Summary

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. Health-related decisions became no longer the exclusive responsibility of researchers and health professionals. This attention for involving patients in health-related decisions is related to two major developments in society. The first development 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. 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.

The involvement of patients in health-related decisions can be defined as patients being actively involved in, and having influence on, decision-making processes in health research, policy and care practice. Patients can be involved in different types of health research, and involvement in decision-making could occur in all phases of the research cycle. Also in health policy and care practice, patient involvement covers a broad area, ranging from involvement in procedures of funding agencies, to clinical guideline development and shared decision-making.

The involvement of patients in health-related decisions can be 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). Second, patients have gained specific knowledge – experiential knowledge – since they are confronted on a daily basis with the consequences of their disease/condition. This experiential knowledge complements the knowledge of researchers and health professionals, and thereby improves the quality of health-related decisions (substantive argument). Third, the legitimacy of health-related decisions increases when patients are involved (political argument).

The involvement of patients in health-related decisions implies that patients should be included in the process of knowledge co-production. Knowledge co-production in the context of health represents the development of knowledge 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.

The process of knowledge co-production has three key elements: (1) knowledge articulation, (2) knowledge integration and (3) knowledge embedding. The process entails the development of appropriate strategies for articulating, integrating and embedding the knowledge of the different actors (e.g. researchers, health professionals and patients). 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. By means of an interactive process, patients’ lived experiences are converted from implicit knowledge into explicit knowledge (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). The knowledge is embedded in health research, policy and care practice.

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 health-decision processes. 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.

Therefore, 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 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. In line with this primary aim of the thesis, the research in this thesis 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, 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?

Research design

The research question in this thesis is addressed using a multiple case study approach. In total, four cases have been selected, each addressing different elements of the knowledge co-production process and situating the field of health research, policy and care practice. 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 first three cases, concerning patient involvement in (1) a policy advisory process, (2) a clinical guideline development and (2) clinical trials, are single cases and provide insights into knowledge articulation, knowledge integration and knowledge embedding. The last case is a multiple case study that evaluated patient involvement in 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, as described above. In the four cases knowledge co-production was applied by a range of multi-phased, qualitative approaches with a variety of data collection methods, ranging from desk studies, observations, semi-structured interviews, focus group discussions and dialogues between actors. A more detailed description of the research approaches, data analyses and results can be found in the next section.

Findings of the cases

This section presents the findings from the four cases about patient involvement in health-related decisions.

The first case (Chapter 4) discusses the involvement of a broad range of patients in a Dutch policy advisory process concerning a research agenda for the future development of medical products for 15 disease domains. The approach to knowledge co-production applied in this case was based on the Dialogue Model, an approach which operationalizes knowledge co-production in the field of research agenda setting. The approach comprised four phases: (1) exploration, (2) consultation and prioritisation, (3) integration, and (4) follow-up. 119 (expert) patients and 92 non-patient representatives were consulted using interviews and focus group discussions. This study showed two dilemmas. First, finding a balance between a predefined focus (medical products) and being sufficiently broad to enable patients and patient representatives to contribute, and second, finding a balance between relevance for many patients groups and saturation of data for a lower number of patient groups. By taking the context of patients’ daily life as a starting point, patient groups provided new insights. The predefined focus was sometimes perceived as constraining by patients. The commissioner of the advisory process, the Health Council of the Netherlands, considered the articulated needs constructive and incorporated patients’ input in their advice to the Minister of Health.

The second case (Chapter 5) focuses on the involvement of patients in the development of a clinical guideline for resumption of employment after gynaecological surgery. The effectiveness of patient involvement in this knowledge co-production process was assessed by means of Reflexive Monitoring in Action. To this end, a monitoring and evaluation framework was used, comprising pre-defined criteria for the process and the generated outcomes. Patients were involved in the guideline development process at three different stages: (1) 21 patients participated in three focus group discussions which were organized to identify the problems and needs of patients regarding peri-operative care and resumption of employment; (2) three 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) 15 patients tested and evaluated the web-based patient version of the clinical guideline. The guideline development process was divided in two parallel trajectories in which patients and professionals were consulted separately. Patients were primarily consulted for the development of the patient version, although their input also influenced the recommendations for resumption of (work) activities after surgery. Professionals were mainly involved in the development of the recommendations of the clinical guideline. The involvement of gynaecological patients in the guideline development for resumption of (work) activities after surgery was successful in many respects. Consultation of individual patients by means of focus group discussions and with regular feedback moments has been rather effective for a guideline development process related to an incidental, non-threatening disease for which there is no patient organization. Patients’ input contributed to applicability of the clinical guideline in daily practice, which positively contributed to the embedding of the developed knowledge. Increased patient involvement could be achieved by integration of the two parallel trajectories with additional participatory activities, such as a dialogue meeting.
The third case, presented in Chapters 6 and 7, focuses on patient involvement in the field of breast cancer 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). A knowledge co-production process was developed based on two models: the Dialogue Model which provides tools for consultation and dialogue between actors and the FIRST-model which provides guidelines for patient involvement in a research project over a longer period of time. The approach comprised four phases: (1) exploration, (2) consultation of 28 patients and 18 professionals, (3) advisory by a patient advisory board of 7 expert patients and (4) a multi-actor dialogue (17 participants). Chapter 6 presents personal experiences of patients with partaking in breast cancer clinical trials, and points of improvements were identified. The findings revealed that overall patients were positive about clinical trials. Experiences were mostly related to recruitment and information supply, focusing on motivation to take part, decision-making, and random assignment. Support and communication skills of clinicians were essential. Written information was considered too scientific, general and juridical, and random assignment caused insecurity in some patients. Although the decision to take part was dependent on many factors, it appeared to be largely an intuitive decision. And although experiences were generally positive, there is room for improvement. Proposed improvements include a more individual approach, providing feedback of results, and reminding patients being part of a clinical trial, since patients often are not aware of their participation. Chapter 7 discusses possibilities for patient involvement in breast cancer clinical trials from the perspectives of patients and health care professionals. Three possible forms for patient involvement were formulated: (1) an advisory committee of patients, (2) patient research partners in clinical trial teams, and (3) patient representatives for individual and specific tasks. Involved patients could perform tasks in different stages of a clinical trial, e.g. think about and reflect on research designs, improve patient informed consent forms, write and reflect on feedback of the clinical trial findings which could be sent to patients. The formulated possibilities for patient involvement are sustained during a clinical trial and across clinical trials, and could be used as a ‘template’ for patient involvement in clinical trials in general. The added value of the applied multi-layered approach for knowledge co-production was that the different forms of involvement complemented each other and provided different opportunities for patients and professionals to be involved. In the consultation phase, experiences and ideas of a broad group of patients and professionals were collected. The advisory board structured, deepened and made more concrete the general overview identified in the consultation phase. During the dialogue meeting, different actors reflected on possibilities of implementation, covering different perspectives. The dialogue meeting resulted into a shared vision for patient involvement and agreements on how to proceed.

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. The knowledge co-production processes in the nine cases was assessed by the responsive evaluation methodology. All nine cases involved patients and used the Dialogue Model. 54 semi-structured interviews were held with different actors (patients, researchers, funding agencies). Three focus group discussions with patients, funding agencies and researchers (16 participants) were organized to validate the findings. Chapter 8 provides insights into whether patients’ topics are translated into a funding program and are taken up by researchers. Three strategies for the translation (embedding) of research agendas into funding programmes were identified: (1) one-on-one translation, (2) agendas were used to adapt general policies, and (3) no translation. A number of factors, facilitating or impeding this translation, were identified, relating to the context or the process of programming and implementation. Context appeared to be crucial: positive attitudes towards patient involvement, good relations between actors and supportive characteristics of organizations. Patient involvement was rarely sustained during programming and implementation. Chapter 9 describes how the Dialogue Model can be optimized in order to increase patient involvement in follow-up activities of research agenda setting, and thereby contribute to improvement of knowledge embedding. Optimization of the
model is possible by attending to the nature of the agenda and its intended use in earlier phases. Attention should also be given to the ambassadors and intended users of agenda topics. Support is needed during programming and implementation to organize patient involvement and adapt review procedures. In all phases the attitude to patient involvement, actor participation and formal and informal relationships between parties need to be addressed to build a strong relationship with a shared goal.

Conclusions and discussion

The process of knowledge co-production in health-related decision making is complex and not automatic. The four cases provide insights into how knowledge co-production processes could be more optimally structured by studying the three elements of the process in detail. However, it should be emphasized that knowledge co-production is not a linear process but should be considered as an iterative process with interaction between a wide variety of actors and ongoing learning processes.

In three cases, specific attention was paid to the articulation of experiential knowledge of patients. It revealed that knowledge articulation can be conducted in many ways. The use of interviews and particularly focus group discussions appeared very effective methods for articulation of experiential knowledge. To convert the ‘I-voice’ into a ‘we-voice’, quantitative methods have generally been proposed, but the research in this thesis reveals that qualitative methods involving patient representatives could also be used for the successful conversion of the ‘I-voice’ into a ‘we-voice’. Lay-patients and patient representatives complemented each other, and patient representatives were able to deepen and broaden the experiences of lay-patients, leading to increased diversity and representation of the patient community. However, it is also revealed that in a knowledge co-production process, which is constrained by time and money, it is very complicated to include all different sub-groups within an actor group. This creates a dilemma regarding the realization of legitimate knowledge articulation. Explicit attention should be paid to maximizing diversity and representation of the actor groups within the constraints of time and money. Exclusion of specific groups should be transparently communicated, including the reasons for exclusion.

The first three cases illustrate different approaches to knowledge integration. Integration can be brought about by the actions of an independent facilitator or by means of interaction between actors. Interaction between actors can be operationalized by a working group comprising representatives of the actor groups or by a dialogue meeting between actors. The three cases also showed that the end-product of knowledge integration can be constructed in different ways. The cases reveal that although many ways of integration could be successful, it is desirable that actors agree on the content of the knowledge and underlying values and assumptions. For this reason, interaction between actors is preferable because it stimulates reflexive learning. It makes different perspectives and underlying assumptions visible. It can broaden and enrich debates, may avoid the exclusion of actors and bring forward creative ideas. Moreover, to ensure acknowledgement and appreciation of experiential knowledge, strong facilitation is necessary to create conducive social conditions and build trust between actors.

All cases reveal that the process of knowledge embedding is complicated, because of the required systemic changes. Several factors have been identified which particularly influence knowledge embedding: attitude towards patient involvement, relations between actors, characteristics of organizations (e.g. available resources, presence of expertise, structures and procedures), and availability of time for embedding of the research agendas. The cases show that knowledge embedding should become a more integral part of projects for patient involvement and that already at the start of a knowledge co-production process there should be more anticipation on factors influencing
knowledge embedding. Also, more attention should be paid to the development of know-how among actors by continuing to experiment with patient involvement in health-related decisions, including ‘quick wins. This further stimulates reflexive learning processes, which contributes to competence building among actors and the realization of an ongoing means of knowledge co-production. Moreover, the presence of change agents can promote change in health systems and stimulate embedding of knowledge. Early identification and support of change agents could enhance knowledge embedding. These insights could contribute to improvement of, not only the Dialogue Model, but also other participatory approaches to knowledge co-production.