SUMMARY

Our Western health research system is under pressure. Certain aspects of the system and its prevalent paradigm are increasingly felt to compromise its competency to improve patients’ health-related quality of life. The meaningful and sustainable involvement of patients in health research decision-making can aid in making the health research system more responsive to the perspectives of those people it ultimately concerns: patients.

First, the biomedical model, prevalent in our health research system, considers accumulated research evidence superior to clinical expertise and patient values. The diversity of patient perspectives is of secondary importance in this model. Related to this focus on research evidence, the biomedical research paradigm favours monodisciplinary over multidisciplinary research, thereby insufficiently acknowledging the complexity of the individual’s contextualized experience of illness. Second, mismatches have been identified between the research agendas and activities of researchers and the (research) needs of patients. Newly developed innovations with an interventionist focus do not always address relevant problems in current health practices, nor do they meet the needs of patients. Third, these knowledge-hierarchical, monodisciplinary and interventionist aspects of health research are reproduced through a self-governing system of peer evaluation. This enhances researchers’ interdependence, encouraging them to align to the dominant paradigm and impeding them to contribute to a system innovation. Last, the health research system can be characterized as supply-driven, where an elite group of stakeholders, mainly comprising of qualified academics and medical specialists, has taken the lead in health research decision-making. This can be associated with limited attention to the contextualisation of the produced knowledge and innovations, and poor responsiveness to the concerns and needs of patients.

One avenue for improvement is more reciprocal communication between researchers, healthcare professionals and patients to improve the applicability, utility and acceptability of health research; i.e. by involving patients in health research decision-making. Three arguments advocating patient involvement can be discerned. First, there is a democratic, normative imperative to involve those who are most directly affected by health research outcomes. Second, according to the instrumental argument, societal support for research is enhanced if patients are involved in its decision-making. Third, the substantive argument emphasizes the unique perspective patients can offer on health and disease. Patients’ daily experience of living with a disease and its consequences –their experiential knowledge- is complementary to the knowledge of researchers and healthcare professionals.

Despite these arguments in favour of involving patients, mainstreaming meaningful patient involvement in health research decision-making has proven problematic. Initiatives are often one-off events, their success remains dependent on enthusiastic individuals and the organizational embedding of the initiatives proves challenging. In other words, the
sustainability of the patients’ involvement is often limited. Additionally, tokenism is a recurring issue in patient involvement; window-dressing occurs when patients are asked and/or allowed to contribute only to aspects of the research process with little significance to the researchers, or when their input is side-lined or ignored.

The difficulty of meaningfully and sustainably involving patients can be understood by considering the systemic mechanisms that dominate health research decision-making. The current regime of the health research system is not configured to incorporate the patients’ perspective as common practice. Regime players share a dominant way of thinking (culture), organising (structure) and enacting (practice) health research decision-making, which hinders meaningful and sustainable patient involvement. For example, in the biomedical research paradigm the patients’ perspective is undervalued, obstructive or unsupportive resource infrastructures can be identified, and actors lack insight and/or willingness to involve patients meaningfully in decision-making processes. Moreover, these features of the health research system are systemic, meaning that a resistance to change from within the system counteracts the mainstreaming of niche-initiatives in which an alternative constellation is employed. To meaningfully and sustainably involve patients in health research decision-making, a system innovation is required in which these systemic mechanisms are overcome.

Based on these insights, this thesis aims to analyse and enhance meaningful and sustainable patient involvement in health research decision-making, in order to make the health research system more responsive to the perspectives of patients. To achieve this aim, we argue that there is a need to focus on the meaningfulness of patient involvement, and to unravel and overcome the systemic features which influence the sustainability of patient involvement in health research decision-making. The aim of this thesis was therefore divided into two research questions;

1. How can patients be meaningfully involved in health research decision-making?
2. How can the sustainability of patient involvement in health research decision-making be enhanced?

Case studies

In order to answer these research questions we conducted four qualitative, real-life transdisciplinary case studies, which provided the opportunity to facilitate and critically examine patient involvement in various stages of the health research decision-making cycle. In all case studies we took an action-oriented approach, which allowed us to intervene based on our preliminary insights and to stimulate stakeholders’ reflexivity.

Explicating patients’ research needs

To incorporate patients’ research needs meaningfully in health research decision-making, we investigated what these needs and their contexts entail. Therefore, in two case studies patients’ research needs were put centre stage: two research agendas were articulated,
one for patients with haematological cancers and one for people with visual impairments. The methodological challenges associated with setting a research agenda in a divided patient community were also discussed.

In the first case study, a research agenda from the patients’ perspective was set in collaboration with and intended for people with a haematological cancer. To identify and prioritize their everyday problems and research needs, we adopted the Dialogue Model. Patients’ everyday problems can be categorized as physical discomfort, psychosocial issues, problems with the healthcare system, and policy issues. Regarding research needs, respondents prioritize research aimed at factors potentially influencing survival, and research aimed at improving patients’ quality of life. We identified an underrepresentation of research of a social scientific character and as such, patients’ everyday problems are not all directly reflected in the research agenda. Our findings indicated that patients, besides emphasizing the importance of improving survival, have a desire to increase control over their lives.

The second case study aimed to identify and contextualize the research wishes of people with a visual impairment. Here we carried out the first four phases of the Dialogue Model. Patients were divided according to the aetiology of their ophthalmological disease or severity of visual impairment, and consulted in focus group discussions in which they identified everyday problems and research needs. Subsequently, patients were asked to prioritize the research needs on the medical domain and socio-psychological domain separately. For the medical research agenda, patients considered research aimed at treating the cause of ophthalmological disorders more important than research aimed at improving their quality of life. For the socio-psychological agenda, respondents considered needs concerning the improvement of technologies for people with visual impairments and navigation, orientation, and accessibility of public space top priorities. We also stratified the results regarding ophthalmological disorder, severity of visual impairment, age, age of onset, gender and membership of a patient organisation.

During this second case study, it became apparent that the heterogeneous patient community could be considered divided: especially patient representatives (rather than patients without a representative role) tended to defend the interests of patients with their type of ophthalmological disease, rather than unite forces with representatives from other patient communities. We analysed the participatory research agenda-setting process, employing the analytical framework of boundary work. We distinguished both our own and patient representatives’ efforts to create, manage or disrupt boundaries between the patient communities. Boundaries were reinforced by our strategies acknowledging the differences, to maintain support amongst patient representatives. Our management of boundaries concentrated on facilitating opportunities of reconciliation and on depoliticizing the study. Disruption of the boundaries was attempted in the heterogeneity of the project team’s composition. We conclude that the Dialogue Model is mainly effective in reducing
moderate substantive conflict, but that additional affective conflict-reducing strategies are required when a research agenda is set in a divided patient community.

Unravelling and overcoming systemic features which influence the sustainability of patient involvement

To sustainably involve patients in health research decision-making, a new way of organizing, thinking and acting is required. The systemic features influencing the adjustment of the health research system received explicit attention in the third and fourth case study.

In the third case study, the involvement of patient reviewers in proposal appraisal at the Dutch Cancer Society (DCS), a private research funding organization, was supported and reflexively monitored. The emergent and participatory research approach Reflexive Monitoring in Action (RMA) was applied in three phases. The inventory of the starting situation revealed a positive stance of stakeholders towards patient involvement, which was, however, accompanied by a lack of tools and structures. Based on the findings in this phase, interventions were designed and monitored during their execution. In the evaluation phase, patient reviewers and staff members reflected on the developments regarding patient involvement in proposal appraisal at DCS. Analysis revealed that structural interventions especially succeeded in the relatively bureaucratic context. We also addressed the cultural shift required at the funding organization, but the effects of these efforts took longer to materialize. This may have reduced the meaningful involvement of patients in proposal appraisal in practice. To overcome the underlying systemic barriers, an approach is needed that encourages stakeholders’ reflexivity in the structural, cultural, and practical domain.

The fourth case study aimed to involve patient representatives meaningfully and sustainably in research decision-making at a research department of an academic hospital. A collaboration between patient representatives and researchers was initiated, facilitated and reflexively monitored following the research approach RMA. Participants’ expectations regarding the collaboration were explicated. Subsequently, a process facilitator prepared and facilitated several dialogue meetings at which all participants were present, as well as numerous intermediate meetings with a subset of participants. All dialogue meetings concluded with a reflective component, in which participants explicitly evaluated the progress of the participatory practice. In evaluative interviews, the participants reflected on the participatory practice. In this study, we found that participants valued the patient perspective and were motivated to collaborate, but the project contributed relatively little to the institutionalization of patient involvement in research decision-making. This was due to several barriers such as difficulties to transfer the sense of ownership to staff of the organization. Also, the collaboration was not incorporated into the daily routines of the involved researchers, and participants did not adequately regard the collaboration as a learning experience on a reflexive level. Outside the sphere of influence of this collaboration, the rigidity of the medical research financing system limited the flexibility required to
meaningfully involve patients in research decision-making. To enhance the sustainability of meaningful patient involvement, the following elements are important conditions: (1) second-order reflexivity and an action-oriented stance of the participants to overcome the systemic barriers of a collaboration, (2) a sense of ownership within the organization, providing sufficient organizational and financial support to guide the required structural adjustments, and (3) lasting organizational flexibility to sustainably implement the results of the collaboration effectively.

**Meaningful patient involvement in health research decision-making**

The first research question of this thesis revolved around the meaningfulness of patient involvement in health research decision-making. To analyse this meaningfulness of patient involvement in our case studies, we operationalised this concept in terms of process and outcome objectives: stakeholder representation, supporting process structure, supporting process management, and direct and indirect outcomes.

To adequately voice the patients’ perspective, it is of key importance to involve a diverse group of patients. Also, patients need to be empowered in articulating their implicit experiential knowledge. Although we agree with a need for support for patient representatives in this respect, we argue for some restraint with regards to the extensive training of patients regarding (biomedical) scientific knowledge. Patient representatives may welcome such support, but unintentional side-effects may include the drifting apart of patient representatives from their grass-root support and the exclusion of less well-informed patient representatives. Furthermore, the process of empowerment goes both ways, in that articulating knowledge also contributes to empowerment. In this light, we fully acknowledge the importance of dialogue sessions, both between patient representatives and their grass-root support, as well as between patient representatives and other stakeholders.

In all case studies, a neutral process facilitator proved vital for supporting meaningful patient involvement. Such a facilitator can establish a transparent process structure by formulating the objectives and scope of the patient involvement and by clarifying patients’ and other stakeholders’ roles. The organisational context in which the facilitator operates also influences the meaningfulness of patient involvement in the decision-making processes. Furthermore, the value of direct interpersonal contact was frequently mentioned in our case studies; patient representatives in particular indicated that meeting other stakeholders in real life felt different from online contact, and that their involvement benefited from forming personal connections. In this regard, a process facilitator has a responsibility in nurturing an atmosphere of mutual respect, trust, openness and constructive interaction. It is important that the facilitator takes the affective relationships between stakeholders more explicitly into account than previously assumed; there is a need to enrich patient involvement methodologies using affective conflict management strategies, especially when a divided patient community is involved. In contrast to previous findings, our research
reveals that not all patient representatives (nor other stakeholders) felt the need to reflect and provide feedback on the process of patient involvement.

The aim of reaching consensus between stakeholders contributes to the formation and maintenance of a long-term partnership between stakeholders. Reaching such mutual agreement was indeed prominent throughout all case studies. However, the extent to which patients’ perspectives were incorporated in the decisions taken and the chances of implementation of these decisions varied. The research methodology RMA enabled us to convert such insights into recommendations to be applied on the very short term.

In all case studies, stakeholders especially built trust and learned from each other when they directly interacted over a sustained period of time. Learning mainly concerned substantive matters and reflection focused on defining concrete problems, to which concrete solutions were sought. On the procedural and reflective level, mutual learning took place to a lesser extent. Similarly, second-order reflection, which enhances the mutual understanding of stakeholders’ argumentations and their underlying values and assumptions, was generally limited.

**Sustainability of patient involvement in health research decision-making**

The second research question of this thesis focused on how to enhance the sustainability of meaningful patient involvement. We argue that the current health research system impedes meaningful and sustainable patient involvement in health research decision-making. Therefore, a system innovation (including changes in structure, culture and practice of the current regime) is required to make such involvement a common practice.

In the prevalent biomedical research paradigm, patient expertise is little appreciated. A deep-seated change in health research culture is needed. However, this is not yet acknowledged by all stakeholders and more reflection on the cultural shift regarding the values and assumptions underlying patient involvement is thus necessary.

First, most stakeholders in the case studies expressed that they were, in principle, supportive of patient involvement. However, despite this seeming enthusiasm, patients’ contributions were considered valuable only when researchers welcomed them as such. This shows that patient involvement is not yet an axiomatic component of the knowledge production process. Second, in some case studies researchers and patient representatives criticised the employed methodology because of its qualitative focus on contextualised data, maximising diversity and achieving data saturation, at the expense of statistical representativeness. This demonstrates that not only most scientists adhered to the positivist paradigm, but many patients seemingly did so as well. Third, in practice, involving patients in a meaningful and sustainable way in research decision-making requires researchers to adjust their scientific discourse. However, in our case studies patients were often expected and inclined to adjust themselves to the prevalent language and social
norms, rather than partaking in a mutual learning process. This demonstrates how culture (regarding the scientific paradigm) influences etiquette (regarding discourse), thus playing out in practice, and how both culture and practice need to change in order to involve patients meaningfully in decision-making processes.

To address this required change in health research culture, we attempted to improve stakeholders’ ability to reflect on the process on a second-order level. We introduced and facilitated recurring active moments of reflection in our case studies. Simultaneously, we aimed to align the context and the patient involvement initiative to maximize the system learning of involved stakeholders. However, such alignment with a results-oriented medical culture proved counterproductive; our attempts to stimulate reflectivity encountered opposition precisely because it did not yield any concrete results. As we have found few additional methodological directions in the existing literature on how to both stimulate reflectivity and align with the initiative’s context, we call for more research on this issue.

In addition to the need for adjustment of the health research culture, the health research infrastructure also needs to be reconfigured. This entails the adjustment of daily working routines to involve patients in decision-making, the allocation of budgetary and organisational resources to organise meaningful and sustainable patient involvement, and the responsiveness of the health research funding system to patients’ perspectives.

Encountered barriers in structure and practice are often related to logistics, such as the extra time, labour and money that patient involvement requires. In most of our case studies, the incorporation of patient involvement activities into the daily routines of involved researchers indeed proved problematic. Only when procedures and regulations were sufficiently bindingly altered, involving patients as common practice became a success. This demonstrates that sustained changes in structure are necessary to sustainably change practice. Also, allocating budgetary and organisational resources to appoint a neutral process facilitator is required for sustained patient involvement. From an organizational perspective, a sense of ownership over patient involvement activities is especially needed.

Last, the rigidity of the health research funding system has an impeding effect on patient involvement. To acquire funding in the current financing system of grant-allocated research, applicants must commit to predefined performance indicators. This complicates the adjustment of outcome measures or research set-up, limiting the flexibility that is required to involve patients meaningfully in research. Research funding agencies, which are increasingly supporting patient involvement, should therefore adopt a more lenient and encouraging position towards changes proposed during research conduct on the basis of patients’ input.