Knowledge co-production in health research, policy and care practice

Patient involvement in health-related decisions

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INTRODUCTION
Introduction
Chapter 1 – Introduction

The emergence of complex, uncertain health innovations over the past four decades has been responsible for a greater need for exchange and integration of knowledge of various societal actors, also known as knowledge co-production. Health-related decisions are no longer the exclusive responsibility of researchers and health professionals. The shift to multi-actor involvement in health-related decisions has been especially visible in the case of patients. Patients are newcomers in the decision-making arena on health research, policy and care practice, only making their entrance as active players some two decades ago. In this thesis, I explore how knowledge co-production processes could be more optimally structured to enhance effective patient involvement in health-related decisions.

1.1 Patient involvement in health-related decisions

Leading health organizations in Western countries are increasingly recognizing the role of patients in the formulation of health policies and visions on health-related decisions (World Health Organization 1978; Coulter 2003). For instance, the Council of Europe considers that patient involvement in health-related decisions is a basic and essential part of a democratic society (Council of Europe 2000). In a similar vein, the National Health and Medical Research Council of Australia recognizes that public scrutiny and debate could help to enhance the integrity and accountability of research (National Health and Medical Research Council 2002). These examples demonstrate increased attention for patient involvement in the international health scene.

Patient involvement in decision-making can be defined as patients being actively involved in and having influence on decision-making processes in health research, policy and care practice (e.g. Oliver 2008; Tritter 2009; Boote et al. 2010; Elberse 2012). Patients can be involved in different types of health research, varying from biomedical research (Caron-Flinterman 2005; Elberse 2012), more applied and clinical research (Epstein 1995; Marsden et al. 2004; PatientPartner 2011) to health service research (McIntyre et al. 2010). Moreover, patient involvement
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occurs in all phases of the research cycle: agenda setting (Caron-Flinterman et al. 2005; Mitton et al. 2009; Stewart et al. 2011; Nasser et al. 2012), research designs (Marsden et al. 2004; McCormick et al. 2004; Ali et al. 2006; Boote et al. 2012), proposal appraisal (O'Donnell and Entwistle 2004; Teunissen et al. 2011), conduct of research (Faulkner 2006; Morrow et al. 2010; de Wit et al. 2013) and dissemination of end-results (McLaughlin 2006). In health policy, patients can, for instance, become involved in procedures of funding agencies (O'Donnell and Entwistle 2004) and insurance agencies (Judd and Rienstra 2013). Governments involve patients in all phases of the policy process: agenda setting, policy formulation, implementation and evaluation (Broerse et al. 2009). In care practice, patient involvement in decision making also covers a broad area, ranging from clinical guideline development (Boivin and Legare 2007; Bovenkamp and Trappenburg 2009), shared decision making (van der Weijden et al. 2011), criteria for ‘good care’ (Teunissen et al. 2011; Broerse et al. 2012) to tools for self-management (Warsi et al. 2004; Steffelson et al. 2010).

The attention for patient involvement is related to two major developments in society. The first concerns the profound impact of health innovations on health practice and policy-making. The emergence of complex health innovations, such as personalized medicine and genetic testing, are associated with a high degree of uncertainty regarding their societal effects. As a consequence, it has been argued that health-related, value-based or contextual decisions should be made in discussion with patients, and not by researchers and health professionals alone (Oliver et al. 2004; Pivik et al. 2004; Hansen Børsen 2006). Changes within society, including consumerism, public accountability and democratization of science, also facilitate openness towards patient involvement (Fuller 2000; Nowotny et al. 2001; Chopyak and Levesque 2002; Scott 2007; Broerse et al. 2010).

The second societal development relates to the shift from a supply-driven towards a demand-based health practice. Over the last decades, health-related decisions have become more influenced by the needs and preferences of patients. This shift can be explained by advances in democracy and the disability rights movement (Oliver et al. 2004) and, more recently, by the push to restructure health systems due to policy failures, perpetual crisis and tight budgets (Tomes 2006). Another explanation involves the increased emphasis on empowerment of patients in their treatment and care, but also in higher levels of decision making, for example in discussions regarding insurance coverage, guideline development, agenda setting,
and funding of research (Caron-Flinterman et al. 2005; Abma 2006; Schipper 2012).

In the scientific literature, patient involvement in health-related decisions is justified by three main arguments. First, patients are primary actors and end-users of health-related decisions, and therefore have the right to be involved (normative argument) (Goodare and Smith 1995). Second, patients are confronted on a daily basis with the consequences of their disease/condition and, consequently, gain specific knowledge – experiential knowledge – of their disease. This experiential knowledge complements the knowledge of researchers and health professionals, and thereby improves the quality of health-related decisions (substantive argument) (Telford et al. 2002; Caron-Flinterman et al. 2005; Needham 2008). Third, the legitimacy of health-related decisions increases when patients are involved. The involvement of patients increases the chance of successful implementation, since the decisions are more in line with the needs and wishes of patients (political argument) (Telford et al. 2002; Rowe and Frewer 2004; Abma and Broerse 2010).

On the international health scene, patient involvement in health-related decisions represents a major feature and has led to a variety of initiatives. The UK is a frontrunner and several institutions, such as Involve (INVOLVE 2013) and the James Lind Alliance (Alliance 2013), have been established to facilitate patient involvement. Elsewhere, various health authorities and organizations are stimulating patient involvement, such as the Consumer Health Forum of Australia (Consumer's Health Forum of Australia and National Health & Medical Research Council 2001), the Canadian Coordinating Office for Health Technology Assessment (Pivik et al. 2004) and the Cochrane Collaboration (Shea et al. 2005). The Netherlands is also considered an important player, since relatively many actors (health authorities and disease-specific foundations) have taken up patient involvement and are experimenting with various approaches to patient involvement (ZonMW 2006; Elberse et al. 2009; Bovenkamp 2010; Elberse et al. 2011; Nierse and Abma 2011; de Wit et al. 2013).
1.2 Knowledge co-production: approaches and challenges

The involvement of patients in health-related decisions implies that patients should be included in the process of knowledge co-production. Knowledge co-production represents the development of knowledge between science and society (Regeer 2009; Betten et al. 2013; Broerse 2013). The involvement of patients in this process implies that patients’ lived experiences are converted from implicit knowledge into explicit knowledge (knowledge articulation), are subsequently integrated with the knowledge of researchers and health professionals (knowledge integration) and are embedded in health research, policy and care practice (knowledge embedding). The process of knowledge co-production entails the development of appropriate strategies for articulating, integrating and embedding the knowledge of the different actors.

However, research has shown that despite the development of appropriate approaches and many activities involving patients, these initiatives generally have had a low impact on decision-making processes (Boote et al. 2002; Oliver et al. 2008; Staniszewska et al. 2008). Apparently, knowledge co-production processes are not entirely successful and various difficulties are experienced in relation to the implementation of the three elements of the knowledge co-production process (Boote et al. 2002; Stevens et al. 2003; Caron-Flinterman et al. 2007; Bijker et al. 2009; Jinks et al. 2009; Broerse et al. 2010; Elberse et al. 2011).

This thesis aims to acquire insights into how knowledge co-production processes could be more effective in terms of improving the influence of patients on decision-making processes in health research, policy and care practice. Specific attention is paid to constraints encountered and strategies that could be applied in the three key elements of knowledge co-production: (1) knowledge articulation (particularly the knowledge of patients), (2) integration of the knowledge of researchers, health professionals and patients, and (3) embedding of knowledge co-production and its outcomes in health research, policy and care practice.
THEORETICAL BACKGROUND
Chapter 2 - Theoretical background

In this chapter, background is provided to relevant concepts in knowledge co-production processes. First, I elaborate on knowledge co-production processes in general. After conceptualizing knowledge co-production, the key elements of knowledge co-production processes, namely articulation, integration and embedding are discussed in more detail. Thereafter, insights are provided into how knowledge can be articulated, knowledge as a concept is discussed, as well as the different types of knowledge and their distribution among various actors. Also, strategies for knowledge articulation are presented. Next, ways of bringing about knowledge integration and its challenges are discussed. And, the process of knowledge embedding is described. Finally, the difficulties concerning on-going co-production of knowledge are discussed.

2.1 Knowledge co-production

Patient involvement in health-related decision making is not an isolated development. It is part of a larger trend to involve a broad range of actors with different perspectives and types of knowledge in the development of emerging innovations and solving complex problems (In ’t Veld 2000; Klein et al. 2001; Boon 2008; Regeer and Bunders 2009; Betten et al. 2013). Many innovations lead to uncertainties and concerns, making their development a meandering and complex process. Uncertainties and concerns relate to the nature and development of innovations; the lack of clear consensus on end goals or preferred solutions; and the difficulties of obtaining clear indications on how to bring about possible solutions (Merkx 2012). The uncertainties and concerns emerged from innovations have become so complex that they cannot be solved from a single perspective. They demand a different solution-oriented problem approach in which the traditional distinction between science and society is no longer appropriate. Therefore, a different form of knowledge development, in which different disciplines and societal actors work together, is required (Gibbons 1994; In ’t Veld 2000; Klein et al. 2001; Nowotny et al. 2001; Jasanoff 2004; Regeer and Bunders 2009; Betten et al. 2013).
The emerging uncertainties and concerns also correspond to so-called ‘unstructured policy problems’ (Hisschemöller and Hoppe 1996). Policy problems are unstructured when science disagrees about the relevant scientific facts and when societal actors disagree about values and objectives. Given the increase in the complexity of problems and innovations, it has been argued that value-based or contextual decisions should be made in discussion with end-users (including patients), and not by researchers and health professionals alone (Oliver et al. 2004; Pivik et al. 2004; Hansen Børsen 2006). Changes within society, including consumerism, public accountability and democratization of science, have encouraged the involvement of end-users in decision-making (Harvey 1989; Fuller 2000; Nowotny et al. 2001).

The developments described above have had a profound impact on scientific knowledge production. While scientific knowledge production used to be primarily located in academic institutes, knowledge production is now increasingly taking place in transdisciplinary, heterogeneous collaborations (Jasanoff 2004; Hessels 2008; Regeer and Bunders 2009). Transdisciplinarity can be defined as

“A new form of learning and problem-solving, involving co-operation between different parts of society and science in order to meet complex challenges of society. Transdisciplinary research starts from tangible, real-world problems. Solutions are devised in collaboration with multiple actors” (Klein et al. 2001, p. 7).

Thus, the responsibility for solving complex health-related problems no longer lies solely within the scientific domain, indicative of a changed relationship between science and society. In the past, scientific knowledge development was considered to be autonomous and divorced from society and, at the same time, assumed to automatically engender societal and economic progress. However, it is now increasingly understood that science and society should co-operate intensively in joint knowledge production to solve complex and uncertain problems (Gibbons 1994; Hessels 2008; Regeer and Bunders 2009). In his reference work The New Production of Knowledge, Gibbons (Gibbons 1994) explains the changing relationship between science and society by presenting different modes of scientific knowledge development which have evolved over time (see Table 2.1) (Regeer and Bunders 2009). Over the past decades, the relationship between science and society has evolved from science and society being separate domains (mode-0), to being connected (mode-1) and interactive (mode-2).
Table 2.1. Different modes of knowledge development (Regeer and Bunders 2009)

<table>
<thead>
<tr>
<th>Mode</th>
<th>Relationship between science and society</th>
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<tbody>
<tr>
<td>Mode – 0</td>
<td>Separate domains</td>
</tr>
<tr>
<td>Mode – 1</td>
<td>Co-operation between domains, but no change in working methods within domains</td>
</tr>
<tr>
<td>Mode – 2</td>
<td>Co-production: science and society are jointly producing knowledge</td>
</tr>
</tbody>
</table>

The increased attention for involving a broad range of actors in health-related decisions is not only framed in the context of mode-2 knowledge production (Gibbons 1994; Nowotny et al. 2001) but also in various other constructivist areas, like transdisciplinary research (Klein et al. 2001), Third Wave of science studies (Collins and Evans 2002), strategic science (Rip 2004) and post-normal science (Funtowicz and Ravetz 1993). These different constructivist areas have a comparable underlying assumption, namely that the emergence of a knowledge production process is ‘socially robust’ and requires a heterogeneous practice (Nowotny 2003; Hessels 2008). In this thesis, the involvement of multiple actors in health-related decisions is referred to as knowledge co-production, consistent with definitions of transdisciplinary research (Klein et al. 2001). Knowledge co-production can occur at many different levels and in many different systems (e.g. economic, political, social, or scientific). Given the focus on patient involvement, this thesis proposes a closer look at knowledge co-production within the health domain. The concepts underlying knowledge co-production, its challenges and potential success factors are described in detail in the following sections.

**What is knowledge co-production?**

Knowledge co-production relates to knowledge development between science and society. In the context of health, it involves knowledge development between science, health professionals and patients. For this thesis, knowledge co-production is defined as: A form of knowledge development in which different scientific disciplines work with societal actors, to develop concrete solutions to complex problems. In the health domain, this involves health professionals, patients and uncertain health-related innovations.
This represents a further specification of the previously mentioned definition of transdisciplinary research of (Klein et al. 2001).

![Diagram of knowledge co-production process](image)

**Figure 2.1. The knowledge co-production process (Broerse 2013)**

This transdisciplinary process of knowledge co-production has been named knowledge co-creation by other scholars (Regeer and Bunders 2009; Merkx 2012; Broerse 2013), and has three key elements: (1) knowledge articulation, (2) knowledge integration and (3) knowledge embedding (see Figure 2.1) (Regeer and Bunders 2003; Broerse 2013). Knowledge co-production is not a linear process of research that follows from problem definition to problem analysis and problem solution, but a complex co-evolutionary process that proceeds iteratively and is action-oriented (Regeer and Bunders 2009; Merkx 2012). By means of an interactive process, implicit knowledge is made explicit (knowledge articulation). In dialogue between multiple actors, the different perspectives are brought together in a learning process (knowledge integration) that ultimately generates ‘socially robust knowledge’: knowledge that is not only scientifically reliable, but that is also accepted and used in society (knowledge embedding) (Nowotny 2003; Hessels 2008; Regeer 2009). This generation of ‘socially robust knowledge’ is represented in Figure 2.2. Regeer (2009) has also emphasized that knowledge co-production is a communicative process and that the knowledge generated should be seen as the end-product of communication and shared practices (Regeer 2009). This perspective prevents the separation of ‘the knower’ from the context: knowledge is grounded in practice, rather than in objective reality.
Theoretical background

<table>
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<tr>
<th>Knowledge articulation</th>
<th>Knowledge integration</th>
<th>Knowledge embedding</th>
</tr>
</thead>
<tbody>
<tr>
<td>Researchers, health professionals and patients have their own types of knowledge which have to be individually articulated. The knowledge is contextualised, embodied and shared.</td>
<td>The different actors (with their specific knowledge) are brought together in a process of joint problems solving.</td>
<td>'Socially robust knowledge' is generated in which practices and activities are shared.</td>
</tr>
</tbody>
</table>

The various actors are represented in the different figures, e.g. squares, stars, dots.

Figure 2.2. The generation of ‘socially robust knowledge’, (adapted from Regeer and Bunders 2003)

The process of knowledge co-production is however not self-evident because of different procedures, methods, goals, jargon, power and frames of reference\(^1\) (Lakoff and Johnson 2003). Specific approaches are needed to support the process of knowledge co-production and to optimize the outcomes (Merkx 2012). For the operationalization of knowledge co-production processes, scholars have experimented with and developed several multi-phased participatory approaches, e.g. the Interactive Learning and Action Approach (Broerse and Bunders 2000), consensus conferences (Abelson et al. 2003a), Constructive Technology Assessment (Rip and Schot 1995), the Dialogue Model (Abma and Broerse 2010), the Cochrane Agenda and Priority Setting Methods Group (Nasser et al 2012), the Priority Setting Partnerships developed (Alliance 2010), and demand articulation processes (Boon et al. 2011). The multi-phased participatory approaches demonstrate many similarities in their theoretical concepts (focus on dialogue  

\(^1\) Frames of reference can be thought of as mental structures that people use to make sense of complex reality and to guide their actions. They organize central ideas on an issue and are largely metaphorical (Lakoff, 2003)
between actors and learning processes) and guidelines for activities (cyclical and iterative) (Abma and Broerse 2010). Box 2.1 describes a multi-phased participatory approach in the health domain - the Dialogue Model - in more detail.

Often different levels of involvement are distinguished based on the ladder of citizen participation of Arnstein (1969). The ladder, originally including eight ‘sports’, has for application in the health domain been condensed to the following four levels (Caron-Flinterman 2005; Tritter and McCallum 2006; Oliver et al. 2008; Abma et al. 2009; Elberse 2012):

- **Consultation:** Patients are seen as information providers but have little power to influence decisions. It is up to the conventional decision makers whether the input of patients is included in the decision-making process.
- **Advice:** Patients provide advice on health-related decisions, as members of an advisory or decision-making committee. Their influence in the decision-making process is, however, not guaranteed and depends much on the group dynamics.
- **Collaboration/partnership:** This involves the formation of partnerships between health care professionals and patients. Patient inputs are included in decision-making processes; integration of knowledge occurs.
- **Control:** Here a shift in decision-making power takes place; control is transferred from researchers and health professionals to patients. For example, a patient organization commissions and supervises the research.

Many participatory approaches combine different levels of involvement, and separate the different elements of knowledge co-production in different phases. For knowledge articulation, consultation methods are often employed, whereas for knowledge integration, collaboration methods are considered more appropriate. Methods for knowledge embedding are often not explicitly included in approaches. Examples of consultation methods are questionnaires, interviews or focus group discussions with homogenous groups. For collaboration various deliberative methods could be employed, like dialogue meetings, working sessions, citizens juries (Abelson et al. 2003a), citizens panels (Abelson et al. 2003a), the Delphi process (Jones and Hunter 1995) and the Nominal Group Technique (Jones and Hunter 1995).

Good process facilitation by an ‘independent’ facilitator is necessary throughout the entire process to create conditions essential for effective knowledge co-production. An ‘independent’ facilitator has no stake in the outcome, but has interest in safeguarding the quality of the process and is trusted by the different actors. A process facilitator strives to avoid exclusion of perspectives and
particularly, in this context, patients’ perspectives (Caron-Flinterman et al. 2006) and to develop mutual trust between actors (Broerse 1998). The process facilitator should be competent in executing transdisciplinary research, be able to incorporate the interests of all actors, know how to match goals to methods, and take transparency and validity into account (Hewlett et al. 2006; Betten et al. 2013).

**Box 2.1. Dialogue Model**

An example of a multi-phased participatory approach in the health domain is the Dialogue Model. This Model has been developed for involving patients in research agenda setting and is based on the Interactive Learning and Action (ILA) approach (Broerse and Bunders 2000). The ILA approach enables end-users to have a prominent role in decisions regarding innovation processes in all societal domains. The Dialogue Model operationalizes consultation of and collaboration between various actors, and is grounded in the notion that involvement is an interactive process between actors. It emphasizes mutual learning processes between actors by means of on-going dialogues, and endeavours to include the perspectives of all relevant actors (Abma 2005a; Abma and Broerse 2010; Broerse et al. 2010; Elberse et al. 2011; Nierse et al. 2011). The Dialogue Model is based on six underlying key principles which specify how the process needs to be conducted: active engagement of patients, conductive social conditions, respect for experiential knowledge, on-going dialogue, emergent and flexible design, and neutral process facilitation.

The model has an emergent design in which activities are roughly structured in six phases.

1. *Initiation and preparation*: A project team is established and relevant actor groups are identified. By means of a desk study and exploratory interviews, first insights are gained into the needs in terms of the process, the scope of the agenda, the problems, ideas and wishes of patients and other actors. Supportive social conditions for genuine involvement are created.

2. *Consultation*: The actor groups are consulted separately, in order to develop a list of research topics relevant from the perspective of each actor group. Separate consultation is needed to deal with the asymmetry between patients and professionals. Focus group discussions, interviews or internet discussions adapted to the needs of the actor group are frequently employed consultation methods.
### Theoretical background

<table>
<thead>
<tr>
<th>3. <strong>Prioritization:</strong></th>
<th>Actor groups value the identified research topics identified in the previous phase and rank them in order of importance. Appropriate methods are questionnaires for large groups and the Delphi Technique or focus group discussions for smaller groups.</th>
</tr>
</thead>
<tbody>
<tr>
<td>4. <strong>Integration:</strong></td>
<td>The prioritized research topics of each actor group are integrated into a single integral research agenda. Dialogue meetings represent appropriate methods to realize integration through deliberation. Mutual learning and creating shared ownership are important elements.</td>
</tr>
<tr>
<td>5. <strong>Programming:</strong></td>
<td>The research agenda is translated into a research programme or action plan.</td>
</tr>
<tr>
<td>6. <strong>Implementation:</strong></td>
<td>Implementation of the research programme/action plan with a call for research proposals, matching of research subjects and departments, and funding of research. Actors (including patients) implement and take action, monitor progress and evaluate results.</td>
</tr>
</tbody>
</table>

Together, these six phases comprise the process of knowledge co-production. The first three phases concern knowledge articulation, phase 4 concerns knowledge integration, and the last two phases refer to knowledge embedding.

#### 2.2 Knowledge articulation

Within the knowledge co-production process, a critical step involves individual actors making their knowledge explicit. This is particularly crucial for patients because their knowledge is often implicit and not readily made explicit. Patients first need to develop their own voice by making their experiential knowledge explicit to equip them for interaction with other actors (Abma and Broerse 2010). This is also known as ‘enclave deliberation’ (Nierse and Abma 2011). Through ‘enclave deliberation’, patients become aware of their perspectives, needs and preferences, grounded in their personal experiences of living with their disease. During enclave deliberation, problems and concerns experienced by patients in their daily lives are identified, together with possible solutions. Without enclave deliberation, patients tend to replicate either the needs and preferences of professionals or the topics currently receiving attention in the media (Baart and Abma 2011; Elberse et al. 2011).

The following section provides insights into ways of articulating the knowledge of the different actor groups with specific attention to enclave deliberation with
patients. To clearly understand knowledge articulation, I first elaborate on the concept of knowledge and then address the following questions: What types of knowledge can be distinguished and what is the distribution of these types of knowledge among actor groups?

*The concept of knowledge*

In this thesis, I make a distinction between three types of knowledge: (1) scientific knowledge, (2) expert knowledge and (3) experiential knowledge (Lehrer 1990; Caron-Flinterman et al. 2005). **Scientific knowledge** is explicit and explanatory. It is acquired through detached and impersonal study and observation, and strives to generate objective facts, universality and absolute truth (Caron-Flinterman et al. 2005; Schipper 2012; Pols 2013, In Press). Subjectivity is eliminated as much as possible. **Expert knowledge** represents the translation of scientific knowledge to care practice (Pols 2013, In Press). It consists of skills and capacities. This type of knowledge is partly implicit, and must be acquired through training and practice (Schipper 2012). **Experiential knowledge** of patients is defined as

“The often implicit, lived experiences of individual patients with their bodies and their illnesses as well as with care and cure” (Caron-Flinterman et al., 2005, p.2576).

These experiences are converted into personal insights that enable the patient to cope with his or her condition in daily life. These insights provide a wider perspective to health research and thereby contribute to increased quality and relevance of health research (Popay and Williams 1996; Entwistle et al. 1998; Goodare and Lockwood 1999).

The nature and status of the types of knowledge have been subject to on-going academic discussion. As a result, understanding of knowledge and the status of the different types of knowledge have changed during the past century. During the first half of the 20th century, scientific knowledge was seen as the most reliable type of knowledge, followed by expert knowledge, while experiential knowledge had a low status. At that time, scholars followed a positivist discourse and argued that true knowledge is derived from objective observations and based on rational arguments (Carnap 1966). In this discourse, experiential knowledge is considered invalid because it lacks objectivity, verifiability, universality or rationality (Caron-Flinterman et al. 2005). During the second half of the 20th century, scholars began
Theoretical background

to consider knowledge from a more relativist perspective. They argued that the process of knowledge production is always influenced by social contexts, cultural contexts and personal interest (Wittgenstein 1953; Latour and Woolgar 1979; Barnes and Bloor 1982). Following this line of argumentation, contemporary scholars acknowledge that all types of knowledge are socially constructed. Caron-Flinterman et al. (2005) added that experiential knowledge should be considered as relevant and distinguished from ‘non-sense claims’ because of its practical value.

Patients’ experiential knowledge came to attention as early as the 1980s, although the term knowledge was not yet used. Instead, medical sociologists referred to ‘patient perspectives’, ‘patient viewpoints’ or ‘lay health beliefs’ (Prior 2003). They studied the use and expression of this knowledge in particular contexts and circumstances, and how this knowledge should be understood (Morgan and Watkins 1988; Backett 1992; Calnan and Williams 1992; Emslie et al. 2001). In later papers, the term lay knowledge was used (Busby et al. 1997; Popay et al. 1998). Prior (2003) argued that this change in terminology was indicative of a higher status being given to the experiences of patients because what had previously been seen as ‘beliefs’ was being seen as ‘knowledge’. Patients have been transposed to multi-skilled and knowledgeable individuals. In addition, the validity of experiential knowledge has been discussed extensively. Despite these new perspectives on patients’ experiential knowledge, researchers continued to argue that patients lack the objective knowledge that would enable them to make relevant contributions to health research (Oliver et al. 2001; Boote et al. 2002), and that they cannot provide valid insights into causes, course and management of disease because they are not experts in health and clinical research (Prior 2003).

Knowledge of actor groups

Most often, knowledge types are automatically assigned to various actor groups in a one-on-one relationship (Caron-Flinterman 2005; Schipper 2012). For example, common sense relates researchers to scientific knowledge, health professionals to expert knowledge and patients to experiential knowledge. However, the distribution of knowledge among actors is more complex. Patients, professionals and researchers possess all three types of knowledge simultaneously (Caron-Flinterman et al. 2005; Schipper 2012), although the distribution of knowledge types and the way this is developed by the various actor groups may differ, as visualized in Figure 2.3. For example, the first and most basic knowledge of
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Researchers is scientific knowledge. However, researchers could also be themselves patients (and thus have experiential knowledge) or could possess some expertise within the clinical practice (expert knowledge). Also, within actor groups many differences are visible. Within an actor group, individual actors are active in different scientific disciplines (researchers), have different medical specialisms (health professionals) or have experience with different types, severity and course of disease (patients). For this reason, the personal context of individual actors and the differences within an actor group should be taken into account when defining the knowledge of various actor groups, as well as the differences within actor groups. The differences within the actor groups are described in more detail below.

Figure 2.3. Visualization of knowledge distribution among actors groups

For health professionals, a distinction can be made between health professionals with a more ‘propositional’ expert knowledge (such as physicians), which is obtained by written and oral knowledge transfer and experimentation, and health professionals with a more ‘practical’ expert knowledge (such as nurses or physiotherapists) acquired in practical training and professional practice (Caron-Flinterman et al. 2005). The former group is more in contact with researchers and less with patients, whereas the latter group has much contact with patients and less with researchers. Furthermore, differences in knowledge distribution can be made among health disciplines (e.g. neurology, gynaecology, oncology, revalidation, geriatrics, psychiatry). Therefore, it could be argued that specialized health professionals (such as brain, liver or heart specialists) are less familiar with patients’ perspectives, compared to health professionals with a more holistic approach (such as revalidation specialists).
Within the actor group of researchers, several types of researchers can be distinguished by research disciplines (e.g. biomedical sciences, natural sciences, social sciences, clinical studies, health care sciences). In general, it could be argued that basic researchers have less contact with care practice and patients, and are more situated on the left in Figure 2.3., while social scientists often have more contact with care practice and patients, and are therefore more situated to the right.

With respect to patients, the literature distinguishes three types of patient—lay patients, patient representatives and professional patient representatives—each possessing a different combination of knowledge types (e.g. Hogg and Williamson 2001; Caron-Flinterman et al. 2005). Lay patients are able to share their personal and bodily experiences, but are not familiar with the experiences of peers and have little or no professional or scientific knowledge. When patients are able to connect their story with the story of peers, they gain experiential expertise (Caron-Flinterman et al. 2005) and can tell a ‘we-story’, which transcends the ‘I-story’. These patients are often actively involved as patient representatives in patient organizations and are in contact with many peers. As a consequence, they are often also familiar with medical information and possess, to some extent, professional and scientific knowledge. When patients also have experience in acting and speaking on behalf of other patients at the policy level, experiential expertise could be extended to proto-professionalism (Caron-Flinterman et al. 2005). Examples of proto-professional patients include patients who work for patient organizations or have followed courses on health research and/or patient advocacy, and who possess considerable expert and scientific knowledge. However, proto-professionalization has a paradoxical effect: on the one hand, it facilitates inclusion in discussions with professionals but, on the other hand, can result in loss of credibility as representative of the patient community (Van de Bovenkamp and Zuiderent-Jerak forthcoming). Lastly, the term ‘patient’ could be considered to refer more broadly to some non-patients, such as professionals who represent patient organizations on a policy level, or informal carers who represent individual patients. When they are unable to represent themselves (e.g. people with severe intellectual disabilities or Alzheimer’s disease) (Boote et al. 2002; Konijn et al. 2011). However, it should be noted that, although these policy makers and informal carers acquire insights into the lives of patients, they do not have the experience themselves. Furthermore, the content of the knowledge of patients differs among individual actors. Patients have experiences with different types of diseases with differences in severity (from minor impact on daily life to life-
threatening) and time-course (recently diagnosed or diagnosed many years ago). In addition, patients may have different experiences due to the life phase they are in (e.g. childhood, adolescent, adulthood (with or without children, with or without work), elderly person).

*Articulating knowledge*

The process of knowledge articulation involves the identification of scientific, expert and experiential knowledge. To this end, different actors are consulted to determine their most basic, dominant knowledge. In general, this means that researchers are consulted for their scientific knowledge, health professionals for their expert knowledge, and patients for their experiential knowledge. Since patients, professionals and researchers possess all three types of knowledge, it is important that the methodology used for knowledge articulation is designed in such a way that the specific knowledge of various actors is retrieved in a valid and reproducible manner, and results in a legitimate product with attention for the diversity within actor groups. The knowledge of researchers and health professionals is more predominantly explicit, and is therefore easier to articulate. The experiential knowledge of patients is often implicit and needs to be identified and converted into suitable input for health-related decisions.

The knowledge articulation process is mainly consultative (see box 2.1) and often employs a ‘mixed-methods approach’ (Creswell and Plano-Clark 2006). This approach relies on the combination of quantitative and qualitative research methods for in-depth, broad consultation. Qualitative methods involve for instance interviews and focus group discussions, and as Malterud (2001, p.483) indicated:

“It is used in the exploration of meaning of social phenomena as experienced by individuals themselves, in their natural context”.

Thereby, qualitative methods can provide detailed and in-depth insight into the knowledge of the actors, which is often articulated as needs, preferences, ideas or opinions (Abma and Broerse 2010). To articulate the experiential knowledge of patients, patients need to become aware of their perspective and corresponding needs and preferences. Since these are grounded in their personal disease experiences, interviews and focus group discussions often start with identifying
problems and concerns in patients’ daily lives. Subsequently, these problems and concerns are converted into possible solutions, needs and preferences. In this way, implicit knowledge is made explicit and can provide detailed, in-depth insights into the knowledge of actors, often articulated as needs, preferences, ideas and opinions (Abma and Broerse 2010).

Although qualitative consultation aims to obtain maximum variation in perspectives/experiences (diversity within actor groups), a relatively small group of respondents is often involved. For this reason, quantitative methods of data collection, such as questionnaires, are conducted with large groups of respondents to validate the insights obtained from interviews and focus group discussions, to maximize the representation and to increase the diversity of actor groups (Abma and Broerse 2010).

2.3 Knowledge integration

Knowledge integration follows knowledge articulation and can be defined as the process of combining the different, explicitly articulated knowledge of researchers, health professionals and patients. However, as Regeer and Bunders (2009) emphasize, this definition relies on a simplistic conceptualization of knowledge integration. The end-product of knowledge integration is constructed through a process of reflexive learning followed by consensus building. This process is complex and not self-evident. The following section provides insights into the complex process of knowledge integration.

Process of reflexive learning

Knowledge integration could be considered as a learning process (Regeer 2009). Learning involves the acquiring of new, adapted or reinforced knowledge, skills, behaviours or values. Learning does not occur at one time, but builds on steps of acquiring, processing, and storing information. Therefore, learning should be viewed as a process (Argyris and Schön 1978; Bruner 1960). Learning implies that actors change in the process; they change in thinking (Caron-Flinterman et al. 2005). This could be in a substantive way (regarding substantial matters), in a procedural way (regarding knowledge co-production processes) and in a reflexive way (regarding their own and others’ perspectives) (Guston 1999; Irvin and
Stansbury 2004). For knowledge integration, learning implies that actors gain insight into each others’ perspectives and underlying assumptions, which may lead to adjustments of opinions, challenging of prejudices and the development of new ideas (Levine and Moreland 2004; Abma and Broerse 2010).

A learning process can be reflected in three interconnected learning loops: (1) single-loop learning, (2) double-loop learning and (3) triple loop learning (Argyris and Schön 1978). In single-loop learning actors are primarily focused on improving their skills and capabilities (a substantive and procedural way of learning) within the range of their (professional) norms. Double-loop learning implies that actors undergo internal structural change in their beliefs. They are able to reflect on their own perspectives and routines and recognize the underlying patterns of their thinking and behaviour. It stimulates a reflexive way of learning. As Argyris and Schön (1978, p. 21) explain:

“The double loop refers to the two feedback loops that connect the observed effects of action with strategies and values served by strategies. Strategies and assumptions may change concurrently with, or as a consequence of, change in values.”

Double loop learning is reinforced by learning in a group – interaction - which creates the possibility to generate a broad set of perspectives and to become acquainted with and question/inquire each other’s underlying assumptions (Levine and Moreland 2004). Furthermore, interaction could also induce a shift in power relations, since actor groups who are generally not included in decisions are now given a platform for their voice (Ryan and Destefano 2001). Interaction can also result in triple-loop learning, which involves actors reflecting on their own learning. Triple loop learning occurs when the dynamics of the interaction are discussed afterwards. It stimulates meta-learning in which:

“persons reflect on and inquire into the process in which single-loop and double-loop learning take place” (Visser 2007, p. 662)

Conducting integration

The process of knowledge integration (learning) can be conducted in several ways. An independent facilitator could integrate the different types of knowledge derived from the process of knowledge articulation. Although a facilitator can ensure symmetrical knowledge integration in which each knowledge type is
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considered equally valid and true, no interaction takes place between actors despite the integration of feedback from all actors. Since actors do not become directly familiar with each other’s perspectives, their thinking regarding knowledge co-production processes and outcomes may not change (Guston 1999; Irvin and Stansbury 2004; Caron-Flinterman et al. 2007). As a result, learning among actors is mainly restricted to single-loop learning. To a certain extent, however, double-loop learning could occur. Actors could indirectly gain insight into each other’s perspectives and underlying assumptions by, for instance, reading summaries of the knowledge articulation of the other actor groups or by observing focus group discussions of other actor groups. This type of double-loop learning is also referred to as *vicarious learning* (observational learning) (Rosenthal and Zimmerman 1978). Also, after knowledge integration, vicarious learning could occur when the outcomes are disseminated among the different actor groups. Vicarious learning could be enhanced, when the perspectives and underlying assumptions of the different actors are extensively described in written materials (e.g. reports) or audio-visuals (e.g. videos on the internet).

Learning is, however, ‘deepened’ through direct interaction between actor groups. To this end, a working group comprising representatives of the various actor groups, a deliberative dialogue meeting or a conference with different actors could be organized. Interaction between actors on a group level implies that actors change in the process. They listen to each other’s perspectives and reflect on them, which may lead to adjustments of opinions, challenging of prejudices and the development of new ideas (Levine and Moreland 2004; Abma and Broerse 2010). As a consequence, interaction facilitates double-loop and triple-loop learning.

*Facilitation*

However, conducting knowledge integration is by no means easy and self-evident. Although the experiential knowledge of patients is now acknowledged as valid, it is often not (yet) valued nor considered to be of an equal status to scientific and expert knowledge because of its subjectivity (Callaghan and Wistow 2006; Dewey 2008). Moreover, the differences between the various knowledge types could hamper integration due to difficulties experienced with reconciling the experiential knowledge of patients (which is specific, personal and context-related) with the scientific and expert knowledge (which has been gained through scientific training and is generated by objective facts) (Caron-Flinterman et al. 2005; Regeer and
To enhance vicarious learning, the facilitator needs to ensure that different perspectives and underlying assumptions of the actor groups are extensively described, and that the integrated knowledge is subsequently disseminated among the actors.

In interaction with actors, more challenges are faced for knowledge integration. Integration of knowledge of actors with different hierarchical and social status is complicated, because of their divergent expectations, views and language (Abma et al. 2009). In interaction with researchers and health professionals, patients’ input is easily overruled since they are not familiar with scientific language (Thompson et al. 2009). Also, they have to gain a position as a ‘reliable knower’ which is complicated by their personalized and narrative mode of communication (Abma et al. 2009). As a result, the process of knowledge integration may result in an asymmetrical end product in which experiential knowledge is undervalued (Elberse et al. 2011). Therefore, interaction between actors needs to be carefully facilitated. A process facilitator can foster a genuine dialogue between asymmetric actors (Caron-Flinterman et al. 2006). It is essential to facilitate conducive social conditions (e.g. openness, respect, trust between actors), which enable the various actors, particularly patients, to share their perspective and avoid exclusion (Abma 2005b; Caron-Flinterman et al. 2006). Moreover, an important task of the facilitator is to create trust between actors. At the start of a knowledge co-production process, the level of trust is usually low because actors have no experience with such processes. The facilitator, as an independent actor, can enhance ‘intermediated trust’ by facilitating the exchange of perspectives between actors prior to interaction (Broerse 1998).

**End-product**

The end-product of knowledge integration is constructed through the aforementioned process of reflexive learning followed by consensus building. The process of consensus building can be described as

> “Actors who will be affected by a decision, work together to develop a solution that meets as many of their individual and collective interest as possible” (Susskind 1999, p. 599).

Furthermore, consensus building should not be confused with negotiation. Negotiation involves discussion with the intention of producing agreement, while
consensus building involves actors thinking together and identifying where a conclusion might lie (Schipper 2012). Consensus building generally implies that the perspectives of individuals converge and that the ‘borders’ between them are no longer visible (Kerkhof 2006). This harmonization or orchestration of various perspectives is often the desired outcome of health-related decisions. Nevertheless, the joint construction is not restricted to convergence. The increased perspectives and number of actors could lead to more uncertainty and to opposing coalitions of solutions. This could result in the formation of a dominant design in which actors tolerate but do not accept opposing solutions (Boon 2008).

2.4 Knowledge embedding

Knowledge embedding involves the sustainable implementation of the process and outcomes of knowledge co-production. When knowledge is embedded, patients have a structural place in decision-making procedures and there is long-term commitment to collaboration between researchers, health professionals and patients to follow-up on the end-product of knowledge integration. Actor groups have regular interaction and share decision-making power (Beresford 2007; Abma et al. 2009; Ward et al. 2010). Knowledge embedding is a rather neglected element of many participatory approaches. Many approaches only focus on the elements of knowledge articulation and integration. Those that do take knowledge embedding into account, such as the Interactive Learning and Action approach (Broerse and Bunders 2000) and the Dialogue Model (Abma and Broerse 2010), include several strategies to enhance embedding of the process and outcomes of knowledge co-production. This section provides insight into requirements for the sustainable embedding of the process and outcomes of knowledge co-production and strategies necessary to enhance this embedding.

Knowledge embedding requires new and alternative ways of thinking, organizing and doing (Caron-Flinterman et al. 2007). Structural patient involvement requires adjustment of structures and procedures, establishment of long-term relationships between actors, building of competences, willingness to involve patients and availability of resources, inclusive of time and money (Howe et al. 2006; Caron-Flinterman et al. 2007; Thompson et al. 2009; Elberse 2012; Gray et al. 2000; Caron-Flinterman et al. 2007). As Caron-Flinterman et al. (2007) describe, the
characteristics of the research community (which is regarded as reductionist and specialized) needs to change in order to provide space for disease-transcending research topics, such as co-morbidity and fatigue (topics with high relevance for patients). Furthermore, researchers focus on topics and questions that ensure scientific acknowledgement and increase publication rates, but there is usually little attention for patient relevance (Caron-Flinterman et al. 2007). Current financing structures and procedures often support this focus by almost exclusively assessing project proposals on the basis of scientific criteria. In regard to the patient community, scientific literature emphasizes that patients often lack self-confidence. They find it difficult to voice their opinion concerning research and do not always consider their involvement in research a priority (Gray et al. 2000; Boote et al. 2002; Caron-Flinterman et al. 2007).

The many requirements described above indicate that a profound change is needed regarding the primarily supply-driven health system towards a more needs-oriented health system, in which patients have a stronger voice (Broerse et al. 2010). However, every system, including the health system, has fixed routines with clear power relations between actors that structure daily activities, communication and decision-making (Beresford 2007; Bunders et al. 2010). Actors in a system tend to standardize or normalize their actions and interactions. Changes, such as the involvement of patients in knowledge co-production processes, destabilize the shared and fixed routines in a system, and are often countered by actors involved (Caron-Flinterman et al. 2007). Therefore, changing of systems, also called transitions, are often slow processes, which can take one or two generations (Rotmans 2005; Loorbach 2007).

Although insights into enhancement of knowledge embedding are relatively scarce, various strategies have been formulated. First, small, pilot experiments could enhance knowledge embedding, since they provide a safe environment to develop and test new patient involvement practices (Rotmans 2005; Loorbach 2007). Within these experiments, it is important that learning processes continue to further stimulate internal change in beliefs of actors, and improve their skills and capabilities (Regeer 2009). To this end, learning cycles emerge. Furthermore, to stimulate willingness of actors, it is essential that they recognize the additional value of knowledge co-production processes. Therefore, these small experiments should not only have effect in the long-term, but should also result in so-called ‘quick wins’ (results of knowledge co-production that could be realized in the short term) (Roelofsen et al. 2011). Second, coalition building is essential for realizing
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sustainable implementation of the process and outcomes of knowledge co-production (Caron-Flinterman et al. 2007). A broad network of actors with a willingness to involve patients in health-related decisions will stimulate broadening and scaling-up of knowledge co-production experiments. These actors should be thought of as ‘enthusiasts’ (people who want to be involved and to share their perspectives and ideas) (Elberse 2012), and not only involve health professionals, researchers and patients, but also policy-makers of government authorities and funding agencies. The latter actor group could, for instance, place patient involvement on ‘the agenda’ and allocate necessary resources.

2.5 Criteria for effective knowledge co-production

The involvement of patients is expected to improve the quality of health-related decisions. However, knowledge co-production processes require an extra investment of time and money of the different actors. When these extra investments do not improve the quality of health-related decisions or when the additional value is not visible, actors may become less willing to invest in patient involvement in the future (Elberse 2012). For this reason, it is important to assess the effectiveness of knowledge co-production.

To decide whether knowledge co-production processes are effective, various scholars have formulated evaluation criteria or conditions and principles for successful public or patient involvement (Laird 1993; Webley and Tuler 1995; Guston 1997; Guston 1999; Rowe and Frewer 2000; Webley and Tuler 2000; Driessen et al. 2001; Abelson et al. 2003a; Rowe and Frewer 2004; Abelson and Gauvin 2006; Caron-Flinterman et al. 2006; Broerse et al. 2009). Since both the process and the outcomes of knowledge co-production are considered relevant, evaluation criteria detail key measures of whether patients are meaningfully and actively involved (process), and whether their involvement leads to actual change (outcomes) (Driessen et al. 2001; Rowe and Frewer 2004; Abelson and Gauvin 2006; Caron-Flinterman et al. 2006; Oliver et al. 2008; Broerse et al. 2009).

Within the three elements of knowledge co-production (articulation, integration and embedding), patient involvement can be assessed using the following criteria:

- **Involvement of patients**: Attention should be paid to the balance between involved patients, researchers and professionals (Elberse et al. 2011);
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representativeness and diversity among the patient population; saturation of
data, and active involvement of patients (Abma and Broerse 2010).
- **Process structure:** Processes should preferably be multi-phased, have an
  emergent and flexible design, and make use of a variety of tools and methods
  (Abma and Broerse 2010). Furthermore, patients should be informed about
  the aim of their involvement and what influence they have on the decision-
  making (Rowe and Frewer 2000; Webler and Tuler 2000; Caron-Flinterman et
  al. 2006). Also, there should preferably be direct interaction between patients,
  researchers and health professionals during knowledge integration. And
  finally, patients should be involved in significant aspects of decision-
  making to ensure use of patients’ input (Webler and Tuler 2000; Rowe and Frewer 2004;
  Caron-Flinterman et al. 2006).
- **Process management:** Independent process facilitation of patient involvement
  is important to create trust among actors (Broerse 1998), ensure equal
  treatment of patients and respect for patient knowledge, to prevent exclusion
  and to create good social conditions (Abelson and Gauvin 2006; Broerse et al.
  2009; Abma and Broerse 2010). Furthermore, attention should be given to
  support involvement and to adjust activities to the abilities of patients.

Two criteria are particularly relevant for the outcomes of knowledge co-
production:

- **Direct outcomes:** The end-result (decisions and related actions) should reflect
  the input of patients, with particular attention paid to the degree to which
  patients’ inputs are included and what is taken up and why (Driessen et al.
  2001; Irvin and Stansbury 2004). Patients and other involved actors should be
  satisfied and consider the end-result as legitimate and relevant (Abelson and
  Gauvin 2006; Broerse et al. 2009). Moreover, the outcomes should be
  reasonable and well translatable into health-related decisions, and have
  impact on health-related decisions (Driessen et al. 2001; Caron-Flinterman et
  al. 2006).
- **Indirect outcomes:** Indirect outcomes are related to the stimulation of learning
  processes and the achievement of double-loop and triple-loop learning,
  resulting in changes to the thinking of both patients, researchers and health
  professionals (Argyris and Schön 1978; Caron-Flinterman et al. 2006; Broerse
  et al. 2009).
Despite the growing attention for involving patients in health-related decisions and the development of specific approaches, the impact of patient involvement on decision-making processes and the outcomes remain relatively low (Boote et al. 2002; Oliver et al. 2008; Staniszewska et al. 2008). Apparently, knowledge co-production processes are not entirely effective. Several constraints are observed in the three elements of the knowledge co-production process.

Knowledge articulation receives much attention in international literature and a wide range of appropriate and validated methods are available (Boote et al. 2010; Barber et al. 2011). Despite this attention, many difficulties are still experienced in practice. In particular, scholars struggle with representativeness and diversity of the actors. Consultation initiatives generally aim at maximum diversity of respondents but, often, a relatively small group is included. For health professionals, often nurses and physiotherapists are neglected, as well as general practitioners. Most often, researchers involved in for instance implementation research are not included. And for patients, the most vulnerable groups, such as seriously ill patients, patients with a low socio-economic status and patients from ethnic minorities, are often difficult to reach and thus not included (Hogg and Williamson 2001). The inclusion of a relatively small group is often also related to the limited availability of financial resources and time (Elberse 2012). To this end, scholars often struggle in finding a balance between, on the one hand, acquiring maximum representativeness and diversity of the actors with, on the other hand, limited time and money, in order to perform a legitimate knowledge articulation process.

Knowledge integration is not a self-evident step in the knowledge co-production process, and the integration process is often constrained by asymmetry. For instance, patient involvement can create dilemmas in terms of loss of scientific quality and authority (Bijker et al. 2009). Patient knowledge is still often considered to be subjective (Telford and Faulkner 2004). Experiential knowledge is often assigned a lower scientific status which can lead to exclusion of the patient perspective from the process of knowledge production (Elberse et al. 2009). The asymmetrical relationships between parties – often caused by differences in hierarchical position and status – further complicate this dilemma and add to concerns regarding the representativeness of patient involvement (Boote et al. 2002; Oliver et al. 2008; Staniszewska et al. 2008).
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In addition, the different types of knowledge differ greatly from each other, since they are formulated from different backgrounds and in different contexts. For instance, knowledge of researchers and health professionals is often very specifically formulated and focused on medical applications, whereas the knowledge of patients is often formulated starting from a problem they experienced or from a deficiency in daily practice, rather than being specifically focused on medical applications (Caron-Flinterman et al. 2005). As a consequence, it is considered difficult to integrate the different knowledge types into one outcome.

Knowledge embedding is particularly challenging. In the last decades, patients, researchers, health professionals and policy makers have had the opportunity to become familiar with the concept of patient involvement, to experiment with appropriate strategies and methodologies, and to experience the additional value of experiential knowledge on health research, policy and care practice. One difficulty facing knowledge embedding concerns the continuation of patient involvement once the knowledge integration process has been completed. Currently, patient involvement initiatives are often one-off events with a limited time frame (Stevens et al. 2003; Caron-Flinterman et al. 2007; Broerse and Bunders 2010). Finance generally stops once the process of knowledge integration has been completed. It is often assumed that knowledge embedding will follow naturally but this rarely happens in practice. For the structural involvement of patients during knowledge embedding, long-term commitment for collaboration is required. Changes to structures and competences are required to maintain an ongoing dialogue between patients and professionals, and to give patients a sustained place in decision-making structures. However, these changes are usually not put into practice because social systems are resistant to change and few strategies are available to overcome resistance (Caron-Flinterman et al. 2007; Jinks et al. 2009; Broerse and Bunders 2010). As a consequence, after knowledge integration, each actor group goes back to their day-to-day routines; it is business-as-usual.

The above indicates that more insights are needed into constraints in, and strategies for, realizing effective knowledge co-production processes in health research, policy and care in the three elements of the process.
RESEARCH DESIGN
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Chapter 3 - Research design

In this chapter, the research design is presented. First, the objectives and the main research question are presented. Next, I describe the research approach, followed by a description of the various cases and research methods. The chapter ends with issues of validity and the outline of the thesis.

3.1 Main research question and study questions

The difficulties in involving patients in decision-making processes in health research, policy and care practice, presented in Chapter 2, suggest that new insights are needed into how knowledge co-production processes could be optimally structured. In line with these findings, and with the primary aim of the thesis, as presented in Chapter 1, the current research has been guided by the following research question:

*How can knowledge co-production processes be structured so that patients’ knowledge is effectively articulated and integrated with knowledge of researchers and health professionals, and can be embedded in health research, policy and care practice?*

The three main concepts in the research question correspond with the key elements of knowledge co-production, namely (1) knowledge articulation, (2) knowledge integration, and (3) knowledge embedding. Against the backdrop of these three elements, presented in Chapter 2, the main research question is divided in three study questions:

1. What strategies can best be employed to articulate the knowledge of researchers, health professionals and, in particular, patients?
2. How can patients’ knowledge be effectively integrated with the knowledge of researchers and health professionals?
3. How can the embedding of knowledge co-production processes in health research, policy and care practice be enhanced?

3.2 Research approach

In the thesis, the study questions are addressed using a multiple case study approach (Eisenhardt and Graebner 2007; Yin 2009). Case studies are often used in social sciences to gain a better understanding of complex processes in relation to their context. They provide the opportunity to apply different methodologies, such as desk studies, interviews, observations, focus group discussions and dialogue meetings, often in various combinations.

Patient involvement is a complex, heavily context-dependent phenomenon and, in combination with the main question posed in this research (a ‘how’ question), is very suitable for a case study approach. The complexity of patient involvement is a result of the diversity of actor groups (e.g. research institutes, funding agencies, health authorities, patient organizations), research areas and diseases. For this reason, patient involvement initiatives
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represent a broad, complex real-life context, with different challenges and difficulties. The holistic approach of case studies provides the opportunity to conduct in-depth analyses, to validate and to understand the complexity of knowledge co-production processes in the context of patient involvement initiatives. This approach takes into account this complexity, not only by acknowledging the importance of various elements (such as the different actors, research area or diseases) but also by providing insights into the decisions that are made and other specific characteristics of the cases.

3.3. Case selection and research methods

The research in this thesis is based on multiple cases, each addressing different elements of the research question. Together, the cases provide a frame of analysis which allows an in-depth insight into ‘how’ knowledge co-production processes take place within the area of patient involvement and ‘how’ these may be improved.

The cases were selected using the following criteria:
1. The cases are situated in the field of health research, policy or care practice, where knowledge articulation, integration and embedding occur.
2. In the cases, patients make a significant contribution to the end-product and are involved, as a minimum, at the level of consultation.
3. The cases differ in terms of their contexts and strategies of patient involvement, making it feasible to compare different approaches.
4. The cases take place in a real-life setting which implies that actors in the field took the initiative for patient involvement.
5. The case are accessible for in-depth investigation.
6. Each case provides the opportunity to answer at least one study question.
7. The cases together cover all study questions posed in this thesis.

Based on these criteria, four (single and multiple) cases were selected for this thesis. The relationship between the four cases presented in this thesis, on the one hand, and the health domains and key concepts of the three study questions, on the other hand, are summarized in Table 3.1 and described in detail in the following paragraphs.

Table 3.1. Selected characteristics of the four cases included in the thesis

<table>
<thead>
<tr>
<th>ID</th>
<th>Topic</th>
<th>Type of case</th>
<th>Health domain</th>
<th>Articulation</th>
<th>Integration</th>
<th>Embedding</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Policy advisory process</td>
<td>Single</td>
<td>Health policy and research</td>
<td>Yes</td>
<td>Yes. Externally by a working group, including a patient representative</td>
<td>Add on. Attention for dissemination of the end-results</td>
</tr>
<tr>
<td>2</td>
<td>Clinical guideline development</td>
<td>Single</td>
<td>Care practice</td>
<td>Yes</td>
<td>Yes. Externally by an independent process facilitator</td>
<td>Yes. Integrated in the approach</td>
</tr>
</tbody>
</table>
The first three cases – policy advisory process, clinical guideline development and clinical trials – are single cases and provide insights into knowledge articulation, knowledge integration and knowledge embedding. They, thus, contribute to answering all three study questions proposed in this thesis. Although knowledge embedding was not an integral part of the approach in cases 1 (scientific advisory process) and 3 (clinical trials), these cases do provide interesting insights into how attention was paid to this element of the knowledge co-production process. The last case is a multiple case study that evaluated follow-up activities after agenda setting (knowledge integration) in nine cases. This multiple case study focused specifically on the embedding of patient involvement in procedures and structures of funding agencies with respect to programme development and implementation, providing insights that were particularly relevant for the last study question. Processes of knowledge articulation and integration were not explicitly studied in this case study, although many of the cases had previously been undergone thorough evaluation of articulation and integration.

In line with the case study methodology described above, the four cases in this thesis employ a range of multi-phased, qualitative approaches (table 3.1) with a variety of data collection methods, ranging from desk studies, observations, semi-structured interviews, focus group discussions and dialogues between actors. A case description of each case and an overview of the research methods used in each case are presented in the following paragraphs. A detailed description of the research approaches, data analyses and results can be found in Chapters 4 to 9.

**Case 1: Patient involvement in a policy advisory process**

The first case, presented in Chapter 4, concerns the involvement of a broad range of patients in a Dutch policy advisory process for the development of medical products. This resulted in an advisory research agenda for the future development of medical products for 15 different disease domains. Box 3.1 describes the context and questions that guided the research carried out in case 1.

**Box 3.1. Patient involvement in a policy advisory process - Case description**

In 2010, the Dutch Minister of Health, Welfare and Sport (in Dutch: Volksgezondheid, Welzijn en Sport, VWS) requested advice from the Health Council (in Dutch: Gezondheidsraad, GR) on a research agenda for the development of medical products (drugs, medical devices related to diagnosis and care, and tissue replacement products). The GR is an independent advisory board, which provides advice on public health policy, both on request and spontaneously, to the Minister.
The appointed GR committee, comprising of twelve members \(^2\) of which one was a patient representative, designed a project to address the Minister’s request (Gezondheidsraad 2010; Gezondheidsraad 2011). The project consisted of three parts that answered the following questions:

1. What is the current position of the Netherlands in the field of medical product development?
2. Which medical products are needed according to patients, professionals and informal carers?
3. How can medical product development be stimulated in the Netherlands?

Question 1 was addressed by the GR staff. To answer question two, the GR approached the Athena Institute (VU University Amsterdam). Although consultation of important actors in the field, such as scientific and industrial experts on the supply side, would have been more common, the committee decided that also the involvement of patients, informal carers and health professionals was essential for establishing an agenda responsive to the needs of the ‘users’ (demand side). Representatives of industry (‘producers’) were consulted by the GR Secretariat in a process parallel to the consultation of ‘users’. Advice on how to stimulate development of medical products was collected by the GR (Question 3).

Case 1 provided the opportunity to identify the medical products needed by patient groups of fifteen disease domains. In this case, the researchers had a central role in designing and conducting the research, as well as in analyzing the results and evaluating the process. This central role provided opportunities to examine the data from a close proximity and analyse the data in the context they took place.

The approach to knowledge co-production applied in the case was based on the Dialogue Model, an approach which operationalizes knowledge co-production in the field of research agenda setting (see Box 2.1 in Chapter 2) (Abma and Broerse 2010), and comprised four phases: (1) exploration, (2) consultation and prioritisation, (3) integration, and (4) follow-up. The exploratory phase consisted of a desk study and 29 interviews with patient representatives of the 15 disease areas. Knowledge articulation took place in the second phase and involved the separate consultation of patients. In total 15 focus group discussions were organized with 97 patients, three patient representatives and 49 informal carers; and 33 interviews with 16 patients, 15 patient representatives and two informal carers. Knowledge integration occurred in the third phase in which the researchers facilitating the process combined the priorities of the 15 disease domains into a single patient research agenda, providing input to the advisory report of the GR. The follow-up phase gave insight into knowledge embedding, since it provided insights into how the process and outcomes were perceived by the actors. This involved additional interviews with three members of the GR committee and informal conversations with consulted patients, following interviews, focus group discussions and a feedback meeting (organized by the GR committee).

**Case 2: Patient involvement in clinical guideline development**

The second case, presented in Chapter 5, focuses on the involvement of patients in the development of a clinical guideline for resumption of employment after gynaecological surgery. The context of this case is presented in Box 3.2. The research conducted within this thesis, as part of case 2, is detailed below.

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\(^2\) The GR committee comprised twelve members, of which six were professors in different medical disciplines, two were representatives of the industry, three were representatives of the government (ministry of Economics and Health, Welfare and Sport), one was a patient representative, and two were from the GR staff.
The development of the clinical guideline for resumption of employment after gynaecological surgery was initiated by the department of Obstetrics and Gynaecology and the EMGO Institute for Health and Care Research, both of the VU University Medical Centre (Amsterdam, the Netherlands). The clinical guideline development is financed by the Netherlands Organization for Health Research and Development, ZonMw.

The guideline development process consisted of two parallel processes which resulted in two products: (1) a clinical guideline with recommendations for resumption of employment after gynaecological surgery, and (2) a web-based patient version (e-health intervention) contributing to implementation of the clinical guideline. Both processes comprised several stages and were interconnected. Knowledge articulation of patients and health professionals took separately place. Health professionals were consulted by means of a Delphi Study and the outcomes formed the primary input for the development of recommendations for the clinical guideline. Patients were mainly consulted for the development of the web-based patient version of the guideline (knowledge articulation). Knowledge integration occurred by one clinician and two project leaders who monitored the process and integrated the data obtained from consultation of patients and professionals. The recommendations of the clinical guideline were integrated into the e-health intervention (knowledge embedding), and patients’ needs and preferences influenced the topics of the recommendations.

Patients were involved in the guideline development process at three different stages: (1) patients participated in three focus group discussions which were organized to identify the problems and needs of patients regarding peri-operative care and counselling on resumption of employment; (2) patients were involved in the development of the script for an instruction video, part of the web-based patient version of the clinical guideline, and (3) patients tested and evaluated the web-based patient version of the clinical guideline.

The effectiveness of patient involvement in this case was assessed by means of Reflexive Monitoring in Action (Grin and Weterings 2005; Regeer et al. 2009). Reflexive Monitoring in Action stimulates learning processes by enhancing reflection and dialogue between actors on the process and the outcomes of projects. This method is facilitated by a monitor who observes the process, gathers relevant data and reflects with actors on the activities, in order to optimize the further process.

In this case, the involvement of gynaecological patients was assessed using a monitoring and evaluation framework, which was developed based on a literature review. The framework comprised pre-defined criteria for the process and the generated outcomes (Laird 1993; Webler 1995; Guston 1997; Guston 1999; Rowe and Frewer 2000; Webler and Tuler 2000; Driessen et al. 2001; Abelson et al. 2003a; Rowe and Frewer 2004; Abelson and Gauvin 2006; Caron-Flinterman et al. 2006; Broerse et al. 2009), which were divided in several sub criteria. Process criteria were subdivided in involvement of actors, process structure and process management. Within outcome criteria, a distinction was made between direct and indirect outcomes. The involvement of gynaecological patients was considered effective when the criteria of the framework were met. The monitor observed the process, gathered the data for the assessment and reflected with the actors on the activities. To this end, a triangulated method was used, involving direct observations, document analysis, semi-structured interviews, informal conversations with patients and with guideline development project leaders, and evaluation forms after the focus group discussions.
Case 3: Patient involvement in breast cancer clinical trials

The third case, presented in Chapters 6 and 7, took place in the field of clinical trials. Patients were involved to identify potential improvements to breast cancer clinical trials (Chapter 6) and possibilities for patient involvement in the design and conduct of breast cancer clinical trials (Chapter 7). The case is described in Box 3.3, and the research is detailed below.

Box 3.3. Patient involvement in breast cancer clinical trials - Case description

The Dutch Cancer Society (KWF Kankerbestrijding) aims to give patients a stronger voice in its research policy by actively involving patients in clinical research. In order to improve the quality of breast cancer clinical trials, a collaboration was established between the Dutch Breast Cancer Trialists Group (BorstkankerOnderzoeksgroep, BOOG) which coordinates breast cancer clinical trials in the Netherlands, and the Breast Cancer Patient Organization (Borstkanker Vereniging Nederland, BVN). Given that patient involvement in clinical trials is a relatively new field with few best practices, a research study was first undertaken by the three organizations to identify patients’ experiences with clinical trials. The three organizations then approached the Athena Institute (VU University Amsterdam) to design and conduct a qualitative participatory study (a knowledge co-production process). The study aimed to identify potential improvements to breast cancer clinical trials and the possibilities for patient involvement in the design and conduct of these trials, including both the perspectives of patients and health care professionals. The study was financed by the Dutch Cancer Society.

Given that the research project aimed to identify possibilities for patient involvement in breast cancer clinical trials, the active involvement of patients was seen as a necessity and patients were given a central role in data processing. An advisory board of patients was established to develop an advice based on the outcomes of the patient consultation. Moreover, BOOG and BVN were committed to implement the outcomes into daily practice. They therefore decided to pay attention to concrete ideas for implementation of the advice of patients.

A knowledge co-production process was developed based on two models: the Dialogue Model which provides tools for consultation and dialogue between actors (see Box 2.1, Chapter 2) (Abma and Broerse 2010) and the FIRST-model which provides guidelines for patient involvement in a research project over a longer period of time (Hewlett et al. 2006; de Wit et al. 2013). FIRST stands for the elements Facilitate, Identify, Respect, Support and Training, for which conditions are described to achieve successful patient involvement.

The research project comprised four phases: (1) exploration, (2) consultation, (3) advice and (4) dialogue. In the exploration phase, three semi-structured interviews were held with patients. In the consultation phase, knowledge articulation took place by separately consulting patients and health professionals. One focus group discussion was organized with eight patients who had taken part in a clinical trial in the last four years. In addition, 20 semi-structured interviews were held with 14 primary diagnosed female patients who participated in a clinical trial and had no metastases; three patients who had decided not to participate in a clinical trial,; and three male patients. For the consultation of professionals, two focus group discussions (respectively four clinicians and nine research nurses) were organized. In the next phase – advice – knowledge articulated was further deepened. To this end, the outcomes of the consultation phase were used to develop advice on how clinical trials could be optimized and how patient involvement could be brought about. The advice was developed by an advisory board of seven patient representatives. Knowledge integration occurred during the dialogue meeting, in which the final advisory report
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was discussed with different actors who identified opportunities for implementation. Knowledge embedding was in this case limited to planning of implementation of the advice.

Case 4: Evaluation of patient involvement in follow-up of agenda setting

The fourth case, presented in the Chapters 8 and 9, consisted of the evaluation of patient involvement in nine research agenda setting projects. Specific attention was paid to the sustainability of patient involvement in the follow-up phases of programming and implementation. Chapter 8 provides insights into whether patients’ topics are translated into a funding program and are taken up by researchers. Chapter 9 describes how the Dialogue Model (see Box 2.1) can be optimized in order to increase patient involvement in follow-up activities of research agenda setting. This case is described in Box 3.5, and the research is detailed below.

Box 3.5. Evaluation of patient involvement in follow-up activities of agenda setting - Case description

Between 2003 and 2009, the patient perspective was included in the research agenda setting to design the research policy of six disease-specific charity foundations and a governmental funding agency (the Netherlands Organization for Health Research and Development, ZonMw). This resulted in nine research agendas focusing on the following diseases: asthma/Chronic Obstructive Pulmonary Disease (COPD) (Caron-Flinterman et al. 2005), update and extension of asthma/COPD and rare lung disease (Elberse et al. 2012b), renal failure (Abma et al. 2009; Schipper and Abma 2011), diabetes (Broerse et al. 2006), burns (Broerse et al. 2010), congenital heart disease (Elberse et al. 2011), spinal cord injuries (Abma 2005a); intellectual disabilities (Nierse et al. 2006); and neuromuscular disease (Nierse et al. 2012). The first six projects were commissioned by disease-specific charities, while the latter three projects were initiated and financed by a governmental research funding agency.

To include multiple actors in research agenda setting, the Dialogue Model was developed (see Box 2.1). The first four phases of the Dialogue Model have often been executed by an academic institute and have been validated and improved extensively in the settings described above. The subsequent phases of programming and implementation of the research agendas are often mainly the responsibility of the bodies commissioning the research. However, experiences so far indicate that patient involvement in research agenda setting is not sufficient in itself to bring about more needs-oriented health research, because patient involvement in subsequent phases of programming and programme implementation seems limited. Therefore, the programming and implementation phases of these nine Dutch research agenda setting projects were assessed/studied in order to increase insights into follow-up activities.

The study took place between November 2010 and April 2012, and used the responsive evaluation methodology (Stake 1975; Guba and Lincoln 1989; Greene 2001; Abma 2005b). Responsive evaluation is a process-oriented approach and is based on a constructivist theory. It assumes that all human beings give meaning to their experiences. Construction is influenced by personal background but outcomes of a socio-cultural process are also influenced by socio-structural and actor positions. The approach focuses on the involvement of actors and criteria for evaluation are based on the dialogue between these actors. The aim of this dialogue is to make the issues of different actors explicit and to facilitate the joint discussion of these issues. Together, the various complementary actor perspectives will lead to a more informed understanding of what is evaluated.

Nine research agenda setting projects were selected for the study. They all involved patients and used the Dialogue Model. The assessment of the nine cases focused on the follow-up activities (programming and
implementation) of research agenda setting, and consequently focused on the element of knowledge embedding. The assessment comprised four phases: (1) exploration, (2) in-depth evaluation, (3) validation and integration, and (4) implementation. The first phase consisted of document analysis and eight exploratory interviews. The second phase was carried out through 46 in-depth interviews with 12 policymakers, 12 representatives of patient organizations, 12 health-specific researchers, four qualitative researchers (involved in the execution of the research agenda setting), and six project leaders. The third phase consisted of three (heterogeneous) feedback meetings with 23 participants (11 representatives of patient organizations, 11 policy makers of funding agencies and one researcher). In the fourth phase, a conference was organized with 33 representatives of patient organizations, funding agencies and research institutes.

3.4 Validity

Qualitative research requires specific procedures to enhance the validity of the outcomes, which stimulates researchers to be critical towards data collection, analysis and interpretation. In this thesis, internal and external validity were enhanced using several procedures (Guba and Lincoln 1989).

**Internal validity**

Internal validity concerns the ‘true value’ of the results of a qualitative study and the extent to which they correspond to the broader environment (Guba & Lincoln, 1989). In order to minimize the effects of researchers’ bias and to enhance the internal validity of the results, the following strategies were used:

- **Triangulation of data:** Several data collection methods, such as desk studies, interviews, focus group discussions, and observations, were used in order to reduce researchers’ bias and limitations of a single method. In all cases, two or more researchers were involved in conducting the research, and analyzing and interpreting the findings, focusing on comparing contested findings.

- **Data saturation:** In all cases, saturation of data was sought, as far as possible. As such, interviews and focus group discussions were organized until no new perspectives emerged. Moreover, recruitment aimed to obtain maximum variation so that insights could be obtained on the entire range of perspectives/experiences. According to literature, data saturation happens after approximately 12 in-depth interviews (Guest et al., 2006).

- **Data processing and analysis:** In all cases, data was extensively documented. Interviews, focus group discussions and dialogue meetings were audio-taped and verbatim transcribed. Research logs were kept to document observations and informal conversations. Analysis of the data occurred by an inductive approach, comprising three steps: (1) open coding (identifying, categorizing and describing of concepts), (2) axial coding (creating subthemes by relating codes to each other) and (3) selective coding (developing storyline by relating subthemes to main themes).

- **Member checks:** Respondents of interviews, focus group discussions and dialogue meetings received the summary of the respective transcripts and were invited to react and reflect on the researcher’s interpretations of the data (member checks) (Meadows & Morse, 2001). Also, in all cases, except for one (namely, case 2), preliminary findings from the study were presented to and discussed with respondents in later phases of the research, in order to validate findings and to minimize misinterpretations and misunderstandings.
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External validity

External validation concerns the generalization of the data and whether the findings are transferable to other contexts. The cases included in this thesis differed in terms of contexts and strategies used for patient involvement. However, each case included an extensive description of the research context, approach, analysis and results, in order to allow readers to determine the extent to which results presented here apply to the characteristics and/or conditions of their contexts ('petite generalizations') (Abma 2005b). Furthermore, by comparing findings to similar studies in scientific literature, the validity of the findings was further strengthened beyond the cases studied.

Ethical considerations

The current research took ethical considerations into account. To this end, all cases aimed to obtain consent for involvement, self-determination, minimization of harm, anonymity and respect for privacy and confidentiality. The researchers provided the respondents with sufficient information about the project to allow them to make a well thought through decision (consent for involvement). Respondents had the opportunity to withdraw at any time during the research and they could decide for themselves which knowledge they wanted to share (self-determination). Respondents were not put at risk or were done any harm (minimization of harm). Data were processed anonymously to protect identity of respondents (anonymity) and was kept confidential (respect for privacy and confidentiality).

Accredited Medical Research Ethics Committee approval was not needed for this research because involvement was not related to treatment or gathering of medical data, was non-invasive, done on a voluntary basis and guaranteed the anonymity of respondents.

3.5 Research teams

The individual cases were executed by a research team. The research teams varied in composition and are presented in Box 3.6. In all case studies, the author of this thesis was involved as one of the main researchers.

<table>
<thead>
<tr>
<th>Box 3.6. Overview of research team members (and their roles) within each case</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Case 1: Patient involvement in a policy advisory process</strong></td>
</tr>
<tr>
<td>- Carina Pittens, MSc a (researcher)</td>
</tr>
<tr>
<td>- Dr. Janneke Elberse b (researcher)</td>
</tr>
<tr>
<td>- Janine van de Kraats, MSc c (researcher)</td>
</tr>
<tr>
<td>- Marjolijn van Wijk a (research intern, master student)</td>
</tr>
<tr>
<td>- Prof. Dr. Tjard de Cock-Buning a (facilitator)</td>
</tr>
<tr>
<td>- Prof. Dr. Jacqueline Broerse a (project leader and facilitator)</td>
</tr>
<tr>
<td><strong>Case 2: Patient involvement in clinical guideline development gynaecological surgery</strong></td>
</tr>
<tr>
<td>- Saskia van Veen, MSc a (researcher, monitor and evaluator)</td>
</tr>
<tr>
<td>- Carina Pittens, MSc a (researcher, monitor and evaluator)</td>
</tr>
<tr>
<td>- Prof. Dr. Maurits van Tulder b (advisor)</td>
</tr>
<tr>
<td>- Prof. Dr. Jacqueline Broerse (project leader)</td>
</tr>
</tbody>
</table>
3.6 Outline of the thesis

This thesis is organized as follows. The first three chapters introduce the rationale and the aim of this thesis (Chapter 1), present the theoretical framework and practical context (Chapter 2), and clarify the research design by briefly describing the research approach and the four cases (Chapter 3).

The Chapters 4-9 of this thesis present the findings of the study. These chapters have already been published, are accepted for publication or submitted for publication (and currently under review). They are slightly adapted to ensure consistency of terminology and reference style.

The first case – a policy advisory process – in which a broad range of patients was involved in a policy advisory process for the development of future medical products is analysed in Chapter 4. Chapter 5 presents the second case, which focuses on the involvement of patients in the development of a clinical guideline for resumption of employment after gynaecological surgery. The third case about clinical trials is presented in Chapters 6 and 7. Chapter 6 presents the involvement of patients in the identification of potential improvements to breast cancer clinical trials, whereas Chapter 7 describes the possibilities for patient involvement in the design and conduct of breast cancer clinical trials from the perspective of patients. Chapters 8 and 9 present the fourth multiple case
study of an evaluation of follow-up activities in nine research agenda setting projects. **Chapter 8** provides insights into whether patients’ topics are translated into a funding programme and are taken up by researchers. **Chapter 9** describes how the Dialogue Model can be optimised in order to increase patient involvement in follow-up activities of research agenda setting.

In **Chapter 10** the main research questions and the three study questions are answered based on the findings of the four cases. These answers are subsequently discussed and placed in a broader context.