focussing more specifically on patients' self-management strategies and healthcare professionals' attitudes, our systematic review provides novel insights compared to the two prior review studies on self-management in advanced cancer care(19,20). The scoping review of Budhwani and colleagues merely distinguished between physical and psychosocial domains(19), while our review also describes domains of lifestyle, knowledge and information, navigation and coordination and medical decision-making. Furthermore, we provide a more detailed overview of the full range of self-management strategies and experiences in these domains, thereby also revealing divergence and ambivalence between these strategies and experiences. Only a few studies explicitly defined self-management (or self-care). Although these definitions were not univocal regarding the nature of self-management, our review's findings on the divergence of strategies and experiences suggest that the question should not be whether or not, but how someone engages in self-management. This corroborates the previously proposed idea that 'one cannot not self-manage'(15) and argues against a normative approach to self-management (support). It also implies that self-management, although affected by personal and contextual factors, is not restricted to particular settings or disease stages. It may thus be facilitated anywhere and anytime (until the very end), if tailored to individual circumstances, abilities and preferences.

The seven domains we distinguished partly overlap with those observed within chronic disease populations, the field on which studies about disease self-management have thus far predominantly focused. Many of the models proposed to describe and enhance chronic disease self-management also include domains of social support, information, navigation and decision-making(10,11,17,18). Remarkable, however, is that the self-management strategies and experiences we identified among patients with advanced cancer are generally more divergent and ambivalent than those described for patients with chronic, generally less advanced diseases. For example, while we found that avoiding or ignoring (medical) information may be a self-management strategy among patients with advanced cancer, reviews about self-management among patients with chronic diseases merely report on the opposite, that is, seeking and obtaining information(10,68–71). Possibly, the prospect of imminent deterioration and, ultimately, death adds to the complexity of self-management. After all, these prospects can drastically change the way someone relates to space, place, time, self and others. These changes, in turn, interact and consequently affect self-management, thus complicating its dynamics and increasing its susceptibility to different, potentially competing values, priorities and interests. This interpretation is supported by recent chronic disease studies showing that self-management is perceived to be more challenging during periods of transition, disease progression or acute events (e.g. exacerbations of chronic obstructive pulmonary disease)(16,72,73). It also resonates with findings from several advanced disease studies(22,74–78). Two recent systematic reviews indicated that patients with advanced diseases were ambivalent towards discussing mortality and advance care planning(77,78). Furthermore, a recent literature review on life values of elderly people with advanced cancer showed that these patients often fluctuated between avoiding and facing the truth about their medical condition(75). Many of them also considered maintaining independence and withholding emotions to contribute to a good death, as this was perceived to minimise strain on their loved ones.

In six studies, we identified several sociodemographic and functional patient characteristics that might affect self-management of patients with advanced cancer. These results were derived from non-experimental quantitative studies with moderate sample sizes and limited adjustments for confounding and selection bias, and should thus be interpreted cautiously. Nevertheless, they provide some explorative insights that could generate hypotheses for future research. Younger patients and patients with higher levels of physical functioning might, for instance, be more likely to adopt physically and socially active self-management strategies (e.g. yoga, physical exercise, participation in cancer support groups) than older patients and patients with lower levels of physical functioning(43,56). Another study suggested that fewer depressive symptoms, higher educational levels(57) and greater self-efficacy(57) positively predicted self-perceived effectiveness of applied self-management strategies(57). Resigned acceptance and negative mood were also associated with more self-management difficulties(38). One of the explanations could be that patients with more depressive symptoms and a negative mood lack motivation and energy to actively self-manage(79). In addition, higher educated patients are generally more health literate(80), and might therefore be more likely than lower educated patients to take a proactive approach in self-management and acquire accurate knowledge and understanding of their condition and self-management possibilities(16,81). Furthermore, patients with higher self-efficacy levels are more likely to perceive symptoms and other disease consequences as modifiable and might therefore invest more in self-management strategies than patients with lower self-efficacy levels(82).

Only three qualitative studies assessed attitudes of healthcare professionals towards patient self-management(53,55). Healthcare professionals in these studies were generally optimistic about patient self-management, but also set conditions for its effectiveness, such as solid patient-professional partnerships and sufficient skills, knowledge and information. Suggestions to provide patients with more education, introduced early in the disease trajectory and in a range of different formats, are in line with remarks made by nurses participating in a recent online focus group study on self-management support and eHealth for patients and relatives confronted with advanced cancer(83). We identified a lack of studies among relatives, and inconsistencies regarding the roles and responsibilities assigned to others (i.e. healthcare professionals, relatives) in the self-management definitions provided by some of the included studies. Nevertheless, our findings evidently show that important others play a crucial role in patient self-management, and should thus somehow be involved in self-management support. However, also fears of becoming a burden to relatives were commonly described by patients in our review as well as in previous studies(84,85). Together with findings that some patients appreciated each other's company and derived satisfaction from participation in cancer-related charity activities, this suggests that peer support could be a promising pillar in patient self-management. The mutual benefits of peer support are increasingly recognised, also among patients with advanced cancer(86,87).

Strengths and limitations

To our knowledge, this is the first systematic review on patient self-management in advanced cancer care that includes perspectives of both patients and healthcare professionals. A strength is that we used a comprehensive search strategy with a broad operational definition of self-management. Nevertheless, we may have missed studies reporting on self-management without labelling it as such (or as a related term, e.g. self-care). The moderate quality of some of the included individual studies affected the evidential value of this systematic review, especially regarding selection bias: patients who had participated in the included studies may have been more able and willing to engage in the self-management strategies studied. This may have caused an overestimation of the prevalence and impact of self-management strategies.

Relevance for clinical practice

Our review provides several clinically relevant insights into self-management in the context of advanced cancer. Compared to care provided by healthcare professionals, patients' self-management strategies may be substitutional (such as self-administering pain medications at home), additional and distinctive (such as mobilising peer support) or even conflicting (such as refraining from taking prescribed medications). This highlights the importance of solid patient-professional partnerships, in which preferences, experiences and expertise are mutually shared. Our review also provides a foundation for the development of self-management support interventions. Such programmes should be tailored to the domains in which patients need additional support. The findings on sociodemographic and functional characteristics associated with self-management strategies also provide useful preliminary targets for self-management support programmes. For example, patients with low levels of education may need additional support in self-management domains of information, and navigation and coordination. Finally, a key challenge in the development of self-management support programmes will be to incorporate them into existing models of care(19).

Implications for future research

To enhance conceptual clarity regarding self-management in the advanced cancer context, we recommend consistency in terminology used as well as further elaboration of its definition and conceptual framework. Given the important role of formal and informal caregivers in supporting patient self-management, additionally, we recommend that future studies identify their experiences and attitudes as well. Finally, we recommend that future studies investigate the effectiveness and working mechanisms of self-management strategies used by patients with advanced cancer at the levels of patients, communities and healthcare organisations and policy.

CONCLUSION

This systematic review shows that self-management of patients with advanced cancer is complex and multifaceted, covering multiple domains and a broad range of strategies and experiences that are highly dependent on individual preferences and characteristics. Possibly, the prospect of imminent deterioration and death adds to its complexity, as subsequent changes in values, priorities, interests and social interactions are likely to affect self-management strategies and experiences. Therefore, self-management support programmes for this vulnerable group could benefit from an individualised approach that re-evaluates patients' needs and wishes, is embedded in solid partnerships with relatives and healthcare professionals, and is incorporated into existing models of care.

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SUPPLEMENTARY MATERIAL

Table 1. Search terms.

1 .Embase.com

('advanced cancer'/de OR (((advance*) NEAR/4 (cancer* OR neoplas* OR carcinoma* OR tumor* OR tumour* OR malignan*))):ab,ti OR ((Neoplasm/mj/exp OR 'cancer patient'/de OR (neoplas* OR cancer* OR carcinoma* OR oncolog* OR tumor* OR tumour* OR malignan*):ab,ti) AND ('metastasis'/exp OR 'palliative therapy'/exp OR 'palliative nursing'/de OR 'terminal care'/exp OR 'terminally ill patient'/exp OR (metasta* OR palliati* OR terminal* OR 'end of life' OR hospice* OR incurable*):ab,ti))) AND ('self care'/exp OR 'self monitoring'/de OR 'drug self administration'/de OR (((self) NEXT/1 (car* OR help* OR manag* OR medicat* OR treat* OR monitor* OR administrat*))):ab,ti) NOT ('Conference Abstract'/it)

2. Medline Epub (Ovid)

(((advance*) ADJ4 (cancer* OR neoplas* OR carcinoma* OR tumor* OR tumour* OR malignan*)).ab,ti. OR ((exp* Neoplasms/ OR (neoplas* OR cancer* OR carcinoma* OR oncolog* OR tumor* OR tumour* OR malignan*).ab,ti.) AND (exp "Neoplasm Metastasis"/ OR "Palliative Care"/ OR "Palliative Medicine"/ OR "Hospice and Palliative Care Nursing"/ OR exp "Terminal Care"/ OR "Terminally Ill"/ OR (metasta* OR palliati* OR terminal* OR ((end) ADJ2 (life)) OR hospice* OR incurable*).ab,ti.))) AND (exp "Self Care"/ OR (((self) ADJ (car* OR help* OR manag* OR medicat* OR treat* OR monitor* OR administrat*))).ab,ti.)

3. PsycInfo (Ovid)

(((advance*) ADJ4 (cancer* OR neoplas* OR carcinoma* OR tumor* OR tumour* OR malignan*)).ab,ti. OR ((exp* Neoplasms/ OR (neoplas* OR cancer* OR carcinoma* OR oncolog* OR tumor* OR tumour* OR malignan*).ab,ti.) AND ("Metastasis"/ OR "Terminal Cancer" OR exp "Death and Dying"/ OR "Palliative Care"/ OR "Hospice"/ OR "Terminally Ill Patients"/ OR (metasta* OR palliati* OR terminal* OR "end of life" OR hospice* OR incurable*).ab,ti.))) AND (exp "Self Management"/ OR "Self-Monitoring" OR (((self) ADJ (car* OR help* OR manag* OR medicat* OR treat* OR monitor* OR administrat*))).ab,ti.)

4. Cochrane Central

((((advance*) NEAR/4 (cancer* OR neoplas* OR carcinoma* OR tumor* OR tumour* OR malignan*))):ab,ti OR (((neoplas* OR cancer* OR carcinoma* OR oncolog* OR tumor* OR tumour* OR malignan*):ab,ti) AND ((metasta* OR palliati* OR terminal* OR 'end of life' OR hospice* OR incurable*):ab,ti))) AND ((((self) NEXT/1 (car* OR help* OR manag* OR medicat* OR treat* OR monitor* OR administrat*))):ab,ti)

5. Cinahl (EBSCO)

(((advance*) N4 (cancer* OR neoplas* OR carcinoma* OR tumor* OR tumour* OR malignan*)) OR ((MM Neoplasms+ OR MH "Cancer Patients" OR (neoplas* OR cancer* OR carcinoma* OR oncolog* OR tumor* OR tumour* OR malignan*)) AND (MH "Neoplasm Metastasis+" OR MH "Hospice and Palliative Nursing+" OR MH "Terminal Care+" OR MH "Terminally Ill Patients+" OR (metasta* OR palliati* OR terminal* OR "end of life" OR hospice* OR incurable*)))) AND (MH "Self Care+" OR (((self) N1 (car* OR help* OR manag* OR medicat* OR treat* OR monitor* OR administrat*)) OR (patient controlled N1 analgesi*)))

6. Web of Science

TS=(((((advance*) NEAR/3 (cancer* OR neoplas* OR carcinoma* OR tumor* OR tumour* OR malignan*))) OR (((neoplas* OR cancer* OR carcinoma* OR oncolog* OR tumor* OR tumour* OR malignan*)) AND ((metasta* OR palliati* OR terminal* OR "end of life" OR hospice* OR incurable*)))) AND ((((self) NEAR/1 (car* OR help* OR manag* OR medicat* OR treat* OR monitor* OR administrat*))))) AND DT=Article

7. Google Scholar

"advance|advanced|metastases|metastatic|palliative|terminally|"end of life"|hospice|incurable cancer|neoplasm|neoplasms|carcinoma|tumor|tumour" "self care|help|management|medication|treatment|monitoring|administration"

Table 2. Non-experimental qualitative study results on self-management strategies and corresponding experiences of patients with advanced cancer (23 studies)

First author, year Patient population: type(s) of advanced cancer (N)	Self-management strategies and corresponding experiences	Self-management domains
Bennett, 2006[44] Breast cancer (N=8)	• Patients maintained daily activities to keep living in the present, although this was complicated by perceived physical and emotional constraints. • They used physical as well as emotional self-isolation to protect self, relatives and friends and/ or exert control. This also had negative consequences, e.g. anxiety about losing friends and support.	• Lifestyle • Social support
Campling, 2017 [39] Various types, i.e.: bile duct, breast, colon, lung, and other (N=15)	Medication management of patients approaching the end of life varied from full to no engagement: some patients accepted and preferred full responsibility in their medication management, whereas others limited their autonomy and decision-making. Patients adopted the following roles in medication management: • Advocate, e.g. by requesting alternative opioids if side effects were not acceptable; • Educator of relatives, anticipating future changes so that they could rely on relatives in case of worsening condition; • Facilitator, facilitating relationships with their healthcare professionals and relatives to aid access to their medicines; • Problem-solver, striving to navigate the difficulties posed by the medicine supply system and to problem solve the side effects of their opioids, e.g. by titrating laxatives; • Communicator of relevant information to relatives and healthcare professionals; • Goal-setter, self-planning or planning with a general practitioner or clinical nurse specialist; • Monitor, writing down breakthrough doses and noting effectiveness; • Reporter of relevant symptom experiences and side effects. The continual disease progression led to rapid changes in symptoms and side effects experienced by patients, which led to fluctuations in medication management roles and capabilities.	• Medicine and pharmacology • Social support • Knowledge and information • Navigation and coordination • Medical decision-making
Correa-Velez, 2005[51] Various cancer types, i.e.: bladder, breast, gastrointestinal, lung,	• Patients used CAM for three main reasons, i.e. to: - control the cancer and prolong life; - relief cancer symptoms and side effects of conventional treatments; - detoxify the body, boost immunity, boost energy, and enhance quality of life.	• Lifestyle

melanoma, ovarian, and prostate (N=39)		
Devik, 2013[60] Various cancer types; not specified (N=5)	To keep hope alive, cope with doubts and anxiety, and exert control, patients • Underwent chemotherapy; • Maintained daily life, autonomy and independence as much as possible; • Focused on the positive sides and kept on dreaming. • They asked few questions, which resulted in a lack of knowledge and confusion concerning symptoms, disease progression and treatment. • Navigating through the healthcare system alone (e.g. ordering tests, arranging treatment schedules, and making practical plans) required much time and energy. Patients experienced limited control over practical arrangements (e.g. long waiting times for a bed, meal or taxi home). • Patients sought support from relatives and especially valued the emotional aspects of this support. They were reluctant to ask for assistance from relatives and healthcare professionals because of fears of becoming a burden and/ or a demanding patient.	• Medicine and pharmacology • Lifestyle • Psychology/ Mental health • Social support • Navigation and coordination
Eliott, 2008[40] Various cancer types, i.e.: breast, colorectal, kidney, lung, lymphoma, and other (N=28)	• Patients used CAM because of a desire for cure, social reasons and improved quality of life through control over the disease and its side effects. They considered integrating it into conventional medicine. • Patients who did not use CAM framed it exclusively in terms of (unachievable) curative treatment. They perceived it as an affront to medical expertise and thought healthcare professionals would perceive it similarly.	• Lifestyle
Erol, 2018[41] Various cancer types, i.e.: colorectal, gastric, and lung (N=16)	Patients described pain at the end of life as an extremely difficult experience, which affected their daily life and social relations. They could not manage daily household chores and became dependent on their family members. Patients: • Used non-pharmacological approaches to relieve pain, e.g. praying, changing position, having a hot bath, walking, watching TV, and engaging in crafts. Praying to God for healing and Namaz (an Islamic pray ritual) were the approaches most commonly used. Herbal product use was rare. • Generally avoided using pharmacological agents until the intensity of their pain increased and became unbearable. The most common problem was poor sleep; several patients used sleeping pills. • Less than half of the patients immediately informed the nurses about their pain and need for medication, others tried to be calm and patient, and informed healthcare professionals when the severity increased.	• Medicine and pharmacology • Lifestyle • Social support

Gibbins, 2014[61] Various cancer types, i.e.: breast, lung, melanoma, mesothelioma, ovary, prostate, and sarcoma (N=12)	Self-management of pain was largely inspired by the wish to maintain role, self and independence. Patients • Did not use pain medications as prescribed, but rather took the amounts perceived sufficient to 'keep a grip' on the pain without causing too many sedative side effects (which were considered a threat to self); • Tried to continue normal activities and everyday tasks (e.g. household chores, hobbies); • Adjusted activities to pain; • Adjusted expectations from becoming pain free to alleviating pain; • Sought help from others, but only when normal activities were threatened. Patients considered maintaining relationships important, but difficult due to discomfort caused by physical dependence and seeing relatives distressed by their pain. They found it challenging to communicate pain to healthcare professionals due to a perceived lack of interest from healthcare professionals. • Patients considered knowledge about causes of pain even more important than pain relief (possibly due to fear of disease progression).	• Medicine and pharmacology • Lifestyle • Psychology/ Mental health • Social support • Knowledge and information
Hansen, 2015[49] Hepatocellular cancer (N=14)	• Patients searched alternative sources (i.e. internet, books and friends) for information. • They compared their cancer to other diseases and examined changes in physical condition to understand and describe their disease. • Patients started medical treatments because these provided a sense of control. Over time, they more often decided to stop or refuse (additional) treatments and/ or regretted having ever started treatments because of adverse side effects, uncertainty about treatment efficacy and an increased focus on quality of life.	• Medicine and pharmacology • Knowledge and information • Medical decision-making
Hopkinson, 2007[54] Various cancer types, i.e.: breast, gastrointestinal, head and neck, lung, prostate, and other (N=30)	• Patients ate nutritious, tolerable food to retain a sense of control over changing eating habits. • They also ate to please relatives.	• Lifestyle
Johansson, 2006[62] Various cancer types, i.e.: bile duct, bladder,	Patients highly valued ordinariness and found it important to live their lives more or less as they did before they became ill. They • Took extra drug doses in case of breakthrough pain; • Balanced rest and physical activity to alleviate suffering;	• Medicine and pharmacology • Lifestyle

breast, and prostate (N=5)	• Maintained daily activities; • Kept positive memories alive and increased other positive coping skills; • Kept occupied with tasks and being important to others. Emotional support from relatives and friends was considered essential and provided distraction. Support from healthcare professionals was perceived as positive, committed and testifying to control over the situation, which gave a sense of security. • Patients prepared themselves and their relatives for how they wanted things to be after their death; • They independently took important decisions about their present lives and the future. They still had a clear mind to do this.	• Psychology/ Mental health • Social support • Navigation and coordination • Medical decision-making
Johnston, 2012[55] Various cancer types, i.e.: breast, colorectal, gastrointestinal, hematological, lung, and urogenital (N=20)	• Patients tried to wash, dress, shop and cook themselves. They wanted to be seen as normal and not as an illness. • They used CAM. This was considered a beneficial strategy mainly by patients who lived in urban areas or attended in hospice day care. • Patients practised religion. • They focused on acceptance, positivity, and their rights of choice and control. • They sought support from other cancer patients. Sometimes they received support from a clinical nurse specialist; only few patients received care to stay at home. They wanted to stay at home. • They sorted out affairs, planned their funeral and/ or discussed what to expect and how to prepare for death. Healthcare professionals were only rarely involved in these discussions.	• Lifestyle • Psychology/ Mental health • Social support • Navigation and coordination
La Cour, 2009[59] Various cancer types, i.e.: breast, colon, lung (N=45)	• Patients maintained their daily activities by avoiding activities that might cause them pain or harm. • They undertook novel activities to enrich their daily routine. • Patients limited social networks to relatives and close friends. They did not want to be a burden to their relatives and friends. They expressed a desire to be with others with the same condition, but were also concerned that this would lead to too much talking about the disease.	• Lifestyle • Social support
Levy, 2015[48] Prostate cancer (N=5)	• Patients did new and familiar leisure activities to live in the present and be distracted from pain. • Patients did meaningful activities, such as volunteering, to renegotiate their sense of who they were. The benefits of new activities gave them a sense of belonging and connection, which counteracted the feelings of separation that their 'between life and death' state entailed. They channelled thoughts of their own death towards their family's future well-being and tried to maintain their role as head of the family even after death, thereby enhancing this role in the present. These strategies were sometimes incompatible, causing psychological turmoil.	• Lifestyle • Psychology/ Mental health • Social support • Knowledge and information • Navigation and coordination

	• Patients socialised and spent time with relatives. They took control of communication with relatives about their cancer and impending death. • Patients looked for information about treatment. • They made financial and practical plans to control their death. • They strove to move forward in a goal-oriented and active way, by using short-term goals (‘stepping stones’) as bridges between time and achievement of life purposes.	• Medical decision-making
Lewis, 2016[45] Breast cancer (N=18)	Three self-management styles: 1) Patients who strove for normality • These patients did not make any major lifestyle changes because continuing life as it used to be was so important to them. • They tried to minimize the negative social impacts of having metastatic disease. • They tried not to think, talk or read about their cancer to live in the present, exert control over their disease experience and protect themselves from feelings of unhappiness. 2) Patients who re-evaluated life • These patients re-evaluated what was most important to them. A positive mind-set contributed to keeping control over their disease. • They devoted more time with relatives and friends. The emotional support received by talking with relatives and friends was of great importance. 3) Patients who felt restricted • These patients described lifestyle strategies that were continuously changing because their health was so variable. Although they tried to get on, they felt limited by disease symptoms and side effects of treatment.	• Lifestyle • Psychology/ Mental health • Social support • Knowledge and information
Liu, 2017[42] Various cancer types, i.e.: breast, lung, pancreatic, prostate, and other (N=9)	Patients described that breakthrough cancer pain adversely affected daily activities, sleep, and feelings; they could not eat, walk, and run. Sometimes pain even caused extreme difficulties with sitting down and getting up. Patients • Endured their pain, because this is encouraged by the traditional Chinese culture, in which pain is part of the disease process, and because they did not want to be labelled as hypochondriacs or complainers. • Used massage, acupuncture, and other non-pharmacological pain management methods. • Took nonsteroidal anti-inflammatory medicines or visited pain clinic doctors when they felt that these medicines were ineffective.	• Medicine and pharmacology • Lifestyle • Psychology / Mental health • Knowledge and information • Navigation and coordination

	• Sought support from the healthcare system to understand and control breakthrough pain and to communicate effectively with healthcare professionals.	
McPherson, 2014[63] Various cancer types, i.e.: breast, gastrointestinal, respiratory/thoracic, urogenital, and other (N=18)	Patients had a strong desire to be independent in their pain management. They often • Rejected pharmacological approaches because of the side effects; • Delayed or omitted their medication intake and endured pain; • Tried to relief pain with non-pharmacological approaches, such as lying down, sleeping, changing position, not moving and distraction. • Patients sought support from others, but only mentioned their pain when it became too difficult to manage. They did not always relatives in discussions with professional caregivers visiting them at home, also because did not want to bother their relatives. • Patients sometimes delegated roles in in pain control (e.g. deciding on pain treatment approach, taking responsibility for obtaining medications and attending appointments) to their relatives (depending on their physical and psychological functioning).	• Medicine and pharmacology • Lifestyle • Social support • Navigation and coordination
Pacsi, 2015[46] Breast cancer (N=6)	• Patients made extensive dietary changes. • They emphasised the power of prayer, which gave the strength to deal with the treatment and its side effects while focusing on living in the moment. • They adjusted to changes in body image. • They promoted breast cancer awareness. • They maintained a positive attitude. • They tried to participate actively in their children's lives. • Patients made informed decisions about the treatments.	• Lifestyle • Psychology/ Mental health • Social support • Medical decision-making
Peoples, 2017[66] Various cancer types, i.e.: breast, cervix, colon, lung, prostate, and other (N=73)	To manage daily activities, patients: • Took breaks during the day or divided activities into smaller parts; • Sometimes asked for assistance from others (mostly relatives, friends and neighbours) to manage their activities, required social services, and/ or used assistive devices; • Limited their social interactions to close friends and relatives; • Set short-term goals to fulfil wishes and reach long-term goals.	• Lifestyle • Social support • Medical decision-making
Sand, 2009[50] Various cancer types, i.e.: breast, gastrointestinal, lung,	To manage and remain in control over medication intake, patients: • Questioned whether they actually needed prescribed medication; • Discontinued or titrated the amount of medication; • Sometimes delayed medication intake until they were no longer able to hold the pain.	• Medicine and pharmacology • Social support • Knowledge and information

melanoma, and urogenital (N=15)	• Patients with self-regulation problems let their relatives take over the responsibility for their medication. Support from others was considered helpful. Patients wanted healthcare professionals to listen better. • They wanted to be informed about the side effects and importance of their medication and the time it would take to reach the maximum dosage. Some patients wondered whether better medicines might have come available and why they had not received palliative medication earlier.	• Navigation and coordination
Schulman-Green, 2011[47] Breast cancer (N=15)	• Patients adhered to treatment regimens. • They engaged in exercise and nutritional regimens. • They kept life as normal as possible. • They sought support from friends and relatives and evaluated and ensured support at home. • They learned about cancer. • They coordinated medical services. • They participated in advance care planning. • They made treatment decisions. They reported barriers of symptom distress, limited time and energy, competing priorities, lack of understanding of the different forms of care, and fears to make the right decisions.	• Medicine and pharmacology • Lifestyle • Social support • Knowledge and information • Navigation and coordination • Medical decision-making
Stephens, 2014[52] Myeloma (N=11)	• Patients manipulated medication schedules to manage the side effects of treatment. • They tried to get an adequate fluid-intake, made dietary changes and exercised to rebuild strength and reduce complications. • They gathered information about their disease. • They rather let primary support persons (usually a partner or child) make disclosures on diagnosis or disease progression to other family members to manage their distressing emotions.	• Medicine and pharmacology • Lifestyle • Knowledge and information • Social support
Thomas, 1999[65] Various cancer types; not specified (N=19)	• Patients adopted one or more CAM treatments. • They reached out to a wide-ranging network of relatives, friends and other support systems, but found it difficult to accept the encouragement and positivity. Patients perceived their healthcare professionals as supportive, but others felt that healthcare professionals ignored their emotional needs. They mentioned that listening to others with cancer helped them to understand their disease better. • Patients wanted to know more about their cancer. • They weighed up options, made choices, participated in decisions affecting themselves and others, set goals, and gauged future outcomes.	• Lifestyle • Social support • Knowledge and information • Medical decision-making

CAM: Complementary and alternative medicine


3

CHAPTER 3

Self-management of patients with advanced cancer: A qualitative interview study

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Submitted

ABSTRACT

Objective

Little is known about self-management of patients with advanced cancer. Insight in patients' self-management behaviors and the factors associated with self-management is crucial for developing adequate self-management support.

Methods

Qualitative semi-structured interview study among patients with advanced cancer. Transcripts were inductively and thematically analyzed, using open, axial and selective coding.

Results

We interviewed 33 patients with advanced cancer (52% female, aged 33-85 years). Patients described self-management challenges in the medical, psycho-social, existential and daily living domains. Self-management behaviors were personal and often creative, and included the management of physical and psychological symptoms in the home setting, navigating the medical system and coordination of appointments, investing in a good relationship with healthcare professionals -including dealing with incidental conflicts and contributing experiential knowledge to the decision-making-, managing social contacts, and planning -including preparing for the end of life-. Some patients were proactive and articulate in their self-management behaviors, and keen on maintaining autonomy and finding the best possible information and treatments themselves. Most self-management behaviors were aimed at maintaining "normality". Patients' self-management behaviors seemed to be associated with their personality, life history, the moment in the illness trajectory and their social environment.

Conclusions

Self-management behaviors of patients with advanced cancer are personal, often creative, and situate largely beyond the medical domain. They are typically aimed at maintaining a normal life. Self-management support should be tailored to patients' specific problems, needs and personal history. It should be aimed at consolidating effective self-management behaviors and be embedded in trustful partnerships with patients and relatives.

BACKGROUND

Advanced cancer has a large impact on patients' lives(1), and patients deal with its impact in their own, personal ways. Self-management in this context has been defined as "the strategies used by persons with the aim of managing the physical, psychosocial and existential consequences of living with a progressive, life-threatening disease and its treatment"(2). Most research about self-management has been conducted in patients with chronic diseases.

Patients with progressive, life-threatening illness, such as advanced cancer(1), may face other challenges(3-7). Little is known about self-management behaviors of patients with advanced cancer(1). Studies that have been done often focus on self-management (of medication) to alleviate physical symptoms, such as fatigue, shortness of breath, or pain. Fewer studies have been conducted on psychosocial and existential self-management(8-11). In order to provide patients with adequate self-management support that is tailored to their needs, comprehensive insight into the consequences of their illness on their day-to-day lives and their self-management behaviors is needed. Further, only a few studies explored *why* patients self-manage in a certain way. These studies provide some preliminary, partially conflicting evidence that age, level of physical functioning and education might be associated with self-management behaviors(12, 13). Insight in these associated factors could help to elucidate which patient groups might experience difficulties in self-management and how they could best be supported(14, 15).

In this qualitative study, we aim to 1) explore and describe the consequences of living with advanced cancer and its treatment and patients' self-management behaviors to address these consequences, and 2) examine which factors may influence self-management behaviors.

METHODS

Study design and sample

This is a qualitative semi-structured interview study in which we interviewed patients with advanced cancer. Patients were eligible when they were older than 18 years, were diagnosed with advanced cancer (any type), and were able to provide written consent. Advanced cancer was defined as cancer that can no longer be cured (16). We recruited patients purposively, to include a diverse patient group with patients from different cultures and educational levels. Oncologists, general practitioners and nurses from one academic and one general hospital and two hospice care facilities asked eligible patients whether they were interested in participating in the study. Also, calls for participation were circulated via websites of a patient organization, the main research center (Erasmus University Medical Center) and social media platforms. We contacted potentially interested patients by phone and sent them extra information by postal mail. Patients signed for informed consent to participate prior to the interview. We conducted interviews on a location of the participant's choice (mostly at their homes). Participants who needed to travel received reimbursement for their travel expenses. Interviews were conducted until saturation, that is, until no new information arose from the interviews. The Erasmus University Medical Center research ethics committee granted approval of the study (METC-2018-1368).

Data collection

We developed an interview guide prior to starting the interviews, partly based on our systematic review on self-management experiences of patients with advanced cancer (Box 1)(1). We conducted the interviews between September 2018 and July 2019. The interviewers RS, HN, JR, MA and KL took field notes. One interview was conducted and analyzed in Turkish by a Turkish speaking member of the research team (MA). After the interviews, patients completed a questionnaire on demographic characteristics (e.g. type of cancer, year of diagnosis, current treatment). Interviews were audiotaped and transcribed.

Box 1. Interview guide used in conducting the interviews

1. Can you tell me something about yourself and the situation that you are in?
2. What consequences does the illness have on your life?
3. What helps you to deal with the consequences you described?
4. Which of the consequences bothers you most?
5. What could help you to better manage your illness and its consequences?

Data analysis

We performed an inductive thematic analysis, using open, axial and selective coding(17), supported by NVivo 12 pro. Two researchers (RS and HN) read five of the transcripts from the patient interviews line by line and identified initial themes, which were discussed with other researchers (JR, SD, MA, KL). We developed an initial coding tree based on this process and *a priori* (from the interview guide) and adjusted it by going back and forth through the transcripts using the constant comparative method. The coding tree was tested by RS and HN on another five transcripts, and was discussed and adjusted. Subsequently, all transcripts were analyzed by RS and HN. The research team met frequently to refine the initial themes and subthemes by discussing the coded transcripts, themes, subthemes, and each theme in relation to the other themes until no new information arose.

RESULTS

We interviewed 33 patients. Interviews lasted an average of 50 minutes (SD 14 minutes). Seventeen (52%) patients were female (Table 1). The age of participants ranged from 33 to 85 years. Thirty-two patients (97%) had the Dutch nationality, of whom one had a Moroccan background and one a Surinam background. One patient had the Turkish nationality. Most patients resided at home (91%) together with a partner (76%). Most patients had a high school degree or higher (93%). Eighteen patients were recruited in the general hospital, 11 in the academic hospital, two in the hospice, one through social media and one through the website of a patient association.

The results are described in two parts. In part I, we describe the consequences of having advanced cancer and patients' self-management behaviors. In part II, we describe which factors influence patients' self-management behaviors.

Table 1. Characteristics of patients enrolled

	Patients (n=33)
Gender	
Male	16 (49%)
Female	17 (52%)
Age	
18 – 40 years	1 (3%)
41 – 60 years	9 (27%)
61 – 80 years	21 (64%)
> 80 years	2 (6%)
Nationality	
Dutch[1.]	32 (97%)
Other	1 (3%)
Education	
Less than high school	2 (6%)
High school graduate	14 (42%)
Some college	6 (18%)
College graduate	5 (15%)
University	6 (18%)
Living situation	
At home	30 (91%)
Healthcare institution	2 (6%)
Other	1 (3%)
Marital status	
Married/living together	25 (76%)
Not married	5 (15%)
Divorced	2 (6%)
Widowed	1 (3%)
Children	23 (70%)
Of which living at home	6 (18%)
Religious (yes)	12 (36%)
Type of cancer	

Colon	7 (21%)
Lung	3 (9%)
Breast	13 (39%)
Prostate	3 (9%)
Other[2.]	7 (21%)

[1.] One patient had a Moroccan background, one patient had a Surinam background

[2.] Other cancer types were: Liver, peritoneal, penile, cervix, esophagus, melanoma, ovary

Part I – Consequences of having advanced cancer and self-management behaviors

Consequences

Patients described consequences of having advanced cancer and its treatment, in different domains: the physical domain (e.g. nausea, fatigue, changes in appearance), psychological domain (e.g. sadness, feelings of insecurity), social domain (e.g. less social contact, unwanted advice), existential domain (e.g. loss of meaning in life, fear of dying), medical care domain (e.g. complex medication schemes, contact with healthcare professionals), and the daily life domain (e.g. loss of work, difficulties in household activities).

Self-management behaviors

We identified six categories of self-management behaviors in patients' narratives: health behaviors, psychological strategies, seeking and providing social support, collaborating with healthcare professionals, obtaining knowledge and information, and planning. Self-management behaviors were often used in multiple domains. For instance, patients sought social support for help with daily living activities (e.g. household activities) but also for psychological relief (e.g. talking about negative thoughts). Moreover, patients often used different self-management behaviors in the same domain. For instance, psychological consequences of the disease could be addressed by health behaviors (e.g. walking), psychological strategies (e.g. writing down thoughts) and seeking social support. Some self-management behaviors facilitated other self-management behaviors. For instance, adequate planning facilitated patients to engage in hobbies.

Health behaviors

Many patients actively engaged in maintaining a healthy lifestyle, such as staying physically active and maintaining a healthy diet. It gave them the feeling to be 'in charge' of their health. Patients often mentioned walking to both manage physical and psychological discomfort.

> *"If I want to take a break from it all, I put on my good boots […] and I will go out into the polder […] fantastic right? Into the forest. Alone. I think that is the best psychiatrist there is" – P012, male, 73, prostate cancer*

Psychological strategies

Many patients used a variety of personal psychological strategies. Some patients sought help from a psychologist to support them dealing with the emotional burden of their illness, others did not. Sometimes they used cognitive strategies, such as trying to ignore, nuance, or actively deal with negative thoughts. Others described behavioral strategies, for instance seeking distraction by practicing hobbies or writing down thoughts and different scenarios about how their illness might progress.

> *"Usually I use my notebook to come up with a couple of scenarios, ranging from bad to less bad, and then I can read that back. That is my way of managing with everything, to get it out of my head […] It is about all the things that come to mind, doom scenarios, but also hope you cherish" – P034, male, 58, lung cancer*

Seeking and providing social support

Many patients used support from their social network to deal with the consequences of their disease. This sometimes served a practical goal (e.g. helping with household activities, going to clinical appointments), but often it served a more psychological goal. Talking with friends gave relief to many patients. However, some other patients were not always positive about engaging with their social environment. Some said others could be intrusive or make inappropriate comments (e.g. "Hey, you are still here?").

Some patients provided social support to fellow patients. This could include participation in a peer support group, volunteering in a patient association, or being active on social media. Patients were often grateful if they were able to help someone else.

> *"When I speak to a fellow patient and I am able to give some advice or support, that gives me a lot of satisfaction" – P019, female, 58, lung cancer*

Not everyone was interested in peer support groups, as they indicated that talking about their illness would probably only made them feel down.

Collaborating with healthcare professionals

Several patients indicated they invest in a good relationship with their healthcare professional. This provided a trustworthy environment for the discussion of wishes and preferences for medical care. Several patients stressed the importance of their experiential knowledge.

> *"I know a lot about it; therefore I can challenge the doctor. If he says something, I will tell him 'that is not how it works' […] I am no doctor, do not have that expertise […], but I am an expert by experience. I do not know everything, but I do know what the medication is doing with my body" – P021, female, 63, breast cancer*

Some patients noticed that not all healthcare professionals were fully aware of the impact of their illness on their daily life, and that they had to actively bring this up. Most patients were satisfied about the collaboration with their healthcare professionals and the care they received. Some patients indicated they had to deal with conflicts with a healthcare professional, for instance due to different opinions about treatment.

Seeking information

All patients saw their healthcare professionals as the main source of information. For some, the information they received was sufficient. Others used the internet to find more information, although many pointed to possible incorrect information on the internet, or a focus on negative consequences of their illness. Some patients indicated they wanted more information about their illness or (alternative) treatments:

> *"I think that [information about the illness] is highly interesting […]. Scientifically, I have a very broad interest [in my illness] despite the fact that this affects me" – P003, male, 65, prostate cancer*

Planning

Having advanced cancer largely dictated patients' agenda, something some patients struggled with. Planning was a necessary skill for most patients. Many patients valued a daytime routine and tried to live their lives as normally as possible, for instance by continuing their hobbies and usual activities.

> *"I want to live as if everything is just right […] I want to be normal […]. Your life continues and you want to live a normal life" – P007, female, 33, breast cancer*

However, patients were not always able to continue their usual activities in the way they were used to. They sometimes had to distribute their activities over the day, find substitute activities, and actively plan moments of rest, for instance by cooking for multiple days or clustering household activities.

> *"You have to adjust everything to it. I now make sure that if I have to do something upstairs, I immediately also do the laundry, clean the bathroom, iron, so everything is finished upstairs and I do not have to unnecessarily burden myself" – P010, female, 56, breast cancer*

Some patients had difficulties planning their medical care. Some had to check and adhere to difficult medication schemes. Many patients indicated they had to take charge of their clinical appointments, as these were not always well coordinated or did not fit in their personal agenda.

> *"It is complicated, you are right about that, it is complicated. You have an appointment here, another appointment there, another appointment, and another one, you will be in the hospital 100.000 times" – P015, female, 68, cervical cancer*

Some patients also engaged in planning their last phase of life. This ranged from talking with their loved ones about their approaching death to arranging practical matters, such as their finances, mortgages, and buying a grave.

Part II – Factors of influence of self-management behavior

Patients were often unaware of *why* they performed certain behaviors; they described their behaviors as being 'normal' to them. No clear association was found between patients' self-management behaviors and their age, gender, or educational level. We could distill four general factors that influenced self-management behaviors: personality, life history, the moment in the illness trajectory, and the social environment.

Personality

Some patients explained that the way they dealt with their illness was rooted in their personality. Often heard phrases were "That is just how I am", "It is in my nature", and "I have always dealt with things like this".

> *"I do not know; it is just who I am. And everybody says I am managing so well and I am so strong. Well, it is not something I have to work for, I think it is just in my nature" – P006, male, 57, peritoneal cancer*

Many patients described themselves as being assertive. They described that this helped them to protect their boundaries, to stay in charge of their own health, and to discuss concerns and disagreements with healthcare professionals. Other important personality characteristics, according to patients, were to stay realistic, put things in perspective, be flexible, and have a practical mindset without getting carried away by emotions. Also, a positive outlook was important for many.

> *"I am more conscious about it [about being positive]. I was not necessarily negative […] I always danced through life actually. […] But now I think it is actually a survival strategy, positivity. I think if a human can be positive, and is not always grumbling, that you can get very far" – P017, female, 58, breast cancer*

However, some patients indicated that their personality affected their self-management behaviors in a more negative way. One patient for instance described that she always tends to say 'yes' when people ask her for help, which makes her cross her boundaries often, leaving her fatigued and saddened that she cannot do the things she used to do.

Life history

Self-management behaviors were also rooted in how people were raised. Some were born in the second world war and raised with the adage 'not to whine'. Previous experiences with cancer in the family or amongst friends also affected patients' self-management behaviors. Some were strengthened by having witnessed these previous experiences, because people may have survived cancer or were able to cope with the illness in a positive way. Other patients had negative experiences. They described how they saw a loved one poorly managing their illness, and stated that they wanted to do better themselves. Some interviewed patients were healthcare professionals themselves. They described that their professional experiences had helped them in managing their own disease. Some mentioned that they were sometimes approached as a healthcare professional, while they preferred to be approached as a patient.

> *"I am a general practitioner by training, [..] so I am very stubborn in wanting to do things myself […] A negative side is that I have easy access to the medical literature, which means that you sometimes realize things are not as positive as you initially thought […] Another negative side is in the contact with medical specialist, because they will often say 'you understand this, right?'." – P014, male, 59, colon cancer*

Moment in the illness trajectory

Patients often showed a similar pattern in responding to their illness in the beginning of the illness trajectory. Shortly after the diagnosis of advanced cancer, many patients indicated arranging practical matters to seek a feeling of control. Examples are the arrangement their finances, drafting an advance care directive, and obtaining all available information about their illness, (alternative) treatment, and life expectancy. However, as the illness progresses, patients seemed to focus more the integration of their illness into a normal, daily life.

> *"You really have a life before and after your diagnosis, that is really true […]. In the beginning you are changing everything. The moment you hear it, you will drop everything, then slowly you will rebuild everything" – P011, female, 50, breast cancer*

Social environment

Apart from being a source of support itself, the social environment, such as family, friends and colleagues, also affected how patients self-managed their disease and its consequences. For some patients, the social environment was a motivation to try to manage their illness as actively as possible. Some said their family and friends were worth living for or motivated them to engage in certain self-management behaviors:

> *"There are enough people in our lives, friends, who are worth living for, they are sweet and kind and nice" – P001, male, 69, lung cancer*

Furthermore, some patients described that the practical social support they sometimes received meant that they had to do less themselves:

> Q"*It is actually very relaxed, because you do not have to do anything anymore, because nobody is expecting it from you. So it does have something very easy, everybody is very eager to help and support you, because they feel sorry for you*" – P011, female, 50, breast cancer.

DISCUSSION

Having advanced cancer has many consequences for patients in the medical, psycho-social, existential and daily living domains. Our study reveals the full width of the "work" patients are faced with in these domains, which has partly also been shown for patients with chronic diseases(7, 18-20). We found that particularly challenging aspects of living with advanced cancer are having to deal with physical and psychological symptoms in the home situation, navigating the medical system and managing one's medical appointments, maintaining a good relationship with healthcare professionals (which may include dealing with conflicts and contributing experiential knowledge), managing ones' social network, and dealing with existential questions including impending death. The six types of self-management behavior we identified (health behaviors, psychological strategies, seeking and providing social support, collaborating with healthcare professionals, obtaining knowledge and information, and planning) were often used in multiple domains and patients often used different self-management behaviors in the same domain.

Our study shows that patients deal with the consequences of their illness in various, personal and often creative ways. The way in which patients approached their illness and its consequences often seemed to be rooted in their personality and previous life experiences: patients indicated to always have dealt with challenges in a certain way, learned this along the way, or recognized the need to deal with the consequences of their illness in response to a personal experience. A subgroup of patients adopted proactive and articulate self-management behaviors: they actively search for information to obtain an extensive understanding of their illness, they orientate on the best possible treatments, and proactively (re)schedule their medical appointments for a better fit in their personal agenda. This group has previously been referred to as “e-patients”: patients who are “equipped, enabled, empowered and engaged in their health and health care decisions”(21). It may not always be easy for clinicians to relate to e-patients, as their behavior may challenge them to adopt less traditional roles in clinician-patient partnerships(22). For instance, research has shown that many clinicians underestimate the benefits and overestimate the risks of online health resources for patients, and that many patients are hesitant to share internet-based information with their doctors(23). This lack of openness could harm patient-doctor communication and their relationship.

A consistent finding in our study is that patients' self-management behaviors are typically aimed at maintaining normality. Many patients tried to continue the hobbies and usual activities they engaged in before they became ill. Interestingly, patients' desire for maintaining normality seems to be contrary to the common notion that patients want to draft and complete a bucket list, to ‘get the most out of their lives’, and plan e.g. on accomplishing personal goals(24). For several patients, living a normal life was threatened by the medical management of their disease which sometimes largely dictated their agenda. For instance scheduling and attending hospital visits and taking the right medicines at the right time sometimes impaired their normal and usual daily living activities. This is also apparent in other studies(7, 19, 20). Corbin and Straus postulated that chronic illness involves medical work, daily life work and biographical work (18). Being overwhelmed by the medical work might mean that there is less time and energy to be spent on daily life work and biographical work, including family gatherings, placing the illness in ones' biographical context and meaning making of ones' situation. May et al called for *minimally disruptive medicine* as a response to the work that is delegated to patients and families(25). Critically, this involves respecting patients for what they do, as well as for who they are. In their Burden of Treatment Theory they suggest that by redesigning healthcare services so that they are better coordinated, more patient-centered, and acknowledge patient complexity, patients could be better equipped to handle their health problems(26).

Clinical implications

Our study shows that patient self-management largely takes place beyond the classical professional care setting. The consequence is that the experience of illness and the changes it brings to patients' lives often remain unacknowledged by healthcare professionals, while patients' self-management nevertheless affects the care and treatment within the medical setting(27). Healthcare professionals need to relate and respond to the large diversity of self-management strategies they encounter, for instance when patients engage in certain diets or alternative treatments. Moreover, they have the opportunity to support patients in their self-management. Our study provides several insights that they can leverage. First of all, the initiation of open conversations with patients and their relatives is crucial to broach their experiential knowledge of their illness and to identify what consequences of the disease and its treatment patients are faced with, how proactive they are, and what self-management support needs they have. Healthcare professionals could adjust their own style of guidance accordingly. Secondly, discussing to what extent patients attach importance to "maintaining normality" is important, including exploring what normality means, and how medical care and treatment could be provided in such a way that it is minimally disruptive to patients' lives(25). Here, timing is crucial: right after the diagnosis of advanced cancer, many patients tend to focus on emotional and practical matters, but as illness progresses, patients focus more on the integration of their illness into a normal, daily life. Third, we recommend that healthcare professionals map, together with the patient, patients' self-management palette. What self-management behaviors does a patient use commonly, now and in the past? Could the patient leverage these successfully in other domains? What is the role of the family in supporting the patient's self-management? And can the patient, within certain domains, develop new self-management behaviors?

Furthermore, peers could support patient self-management, individually, through social media, or peer support self-management groups. In our study, not everyone was interested in giving or receiving peer support, but some were. As clinical services are unlikely to meet all the needs of patients with advanced cancer, some of patients’ needs could potentially also -or even better- be met outside formal healthcare systems, especially those needs where peers could contribute by sharing their experiential knowledge and creative self-management strategies(28). While peer support is gaining increasing momentum, it is largely neglected -or even raises resistance- in formal healthcare systems(28). Its potential, challenges and potential integration with healthcare systems deserve further study.

Study limitations

This study has some limitations. First, it might be possible that participants with a special interest in the topic, with particular negative or positive experiences towards the care they received, or who were still functioning relatively well, were overrepresented, thereby giving a somewhat biased picture. Furthermore, although we purposively included a relatively large group of patients from a variety of backgrounds, it was difficult to recruit patients from varying cultural backgrounds. This should be considered in future research.

Conclusion

Self-management behaviors of patients with advanced cancer are personal, often creative, and situate largely beyond the medical domain. They are typically aimed at maintaining a normal life. Self-management support should be tailored to patients’ specific problems, needs and personal history. It should be aimed at consolidating effective self-management behaviors and it should be embedded in trustful partnerships with patients and relatives. Self-management of patients with advanced cancer will become increasingly important, partly due to novel treatments that increasingly turn advanced cancer into a chronic condition(29, 30), and partly because clinical services may not be able meet all the needs of those with advanced cancer. Therefore, future studies should focus on innovative methods that support patients in dealing with their illness, that leverage their experiential knowledge of their illness.

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CHAPTER 4

The views of healthcare professionals on self-management of patients with advanced cancer: An interview study

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ABSTRACT

Objectives: Self-management of patients with advanced cancer is challenging. Although healthcare professionals might play a crucial role in supporting these patients, scant scientific attention has been paid to their perspectives. Therefore, we examined healthcare professionals' views on self-management and self-management support in this population.

Methods: We conducted qualitative interviews with 27 purposively sampled medical specialists (n=6), nurse practitioners (n=6), general practitioners (n=8) and homecare/ hospice nurses (n=7) in the Netherlands. Transcripts were analysed using thematic analysis.

Results: Healthcare professionals experienced self-management of patients with advanced cancer to be diverse, dynamic and challenging. They adopted instructive, collaborative and advisory roles in self-management support for this population. Whereas some professionals preferred or inclined towards one role, others indicated to switch roles, depending on the situation.

Conclusions: Just like patients with advanced cancer, healthcare professionals differ in their views and approaches regarding self-management and self-management support in this population. Therefore, instructive, collaborative and advisory self-management support roles will all be useful under certain circumstances.

Practice implications: Healthcare professionals can support self-management by being aware of their own views, and communicating these clearly to their patients and colleagues. Education in self-management support should include self-reflection skills and discuss the relation between self-management and professional care.

INTRODUCTION

In the past decades, there has been a paradigm shift from paternalistic towards more participatory, patient-centered healthcare(1-3). This shift was fueled by broader societal developments. In the Netherlands, for instance, there has been a gradual shift from a welfare state into a “participation society”, where individual wellbeing is no longer primarily a state responsibility and people should create their own support and safety nets(4). Consequently, patients are expected to take a more active role in their health and care. Together with the growing prevalence of chronic diseases and the subsequent need for patients to integrate illness into their daily lives(3, 5), these developments have drawn the attention to self-management. In the context of disease, self-management is commonly defined as ‘the strategies used by persons to manage the symptoms, treatment, physical and psychosocial consequences and lifestyle changes inherent in living with the disease’(6). As such, self-management refers to a comprehensive and interactive process that is not limited to medical management, but also includes daily life management and emotional or identity management(7–9).

The concept of self-management was developed and has mainly been used within the field of chronic diseases. However, also for patients with advanced diseases, such as advanced cancer, self-management concerns a major issue: their disease consequences can be heavily fluctuating and disruptive, involving (invasive) treatment regimens(10), increasingly disabling symptoms(11, 12), and social and existential implications of altered normality and impending death(13, 14). Moreover, as medical and technological advances have increased the average life expectancy of patients with advanced cancer, they generally need to manage these consequences during more protracted, yet uncertain illness and end-of-life trajectories(15). A recent systematic review showed that their self-management strategies are highly personal, varying from self-administering medications to joining peer support groups(16). Additionally, they include specific strategies to manage the end of life, such as sorting out financial affairs and making funeral preparations(16–18). Furthermore, within each self-management domain, strategies are divergent, ranging for instance from coordinating care to fully delegating it to others(16). The accounts of patients with advanced cancer have revealed that adopting and combining strategies can be highly challenging and is pervaded by ambivalence and changing or competing priorities, for example when prescribed medications are experienced to threaten usual daily routine(10, 14, 16, 19, 20).

Ineluctably, self-management of patients with advanced cancer also requires efforts from their healthcare professionals, who need to relate and respond to the large diversity of self-management strategies they encounter. They may be confronted with patients who request or refrain from shared decision-making(16, 21, 22), request access to their medical records (23) or start practising complementary and alternative medicine(16). Besides relating and responding to such strategies, healthcare professionals have the opportunity to support patients in their self-management. Nevertheless, studies to date have mainly addressed patient perspectives. The few studies that did assess professional perspectives focused on nurses(24–29), who are traditionally most closely involved with patients' daily lives, connect the multiple aspects of advanced cancer care, and have therefore been assigned a key role in self-management support(30, 31). Yet, other healthcare professionals, such as medical specialists and general practitioners, need to deal with and support self-management as well. Examining professional views from a broader perspective will enhance our understanding of self-management and self-management support in patients with advanced cancer, and might identify potential targets for improving self-management support. Therefore, we aimed to answer the following research questions:

1. What are healthcare professionals' experiences, attitudes and challenges regarding self-management of patients with advanced cancer?
2. How do they view their own roles in self-management support for patients with advanced cancer?

METHODS

Study design and sample

We conducted a qualitative semi-structured interview study among medical specialists, nurse practitioners, general practitioners and nurses who are involved in care for patients with advanced cancer. These healthcare professionals were recruited purposively via the clinical network of the research group and through the snowball method, thus ensuring variation in characteristics of healthcare professionals (e.g. age, profession, work setting) as well as their patient populations (e.g. cancer type). A multistage sampling approach was adopted, directing recruitment more specifically at healthcare professionals holding deviant views and working in particular settings (e.g. a socio-economically deprived neighbourhood) or fields of expertise (e.g. palliative care, experimental clinical trials) as data collection proceeded(32). Healthcare professionals were invited to participate in the study via email. Those who were interested were contacted again by phone or email to provide them with additional study information and schedule an interview appointment.

Measurements and data collection

Prior to the interviews, a semi-structured interview guide was developed (see Box 1). This interview guide was tested with two colleagues who have clinical work experience and were not otherwise involved in the study. To allow for differences in concept, but also ensure validity and comparability of interview data, we first inquired for participants' views on the definition and meaning of self-management among patients with advanced cancer, and, subsequently, proposed a broad and commonly used self-management definition, which had been adapted to the context of advanced cancer(33). Interviews were administered between March 2018 and July 2018 by three researchers (SD, RS, JR). Healthcare professionals were interviewed face-to-face (mean duration: 1 hour) at a location of their choice, which generally was their work setting. All interviews were audio recorded. At the beginning of each interview, participants provided verbal informed consent. After the interview, they completed a short survey on sociodemographic characteristics. All interviews were transcribed verbatim and anonymised.

Box 1. Interview guide for semi-structured interviews among healthcare professionals

6. What does self-management mean when it concerns patients with advanced cancer?
   - What are your associations and connotations?
   - Introduction of self-management definition: 'the strategies used by persons with the aim of managing the physical, psychosocial and existential consequences of living with advanced cancer and its treatment' (33).
7. What do patients with advanced cancer do to manage the consequences of their disease?
   - What do they generally not do themselves? Are there tasks they mainly leave to you or other healthcare professionals?
8. Do patients with advanced cancer differ in their self-management? If so, how do they differ from each other?
   - Do they differ in their self-management behaviours? Or in their self-management preferences and skills?
9. What factors complicate or facilitate self-management of patients with advanced cancer?
   - E.g. characteristics of patients, their informal caregivers or professional care?
10. Has self-management of patients with advanced cancer changed over the past years, and, if so, how has it changed? How do you experience these changes?
11. How does self-management of patients with advanced cancer affect you and your work?
    - Does it for example influence your professional activities, responsibilities or workload?
12. What is your impression of the consequences of self-management for patients themselves?
    - When do you think self-management has succeeded?
13. What are your ideas about self-management support for patients with advanced cancer?
    - How do you see your own role in self-management support for these patients?

Data analysis

Interview data were inductively analysed following the principles of thematic analysis, using open coding(34, 35). Transcripts were read thoroughly and multiple times to get familiar with the data. Initial codes were assigned and collated into potential themes. Emergent themes were subsequently reviewed, redefined and described in relation with the coded extracts(34). A sample of 10 transcripts were coded by two researchers (SD, RS) independently. The analysis of the remaining transcripts was performed by one researcher (SD) and checked by another (RS). Codes, themes, their interpretations and the clustering of quotes were regularly discussed within the project team. Through constant comparative analysis, experiences, attitudes and challenges regarding patient self-management as well as professional self-management support roles were compared both within and between participants(35). Data on healthcare professionals' roles in self-management support for patients with advanced cancer were classified based on their statements about a) the degree of initiative and control they reported to take in supporting self-management (i.e. self-management support style) and b) the scope of the self-management support activities they described (i.e. the content of self-management support). Sociodemographic characteristics were summarised using descriptive statistics.

RESULTS

Study population

Of the 33 healthcare professionals who were approached, 27 eventually participated. Four professionals did not respond to the invitation emails and reminders, and two refused to participate because of a lack of time. The participating 27 healthcare professionals (74% female; mean (SD) age: 50 (11) years) worked as general practitioner (n=8), medical specialist (n=6), nurse (n=7) or nurse practitioner (n=6) in the hospital, home or hospice setting in various regions of the Netherlands (see Table 1).

Table 1. Background characteristics of healthcare professionals included (n = 27)

Characteristic	n (%) Mean (SD); [min – max] [a]
Age in years	50 (11); [30 – 64]
Sex	
Male	7 (26)
Female	20 (74)
Profession	
General practitioner	8 (30)
(Trainee) Medical specialist (oncology, internal medicine, otolaryngology, head and neck surgery, pulmonology)	6 (22)
Nurse	7 (26)
Nurse practitioner	6 (22)

Work setting	
General practice	8 (30)
Academic hospital	9 (33)
General hospital	3 (11)
Home care	5 (19)
Hospice care	2 (7)
Years of working experience	22 (12); [2 – 39]

[a] Categorical characteristics presented as n (%); Continuous characteristics presented as mean (SD); [min – max]

Abbreviations: n = number; SD = standard deviation; min = minimum; max = maximum

Experiences with self-management of patients with advanced cancer

Healthcare professionals associated self-management of patients with advanced cancer with choices and behaviours in various domains. Some of them described it primarily in clinical terms, focusing on strategies related to disease management and medical care, such as complying with treatment regimens, monitoring symptoms and navigating the healthcare system. Others adopted a broader perspective, addressing choices and behaviours regarding life in general, including domains of social support and existential wellbeing. According to some of them, patients' strategies were not always compatible with their professional care, for example when patients adopted stringent diets while chemotherapy required increased energy intake, or when they deliberately withheld information about their symptoms for fear that professionals would decide to discontinue their treatment. Without exception, professionals mentioned the use of alternative medicine among a considerable proportion of their patients with advanced cancer, including strategies that could alter the safety or effectiveness of regular therapies. Some of them discussed their experiences with patients who substituted regular care with alternative care strategies:

> *'I have encountered patients who decided to quit medical treatment because their faith healer had told them their cancer was gone.' – HCP02, medical specialist*

Participants experienced large differences in self-management between patients with advanced cancer, describing a variety of different patient roles and styles. On the one hand, they provided examples of patients showing proactive behaviours, and, on the other hand, they distinguished patients who adopt more passive or even avoidant approaches in their presence:

> *'Some patients are very determined. Patients who have mapped out a detailed route for their disease trajectories and might go very far to realise the intended route, by searching and reading*

> *scientific literature themselves, networking and approaching people, arranging second opinions and things like that. Other patients are much more passive, just like... "Doctor, or nurse, whatever you say, that is how we are going to do it." – HCP07, medical specialist*

Healthcare professionals shared the experience that recent healthcare and broader societal developments have urged patients to be more proactive in their self-management. They described the implementation of online patient portals that contained increasing amounts of medical correspondence and sometimes even disclosed clinical test results before these had been discussed between patient and professional. They also spoke about the emergence of a 'self-management industry', in which business people try to advertise their self-management support programmes within medical settings:

> *'I receive emails saying "We know you work at the oncology clinic, I have written a book about breast cancer and would like to bring it to your attention, can I come visit?" […] Coaches who visit the hospital and ask me whether they may distribute some leaflets here (shows a brochure). […] That is certainly different from 10 years ago.' – HCP21, nurse practitioner*

Professionals with many years of work experience saw these developments reflected in the behaviours of their patients, who generally had become more assertive over time. Examples included patients who initiated mobile recordings of their medical consultations, ordered wound care materials themselves or turned down supportive care offered by the treating healthcare institution because they had already arranged it elsewhere. However, professionals also expressed concerns about patients considered 'unwilling or unable to be in charge', as these patients are 'disadvantaged by the shift away from traditional, paternalistic medicine' and might subsequently 'fall behind', resulting in increased health inequalities between patients:

> *'Self-management has become increasingly proactive, because that is what the current healthcare system and society in general demand from patients. However, not all patients can keep up. […] So we run the risk that people are being left behind, especially the low-literate and the people who do not see any examples in their environment or just do not have the intelligence to manage it all.' – HCP01, medical specialist*

As shown by abovementioned statement, healthcare professionals experienced self-management to be highly dependent on patients' skills and available resources. Multiple other patient characteristics were considered important as well. While professionals considered some associations clearly present at a population level, such as the use of proactive and persevering strategies by patients with young children, they frequently noted the pitfalls of generalisation in individual patient care. They illustrated this with examples of patients who had surprised or otherwise impressed them with their self-management:

> *'Currently, I am treating a patient whose oncologist gave him a free hand to take pain medications. A high-educated person. Yet, last week, he told me he is not able to do it, because he cannot estimate what to do.' – HCP08, nurse practitioner*

> *'We once treated a lady with ovarian cancer [...]. She had two little children. Probably, she could have survived, but she decided to forgo chemotherapy. A pretty woman with beautiful hair, who just absolutely did not want to lose that hair. While we assumed... well, I do not care about my hair so much... so I expected another choice.' – HCP20, nurse practitioner*

The nearing end of life was experienced to have a big impact on patients' self-management, especially when it came rather unexpectedly and patients were still actively involved in life. According to some professionals, a persistent will to survive or fear of death could cause patients to conduct 'desperate', 'extreme' or 'unrealistic' searches for potentially life-prolonging, yet risky and costly treatment options:

> *'With the cannabis oil, for example. Patients go looking for it themselves, but everyone can put anything on the internet and claim to be an expert. And if you are a layman and you have just been told that you are incurably ill, then you are amenable to everything. […] Those are the people who go to Germany for unproven invasive treatments.' – HCP21, nurse practitioner*

Conversely, healthcare professionals also experienced that for other patients, the news about their prognosis was so overwhelming that it 'confused' or even 'paralysed' them. Furthermore, professionals described how the prospect of impending death could lead patients to reconsider their values and priorities and rearrange their lives, resulting in changing ways of self-management:

> *'I treated this neurologist, the most rational neurologist ever. He got a tumour and became interested in spirituality and started using alternative medicine. He himself found it strange as*

> *well, but told me he felt a very strong need to try it. So sometimes, you see people change when the end is near.' – HCP09, general practitioner*

Attitudes and challenges regarding self-management of patients with advanced cancer

Some professionals were optimistic about self-management of their patients with advanced cancer, considering them 'to know more than they think they know' and 'to be less and less surrendered to the almighty medical world and become more and more empowered'. Others, who focused more on patients' participation in medical care and treatment, were sceptical, indicating that they 'noticed little self-management' and that patients who do participate are 'the ones they remember at the end of the day'.

Irrespective of their preponderant attitudes, healthcare professionals indicated that self-management of these patients posed considerable challenges for professional care and treatment, arguing that their jobs had become 'bigger' and 'more varied'. Furthermore, patients' self-management prevented them from 'working on the autopilot', as they 'had to keep up to date', also about matters that used to be outside of their professional scope, such as lifestyle hypes. According to different healthcare professionals, dealing with self-management was especially challenging or even disturbing in case of incongruity between patients' strategies and their professional circumstances, capabilities, opinions or responsibilities (see Box 2). Difficulties also arose when care and support from informal caregivers were experienced inconsistent with the patient's best interest.

Box 2. Healthcare professionals' challenges to dealing with self-management of patients with advanced cancer

1. Institutional or educational barriers, e.g. insufficient opportunities (e.g. time), knowledge or skills:

> *'Just giving a patient his diagnosis, treatment plan and a bit of explanation about the side effects versus additionally discussing his priorities, preferences, values, meanings, [...]. The latter is a completely different conversation, which takes more time. However, in the current hospital payment structure, there is no reward for scheduling extra consultations, except for my own feelings of integrity.' – HCP01, medical specialist*

2. Discrepancies between patients' self-management strategies and professional opinions or responsibilities:

> *'One of my patients has advanced colon cancer and she is doing all kinds of vague alternative and experimental treatments, also abroad. For us that is very difficult, because there is no transparency about what happens. Yet, if complications occur, these patients come to our hospital, expecting us to resolve them. This becomes increasingly problematic, especially since it concerns a growing patient group.' – HCP05, nurse*

3. Doubt or disapproval regarding the influence of informal caregivers on patients' self-management:

> *'I just asked a patient about her medication and then that lady said: "My husband knows all that". Or when I ask a patient about his stools and he looks at his partner. […] I have difficulty with that, because it means that someone has fully relinquished his self-management to relatives. Also, I see many relatives who completely take over self-management. By forcing a patient to eat, for instance.' – HCP08, nurse practitioner*

Healthcare professionals' views on their own roles in self-management support for patients with advanced cancer

In reaction to their experiences, attitudes and challenges regarding self-management of patients with advanced cancer, healthcare professionals expressed different views on their own roles in self-management support for these patients. We identified the following three roles: 1) instructive; 2) collaborative and 3) advisory (see Box 3). Whereas some healthcare professionals preferred or inclined to adhere to one of these three roles, others indicated to switch their roles, depending on the situation.

Box 3. Healthcare professionals' roles in self-management support for patients with advanced cancer

1. The instructive role:

Directing patient self-management based on professional expertise; the healthcare professional is leading.

> *'Self-management works well when patients have the instructions and the recipes and have already read those when coming to the hospital, so that they can follow our steps. Patients who can accept and follow advice… If a patient allows you to take him by the hand, I don't mean to be patronizing, he will deprive himself a little less.' – HCP08, nurse practitioner*

2. The collaborative role:

Integrating professional-directed and patient-directed strategies, using a clear and well-delineated division of roles and tasks between professional and patient.

> *'Patient and professional both need something from each other. The patient needs my knowledge and the pros and cons of everything and I need to know what suits the patient, what type of person he or she is and how he or she lives and used to live.' – HCP15, medical specialist*

3. The advisory role:

Following patient-directed strategies using a holistic approach; the patient is leading.

‘You should listen rather than talk to your patients. The most important thing is to give them the opportunity to bring up whatever they want to discuss, because these are often things you would not have thought about.’ – HCP12, general practitioner

The instructive role

In the instructive role, healthcare professionals tried to direct and stay in charge of self-management of patients with advanced cancer. Directions mostly concerned strategies related to managing the disease and medical care, such as monitoring and controlling symptoms and maintaining activities of daily living. Healthcare professionals often expressed a large sense of responsibility for their patients to understand and comply with their instructions:

> *‘I have created treatment diaries, in which patients should monitor their symptoms and compliance with therapy. Yet, if I do not explain this, nothing happens. You need to keep repeating and checking whether the patient can follow you.’ – HCP08, nurse practitioner*

Instructions could pose limits to patient-directed self-management activities, but were considered necessary to achieve individual healthcare and treatment goals, which were also mainly focused on disease and medical care and included ‘improving symptom control and quality of life’, ‘keeping a patient on treatment as long as possible’ and ‘increasing patients’ awareness of their medication regimens and reasons to call for medical assistance’. Furthermore, ‘optimal’ self-management would contribute to more efficient and effective care, thus also benefiting the healthcare professional and the overall healthcare system. Hospital culture was mentioned to foster instructive approaches. Compared to the other professional disciplines, nurse practitioners referred to these approaches most often, especially if they cared for patients receiving experimental clinical treatments:

> *‘Patients have signed a treatment agreement, which involves expectations and obligations. Of course I want to think along about how it suits them best, but the fact remains that they are on a leash, because otherwise the treatment will be at risk.’ – HCP18, nurse practitioner*

The collaborative role

In the collaborative role, healthcare professionals integrated their professional support strategies with patient-directed strategies. Collaboration with patients was realised by establishing a clear and well-delineated division of complementary tasks between professional and patient. Healthcare professionals wanted patients to reflect on their personal circumstances, needs, values and preferences and considered these in their care and treatment policies. Yet, they also expected patients to rely on their professional expertise and consider this in their daily life:

> *'Patients who research scientific studies down to the smallest detail […]. They function on a high level and find a lot of information, but still lack the medical knowledge to interpret it correctly. […] That is something we should try to avoid.' – HCP01, medical specialist*

If they found patients' self-management behaviours incompatible with their professional expertise or protocols, they would intervene by finding an alternative everyone agreed on:

> '*The art is to make sure that patients are satisfied when they leave the room, yet, without that referral letter or that pot of weed they insisted on, because (providing) that would be such a waste of time, or a waste of the drug.' – HCP21, nurse practitioner*

Healthcare professionals who adopted a collaborative role indicated that self-management support had succeeded if patients would have 'received care and treatment that incorporated their personal values and circumstances'. Better interprofessional collaboration, for instance by means of 'more appropriate referral to supportive care disciplines, such as psychology', was also explicitly mentioned as a goal that would benefit both patient self-management and professional self-management support. In their stories about the work experience they had gained, healthcare professionals often signified a shift from instructive towards more collaborative self-management support roles:

> *'Filling your backpack is something you do throughout your career. At a certain point, this baggage gives you an overall picture, which makes you understand that you do not always have to stick to protocols and decision trees. It is not either A or B; it may be a combination of both. Gradually, you learn to make those combinations faster and deal more flexibly with the patient and his personal wishes.' – HCP05, nurse*

The advisory role:
In the advisory role, healthcare professionals adjusted their professional support strategies to patient-directed strategies, organising and arranging care and treatment starting from the needs and preferences articulated by the patient. Patient self-management as well as professional self-management support were approached holistically and could also include more personal aspects of a patient's life:

> *‘Currently, I am treating a patient with advanced hereditary breast cancer, who has entered the rollercoaster of radiotherapy, surgery, chemotherapy, everything. However, with me she prefers to talk about ordinary things, such as the wig. So for some patients, you are more like a confidant.’ – HCP06, general practitioner*

The personal, holistic approach of healthcare professionals who adopted an advisory role was also reflected by their statements about the goals of self-management and self-management support, which included that patients would ‘make choices that suit their personalities’ and ‘keep living besides their illness’. As long as neither the patient’s safety nor their own feelings of integrity were compromised, healthcare professionals with an advisory style left patients free in their self-management behaviours and their decisions about the frequency and intensity of professional support:

> *‘One of my patients had a colossal tumour just below the knee. […] She did not want to be referred […] or talk about death. As soon as I steered the conversation in that direction, she cut it short in all kinds of creative ways. [...] I think that is something we (healthcare professionals) have to respect.’ – HCP12, general practitioner*

Some professionals suggested that gaining life experience had encouraged them to adopt more advisory approaches:

> *‘My own mother passed away last year. […] It taught me things I now apply in my work, like starting from the needs patients express. Because now I know that it was most supportive if healthcare professionals just asked us how we were doing. I already knew that, but now I feel it even more.’ – HCP26, nurse*

Determining the appropriate self-management support role

Healthcare professionals often explained that they based their self-management support roles on specific characteristics of their patient populations. However, because of deviating views and preferences, they responded differently to similar populations:

> *'Among patients with head and neck cancer, there are quite some people who avoid regular care and have unhealthy lifestyles. […] We struggle with the responsibility for these patients and then often take on the paternal role.' – HCP02, medical specialist*

> Versus:

> *'Compared to general populations, homeless people tend to be even more autonomous in their self-management. For example, that woman who seized every opportunity to obtain drugs. [...] Of course, we tried to address the issue, but [...] it would happen anyway. Moreover, it was her way of self-managing.' – HCP20, general practitioner and street doctor*

Likewise, the fact that patients were incurably ill and at the end of life influenced professional support roles in various ways. Several participants explicitly mentioned that they approached patients with advanced diseases less instructive compared to patients with chronic diseases:

> *'In chronic care, you try to let people live as healthy as possible. In advanced cancer care, the goal is to promote quality of life in the last phase [...]. It (self-management support) really serves a different purpose.' – HCP06, general practitioner*

Another reason for being less instructive was a lack of professional consensus about the options for end-of-life care and treatment:

> *'Of course, I think patients should know the consequences of their actions, but the thing is that with these patients (with advanced cancer), we often have no idea (about the consequences). So who am I to say what someone should or should not do?' – HCP14, medical specialist*

On the contrary, other professionals pointed out that the enormous impact of advanced cancer on patients' lives led them to adopt a more instructive support role:

> *'Many patients, including clever and organised people, are so overwhelmed by emotions that they are trapped in a state of collapse. […] They need a professional who tells them "this is how we are going to do it".' – HCP15, medical specialist*

Eventually, the match between the various characteristics of professional and patient determined whether healthcare professionals perceived their self-management support as successful and satisfactory:

> *'I am lucky with my patients, because I like to explain things and give advice, while they like to get explanations and follow advice. But should I change my approach if a patient would not like it? That would be very hard.' – HCP18, nurse practitioner involved in experimental clinical trials*

Many healthcare professionals attempted to enhance this match by tailoring their support role to the individual patient they were working with. They did so by 'exploring the patient's background and context', 'observing a patient's body language', 'feeling and reading the patient', 'identifying the question behind a question', and 'building a trusted relationship with the patient'. Furthermore, self-reflection and becoming aware of one's own preferences, expectations and convictions were declared essential for determining the appropriate self-management support role:

> *'Ideally, self-management will make patients feel resignation, catharsis. However, that is very much how I see it. […] By contrast, many of my patients want to have tried everything and remain in control until their last breath. As healthcare professionals, we need to become aware of our own beliefs and realise that these do not necessarily have to coincide with those of our patients or colleagues. […] Therefore, we must be careful with our own projections.' – HCP14, medical specialist*

DISCUSSION AND CONCLUSION

Discussion

Experiences, attitudes and challenges regarding self-management of patients with advanced cancer

This interview study showed that healthcare professionals in various medical and nursing disciplines experienced self-management of patients with advanced cancer to be highly individual, diverse and potentially dynamic. Their attitudes towards self-management varied from mostly optimistic to sceptical, with more scepticism observed among professionals who concentrated on medical aspects. Professionals unanimously mentioned that self-management posed significant challenges to their care and treatment. Despite observing this population becoming increasingly proactive and enterprising in its self-management, they expressed concerns about patients who cannot or do not want to live up to the expectation of being actively engaged in their healthcare. Moreover, proactive, patient-directed strategies conveyed substantial difficulties for them as well. Our study confirms previous findings indicating that self-management of these patients is not always compatible with professional care and expertise(16, 22, 36–40), for example, when patients choose to withhold relevant information about their symptoms. Also, patient self-management goes largely beyond the professional care setting, while nevertheless affecting care and treatment within that setting. In this regard, professionals frequently discussed their experiences with patients who made rigorous lifestyle changes or started using complementary and alternative strategies that could alter the safety and effectiveness of regular medical therapies. In general, healthcare professionals seemed to have more difficulty with self-management when they could not reconcile patients' or their relatives' strategies with their own professional circumstances, capabilities or responsibilities, or their ideas about what sensible self-management would entail.

Roles in self-management support for patients with advanced cancer

In this study among healthcare professionals from various medical and nursing disciplines, we identified three different roles in self-management support for patients with advanced cancer: 1) instructive, directing self-management based on professional expertise; 2) collaborative, using a well-delineated division of complementary tasks between patient and professional; and 3) advisory, following patient-directed strategies using a holistic approach. Previous studies have also used typologies to describe healthcare professionals' self-assigned and performed roles in patient-professional relationships(26, 28, 41–46). However, these studies differ from our study in several ways: they examined these roles from other, either narrower or wider angles, such as decision-making (44), goal-setting (43) and patient-professional communication in general(41, 42, 45); focused on chronic disease populations (26, 28,) or general patient populations(41, 42, 44, 45); included only a single type of healthcare professionals, such as nurses (26, 28, 46) or physicians(41, 42, 45); or based typologies on theoretical rather than empirical findings(41, 42, 46). Consistent with two studies that examined nurses' self-perceived roles in self-management support for patients with chronic diseases(26, 28), we found that the scope of self-management support varied from predominantly biomedical (in the instructive role) to holistic (in the advisory role). However, while both nursing studies derived their typologies primarily from such differences in self-management support content, in our study, differences in self-management support style, i.e. the degree of initiative and control taken by healthcare professionals, emerged as the main and most univocal criterion for classification.

The healthcare professionals in our study indicated that their self-management support roles were shaped by patient characteristics. The influence of the end of life of patients seemed to be bidirectional. On the one hand, a lack of professional medical consensus and a shifted focus towards preference-sensitive care and treatment decisions could cause professionals to take on a more accommodating, advisory support role. On the other hand, the ‘extreme’, ‘desperate’ or ‘paralysed’ ways in which some patients were considered to self-manage at the end of life, resulted in more controlled, instructive support roles. Differences in professional self-management support styles thus seemed to be a response to the divergence experienced in self-management strategies of patients with advanced cancer. However, professionals attributed preferred and adopted roles not merely to their patients, but also to other, work-related and personal factors, such as their work setting and the life experience they had gained. These findings endorse the hypothesis raised by Been-Dahmen et al., who questioned whether the absence of an expected association between nurses’ self-management support views and their patients’ characteristics could be explained by an attenuating or overruling effect of nurses’ personal characteristics(26). Our results also corroborate prior findings indicating that physicians assess self-management support from a broader, more holistic perspective if they have experienced adversity themselves(47). Most importantly, adopted support roles were not static, but rather dynamic, resulting from interactions between professionals, patients and their environments.

Methodological considerations and future research

This study expands the scope of our knowledge and understanding of patient self-management and professional self-management support by adding novel insights from the context of advanced disease and the perspective of different healthcare disciplines involved. Its qualitative design and heterogeneous study population enabled a phenomenological examination of the variation in healthcare professionals’ views. However, this method also has limitations, as it may fail to uncover unconscious, implicit ideas and actual behaviours during patient-professional encounters. Furthermore, a lack of saturation within categories of certain characteristics, such as professional setting, hampers firm conclusions about the nature and direction of their associations with self-management support roles. Further research needs to test the hypotheses generated in this study, which for instance suggest that instructive self-management support roles are more prevalent within hospitals compared to other care settings. Future studies could also investigate distributions of the different support roles in a larger sample, including professionals from supportive care disciplines, such as psychologists and chaplain workers. Additionally, future studies using longitudinal methods and audio-visual aids might be able to scrutinise the dynamics between patient self-management and professional self-management support in actual care practice and over time. Finally, it is important to examine what healthcare professionals in different self-management support roles need in order to fulfil these roles in a satisfying, self-congruent and effective way.

Conclusion

Self-management is not only challenging for patients with advanced cancer, but also for the professionals involved in their care: they need to support a wide variety of patients with versatile and potentially dynamic self-management styles, activities, preferences and needs. While some healthcare professionals predominantly applaud patients' strategies, others express more hesitant attitudes. Healthcare professionals hold different views on self-management support in advanced cancer, adopting instructive, collaborative and advisory roles. Whereas some of them prefer or incline towards one of these roles, others adjust or switch roles, depending on the interactions with patients and their environments.

Practice implications

Just as patients with advanced cancer have different approaches to self-management, healthcare professionals differ in their approaches to self-management support for these patients. Instructive, collaborative and advisory self-management support roles will therefore all be appropriate and useful under certain circumstances. Also, it might be unfeasible and even undesirable to expect all healthcare professionals, regardless of their work setting and discipline, to become proficient in all self-management support roles. Nevertheless, professionals who are able and willing to adopt multiple self-management support roles can meet the needs and preferences of a larger diversity of patients. Regardless of their preferred or adopted support roles, healthcare professionals could facilitate patient self-management by being aware of their own preferences, expectations and convictions, and communicating these clearly to their patients and colleagues. We therefore recommend that all healthcare professionals involved in care for patients with advanced cancer receive education and training that, besides practical self-management support skills, teach skills to increase self-knowledge and critical self-reflection. Within education, the debate about how patient self-management relates to professional care and treatment, and what this means for the organisation and responsibilities of healthcare, should be encouraged.

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CHAPTER 5

The incidence and impact of implantable cardioverter defibrillator shocks in the last phase of life: An integrative review

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ABSTRACT

Background

Although the implantable cardioverter defibrillator is successful in terminating life threatening arrhythmias, it might give unwanted shocks in the last phase of life if not deactivated in a timely manner.

Aims

This integrated review aimed to provide an overview of studies reporting on implantable cardioverter defibrillator shock incidence and impact in the last phase of life.

Methods and results

We systematically searched five electronic databases. Studies reporting on the incidence and/or impact of implantable cardioverter defibrillator shocks in the last month of life were included. Fifteen studies were included. Two American studies published in 1996 and 1998 reported on the incidence of shocks in patients who died non-suddenly: incidences were 24% and 33%, respectively, in the last 24 hours, and 7% and 14%, respectively, in the last hour of life. Six American studies and one Danish study published between 1991–1999 reported on patients dying suddenly: incidences were 41% and 68% in the last 24 hours and 22–66% in the last hour. Four American studies and two Swedish studies published between 2004–2015 did not distinguish the cause of death: incidences were 17–32% in the last month, 3–32% in the last 24 hours, and 8% and 31% in the last hour of life. Three American studies published between 2004–2011 reported that shocks in dying patients are painful and distressing for patients, and distressing for relatives and professional caregivers.

Conclusion

If the implantable cardioverter defibrillator is not deactivated in a timely manner, a potentially significant proportion of implantable cardioverter defibrillator patients experience painful and distressing shocks in their last phase of life.

INTRODUCTION

During the last phase of life, goals of care usually shift from prolonging life to maintaining quality of life.(1) Many patients with heart failure eventually die due to progression of their underlying heart condition, despite the many treatments that are currently available.(2) Whether certain life-sustaining treatments are still appropriate in this last phase of life is an important topic of discussion. The implantable cardioverter defibrillator (ICD) is one of these life prolonging treatments that may pose difficult dilemmas in the last phase of life. The ICD is currently the treatment of choice in patients who are at risk of sudden cardiac death because of ventricular arrhythmias.(3) In Europe, more than 85.000 ICDs were implanted in 2013, based on 46 countries, mainly in Western Europe.(4) The number of new implants is still increasing, due to both an ageing population and expanding of the indication for ICD implantation, shifting from secondary prevention (implantation in patients that already have experienced life-threatening arrhythmias or aborted cardiac death) to primary prevention (implantation in patients with an elevated risk of arrhythmias or cardiac death who have not yet experienced such an episode).(5)

Although the ICD is effective in prolonging life, it poses challenges to the patient, such as (fear of) experiencing painful shocks and feelings of helplessness, because of the unpredictable nature of the arrhythmia and subsequent shocks.(6) During the last phase of life, the benefits of the ICD may no longer outweigh these challenges and it may be important to discuss whether or not to deactivate the device.(1) However, such discussions can be challenging and complex for healthcare professionals, patients, and their relatives.

It is however unclear to what extend ICD patients experience shocks in the last phase of life, and what the impact of these shocks is, specifically at the end of life, on the patient, the patient's relatives and the professional caregivers. Therefore, the following research questions will be addressed: (a) what is the incidence of appropriate and inappropriate ICD shocks in the last phase of life?; and (b) what is the impact of ICD shocks in the last phase of life on patients, their relatives, and their professional caregivers?

METHODS

Data sources and search strategy

Research questions, search strategy and inclusion criteria were specified in advance and documented in a protocol. A search query was developed to identify relevant papers. In August 2016, we systematically searched five electronic scientific databases: Embase, Medline, Cochrane Central, Web of Science and Google Scholar. In order to find all relevant literature, we did not place a limitation on year of publication, all articles up to August 2016 were considered for inclusion. Studies were identified with the following keywords: defibrillator, defibrillator pacemaker, implantable cardioverter defibrillator, internal defibrillator, ICD, AICD; and shock, electric shock, electroshock, countershock, cardioversion, convulsive therapy; and terminal care, terminally ill patient, EOL, death, dying, palliative, hospice, last phase, last year, last month, last week, last day. Boolean operators were used in between key words. The detailed search queries can be found in Supplementary Material Appendix 1.

Study selection and eligibility criteria

Articles were reviewed by RS and AB via a stepwise procedure according to the PRISMA guidelines.(7) First, studies were screened on title and abstract. Selected studies were subsequently reviewed on full text and either included or excluded. In case of disagreement, consensus was sought and achieved. Table 1 describes the inclusion and exclusion criteria. Studies were included when they reported on deceased patients with an active ICD in the last phase of life, either on the incidence of ICD shocks or on the impact of shocks on patients, relatives or professional caregivers. The last phase of life was operationalised as the last month preceding death. This was based on a study that reported on shocks one month before death.(8) Relatives were not limited to family members but could also include others, such as close friends or other loved ones. Studies had to be written in English, and the full-text of the article had to be available. Studies were excluded when they reported on minors or were case reports. References lists from the included studies were examined to identify additional relevant studies.

Table 1. Inclusion and exclusion criteria.

Inclusion criteria

1. Studies are empirical (both quantitative and qualitative) and report on deceased ICD patients that had an active ICD in the last month preceding death.
2. Studies report on the incidence and/or impact of ICD shocks in the last month preceding death.
3. Studies are in English.

Exclusion criteria

1. Studies in minors (<18 years of age).
2. Study is a case report.
3. Full text unavailable.

ICD: implantable cardioverter defibrillator.

Data extraction

Data of the studies that were included in the review were extracted via an extraction form. This form was developed by RS, AB and JR and piloted by RS and AB. The eventual data extraction was completed by RS.

The following data were extracted:

1. Shock incidence, where possible stratified by type of death. Type of death could be non-sudden death (NSD), defined as death occurring more than one hour after the onset of new symptoms, or sudden death (SD), defined as death that occurs less than one hour after the onset of new symptoms.(9) Shock incidence was calculated by dividing the number of patients in a specific group receiving shocks by the total number of patients in that group.
2. Timing of shocks, categorised in three time periods: the last month, the last 24 hours and the last hour preceding death. When a study reported on multiple time periods, patients who experienced shocks during the last hour or last 24 hours preceding death were also included in the incidence in the last 24 hours and last month before death.
3. Appropriateness of shocks. An appropriate shock was defined as a shock for ventricular tachycardia (VT) or ventricular fibrillation (VF). An inappropriate shock was defined as any shock not delivered for VT or VF.
4. Impact of shocks on either patients, relatives, or professional caregivers. All available data were extracted.
5. Characteristics of the study, such as year of publication, study design, aim, year of implantation, year of death, and characteristics of the participants.

Quality assessment

Methodological quality was assessed with the Quality Assessment Tool.(10) Via this tool, studies were evaluated regarding nine items: abstract and title, introduction and aims, methods and data, sampling, data analyses, ethics and bias, results, generalisability, and implications. Each criterion was scored on a four-point Likert scale, ranging from one (very poor) to four (good). In total, a summed score of 9–36 was calculated (9=very poor, 36=good). Studies with scores between 30–36 were assessed as high quality, studies with scores between 24–29 were assessed as moderate quality, and scores lower than 23 were assessed as low quality. Studies were not excluded based on their methodological quality.

RESULTS

The search yielded a total of 4246 studies. We removed 1875 duplicates. All titles and abstracts were reviewed, after which 82 articles were available for full text assessment. Eventually, we included 15 studies (see Figure 1), comprising a total population of 1362 (range 4–558) patients. All studies were observational. The majority of the studies were conducted in the USA (12), the remainder in Europe (three). A total of 12 studies reported solely on shock incidence (80%), two solely on the impact of shocks (13%) and one on both the incidence and impact of shocks (7%) (see Table 2). Studies scored moderate to high on methodological quality (Tables 3 and 4). No additional studies were identified after examination of the reference lists of the included articles.

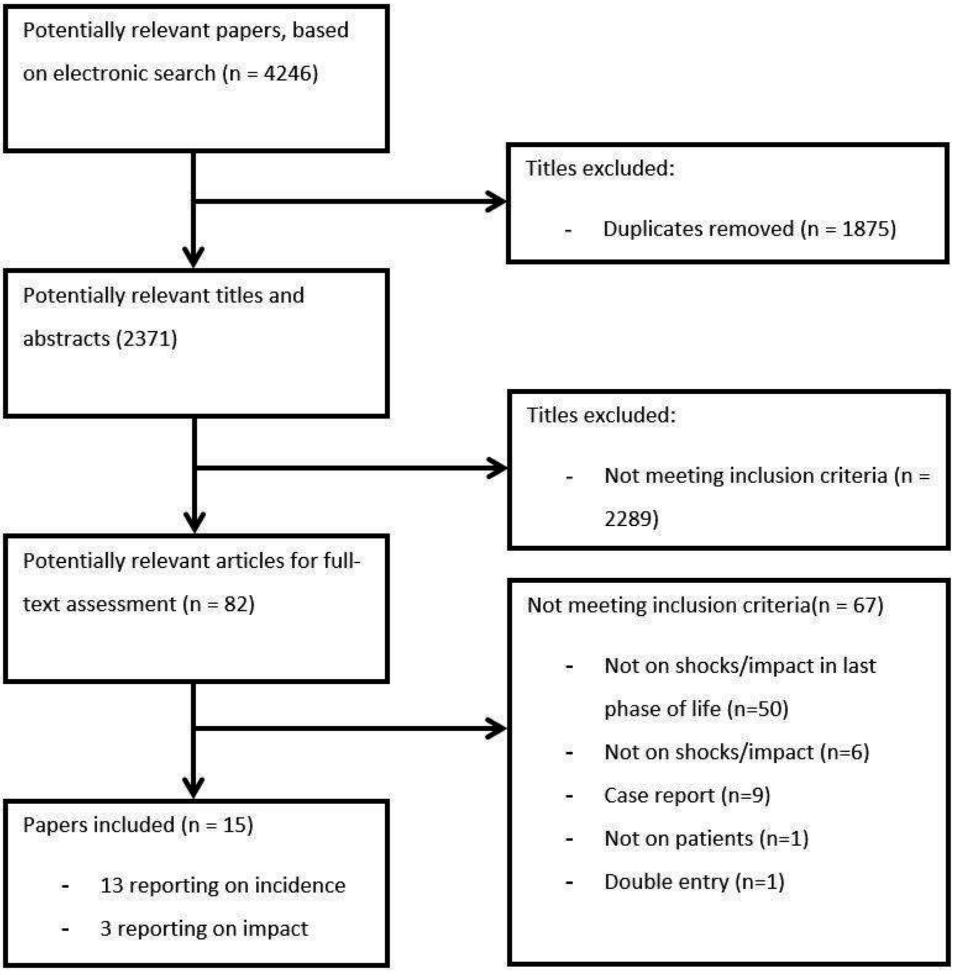

Figure 1. Flow diagram of literature search to identify articles reporting on the incidence and/or the impact of ICD shocks in the last phase of life.

Table 2. Characteristics of the included studied (*n* = 15)

Study characteristics		*n* (%)	
Type of study	Quantitative	15	(100%)
Country	USA	12	(80%)
	Sweden	2	(13%)
	Canada	1	(7%)
	Denmark	1	(7%)
	New Zealand	1	(7%)
Study on	Shock incidence	13	(87%)
	Impact	3	(20%)
Number of patients in study	0-50	7	(47%)
	50-100	7	(47%)
	>100	1	(7%)

Incidence of ICD shocks

A total of 13 studies reported on shock incidence (Table 3). The year of publication ranged from 1991–2015. A prospective and a retrospective study, both high quality studies published in 1996 and 1998, reported on shock incidence in NSD patients.(11, 12) All deaths were of cardiac origin. In these studies, 24% and 33% of patients respectively experienced one or more shocks in the last 24 hours, and 14% and 7% experienced one or more shocks in the last hour preceding death. One study reported on the appropriateness of the shocks,(12) and found that all shocks were appropriate.

Seven studies reported on shock incidence in SD patients.(11-17) Year of publication ranged from 1991–1999. Studies scored moderate to high on methodological quality. All deaths were cardiac of origin. Two studies reported on shock incidence in the last 24 hours preceding death,(11, 12) one prospective study showed an incidence of 41%, a retrospective study showed an incidence of 68%. All seven studies reported on shock incidence in the final hour of life, showing an incidence ranging from 22–66%. One study reported on the appropriateness of the shocks,(16) and found that all shocks were appropriate.

Two studies from 1996 and 1998 specifically reported on patients dying of noncardiac causes.(11, 12) The first study showed that 20% of patients dying of a noncardiac cause experienced shocks in the last 24 hours, and 8% experienced a shock in the last hour preceding death.(11) The second study showed that one out of 36 patients (3%) experienced a shock in the final hour of life.(12)

Table 3. Information on studies reporting on implantable cardioverter defibrillator (ICD) shock incidence at the end of life.

Author	Year of publication	Year of implantation	Year of death	Country	Study design	Grade [a]	Patient population (N)	Shock incidence [b]			Shock type
								30d	24h	1h	
Pratt et al.(11)	1996	[c]	1990-1994	USA	Observational Longitudinal Prospective	31	NSD (51)	-	24%	14%	-
							SD (17)	-	41%	29%	
							NCD (40)	-	20%	8%	
Grubman et al.(12)	1998	1989-1993	1989-1995	USA	Observational Cross-sectional Retrospective	32	NSD (55)	-	33%	7% [d]	appropriate
							SD (28)	-	68%	22% [d]	
							NCD (36)	-	-	3%	
Lehmann et al.(13)	1994	1982-1988	[c]	USA	Observational Cross-sectional Retrospective	27	SD (32)	-	-	66%	-
Mosteller et al.(14)	1991	1982-1988	1982-1988	USA	Observational Cross-sectional Retrospective	29	SD (5)	-	-	60%	-
Pires et al.(15)	1999	1990-1996	[c]	USA	Observational Cross-sectional Retrospective	33	SD (60)	-	-	62% [d]	-
Nielsen et al.(16)	1997	1989-1996	[c]	Denmark	Observational Longitudinal Prospective	28	SD (4)	-	-	50% [d]	appropriate
Gross et al.(17)	1991	1982-1990	1982-1990	USA	Observational Longitudinal Retrospective	31	SD (5)	-	-	40% [d]	-
Westerdahl et al.(18)	2015	1998-2010	2003-2010	Sweden	Observational Cross-sectional Prospective	34	All deaths (42)	-	24%	-	-
Sherazi et al.(19)	2013	1997-2001	1997-2001	USA	Observational Longitudinal	31	All deaths (83)	17-32%	3-19%	-	-

					Retrospective					
Westerdahl et al.(20)	2014	1998-2010	2003-2010	Sweden	Observational Cross-sectional Prospective	33	All deaths (97)	-	32%	31% [e]	13% inappropriate in last 24 hours
Lewis et al.(21)	2006	[c]	1994-2004	USA	Observational Longitudinal Retrospective	28	All deaths (43)	21%	-	-	-
Goldstein et al.(8)	2004	[c]	1997-2002	USA	Observational Cross-sectional Retrospective	31	All deaths (100)	27%	-	8% [d]	-
Poole et al.(22)	2008	1997-2001	[c]	USA/ Canada/ New Zealand	Observational Longitudinal Retrospective	29	All deaths (64)	-	31%	-	Appropriate

1h: Last hour before death; 24h: Last 24 hours before death; 30d: Last month before death; NSD: Nonsudden death; SD: Sudden death; NCD: Noncardiac death.

[a] Score classification on Quality Assessment Tool(10): 30-36=high quality; 24-29=moderate quality; <23=low quality

[b] Reported as time period before death.

[c] Unknown.

[d] Shocks were actually in the last minutes before death.

[e] *n*=87.

In six studies, the type of death was not specified.(8, 18-22) Year of publication ranged from 2004–2015 and all studies scored moderate to high on methodological quality. Three of these studies reported that 17–32% of patients experienced shocks in the last month of life.(8, 19, 21) Four studies showed that in the last 24 hours of life, 3–32% of patients experienced shocks.(18-20, 22) Two studies showed that in the last hour of life, 8% and 31% of patients experienced shocks.(8, 20) Two studies reported on the appropriateness of the shocks.(20, 22) In one study, all shocks were appropriate.(22) In the other study,(20) four of the 31 patients (13%) receiving shocks in the last 24 h were shocked inappropriately.

Impact of ICD shocks

A total of three studies reported on the impact of shocks in the last phase of life (Table 4). Date of publication ranged from 2004–2011. All studies were quantitative.(8, 23, 24)

Impact of shocks on patients

Two studies reported on the impact of shocks on patients in the last phase of life.(23, 24) In these studies, physicians and hospice administrators were surveyed. In the first study, physicians were asked whether they thought shocks are distressing to the patient. Seventy-six per cent agreed with this statement.(23) In the second study, hospice administrators reported that 74% of patients receiving shocks in the last phase of life were distressed by these shocks.(24)

Impact of shocks on relatives

Three studies reported on the impact of shocks on relatives.(8, 23, 24) In one study,8 100 next of kin were surveyed. The next of kin who witnessed the patient being shocked at the end of life reported that this was distressing to see. One relative reported in an interview that the patient experienced shocks every 20 minutes at the end of life, and reported it was like seeing the patient wake up from a 'really bad dream type of thing', after which the patient lost consciousness again. In a survey study, 76% of physicians agreed with the statement that shocks in patients at the end of their lives are distressing for the patients' loved ones.(23) A study in hospices reported that 92% of family members of patients receiving shocks found this distressing to witness. In one case, a patient experienced shocks during the dying process and, immediately after, the nurse had to wrap her body around the patient to stop the flailing of the body, which was distressing to witness.(24)

Table 4. Information on studies reporting on implantable cardioverter defibrillator (ICD) shock impact at the end of life.

Author	Year of publication	Country	Study design	Grade [a]	Respondents	Reporting impact on	Number of patients	Outcome
Kelley et al.(23)	2009	USA	Observational Cross-sectional Retrospective	33	Physicians	Patient and relatives (loved ones)	558 [b]	Seventy-six percent of physicians believed that shocks at the end of life are distressing for both patient and their loved ones
Fromme et al.(24)	2011	USA	Observational Cross-sectional Retrospective	32	Hospice administrator	Patient, relatives (family members) and professional caregivers	42 [c]	In 96% of cases, shocks were distressing to the patient and/or relatives. Also, present professional caregivers were distressed by shocks
Goldstein et al.(8)	2004	USA	Observational Cross-sectional Retrospective	31	Relatives	Relatives (next of kin)	100	Shocks were distressing to witness for the next of kin.

[a] Score classification on Quality Assessment Tool(10): 30-36= high quality; 24-29= moderate quality; <23= low quality.

[b] These are the number of physicians reporting on patients, not the actual number of patients.

[c] These are the numbers of hospices who participated in the study, not the actual number of patients.

Impact of shocks on professional caregivers

One survey study reported on the impact of shocks on professional caregivers.(24) Shocks were not only distressing for the hospice team to witness, but they must also deal with pain and panic induced by the shocks and loss of control in the patient and family. In the study, a situation was reported by a hospice administrator in which a patient was shocked multiple times during dying. The body was lifted off the bed due to the force of the shocks, which was disturbing for the nurse to witness.(24)

DISCUSSION

This review suggests that shocks in the last month of life are common in patients with active ICDs. An important finding of this review is that of patients dying non-suddenly, a quarter to a third experienced shocks in the last 24 hours of life. In patients dying suddenly, this was a third up to nearly 70%. The number of studies on the impact of shocks in the last month of life on patients, relatives and professional caregivers was limited, but they suggest that shocks are painful for the patient and distressing for patients, relatives and professional caregivers.

While the shock incidences found in this review are rather high, we found some variation between studies. There are several possible explanations for this variation. First, studies reporting on SD patients seem to report higher shock incidences than studies reporting on NSD patients.(11-17) This is because patients in these studies mostly died of sudden cardiac causes, predominantly due to VT(14) or VF,(16) resulting in the ICD delivering shocks. In patients dying non-suddenly, death is less often the direct result of a tachyarrhythmia, but rather of the underlying heart disease or bleeding, resulting in the ICD intervening less often. Second, the indication for ICD implantation has shifted over time. Seven studies reported on patients who had their ICD implanted before the year 2000. In this period, ICDs were mainly implanted in patients for secondary prevention. These patients have a higher risk of recurrent VTs than patients with ICDs for primary prevention, leading to higher shock incidences.20 From 2006 onwards, primary prevention was more often the indication for implantation than secondary prevention.(25) Only one study solely reported on patients with ICDs for primary prevention, showing that in the last 24 hours of life, 31% of patients experienced a shock.(22) This is still high, which might be due to the fact that in this particular study, patients with known sustained VT were excluded from the study, and only a single zone of therapy was used, so no antitachycardia pacing (ATP) was allowed. Third, in recent years, advances have been made to further optimise ICD programming to minimise inappropriate and appropriate therapy.(26) Developments such as ATP, longer detection times and high rate cut-offs all contribute to a reduction in shock therapy, by allowing more time for the arrhythmia to terminate spontaneously.(26, 27) These improvements in programming the ICD could mean that the current incidence of shocks might be lower than reported in the studies in this review. Fourth, studies we found were mainly concerned with patients dying of cardiac diseases, which might be an explanation for the high incidences as well. Only two studies specifically reported shock incidences on patients dying of noncardiac causes. Incidence in these populations seem to be lower than in the populations of NSD and SD patients.

Few studies were conducted on the impact of ICD shocks on patients in the last phase of life, their relatives and professional caregivers. From the literature on patients in earlier stages in their disease, it is known that shocks are painful and are associated with a diminished self-reported physical, emotional and social functioning, and symptoms of anxiety.(28) Although we found little detailed information on the impact of shocks in the last phase of life, this could also be the case in dying patients. In order to promote a peaceful death, quality of life should be pursued and the risk of developing symptoms of anxiety and depression should be reduced to a minimum. Timely deactivation of the ICD could help in promoting this peaceful death.

Communication between the professional caregiver and patient might be an important factor in preventing unwanted shocks at the end of life by deactivating the ICD. It has been shown that, when deactivation is discussed, a large proportion of patients decide to do so.(8) However, these conversations are rare.(29, 30) Professional caregivers often struggle with these conversations, because they feel they have insufficient knowledge about end of life care,(31) they feel uncomfortable discussing the topic,(32) or because they think talking about deactivation would take away hope from the patient.(33) A stronger collaboration with palliative care professionals might help professional caregivers to feel less uncomfortable initiating discussions about end of life.(34, 35) Also, policies can be developed for caregivers who do not frequently attend ICD patients, explaining the importance of discussing and deactivating the ICD, with specific opening questions to address the topic.(1, 36, 37)

More attention should be paid to inform the patient about the possibility of ICD deactivation, preferably starting before implantation of the device. Such discussions should be tailored to the patients' health literacy, communication style and personal values. Physicians and nurses should be trained to obtain the necessary skills to discuss delicate end-of-life issues, such as possible deactivation of ICDs.(38)

This study has some limitations. As with any review, it is possible that relevant studies were missed in conducting the search. In addition, publication bias is a possible limitation as well, leading to finding only articles which show notable results.7 Also, included studies had relatively small study populations, only three included more than 100 participants.(11, 12, 23) Further, seven of the 13 included studies on shock incidence were conducted 20 years or longer ago. Shock incidences might currently be lower than reported in this review, as described above. Finally, the majority of the studies were conducted in the USA, which might reduce the generalisability to a European population.

Future research on ICD management in the last phase of life should focus on determining shock incidences in both patients dying of noncardiac causes and patients dying with ICDs implanted for primary prevention, since these are not well known. Also, few studies are conducted on the impact of shocks at the end of life on patients, relatives and professional caregivers. This is a topic to be further examined. Also, it should be further examined what can be done to promote discussions before implantation and in early stages of the disease.

CONCLUSIONS

This is the first integrated review on the incidence and impact of ICD shocks in the last phase of life, providing a thorough overview of all the available evidence on these topics. Shocks were found to occur commonly, both in patients dying non-suddenly and patients dying suddenly. Shocks are painful and distressing for patients, but also distressing to witness for relatives and professional caregivers. The evidence summarised in this review should raise awareness among healthcare professionals of the negative consequences of having an active ICD at the end of life. Our findings emphasise that it is important for healthcare professionals to discuss ICD deactivation with the patient in an early stage of the disease. This can contribute to a timely deactivation of the ICD, and therefore can minimise possible suffering due to shocks and help promote a calm and peaceful death.

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SUPPLEMENTARY MATERIAL

Supplementary Appendix 1. Detailed search queries for all databases.

I. Embase

('defibrillator'/de OR 'defibrillator pacemaker'/de OR 'implantable cardioverter defibrillator'/de OR 'internal defibrillator'/de OR (((implant* OR intern* OR pacemaker* OR therap*) NEAR/4 defibrillator*) OR ICD OR AICD):ab,ti) AND ('convulsive therapy'/de OR 'electric shock'/de OR 'cardioversion'/de OR (shock OR shocks OR electroshock* OR countershock* OR cardioversion* OR cardioconversion* OR electroversion* OR electroconversion*):ab,ti) AND ('terminal care'/exp OR 'terminally ill patient'/exp OR (terminal* OR ((end OR last) NEAR/4 (life OR living OR stage)) OR EOL OR death* OR dying* OR palliati* OR hospice* OR (last NEXT/1 (phase OR year* OR month* OR week* OR day*))):ab,ti) AND English:la NOT ('conference abstract'/it)

II. Medline

("Defibrillators, Implantable"/ OR (((implant* OR intern* OR pacemaker* OR therap*) ADJ4 defibrillator*) OR ICD OR AICD).ab,ti.) AND ("Electric Countershock"/ OR (shock OR shocks OR electroshock* OR countershock* OR cardioversion* OR cardioconversion* OR electroversion* OR electroconversion*).ab,ti.) AND (exp "Terminal Care"/ OR "Terminally Ill"/ OR (terminal* OR ((end OR last) ADJ4 (life OR living OR stage)) OR EOL OR death* OR dying* OR palliati* OR hospice* OR (last ADJ1 (phase OR year* OR month* OR week* OR day*))).ab,ti.) AND English.lg. NOT (abstract).pt.

III. Cochrane Central

((((implant* OR intern* OR pacemaker* OR therap*) NEAR/4 defibrillator*) OR ICD OR AICD):ab,ti) AND ((shock OR shocks OR electroshock* OR countershock* OR cardioversion* OR cardioconversion* OR electroversion* OR electroconversion*):ab,ti) AND ((terminal* OR ((end OR last) NEAR/4 (life OR living OR stage)) OR EOL OR death* OR dying* OR palliati* OR hospice* OR (last NEXT/1 (phase OR year* OR month* OR week* OR day*))):ab,ti)

IV. Web of Science

TS=(((((implant* OR intern* OR pacemaker* OR therap*) NEAR/4 defibrillator*) OR ICD OR AICD)) AND ((shock OR shocks OR electroshock* OR countershock* OR cardioversion* OR cardioconversion* OR electroversion* OR electroconversion*)) AND ((terminal* OR ((end OR last) NEAR/4 (life OR living OR stage)) OR EOL OR death* OR dying* OR palliati* OR hospice* OR (last NEAR/1 (phase OR year* OR month* OR week* OR day*))))) AND LA=English AND DT=Article

V. Google Scholar

"implantable|internal|pacemaker defibrillator"|ICD|AICD

shock|shocks|electroshock|countershock|cardioversion|electroversion|electroconversion

terminal|"end|last life|living|stage"|EOL|death|dying|palliative|hospice|"last phase|year|month|week|days"

6

CHAPTER 6

Trends in time in the management of the implantable cardioverter defibrillator in the last phase of life: a retrospective study of medical records

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ABSTRACT

Background

The implantable cardioverter defibrillator (ICD) might give unwanted shocks in the last month of life. Guidelines recommend deactivation of the ICD prior to death.

Aims

The aims of this study were to examine trends in time (2007–2016) in how and when decisions are made about ICD deactivation, and to examine patient- and disease-related factors which may have influenced these decisions. In addition, care and ICD shock frequency in the last month of life of ICD patients are described.

Methods

Medical records of a sample of deceased patients who had their ICD implanted in 1999–2015 in a Dutch university (*n* = 308) or general (*n* = 72) hospital were examined.

Results

Median age at death was 71 years, and 88% were male. ICD deactivation discussions increased from 6% for patients who had died between 2007 and 2009 to 35% for patients who had died between 2013 and 2016. ICD deactivation rates increased in these periods from 16% to 42%. Presence of do-not-resuscitate (DNR) orders increased from 9% to 46%. Palliative care consultations increased from 0% to 9%. When the ICD remained active, shocks were reported for 7% of patients in the last month of life. Predictors of ICD deactivation were the occurrence of ICD deactivation discussions after implantation (OR 69.30, CI 26.45–181.59), DNR order (OR 6.83, CI 4.19–11.12), do-not-intubate order (OR 6.41, CI 3.75–10.96), and palliative care consultations (OR 8.67, CI 2.76–27.21)

Conclusion

ICD deactivation discussions and deactivation rates have increased since 2007. Nevertheless, ICDs remain active in the majority of patients at the end of life, some of whom experience shocks.

INTRODUCTION

The implantable cardioverter defibrillator (ICD) is effective in terminating life-threatening cardiac arrhythmias by giving electric shocks(1). When death is imminent due to irreversible terminal illness, shocks might be unwanted and can be a factor of distress to the patient and his or her family that disturbs the dying process(2).

A recent integrated review using a systematic approach shows that ICD shocks occur rather frequently on the last day of life, ranging from 24% to 33% in patients who die nonsuddenly up to 41-68% in patients who die suddenly(2). However, 7 of 13 studies in this review were conducted two decades or more ago, and studies were mainly based on patients with a secondary prevention indication, who are more prone to experiencing shocks due to their cardiac history(3).

Important developments in ICD management have taken place. First, international expert consensus statements on the management of the ICD in the last phase of life have been published(1, 4-6). In these statements it is discussed that shocks in the last phase of life may be physically and psychologically stressful, and it is recommended to deactivate the ICD at the end of a patient's life(1). It is also recommended that professional caregivers have early and recurring discussions on the possibility and desirability of deactivating the ICD. These statements might have established an increase in ICD deactivation discussions and actual deactivation. Second, ICD programming has been further optimized. A longer detection time and a higher rate cutoff have shown good results in reducing the number of inappropriate shocks(7, 8). This might have established a decrease in patients receiving shocks at the end of life.

The aims of this study were to:

1. Examine the decision-making process concerning ICD deactivation and to examine patient- and disease- related factors influencing the decision to deactivate.
2. Describe the care and ICD shock frequency in the last month of life of ICD patients, in particular the presence of do-not-resuscitate (DNR) and do-not-intubate (DNI) orders and the involvement of palliative care teams.

For both questions we will describe the trends in time (periods 2007-2009, 2010-2012 and 2013-2016).

METHODS

Patients and setting

Patients older than 18 years of age, who had an ICD implanted in a large Dutch university hospital or in a general hospital and who died between 2007 and 2016 were eligible for inclusion in the study. In the university hospital, a total of 544 patients with an ICD had died in the specified time period. These patients had their first ICD implanted between 1999 and 2015. Medical records of 25 deceased patients per year of implantation were randomly selected to be assessed for this study in order to obtain a representative sample that covered all years of implantation. If there were less than 25 deceased patients for an implantation year, all patients were included. A total of 308 patients in the university hospital were eventually included. In the general hospital, 72 patients had died. These patients had their ICD implanted between 2002 and 2015 and all were included.

Data collection

Data were collected by AB, RS and AD from three sources:

1. ICD registries in both hospitals contained clinical information, demographics, and ICD-related information such as indication for implantation, categorized as primary (implantation in patients with elevated risk of arrhythmias or cardiac death) or secondary prevention (implantation in patients who already experienced life-threatening arrhythmias or aborted cardiac death)(3), implantation date, type of ICD and occurrence of shocks in the last month of life, categorized as appropriate (shock for ventricular fibrillation or ventricular tachycardia) or inappropriate (any other shock).
2. From the electronic and paper medical records we extracted the following data: the number and timing of ICD deactivation discussions, the presence DNR and DNI orders, consultations with palliative care teams (both hospitals had such a team), reasons for consultation, occurrence and characteristics of ICD deactivation, and occurrence and timing of shocks. A checklist was developed and pilot tested in 10 patients by RS and AB.
3. If a patient had died outside hospital, we contacted the patient's general practitioner by telephone to collect data on the cause and place of death, whether or not the ICD was deactivated preceding death, and the occurrence of shocks in the last month of life.

Cause of death was classified by RS and DT in four categories, using and adapted version of the classification schemes of Hinkle and Thaler and Epstein et al.(9, 10): s*udden cardiac death* – cardiac death that occurs within 1 h after the onset of new symptoms; n*onsudden cardiac death* – cardiac death that occurs more than 1 h after the onset of new symptoms; *Noncardiac death* – death from other causes; and *unknown*. Our study conforms with the principles outlined in the *Declaration of Helsinki*(11). Approval of the local ethical committee (METC-2016-453) was obtained before starting the data collection.

Data analysis

Descriptive statistics were used to summarize demographic data. Mean scores and standard deviations were calculated for normally distributed data and medians and interquartile ranges (IQRs) for non-normally distributed data. Differences in medians between patients dying during 2007-2009, 2010-2012, and 2013-2016 were examined with Kruskal Wallis-tests. Categorical variables were compared with the Pearson Chi-square test, using a Bonferroni correction when appropriate. Differences between the two hospitals participating in this study were explored with Pearson Chi-square tests. Logistic regression analyses were conducted for the variables that were expected to possibly predict deactivation of the ICD(12): age, gender, New York Heart Association (NYHA) classification, myocardial infarction, diabetes mellitus, chronic kidney disease, left ventricular ejection fraction (LVEF), indication for ICD implantation (primary vs. secondary prevention), occurrence of ICD discussion (no discussion vs. discussion before implantation vs. discussion after implantation), occurrence of palliative care team consultation (yes vs. no), presence of DNR and DNI order (yes vs. no), the occurrence of shocks (yes vs. no), and the year of death (divided in periods 2007-2009, 2010-2012, 2013-2016). A *p*-value of <0.05 was used to indicate statistical significance. Analyses were performed using SPSS version 24.

RESULTS

Demographics

Of the 380 patients examined in this study, 333 (88%) were male. Of patients, 110 had their first ICD implanted before 2005 (24 (22%) for primary prevention), 137 patients between 2005 and 2010 (95 (69%) for primary prevention), and 133 patients after 2010 (99 (74%) for primary prevention). There was an increase in ICDs being implanted for primary prevention ($p < 0.01$) Patients had their first ICD implanted at a median age of 67 (IQR 58-73) years. After 2010, ICDs were implanted more frequently in older patients (median age at implantation 70 years) as compared to before 2010 (median age at implantation 65 years) ($p < 0.01$). Also, more ICDs were implanted for primary prevention in the general hospital (78%) compared to the university hospital (53%) ($p < 0.01$).

Patients died at a median age of 71 (IQR 63-78), due to nonsudden cardiac causes (n=122, 32%), sudden cardiac causes (n = 47, 12%), or noncardiac causes (n = 121, 32%) (Table 1). For 90 (24%)

patients, cause of death was unknown. A total of 183 (48%) patients died in the hospital, 85 (22%) at home, and 30 (8%) in a facility (including nursing home and inpatient hospice).

Table 1. Characteristics of ICD patients.

	2007-2009 (*n* = 96)	2010-2012 (*n* = 108)	2013-2016 (*n* = 176)	All (*n* = 380)	*p*-value[a]
Gender (Male)	89 (93%)	90 (83%)	154 (88%)	333 (88%)	0.13
Age at ICD implantation (median, IQR)	67 (58-73)	65 (57-72)	68 (59-74)	67 (58 – 73)	0.14
Implantation for primary prevention	38 (40%)	57 (53%)	123 (70%)	218 (57%)	<0.01
Type of ICD at first implant					0.35[b]
Single chamber	31 (32%)	30 (28%)	41 (23%)	102 (27%)	
Dual chamber	37 (39%)	39 (36%)	61 (35%)	137 (36%)	
CRT-D	28 (29%)	36 (33%)	69 (39%)	133 (35%)	
Subcutaneous ICD	0	1 (1%)	4 (2%)	5 (1%)	
Unknown	0	2 (2%)	1 (1%)	3 (1%)	
Type of ICD at death					0.26 [b.]
Single chamber	27 (28%)	31 (29%)	35 (20%)	93 (25%)	
Dual chamber	37 (39%)	32 (30%)	54 (31%)	123 (32%)	
CRT-D	31 (32%)	41 (38%)	81 (46%)	153 (40%)	
Subcutaneous ICD	0	1 (1%)	4 (2%)	5 (1%)	
ICD extracted	1 (1%)	1 (1%)	2 (1%)	4 (1%)	
Unknown	0	2 (2%)	0	2 (1%)	
NYHA					0.53[b]
Class I	6 (6%)	8 (7%)	21 (12%)	35 (9%)	
Class II	59 (61%)	62 (57%)	88 (50%)	209 (55%)	
Class III	27 (28%)	32 (30%)	44 (25%)	103 (27%)	
Class IV	0	1 (1%)	1 (1%)	2 (1%)	
Unknown	4 (4%)	5 (5%)	22 (13%)	31 (8%)	
Comorbidities					
Myocardial infarction	65 (68%)	72 (67%)	105 (60%)	242 (64%)	0.31
Diabetes Mellitus	21 (22%)	28 (26%)	61 (35%)	110 (29%)	0.06
Chronic kidney disease	39 (41%)	48 (44%)	79 (45%)	166 (44%)	0.78
LVEF (≤ 30)	54 (56%)	57 (53%)	94 (53%)	205 (53%)	0.64
Age at death (median, IQR)	70 (62-76)	70 (61-78)	72 (64-79)	71 (63-78)	0.19
Classification of death					0.82[b]
Nonsudden cardiac	28 (29%)	30 (28%)	64 (36%)	122 (32%)	
Sudden cardiac	11 (12%)	15 (14%)	21 (12%)	47 (12%)	
Noncardiac	24 (25%)	34 (32%)	63 (36%)	121 (32%)	
Unknown	33 (34%)	29 (27%)	28 (16%)	90 (24%)	
Place of death					0.47[b]
Home	16 (17%)	22 (20%)	47 (27%)	85 (22%)	
Hospital	44 (46%)	54 (50%)	85 (48%)	183 (48%)	
Nursing home	2 (2%)	9 (8%)	15 (9%)	26 (7%)	
Hospice care	0	1 (1%)	3 (2%)	4 (1%)	
Other[c]	2 (2%)	3 (3%)	2 (1%)	7 (2%)	
Unknown	32 (33%)	19 (18%)	24 (14%)	75 (20%)	

Note: Percentages may not add up to 100% due to rounding.

IQR: Interquartile range; LVEF: Left ventricular ejection fraction; NYHA: New York Heart Association; CRT-D: Cardiac Resynchronization Therapy – Defibrillator; S-ICD: Subcutaneous ICD.
[a] *p*-values calculated with Pearson Chi-Square test.
[b] The "unknown" category was not used in calculating the statistical significance of differences between groups.
[c] Other places of death were: in the ambulance on the way to hospital ($n = 2$), in a rehabilitation center ($n = 2$), outside ($n = 2$) and in a care hotel ($n = 1$).

Decision-making about ICD deactivation

Discussions on ICD deactivation

In 81 (21%) medical records, discussions between a professional caregiver and the patient on ICD deactivation were reported (Table 2). Sixty (16%) patients had one discussion with a professional caregiver, and 21 (6%) patients had more than one discussion (range 2-7). Twenty (5%) patients had the discussion as part of a "pre-implantation conversation" with an ICD nurse, all of which were held in the university hospital. For 13 of these patients, this was the only occurrence that a discussion on ICD deactivation was reported in the medical record. Sixty-eight (18%) patients had a discussion after ICD implantation, for 47 patients this was the only time deactivation was discussed. Seven (2%) patients had discussions both before and after implantation. A total of 37 (10%) discussions were held on the day of deactivation. For 25 (7%) patients, this was the only time deactivation was discussed. Fifteen (19%) patients who had a discussion about ICD deactivation, did not have their ICD deactivated. In patients who died between 2007-2009, discussions on ICD deactivation were reported in 6 (6%) medical records, compared to 13 (12%) for 2010-2012, and 62 (35%) for 2013-2016 ($p < 0.01$).

Table 2. Characteristics of ICD deactivation discussions.

	2007-2009 ($n = 96$)	2010-2012 ($n = 108$)	2013-2016 ($n = 176$)	All deaths ($n = 380$)	*p*-value[a]
Discussions about ICD deactivation	6 (6%)	13 (12%)	62 (35%) [b.]	81 (21%)	<0.01
Before ICD implantation	0	0	20 (11%)	20 (5%)	<0.01
After ICD implantation	6 (6%)	13 (12%)	49 (28%)	68 (18%)	<0.01
Number of days between discussion and ICD deactivation (median, IQR)[c]	0 (0-1)	0 (0-3)	0 (0-1)	0 (0-1)	0.78[d]
Number of days between discussion and death (median, IQR)[c]	6 (3-32)	5 (1-72)	9 (2-66)	8 (2-55)	0.80[d]

Note. IQR: Interquartile range.
[a] *p*-values calculated with Pearson Chi-Square test.
[b] Some patients had discussions both before and after ICD implantation.
[c] Only patients with discussions after ICD implantation ($n = 68$) were included in calculating medians.
[d] Difference between medians calculated with Kruskal-Wallis test.

Characteristics of ICD deactivation

In 112 (30%) medical records, it was reported that the ICD was deactivated before death (Table 3). Sixty-six (59%) of these patients had a previous discussion about deactivation. ICD deactivation was performed through reprogramming the device for 70 (63%) patients, through placing a magnet for 15 (13%) patients, and 4 (4%) devices were completely removed due to infections. For 23 (21%) patients, it was unknown how the ICD was deactivated. The ICD was mostly deactivated in the hospital in 92 (82%) patients. Four patients had their ICD deactivated at home, three in a nursing home, one in an ambulance and one in a revalidation center. ICDs were mostly deactivated by ICD technicians (69 patients, 62%) and cardiologists (10 patients, 9%). Deactivation was more rarely conducted by emergency physicians (*n* = 2), internists (*n* = 1), anesthesiologists (*n* = 1), oncologist (*n* = 1), and ambulance staff (*n* = 1). Patients' median age at deactivation was 72 (IQR 65 - 78) years. Patients died a median of 3 (IQR 0 – 18) days after deactivation. In 15 (16%) patients who died between 2007-2009 the ICD was deactivated, as compared to 24 (22%) deactivations in patients who died between 2010-2012 and 73 (42%) who died between 2013-2016 ($p < 0.01$).

Table 3. Characteristics of ICD deactivation.

	2007-2009 (*n* = 96)	2010-2012 (*n* = 108)	2013-2016 (*n* = 176)	All years (*n* = 380)	*p*-value[a]
Total number of deactivations	15 (16%)	24 (22%)	73 (42%)	112 (30%)	<0.01
Type of deactivation					0.39[b]
Reprogramming	11 (73%)	13 (54%)	46 (63%)	70 (63%)	
Magnet	0	5 (21%)	10 (14%)	15 (13%)	
Extraction	1 (7%)	1 (4%)	2 (3%)	4 (4%)	
Unknown	3 (20%)	5 (21%)	15 (21%)	23 (21%)	
Location of deactivation					0.75[b]
Hospital	12 (80%)	17 (71%)	63 (86%)	92 (82%)	
Home	0	0	4 (5%)	4 (4%)	
Nursing home	0	1 (4%)	2 (3%)	3 (3%)	
Other[c]	0	0	2 (3%)	2 (2%)	
Unknown	3 (20%)	6 (25%)	2 (3%)	11 (10%)	
ICD deactivated by					0.80[b]
Cardiologist	1 (7%)	3 (13%)	6 (8%)	10 (9%)	
ICD technician	7 (47%)	12 (50%)	50 (68%)	69 (62%)	
Other[d]	0	1 (4%)	5 (7%)	6 (5%)	
Unknown	7 (47%)	8 (33%)	12 (16%)	27 (24%)	
Age at deactivation (median, IQR)	62 (57-71)	75 (68-80)	73 (66-78)	72 (65-78)	0.02[e]
Number of days from deactivation to death (median, IQR)	3 (0-23)	2 (0-24)	4 (1-17)	3 (0-18)	0.89[e]

Note. Percentages may not add up to 100% due to rounding.
IQR: Interquartile range.
[a] *p*-values calculated with Pearson Chi-Square test.

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[b] The "unknown" category was not used in calculating the statistical significance of differences between groups.
[c] Other locations of deactivation were: in the ambulance on the way to the hospital (n = 1), in a revalidation center (n = 1).
[d] Other persons who deactivated the ICD were: physician other than cardiologist (n = 5), ambulance staff member (n = 1).
[e] Difference between medians calculated with Kruskal-Wallis test.

Care and shocks in the last month of life

DNR- and DNI-orders and palliative care teams

In 120 (32%) medical records a DNR-order was documented (Table 4); this concerned 9 (9%) patients who died between 2007-2009, 30 (28%) patients who died between 2010-2012, and 81 (46%) patients who died between 2013-2016 ($p < 0.01$). In 78 (21%) medical records a DNI-order was documented; this concerned 5 (5%) patients who died between 2007-2009, 25 (23%) patients who died between 2010-2012, and 48 (27%) patients who died between 2013-2016 ($p < 0.01$). Palliative care team consultations were documented in 17 (5%) medical records, all consultations occurred after 2010. Teams were consulted about medication management or the possible use of palliative sedation. In one case the team was consulted because of a euthanasia request.

Occurrence of shocks

A total of 268 (71%) patients died with an active ICD. In 114 (43%) of these medical records, one or more shocks were reported (Table 4). Of the patients who experienced one or more shocks, 89% experienced an appropriate shock, and 24% an inappropriate shock. In 20 (7%) medical records, shocks were reported in the last month of life (90% appropriate and 15% inappropriate) and in 5 (2%) on the last day of life (100% appropriate and in 20% inappropriate). Occurrence of shocks in the last month and on the last day of life did not significantly differ in patients dying between 2007-2009, 2010-2012 or 2013-2016. Of the patients who died with an active ICD, 71 (26%) deaths were classified as nonsudden cardiac, 39 (15%) as sudden cardiac, and 76 (28%) as noncardiac. For 82 (31%) patients, cause of death was unknown. In patients whose deaths were classified as sudden cardiac, shocks in the last month of life were significantly more often reported (28% shocks) as compared to patients whose deaths were described as nonsudden cardiac (4% shocks), noncardiac (3% shocks) or unknown (5% shocks) ($p < 0.01$).

Table 4. Presence of DNR and DNI orders, palliative team consultations and shock incidence.

	2007-2009 (n = 96)	2010-2012 (n = 108)	2013-2016 (n = 176)	All deaths (n = 380)	p-value[a]
Presence of DNR order	9 (9%)	30 (28%)	81 (46%)	120 (32%)	<0.01
Presence of DNI order	5 (5%)	25 (23%)	48 (27%)	78 (21%)	<0.01

Consultation with palliative care team	0	2 (2%)	15 (9%)	17 (5%)	<0.01
Shocks 30 days before death[b]	5 (6%)	6 (7%)	9 (9%)	20 (7%)	0.80
Shocks 24 h before death[b]	0	2 (2%)	3 (3%)	5 (2%)	0.32
All shocks[b]	45 (56%)	31 (37%)	38 (37%)	114 (43%)	0.02

Note. DNR: do-not-resuscitate; DNI: do-not-intubate.

[a] *p*-values calculated with Pearson Chi-square test.

[b] Only patients with an active ICD at time of dying were included in these calculations: between 2007-2009, *n* = 81; between 2010-2012, *n* = 84; between 2013-2016, *n* = 103.

Predictors of ICD deactivation

A total of 14 factors were examined to determine their relationship with ICD deactivation (Table 5). Significant predictors of ICD deactivation were the occurrence of ICD deactivation discussions after implantation (odds ratio (OR) 69.30, confidence interval (CI) 26.45-181.59), the presence of a DNR (OR 6.83, CI 4.19-11.12) or DNI order (OR 6.41, CI 3.75-10.96), the occurrence of a palliative care team consultation (OR 8.67, CI 2.76-27.21), and year of death (OR 2.05, CI 1.51-2.78).

Table 5. Clinical and nonclinical predictors of ICD deactivation[a]

Predictor Variables	OR	95% CI	*p*-value
Age	1	0.98-1.02	0.94
Gender	0.70	0.37-1.34	0.28
NYHA	1.05	0.72-1.54	0.79
Myocardial infarction	0.98	0.62-1.55	0.94
Diabetes Mellitus	0.92	0.56-1.50	0.72
Chronic kidney disease	1	0.64-1.57	0.99
LVEF (≤ 30)	1.33	0.85-2.08	0.21
Indication (primary prevention)	1.38	0.88-2.18	0.16
ICD discussions			<0.01
Before implantation	1.65	0.44-6.23	0.46
After implantation	69.3	26.45-181.59	<0.01
DNR-order	6.83	4.19-11.12	<0.01
DNI-order	6.41	3.75-10.96	<0.01
Palliative care team consultation	8.67	2.76-27.21	<0.01
Shocks	0.81	0.52-1.28	0.36
Year of death	2.05	1.51-2.78	<0.01

Note. OR: Odds Ratio; 95% CI: 95 percent confidence interval; NYHA: New York Heart Association; LVEF: Left ventricular ejection fraction; DNR: Do-not-resuscitate; DNI: Do-not-intubate.

[a] Univariable logistic regression analyses.

DISCUSSION

Decision-making about ICD deactivation

In this study, discussions about ICD deactivation between professional caregivers and the patient were reported in 21% of the patients. Most of these patients had only one discussion reported in their record, occurring after implantation of the device. There was however a steep increase in discussions over the years, from 6% for patients dying between 2007-2009 up to 35% for patients dying between 2013-2016. Discussing ICD deactivation was significantly associated with ICD deactivation, which was reported in 30% of the medical records overall, also with an increase over the studied years from 16% up to 42%. The increase in deactivation discussions and actual deactivation might be attributable to the increased attention to ICD management in the last phase of life, such as the development of expert consensus statements(1, 4-6), and more general to an increased attention to advance care planning(13). The percentages of deactivation discussion and deactivation are in line with a retrospective study conducted in the UK in patients (*n* = 44) who died in 2012 and 2013, showing that 39% of patients had documentation of a discussion about ICD deactivation after implantation(12). A recent prospective American study in which 51 ICD patients were prospectively followed showed that 26% of the patients discussed the possibility of deactivation with their cardiologist(14). Two Swedish studies published in 2014 and 2015 showed that 30-49% of patients had their ICD deactivated(3, 15).

Despite an increase in ICD deactivation discussions and actual deactivations, current rates are still rather low. Several factors may explain this. It might be possible that actual deactivation discussions were not reported in the hospital medical records(16), or that these discussions were described in another medical file by another professional caregiver (such as the general practitioner). It is however of utmost importance that conversations about this topic are reported in the medical record, so that other professional caregivers are aware of the knowledge level and preferences of the patient. Secondly, while studies show that many professional caregivers think that the possibility of future deactivation should be discussed in an early stage of the disease(17), other studies report on barriers on actually engaging in such discussions(17). Professional caregivers are often unsure about the right time to discuss deactivation(18), sometimes feel like they do not have a well enough established relationship with the patient(19), have too little time to accurately discuss deactivation(18), find it difficult to predict the end of life(20) or feel that talking about deactivation might be inappropriate, since the ICD was implanted to ‘safe lives’(19). Also, a recent international survey study from the UK, conducted in 262 professionals caregivers, shows that professional caregivers who are physicians or American, and had initiated a deactivation discussion before, were more likely to discuss ICD deactivation(21). This study also identifies that, even though only 30% of nurses were involved in ICD deactivation discussion, 81% of professional caregivers felt that nurses do have the necessary skills to start these conversations, and might therefore play a bigger role in informing patients about ICD deactivation. Further, the study identified that nurses were in favor of informing the patients about ICD deactivation before implantation, which is in line with current guidelines, which advise to discuss ICD deactivation early in the disease process, preferably before implantation, to make the patients aware of the consequences of having an active ICD at the end of life(1, 4, 6).

Although the patient's willingness to discuss ICD deactivation varies, previous studies showed that patients are sometimes hesitant to discuss ICD deactivation, or might even be unaware about the option of deactivation at all(22). Some patients may misunderstand the role and function of the ICD(17), the distinction between the bradycardia- and tachycardia therapy, and are not always sure why the ICD might intervene(23). Patients sometimes consider deactivation to be similar to the active ending of life(23). The active ending of life, or euthanasia, is however defined as medication administered by a physician, with the explicit intention of hastening death at the explicit request of the patient. Euthanasia is legal in the Netherlands, under strict criteria stipulated by the Dutch euthanasia law, and only allowed for patients who are suffering unbearably with no prospect on relief(24). Deactivating the ICD is fundamentally different from euthanasia, since no medication is administered to hasten death. Also, deactivating the ICD will not cause imminent death, contrary to what patients sometimes believe(25). It is important to inform patients about ICD deactivation adjusted to their willingness to engage in these conversations, their knowledge level, and stage of disease(13).

Care and shocks in the last month of life

In our study, DNR orders were present in 32% of medical records of deceased ICD patients, which is a significant increase since 2007, in which only 9% of patients had a DNR order. Discussing a DNR order has been suggested to be a good opportunity to also discuss possible ICD deactivation(1, 4, 6). Nevertheless, while in our study DNR orders were associated with ICD deactivation, almost half of the patients with a DNR order had an active ICD at time of death. A full advance care planning process including discussions between patients with an ICD, their next of kin and professional caregivers has been suggested to be a be more effective than merely the completion of a DNR order(13, 26).

Palliative care teams were rarely consulted (5%), possibly because professional caregivers are unsure about the possible contribution of these teams to patient care(27). We did however see, that palliative care team consultations was associated with ICD deactivation. There is an unmet need for palliative care in patients with heart failure(28). Not all cardiologists consider end-of-life care to be part of their responsibility(29). Yet, palliative care teams can help to clarify goals of care of ICD patients, and might contribute to patient-centered end-of-life care(30).

In patients in whom the ICD was not deactivated, shocks in the last month of life were reported in 20 (7%) patients and shocks in the last 24 hours were reported in 5 (2%) patients. Previous studies showed a higher incidence of shocks, namely up to 32% in the last month of life and up to 68% in the last 24 hours of life(2). This might be due to different factors, such as the high proportion of patients who have their ICD implanted for primary prevention in our study, which is the main indication for ICD implantation since 2006(31). Previous studies that reported on shock incidence are generally older and have high proportions of secondary prevention patients(3, 15). Furthermore, developments in optimizing ICD programming have resulted in reducing the number of inappropriate shocks at the end of life(7, 8).

Strengths and limitations

To our knowledge, this is the first study that examines the trends in time of ICD deactivation discussions, ICD deactivation and ICD shocks and that identifies factors which possibly predict ICD deactivation. Also, this is the largest study to examine shocks in the last month of life. This study does however have some limitations. This study was a retrospective study which relied on the medical records of patients. These medical records could be incomplete(16), which could have led to underreporting of discussions regarding ICD deactivation. Also, we did not have insight in the exact content of the deactivation discussions.

Recommendations and future research

Guidelines recommend that discussions on ICD deactivation occur early and on set times during the disease trajectory(1, 4-6). Educating patients about the consequences of an active ICD in the last phase of life is an important task for the health care professional and has been proven to increase device deactivation(23, 32). These discussions should include an extensive exploration of the patient’s personal values and future goals of care, and should frequently be reassessed, since patients might be subject to changing preferences(13). Professional caregivers might be supported in conducting these conversations by palliative care teams, and practical decision aids, such as checklists(30). Documentation of these discussions is crucial, and decisions should be communicated with all involved professional caregivers to avoid misunderstanding and enable good decision-making(5). Future research might focus on barriers and facilitators of professional caregivers and patients to discuss ICD deactivation.

CONCLUSIONS

The occurrence of ICD deactivation discussions, both before and after implantation, the number of ICD deactivations, the presence of DNR and DNI orders and the number of palliative care team consultations have increased since 2007. However, ICDs still remain active in the majority of patients at the end of life, who as a result may experience shocks. This study underlines the importance of timely discussions between professional caregivers and patients.

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CHAPTER 7

Implantable cardioverter defibrillator deactivation and advance care planning: a focus group study

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ABSTRACT

Objective

Implantable cardioverter defibrillators can treat life threatening arrhythmias, but may negatively influence the last phase of life if not deactivated. Advance care planning conversations can prepare patients for future decision-making about implantable cardioverter defibrillator deactivation. This study aimed at gaining insight in the experiences of patients with advance care planning conversations about implantable cardioverter defibrillator deactivation.

Methods

In this qualitative study, we held five focus groups with 41 patients in total. Focus groups were audio-recorded and transcribed. Transcripts were analyzed thematically, using the constant comparative method, whereby themes emerging from the data are compared with previously emerged themes.

Results

Most patients could imagine deciding to have their implantable cardioverter defibrillator deactivated, for instance because the benefits of an active device no longer outweigh the harm of unwanted shocks, when having another life-limiting illness, or when relatives would think this would be in their best interest. Some patients expressed a need for advance care planning conversations with a healthcare professional about deactivation, but few had had these. Others did not, saying they solely focused on living. Some patients were hesitant to record their preferences about deactivation in advance care directives, because they were unsure whether their current preferences would reflect future preferences.

Conclusions

Although patients expressed a need for more information, advance care planning conversations about implantable cardioverter defibrillator deactivation seemed to be uncommon. Deactivation should be more frequently addressed by healthcare professionals, tailored to the disease stage of the patient and readiness to discuss this topic.

INTRODUCTION

The Implantable Cardioverter Defibrillator (ICD) treats potentially lethal arrhythmias by either antitachycardia pacing or delivering an electrical shock(1). About half of ICD patients experience potentially painful shocks during their life(2). The number of ICD patients has grown, as the indication for implantation has been extended from secondary to primary prevention(3). While the ICD is effective in treating arrhythmias, patients eventually die due to deterioration of their underlying heart disease or another illness(4). An active ICD delivers shocks in the last 24 hours of life in up to 33% of patients dying nonsuddenly(5). Shocks are potentially painful and a source of distress and anxiety for patients and relatives(5, 6). Shocks can be avoided by timely deactivating the shock function of the ICD(7).

International expert consensus statements from the European Heart Rhythm Association and Heart Rhythm Society recommend to timely and repeatedly discuss ICD deactivation with the patient and relatives(1, 8). This is in line with the international advance care planning (ACP) consensus statement(9). ACP enables individuals to define and discuss goals and preferences for future medical treatment and care, and to record and review these if appropriate(9). Previous studies have shown that only a minority of patients (27% in a study from 2004(10), up to 35% in a study from 2018(2)) had discussed ICD deactivation with their healthcare professional and had their ICD deactivated prior to death(2, 11). It is unknown why ICD deactivation is infrequently discussed, and what patients would want with their device when approaching the end-of-life. It is known that many patients are confused about the role of the ICD, especially in the last stages of life(12, 13).

Insight into the experiences of ICD patients with ACP conversations about ICD deactivation is limited. Having more insight into these experiences and on how ICD patients reflect on the end-of-life might help to increase the understanding of how patients think, and how they could best be approached in having an ACP conversation. Therefore, we examined ICD patients' experiences with ACP conversations about ICD deactivation in a qualitative focus group study.

METHODS

Study design and sample

We conducted focus groups with ICD patients, recruited via the Dutch national ICD recipient association ("STIN"). A call for participants was published in their magazine, on their website and on social media channels. Patients were eligible when they had an active ICD, were older than 40 years, able to speak and understand Dutch and to provide written consent. Patients who were interested in participating in the study received information via e-mail, accompanied with an informed consent form. Focus groups were organized in the Erasmus University Medical Center Rotterdam, the Netherlands. Patients were compensated for their time with a gift card of €25, and received reimbursement for travel expenses.

Forty-one patients agreed to participate in the study. Five focus groups were organized with respectively 9, 8, 9, 9, and 6 patients. Approval of the study was obtained from the local research ethics committee (METC-2017-357).

Data collection

Focus groups were conducted in September 2017, and were led by senior researchers with experience in leading focus groups (AB, AH, JR), and supported by two researchers (RS or AB) who took field notes. Participants completed a questionnaire on demographic characteristics prior to the focus group. Focus groups were recorded and subsequently transcribed. Patients received a summary of the transcripts after the focus groups were conducted.

An interview guide was used to guide the focus groups (Box 1). This semi-structured guide was developed by the research group, based on expert opinion and previously published literature(13, 14). Topics discussed were: 1) information provision and communication about the ICD at the end-of-life, 2) attitudes towards ICD deactivation and 3) suggestions for improvement of information provision and communication.

Box 1. Interview guide used to facilitate focus groups

- ► Information provision and communication about the ICD at the end-of-life:
    - Did you ever had a conversation with your healthcare professional about what to do with your ICD when you get older or sick? What was discussed?
    - How do you value the quality of this conversation?
    - How would you prefer to get informed about the ICD at the end-of-life?
- ► Attitudes towards ICD deactivation
    - Did you ever think about what to do with your ICD when you get older or sick?
    - Would you deactivate the device? Why (not)?
- ► Points for improvement
    - Looking back at the conversation you had with your healthcare professional, are there things you would like to see improved?

ICD, implantable cardioverter defibrillator

Data analysis

Transcript were analyzed thematically, using the constant comparative method, a data-analytic process whereby each interpretation and finding emerging from the data is compared with previous findings(15). Transcripts were read by JR and RS and meaningful themes were inductively identified and summarized in a preliminary coding tree. The coding tree was discussed with the coauthors, tested on one of the transcripts, refined and finalized. Subsequently, all transcripts were coded by RS, and checked by JR. The two researchers met frequently to discuss the coded transcripts, and to discuss and resolve minor disagreements.

Patient and Public Involvement

A patient advisory group met frequently for the duration of the study and provided feedback on informational materials and interview guides. At the end of the study, they commented on the findings.

RESULTS

Focus groups lasted an average of 97 minutes (range 89-107). Patients were more often male (56%) and had a mean age of 64 (SD 9.7). A majority had their ICD implanted for primary prevention (59%), and 44% had a Cardiac Resynchronization Therapy – Defibrillator implanted, on average 6.4 (SD 4.8) years before participation in the study (Table 1).

The results are described along the line of the three key elements of ACP: 1) Reflection on wishes and preferences; 2) discussing preferences with healthcare professionals and relatives; 3) recording and reviewing preferences(9). Illustrative quotes per key element are presented in Table 2.

Table 1. Characteristics of patients enrolled in the focus groups (n=41)

Gender (male)	23 (56%)
Mean age (SD)	64.3 (9.7)
Marital status	
Unmarried	2 (5%)
Married	30 (73%)
Divorced	6 (15%)
Widowed	3 (7%)
Education	
Less than high school	1 (2%)
High school graduate	10 (24%)
Some college	8 (20%)
College graduate	15 (37%)
University degree	7 (17%)
Indication for ICD (primary prevention)	24 (59%)
Type of ICD	
Single chamber	9 (22%)
Dual chamber	6 (15%)
CRT-D	18 (44%)
S-ICD	1 (2%)
Unknown	2 (5%)
Mean years implant (SD)	6.4 (4.8)

CRT-D, cardiac resynchronisation therapy-defibrillator; ICD, implantable cardioverter defibrillator; S-ICD, subcutaneous-ICD.

Table 2. Illustrative quotes regarding ACP

Elements of ACP	# Quote	Quote
1) Reflection on wishes and preferences regarding future ICD deactivation	Q1	*I thought they implanted the ICD so I was not able to die?* – Male patient, focus group 5
	Q2	*“When do I have to do this (deactivate – RS)? If I have problems with my heart? Should I do it if I get some sort of cancer? Or suffer from dementia? When to deactivate the ICD, that is a difficult question”* – Male patient, focus group 4
	Q3	*“I have that fear (of shocks – RS) as well, and based on that fear I would say deactivate my ICD as quickly as possible when something happens. If I get sick or whatever, done with it”* – Male patient, focus group 2
	Q4	*“Well I have the ICD simply to ensure that it intervenes if […] a cardiac arrest would occur. […] and there is no other way (to terminate the cardiac arrest – RS) than by getting a shock. So I think I’ll welcome it then”* – Male patient, focus group 2
	Q5	*“There is of course a difference when you are terminal due to cancer […]. Because if you are terminal due to your heart disease, then such device is much more important. If you would deactivate then, then you will certainly go (die – RS). And with cancer, it can be very different* – Female patient, focus group 5
	Q6	*“I do not want my wife, children and grandchildren to stand beside my bed, while I am bouncing up and down in my last moment. I do not want that […] so for me, it is clear what I want.”* – Male patient, focus group 4
2) Discussing preferences	Q7	*“Then he (the cardiologist – RS) told me […] we could also deactivate halfway through. Well, I thought that was a wonderful thought”* – Male patient, focus group 3
	Q8	*You come in as a patient, you feel like you are giving your life to such a doctor, because that is what it comes down to […] and he does not take sufficient time to explain it properly, such an important issue as the end-of-life. And that always annoys me […] they just do not take the time for it. You are there for ten minutes and that’s it* – Male patient, focus group 4
	Q9	*[…] I think the cardiologist should pay more attention to that (the ICD at the end-of-life – RS), they should realize that it is a very important topic to us* – Male patients, focus group 4
	Q10	*“I did not have this conversation about when to turn it off, but I do not miss it at all. Because then I think well, I really am in a phase of getting better and cure, so then I think yes, I am not waiting for this (conversation about ICD deactivation – RS) at all”* – Female patient, focus group 2
	Q11	*“I have talked about it with my children, but they really do not want to know about it yet” – Female patients, focus group 5*
3) Recording and reviewing preferences	Q12	*“I do think about it (deactivation – RS) every now and then, but to record my preferences, let me just say it, that is going too far, because you do not know what it is like when you are at that moment, that you are what you recorded* – Female patient, focus group 3

	Q13	*"I will make an advance care directive, not so much because of own situation, but because of the fact that my father died six months ago and he had one. Back then I though well, everyone should have such a document because… well, for me it felt good and I will make one myself as well"* – Female patient, focus group 2

Note. ACP: Advance Care Planning; ICD: Implantable Cardioverter Defibrillator

Reflection on wishes and preferences regarding future ICD deactivation

Some patients were not aware that ICD deactivation was an option, and one patient thought he was not able to die with an ICD (Q1 in Table 2). Of those who were aware of ICD deactivation, wishes and preferences differed. Most could imagine deciding to have their ICD deactivated one day. However, some patients indicated they could not imagine ever asking for deactivation. One patient for instance indicated that deactivating the ICD could feel like “euthanasia”. Euthanasia, defined as ending a patient’s life by administering medication by a physician with the explicit intention of hastening death, at the explicit request of the patient, who suffers unbearably without prospect on relief, is legalized in the Netherlands under strict criteria stipulated by the Dutch law(16). Some patients were uncertain about in what situation deactivating their ICD would be appropriate, stating that the decision to deactivate is complex and dependent of multiple factors (Q2).

Patients often considered the balance between quality versus length of life when reflecting on possible future ICD deactivation. This balance differed between people, but generally patients indicated that postponing death – just because it is possible – without taking quality of life into account does not make sense. The ICD was considered a lifesaving device by many patients, which was also apparent in how patients talked about their device. They often used words such as ‘angel’, ‘guard dog’ and ‘safety net’. However, some patients described a ‘love-hate’ relationship with their device: previous shocks had saved them from a sudden death, but were painful, often a source of concern, and a personal confrontation with being ill and the finiteness of life. The possibility of experiencing unwanted shocks while dying was often brought up as an important factor when reflecting on the role of the ICD at the end-of-life. One patient described that their fear of receiving shocks was greater than the fear of dying (Q3). However, another patient indicated to always want arrhythmias to be treated by shocks, even if death was imminent (Q4).

Some patients stated that living longer with a compromised quality of life was undesirable. Losing independency and being diagnosed with a life-limiting illness were amongst the most frequently mentioned factors threatening quality of life. However, there was a distinction made in the nature of the disease. Advanced cancer or advanced lung disease was brought up by several patients as a clear indication to ICD deactivation, since an active ICD would potentially mean having to live longer with symptoms such as pain and discomfort, but, as one patient described, a progression of heart disease could make one hesitant about ICD deactivation (Q5).

Patients indicated that their relatives would play an important role in the decision-making about ICD deactivation. Several patients indicated that they did not want their relatives to witness them whilst receiving shocks at the end-of-life (Q6). On the other hand, some others mentioned that if their relatives would want them to continue ICD therapy, they would.

Discussing preferences

Although patients used different sources of information, including the internet and patient folders from the hospital, the preferred mode to receive information on ICD deactivation was by having conversations with the healthcare professional. Some patients had had this conversation, mostly with their general practitioner or cardiologist and on their own initiative. Such conversations gave relief to some patients (Q7). The vast majority of patients however, indicated they never had had such conversations, which was source of discontent among some patients (Q8).

Patients described their cardiologist as very knowledgeable on a medical-technological level, but some indicated they felt there was not always the opportunity to discuss issues with a strong emotional component. Various reasons were suggested for this, such as a perceived lack of time or willingness of the cardiologist, as well as unawareness of importance of the healthcare professionals (Q9). These patients envisioned a greater role for other healthcare professionals to support the cardiologist in conducting these conversations, such as nurses or the general practitioner.

There was debate on the timing of conversations about ICD deactivation. Some patients thought it was best to engage early in ACP conversations about ICD deactivation, starting before implantation. However, others felt that bringing up issues related to the end-of-life at this stage might be inappropriate, since implantation of the ICD felt like a ‘second chance at life’. Therefore, some suggested to start up such ACP conversations during follow-up visits. However, others disagreed, since patients might get worried about why the physician would bring up the topic at that particular moment. Other moments were also identified to discuss ICD deactivation: when faced with a life-limiting illness, or when a do-not-resuscitate order is being discussed.

While most patients had positive attitudes towards discussing the end-of-life and ICD deactivation, this was not true for all patients. Some described they focused on living and getting better, and that they felt no need to think about future ICD deactivation (Q10).

Several patients mentioned that they had discussed possible future ICD deactivation with their relatives. Some considered such conversations to be more important than conversations with healthcare professionals. Most patients had positive experiences with talking with their relatives about ICD deactivation. Yet, some patients indicated that their relatives did not want to engage in such conversations (Q11).

Recording and reviewing preferences

Some patients had their preferences for future medical care and treatments recorded in an advance care directive, or were planning on doing so. Not everyone was in favor of recording their preferences about ICD deactivation in a document, because they were not sure what they would want when their disease would progress. It was also described that, even if someone has well-considered ideas and whishes about what to do in specific situations, these ideas could be opposite of how someone acts when they are actually in that situation, and that people adapt to their current situation (Q12). They said that multiple factors influence the decision to deactivate, such as prognosis, age, and severity of illness. Also, they were doubtful whether their wishes would be respected. On the contrary, others were in favor of reporting their preferences about ICD deactivation in an advance care directive. One patient had an earlier experience with a loved one at the end-of-life, and indicated that an advance care directive gave some clarity (Q13).

DISCUSSION

Little research has been conducted on the experiences of ICD patients with ACP conversations about ICD deactivation. Although international expert consensus statements recommend to timely and frequently discuss ICD deactivation with the patient(1), recent studies show these conversations are scarce(2). Several patients in our study indicated that their healthcare providers are capable regarding medical-technological issues, but felt there was not always the opportunity to discuss topics with an emotional component, such as the end-of-life. This was also shown in an American focus group study with nurses, where nurses stated that physicians sometimes ‘fail’ in considering psychosocial, economic, and ACP aspects of living with an ICD(14). Another American interview study with ICD patients showed that patients seemed to deprioritize ACP conversations, and overemphasize the lifesaving abilities of the ICD(17). In our study, we saw that some patients were unaware of the option of ICD deactivation. This lack of knowledge in the patient could decrease the willingness of ICD patients to engage in ACP and discuss deactivation(18, 19). A lack of ACP conversations might impair the decision-making at the end-of-life, leading to reactive decision-making concerning ICD deactivation(14). Several patients indicated that it is important to discuss ICD deactivation with their relatives. However, involving families could also cause conflicts, such as relatives wanting the patient to keep the ICD active(14). This could be due to a knowledge deficit in the relatives as well, and could be reduced by including them in ACP conversations(14). The concern of family conflicts was not mentioned in our focus group discussions, although one patient mentioned to keep the ICD on if her family would want so.

There was debate on the timing of discussing ICD deactivation. Some patients were hesitant to discuss deactivation before implantation, although this would make it easier to start conversations when deactivation becomes more directly relevant(19). Previous research showed that conversations about ICD deactivation often only occurred when indicated during follow-up or at the end-of-life(11, 20). Postponing the discussion until the end-of-life is not recommended, since patients might have too little time to reflect upon their decision, and the last phase of life is hard to identify in heart failure patients(21), as shown in a previous study, in which only 15.7% of included healthcare professionals were confident in predicting death, which might impair the timing of ACP conversations(22).

Previous research showed that ICD patients are more reserved than other patient groups towards ACP conversations, amongst others because the ICD is often implanted before patients perceive themselves as being seriously ill(13). Also, the ICD is often advocated as a solely life-saving device(23). Also in our study, some patients developed a complex psychological relationship with their ICD, viewing it as a ‘trusted friend’ and an integral part of their body, which might make it difficult for patients to talk or even think about deactivating the ICD(13, 24). In our study, some patients indicated they do not want a conversation about ICD deactivation. Although this should be respected to some extent, since ACP conversations should be tailored to the readiness and the phase of life of the patients(9), we do feel that the healthcare professional also has an informative role, in which all benefits, harms, and future perspectives of a treatment should be discussed. Also, patients should have the opportunity to elicit general treatment preferences and goals of care in addition to their deactivation preferences, since these might play a role in their later decisions about ICD deactivation(25).

Possibly helpful for patients might be to record their wishes and preferences in an advance care directive. In our study, only few patients did so. This was also apparent in other studies that showed that patients do often have preferences recorded on for instance feeding tubes or respirators, but are hesitant to report preferences on ICD deactivation(26, 27). Possible explanations for this might be that preferences are subject to change, and patients are not certain whether their current wishes would represent their wishes at the end-of-life. However, recording preferences might help patients to actively think about their preferences, and could reduce ethical dilemmas or moral distress in relatives or healthcare professionals(28). Also, if patients are informed that the advance care directive is a ‘living’ document, which can always be adjusted, they might be less hesitant to record their wishes.

This is one of the few studies exploring the experiences of ICD patients with ACP conversations in depth. A strength is its large number of participants(29) recruited in multiple centers. Yet, some limitations have to be addressed. Participants were recruited via the Dutch ICD patient association. It is possible that only people responded with special interest in the topic or with negative experiences, and that ACP conversations occur more often in practice. Also, it should be noted that most patients in the focus groups were in general good health, and not at the end of their lives, which might influence their wishes and preferences. Our study was conducted in the Netherlands, where there is ample attention in the public and medical-professional domain to support ACP. More generally speaking, it is a country with a rather open debate about end-of-life decision-making, also showing from the issue of euthanasia that was mentioned in one of the focus groups. This means that our findings need replication in other countries and cultures.

Implications

This study and previous studies showed a variability in how and when patients want to be informed and in their attitudes towards deactivation. In line with the international ACP consensus statement(9), we recommend healthcare professionals to explore the patient's readiness to talk about end-of-life and ICD deactivation, so that information can be tailored to the needs of the patient. Specific time points to explore this are before implantation, at battery replacement, when health deteriorates, or when a patient is referred to palliative care(1). Such conversations about ICD deactivation could be incorporated in more general conversations about goals of care, values, and preferences, so that a clear understanding of the patient's wishes could be established. We recommend to involve relatives in these conversations as well(9), as also indicated by the patients in our study.

CONCLUSION

Many patients reflected on the role of their ICD at the end-of-life and report a need to be better informed about this topic. However, ACP conversations with the healthcare professional about treatment preferences and ICD deactivation seemed to be uncommon. Preferences about ICD deactivation were personal and dependent on the situation. Therefore, caution is advised in using one-size-fits-all approaches in informing the patient about deactivation. Some patients were hesitant to record their preferences in an advance care directive, since they were unsure whether their current preferences would reflect their future preferences.

FOOTNOTES

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Research ethics and patient consent

Before conducting this study, approval of the ethics committee of the Erasmus MC, Erasmus University Medical Center Rotterdam, was obtained (METC-2017-357). All patients were given both written and verbal information on the study and provided consent in writing.

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8

CHAPTER 8

Relatives' experiences with advance care planning and the last phase of life of patients with an implantable cardioverter defibrillator: A mixed-methods study

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Under review

ABSTRACT

Background Little is known about the last phase of life of patients with implantable cardioverter defibrillators and the practice of advance care planning in this population.

Aim To study bereaved relatives' experiences of the last phase of life of implantable cardioverter defibrillator patients, advance care planning conversations and satisfaction with care.

Design Mixed-methods study, including a survey and focus group study.

Setting/participants A survey among 170 relatives (response rate 59%) reporting about 154 deceased patients, and 5 subsequent focus groups with 23 relatives.

Results Relatives reported for 38% of patients that they had had a conversation with a healthcare professional about deactivation. Patients' and relatives' lack of knowledge about implantable cardioverter defibrillator functioning and the perceived lack of time of healthcare professionals were frequently mentioned barriers to advance care planning. For 24% of the patients, relatives reported that they had received shocks in the last month of life, invoking distress in 74% of these patients and 73% of their relatives. 42-61% of relatives reported to be satisfied with different aspects of end-of-life care, such as the way in which wishes of the patient were respected. Patients' quality of death was scored relatively low, but was higher for patients with a deactivated device than those with an active device (mean 6.74 versus 5.67 on a 10-point scale, p= 0.012).

Conclusions Implantable cardioverter defibrillator deactivation was discussed with a minority of patients, and one in four patients received shocks in the last month of life that were distressing to patients and relatives.

INTRODUCTION

An implantable cardioverter defibrillator (ICD) treats potentially lethal arrhythmias by delivering electrical shocks, thereby prolonging life[1]. At the end of life, shocks may no longer be appropriate, since prolonging life might also mean possible prolongation of suffering. This can be prevented by timely deactivating the ICD. Guidelines recommend to timely and frequently discuss ICD deactivation with the patient and relatives throughout the disease trajectory[1, 2]. Advance care planning (ACP), the process of defining, discussing and reviewing goals and preferences for future medical treatment and care[3], might help patients and relatives making a well-informed decision about ICD deactivation. Previous research showed that these conversations are conducted in up to 21% of patients[4, 5], ICDs are deactivated in 30%, and 7% of patients experience shocks in the last month of life[5]. However, studies often rely on data from hospital medical records[6], which may not be complete. They might lack important information about the end of life of patients, especially of those who die outside the hospital, and about the underlying reasons for why ICD deactivation was discussed. Relatives usually have an important role in the care for patients, and are usually an important partner in making decisions about care and treatment in the last phase of life[3, 7-9]. Examining their views and experiences will give unique insight into the last phase of life of the patient, including the decision-making about ICD deactivation[4, 10].

We performed a mixed-methods study, including a survey and focus groups among relatives of deceased ICD patients to: (1) examine ACP conversations about ICD deactivation, including barriers and facilitators; (2) examine the last phase of life of ICD patients; and (3) examine the satisfaction with treatment and care in the last phase of life of ICD patients and relatives.

METHODS

Study design

This study had a mixed-methods design. Approval of the ethical committee of the Erasmus University Medical Center Rotterdam (MEC-2017-357) was obtained before starting the data collection. All participants provided written informed consent.

Survey study

Sample

Relatives were identified in the medical records of ICD patients deceased between 2012 and 2017 from a university hospital and a general hospital in the Netherlands. Relatives were contacted by phone between December 2017 and March 2018 and were invited to participate in the study. Relatives were eligible when they were the relative of the deceased ICD patient, were able to speak Dutch, were older than 18 years, and were able to provide written consent. Relatives who were interested in participating received additional information, an informed consent form and the survey by postal mail. Those who did not return the survey, received a reminder one month after the initial mailing. A total of 419 relatives were identified in the medical records of 297 deceased patients.

Measurements

The survey included 52 questions. Questions were based on a previously conducted phone survey by Goldstein[4], which was adapted from the After-Death Bereaved Family Interview[11]. Questions were translated into Dutch. Self-constructed questions on the impact of ICD shocks on both the patient and the relative were added. The survey was pilot tested in eight patients and adjusted. The last phase of life was operationalized as the last month preceding death[4]. The survey consisted of eight sections (see Supplementary Appendix 1), with - amongst others – sections about the last phase of life and death of the patient, ACP conversations about the ICD, and statements on satisfaction about care in the last phase of life (on a five point Likert scale, 1 meaning totally agree, 5 totally disagree) and quality of death (rated on a scale from 1-10)

Data analysis

One-hundred-seventy relatives participated in the survey study, reporting on 154 patients. For 15 of the 154 patients more than one relative responded (for 14 patients, two relatives responded, for one patient, three relatives responded). For these 15 patients, we used the questionnaire of the relative closest to the patient based on their relationship (in order: spouse, child) for analyses. For the analyses of questions related to the personal opinion of the respondents (e.g. questions on satisfaction with treatment and care), we used all 170 respondents. Descriptive statistics were used to present the data. For the statements on satisfaction, scores of 1 (‘totally agree’) and 2 (‘agree’) represented the category ‘agree’. Reported quality of death was compared by reported shocks in the last month of life, occurrence of ACP conversations and ICD deactivation with student t-tests. A p-value of <0.05 was considered statistically significant. Analyses were performed using SPSS version 24.

Focus group study

Sample

Relatives who completed the survey were invited to participate in focus groups held in the Erasmus University Medical Center. If they were, they received the study information sheet. Those participating received a €25 gift card and travel reimbursement.

Measurements

An aide memoir was used in conducting the focus groups. Topics discussed were 1) information provision and communication about ICD deactivation; 2) experiences with patient’s last phase of life; and 3) suggestions for improving end-of-life care. Focus groups were led by experienced moderators (JR, AB or RS), and supported by AB, RS or HE.

Data analysis

Transcripts were read and analyzed using the constant comparative method[12]. Meaningful themes were identified and a coding tree was developed. The coding tree was discussed with the coauthors, and subsequently tested and refined. Transcripts were analyzed by RS, and checked by AB and JR. The three researchers met frequently to discuss the transcripts. Consensus was sought and found in case of disagreements.

RESULTS

Study population

Survey study

Of the 419 identified relatives, we contacted 289; 247 agreed to participate and 170 returned a completed survey (response rate 59%) (Figure 1). They reported on 154 unique patients who died a median of 27 months (IQR 17-44) before completion of the survey, at a median age of 74 (IQR 66-79). 87% of patients were male. Relatives had a median age of 62 years (IQR 50-75) and 22% were male (Table 1). Of the relatives, 52% were spouses, 33% children, 13% other relatives, 1% friends, and 1% neighbors.

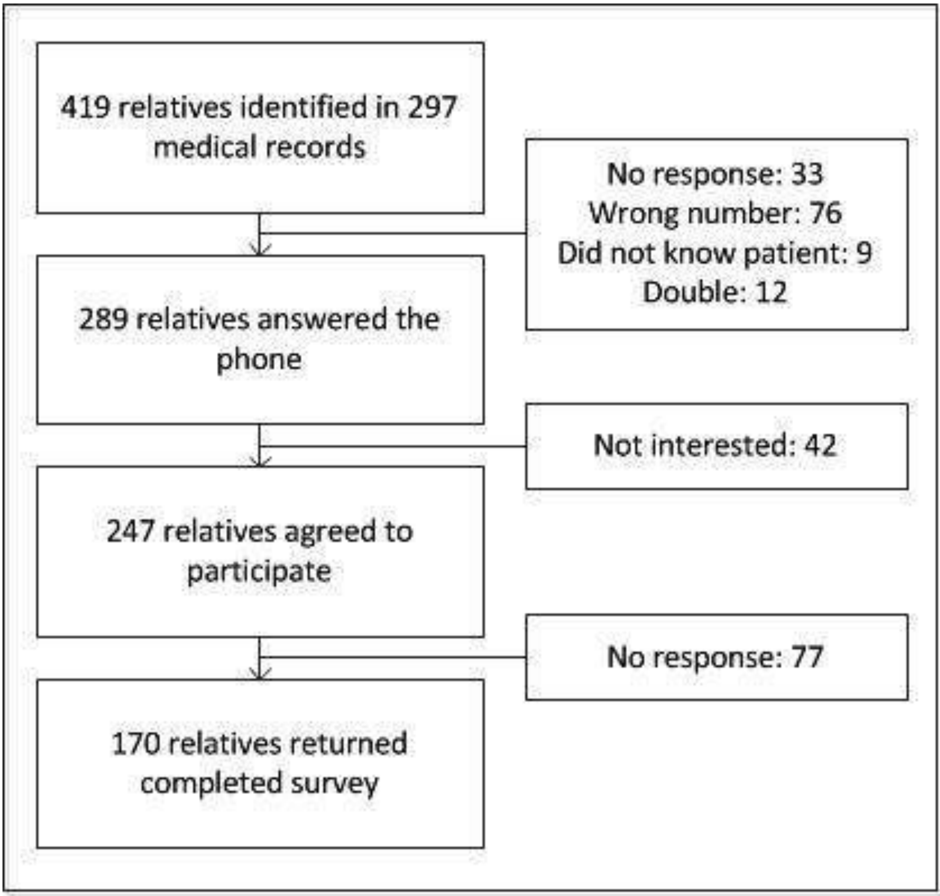

Figure 1. Inclusion flowchart

Table 1. Characteristics of relatives of decease ICD patients (N=170), and the patients they report about (n=154) (survey study)

Relatives (n=170)	n (%)
Gender (male)	37 (22%)
Age (median, IQR)	62, 50-75
Patient was his/her[1]	
Spouse	89 (52%)
Parent	2 (1%)
Child	56 (33%)
Other	22 (13%)
Ethnicity[1]	
Dutch	163 (96%)
Other	6 (4%)
Education level[2]	
Low	59 (35%)
Medium	65 (38%)

High	44 (26%)
Unknown	2 (1%)
Patients (n=154)	
Gender (male)	134 (87%)
Age at death (median, IQR)	74, 66-79
Year of death	
2012-2013	33 (21%)
2014-2015	58 (38%)
2016-2017	63 (41%)
Ethnicity	
Dutch	146 (95%)
Other	8 (5%)
Education level[2]	
Low	83 (54%)
Medium	41 (27%)
High	24 (16%)
Unknown	6 (4%)
WHO-performance status last month before death[1]	
0 (able to carry out all normal activity)	5 (3%)
1 (restricted in activity, but able to carry out light work)	45 (29%)
2 (Capable of self-care, unable to carry out work activities)	31 (20%)
3 (Limited self-care, confined to bed or chair >50% of waking hours)	48 (31%)
4 (Completely disabled)	24 (16%)
Cause of death[1]	
Cardiovascular	91 (59%)
Cancer	21 (14%)
Other	34 (22%)
Unknown	7 (5%)
Place of death[3]	
Hospital	79 (51%)
Home	47 (31%)
Nursing home	15 (10%)
Hospice care facility	8 (5%)
Other[4]	3 (2%)

Notes: ICD: Implantable Cardioverter Defibrillator; IQR: Inter Quartile Rate; WHO: World Health Organization

[1] One response missing

[2] Low: Primary school or lower general secondary education; Medium: Intermediate vocational education or pre-university education; High: University or university of applied sciences

[3] Two responses missing

[4] Other places of death were: rehabilitation center (n=2); care hotel (n=1)

Focus group study

23 of 58 interested relatives participated in a focus group. We held five focus groups with respectively 4, 5, 5, 7, and 2 relatives. Relatives were most often the child (48%) or spouse (39%) of the deceased patient. Mean age was 56 years (SD 14.1) and 22% were male.

Advance care planning in ICD patients

Survey study

Fifty-eight of 170 relatives (34%) indicated having had a conversation with the patient about ICD deactivation (Table 2). According to the relatives, 59 of 154 patients (38%) had had a conversation about ICD deactivation with a healthcare professional, 15 (25%) in the last month, and 11 (19%) in the last week of life. Forty-seven (80%) relatives were present during these conversations. In 11% of the patients who did not have a conversation about ICD deactivation the relative indicated that such a conversation would have been appreciated. Relatives reported that 18 patients (12%) had recorded their preferences for medical treatment and care in an advance care directive. Five of these documents addressed ICD deactivation.

Table 2. Advance care planning in ICD patients (N=154) (survey study)

ICD deactivation conversation between relative and patient[1,2]	58 (34%)
ICD deactivation conversation between patient and healthcare professional	59 (38%)
Patient in need of ICD deactivation conversation, according to relative[3]	10 (11%)
Timing of conversation between patient and healthcare professional	
Pre-implantation	17 (11%)
More than one month before death	22 (14%)
Last month before death	15 (10%)
Last week before death	11 (7%)
Last day before death	4 (3%)
Last hour before death	2 (1%)
Conversation between patient and healthcare professional initiated by[4]	
Patient	12 (8%)
One of the relatives	8 (5%)
A physician	27 (18%)
A nurse	3 (2%)
Someone else	5 (3%)
Not sure	3 (2%)
Patient discussed preferences regarding medical treatment and care[3]	
With relative	51 (33%)
With other family members	36 (23%)
With physician	39 (25%)
With nurse	11 (7%)
With someone else[5]	1 (1%)
Patient had documented an advance care directive[6]	18 (12%)

Patient had documented wishes on ICD deactivation	5 (3%)
Patient documented a healthcare proxy[2]	64 (42%)
DNR-order at time of death[4]	103 (67%)

Notes: ICD: Implantable cardioverter defibrillator; DNR: Do-not-resuscitate

[1] All 170 relatives were included in calculating this variable

[2] Three responses missing

[3] Only patients who did not have an ICD deactivation discussion were included in calculating this variable (n=95)

[4] One response missing

[5] Home care nurse

[6] Not including DNR-order

Focus group study

In the focus groups, barriers and facilitators towards ACP conversations were discussed (Table 3). We categorized these into patient and relative related factors, and healthcare provider and system related factors. Some relatives reported that the patient's willingness to discuss deactivation and their level of curiosity regarding understanding their device were important facilitating factors in having ACP conversations, as one relative described:

> *"As a patient, at a certain moment, you do have the obligation to ask a little more proactively about what a certain product does, what it is, and how to deal with certain things […] I think he did not even have the ICD for a month when we already had that conversation (about ICD deactivation – RS). And that makes you aware about the ICD, how it works and what it does"*
>
> – Focus group 4, participant F032, 33 y/o son

Other facilitating factors were a good patient-doctor relationship, and the involvement of relatives to empower them to be involved in the decision-making process about ICD deactivation. Some relatives reported that a lack of knowledge about the functioning of the ICD, and a focus on its lifesaving capacity could act as barriers.

Some relatives mentioned they thought that healthcare professionals other than cardiologists (such as the GP) were not always knowledgeable about the functioning of the ICD and the possibility to deactivate. Also, some relatives perceived ACP conversations about ICD deactivation to be ‘routine practice’, which made some feel they were ‘one of many’, as one relative described:

> *“Sometimes you get the feeling like you are walking into a factory, and you are just one of many… yes I do understand that the doctor may have this conversation 365 times a year, but well […] for us it is only one time”* – Focus group 3, participant F127, 30 y/o son

At the same time, routineness was valued by others, as this would normalize the topic and ensured its discussion. Another discussed barrier was the perceived lack of time of the healthcare professional, while some personal characteristics of the healthcare professional, such as a younger age, were sometimes perceived to be a facilitator.

Table 3. Barriers and facilitators of ACP conversations with the healthcare professional, according to relatives of deceased ICD patients (focus group study)

Patient and relative related barriers	
Theme	Quote
Willingness to talk about ICD deactivation	*"You were not able to talk with my father-in-law about death at all, that was taboo"* – Focus group 1, participant F079, 56 y/o daughter *"My parents were already of age, and if you do it (conversation about deactivation – RS) right from the start (implantation – RS), so with older people, so much information comes at you… you do not remember half of it. And of course you also think a bit slower, so the impact of all those choices… it is just too overwhelming"* – Focus group 5, participant F126, 36 y/o daughter
Relationship with the healthcare professional	*"My parents also did not ask the doctor questions. I said 'did you…?', (they responded – RS) 'well the doctor said it is like this, so…'"* – Focus group 3, F108, 50y/o daughter *"He did not have a nice doctor with whom you could have a nice conversation […] the 'how and what', if you asked about that, he would not discuss this"* – Focus group 4, participant F128, 53 y/o daughter
Not the right moment to talk about ICD deactivation	*"To be honest, he was doing so well, we never thought about it (deactivation of the ICD – RS) when we had our check-up at the ICD-technician […] never came up with "well, if something happens, what are you going to do?" Because he was… he was doing so well, made so much progress and had so many plans… You do not think about it, including myself"* – Focus group 4, participant F061, 65 y/o wife
Lack of knowledge about the ICD	*"Three weeks prior to his death we knew that the last phase was upon us, but he still wanted every time… because he thought that if the ICD is deactivated, it is over, because that had always been his lifeline"* – Focus group 3, F108, 50y/o daughter *"You are so happy that such a thing (the ICD – RS) exists, you won't think about the last phase of life, because you assume you won't be in the last phase of life anytime soon, since that thing (the ICD – RS) was implanted"* – Focus group 1, participant F013, 59 y/o daughter
Patient and relative related facilitators	
Willingness to talk about ICD deactivation	*"My father was a very open man, and spoke very easily about everything, including the end and death"* – Focus group 1, participant F013, 59 y/o daughter
Relationship with healthcare professional	*"But then (to discuss ICD deactivation – RS) you should also have a good relationship with your doctor"* – Focus group 1, participant F0792, 59 y/o son
Involvement of relatives in ACP conversations	*"But that is why I think it is wise to always be with two people at such a conversation (about ICD deactivation – RS), so you know what is being asked […] you were the one who was alert on that moment (at the end of life – RS), so you can indicate it (to deactivate the ICD – RS)"* – Focus group 2, participant F138, 75 y/o wife

	"We are also working in healthcare ourselves, my sister and I and my cousin, so we thought in his last phase 'oh wait, I think we should do something with this' (with the ICD – RS)" – Focus group 3, F108, 50y/o daughter
Healthcare provider and system related barriers according to relatives	
Knowledge about the ICD	*"His own GP was allowed to come […], but his GP also did not have the information available. And I think that might be something that could be improved in terms of information provision, that the GP also knows how important it is to be aware of the ICD, so they can act accordingly"* – Focus group 2, participant F113, 60 y/o daughter
Routineness of conversation	The hospital is sometimes compared to a factory, it is so big, so immense […] You will get 20 minutes and then it is 'bam, bam, bam', they (cardiologists – RS) need to see as many patients as possible on one day" – Focus group 3, participant F127, 30 y/o son
Perceived lack of time for ACP conversations	*"I assume that every doctor who works here is pretty good in his field […] but I think the pressure on doctors is just too great, that there are too many patients […] you will get 15 minutes […] I think they do not really have the time to sit with you and to explain how everything works"* – Focus group 3, F030, 33 y/o son
Healthcare provider and system related facilitators according to relatives	
Knowledge about the ICD	*"He (the patient – RS) once received a letter from the hospital after 5 years or so about when you decide to deactivate that thing (the ICD – RS) […] from there on we actively continued at the hospital and said 'we want to record when we want to deactivate it (the ICD – RS)'"* – Focus group 1, participant F066, 49 y/o daughter *"I thought the information meeting was really great. That is a very neutral moment, you are just like: okay, when the time comes, we must think about it"* – Focus group 3, F108, 50y/o daughter
Routineness of conversation	*"Isn't it an idea to […] just have a 5-minute-conversations about that (ICD deactivation – RS)? So that you as a doctor at least know you that have discussed it […], also recurrent. You can sometimes switch cardiologist […] this is the day we had a conversation, let's do that again in a years and a half or two years. Well, you can easily plan that"* – Focus group 4, participant, F032, 33 y/o son
Characteristics of healthcare professional	*"So as a healthcare professional, I think you should alert a patient. And then in a friendly manner, not that you think… you know I do not need to know all that, but do you have the need for us to explain how it will go in the future if you may not have much longer to live?"* – Focus group 4, participant F128, 53 y/o daughter *"I also have the idea that younger doctors can more easily deal with this… anyway in the overall communication compared to the somewhat older doctors […] that is a world of difference, that (communication with older doctors – RS) was clinical"* – Focus group 3, F030, 33 y/o son

The last phase of life

Survey study

Relatives reported that 113 (73%) patients were admitted to a hospital at least once in the last three months of life (Table 4). For 125 (74%) relatives and 108 (71%) patients, it had been clear that the patient was going to die within several days. In 84 (55%) patients, the ICD was deactivated, 35% in the last week and 32% on the last day of life, in most cases by the ICD technician (49%) in the hospital (74%). Relatives reported that 37 (24%) patients experienced one or more shocks in the last month of life, 25 (16%) in the last week, and 16 (10%) on the last day of life. Relatives reported that 74% of patients were distressed by these shocks, and 73% of relatives reported to be distressed themselves.

Table 4. The last phase of life of ICD patients (N=154) (survey study)

Hospital admission in last three months of life (yes)	113 (73%)	
It was clear to the relative that death was imminent[1,2]	125 (74%)	
It was clear to the patient that death was imminent[2]	108 (71%)	
ICD shocks		
During last month	37 (24%)	
During last week	25 (16%)	
On last day	16 (10%)	
Effect of shocks in last month of life	On relative[3]	On patient[4]
Pain	-	13 (35%)
Fear	16 (41%)	13 (35%)
Sadness	7 (18%)	11 (30%)
Stress	8 (21%)	9 (24%)
Worry	23 (59%)	11 (30%)
Helplessness	16 (41%)	7 (19%)
Anger	3 (8%)	3 (8%)
Confusion	3 (8%)	6 (16%)
Not sure	2 (5%)	2 (5%)
No effect	3 (8%)	1 (3%)
ICD deactivated	84 (55%)	
Timing of ICD deactivation[5]		
More than a month before death	12 (14%)	
Weeks before death	16 (19%)	
Days before death	29 (35%)	
Last day of life	27 (32%)	
ICD deactivated by[5]		
Physician	20 (24%)	
ICD technician	41 (49%)	
Industry representative	1 (1%)	
Unknown	22 (26%)	
Location of deactivation[5]		

Home	13 (16%)
Hospital	62 (74%)
Nursing home	4 (5%)
Hospice	3 (4%)
Care hotel	1 (1%)
Not sure	1 (1%)
Form of deactivation[5]	
Reprogramming device	23 (27%)
Magnet	29 (35%)
Surgical extraction	2 (2%)
Not sure	30 (36%)

Notes: ICD: Implantable cardioverter defibrillator

[1] All 170 relatives were included in calculating this variable

[2] One response missing

[3] Only the 39 relatives of patients who had a shock in the last month of life were included

[4] Only the 37 patients who had a shock in the last month of life were included

[5] Only the 84 patients who had their ICD deactivated were included

Satisfaction with treatment and care in the last phase of life

Survey study

Of the 170 relatives, 104 (61%) agreed with the statement that healthcare professionals had sufficiently informed them and the patient about the patient's prognosis. Seventy-one (42%) relatives agreed with the statement that issues related to the ICD in the last phase of life were sufficiently discussed by the healthcare professional. Ninety-two (54%) relatives felt that the healthcare professional respected the wishes of the patient, and 88 (52%) felt that they received good emotional support from the healthcare professional. For the 59 patients who had an ICD deactivation conversation with the healthcare professional and the 47 relatives who were present during this conversation, respectively 42 (71%) and 38 (81%) were satisfied with this conversation. Relatives rated the quality of death of the patient with a mean score of 6.4 (SD 2.5). Relatives of patients who had their ICD deactivated reported a significantly higher quality of death (6.74) as compared to relatives of patients who did not have their ICD deactivated (5.67) ($p=0.012$). The occurrence of shocks (reported quality of death with shocks 5.89 and without 6.60) and the occurrence of ACP conversations (reported quality of death with ACP conversations 6.72 and without 5.97) was not significantly associated with quality of death ($p=0.163$ and $p=0.077$ respectively).

Focus group study

Satisfaction with treatment and care in the last phase of life of the patient varied (Table 5). Three themes emerged in the focus groups concerning the satisfaction with treatment and care in the last phase of life. First, the relationship between the patient and relative and the healthcare professional was considered important by many relatives. They greatly appreciated emotionally involved healthcare professionals, respect, honesty and kindness, while others described missing these features.

> *"Honesty is also important, what the doctor had always been, he was always clear and honest, what they (patient and the doctor – RS) really valued in each other"* – Focus group 5, participant F126, 36 y/o daughter

Second, communication between the patient and relative and the healthcare professional was often mentioned. Some relatives described that healthcare professionals were always available for questions and took the time to answer all questions, which was appreciated. However, others described that healthcare professionals never educated the patient about the ICD and the last phase of life, and communication lacked.

> *"We have never heard anything about the last phase of life [...] I feel it (communication about the ICD in the last phase of life – RS) went so wrong with us and I still feel bad about that"* – Focus group 1, participant F079, 56 y/o daughter

Third, communication between healthcare professionals was often mentioned in the focus groups. Some described that this communication lacked, especially when healthcare professionals worked in different institutions, which made relatives feel responsible for the patient's medical care. Some relatives described missing a ‘coordinator’, who could act as a contact person and take the lead in organizing end-of-life care, including ICD management.

> *“There was no information transfer whatsoever, not from the hospital, not from the care facility, nobody… We were the only ones who had information, but only from what we had heard. We did not have the medical records.” – Focus group 2, participant F113, 60 y/o daughter*

Table 5. Satisfaction with treatment and care in the last phase of life according to relatives of deceased ICD patients (focus group study)

	Positive experiences	Negative experiences
Relationship between patient and relative and healthcare professional	*"We are very pleased […] that the doctor was so incredibly nice. Yes, then you can indeed close it in some sort of way"* – Focus group 3, participant F108, 50 y/o daughter *"That good contact with the doctor… He really called two, three times, also to me asking 'how are you really doing?' […] you really felt heard […] very safe and familiar"* – Focus group 3, participant F108, 50 y/o daughter	*"Then (during the last moments – RS) she (the nurse – RS) was there as well. And I did not really liked that, the last moments with my husband and my daughter […] I experienced that as unpleasant […] It was a violation of our privacy, because he looked more at her than at me"* – Focus group 2, participant F005, 73 y/o wife *"I cannot compete with the cardiologist. We did not go along well […] because he did not understand me"* – Focus group 2, participant F005, 73 y/o wife
Communication between patient and relative and healthcare professional	*"I had a good experience with that, when he (the patient – RS) came in so critically, they immediately started talking about the ICD, what are we doing with that?"* – Focus group 4, participant F032, 33 y/o son *"So then (in the last phase of life – RS) I got the GP to explain that, I thought I would find it difficult myself […] and the GP said whenever you need me, I will provide support. So that is what she did, and I really appreciated that"* – Focus group 1, participant F013, 59 y/o daughter *"Whatever you asked, whenever, whatever moment you dropped by, you always could ask the questions you were worried about and you would get a correct answer […] They really took the time for us"* – Focus group 2, participant F094, 62 y/o niece	*"... poor staff, or at least a lot of changes in the staff, that is where I think it went wrong"* – Focus group 4, participant F128, 53 y/o daughter *"I regularly had conversations with the doctors. And they just sat down at his bedside and said 'sir, it is end of story, you are going to die', that was how it was being said. I thought that was very confronting at the time for someone who still had some hope"* – Focus group 2, participant F094, 62 y/o niece *"It should not have been me who started about deactivating it (the ICD – RS). They (the doctors – RS) should have told me that now was the moment to take this step (deactivating the ICD – RS)"* – Focus group 2, participant F094, 62 y/o niece
Communication between healthcare professionals and institutions	*"They (the general practice – RS) had all the information send to them, they exactly knew what was going on […] It was a general practice with 5 GPs, and if she (their own GP – RS) was not there then somebody else took over […] So if you would see another GP, then he would exactly know what was going on, you never*	*"It is the communication, the consultation from one institution to another, yes, that is really lacking. There are mistakes being made" – Focus group 5, participant F126, 36 y/o daughter*

	had to say anything. And whenever there was something special, they would look in the medical record and come instantly" – Focus group 2, participant F031, 75 y/o wife

DISCUSSION

In this study, relatives reported that 38% of patients had an ACP conversation with the healthcare provider, 55% had their ICD deactivated, and 24% experienced shocks in the last month of life. These shocks were distressful for 74% of patients and 73% of relatives. About half of the relatives reported to be satisfied with different aspects of care and treatment in the last phase of life, such as the way in which relatives felt healthcare professionals respected the wishes of the patient. Relatives reported a rather low quality of death, that was somewhat higher when the ICD was deactivated.

A comparable American study conducted by Goldstein and colleagues among relatives of deceased ICD patients in 2004, showed that 27% of ICD patients had had a conversation about ICD deactivation[4]. In our study, this was 38%. This is consistent with the trend of increasing rates of deactivation discussions[5]. It is remarkable that in our study only 11% of the relatives of patients who had not had a conversation about ICD deactivation thought the patient would have been interested in having had this conversation. Studies from the perspective of the patient had contradictory results. They mostly show that patients are in need of and positive about these conversations[8, 13, 14]. However, other studies reported that patients are reluctant to discuss deactivation, partly due to inadequate knowledge of how the ICD works[13, 15], which emphasizes the need to educate patients about the consequences of having an ICD[3]. These contradictory results also show that ACP is not a one-size-fits-all approach, and that the readiness and willingness of the patient to discuss ICD deactivation should be explored[3]. In our study, most relatives wanted to be involved in ACP conversations, although only 28% were present during these conversations. Previous research confirms this and shows that relatives often feel excluded[16]. Both patients and healthcare providers agree that relatives should be involved in ACP conversations[13, 17-19].

We identified several barriers and facilitators towards conducting ACP conversations. Some relatives reported a lack of knowledge in both patients and relatives about the ICD, and patients often focused on the lifesaving capacity of the ICD[20, 21]. This might impair their willingness to engage in conversations about deactivation. In addition, relatives reported that the knowledge about the ICD of healthcare providers who work in specialties outside cardiology (e.g. general practitioners), seemed to be limited as well. Some personal characteristics of the healthcare professionals (e.g. honesty, younger age), and the perceived quality of the patient-doctor relationship acted as facilitators for having ACP conversations[22]. Some of these barriers and facilitators are comparable to barriers and facilitators towards ACP as mentioned by patients with cancer[23]. However, ICD patients are a unique population, amongst others because the disease trajectory in heart failure is much more unpredictable[24, 25], and the fact they have a potentially lifesaving device implanted in their body, which can be deactivated[26], adding to the complexity of decision-making at the end of life.

In our study, 55% of the patients had their ICD deactivated, compared to 21% in Goldstein's study[4]. The increase might be the result of enhanced awareness about this topic and recent publication of guidelines[1, 2]. It is remarkable that the incidence of ICD deactivation in our study is higher than the occurrence of conversations about ICD deactivation between the patient and healthcare professional (38%). This might be due to the possibility that the relative was not aware or had forgotten that the patient had a conversation about ICD deactivation with the healthcare professional. Relatives in our study reported that 24% of patients experienced shocks in the last month of life, and 10% on the last day of life, compared to respectively 27% and 8% of patients in Goldstein's study[4], which is comparable[6]. Studies of medical records reported lower shock incidences, but these might have underestimated the shock incidence[5, 6]. Our study showed that 74% of patients and 73% of relatives were distressed by shocks in the last month of life. The high distress rate amongst patients and relatives, in addition to our finding that relatives reported a higher quality of dying for the patient when the ICD was deactivated, conforms the importance of conducting ACP conversations. Less than half of the relatives reported to be satisfied about the way in which they were informed about the ICD and the last phase of life. This is comparable to previous studies[27, 28], which showed that the information patients received about psychosocial consequences of living with the ICD and about the end of life was considered insufficient by the patient.

Strengths and limitations

This study is the first mixed-methods study about ACP in ICD patients, from the perspective of relatives. Some limitations have to be considered. Selection bias may have played a role. It is possible that only relatives participated with either strong positive or negative experiences. This is partly accounted for by identifying relatives via medical records and the reasonable response rate. Also, recall bias should be considered, as we selected relatives of patients who had died between 2012-2017.

Implications

ACP conversations are aimed at eliciting preferences of ICD patients towards ICD deactivation. However, patients and relatives experience barriers towards having these conversations. Healthcare professionals should be attentive of these barriers and take the initiative to start the conversation. Since patients' willingness towards having these conversations may vary, and healthcare professionals themselves might experience barriers as well, educational programs for healthcare professionals to conduct ACP conversations should be developed and implemented[29]. Healthcare professionals have to be aware that patients and relatives might struggle with the idea of deactivation. ACP conversations with patients and relatives should therefore be carefully tailored to their willingness to engage in these, but should provide them with the information needed to formulate their future wishes.

Conclusion

ICD deactivation was discussed with a minority of patients. Half of the patients had their ICD deactivated, and up to a quarter received shocks in the last month of life, which were reported to be distressful for both patients and relatives. ACP conversations, tailored to the need of the patient, can help to make a timely and well-informed decision to deactivate the ICD, to avoid receiving distressful shocks at the end of life, and to promote a peaceful dying.

Declarations

Research ethics and patient consent

Before conducting this study, approval of the ethics committee of the Erasmus MC, Erasmus University Medical Center Rotterdam, was obtained (METC-2017-357). Participants were given both verbal and written information on the study and all provided written consent.

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SUPPLEMENTARY MATERIAL

Survey

1) Demographic characteristics of both respondent and patient		
1A) Patient		
#	Question	Answer categories
1	What was the date of birth of your relative?	Open question
2	On what date did your relative die?	Open question
3	Was your relative a man or a woman?	1. Man 2. Woman
4	What was the living situation of your relative at time of death?	1. At home, alone 2. With partner, no children 3. With partner and children 4. Nursing home / home care 5. Other, namely
5	What was the highest completed education of your relative?	1. Primary school 2. Lower vocational education 3. Pre-vocational secondary education 4. Secondary vocational education 5. Senior general secondary education / pre-university education 6. Higher professional education 7. University education 8. Other, namely
6	What was the nationality of your relative?	1. Dutch 2. Other, namely
7	Was your relative religious?	1. Yes, Catholic 2. Yes, Protestant 3. Yes, Islamic 4. No 5. Don't know 6. Other, namely
1B) Respondent		
8	What is your age?	Open question
9	What is your gender?	1. Male 2. Female
10	What was your relationship with your deceased relative? He/she was my:	1. Husband/wife/life partner 2. Son/daughter 3. Father/mother 4. Other, namely
11	What is your highest completed education?	1. Primary school 2. Lower vocational education 3. Pre-vocational secondary education 4. Secondary vocational education

		5. Senior general secondary education / pre-university education 6. Higher professional education 7. University education 8. Other, namely
12	What is your nationality?	1. Dutch 2. Other, namely

2) The last phase of life and death of your relative		
#	Question	Answer categories
13	In the last three months of his or her life, was your relative ever admitted to hospital?	1. Yes 2. No 3. Don’t know
14	What was the cause of death of your relative?	1. Heart disease 2. Lung disease 3. Cancer 4. Dementia 5. Don’t know 6. Other, namely
15	Where did your relative die?	1. At home 2. At the hospital 3. In a nursing home/home care 4. In a hospice 5. Don’t know 6. Other, namely
16	Was your relative, a few days before he or she passed away, aware that he or she would die soon?	1. Yes 2. Yes, sort of 3. No 4. Don’t know
17	Were you, a few days before your relative passed away, aware that he or she would die soon?	1. Yes 2. Yes, sort of 3. No 4. Don’t know
18	Was a decision made to not resuscitate?	1. Yes 2. No 3. Don’t know
19	Can you indicate for the following statements which best suits the health condition of your relative in <u>the last month</u> before death?	1. My relative was able to carry out all normal activities without restriction 2. My relative was restricted in physically strenuous activity, but mobile and able to carry out light activities 3. My relative was capable of self-care, but unable to carry out any other activities. In bed less than 50% of the day

		4. My relative was capable of only limited self-care, confined to bed more than 50% of the day 5. My relative was completely disabled, confined to bed all day

3) ACP conversations about the ICD		
#	Question	Answer categories
20	Have you ever had a conversation with your relative about turning off the ICD?	1. Yes, please comment 2. No
21	Did a healthcare professional, as far as you know, ever had a conversation with your relative about turning off the ICD?	1. Yes (go to #23) 2. No (go to #22) 3. Don't know (go to #22)
22	Do you think your relative had a need for this?	1. Yes, please explain (go to #30) 2. No, please explain (go to #30) 3. Don't know (go to #33)
23	When did this conversation take place? (multiple answers possible)	1. Before ICD-implantation 2. More than a month before death 3. In the last month before death 4. In the last week before death 5. On the last day before death 6. In the last hour before death 7. Don't know
24	Who took the initiative to talk about turning of the ICD?	1. My relative 2. A family member 3. Myself 4. The cardiologist 5. The GP 6. A nurse 7. Don't know 8. Other, namely
25	Who were present during this conversation? (multiple answers possible)	1. My relative 2. A family member 3. Myself 4. The cardiologist 5. The GP 6. A nurse 7. Don't know 8. Other, namely
26	In case your relative was not present during this conversation: What was the reason for this? (multiple answers possible)	1. He/she was too confused 2. He/she was unconscious 3. It was too stressful for him/her 4. Not applicable, he or she was present 5. Other, namely
27	Was your relative satisfied with this conversation, as far as you know?	1. Yes 2. No, please comment

		3. Not applicable, he or she was not present 4. Don't know
28	Were you satisfied with this conversation?	1. Yes 2. No, please comment 3. Not applicable, I was not present
29	During this conversation, was a decision made about whether or not to turn off the ICD?	1. Yes, the ICD would be deactivated 2. Yes, the ICD would remain active 3. No 4. Don't know 5. Other, namely

4) Deactivation of the ICD		
#	Question	Answer categories
30	Was the ICD turned off before death?	1. Yes (go to #31) 2. No (go to #35) 3. Don't know (go to #35)
31	How long before your relative passed away was the ICD turned off?	1. A few minutes before death 2. A few hours before death 3. A few days before death 4. A few weeks before death 5. More than a month before death 6. Don't know
32	Who turned off the ICD?	1. A doctor, what kind of doctor?: 2. A representative of the supplier of the ICD 3. An ICD-technician 4. Don't know 5. Other, namely
33	Where was the ICD turned off?	1. At home 2. In the hospital 3. In the nursing home/home care 4. In the hospice 5. Don't know 6. Other, namely
34	How was the ICD turned off?	1. With a magnet that was placed on the chest 2. With a computer 3. Don't know 4. Other, namely

5) Shocks of the ICD		
#	Question	Answer categories
35	Did your relative ever receive a shock from the ICD?	1. Yes (go to #36) 2. No (go to #41) 3. Don't know (go to #41)

36	Did your relative receive any shock during the last day of his/her life?	1. Yes, how many? (estimate) 2. No 3. Don’t know
37	Did your relative receive any shock during the last week of his/her life? (note: this is excluding potential shocks during the last day of his/her life)	1. Yes, how many? (estimate) 2. No 3. Don’t know
38	Did your relative receive any shock during the last month of his/her life? (note: this is excluding potential shocks during the last week of life)	1. Yes, how many? (estimate) 2. No 3. Don’t know
39	In case your relative received a shock during the last month of his/her life: What was the effect of these shocks on your relative? (multiple answers possible)	1. Pain 2. Fear 3. Sadness/gloom 4. Stress 5. Worry 6. Helplessness 7. Anger 8. Confusion 9. Don’t know 10. No effect 11. My relative did not experience shocks during the last month of his/her life 12. Other, namely
40	In case your relative received a shock during the last month of his/her life: What was the effect of these shocks on yourself? (multiple answers possible)	1. Fear 2. Sadness/gloom 3. Stress 4. Worry 5. Helplessness 6. Anger 7. Confusion 8. Don’t know 9. No effect 10. My relative did not experience shocks during the last month of his/her life 11. Other, namely

6) Wishes and preferences for the last phase of life		
#	Question	Answer categories
41	Did your relative have certain wishes or preferences regarding medical treatments in the last phase of life?	1. Yes (go to #42) 2. No (go to #45) 3. Don’t know (go to #45)
42	Did your relative discuss these preferences with someone? (multiple answers possible)	1. Yes, with me 2. Yes, with other family members 3. Yes, with a doctor 4. Yes, with a nurse 5. No 6. Don’t know 7. With someone else, namely
43	Did your relative recorded these preferences in writing?	1. Yes (go to #44)

		2. No (go to #45) 3. Don’t know (go to #45)
44	Did this written statement include anything about whether or not to turn off the ICD?	1. Yes, please explain 2. No 3. Don’t know
45	Did your relative appoint someone who could make decisions on his/her behalf should there come a time when he/she is no longer able to do this him/herself?	1. Yes 2. No 3. Don’t know

7) Statements		
Below you will find a number of statements about the last month of your relatives’ life. Tick the answer that best fits the situation of your relative and yourself. If you cannot answer a statement, you can tick ‘I don’t know’		
#	Question	Answer categories
46	Healthcare professionals adequately informed my relative and me about the disease and the prospects of my relative	1. Totally agree 2. Agree 3. Neither agree nor disagree 4. Disagree 5. Totally disagree 6. Don’t know
47	The way in which healthcare professionals discussed issues concerning the ICD in the last phase of life was good	1. Totally agree 2. Agree 3. Neither agree nor disagree 4. Disagree 5. Totally disagree 6. Don’t know
48	Healthcare professionals respected the wishes of my relative	1. Totally agree 2. Agree 3. Neither agree nor disagree 4. Disagree 5. Totally disagree 6. Don’t know
49	Healthcare professionals provided good emotional support to my relative and myself	1. Totally agree 2. Agree 3. Neither agree nor disagree 4. Disagree 5. Totally disagree 6. Don’t know
50	How do you rate the quality of death of your relative?	Scale from 1-10 (1 meaning very poor, 10 meaning very good)

8) After your relative died		
#	Question	Answer category
51	Did you have a follow-up conversation after the death of your relative?	1. Yes, please explain (end of survey) 2. No (go to #52) 3. Don’t know (go to #52)
52	Would you have liked to have had this conversation?	1. Yes 2. No 3. Don’t know
End of survey		


9

CHAPTER 9

Implantable cardioverter defibrillators at the end of life: Future perspectives on clinical practice.

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ABSTRACT

The implantable cardioverter defibrillator (ICD) is effective in terminating life-threatening arrhythmias. However, in the last phase of life, ICD shocks may no longer be appropriate. Guidelines recommend timely discussion with the patient regarding deactivation of the shock function of the ICD. However, research shows that such conversations are scarce, and some patients experience avoidable and distressful shocks in the final days of life. Barriers such as physicians' lack of time, difficulties in finding the right time to discuss ICD deactivation, patients' reluctance to discuss the topic, and the fragmentation of care, which obscures responsibilities, prevent healthcare professionals from discussing this topic with the patient. In this point-of-view article, we argue that healthcare professionals who are involved in the care for ICD patients should be better educated on how to communicate with patients about ICD deactivation and the end of life. Optimal communication is needed to reduce the number of patients experiencing inappropriate and painful shocks in the terminal stage of their lives.

The implantable cardioverter defibrillator at the end of life

The implantable cardioverter defibrillator (ICD) is highly effective in terminating potential life-threatening arrhythmias. However, for patients at the end of life, the goal of the ICD – prolonging survival – may no longer be appropriate. Guidelines recommend early and regular discussion of the appropriateness of the ICD shock function throughout the disease trajectory and subsequently deactivation of the ICD at the end of life[1, 2]. The Dutch Association of Cardiology (NVVC) released a guideline in 2013[3] discussing the implications of the ICD at the end of life as well as the indication for and consequences of deactivating the device, but above all stresses the importance of discussing deactivation. Such discussions could be initiated before implantation (supported by a written patient folder), at follow-up (e.g. changing health, ICD battery change), and when entering the palliative and terminal phase. A recent Dutch study showed that only 35% of deceased ICD patients had discussed ICD deactivation. Further, 42% of patients had their ICD deactivated, and 9% experienced shocks in their last month of life[4], which have been reported as distressing for both patients and relatives[5]. In this point-of-view article, we discuss the barriers experienced by Dutch healthcare professionals to discussing ICD deactivation and provide future perspectives on clinical practice.

Difficulties in discussing ICD deactivation

It is important that discussions on ICD deactivation are conducted before the actual deactivation[4]. However, healthcare professionals struggle with such discussions, while many patients indicate that they want to be informed about this topic[6]. Some research has described the potential barriers healthcare professionals experience to discussing ICD deactivation[7-9]. These include having too little experience or knowledge with regard to talking about deactivation[10-16], not feeling comfortable discussing the topic[14, 17-20], a predominant focus on cure and on the benefits of ICD therapy[10, 12, 18, 21, 22], having too little time to hold this conversation[12, 18, 19, 21], uncertainty about predicting patients' disease trajectory[10, 12, 13, 21], being afraid of taking away hope[10, 12, 13, 19, 21], not knowing at what stage this conversation is appropriate[10], not knowing who is responsible for these conversations[10, 15], a lack of multidisciplinary cooperation[18], and a stressful work environment and high workload[23]. However, almost all of these studies were conducted in the USA, and no research has yet been performed in the Netherlands. Given the open culture in the Netherlands with regard to end-of-life decision-making[24], it is remarkable to see that ICD deactivation discussions do not occur often.

Barriers hindering Dutch healthcare professionals from discussing ICD deactivation

We examined the experiences of Dutch healthcare professionals with ICD deactivation discussions in focus group meetings and individual interviews. Healthcare professionals were recruited via e-mail in an academic and non-academic hospital and subsequently by using a snowball approach. Participants had to have experience in care for ICD patients at the end of life. Eighteen healthcare professionals participated in individual interviews, and two focus group meetings were held with four participants each. Interviews and focus groups were conducted between October 2017 and January 2018 using a predefined semi-structured topic list. Focus groups were led by experienced moderators (A.v.d.H. and J.A.C.R.), and the interviews were conducted by a psychologist (R.S.). Data were analysed using the constant comparative method. The characteristics of the participating healthcare professionals can be found in Tab. 1.

Table 1. Characteristics of participating healthcare professionals (*n* = 26)

Male gender	14 (54%)
Age (mean, SD)	47.0 (11.3)
Years of experience (mean, SD)	14.1 (8.5)
Profession	
- Physician[a]	11 (42%)
- Nurse[b]	11 (42%)
- ICD technician	4 (15%)
Work place	
- Non-academic hospital	12 (46%)
- Academic hospital	8 (31%)
- Hospice	3 (12%)
- General practitioner office	2 (8%)
- Care home	1 (4%)

ICD Implantable cardioverter defibrillator

[a] 4 cardiologists, 4 elderly care physicians, 2 general practitioners, 1 oncologist

[b] 5 cardiology nurses, 4 nurse specialists, 2 ICD nurses

All healthcare professionals reported to experience some barriers to discussing ICD deactivation. Perceived barriers were related to clinical practice, the patient, or societal factors (Tab. 2). A frequently reported barrier relating to clinical practice was experiencing a lack of time to discuss this topic. A nurse specialist in palliative care observed: ‘It calls for a different planning of consultation hours, because those conversations take more time, and the consultation hours are not really designed for that’. Other healthcare professionals described having a lack of knowledge about the ICD and the end of life, such as two elderly care physicians, who indicated that they were ‘unconsciously incompetent.’ A cardiologist pointed out that it is difficult to find the right moment to discuss ICD deactivation, and that it might be inappropriate to discuss this topic during routine appointments. However, the pre-implantation conversation held with all patients was also regarded as inappropriate by some, who stated that this conversation should focus on practical aspects of the ICD. Another barrier reported was the fragmentation of healthcare and the predominant focus of healthcare professionals on their own discipline, which sometimes leads to overlooking the patient as a person.

A nurse specialist remarked: ‘The cardiologist only looks at the heart, the pulmonologist only looks at the lungs… everybody is looking at his own small piece… we need to look at the patient as a whole. And that is sometimes not done’ . Other barriers related to clinical practice were concerns about taking away patients’ hope, difficulties in predicting the disease trajectory, a lack of experience in conducting such conversations, a lack of (awareness of) guidelines (only nine healthcare professionals mentioned the guideline of the NVVC), protocols which focus only on medical issues, and frequent staff turnover.

Healthcare professionals also experienced barriers related to the patient: they felt that patients are reluctant to discuss ICD deactivation and the end of life. Some argued that patients tend to overestimate the life-saving ability of the ICD and think that, if the ICD is deactivated, they will immediately die. Also, many patients do not seem to think the topic is relevant yet. At the start of the treatment, patients are focussed on living and the practical implications of the ICD. Later on in the disease trajectory, however, many patients are still not thinking about the end of life. A cardiologist compared this with retirement income: *‘There are certain things, and the same goes for retirement income, we know it is important, but did you ever delve into how much you will actually receive?… It is very difficult to motivate yourself to think about that in depth’.*

Attitudes in society towards death and dying were also mentioned as barriers to discussing ICD deactivation. Several clinicians indicated that medicine and society are predominantly focussed on treating and curing illness, and length of life is often viewed as being more important than quality of life. An oncologist described*: ‘Death should be a much more integral topic during life. We all want to be young forever, have no wrinkles, and whatever… We want to overcome everything, overcome cancer, cancer out of the world… it is nonsense… We get cancer. It is part of our lives’.* A nurse specialist in palliative care said: *‘We can do everything, but not everything we can do is always appropriate. When you are 92, do you have to have a new aortic valve or a new ICD? And another, and another? How realistic is that?’*. It was pointed out that no one can live forever, and that we need a different approach towards death and dying.

Table 2. Barriers for healthcare professionals towards discussing deactivation of the implantable cardioverter defibrillator

Clinical practice
Lack of time
Lack of knowledge about ICD in last phase of life
Difficulty finding the right moment to discuss deactivation
Lack of communication/coordination between healthcare professionals
Little insight into what other healthcare professionals do
Focus on practical matters
Focus on own discipline (fragmentation)
Being afraid to take away patients’ hope
Focus of cardiology on saving lives

Difficulty predicting patients’ disease trajectory
Feeling uncomfortable discussing last phase of life
Lack of experience discussing last phase of life
Focus on life-saving potential of ICD
Too little education on last phase of life, palliative care, and communication
Lack of (awareness of) guidelines
Uncertainty about who is responsible
Poor relationship with the patient
Lack of facility for a calm conversation
Difficulty stopping treatments
Protocols focus on medical aspects
Too much staff turnover
Patients
Reluctance to discuss/think about topic
Last phase of life not yet relevant/focused on practical matters
Overestimating life-saving character of ICD
Lack of knowledge about deactivation
Young age of patient
Topic too emotional
Culture/religion
Association with euthanasia
Lack of knowledge about what is medically possible
Society
Medicine/society focused too much on treatment/cure

Future perspectives

Despite guidelines on how to adequately address deactivation of the ICD shock function at the end of life, many patients never discuss ICD deactivation and die with an active ICD, some after experiencing painful shocks[4-6]. Palliative care, the end of life and advance care planning[25] are atypical subjects in the highly technological field of cardiology, but are of great importance[26]. However, healthcare professionals experience barriers to discussing these topics. Action is needed to increase attention to these topics and to overcome barriers. Educating healthcare professionals about the importance of discussing ICD deactivation and the last phase of life is needed, and can increase their involvement in advance care planning[27, 28].

A recent UK study by Javaid and colleagues reported on an easy-to-implement but effective programme to improve the attention given to ICD deactivation[29]. This programme encompassed: (1) raising awareness and increasing knowledge about ICD deactivation by presenting research and guidelines to different medical departments; (2) e-mailing all staff who were not able to be present during these presentations; (3) developing and distributing informative posters about the ICD and end-of-life care on medical wards; and (4) offering teaching and checklists to staff working on medical wards. After implementation of this programme, ICD deactivation increased from 0 to 54% and none of 13 patients studied experienced painful shocks during the last month of their lives. Although this study was small, the results are promising. Healthcare professionals in the Netherlands taking care of ICD patients should critically review how care at the end of these patients' lives is organised. To further facilitate and initiate advance care planning discussions, consultation schedules and the role of nurses might be revisited, since they might be well suited to initiate discussions with patients regarding the future role of their ICD[18, 21, 30]. Advance care planning and discussions about end-of-life care should become an integral part of cardiological care. Only then can we reduce the number of patients experiencing inappropriate and painful shocks at the end of their lives.

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CHAPTER 10

General discussion

The overall aim of this thesis was to provide insight into self-management behaviours and advance care planning (ACP) practices of patients living with an advanced illness, by studying patients with advanced cancer and patients with an implantable cardioverter defibrillator (ICD). In this general discussion, the main findings of this thesis are described and interpreted, methodological considerations are discussed, and recommendations for clinical practice and future research are given.

MAIN FINDINGS

Part I

In part I of this thesis, self-management was examined in a population of patients with advanced cancer. The aim was to better understand the consequences of having advanced cancer and patients' self-management to deal with these consequences. Further, the aim was to examine healthcare professionals' experiences and attitudes towards patients' self-management, and their perceived roles in self-management support. Self-management was defined in our studies as "*the strategies used by persons with the aim of managing the physical, psychosocial and existential consequences of living with a progressive, life-threatening disease and its treatment"*(1).

Research question 1: How do patients with advanced cancer deal with the consequences of their illness and its treatment?

Patients with advanced cancer who participated in interviews about self-management described a large number of consequences of living with advanced cancer and its treatment in multiple domains, such as the physical domain, the psychological domain, the social domain, the existential domain, the medical domain, and the domain of daily life. In our systematic review of the international literature and in our interview study with patients with advanced cancer, a large number of self-management behaviours were identified (Table 1). As a whole, these self-management behaviours can be synthesized into six categories: health behaviours; psychological strategies; behaviours related to seeking and providing social support; behaviours related to collaborating with healthcare professionals; behaviours related to obtaining knowledge and information; and behaviours related to planning. Self-management behaviours were often used in multiple domains. For instance, planning was used to organize medical appointments, but also to organize household activities. Also, self-management behaviours were used complimentary in the same domain. For instance, in order to deal with sadness, patients used health behaviours, such as staying physically active, but also psychological strategies, such as keeping a diary. Patients generally tried to live as normally as possible. However, some struggled with this, since their illness largely dictated their agenda. They indicated to spend, for instance, much time on adhering to sometimes complex medication schemes and coordinating their medical appointments.

Table 1. Self-management behaviours of patients with advanced cancer

Category of behaviour	Self-management behaviours
Health behaviours	• Dieting • Monitoring symptoms • Relaxation • Self-administering and adjusting medication • Staying physically active • Using complementary and alternative treatments
Psychological strategies	• Accepting, ignoring or downplaying illness • Keeping a diary • Practicing religion • Seeking distraction • Using mindful self-help strategies
Behaviours related to seeking an providing social support	• Limiting social interactions • Providing social support to others • Seeking support from family, friends or others • Volunteering in patient associations
Behaviours related to collaborating with healthcare professionals	• Engaging in ACP • Investing in relationship with healthcare professional • Making decisions about treatment • Seeking support from healthcare professionals
Behaviours related to obtaining knowledge and information	• Avoiding or neglecting information • Coordinating information dissemination to relatives • Seeking information about illness, treatment and self-care on the internet
Behaviours related to planning	• Adhering to medication schemes • Coordinating medical services • Dividing and planning activities over the day • Maintaining normal daily routine • Making financial and practical plans • Planning moments of rest • Short-term goal setting

Research question 2: What factors influence self-management of patients with advanced cancer?

In our review of the international literature on self-management, different factors that might influence self-management of patients were identified, such as age: younger patients performed more physically and socially active self-management behaviours; educational level: higher educated patients were more proactive and reported a higher self-perceived effectiveness of their self-management; and self-efficacy: patients with higher levels of self-efficacy perceived their symptoms more often as modifiable. The studies in our review were however non-experimental studies, with moderate sample sizes, some extent of selection bias, and limited adjustment for confounding, due to which the results should be interpreted with caution. In our qualitative interview study, we found no evidence that age, gender or educational level affected patients' self-management. Rather, we found that self-management is influenced by patients' personality, their life history, the moment in the illness trajectory and the involvement of their social environment. Most patients indicated not to be aware of *why* they dealt with consequences in certain ways, and indicated they 'always did things like this'. This suggests that patients often draw from their usual self-management behaviours, and try to apply and adjust these to their new situation of dealing with advanced cancer.

Research question 3: What are the experiences and attitudes of healthcare professionals towards self-management of patients with advanced cancer, and what self-management support roles can be distinguished?

Healthcare professionals who participated in our interview study experienced patients' self-management as highly individual and diverse. Most healthcare professionals where optimistic about patients' self-management abilities and behaviours, although some mentioned concerns about the patients who seemed less able to engage in self-management. Healthcare professionals adopted different roles in supporting patients' self-management. These were: the instructive role, in which the healthcare professional is leading, and directs patients' self-management based on their own professional expertise; the collaborative role, in which a well-delineated division of tasks between patient and professional is apparent and both patient and healthcare professional directed strategies are integrated; and the advisory role, in which the patient is leading, and a more patient-directive strategy is apparent, in which the healthcare professional is more 'serving' the patient. The role of the healthcare professional was dependent on their working conditions and clinical experience, their personality and life experience. Some healthcare professionals preferred or inclined towards one of the three roles, although mostly they seemed to switch between these, based on the needs of the patient.

Part II

In part II of this thesis, advance care planning (ACP), an expression of self-management which is very relevant for patients with advanced illness, was examined. ACP is commonly defined as *"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"*(2). ACP was examined in a population of patients with an ICD. The aim of this part was to examine the last phase of life of patients with an ICD, and to examine the experiences of patients, relatives, and healthcare professionals with ACP conversations about whether or not to deactivate the shock function of the ICD.

Research question 4: What is the impact of having an active ICD in the last phase of life?

In our review of the international literature, up to 33% of patients who died due to non-sudden causes experienced shocks from their ICD on the last day of life, and up to 14% in the last hour of life. Up to 68% of the patients who died due to sudden causes experienced shocks on the last day of life and up to 66% in the last hour of life. Studies that did not differentiate by cause of death showed that up to 32% of patients experienced shocks in the last month of life, up to 32% on the last day of life and up to 31% in the last hour of life. In the medical record study in which we examined the records of 380 deceased ICD patients, 70% of patients died with an active ICD. In 7% of these patients, their medical record included reports about shocks in the last month of life. However, since most patients had died outside the hospital, we assumed that the shock incidence might be higher, and reports of bereaved relatives might possibly be more accurate. This was examined in our mixed-methods study in which 170 relatives of 154 deceased ICD patients participated. They reported that of all patients, 24% had experienced a shock in the last month, and 10% on the last day of life. Experiencing shocks in the last phase of life had a negative impact on both patients and relatives. Shocks were distressing for 74% of patients and 73% of relatives. Most often reported sources of distress were pain and fear in patients, and fear and worry in relatives.

Research question 5: How and when are decisions about ICD deactivation made, and what factors influence these decisions?

ICD deactivation was discussed with 35% of patients who had died between 2013 and 2016, based on the notes in the medical records of these patients. This was comparable to what bereaved relatives reported in our mixed-methods study: 38% of patients discussed ICD deactivation with their healthcare professional. These discussions occurred late in the illness trajectory, often in the last week of life. Only 11% of patients had discussed deactivation during the pre-implantation conversation. In the medical record study and in the mixed-method study among relatives of deceased ICD patients, it was found that between 30% and 55% of ICD patients had their ICD deactivated before death. The ICD was deactivated late in the illness trajectory, most often in the last days of patients' lives. Patients who had had a conversation about ICD deactivation were 69 times more likely to have their ICD deactivated compared to patients who did not have this conversation. Other factors that positively influenced the decision to deactivate the ICD were having a do-not-resuscitate or do-not-intubate order, and the involvement of a palliative care team.

Research question 6: What barriers do patients and relatives experience towards ACP conversations about ICD deactivation?

Many ICD patients in the focus groups indicated to be interested in having ACP conversations about ICD deactivation with their healthcare professional, although few patients had had this conversation. Patients described that their healthcare professionals were knowledgeable about many medical issues, but some patients indicated there was not always the opportunity to discuss topics with a strong emotional component, such as the end of life. Preferences about the timing of these conversations were varied: some wanted this conversation during the pre-implantation phase, others would want this when ICD deactivation becomes more directly relevant. In our focus groups with relatives of deceased ICD patients, numerous barriers towards having ACP conversations about ICD deactivation were identified. Some of the barriers were related to the patient and relatives, such as a lack of willingness to discuss ICD deactivation, thinking the topic is not relevant yet, having a lack of knowledge about the functioning of the ICD and having a disturbed relationship with the healthcare professional. Other barriers were related to the healthcare professional and the healthcare system, such as a perceived lack of knowledge of the healthcare professional about the functioning of the ICD, a perceived lack of time of the healthcare professional to engage in ACP conversations, and a lack of patient-centeredness.

Research question 7: What barriers do healthcare professionals experience towards ACP conversations about ICD deactivation?

Healthcare professionals who were interviewed and participated in focus groups about ICD deactivation experienced several barriers towards having ACP conversations about ICD deactivation with their patients. Barriers were experienced on three levels: their clinical practice, the patient and society. Most often reported barriers on the clinical practice level were having a lack of time and knowledge about the ICD and the last phase of life, and having difficulties finding the right time to start an ACP conversation. On the patient level, healthcare professionals sometimes experienced reluctance of the patient towards discussing the topic, or thought the last phase of life was not yet relevant to discuss. On the societal level, healthcare professionals indicated that medicine in general and society is too much focused on curing patients. They also noticed that healthcare professionals were often mainly focused on their own discipline, which could lead to fragmentation of care and uncertainty about who is responsible for conducting ACP conversations.

METHODOLOGICAL CONSIDERATIONS

A variety of methodologies were used to examine the research questions outlined in the introduction. **Chapter 2 and 5** both described literature reviews (of which one systematic and one integrative). **Chapter 3, 4, 7 and 9** described studies with a qualitative design (of which three individual interview studies and one focus group study). **Chapter 6** described a medical record study. **Chapter 8** described a mixed-methods study, including a survey and a qualitative focus group study. These methodologies will be briefly discussed in this section. The variety of research methods used is a strength of the studies in this thesis. Self-management and ACP were first examined by summarizing all existing literature in two reviews, and subsequently by using quantitative and qualitative research methods. The quantitative studies provided us with clear numbers, while the qualitative studies provided us with a detailed and in-depth understanding of the studied concepts(3-5). Especially with issues that are difficult to describe or understand in numbers, and largely depend on the subjective experiences and attitudes of those involved, such as self-management and ACP, qualitative methods are ideally suited to investigate this(6). This meticulous approach expands our knowledge and provided us with a deep understanding of self-management and ACP in the two specific populations.

Reviews

A potential limitation of the reviews in **chapter 2** and **chapter 5** is that some studies might have been missed, for instance because they were not indexed in the scientific databases that were searched, because certain keywords were missed, or because studies were published in languages other than English. In **chapter 2**, we used a broad definition and a variety of search terms for ‘self-management’, but it is possible that we missed research that used different terms for the same concept. In this review, the included studies were often of moderate quality. Studies were especially affected by selection bias. Little information was provided on inclusion and exclusion criteria and response rates. It is possible that the included studies largely involved patients who were more than the average patient able and willing to engage in self-management, thereby impairing the generalizability to a larger population. In the review on ICD shock incidence in **chapter 5**, the included studies had relatively small study populations and were relatively old, often conducted more than 20 years ago. In recent years, we have seen large improvements in ICD technology, such as antitachycardia pacing (ATP), longer detection times and high rate cut-offs(7, 8). Also, ICDs are currently more often implanted in patients for primary prevention instead of secondary prevention, and these patients have a lower risk of recurrent ventricular tachycardias(9, 10). Lastly, there is increasing attention for ICD deactivation(11). This could mean that the current shock incidence is lower than reported in our review.

Qualitative studies

In **chapter 3, 4, 7, 8 and 9**, qualitative research methods such as interviews and focus groups were used. Such methods rely on the subjective experience of the participants, and caution is advised in generalising the results to a larger population(3, 4, 12). Participation bias should be considered, for instance in **chapter 3**, **7** and **8** in which participants were recruited via calls for participation in magazines, websites and social media channels. This might mean that only people enrolled with a special interest in the topic or with negative experiences. Recall bias should also be considered, for instance for the interview study reported on in **chapter 8**, in which relatives of deceased patients participated. It might be possible that participants were not always able to remember important information. Generally, the longer the time between the death of the patient and the timing of the interview, the more information is lost(13). However, past experiences that are significant to the respondent or have a long duration are usually less prone to recall bias(14). The death of a loved is such an experience, and therefore, recall bias may be minimal in our studies that rely on the relatives' memory, such as in **chapter 8**.

Medical record studies

In **chapter 6**, the results of a retrospective study of medical records were described. In this study, medical records of patients who had their ICD implanted from 1999 onwards were examined. Although this is an effective method to examine ICD deactivation and ICD shocks, some limitations have to be considered. Many medical records consisted of a large amount of scanned, handwritten notes from healthcare professionals, which were sometimes hard to read and poorly organized. Also, it might have been possible that medical records were incomplete, which could also have led to an underreporting of the studied variables(15). Although the data was meticulously collected, it is possible that some information was missed, especially concerning the occurrence and exact content of ICD deactivation discussions with the patient. This could mean that in reality, ICD deactivation discussions, ICD deactivation, and shocks occurred more often than we could demonstrate. Also, in the patient sample in **chapter 6**, more than half of the patients died outside the hospital, and the last days of life were not always recorded in the medical record. We therefore sometimes missed data from the last months of life of these patients. This was partly accounted for by contacting the GP of these patients, although not all GPs were able to complete this information. Lastly, patients' medical records in only two hospitals were examined. In order to gain a broader insight in the studied variables, replication is necessary in other hospitals in the Netherlands and beyond.

Survey

In **chapter 8**, the results a mixed-methods study were reported, in which relatives completed a survey. Relatives were recruited via the hospital medical records of deceased patients. However, a large number of telephone numbers were no longer used or were incorrect. Nonetheless, we had a reasonable response rate in this study (59%), thereby reaching a large group of relatives. Relatives most often were the spouse of the deceased patient and were closely involved in the last phase of life of the patient, thereby providing us with first-hand information. However, recall bias should also be considered. It might be possible that relatives were unable to recall certain events that found place in the last months of life of their loved ones.

INTERPRETATION AND INTEGRATION OF THE FINDINGS

Living with a progressive, life-threatening illness

The vast and hard work of being ill

Having a progressive, life-threatening illness, such as cancer or cardiovascular disease, greatly disrupts patients' lives. In the interview study presented in **chapter 3**, patients described experiencing numerous consequences of having advanced cancer and its treatment on many domains of life. They experienced consequences in the physical, psychological, social and existential domain, but also in the medical care domain and in their daily living activities. Previous research, as summarized in the systematic review described in **chapter 2**, showed similar consequences. Patients engage in numerous activities to deal with the consequences of their advanced illness. These activities are diverse and individually determined, and are influenced by the patients' personality, their life history, the phase of illness and the involvement of the social environment. Other studies that used quantitative research methods also showed that age, level of physical functioning, income, education, self-efficacy and symptoms of anxiety and depression influence the patient's self-management(16-19). Mostly, patients in our studies indicated to be able to effectively deal with their illness and its consequences. However, some patients indicated to sometimes struggle with self-management, which was also confirmed by some of the healthcare professionals who participated in our interview study about self-management (**chapter 4**). These patients experienced a high treatment burden. Treatment burden refers to the work patients have to perform in order to organize and coordinate their care, to adhere to treatment regimens, and to self-manage their symptoms, next to the demands of everyday life(20). A high treatment burden leads to the patient feeling overwhelmed, noncompliance to treatment, poorer health outcomes, and an impaired quality of life(20-23).

Living life as normally as possible, for instance by continuing to practice hobbies or usual daily living activities, was frequently mentioned as important in the interviews by patients with advanced cancer. Some patients had difficulties with this. These patients indicated that their illness and its treatment largely dictated their agenda, and said they were sometimes approached differently by others. The patients with an ICD who participated in our focus groups had similar experiences, and indicated the need to live their lives as normally as possible. Other research also showed that patients want to continue living their normal life with reasonable independence, and do not always want to be approached as a patient(24). However, for some patients, living a normal life is threatened by the medical work they have to perform. As Corbin and Strauss postulated, and as was described in **chapter 1**, managing an illness requires ‘medical work, daily life work, and biographical work’(25). When patients’ lives are overwhelmed with medical work, patients are hampered in living their usual daily lives. Concluding, having a progressive, life-threatening illness is hard work which potentially threatens patients from living their normal lives(1, 26). Patients should be supported and empowered in making sure they are able to deal with the consequences of their illness(27).

Living in the ‘here and now’ versus preparing for the future

As described in the general introduction in **chapter 1**, ACP can be considered an expression of self-management. ACP consists of three key elements: 1) reflection on wishes and preferences for future medical treatment and care; 2) discussing these with healthcare professionals and relatives; and 3) recording and, if necessary, reviewing these. In other words, ACP can help patients to prepare for future healthcare decision-making. But are patients willing to engage in ACP? In our systematic review and interview study on patients with advanced cancer (**chapter 2 and 3**) and our interview study with patients with an ICD (**chapter 7**), we saw a variety in the needs and willingness to engage in ACP. Some patients were actively thinking about the end of life and future healthcare preferences, and were interested in discussing these with healthcare professionals, although ACP conversations did not occur often (**chapter 5 to 9**). Other patients were more reserved towards thinking about future treatment and care and the end of life. They were focussed on living in the ‘here and now’, and not on preparing for the end of life. Other studies also found that patients with a progressive illness are sometimes ambivalent towards discussing the end of life and ACP(28, 29). Moreover, they sometimes seem to fluctuate between actively thinking about their illness and future deterioration on the one hand, and ignoring it on the other hand(30). This variety in willingness to engage in thinking about the end of life could hamper actual ACP conversations, since it can make healthcare professionals uncertain about when to start this conversation(31). An important element of ACP is to explore the needs and readiness of patients to engage in ACP(2). Research showed that ACP is only beneficial when patients are ready to engage in this(32). However, there is also evidence that, even if patients do not seem ready to have this conversation, they in hindsight agree that the conversation was beneficial(32). This means that healthcare professionals should not be hesitant to broach this topic when patients do not seem to be fully ready to engage in ACP, especially since the ACP process itself could promote patients' readiness to engage in ACP(29). The fact that patients are ambivalent towards having this conversation should not mean that this conversation should not occur. Actually, the fact that something is difficult or uncertain should be a trigger for exploration instead of avoidance(33).

Working together to promote self-management and ACP

How does the healthcare professional appreciate patients' self-management?

The healthcare professionals we interviewed about self-management (**chapter 4**) were generally positive about their patients' self-management, but also worried about the patients who seemed less able to adequately deal with the consequences of their illness. They indicated that some patients are unwilling or not able to deal with their impaired health, and that some patients may be disadvantaged by the shift away from traditional, paternalistic medicine towards a model in which patients are more in charge of their health. Some healthcare professionals indicated that patients struggle with the medical management of their illness. A nurse who was interviewed for instance described how a highly educated man was given the flexibility to determine the amount of pain medication he needed. However, this patient struggled with doing so, since he was not able to decide what to do. It has been shown before that the medical management of an illness is something patients may struggle with, especially when their condition deteriorates, or when they become more bedbound towards the end of life(1, 24, 34). In a large study among 2078 Dutch healthcare professionals, patients and policymakers, it was also found that healthcare professionals are sometimes sceptical about self-management(35). This study showed that 42% of participants doubted whether all patients are capable of self-management, and another 22% doubted whether patients want to take up this responsibility(35). In our interview study among healthcare professionals about self-management (**chapter 4**) and in previous research(36, 37), we saw variations in patients' self-management and self-management support needs. Patients often describe a significant need for support in self-management of the physical consequences of their illness, such as managing symptoms and medications(26, 37, 38). Other studies also showed that patients have a need to be supported in managing the psychosocial consequences of their illness and treatment(24, 37, 39). These self-management support needs should be frequently revisited throughout the illness trajectory.

The importance of the patient – healthcare professional relationship

The participants in our studies indicated that having a good relationship with the healthcare professional is essential, for instance when having ACP conversations, making treatment decisions, or when talking about delicate issues such as the end of life. Healthcare professionals were also aware of the importance of a good patient-healthcare professional relationship (**chapter 4 and 9**). In our interviews with patients with advanced cancer (**chapter 3**), we found that the patient's personality and life history are important factors influencing one's self-management. In order to obtain a clear picture of patients' self-management and self-management needs, it is thus important for a healthcare professional to look 'behind' the illness, and to approach the patient as a whole. This can only be accomplished when having a good relationship based on trust, which is something that should be invested in(40).

In the interview study in which healthcare professionals were interviewed about self-management (**chapter 4**), three roles of self-management support were identified: an instructive role, in which healthcare professionals try to direct and stay in charge of patients' self-management; a collaborative role, in which the professionals and patients work together, but in which the healthcare professional also expects patients to rely on their medical expertise; and an advisory role, in which the patients are more in charge and the healthcare professionals have a more 'serving' role. Similar roles were described by Van Hooft in 2015, who specifically looked at the roles of nurses(41). It is important to note that none of the roles is always 'the best' option. Different patients have different needs, which may also change over time. Most important is looking for a division of tasks that best suits the patient. Most healthcare professionals who participated in the focus groups about self-management were able to do so, and indicated to switch between roles if necessary. However, healthcare professionals sometimes experience difficulties towards supporting self-management and having ACP conversations (**chapter 4 and 9**). Especially concerning ACP, some healthcare professionals experience barriers. Healthcare professionals can be hesitant to talk about difficult topics such as the end of life, especially in medical specialties that are mainly focused on curing illness rather than on providing comfort care(33, 42, 43).

Patients and healthcare professionals have to work together. Creating a mutual participating relationship enhances patients' adherence to treatment regiments, improves health outcomes and increases patients' satisfaction with healthcare(44-46). Further, it reduces hospitalizations and stress and increases patient empowerment and self-management(45, 47). Especially in progressive illness, where patients and healthcare professionals often have a long-term treatment relationship, they are mutually interdependent: the healthcare professionals use their clinical and scientific expertise to treat and support their patients, while the patient's own experiential knowledge could provide important clues for further treatment and can complement the knowledge of the healthcare professional(48, 49). Establishing a good relationship and integrating both the healthcare professionals' and patients' knowledge is vital in making treatment decisions that best suit the patients(49).

Self-management, ACP, and COVID-19

At the time of completing this thesis, the corona virus is spreading across the globe, resulting in a world-wide pandemic. The corona virus causes the infectious disease COVID-19. Around 80% of people recover from the illness, which causes fever, dry cough and tiredness, without needing hospital treatment. However, around 20% of people become seriously ill and develop a difficulty with breathing(50). Due to the strict measures taken, the impact of the pandemic on daily life is large. Measures such as closing down schools, restaurants, limiting non-essential medical visits, and social distancing prevent people from living their usual daily lives(51, 52). Self-management is important to adjust to this new situation, such as working from home, ordering groceries and staying in contact with friends or healthcare professionals via video calling.

For frail people or those with progressive, life-threatening illness, the consequences of the pandemic are even greater. Early reports show that patients with comorbidities such as cardiovascular disease or cancer are at greater risk for developing advance respiratory disease and death, probably due to a less rigorous immune response(53). Also, patients with cancer have an increased risk of needing mechanical ventilation, being admitted to the intensive care unit, and death associated with COVID-19(54, 55). This has a large impact on the sometimes already complex self-management of patients' progressive illness, for instance in staying in contact with healthcare professionals, maintaining treatment regimens, and making decisions about potentially postponing treatment(56). Early qualitative studies of patients with cancer show that they are worried about contracting the virus and developing severe complications(57). Further, social isolation is a problem for these patients. Patients with advanced cancer often already experience feelings of social isolation, and COVID-19 further disrupts their social lives(58). Patients with progressive illness are more confined to their homes compared to the general population, and are recommended to only be exposed to their family, thereby limiting their social interaction(59), while especially for these patients, social interaction is often valued as being very important(22). Concerning ACP, there are discussions in the media and research about its importance, since some patients rapidly deteriorate upon contracting the virus(60, 61). As a response, several guidelines are developed specifically aimed at ACP in patients with COVID-19 or those that are more vulnerable to contract the virus(62, 63). Although social distancing and remote consultation makes ACP challenging, COVID-19 also creates opportunities. ACP conversations do not occur often, and the pandemic may provide a trigger to initiate these discussion(64). Whether these conversations are conducted face-to-face or via remote consultations, most important is that these are conducted in the first place(61). Several online decision aids were developed and are readily available(65, 66).

The COVID-19 pandemic shows the importance, but also the complexity of self-management and ACP. Everybody is introduced to what it is like to rigorously manage their daily life, maintain normality as much as possible, and deal with the challenges involved. For all people, but especially for those living with progressive illness: self-management and ACP are essential, and should be promoted and supported.

RECOMMENDATIONS FOR CLINICAL PRACTICE

Self-management should be individually supported

Self-management support for patients with advanced cancer should include an individualised approach that re-evaluates patients' needs and wishes, which is embedded in solid partnerships with the relatives and healthcare professionals, and which is incorporated in existing models of care. An individualised approach is important, since patients show different needs in the support they want to receive. For instance, some patients indicated to struggle with different aspects of their medical management, such as planning appointments and adhering to treatment regiments (**chapter 3**), while other patients indicated a need for more psychosocial support (**chapter 7**).

Frequent 'self-management check-ups' should be considered

Frequent check-ups by healthcare professionals on how patients are managing their illness, in which ACP is incorporated as well, are appropriate. These check-ups could be conducted by a nurse from the main treating department. This was supported by a study in which 262 physicians and nurses participated. In this study, 81% of healthcare professionals thought that nurses have the necessary skills to conduct such conversations(67). The nurse could play a central, coordinating role in the illness trajectory of the patients and should frequently, in collaboration with the patients, review their self-management, expectations, responsibilities, needs and preferences, and refer the patient to other healthcare professionals if needed, such as the cardiologist, oncologist, palliative care physician or psychologist(67-69). The array of tasks, responsibilities and the needs of the patient should be critically reviewed during these check-ups, rather than assuming the patient is able to self-manage(70).

Teach patients general self-management skills

Patients could benefit from receiving general education about self-management. Numerous self-management support programmes have been described within the literature benefiting the patients(71, 72), for instance in terms of illness related knowledge(73), performance of self-management behaviours(71, 74), and in decreasing symptoms such as fatigue(75) and pain(74), while increasing their self-reported quality of life(76). We recommend to provide patients with a program aimed at education and self-management early in the illness trajectory, integrated into routine practice(77-79). An example of such a program is the 'PEP'-program which is used in cardiac rehabilitation. This program includes four sessions about different topics, such as dealing with stress, emotions, but also resuming household activities, work, and maintaining the relationship with significant others(80). During the illness trajectory, self-management support can be adjusted based on the specific needs of the patient on that point in time.

Consider using eHealth in supporting patients' self-management

eHealth could play a valuable role in supporting self-management. The use of eHealth, such as apps, can be beneficial for patients receiving palliative care(81). One example is an app in which patients register their cancer-related pain, and can have direct contact with a nurse when the patient thinks this is appropriate(82). A meta-analysis showed that the use of eHealth interventions increases the illness-related knowledge of patients, their perceived support and healthcare participation(83). Another recent scoping review showed that web-based ACP programs are feasible and improve patients' ACP knowledge, communication, and documentation(84). However, it should be considered that the use of technology does not only have benefits. Some patients, such as older patients or those who are more ill, might have difficulties in using eHealth, or might not have enough energy to for instance use a laptop or tablet(85, 86). Further, the use of eHealth should not be a substitution for face-to-face contacts and it should be considered that patients might not always have the necessary skills to use and interpret eHealth in light of their own situation(87).

Healthcare professionals should be educated in self-management support and ACP

Healthcare professionals differ in their approach towards supporting patients' self-management, and sometimes struggle with supporting self-management and conducting ACP conversations with their patients. Although adjusting their self-management support approach based on the needs of patients might be most appropriate, we cannot expect all healthcare professionals to be able to switch between these approaches and be skilled in all of these. However, when healthcare professionals are aware of their own preferences and style, and clearly communicate these to their patients and colleagues, they might still be able to adequately support the patient. Therefore, not only education and training on practical self-management support is recommended, but also education to increase self-knowledge and critical self-reflection. Also, healthcare professionals should be educated about the importance of discussing the end of life and eliciting patients' preferences about future treatment, starting in medical school, since this increases the healthcare professionals' engagement in ACP(88, 89).

RECOMMENDATIONS FOR FUTURE RESEARCH

For future research, we suggest to further examine what factors influence self-management of patients at the end of life, and the self-management support needs of patients with advanced cancer and ICDs. Also, a self-management support program specifically for patients with progressive illnesses should be developed and tested, in which ACP should also be incorporated. Specifically, it should be examined how such a program can be integrated in existing care. Having an eHealth component is recommended, amongst other due to its accessibility for patients. Currently, initiatives are ongoing to develop such a program(90). We further suggest to examine whether educational programs and practical decision aids for healthcare professionals, such as checklists or reminders in the medical record of the patient, could promote the occurrence of ACP conversations, especially in medical specialties where end-of-life care is less prevalent. Lastly, relatives might potentially play an important role in supporting patients' self-management, and we recommend to further study their involvement.

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11

CHAPTER 11

Summary

Samenvatting

SUMMARY

Chapter 1 provides an introduction on this thesis about self-management and the practice of advance care planning (ACP) of patients with progressive, life-threatening illness. It elaborates on the shift from healthcare based on the biomedical model, in which the healthcare professional tended to make decisions about treatments in the best medical interest of the patient, but made these decisions unilaterally, often without consulting the patient, towards healthcare based on the biopsychosocial model, in which a larger emphasis is put on individual choice and autonomy of the patient, and in which shared decision making is an important component. In this model, a major role and responsibility is placed on the patient, while at the same time, the healthcare system is becoming more and more fragmented, specialised and complex. It is not known to what extent patients are willing and able to take an active role in their care. Self-management, in this context defined as "*the strategies used by persons with the aim of managing the physical, psychosocial and existential consequences of living with a progressive, life-threatening disease and its treatment*" gained increasing attention in research and in clinical practice, but has rarely been examined in patients with advanced illness. In addition, little is known about the practice of ACP in patients with advanced illness other than cancer. Therefore, the aim of this thesis was to provide insight into self-management behaviours and ACP practices of patients living with an advanced illness. Self-management was examined in a population of patients with advanced cancer, and ACP was examined in a population of patients with an implantable cardioverter defibrillator (ICD).

Part I: Self-management of patients with advanced cancer

Chapter 2 presents the results of a systematic review on the experiences and attitudes of self-management of patients with advanced cancer. Seven databases were systematically searched and 31 out of 1742 identified articles were included. These studies, of which 8 were quantitative and 23 were qualitative, reported on a variety of self-management behaviours in seven domains: medicine and pharmacology, lifestyle, mental health, social support, knowledge and information, navigation and coordination and medical decision-making. We found that the behaviours patients used were individual and sometimes ambivalent. Influencing factors of self-management were age, mental state, physical function, education and self-efficacy: older patients, patients with depressive symptoms and those with lower levels of physical function, education and self-efficacy sometimes struggled with self-management of their illness. Three studies indicated that healthcare professionals thought self-management is desirable and achievable, given that it is based on sufficient skill, knowledge, and a good healthcare professional-patient relationship. This chapter concluded that self-management is

highly personal and multifaceted. Self-management support can benefit from an individualised approach, embedded in solid partnerships with relatives and healthcare professionals.

Chapter 3 includes the results of a qualitative interview study in 33 patients with advanced cancer. Patients indicated that their diagnose of advanced cancer had a major impact on different domains of their life, being physical, psychological, social, existential, medical, and daily life. Patients engaged in different self-management behaviours to deal with these consequences. These were divided in six categories: health behaviours, such as staying physically active and maintaining a healthy diet; behaviours to manage psychological consequences, such as nuancing thoughts or writing down thoughts in a notebook; behaviours related to seeking and providing social support; behaviours related to collaborating with healthcare professionals; behaviours related to obtaining knowledge and information; and behaviours related to planning, such as daily life and adhering to medication schemes. Patients indicated that living as normally as possible, despite their illness, was important. Different factors seemed to influence patients' self-management, such as their personality, their life history, the moment in the illness trajectory and the involvement of their social environment. Most patients were however not always aware of *why* they dealt with the consequences of their illness in a certain way. They often indicated to 'always have done things like this'. This suggests that patients try to draw from their already existing self-management behaviours in order to deal with their new situation of living with advanced cancer.

Chapter 4 reports on a qualitative interview study amongst 27 healthcare professionals caring for patients with advanced cancer. We examined their experiences and attitudes towards self-management of their patients, and their self-management support roles. Generally, healthcare professionals were optimistic about self-management of their patients, although some mentioned concerns about the patients who seemed less able to engage in self-management. Healthcare professionals adopted different roles in supporting self-management. These were: the instructive role, in which the healthcare professional adopts a leading role, and directs patient's self-management based on their own expertise; the collaborative role, in which a well-delineated division of tasks between healthcare professional and patients is apparent; and the advisory role, in which the patient is leading, and the healthcare professional is more serving. Healthcare professionals seemed to attribute their role to patients' characteristics, but also to their own working conditions and experience, their personality, values and life experience. Most healthcare professionals indicated to be able to switch between roles, although others inclined more towards one of the roles. In this chapter we conclude that healthcare professionals, just like patients, differ in their views and approached towards self-management and self-management support. Therefore, the instructive, collaborative and advisory support roles will all be useful under certain circumstances.

Part II: ACP in patients with an ICD

Chapter 5 describes the results of an integrative review which provided an overview of all studies reporting on ICD shock incidence and the impact of these shocks in the last phase of life in the period of 1991 until 2015. Five databases were systematically searched and 15 out of 4246 identified articles were included in the analysis. Two studies from 1996 and 1998 reported on patients who died due to non-sudden causes. They reported that 24% and 33% of patients experienced shocks in the last 24 hours of life, and 7% and 14% in the last hour of life. Seven studies, published between 1991 and 1999, reported on patients who died due to sudden causes. These studies reported that 41% to 68% of patients experienced shocks in the last 24 hours of life and 22% to 66% in the last hour of life. In the 6 studies published between 2004-2015 that did not make a distinction between patients dying non-suddenly or suddenly, it was reported that 17% to 32% of patients experienced shocks in the last month of life, 3% to 32% in the last 24 hours, and 8% to 31% in the last hour of life. Three studies, published between 2004 and 2011, examined the impact of these shocks on the patients, their relatives, and attending healthcare professionals. They reported that shocks were considered painful and distressing to patients, and were distressing for relatives and attending healthcare professionals. These findings emphasise that it is important that healthcare professionals discuss ICD deactivation in a timely manner with the patient and his or her relatives. This can contribute to timely deactivation of the ICD shock function, and therefore can minimise possible suffering due to shocks, thereby promoting a calm and peaceful death.

Chapter 6 includes the results of a retrospective case study in which 380 medical records of deceased patients with an ICD were examined. Trends in time (2007-2016) in how and when decisions were made about ICD deactivation were described and factors related to this decision were outlined. In addition, care and ICD shock incidence in the last month of life were described. In the medical records, we found that the occurrence of ICD deactivation discussions increased over the years. Six percent of the patients who died between 2007 and 2009 had a discussion about ICD deactivation reported in their medical record, compared to 35% of patients who died between 2013 and 2016. Also, the occurrence of actual ICD deactivation increased in this time period, from 16% to 42%. Factors related to deactivating the ICD were: having had an ICD deactivation discussion, a valid do-not-resuscitate order or do-not-intubate order, and the prior involvement of a palliative care team. For patients who died with an active ICD, shocks in the last month of life were reported in the medical records of 7% of these patients. This chapter concluded that, although discussions about ICD deactivation and actual deactivation have increased since 2007, ICDs remain active in the majority of patients at the end of life, some of whom experience shocks.

Chapter 7 elaborates on the results of a focus group study in which 41 patients with ICDs participated. In the focus groups, insight was gained in the experiences of these patients with ACP conversations about ICD deactivation. Although not all patients knew that deactivating the ICD was an option, most patients could imagine deciding to have their ICD deactivated one day. Different reasons were given for this, often encompassing an assessment of the balance between quality and length of life. Some patients indicated to have a need for having ACP conversations with their healthcare professional, but few had had these. Some patients indicated that healthcare professionals were knowledgeable on medical-technological domains, but that there was not always an opportunity to discuss issues with a strong emotional component, such as the end of life. Some patients said they did not have a need for having ACP conversations. These patients said they were solely focussed on living in the ‘here and now’. Patients were often hesitant to record their preferences about ICD deactivation in advance care directives, since they were unsure about whether their current preferences would reflect their future preferences. This chapter concluded that ACP conversations about ICD deactivation should occur more frequently, but should be tailored to the illness stage of the patient and the readiness to discuss this topic.

Chapter 8 describes the results of a mixed-methods study. A total of 170 relatives of deceased ICD patients completed a survey, of which 23 relatives also participated in focus groups. We examined the experiences of these relatives with ACP discussions about ICD deactivation, and with treatment and care in the last phase of life of the patient. We found that, according to the relatives, 38% of the patients had had an ACP conversation with a healthcare professional about ICD deactivation, and that 24% of the patients had experienced one or more shocks in the last month of life, which were distressing for 74% of patients and 73% of relatives. The ICD was deactivated in 55% of the patients. Relatives reported numerous barriers towards having ACP conversations with healthcare professionals. These included, amongst others, patients’ lack of willingness to talk about ICD deactivation, having a bad relationship with the healthcare professional and a perceived lack of time from the professional. Relatives had both positive and negative experiences with the treatment and care in the last phase of life, and 42% to 61% of relatives reported to be satisfied with different aspects of this care. Quality of death was rated relatively low by relatives, but was significantly higher for patients who died with a deactivated ICD (quality of death 6.74 on a 10-point scale) compared to patients who died with an active ICD (quality of death 5.67). This chapter concluded that ACP conversations about ICD deactivation are rare, and suggested that healthcare professionals should be educated about having these conversations with their patients.

Chapter 9 presents a viewpoint on the experienced barriers of healthcare professionals towards having ACP conversations about ICD deactivation with their patients. We could synthesize a total of 31 barriers from interviews with 26 healthcare professionals involved in the care for ICD patients at the end of life. These were related to clinical practice, such as having a lack of time or knowledge, and having difficulties finding the right moment to discuss the topic; the patient, such as experiencing reluctance in the patient to discuss the topic, a lack of knowledge in the patient about the functioning of the ICD, and an overestimation of the life-saving capacity of the ICD; and society, such as a predominant focus of healthcare and society on treating and curing illness. In this chapter, development and implementation of a program aimed at educating healthcare professionals involved in the care for patients with an ICD at the end of their lives was suggested. It is advocated that ACP and discussions about care at the end of life should become an integral part of cardiologic care. Only then the number of patients experiencing inappropriate and painful shocks at the end of their lives can be reduced.

In **chapter 10**, I elaborate on the results of the studies included in this thesis in a general discussion. In this chapter, the main findings of this thesis are summarised and discussed, methodological considerations are outlined, and recommendations for clinical practice and future research are given. I conclude that living with a progressive, life-threatening illness is hard work. Although some patients seem to be able to effectively deal with the many consequences of their illness, for others this can be a burden, and these patients are overwhelmed by the work of having a progressive illness, while mostly they just want to live life as normally as possible. Further, patients show a variety in the need and willingness to engage in ACP. Some patients specifically thought about and discussed their preferences for future medical treatment and care, while others were focussed on living in the ‘here and now’. Self-management is very much individually determined, and so is the need for self-management support and having ACP conversations. Patients can benefit by being educated about general self-management skills, early in their illness trajectory. Subsequently, I advocate an individual approach to self-management support. In order to do so, it is important to have frequent ‘self-management check-ups’, performed by a central, coordinating caregiver. This caregiver, presumably a nurse, discusses with the patient his or her self-management, expectations, responsibilities, needs and preferences, and can refer the patient to other healthcare professionals if needed. During these check-ups, ACP should also be an important topic. In order to have these in-depth discussions with the patient it is important to have a strong healthcare professional – patient relationship, based on trust. In order for healthcare professionals to be skilled in supporting the patients' self-management and conducting ACP conversations, they should be educated about these topics.

SAMENVATTING

Hoofdstuk 1 bevat een algemene introductie op dit proefschrift over zelfmanagement en advance care planning (ACP) van patiënten met progressieve, levensbedreigende ziekten. Het beschrijft de verschuiving van gezondheidszorg gebaseerd op het biomedische model, waarbij de zorgverlener zelfstandig beslissingen nam over behandelingen, vaak zonder de patiënt te raadplegen, naar gezondheidszorg op basis van het biopsychosociale model, waarbij meer nadruk wordt gelegd op individuele keuze en autonomie van de patiënt, en waarin gedeelde besluitvorming een belangrijke rol speelt. In het biopsychosociale model wordt een grote rol en verantwoordelijkheid bij de patiënt gelegd, terwijl tegelijkertijd de gezondheidszorg versnippert en steeds meer gespecialiseerd en complex wordt. Het is niet bekend in hoeverre patiënten een actieve rol in hun zorg willen en kunnen spelen. Zelfmanagement, in deze context gedefinieerd als "*de strategieën die personen gebruiken om te kunnen omgaan met de fysieke, psychosociale en existentiële gevolgen van het leven met een progressieve, levensbedreigende ziekte en de behandeling daarvan*" krijgt steeds meer aandacht in onderzoek en in de klinische praktijk, maar is weinig onderzocht bij patiënten met een gevorderde ziekte. Daarnaast is er weinig bekend over ACP bij patiënten met andere gevorderde ziekten dan kanker. Het doel van dit proefschrift was om inzicht te krijgen in zelfmanagement en ACP van patiënten met een gevorderde ziekte. Zelfmanagement werd onderzocht in een populatie van patiënten met gevorderde kanker en ACP werd onderzocht in een populatie van patiënten met een implanteerbare cardioverter defibrillator (ICD).

Deel I: zelfmanagement van patiënten met gevorderde kanker

Hoofdstuk 2 presenteert de resultaten van een systematisch literatuuronderzoek naar zelfmanagement van patiënten met gevorderde kanker. Zeven databases werden systematisch doorzocht en 31 van de 1742 geïdentificeerde artikelen werden geïncludeerd. Deze studies, waarvan 8 met een kwantitatieve en 23 met een kwalitatieve onderzoeksopzet, rapporteerden over een verscheidenheid aan zelfmanagementgedrag in zeven domeinen: geneeskunde en farmacologie, levensstijl, geestelijke gezondheid, sociale ondersteuning, kennis en informatie, navigatie en coördinatie van zorg en medische besluitvorming. We vonden dat zelfmanagement van patiënten individueel en soms ambivalent was. Zelfmanagement werd beïnvloed door leeftijd, mentale toestand, fysiek functioneren, opleiding en ‘zelf-effectiviteit’, ofwel het vertrouwen dat mensen hebben in hun eigen bekwaamheid om met succes hun situatie te beïnvloeden. Oudere patiënten, patiënten met depressieve symptomen en mensen met een lager niveau van fysiek functioneren, opleiding en zelf-effectiviteit worstelden soms met zelfmanagement. Uit drie onderzoeken bleek dat zorgverleners zelfmanagement wenselijk en haalbaar vonden, mits het gebaseerd is op voldoende vaardigheden, kennis en een goede relatie tussen zorgverlener en patiënt. Dit hoofdstuk concludeert dat zelfmanagement persoonlijk en veelzijdig is. Ondersteuning van zelfmanagement heeft baat bij een geïndividualiseerde aanpak, ingebed in een goede samenwerking tussen patiënt, familieleden en zorgverleners.

Hoofdstuk 3 bevat de resultaten van een kwalitatief onderzoek waarin 33 patiënten met gevorderde kanker werden geïnterviewd. Patiënten gaven aan dat hun diagnose een grote impact had op verschillende domeinen van het leven, namelijk fysiek, psychologisch, sociaal, existentieel, medisch en het dagelijks leven. Patiënten lieten verschillende vormen van zelfmanagement zien om te kunnen omgaan met deze gevolgen. Dit zelfmanagement werd onderverdeeld in zes categorieën: gedrag gericht op gezondheidsbevordering, zoals lichamelijk actief blijven en een gezond dieet aanhouden; gedrag om psychologische gevolgen te beheersen, zoals het nuanceren van gedachten of het opschrijven van gedachten in een notitieboek; gedrag gericht op het zoeken naar en verlenen van sociale steun; gedrag gericht op het samenwerken met professionele zorgverleners; gedrag gericht op het verkrijgen van kennis en informatie; en gedrag gerelateerd aan plannen, zoals het plannen van het dagelijks leven en het volgen van medicatieschema's. Patiënten gaven aan dat ze het belangrijk vinden om een zo normaal mogelijk leven te leiden. Verschillende factoren leken zelfmanagement van patiënten te beïnvloeden, zoals hun persoonlijkheid, hun levensgeschiedenis, het moment in het ziektetraject en de betrokkenheid van hun sociale omgeving. De meeste patiënten wisten niet altijd waarom ze op een bepaalde manier met de gevolgen van hun ziekte omgingen. Ze gaven vaak aan dat ze al op een bepaalde manier omgingen met uitdagingen in het leven. Dit suggereert dat patiënten proberen te putten uit hun reeds bestaande zelfmanagementgedrag om te kunnen omgaan met het leven met gevorderde kanker.

Hoofdstuk 4 rapporteert de resultaten van een kwalitatief onderzoek waarin 27 zorgverleners betrokken bij de zorg voor patiënten met gevorderde kanker werden geïnterviewd. Het doel was om de ervaringen en opvattingen van zorgverleners met betrekking tot zelfmanagement van patiënten met gevorderde kanker te onderzoeken, en een beschrijving te geven van hun rol in zelfmanagementondersteuning. Over het algemeen waren zorgverleners optimistisch over zelfmanagement van hun patiënten, hoewel sommigen bezorgd waren over de patiënten die minder in staat leken te zijn tot zelfmanagement. Zorgverleners namen verschillende rollen aan bij het ondersteunen van zelfmanagement. Dit waren: de instructieve rol, waarin de zorgverlener een leidende rol op zich neemt en zelfmanagement van de patiënt aanstuurt op basis van zijn eigen expertise; de samenwerkende rol, waarin een goed afgebakende taakverdeling tussen zorgverleners en patiënten zichtbaar is; en de adviserende rol waarin de patiënt de leiding heeft en de zorgverlener een meer dienende rol op zich neemt. Zorgverleners schreven hun rol toe aan patiëntkenmerken, maar ook aan hun eigen werkomstandigheden en ervaring, hun persoonlijkheid, waarden en levenservaring. De meeste zorgverleners gaven aan te kunnen schakelen tussen deze verschillende rollen, alhoewel sommigen meer neigden naar een van de rollen. In dit hoofdstuk concluderen we dat zorgverleners, net zoals patiënten, verschillen in hun opvattingen en benadering wat betreft zelfmanagement en zelfmanagementondersteuning. Zowel de instructieve, samenwerkende en adviserende rol zijn allen bruikbaar onder verschillende omstandigheden.

Deel II: ACP bij patiënten met een ICD

Hoofdstuk 5 beschrijft de resultaten van een literatuuronderzoek, welke een overzicht geeft van alle onderzoeken die rapporteren over de incidentie van ICD shocks en de impact van deze shocks op de laatste fase van het leven. Vijf databases werden systematisch doorzocht en 15 van de 4246 geïdentificeerde artikelen, gepubliceerd in de periode van 1991 tot 2015, werden geïncludeerd. Twee studies uit 1996 en 1998 rapporteerden over patiënten die overleden aan niet-plotselinge oorzaken. Deze studies rapporteerden dat 24% en 33% van de patiënten shocks ervaarden in de laatste 24 uur van het leven, en 7% en 14% van de patiënten in het laatste uur van het leven. Zeven studies, gepubliceerd tussen 1991 en 1999, rapporteerden over patiënten die overleden aan plotselinge oorzaken. Deze studies rapporteerden dat 41% tot 68% van de patiënten shocks ervaarden in de laatste 24 uur van het leven en 22% tot 66% van de patiënten in het laatste uur van het leven. In de 6 studies gepubliceerd tussen 2004 en 2015 die geen onderscheid maakten tussen doodsoorzaak, werd gerapporteerd dat 17% tot 32% van de patiënten shocks ervaarden in de laatste maand van het leven, 3% tot 32% in de laatste 24 uur en 8% tot 31% van de patiënten in het laatste uur van het leven. Drie studies, gepubliceerd tussen 2004 en 2011, onderzochten de impact van ICD shocks op patiënten, hun naasten en zorgverleners. Ze rapporteerden dat shocks pijnlijk en stressvol waren voor patiënten en stressvol voor naasten en zorgverleners. De bevindingen in dit hoofdstuk benadrukken dat het belangrijk is dat zorgverleners ICD deactivatie vroegtijdig met de patiënt en hun naasten bespreken. Dit kan bijdragen aan het tijdig deactiveren van de shockfunctie van de ICD, waardoor shocks kunnen worden voorkomen. Dit bevordert een rustig en comfortabel sterven.

Hoofdstuk 6 bevat de resultaten van een retrospectief dossieronderzoek waarin 380 medische dossiers van overleden patiënten met een ICD zijn onderzocht. In dit hoofdstuk wordt beschreven hoe de besluitvorming omtrent het deactiveren van de ICD al dan niet is veranderd in de periode van 2007 tot 2016. Daarnaast wordt gepresenteerd welke factoren verband houden met deze besluitvorming en worden de zorg en incidentie van ICD shocks in de laatste maand van het leven beschreven. Uit het dossieronderzoek bleek dat het aantal gesprekken over ICD deactivatie in de loop der jaren is toegenomen. Bij de patiënten die overleden tussen 2007 en 2009 werd in 6% van de medische dossiers een aantekening over een gesprek over ICD deactivatie gevonden, vergeleken met 35% van de dossiers van mensen die overleden tussen 2013 en 2016. Daarnaast zagen we dat het aantal ICD deactivaties toenam, van 16% in de periode van 2007 tot 2009, tot 42% in de periode van 2013 tot 2016. Verschillende factoren waren geassocieerd met het deactiveren van de ICD, zoals een gesprek over ICD deactivatie, een niet-reanimeren of niet-intuberen beleid en de betrokkenheid van een consultatieteam palliatieve zorg. Van de patiënten die overleden met een actieve ICD, werd bij 7% een aantekening in het dossier gevonden dat deze patiënt een shock heeft gekregen in de laatste maand van het leven. Dit hoofdstuk concludeert dat, hoewel gesprekken over ICD deactivatie en het daadwerkelijke aantal ICD deactivaties sinds 2007 zijn toegenomen, de meerderheid van de patiënten overlijdt met een actieve ICD, van wie sommigen een of meerdere shocks krijgen in de laatste maand voor overlijden.

Hoofdstuk 7 rapporteert de resultaten van een focusgroep onderzoek waaraan 41 patiënten met een ICD deelnamen. In de focusgroepen werd inzicht verkregen in de ervaringen van deze patiënten met ACP gesprekken over ICD deactivatie. Hoewel niet alle patiënten wisten dat het deactiveren van de ICD een optie was, konden de meeste patiënten zich voorstellen dat ze op een dag zouden besluiten hun ICD te deactiveren. Hiervoor werden verschillende redenen gegeven, waarbij vaak een afweging werd gemaakt tussen kwaliteit van leven en levensduur. Sommige patiënten gaven aan behoefte te hebben aan ACP gesprekken met hun zorgverlener, maar slechts weinigen hadden deze gehad. Sommige patiënten gaven aan dat zorgverleners veel kennis hadden op medisch-technologische gebied, maar dat er niet altijd gelegenheid was om zaken te bespreken met een sterk emotioneel component, zoals het levenseinde. Sommige patiënten vertelden dat ze geen behoefte hadden aan ACP gesprekken. Deze patiënten zeiden dat ze uitsluitend gericht waren op het leven in het ‘hier en nu’. Patiënten aarzelden vaak om hun voorkeuren met betrekking tot het deactiveren van de ICD vast te leggen, omdat ze niet zeker wisten of hun huidige voorkeuren hun toekomstige voorkeuren zouden weerspiegelen. Dit hoofdstuk concludeert dat ACP gesprekken over het deactiveren van ICDs vaker zouden moeten plaatsvinden, maar dat deze wel moeten worden afgestemd op het ziektestadium van de patiënt en de bereidheid van de patiënt om dit onderwerp te bespreken.

Hoofdstuk 8 beschrijft de resultaten van een onderzoek met een ‘mixed-methods’ design. Honderdzeventig naasten van overleden patiënten met een ICD vulden voor dit onderzoek een vragenlijst in, waarvan er 23 ook deelnamen aan focusgroepen. In dit onderzoek werden de ervaringen van de naasten met ACP gesprekken over ICD deactivatie en met de behandeling en zorg in de laatste levensfase van de patiënt onderzocht. Volgens de naasten had 38% van de patiënten een ACP gesprek gevoerd met een zorgverlener over het deactiveren van de ICD, en 24% van de patiënten had een of meerdere shocks van hun ICD hadden gekregen in de laatste maand van het leven. Deze shocks waren stressvol voor 74% van de patiënten en 73% van de naasten. Bij 55% van de patiënten was de ICD voorafgaand aan het overlijden gedeactiveerd. De naasten beschreven verschillende barrières voor het hebben van ACP gesprekken, waaronder een gebrek aan bereidheid van patiënten om te praten over het uitschakelen van de ICD, een slechte relatie met de zorgverlener en gebrek aan tijd van de zorgverlener. Naasten hadden zowel positieve als negatieve ervaringen met de behandeling en zorg in de laatste levensfase, en 42% tot 61% van de naasten gaf aan tevreden te zijn met verschillende aspecten van deze zorg. De kwaliteit van sterven werd relatief laag beoordeeld, maar was significant hoger voor patiënten die overleden met een gedeactiveerde ICD (kwaliteit van overlijden 6,7 op een 10-puntsschaal) vergeleken met patiënten die overleden met een actieve ICD (kwaliteit van overlijden 5,7). Dit hoofdstuk concludeert dat ACP gesprekken over het deactiveren van ICDs weinig voorkomen, en raadt aan dat zorgverleners geschoold moeten worden in het voeren van ACP gesprekken met hun patiënten.

Hoofdstuk 9 presenteert de resultaten van een onderzoek naar de barrières die zorgverleners ervaren bij het voeren van ACP gesprekken over ICD deactivatie met hun patiënten. In interviews met 26 zorgverleners die betrokken waren bij de zorg voor patiënten met een ICD in de laatste fase van het leven identificeerden we in totaal 31 barrières voor het voeren van ACP gesprekken over ICD deactivatie. Deze barrières werden onderverdeeld in drie categorieën: barrières gerelateerd aan de klinische praktijk, zoals een gebrek aan tijd of kennis en moeite hebben met het vinden van het juiste moment om het onderwerp te bespreken; barrières gerelateerd aan de patiënt, zoals veronderstelde onwil van de patiënt om het onderwerp te bespreken, een gebrek aan kennis bij de patiënt over het functioneren van de ICD, en een overschatting door de patiënt van de levensreddende mogelijkheid van de ICD; en barrières gerelateerd aan de samenleving, zoals de focus van de gezondheidszorg en samenleving op het behandelen en genezen van ziekten. In dit hoofdstuk wordt voorgesteld om een programma te ontwikkelen en te implementeren dat gericht is op het scholen van zorgverleners die betrokken zijn bij de zorg voor patiënten met een ICD in de laatste levensfase. Daarnaast wordt voorgesteld dat ACP en gesprekken over zorg aan het levenseinde een integraal onderdeel van cardiologische zorg zouden moeten worden. Alleen dan kan het aantal patiënten dat ongepaste en pijnlijke shocks aan het einde van hun leven krijgt, worden verminderd.

Hoofdstuk 10 beschrijft de algemene discussie van dit proefschrift. Dit hoofdstuk bevat een samenvatting en bespreking van de belangrijkste bevindingen van dit proefschrift en de gebruikte methodologie, en bevat aanbevelingen voor zowel de klinische praktijk als voor toekomstig onderzoek. Ik concludeer dat het leven met een progressieve, levensbedreigende ziekte hard werken is voor de patiënt. Hoewel sommige patiënten goed om lijken te gaan met de gevolgen van hun ziekte, ervaren sommigen hun ziekte als een grote last en worden ze overweldigd door het werk wat hierbij komt kijken, terwijl zij vooral een zo normaal mogelijk leven proberen te leiden. Daarnaast verschilden patiënten in hun behoefte en bereidheid om deel te nemen aan ACP. Sommige patiënten dachten specifiek na over hun voorkeuren voor toekomstige medische behandeling en zorg en bespraken deze, terwijl anderen gericht waren op het leven in het 'hier en nu'. Zelfmanagement van de patiënt is individueel bepaald, net zoals de behoefte van de patiënt aan zelfmanagementondersteuning en het voeren van ACP gesprekken. Patiënten kunnen er baat bij hebben om al vroeg in hun ziekteproces te worden voorgelicht over algemene vaardigheden op het gebied van zelfmanagement. Ik pleit voor een individuele benadering van zelfmanagementondersteuning. Hiervoor is het belangrijk dat de patiënt regelmatig gesprekken heeft met een centrale, coördinerende zorgverlener. Deze zorgverlener, bijvoorbeeld een verpleegkundige, evalueert met de patiënt zijn of haar zelfmanagement, verwachtingen, verantwoordelijkheden, behoeften en voorkeuren, en kan de patiënt indien nodig doorverwijzen naar andere zorgverleners. Tijdens deze gesprekken moet ACP ook een belangrijk onderwerp zijn. Een sterke relatie tussen de zorgverlener en de patiënt, gebaseerd op vertrouwen, is een belangrijke voorwaarde om deze diepgaande gesprekken te kunnen voeren. Om ervoor te zorgen dat zorgverleners bekwaam zijn in het ondersteunen van zelfmanagement van de patiënten en in het voeren van ACP gesprekken, is scholing over deze onderwerpen essentieel.