Chapter 9 Towards structural patient participation in health research

Abstract

The role of patients within health research is slowly changing from passive subject towards active partner. In this chapter we focus on a Dutch pilot project, ‘Network patient research partner’, which aims to realize the structural involvement of patients in research projects in the area of rheumatology by means of partnerships between patients and researchers. Twenty-seven patients were trained, and 18 partnerships were established. The network was evaluated over a period of two years to gain insight into facilitators and barriers for a change towards structural involvement. A monitoring and evaluation method was used which was mainly qualitative. The pilot was analysed using a system innovation perspective focusing on ‘culture’, ‘practice’ and ‘structure’ of the health research system. In a ‘protected space’ the partnerships experimented with a new culture, structure and practice to learn what are effective ways to collaborate in the conduct of health research. The presence of factors that enhanced collaboration (facilitators) strengthened the protected space. These included the presence of resources and change agents (proactive people willing to experiment and learn). At the same time, the incumbent culture, structure and practice put pressure on the protected space, which manifested as barriers to developing a new culture, structure and practices. Barriers were the low priority given to the partnership by researchers, lack of initiative and know-how by both partners and researchers, and little interaction within the partnerships. Furthermore, the network structure operated in a rather isolated manner and offered limited support. Due to a combination of these barriers, few partnerships developed new routines to involve partners effectively in the conduct of health research. To enhance structural involvement, strategies need to be developed that focus on strengthening competences (awareness, attitude, skills and knowledge) and building organizational elements in the structure of the health research system.

Introduction

Although patients traditionally have had little influence on health research, their position is slowly but steadily changing from passive subjects and end-users of knowledge to active collaborators (Trivedi and Wykes 2002; Caron-Flinterman, Broerse et al. 2005; Abma, Nierse et al. 2009; Serrano-Aguilar, Trujillo-Martín et al. 2009). It is increasingly acknowledged that patient participation could positively contribute to health research. A growing body of literature describes arguments for patient participation: it could increase the quality and relevance of health research
because patients can complement the scientific knowledge of professionals with their experiential knowledge and can articulate their needs (Entwistle, Renfrew et al. 1998; Tallon, Chard et al. 2000; Boon, Moors et al. 2011; Lloyd and White 2011). The legitimacy of health research can be increased because it becomes more democratic when patients have a voice in decision-making processes and become involved in determining the outcomes that are relevant to them. Another argument mentioned in literature is that participation can lead to empowerment of the patients (Boote, Telford et al. 2002; Williamson 2008).

During the past decade, patients and patient organisations have become involved in a variety of activities related to health research, e.g. setting health research priorities (Nierse, Abma et al.; Oliver, Clarke-Jones et al. 2004; Abma and Broerse 2010; Petit-Zeman, Firkins et al. 2010; Elberse, Caron-Flinterman et al. 2011), being members in (scientific) advisory bodies (Barber, Beresford et al. 2011), reviewing research proposals (O'Donnell and Entwistle 2004) and providing advice on clinical study designs (Hanley, Truesdale et al. 2001). This development is increasingly supported by international health authorities (FDA, EMA), national governments and funding agencies, particularly in Canada, the UK, Scandinavia and the Netherlands. Although consultation is increasingly taking place, the structural collaboration between patients and health researchers seems to be rather limited. Structural patient participation can be defined as patients having a place in decision-making structures and an influence on the decisions taken, whereby both patients and researchers have a long-term commitment to collaboration throughout the research process. In dialogue with researchers, patients provide experiential knowledge which is integrated in research.

Most described initiatives of patient participation are ad hoc or one-off events (Jordan, Dowswell et al. 1998; Stevens, Wilde et al. 2003; Caron-Flinterman 2005; Caron-Flinterman, Broerse et al. 2007) and limited to activities in the periphery of the research process e.g. priority setting. Patient participation is rarely incorporated in the conduct of research. As a consequence, patients’ contributions are often limited (Graham, Broom et al. 2001; Stevens, Wilde et al. 2003; Caron-Flinterman, Broerse et al. 2007), knowledge and skills developed by participating patients are not optimally used, and established relationships between different stakeholders are not sustained. According to Caron-Flinterman et al., the current health research system provides little room for structural patient participation since the current routines, structures, beliefs and values are hardly suitable to integrate the patient perspective structurally, and they are difficult to change due to the rigidity of the system (Caron-Flinterman, Broerse et al. 2007; Broerse, Elberse et al. 2010). The development towards structural participation of patients in the health research system is therefore likely to evolve slowly in small steps. However, little is currently known on how to realize structural involvement, particularly with respect to the actual conduct of research. Therefore, it is adamant to draw lessons from case studies to increase our knowledge on this highly relevant topic.
Towards structural patient participation in health research

The aim of this chapter is to provide insight into facilitators and barriers to structural patient participation in health research by means of partnerships between patients and researchers from a system innovation perspective. To this end we analyse an innovative pilot whereby the Dutch league of arthritis patient associations (Reumapatiëntenbond, RPB) takes up this challenge by establishing a network of trained and supported patients. Trained patients form a partnership with researchers with the aim to become structurally involved in medical and clinical research projects in the area of rheumatology.

A system innovation perspective

To gain insight into facilitators and barriers, a ‘system innovation’ perspective is applied. System innovations are profound changes in dominant cultures, structures and practices which tend to encounter resistance from dominant stakeholders (Geels and Kemp 2000; Grin 2010; Grin, Rotmans et al. 2010). As change is difficult, system innovations are often slow processes, taking 20-30 years. The concepts ‘culture-structure-practice’ are described as relevant notions for system innovations (Rotmans and Loorbach 2006; Rotmans and Loorbach 2010; Van Raak 2010; De Haan and Rotmans 2011). Van Raak (2010) defines culture as ‘a set of values, perceptions and interpretative frames that are shared by most of the involved actors’; structure as ‘the physical, economic, legal, financial, organisational and power structures that facilitate and/or constrain the behaviour of the actors involved’ and practice as the actual actions undertaken by actors which are relevant for the functioning of the system (Van Raak 2010). The culture and structures are shaped by the practices of the stakeholder groups involved, and at the same time, those practices are encouraged or limited by the structures and culture (Loorbach 2007; Van Raak 2010). These three elements are strongly intertwined and reinforce one another. The current culture, structure and practice of the health research system will be indicated with ‘C/S/Pc’.

C/S/Pc developed over time, resulting in a dynamic equilibrium, with stakeholders such as health researchers, funding agencies, research institutes and the government developing routines, rigid relationships and established ways of communication and interaction (Frenk 1992; Caron-Flinterman 2005; Caron-Flinterman, Broerse et al. 2007; Broerse, Elberse et al. 2010). Important characteristics of the culture are: Objective knowledge based on robust and validated research methodologies is key; autonomy and curiosity are important; reward is obtained by publication rates and scientific excellence. In terms of structure, leading researchers are often members of program committees and scientific advisory boards of research funding organisations, and researchers/research department have little interaction with patient organisations. Characteristics regarding practice are the use of validated scientific methods to obtain ‘objective knowledge’, and work being based on protocols and scientific training. Peer review and peer interaction are important ways to judge and discuss findings (Caron-Flinterman 2005; Caron-Flinterman, Broerse
et al. 2007; Broerse, Elberse et al. 2010; Elberse, De Boer et al. Forthcoming). The role of patients has been restricted for a long time to ‘subject of study’ and beneficiaries of developed knowledge.

To induce a change in C/S/Pc, it is important to experiment with new cultures, structures, and practices (C/S/Pn) in a protected space, on a small scale (See Figure 9.1). It is unknown beforehand what this C/S/Pn will look like. By trial and error, involved stakeholders jointly experiment to investigate new practices, adapt and improve them. New practices as well as new values, beliefs and structures may be developed in interaction with each other. Experienced-based learning is essential. Within the protected space, several facilitators are considered key. Facilitators are factors in place or created in the protected space which stimulate the development of C/S/Pn. Important facilitators are: available resources, open learning attitude and commitment, places to meet each other to exchange experiences and ideas, making expectations explicit, having a shared vision, and the presence of change agents (Raven 2005; Raven, Van der Bosch et al. 2010). Change agents are people or organisations who stimulate experimenting with and learning from the novelty at stake. They undertake actions and are willing and able to invest in terms of time, money and/or knowledge.

However, such a protected space experiences pressure from C/S/Pc since the people involved normally function within C/S/Pc. This pressure raises barriers to experimenting with C/S/Pn, pushing things back to ‘business-as-usual’. If too many barriers are experienced by the people involved, the protected space will collapse, and no C/S/Pn develops.

Figure 9.1. Schematic overview of C/S/Pc and C/S/Pn whereby barriers put pressure on the protected space, and facilitators stimulate experimenting with C/S/Pn.
On the other hand, if facilitators are sufficiently effective to overcome barriers, the protected space will become stronger and may expand. To expand, it should offer an attractive alternative (potential) to C/S/Pc. As more people join and learn from C/S/Pn, this increasingly consolidates it. The innovative ‘best practice’ may become normalized, challenging C/S/Pc and adapt C/S/Pc (part of). This is often enhanced by coinciding societal trends (Berkhout, Smith et al. 2004) like in this situation increased patient empowerment (Traulsen and Noerreslet 2004; Barbot 2006; Epstein 2008; Williamson 2008; Williamson 2010), democratization of science, public accountability of science (Frodeman 2000; Fuller 2000; Nowotny, Scott et al. 2001; Chopyak and Levesque 2002; Collins and Evans 2002; Scott 2007) and decline in the authority of experts (Kerr, Cunningham-Burley et al. 2007). They support and induce change towards structural patient participation in health research.

The case described below can be considered a protected space in which patients and researchers experiment with the novel role of patients in the conduct of health research. Since what this new role entails has not yet crystallised, it is a matter of trying, experimenting, learning and adapting in interaction with each other. In the case, stakeholders experiment with C/S/Pn in a real-life setting, thereby offering an unique opportunity to identify facilitators, barriers and strategies that foster structural patient participation (Broerse, Essink et al. 2010).

**Presentation of the transition experiment**

The RPB is a patient association in the Netherlands advocating for the rights and interests of people suffering from a rheumatic condition. They kept on receiving requests from researchers and professional carers to provide input from a patient perspective for research proposals, information materials and questionnaires. Although the RPB considered it of great importance to provide this input and has become a valuable stakeholder in health research, the ad hoc and last minute nature of many of these requests was not considered effective and entailed a danger of tokenism. Therefore, the RPB decided in 2008 to set up a pilot network comprising trained patients that would become structurally involved in research in the area of rheumatology. The FIRST model was used as framework to set up the network (Hewlett, De Wit et al. 2006; De Wit, Elberse et al. submitted). It stands for ‘Facilitation, Identification, Respect, Support and Training’ which are the components considered relevant for enhancing collaboration between patients and professionals. The network was managed by a RPB coordinator for 12 hours a week.

The principal investigators of five rheumatology centers were contacted since they have a good overview of ongoing and future research projects and executing researchers. The principal investigators, together with the network coordinator, selected 18 research projects that seemed suitable for patient participation. Projects varied from care research (e.g. developing e-health
programs), social health research (e.g. relations between rheumatic conditions, work and sports), epidemiological research (e.g. often occurring co-morbidities), medical research (e.g. improvement of early diagnosis) and clinical research (e.g. drug compliance interventions or the influence of co-morbidity on physiotherapy).

Twenty-seven patients were selected based on a motivation letter, a checklist and a ‘job’ interview. They received a 2-day training in ‘What is research’, ‘What is patient participation?’, ‘What is experiential knowledge and how can it be used in research?’ etc. After training, the patients became so-called ‘patient research partners’ and will be referred to as partners in this chapter.

Partners were matched with a research project, often based on their rheumatic condition and the travel distance to the centre. Partners and researchers formed so-called partnerships, comprising preferably two partners and a small number of researchers who collaborate on a research project. In total 17 researchers were involved. Partners received a voluntary contract with the RPB and were reimbursed for expenses incurred.

**Methodology**

**Data collection**

Two researchers (JEE, MPTW) were appointed as ‘monitors’ to conduct a reflexive monitoring and evaluation study over a two-year period, starting in February 2009. Two external advisors (TAA, JEWB) were appointed to provide the monitors with solicited and unsolicited advice and to safeguard their objectivity towards the network. The pilot network was examined in a real-life setting with the aim to gain insight into the barriers and facilitators for structural patient involvement. As described by Barber et al. (Barber, Beresford et al. 2011), many evaluation studies in the area of patient participation are retrospective, potentially losing valuable insights and data. The advantage of a formative evaluation is that direct insight can be obtained of the progress made. Moreover, timely adjustments can be made, and constraining factors may be resolved in an early phase (Regeer and Bunders 2007; Regeer, Hoes et al. 2009). Mixed methods were used (Greene, Benjamin et al. 2001; Stewart, Makwarimba et al. 2008; Feilzer 2010) (See box 9.1), following an emerging research design in which data from earlier stages formed the input for later stages. The data is predominantly qualitative.

The Ethical Committee of VU University Medical Centre was asked if ethical approval was needed for the monitoring and evaluation of the pilot. The study was judged not to be medically oriented since it did not involve treatment, clinical data gathering, interventions or medical data from the
subjects. Furthermore, the monitoring of the pilot was not expected to be a burden for the partners involved.

**Box 9.1 Overview of data collection by the monitors**

- Literature study
- Document analysis including email correspondence
- Participant observations during meetings
  - Introduction meetings whereby partnership meets for a first time
  - Follow-up meetings of partnerships
- Regular updates of functioning of the partners via telephone or email
- Local meetings with partners to exchange experiences
- Local meetings with professionals to discuss patient participation, possibilities for involvement in research projects and progress
- Reflective discussion initiated by two monitors, attended by one partner from each centre, three researchers, the coordinator, a representative of RPB and two external experts
- Electronic survey for partners (response rate 24 out of 27) consisting of 17 topics related to expectations, experiences and benefits of the partnerships
- Electronic survey for professionals (response rate 15 out of 16) consisting of 17 topics related to expectations, experiences and benefits of the partnerships
- Four focus group discussions with partners at the four different locations (27 invited, 20 attended) to discuss the outcomes of the surveys, developments and potential strategies to enhance collaboration.
- Six interviews with professionals, purposive sample to discuss the outcomes of the surveys, developments and potential strategies to enhance collaboration.

All participants (partners and researchers) were informed by email and newsletter in advance about the study, the aim of the monitoring, and what was expected from them as participants. During meetings when the monitors visited the participating department or partnerships for the first time, they introduced themselves and explained their role in the network. All participants consented to participate in the monitoring and evaluation study. After receiving approval from the participants, interviews and focus group discussions held by the monitors were recorded and transcribed. A summary was sent back for member check. Informal conversations, telephone calls and partnership meetings were often not recorded, but detailed notes were made by the monitor(s). Observations of meetings were done to investigate how partners and professionals interacted, collaborated and how the partnerships dealt with the partners’ input. If requested by partnerships, monitors provided input for discussion and suggestions for collaboration. Both monitors documented their activities and findings in a log book, which was regularly discussed
between the monitors, the network coordinator and two external advisors. The monitors had a facilitating role in stimulating dialogue between partners and researchers. To support the partnerships, strategies based on findings from earlier stages and in response to requests from partners and/or professionals were developed and applied if agreed upon by the network coordinator. For the pilot, successful partnerships are defined as ones which were sustained during the course of the project, with the partners involved in different phases of the research project and both partners and researchers experiencing the benefits of collaboration. In addition, in successful partnerships, a start was made with the development of C/S/Pn.

Analysis
The analysis explored the barriers and facilitators to structural collaboration in the partnerships. A first round of thematic content analysis was conducted to identify facilitators and barriers. In a second round of coding, themes related to barriers were coded in relation to ‘culture’, ‘structure’ and ‘practice’. A round of open coding was applied to themes related to facilitators, followed by a comparison with facilitators described in the system innovation literature. The findings were discussed in the larger research team, and consensus was reached on the analysis.

Results
Since forming partnerships was highly innovative and new for all participants, preconditions for successful partnerships were absent. There was a vast diversity in how the partnerships developed. Three of them never materialised or became silent shortly after the kick-off meeting. Three partnerships developed into well-functioning collaborations leading to expanded or prolonged involvement of the partners in new projects. The other twelve partnerships varied enormously in degree of success. Below, the various facilitators and barriers that were experienced in the partnerships are described. All partnerships encountered similar barriers, but in the more successful partnerships many facilitators were present that made it possible to overcome the barriers.

Facilitators
The presence of ‘financial resources’ to set up the network, appoint a part-time coordinator, train partners and arrange reimbursement for the partners’ expenses facilitated the partnerships. For participating departments, there were in principle no costs involved.

When the partnership was perceived as a ‘learning process’, accepting that know-how was something that was not present beforehand but could be jointly explored and discussed, interaction within the partnership took place more regularly. Members were willing to experiment to gain new knowledge, to learn which tasks were suitable for partners, and to understand how
collaboration could be optimized. This included the willingness to invest time and attention; time to get to know each other, meet each other and discuss the research process. Being open-minded towards the partners’ input; making an effort to understand and discuss it; and looking for ways to integrate it in research also facilitated collaboration.

The presence of ‘change agents’ was an important facilitator, which was only seen in a few partnerships. Change agents were proactive, arranged meetings, actively requested information and updates, or suggested possible tasks or improvement for collaboration. In one centre, a partner took up the role of ‘location coordinator’. She checked with researchers about which projects were being executed and which would start in the near future and discussed opportunities for new partnerships. She arranged local meetings for partners to brainstorm and exchange experiences and discussed possibilities for involvement with the principal investigator. This was very effective in fostering partnerships. In another centre, one of the principal investigators made fixed appointments with the partners, inviting them for ‘social happenings’ at the research department and stimulated the partners to make a poster to present at a congress.

The network coordinator regularly sent around a newsletter to inform partnerships about developments, examples of tasks, new partners or new research projects. However, no face-to-face meetings were organized or initiated by partners or partnerships. The monitors decided to organize ‘local meetings with partners’ to exchange experiences and create solidarity within and with the network. Partners expressed that they considered these meetings fruitful to exchange knowledge and ideas, and to get to know each other and have the feeling of being part of a network. Since these meetings were organized quite late in the pilot, the effect was not optimal.

In none of the partnerships were the expectations made explicit at the start resulting in divergent or too ambitious expectations and frustration if ‘reality appeared different’. However, in some partnerships, ‘clear agreements’ were made about contact, responsibilities and potential tasks. This led to stabilized moments of interaction and less frustration. Partners knew that they needed to be patient if the research entered a slow phase, or whom to contact if they wanted an update. Potential tasks were explored by brainstorm and discussion sessions and experimenting. To stimulate making clear appointments in all partnerships as well as discussing the expectations, two forms were developed by the monitors. One could be used during the first meeting to agree on appointments, discuss the research project and make expectations explicit. The other form could be used during follow-up meetings to guide discussions on the progress of the research, potential tasks and trying to make the added value of the partners’ input to the research explicit. This last element was added since making the benefits of collaboration more explicit helped members to become or stay motivated about the partnership. By gaining ‘insight into the added value’, some researchers developed a more positive attitude and became more open-minded. Partners were also motivated by receiving feedback on how their input was used or why it could
not be integrated. This helped them to gain more insight into research procedures and the possibilities and constraints of their perspective.

When there was a ‘personal click’ between partners and researchers, the collaboration was more natural, which made it easier to take the initiative, and both parties reported enjoying the collaboration. Recruitment by the participating departments brought substantial benefits over recruitment by RPB since it increased the chance of a good match and created stronger intrinsic motivation and responsibility on the part of the professionals.

The presence of ‘neutral monitors’ who provided tailor-made advice and support, addressing specific needs or ideas, was another facilitator. With some partnerships, brainstorm sessions were organised to identify suitable tasks, while other researchers were advised to set fixed appointments in the agenda to update the partners. Some researchers were provided with materials like publications, handbooks or research reports describing patient participation in health research to increase their understanding of patient participation and inspire them.

‘Creating more visibility and awareness’ of patient participation and the network among researchers was a significant facilitator as well. A short training module for researchers was developed to address the concepts of patient participation, potential tasks for partners and directions for successful collaboration. The intention was to increase awareness about patient participation and knowhow (skills) on how to involve partners. This training made some researchers less insecure about their collaboration and provided them with tools to enhance the partnership. In addition, two articles were published in the Dutch Journal for Rheumatology. Also, the four ‘best practices’ of the network were showcased at the national rheumatology congress.

If we compare the facilitators identified in the case study with those mentioned in the system innovation literature, we observe that four new facilitators were identified; making clear appointments, a personal click, neutral facilitator, and creating visibility and awareness. What was lacking though is a ‘shared vision’, to which these collaborations should lead, which is considered a very important facilitator in the system innovation literature.

**Barriers in practice**

Difficulties were experienced in ‘identifying suitable tasks for partners’ within the research and creating room for dialogue. The current practices of researchers are activities which are performed in accordance with validated methods and established routines of interaction. Scientific training and scientific competences were perceived as essential for this, and many tasks were not considered to be transferable to or shared with the partners. Identifying new, complementary or additional tasks for partners proved challenging. Some researchers indicated that they had difficulty understanding how to combine ‘scientific knowledge’ and the ‘experiential
knowledge of patients. How can the stories told or the input provided by the partners be used in their research project? Since the partnerships had difficulties identifying tasks, not many meetings were arranged. So a general discussion on the project, talking about research activities, findings and new developments rarely took place. This again led to little dialogue within the partnerships, and thus no potential moment for partners to provide their view and explore new tasks together.

Some partnerships stopped or were kept low key because the researchers felt that the project was no longer suitable. These projects were considered, for example, too fundamental, requiring scientific competences like statistical analysis. Or everything was already agreed upon, and no changes could be made in the research proposal and related documents. According to a principal investigator, it was more difficult than expected to predict which projects were suitable, since there was a lack of criteria and the focus of research projects is often adapted during a study.

For the partners, despite having received training, being involved in research was not as easy as expected by some, and sometimes a ‘lack of insight in research procedures’ was experienced. For example, partners became impatient when little progress was made, due to waiting for funding procedures or approval from the medical ethics committee, or because the phase of data collection took a long time. Partners had not been aware that ‘doing research’ took so much time. Another example is the lack of awareness of the value of validated questionnaires and methods used in research. Partners did not always understand that questionnaires cannot be changed easily. Thus, input provided by partners could therefore not always be incorporated, which led to frustration. It should be noted that in one partnership, the partners indicated that the selected questionnaire was old-fashioned, and they did not recognize themselves in the items asked. They stated that if these questionnaires were used, the questions posed in the research proposal would not be addressed properly. Based on this input, the research team decided to use another questionnaire.

In many partnerships, a ‘lack of taking initiative’ and little interaction were observed. Both partners and researchers were waiting for the other to initiate contact, update each other or plan a new meeting or activity. They also expected the other to come up with potential tasks. Often, there was also a difficulty to maintain interactions. According to some researchers, it was not yet in their routine to regularly contact and update the partners about their project. Some partners with a full-time job preferred to meet in the evenings or weekends, which did not suit their working hours for many researchers. This resulted in long periods of no interaction and dialogue during which partners lost the feeling of being part of a partnership, and researchers returned back to ‘business-as-usual’.

Optimal interaction was hindered by the fact that decisions were often made ad hoc in the research department. There was much ‘quick communication’ during working hours between
researchers. Issues were quickly solved by email or face-to-face interactions between researchers (e.g. at the coffee counter). It is hardly possible to postpone the discussion or decision to the next appointment with partners in order to include their perspective.

**Barriers in culture**
Researchers did not immediately see the ‘added value’ of the partners’ knowledge for their research. Patient participation is a relatively new development, and little attention has been paid to it in the literature. Furthermore, there are few peer researchers who collaborate with patients. So without knowing what it could bring to the project, the researchers were requested to invest time and effort to develop a successful partnership. Researchers indicated that they were busy with their research; they needed to recruit patients (as subjects), finish analyses, had important deadlines for a publication or their PhD thesis, which were considered priorities. The involvement of partners provides little (scientific) reward, according to several researchers, and could even constrain research. The involvement of patients in an active role could conflict with the idea that scientific knowledge is objective; experiential knowledge is subjective and therefore often considered less valuable. Researchers expected their partners to represent a broader patient perspective; partners should have insight into the stories, needs and ideas of a broader patient group to create a more ‘objective’ patient perspective. However, this was rarely the case; partners mainly provided input from their own experiential knowledge. Another question arose: will their research still be considered ‘scientific’ by other researchers if input from patients is integrated into it? For example, researchers were concerned that journals would not accept their papers anymore, since several tasks were executed by ‘partners’ and not by ‘professionals’.

In communication with researchers, ‘much jargon’ is used. Although researchers are often unaware of this aspect, partners experienced difficulties in understanding documents and protocols. Another obstacle for many partners was the amount of English used, instead of Dutch, sometimes even as the main language for the project team. This constrained partners in following discussions, understanding documents they needed to read or questionnaires they wanted to comment on.

**Barriers in structure**
Some partners as well as the professionals indicated that they felt ‘insufficiently supported’. Where to turn to with questions, needs or support was sometimes unclear. Normally, young researchers discussed their work and difficulties with their supervisor. However, the knowledge on how to collaborate effectively with partners and which tasks are suitable for partners had often not yet been developed. The network did not yet have a clear position within the structure of research in the area of rheumatology and functioned in a rather isolated way. In addition, professionals as well as partners rarely looked for help proactively. Often, they let the
collaboration slide without notifying the coordinator or monitors. This indicates that in those cases the urgency to develop an optimal partnership was not high enough.

Not establishing and maintaining a partnership had ‘no negative consequences’. When the network coordinator discussed with principal investigators the possibility of including a partner’s expenses in the budget when applying for grants, they were very reticent to do so. Two reasons were often mentioned. First, when there is room in the budget, they would rather put in the cost for a research nurse instead of a partner. The competences, tasks and responsibilities of a research nurse are clearer. Second, researchers doubt if research funding agencies will accept costs for a partner in a budget, since it is not a standard procedure. And it is not a criterion for receiving funding.

An interesting issue that arose from the data was the question, ‘Who is responsible for the supervision of the partners in the partnership?’ Should this be the network coordinator, providing support and stimulating collaboration, or should this be the principal investigator? This issue was very unclear, despite the fact that partners signed a ‘volunteer contract’ with the RPB. It became apparent that no agreements were made between the principal investigators and the coordinator since they did not foresee this issue. Partners expected the network coordinator to provide the support, training and stimulation of the collaboration and the researchers to provide information regarding research, research procedures, etc. However, several researchers indicated that they expected the training, insight into research processes, facilitation and instructions to be the responsibility of the network.

Although the network was financed by the Dutch Arthritis Foundation, it did not communicate to the researchers that the patients could become actively involved, nor did it create incentives for researchers. The network was not involved in interactions between the established stakeholders. Establishing new relationships proved time consuming, and the network coordinator needed to invest much time in initiating and maintaining contact. The ‘available hours for the network coordinator’ proved to be insufficient to establish and maintain relationships with the different stakeholders involved. Subsequently, new, potentially suitable, research projects started in the participating departments without a partnership. Also, several researchers who were already involved in a partnership did not involve the partners in other or new research projects they were working on. The network coordinator noticed that researchers and principal investigators rarely contacted her to actively ask for the involvement of partners in an upcoming project.
Discussion

This case study is about patients trying to get a place in a relatively closed and stable community – C/S/Pc of health research. It shows that there is no understanding of what C/S/Pn should look like. It also shows the importance of starting with people who are highly motivated and committed to the partnerships. They must be able to tolerate a high level of uncertainty and be willing to invest time and resources to investigate what C/S/Pn could look like, thereby creating a strong protected space. Realizing patient participation in the heart of research, its conduct, proved very challenging since many researchers do feel that it is their domain.

In light of system innovation theory, the question is: How can structural patient participation in the health research system be enhanced? A system innovation can be enhanced if a protected space becomes strong and expands, so C/S/Pn can adapt the system (part of it) from the bottom up. An important prerequisite is that either C/S/Pn offers a clear, beneficial alternative to the ‘normal’ way of doing things or people should be sufficiently open-minded and willing to experiment to see if it offers sufficient benefits. To stimulate more people to experiment with the structural involvement of patients, it is important that best practices be developed which can be adopted by others.

Our study reveals that the role of change agents in the partnerships is essential to make the collaboration successful. Two questions arise. Firstly, can people be trained to become a change agent, or is it more their ‘personality’? People could be challenged to think and act like a change agent with training and support, but ultimately they must be motivated and feel capable of doing so. It is important for both researchers and partners to become change agents to induce a change towards more structural involvement. They can set an example for their peers and inspire others to follow.

Secondly, how can change agents (potential) and other interested people be optimally facilitated to operate in a partnership? To induce new behavior or new practices, it is important that stakeholders ‘know’, ‘can’ and ‘will’ (Coolsma 2008), people should become aware of ‘patient participation in health research’, attitudes towards involvement must be positive, and people need skills, knowledge and a structure to put it into practice. Therefore, we suggest that two types of strategies are needed: ones to develop the necessary competences directed at a new culture and practice, and ones to create strong organizational elements in the health research structure to support the embedding of new competences. Each will be discussed below.

Strategies to develop competences are directed at creating awareness of the issue, stimulating a positive attitude, developing skills and providing knowledge. Training in being proactive, knowledge of effective collaboration techniques, insights into what patient participation entails,
insight into how to provide input (partners) and how to integrate experiential knowledge into research projects (researchers) can improve skills and knowledge. However, one of the main priorities in this regard is gaining more and systematic insight into the impact patient participation has on health research. Insight and visibility of the added value of partnerships in health research may increase the priority for patient participation in research, stimulate a more positive attitude, address a lack of knowhow and encourage researchers. It can stimulate the development of a shared vision on what C/S/Pn should look like and what the benefits of experiential knowledge are for health research. Ideally, added value and lessons learned are published in scientific journals. However, measuring the impact of structural patient participation in health research will be challenging, mainly for two reasons. Firstly, there is no consensus on what impact exactly implies: empowerment of patients, change of research procedures, more needs-oriented health research, better health outcomes or changing values and attitudes of the involved stakeholders (Boote, Barber et al. 2006)? Secondly, it is unclear how to measure impact since it is challenging to (re)assess the influence of involvement. For explicit changes made, like change of questionnaires used, improved intervention or priorities set, the impact can be quite clear (Barber, Beresford et al. 2011), but if patients provided input during decision-making processes, where decisions are made jointly, it is less clear how and how much this decision is influenced by the input of patients. Or what the final health improvement is.

To organize structural patient participation in the conduct of health research, we argue it is important to have central and evident organizational elements embedded in the health research system with the aim to organize patient participation and provide incentives. Such elements should establish enduring relationships with important stakeholders. Ideally, a strong relationship is established with relevant funding agencies, so when grant proposals are submitted, researchers can be directed to this organizational element for the possibilities for patient involvement in the research project. Also, funding agencies can create incentives for researchers to establish a partnership, for example by providing funding for the partners’ expenses, or setting patient participation as a criterion for funding. As shown in the study, the available resources are an important facilitator. In the pilot, a network of trained patients was established as an organizational element and coordinated by a network coordinator. A network seems to have different advantages. With it, partnerships can be coordinated, partners and researchers can be brought together (looking for the right match), or research departments can be instructed on how to select an eligible partner. It can offer support by providing training, tools, information or advice for partnerships on suitable tasks or how to optimize collaboration (De Wit, Elberse et al. submitted). Moreover, it can function as a ‘knowledge junction’ where expertise, experiences, information, knowledge, tools, etc. are collected and made available. Additionally, it can serve as a ‘safety net’. When partners fall ill, the network can look for ways to continue the partnership. Furthermore, it can give more meaning to the people involved; they feel part of a group (Passy and Giugni 2000).
Using a reflexive monitoring approach was very constructive in this study. It gave in-depth insights into barriers and facilitators in this real-life setting which would otherwise have been inaccessible. Meanwhile, reflective monitoring provided a flexible and context-sensitive approach to study the pilot. As a result, some facilitators could be consolidated while some experienced barriers could be addressed (partly) due to developed and applied strategies by the monitors.

The conceptual framework used to analyse the case study was predominantly used in a descriptive way in the areas of transitions in energy (Verbong and Loorbach 2012), agriculture (Klerkx and Leeuwis 2009) and mobility (Geels, Kemp et al. 2012). It offers tools to unravel barriers and clarify facilitators. However, it also appears to be very useful in the domain of health research and can be used in a more prescriptive way. We would recommend considering current ‘culture, practices and structures’ in an early phase to develop strategies, forestall potential barriers, create facilitators and support those already in place.

Change takes time, effort and energy. Some important steps have been made in the past decade. Patients are now increasingly consulted, and the number of initiatives which aim for structural involvement is growing. A next phase could involve sustaining that patient participation, with knowledge of and competences for collaborations of patients and researchers in the conduct of health research being embedded in the organisations involved, and transferred to new research projects, new jobs and new networks.

**Limitations of the study**

An important limitation of the case study is that it is very much context-bound. The network was set up for research in the area of rheumatology, and we do not know to what extent the lessons learned can be generalized to other disease domains or other countries. Also, the data provided too little insight into the suitability of projects for partnerships to experiment with C/S/Pn. Moreover, since the monitors were closely involved with the network and the participants due to participatory observations, there is a risk of observation bias, especially since the network coordinator’s organisational tasks were sometimes taken up by the monitors. Researchers’ bias was reduced by appointing two external senior advisors, keeping a detailed logbook, reflect regularly on findings and developments within the network, and having member checks.

**Conclusion**

In the most successful partnerships, a change agent was present, somebody making an effort and being proactive. Making clear agreements about contact, tasks and responsibilities within the partnership, as well as making expectations and its added value explicit stimulated collaboration,
as did a ‘personal click’ between the partners and researchers. Since C/S/Pn was highly unclear, being open-minded and perceiving the partnership as a learning process were essential.

The aim of the network, to structurally engage partners in the conduct of research, has not yet been realised, but the first important steps have been taken. Many barriers still need to be overcome. The partnership often has a low priority for busy researchers since it has no evident scientific reward. In addition, partnerships interfere in daily practice. Involving patients is new, and researchers are inexperienced about how to do this. The parties did not always feel responsible for the partnership and collaboration was suboptimal. Furthermore, the network operated in a rather isolated manner and offered little support. Due to a combination of the above-mentioned barriers, most partnerships did not start to develop C/S/Pn.

Nevertheless, a growing number of partnerships are starting to function well, are becoming successful and are valued by the partners and researchers. Currently, there are only a few change agents involved in the network, which makes it fragile. Partners are patients, and they can become ill or have to stop being a partner due to disease-related issues. If only a few people take up the role of change agents, and they disappear for any reason, skills and motivation are lost. Therefore, it is important to increase the number of change agents involved (through better recruitment and training), exchange experiences and lessons learned, and build a strong supporting network to fall back on if people drop out.
Part 5. Conclusions
Chapter 10 Conclusions and Discussion

In this part of the thesis, I draw conclusions on the main findings of the research in relation to the three objectives set in Chapter 3:

1. To contribute to an increased understanding of how patient participation in health research can be shaped effectively;
2. To contribute to an increased understanding of the embedding of patient participation in the health research system;
3. To acquire insight into the extent to which increased patient participation may contribute to a transition towards a more needs-oriented health research system.

The following main research question has been formulated:

*How to realise patient participation in health research in such a way that it becomes embedded in the research system and contributes to a system innovation towards a more needs-oriented health research system?*

Based on the conclusions of the inventory study and the transition experiments, the main research question will be answered by discussion the main findings per objective. Then, a reflection on the usefulness of transition theory in light of patient participation in health research will be provided, followed by a description on how the validity in this research was guarded. In the last section, I describe directions for future research in the area of patient participation in health research and round up with a concluding remark.

**How to shape patient participation in health research effectively?**

In 2005 Caron-Flinterman concluded in her PhD thesis that patient participation in health research was ad hoc, marginal, but slowly increasing (Caron-Flinterman 2005). This trend seems to have continued, and now, seven years later, progress can clearly be witnessed and the first steps of a system innovation are taking place. Currently, there seems to be a commitment from a growing number of organizations to effectuate a change towards a more needs-oriented health research system. Front runners can be identified, such as the Netherlands Asthma Foundation, ZonMW and the Dutch Arthritis Patients’ League. They stimulate ‘followers’ in these developments, particularly other funding agencies and patient organizations. Patients are increasingly involved in the appraisal procedures of funding agencies, in clinical trials and in the execution of a broad range of research projects. It indicates that the possibilities for patient participation in health research are wide-ranging. However in the Netherlands, many approaches are still in an ‘experimental phase’; patient participation in health research is not uncontested, and barriers are experienced. Barriers
are related to characteristics of the academic research community (decision-making is dominated by experts in research, strong specialization of the research, and the high valuation of scientific knowledge and excellence), the private research community and industry (no sense of urgency and strict rules and regulations related to contact with patients) and the patient community (not always effectively organized, other priorities, lack of resources and reluctance to become involved).

Different initiatives for putting patient participation in health research into practice are described in this thesis. These transition experiments are set up to stimulate and enhance a system innovation. Related to patient participation in research agenda-setting (‘what is researched’), there is a rapidly growing body of knowledge on how to shape it effectively. The first initiatives had many followers, and every time new initiatives started, the designs were adapted to meet the specific circumstances, and new lessons were learned. In the Netherlands patient participation in agenda-setting has evolved into a ‘best practice’, although there are still challenges to be addressed. In future research agenda setting projects, more attention could be paid to ‘broadening’ and ‘scaling up’. With respect to realizing partnerships in the conduct of health research many barriers were experienced in the pilot project studies in this thesis. How to shape this is less clear and more experiments are needed for all stakeholders to go through a learning process. More insight into how to shape patient participation in the context of how research takes place should be done in a cyclic process of action research: design, experiment, reflect, learn, adapt and share (‘deepening’). This way, a proof of principle can be developed. Also, much can be learned from Participatory Action Research (PAR). PAR focuses on the increased understanding of how changing practices or actions can benefit involved stakeholders (Carr and Kemmis 1986; Kemmis and McTaggart 1988; Kemmis and McTaggart 2000; Reason and Bradbury 2001; McIntyre 2008). PAR has been applied in the field of health care (Cornwall and Jewkes 1995; Dedding 2009), system change (Burns 2007) and policy (Epstein, Mayfield Lynch et al. 2012). This field of study may provide indications on how changing the practices of health researchers to actively involve patients can be stimulated.

**Patient participation in research agenda-setting**

To increase our understanding of how to shape patient participation in health research with respect to ‘what is researched’, we applied the Dialogue Model in three different transition as experiments described in Part 3. This model has already been tested and evaluated in other experiments. It provided the opportunity to analyze patient involvement in agenda-setting in detail. The Dialogue Model has no fixed recipe, its context and requirements always require tailoring as also has been done in the described cases. Applying the Dialogue Model to the different cases provided valuable insights, (described in the next paragraphs) of how to shape patient participation in health research agenda-setting more effectively. The experiments described in Part 3 indicate that the flexibility of an approach is of major importance, so it can be
adapted to the specific circumstances, needs and wishes of the stakeholders involved and requirements from commissioners.

Exclusion mechanism and inclusion strategies: From this transition experiment we wanted to gain detailed insights (deepening) in how to stimulate an inclusive dialogue between patients and professionals. To enhance the quality of dialogue settings between patients and professionals, it is important to be aware of exclusion mechanisms and to apply inclusion strategies. ‘Exclusion’ is defined as the process whereby members of a certain stakeholder group – in this case patients and patient representatives – or their perspectives are not incorporated in the decision-making process, because of actions taken by members of other stakeholder groups or the process facilitator. Applying inclusion strategies to overcome exclusion mechanisms can help to stimulate genuine dialogue between the different stakeholders involved. In Chapter 5, three types of inclusion strategies are described in relation to circumstances, behavior and verbal interaction. In relation to ‘circumstances’, the precautions lie in the sphere of preparation and organisation, creating basic requirements for a genuine dialogue. To stimulate inclusion in relation to ‘behaviour’ and ‘verbal interaction’, strong facilitation is essential for recognizing and correcting exclusion. However, it is important to recognise that exclusion is not merely a matter of one party actively dominating in its position of power. People are often unaware that they exclude others through their use of verbal and non-verbal communication. This is a subtle process; patients and informal carers perceived their participation in the dialogue meeting as inclusive, while some of their specific input was sidelined as became evident in the resulting research agenda. It is suggested that exclusion in this case study was based on embedded assumptions concerning the lower value of experiential knowledge compared to scientific knowledge, and the idea that experts are best suited to define the research agenda. This view was not only observed among professionals, but also among patients and informal carers, who seemed to have a high degree of trust in the experts.

Condensing the Dialogue Model: Designing, experimenting with, and adapting new approaches are time- and money-consuming matters, and first experiments are often quite elaborate, and thus expensive. However, when an approach develops further into a validated method, it is important to test if the approach can become more cost-effective, which would be more suitable for embedding in current structures and practices. The Dialogue Model was sufficiently tested and honed to investigate whether a condensed, more cost-effective method could be developed for updating and extending a shared research agenda. The Netherlands Asthma Foundation considered this a prerequisite for the structural involvement of patients in research agenda-setting. A manageable method was needed which applied the principles of the Dialogue Model but could be conducted in a short period of time with limited resources. In this transition experiment, the Netherlands Asthma Foundation wanted to establish a ‘proof of principle’ on the effectiveness of a condensed method (deepening) and at the same time the main focus was on
‘scaling up’, in order to embed patient participation in the practice and values of the Netherlands Asthma Foundation. The condensed method proved to be successful in the articulation of patients’ and researchers’ needs for health research, resulting in a state-of-the art research agenda reflecting both the needs of patients and researchers. There was much overlap between the priorities of professionals and patients on broader themes, although they differed in details and brought different and challenging perspectives and issues to the table. Since no dialogue meeting took place in the integration phase in this case, a professional patient who works as a policy employee at the Netherlands Asthma Foundation was involved in the research team to ensure that the input of patients was visible in the integrated research agenda (to avoid tokenism). Ideally, even in a condensed approach, a dialogue should have taken place between professionals and patients to discuss differences and to increase mutual understanding. If topics are only considered important by patients, not by researchers, it entails the risk that researchers will not submit research proposals on these topics.

**Having a broad range of disease domains and a pre-framed narrow focus:** In most cases, a research agenda is focused on one disease domain. In the case of the research agenda on medical products, the focus lied on one specific area of research – the development of medical products – but included 15 different disease domains. This provided a perfect opportunity to develop a transition experiment focused on broadening. In this case the commissioner, the Health Council of the Netherlands, set strict criteria for the patients’ input at the request of the Minister of Health. Even though the focus of a research agenda was pre-framed and highly specific, patients were still capable of articulating their needs in a facilitated process. It is important that enough room was provided for patients to share their perspectives and ideas to foster the required trust. The process was designed in such a way that, while keeping the focus and satisfying the criteria set by the commissioner, additional attention was paid to broader issues highly valued by patients. During the focus groups, the context of the patients’ daily life was taken as the starting point for needs articulation. In several steps, specified input corresponding to the criteria set by the Health Council of the Netherlands could be generated. By using the parking spot, we were able to collect and acknowledge at any moment the presence of wider issues such as communication between patient and physician, societal participation, health care organization and health financing. This resulted in participants feeling heard and being motivated to discuss medical products. In this case, the role of the facilitators was crucial to optimally involve patients. Much attention had been paid to realizing social conditions, which were different for all disease domains. It was important to the patients that the commissioner acknowledged their broader input by recognizing the value of these issues as providing context to the advice on medical products. This design revealed that patient participation can be applied a agenda-setting process with a narrow scope, requesting quite specific input, provided that sufficient attention is given to broader issues. A balance can be found between a predefined focus, set criteria, and keeping a mandate for decision-making by a commissioner, while patients are able to provide valuable input from their own perspective.
Based on the lessons learned from previous experiments whereby patients were involved in agenda setting, the Dialogue Model could be successfully applied in a different context. By broadening, system innovation can be further enhanced.

**Patient research partners in the conduct of health research**

In Part 4, a different approach to patient participation in health research was studied. In contrast to patient participation in agenda-setting, where a validated method was already available, less knowledge was available in the Netherlands on how to shape partnerships between patients and researchers to realize patient participation in the conduct of research. The project ‘Network Patient Research Partner’ provided an excellent setting for a case study to learn how to shape participation effectively. Since little was known, the focus was mainly on deepening, to learn as much as possible in a specific context. A reflexive monitoring approach was used which was considered constructive in this study. It gave in-depth insights into barriers and facilitators in this real-life setting that were otherwise inaccessible. At the same time, reflexive monitoring provided a flexible and context-sensitive approach to study the pilot. As a result, some facilitators could be consolidated, while some barriers could be addressed (partly) with strategies developed and applied by the monitors. Also, a neutral facilitator (in this case, a monitor) helped to build bridges between patients and researchers and provide tailor-made advice, support and knowledge.

To realize patient participation as partnerships between patients and researchers in the conduct of research, establishing a network to organize, coordinate and support partnerships provides an interesting opportunity. A network can, besides providing support and coordination, create the feeling of being part of a group. It is important that such a network creates possibilities to meet each other, exchange experiences and develop new ideas and visions. Also, the network coordinator can be a neutral party to facilitate the collaboration between researchers and patients. The FIRST model offered guidance in setting up such a network, but adaptations and elaborations to the model were necessary. The model appeared to be rather static, while the pilot proved that setting up successful partnerships is a dynamic process, and regular reflection on the process is needed. The key is to stimulate learning among the different stakeholders about how they would like to shape participation, to make the added value of involvement explicit and to provide feedback to each other on how they experience collaboration and how experiential knowledge can be integrated in research projects. Furthermore, in the current FIRST model, little attention was paid to supporting researchers in this new endeavor. The FIRST model does not include a system innovation perspective, while the barriers encountered are mostly related to the current culture, structure and practice of the health research system. Especially in the less successful collaborations between patients and researchers, the FIRST model provides insufficient pointers and guidance on how to realize genuine involvement.
The partnerships can be seen as protected spaces in which people (researchers and partners) experiment with new practices, structures and cultures of conducting health research. The presence of facilitators like availability of resources, training and support for patients and researchers, and the presence of change agents committed to the cause were key. Sharing lessons and making the added value explicit helped researchers and patients to stay motivated and increased their understanding of effective collaboration. Partnerships where facilitators were amply present could shield themselves sufficiently against pressure from the incumbent practice, structures and culture and develop new practices, structures and culture through a joint learning process. Barriers were experienced in all partnerships. Due to the lack of initiative and knowhow by both partners and researchers and little interaction within the partnerships, collaboration was often suboptimal. Furthermore, the network operated in a rather isolated way and offered little support and few incentives to the partnerships. Due to a combination of the above-mentioned barriers, most partnerships did not develop new routines and values to involve partners more structurally in research. If few facilitators were in place, the protected space for collaboration did not develop or collapsed, resulting in business-as-usual.

Strategies to enhance partnerships should focus on training in necessary competences, which involve increased awareness what patient participation entails and tasks are suitable for patient research partners, a more positive attitude, skills and knowledge, and building up a strong structure for the network. Creating clarity on expectations and agreements about contact, tasks and responsibilities help to realize a successful partnership. However, in this early phase of experimenting with structural patient participation, it is important that partnerships receive tailor-made advice and support. Nevertheless, such profound change cannot be realized immediately; it emerges by developing trust and self-confidence over a longer period of time. Also, a shared future vision is needed because it provides direction, alignment and can inspire followers.

**Obtaining insight into how to embed patient participation in the health research system**

**Strategies to enhance embedding**

Embedding patient participation in health research means that patient participation becomes included in the research structure, culture and practices, anchored in the system, the ‘normal thing to do’. It is important that it takes place in different phases of the research process and involves sustained collaboration between patients and researchers. The presented research indicates that this is not yet the case in the Netherlands. Even though patient participation in agenda-setting is becoming more established, many initiatives regarding agenda-setting entail one-off events often not being followed up in other stages of the research process. Little momentum for change is created this way since collaboration between patients and researchers
fades after the project is finished; lessons learned can be forgotten, and everybody goes back to ‘business-as-usual’. There is no on-going dialogue between the different stakeholders, implying that the role of patients is again reduced. So there is still a way to go. However, during this research, various strategies have been identified that could enhance embedding.

Embedding of patient participation in the health research system clearly requires a learning process in which all parties need time to adapt to this new patient role. Through experimentation, reflection and learning, more insight will be gained into suitable approaches, projects and tasks. A regular and genuine dialogue between stakeholders will help patients to better understand the principles of research and will help researchers to better understand the patients’ story as well as its value for research. It can stimulate mutual learning and foster trust and respect. Researchers are initially often reluctant, but inviting them to focus groups and dialogue meetings organized during a project can help them to experience what patient participation entails and what patients can bring to the table. Often this results in a more positive view towards patient participation. Furthermore, it is important that support is offered to all parties involved to strengthen the competences to collaborate effectively.

The importance of change agents committed to effectuate change is stressed for both forms of participation. Change agents are people or organizations who are motivated to induce change and willing to invest resources. These change agents may be facilitated by providing appropriate methods, tools and competences. In addition, followers are also important; followers can repeat experiments, copy new practices, or develop new values based on what they see by the frontrunners. If more people follow in the development of change, embedding of patient participation is stimulated.

In addition, the experiments show the importance of alignment of expectations between stakeholders. It is important to understand and shape the different expectations to ensure that they are realistic and to identify at which points they differ. Agreement on the level of involvement and commitment to the experiment and its outcomes are also essential. If little is done with the patients’ input, patients will be reticent to become involved again. Neutral facilitation could help in the alignment of expectations and clarifying agreements. In agenda-setting processes, neutral facilitators are often present to guide the process. In partnerships in the conduct of research, such a person is not always present. Although it could help to align both parties to optimize collaboration, it also adds to the complexity of the research process, since an additional party has to be involved.

The case of the Network Patient Research Partners provided indications that the presence of organizational elements, in this case a network, can help to embed patient participation. A central and organizational element can be part of the structure of the health research system and has a
function to organize patient participation. This element should establish enduring relationships with important stakeholders. Patient participation can be coordinated from a network, and patients and researchers can be brought together (looking for the right match). A network can offer support, training, tools or information or advice. Moreover, it can function as a ‘knowledge junction’ where expertise, experiences, information, knowledge, tools, etc. are collected and made available.

To enhance the embedding of patient participation in health research, it is important that the benefits – the added value – are made explicit and visible. A distinction can be made between personal benefits or benefits for a certain research project and the impact on a larger scale. Making the benefits of engaging patients more explicit on the level of a research project creates a more positive attitude and more open-mindedness towards involvement and motivates researchers to involve patients. Making the added value explicit on a larger scale could serve as an incentive to new actors to start to experiment with patient participation as well.

Stage of embedding

With respect to embedding patient participation in the health research system, it can be observed that patient participation in research agenda-setting seems to be more embedded in the Dutch health research system than partnerships between researchers and patients in the conduct of research. When comparing the transition experiments related to research agenda-setting and the conduct of research described in this thesis, some interesting differences can be seen, which could be explained by characteristics of an innovation that are relevant for the speed of diffusion as distinguished by Rogers’s (2003); observability, relative advantage, complexity/simplicity and compatibility. In the case of patient participation in research agenda-setting, there is a proof-of-principle: involving patients in agenda-setting leads to new research topics, a broader scope of the agenda, but also provides legitimacy to research already financed by the funding agencies (if patients underline the importance of these topics). Also, clear examples of patient participation in agenda-setting initiated by governmental and funding organisations are increasingly available and made visible. With respect to patient participation in partnerships in the conduct of research, there is no proof-of-principle established yet in the Netherlands and examples are hardly visible; it is unclear what the benefits or outcomes for health research will be. The fact that governmental and funding organisations often commission shared agenda-setting projects provides incentives for researchers to be involved. Involving patients in the conduct of research provides hardly any incentives; it does not lead to scientific rewards, and it is not a funding requirement.

Furthermore, with patient participation in an agenda-setting process it is possible to restrict it to an isolated stage and it leads to a tangible, clear end-product, with the input of the involved stakeholders being clearly visible. It creates ownership by the involved stakeholders, resulting in support for the agenda. Additionally, funding agencies are considered better accountable by their members and donors. The isolated phase makes it more compatible with the current structure,
culture and practices of the health research system. Participation in the conduct of research is taken place in the core of research, which makes it more complex, requires a profound change in the incumbent culture, practice and structure. The involvement of patients is not limited to a certain activity and a specific time period and it does not always lead to a tangible end-product so the added value is implicit, since patients’ input is more interwoven with the input of researchers. Patients feel less ownership for the research project since their input is less visible. This makes patient participation more challenging requiring more skills, time and experimenting.

In addition, active consultation of patients (e.g. focus groups, interviews) is often a main approach to involve patients in agenda-setting. This requires no preparation from the patients and only a small amount of time. It can be presumed that the group of patients willing to become involved in such a single event is much larger that the group of patients willing to become structurally involved in the conduct of research. This latter requires regular time investments and more preparation and skills.

Based on the comparison, it can be expected that in the Netherlands the embedding of patient participation in agenda setting in the health research will be quicker and easier than the embedding of partnerships in the conduct of health research and that there is more to it than ‘just a matter of time’. In order to enhance partnerships in the conduct of health research, lessons can be learned from the UK, where the presence of patient research partners in research is already more common. In the UK, patient involvement in the conduct of health research is stimulated by governmental organisations and is more accepted.

**Insights in a shift towards a needs-oriented health research system**

Based on the developments described in this thesis, it may be concluded that a shift towards a health research system in which patients have an important role is emerging. Related to the different phases described in the multi-phase concept of transition theory (Figure 2.1), the points described above are indications that this system innovation is at the stage of take-off. Changes become visible and an increasing number of people and organizations are experiencing a positive effect. But does patient participation result in a more needs-oriented health research system?

Agenda-setting seems, based on the presented experiments as well as the international literature, a useful and suitable step in health research to involve patients and gain insights in their needs. The three experiments described in part 3 demonstrate that patients are capable of articulating their needs for health research in a facilitated process, covering a broad spectrum of topics related to their life with a disease. Patients and their representatives identified and prioritized new research topics, complementing those considered important by experts. In all three cases, patients confirm that topics related to fundamental research are of major importance since they
expect that it has the potential to lead to prevention, cure or improved treatment of the condition for the next generation. However, they often do not articulate their needs regarding basic research in detail. They expect researchers to be able to translate their ‘broad needs’ for basic research into suitable research projects. Professionals often expect patients to be subjective, only articulating and prioritizing needs directly relevant for themselves, and not prioritizing basic or long-term research. These examples indicate that the fears some professionals have are unfounded. Involvement of patients in agenda-setting provides a patient perspective on which topics should be researched.

The niche for patient participation in health research agenda setting has developed quickly in the last years and is still accelerating. An increasing number of patient organizations, funding organizations and governmental organizations established, or are currently establishing, a research agenda which includes the patient perspective. This niche seems to be scaling up since involving patients in setting the research agenda becomes more embedded in the current structure, culture and practice and is increasingly considered the ‘normal’ thing to do. Therefore, patient involvement in health research agenda-setting has great potential to stimulate a system innovation towards a more needs-oriented system, moving away from a supply-driven system where professionals set the research agenda.

However, there are two drawbacks. First, research topics identified and prioritized by patients are not automatically implemented or picked up by researchers. Having topics put on a research agenda does not guarantee that they will indeed be addressed. In a recent project we have observed that researchers tend to continue formulating project proposals on topics they are familiar with and rarely follow up on the ‘gaps’ in scientific knowledge that have been identified by patients (Broerse, Elberse et al. 2009). Second, research projects are usually formulated and executed according to ‘business-as-usual’. It does not lead to a change in how research is conducted. Developing a research agenda is only the beginning of the research cycle, and involvement of patients in this first step is only the start of a shift towards a more needs-oriented research system.

Network Patient Research Partners aimed for patient participation in the conduct of health research and thus tries to influence how research is conducted. There are some examples of how the input of patients influenced the research projects, like adapting questionnaires, improving e-health programs based on their input, and discussing relevant patient-reported outcomes to be included in studies. However, as already stated above, many barriers are experienced in the partnerships between patients and researchers, and not all partnerships were successful. This niche is still small and has had little impact on the regime so far. Several Dutch patient organizations are interested in setting up similar networks, but seem reluctant since there are very few good examples to follow. The regime is quite retentive in the current culture, structure and practice.
Therefore, on the basis of the findings of the research presented in the thesis, it is not yet possible to conclude that this form of structural patient participation will lead to more needs-oriented research on a larger scale although there are indications that this could be the case. Further research is needed to see if the involvement of patients in the conduct of health research can induce a shift to a more needs-oriented health research system.

**Box 10.1 Summary of the conclusion**

- Currently, there seems to be commitment from a growing number of organizations in the system to effectuate a change towards a more needs-oriented health research system. Several frontrunners and followers are experimenting with a variety of approaches to put patient participation into practice. At the same time, patient participation in health research is still not uncontested, and barriers are experienced related to features of the academic and private research community and the patient community. (Chapter 4)
- There is a need for designing, testing, reflecting, adapting and sharing of new approaches to realize patient participation. By doing this in a cyclical way, new best practices can be developed. (Chapter 4, 9)
- Special attention must be paid to exclusion mechanisms and inclusion strategies to stimulate a genuine dialogue between patients, researchers and policymakers. (Chapter 5)
- The Dialogue Model can be condensed for updating and extending a health research agenda, genuinely involving patients and researchers and leading to a state-of-the-art research agenda. (Chapter 6)
- Even when the focus of a research agenda is highly specified and preframed, patients are still capable of articulating their needs and providing useful and valuable input if the process is well facilitated and at the same time room is given to broader issues. (Chapter 7)
- The Dialogue Model is suitable to include a broad range of patients simultaneously, if additional attention is paid to the social conditions for all groups and saturation of the data. (Chapter 7)
- The adapted FIRST model is useful as guidance for establishing partnerships between patients and researchers. Adaptations to the models are made in relation to the acknowledgement that setting up successful partnerships is a dynamic process, and regular reflection on the process is needed as are support and facilitation for both researchers and patients. (Chapter 8)
- Important facilitators for patient participation related to the people involved are open-mindedness, willingness to experiment and learn, recognizing the fact that it takes joint efforts and regular dialogues to build structural relationships, being proactive and motivated. Important barriers for structural involvement are a low priority for researchers, the lack of initiative and know-how by both partners and researchers, little interaction within the
partnerships, and insufficient support from a network. In those cases that few facilitators and many barriers were present, few new routines and values were developed for involving partners. Important strategies to consolidate facilitators and address barriers are directed to building competences (awareness, attitude, skills and knowledge) and establishing a strong organisational element within the health research structure. (Chapter 9)

- The presence of change agents is essential. (This thesis)
- Alignment between stakeholders involved in a patient participation process, as well as making expectations towards each other evident is essential for fruitful collaboration. (This thesis)
- Patient participation in health research agenda-setting does result in insights into patients’ needs and therefore has the potential to enhance a system innovation towards a more needs-oriented system. However, there are two drawbacks. First, research topics identified and prioritized by patients are not necessarily implemented or picked up by researchers; and second, it might not change how research is conducted. (This thesis)

Reflection on the use of transition theory in light of patient participation

For the research described in this thesis, a system innovation perspective was chosen as a conceptual framework. This conceptual framework is predominantly used in a descriptive way in the areas of transitions in energy (Verbong and Loorbach 2012), agriculture (Klerkx and Leeuwis 2009) and mobility (Geels, Kemp et al. 2012) whereby technical developments are often central. It offers tools to unravel barriers and clarify facilitators. However, it also appeared to be very useful in the domain of health (Essink 2012; Schuitmaker 2013) and health research (Caron-Flinterman, Broerse et al. 2007). Also for this PhD research it provided very useful tools to explain the observations made in the different transition experiments. The transition theory provides useful terminology, indications on how to shape experiments and concepts to interpret the findings. Most scholars in the field of patient participation would agree that effectually shaping patient participation in health research will require changes in action and thinking of stakeholders in health research and transition theory provides a perspective to comprehend this needed change.

Another important body of knowledge for this PhD research was patient participation in health research. Combining this field of research with transition theory was sometimes challenging, since it brings together two different fields of knowledge and concepts. Also, articles are limited to a number of words, which does not always provide the opportunity to explain two bodies of knowledge in depth. The articles included in this thesis are therefore not always written in the light of transition theory, but focus mainly on the message for people in the field of patient participation in health research. Although the scientific contribution of this thesis will mainly be in
the area of patient participation, two supplementary notions can be made regarding transition theory. Firstly, transition theory could be used in a more prescriptive way than currently done. If applying transition theory, I would recommend considering current ‘culture, practices and structures’ in an early phase to develop strategies, forestall potential barriers, create facilitators and support those already in place. Secondly, especially network patient research partners indicate that non-powerful individuals as patients or researchers can become important change agents. It is not always necessary to have powerful organisations behind you to start a change. Sometimes the willingness and effort of single individuals can lead to significant change and actions than waiting for an entire organisation to be ready to take action.

Research validity

In this section, issues regarding the internal and external validity of the research findings are discussed. With regard to internal validity, the central role of the researcher will be discussed. Subsequently, the relevance of the research results is discussed in relation to patient participation in other steps of health research as well as the validity of the research for others.

Internal validity

In the described experiments, the research teams, including the author of this thesis, had a central role in designing and conducting the projects, analyzing the results and evaluating the process. In case of Network Patient Research Partners, the appointed monitors took over some tasks of the network coordinator. This central role offers the opportunity of a close proximity to data and a thorough analysis of the results as part of the context in which they took place. At the same time, it implies that a kind of ‘self-evaluation’ is done. Therefore, continuous reflection is required. Various validity checks were built in, as described in Chapter 3, to avoid bias due to self-evaluation, although some bias cannot be avoided.

1. Presenting rich data. Primary data was documented extensively to ensure interpretation within its specific context
2. Triangulation of methods and data sources
3. Using member checks
4. Data was analysed by multiple researchers
5. Regular reflections with colleagues and peers not involved in the projects. Also, on a regular basis, findings are presented at scientific conferences and gatherings.
6. In case of JUMP and Medical Products, there were external project groups that discussed all findings and steps taken. Reflection on data collection and analysis took place during meetings with these groups.
7. In the case of Network Patient Research Partners, two external advisors were appointed to safeguard sufficient distance from the project
With regard to patients involved in interviews, focus groups and partnerships, it is likely that there was an overrepresentation of ‘enthusiasts’: people who want to be involved, to share their ideas and perspectives, and to contribute to health research. In the agenda-setting cases, attention was paid to a broad representation of the patient groups by also recruiting patients via alternative channels (e.g. platforms, support groups). In Network Patient Research Partners, partners were part of the network and no selection took place by the monitors. However, having mostly ‘enthusiastic people’ does not have to be a drawback. These people are motivated and are willing to contribute to health research. Not everybody wants to become involved. And as rightly mentioned by Van de Bovenkamp, patient participation should not be a duty, but a choice (Van de Bovenkamp, Grit et al. 2008; Van de Bovenkamp, Trappenburg et al. 2010).

External validity
The studied transition experiments focus on two approaches for patient participation in health research: agenda-setting and partnerships between patients and researchers in the conduct on research. As explained in Chapter 3, the choices for the transition experiments are made partly pragmatically; due to the availability of suitable experiments and time constraints, it was decided to focus on these projects since they could contribute to the objectives posed in this thesis and address gaps in knowledge regarding patient participation. However, this entails that, based on the experiments described in this thesis, caution is needed to draw conclusions about other approaches of patient participation in the research process. Also, a different selection of experiments could have led to other conclusions. By focusing on ‘just’ these two approaches, an in-depth and detailed study was possible, resulting in findings not only on the macro level, but also on the micro level. As a consequence, the findings will probably be of more relevance for health research practice, and some findings and conclusions could be extrapolated to patient participation in other approaches of patient participation. The findings presented in this thesis can be extended to other countries as well which want to experiment with patient participation in health research, although cultural differences may influence the applicability. For countries like the UK, Canada and Austria where more knowledge is already available on patient participation, useful insights are provided to enhance this development. For countries rather new to this development, the thesis may provide pointers on how to shape such a process. In countries where similar societal trends can be witnessed like science becoming more democratic and participatory (Klein 2001; Nowotny, Scott et al. 2001; Chopyak and Levesque 2002), the public becoming increasingly empowered and demanding more legitimacy and accountability from science (Collins and Evans 2002; Bucchi and Neresini 2008) and the authority of experts declining (Cornwall and Jewkes 1995; Rowe and Frewer 2000; Kerr, Cunningham-Burley et al. 2007), experiments with patient participation in health research can be expected to be useful in enhancing a change towards a more needs-oriented system.
To increase the external validity of the research, I built in several checks. Besides the experiments described in this thesis, a large number of interviews and informal conversations have taken place with other scholars, patient representatives and policymakers in this field. Moreover, I visited many workshops, meetings and conferences related to patient participation in health research where relevant concepts were discussed. This helped me to reflect on findings, place my findings in a broader context and apply other peoples’ finding to my work.

**Future research**

As stated previous, many barriers are still encountered when involving patients in health research. Although much progress has been made in the last decade, there are still many interesting directions to explore to increase our understanding of effective patient participation in health research, and how a system innovation towards a more needs-oriented system can be enhanced. Therefore, I would like to suggest three future research directions: (1) systematic and thorough evaluation of projects, (2) development of new approaches, and (3) development of strategies to enhance a system innovation.

**Systematic and thorough evaluation of patient participation initiatives**

First, researchers, policymakers, and scholars from the field of patient participation in health research are urging a systematic and thoroughly evaluation of patient participation initiatives (Boote, Barber et al. 2006; Wright, Foster et al. 2010; Barber, Beresford et al. 2011). This will contribute to gaining more insight into the impact of patient participation in health research, since this will make the added value (outcomes) more explicit. To address this question, I would like to recommend an evaluation of different initiatives of patient participation in health research to obtain systematic insight into:

1. Impact of patient participation on health research (outcome criteria).
2. Suitability of research projects and tasks for patient participation (process criteria).
3. General criteria versus context-related criteria
4. Lessons learned regarding strategies and process facilitation.
5. Societal cost-benefit analysis

Such a systematic evaluation can contribute to the development of best practices, proof-of-principle and increased shared learning.

While every scholar in the field of patient participation will have sufficient anecdotal examples of impacts of patient participation on research, little systematic evaluation has been done. The first question asked by health researchers when discussing patient participation is often: What are the benefits for research? What will be the added value since time and investment are required for
participation? Can patients have an added value for my type of research? Currently, the issue of impact is becoming visible in the literature (Boote, Barber et al. 2006; Staley 2009; Wright, Foster et al. 2010; Barber, Beresford et al. 2011). I consider it of the utmost importance that scholars in the field of patient participation provide quantitative and qualitative insight on the direct and indirect impact of patient participation in health research. A ‘solid’ proof of added value can help to convince people, especially researchers and funding agencies, to consider patient participation.

Furthermore, not all projects will be suitable for patient participation. As demonstrated in this thesis, the involvement of patients in agenda-setting is probably suited for any type of research and any disease domain. This is likely to differ for patient participation in partnerships in the conduct of research. Hewlett et al. suggest involving patients as partners in research projects with a clinical component (Hewlett, De Wit et al. 2006). Otherwise, little is currently known. Also, not every level of participation will be suitable and applicable for every research project. The intrinsic and potentially utilitarian value of fundamental health research should not be underestimated. Therefore, the question of whether all health research should be submitted to the input of patients is valid. Some scholars argue that more participation will not always result in better health research or care. In every situation the value of participation should be assessed and which form of participation would suit the situation. Patient participation is not a panacea, and its use requires evidence-informed decisions and insight into suitable tasks and projects for patients in that context.

However, measuring the impact of patient participation in health research will be challenging, mainly for two reasons: (1) Impacts on what? and (2) how to measure impact (Barber, Boote et al. 2011). There is no consensus on what impact exactly implies: empowerment of patients, change of research procedures, more patients’ needs-oriented health research, better health outcomes, or changing values and attitudes of stakeholders (Staley 2009; Staniszewska, Adebajo et al. 2011)? Also, the question of ‘How to measure impact?’ has not yet been solved. For some activities in which patients were involved, the impact is quite explicit, like change of questionnaire used, improved intervention or setting priorities (Barber, Beresford et al. 2011). However, when patients provided input during decision-making processes, where decisions were made jointly, it is less clear how and how much this decision is influenced by the input of patients or what the final health improvement is (Epstein 2011). Nevertheless, just because measuring impact is complicated does not mean it should not be done. Especially if scholars want to enhance a system innovation towards a more needs-oriented system, it is highly important to gain insight into whether patient participation indeed can make the system more needs-oriented.

**Development of new approaches and best practices**

More experiments are needed to develop and further validate approaches for patient participation, especially related to patient participation in conducting research. An ongoing and
iterative cycle of planning, experimenting, reflecting, learning, sharing, and replanning can be followed. Applying a system perspective can help to gain more insight into facilitators and barriers, as well provide effective strategies. Also, it can stimulate the development of a shared vision of how the potentially new structures, cultures and practices of a needs-oriented health research system would look like. Within this development of new ‘best practices’ for patient participation in health research, the steering notions ‘deepening, broadening and scaling up’ can assist to effectively shape and experiment with this new role for patients. Also, insights obtained by the systematic evaluations of previous initiatives, as described in the first research direction, could assist in developing these new approaches and best practices. Experimenting could result in surprising insights and ideas for improvement.

Development of strategies to support and enhance a system innovation
A third research direction concerns more research to develop strategies which can support and enhance the system innovation. Barriers and facilitators for patient participation have been identified by different scholars in the patient participation literature (Stevens, Wilde et al. 2003; Caron-Flinterman, Broerse et al. 2005; Hubbard, Kidd et al. 2008; Saunders and Girgis 2010; Ward, Thompson et al. 2010). However, clear strategies to create and support facilitators and address barriers are rare. In Chapter 9, directions for these strategies are already provided. To stimulate a system innovation, more insight is needed into the process strengthening the competences of the people involved and strengthening relevant structures.

Concluding remark
Looking back at the research described in this thesis, ‘open mindedness’ is a frequently used word. For me, it means being open and respectful to each other and each other’s input and knowledge, and to explore what the others have to say. Open minded for new things, for change! Dare to take the risk to experiment with collaboration. But also to enjoy collaboration, value each other, learn from each other, complement each other and work together towards a shared goal: improvement of health, treatment and quality of life. Based on the last five years of research, I believe that the actors involved can induce a system innovation towards more needs-oriented, high-quality research if they become more open-minded. Therefore, I want to invite patients, researchers, policymakers, scholars in the field of patient participation and interested others to collaborate and to join in this change. And when doing this with an open mind, I strongly believe that patient participation can be valuable and indeed enjoyable.