Research agendas involving patients: Factors that facilitate or impede translation of patients' perspectives in programming and implementation

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Patients are increasingly involved in agenda setting in health research policy, but little is known about whether or not patients' topics are translated into a funding programme and taken up by researchers. A qualitative evaluation of nine multi-stakeholder agenda-setting projects in the Netherlands was conducted. Document study and 54 semi-structured interviews with stakeholders were undertaken. Three strategies for the translation of research agendas into research programmes were identified: first, one-on-one translation; second, agendas were used to adapt general policies; and third, no translation. A number of factors, facilitating or impeding this translation, were identified, relating to the context or the process of programming and implementation. Context appeared to be crucial: positive attitudes towards patient involvement, good relations between stakeholders and supportive characteristics of organizations. Patient involvement was rarely sustained during programming and implementation. These insights contribute to more effective procedures for programming and implementing research agendas.

Keywords: Dialogue model; the Netherlands; priority setting; patient involvement; research agenda-setting; patients and researchers' research priorities.

1. Introduction

Traditionally, the health research system was supply driven: for instance, researchers, policy-makers of governmental and charity funding agencies, and the pharmaceutical industry were driving the research process and determining the research topics (Caron-Flinterman et al. 2006, 2007). Over the last decade, the health research systems developed a more needs-oriented focus, in which patients have been increasingly involved in health research policy-making, particularly in setting research agendas (Buckley 2010; Elberse et al. 2012; Oliver and Gray 2006; Staniszewska et al. 2007; Stewart et al. 2011). A research agenda can help to make efficient use of finite resources, to set up policies for research funding and programmes, and to steer funding and lobbying. Active involvement of patients in research agenda-setting gives patient a say and can result in an increased quality of research and relevance to patients' daily lives (Abelson et al. 2003; Caron-Flinterman et al. 2005a; Elberse et al. 2012; Irvin and Stansbury 2004). In addition, involving patients enhances the legitimacy of research policies (Abma and Broerse

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Box 1. The Dialogue Model

The model operationalizes consultation of and collaboration between various stakeholders, and is grounded in the notion that involvement is an interactive process between stakeholders. It emphasizes mutual learning processes between stakeholders by means of ongoing dialogues, and endeavours to include the perspectives of all stakeholders (Abma and Broerse 2010; Elberse et al. 2011; Schipper 2012). The Dialogue Model is based on six underlying key principles, according to which the process needs to be conducted: active engagement of patients, supportive social conditions, respect for experiential knowledge, ongoing dialogue, emergent and flexible design, and neutral process facilitation.

The model has an emergent design whereby activities are roughly structured into six phases:

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

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

(3) *Prioritization:* Stakeholder groups value the identified research topics identified in the previous phase and rank them in order of importance. Appropriate methods are a questionnaire for large groups, and the Delphi Technique or focus group discussions for smaller groups.

(4) *Integration:* The prioritized research topics of each stakeholder group are integrated into one integral research agenda. A dialogue meeting is an appropriate method to realize integration through deliberation. Mutual learning and creating shared ownership are important elements.

(5) *Programming:* The research agenda is translated into a research programme or action plan.

(6) *Implementation:* Implementation of the research programme/action plan with a call for research proposals, matching of research subjects and departments, and funding of research. Stakeholders (including patients) implement and take action, monitor progress and evaluate results.

2007, 2010; Telford et al. 2002). Governments, funding agencies and research departments are able to stimulate research on topics considered important by patients.

Patient involvement in research agenda-setting aims to increase the quality and legitimacy of the research policies of funding agencies and to stimulate research on topics considered important to patients, but there are few insights into whether or not this is achieved. Worldwide, various health authorities have experimented with involving patients in research agenda-setting. To this end, various deliberative approaches and strategies have been developed, varying in structure and using different qualitative and quantitative methodologies (Abelson et al. 2003; Mitton et al. 2009; Oliver and Gray 2006; Stewart et al. 2011). Examples of frequently used participatory approaches are the Priority Setting Partnerships developed by the James Lind Alliance in the UK (James Lind Alliance 2010; Elwyn et al. 2010; Lloyd and White 2011), the Cochrane Agenda and Priority Setting Methods Group in the UK (Bero and Binder 2012; Nasser et al. 2012), the Delphi Method in, for instance, Australia, Japan, USA and the UK (Bayley et al. 2004; Lopez 2003; Malcolm 2009), and the Nominal Group Technique used in the UK, Canada and Australia (Dewar et al. 2003; Gallagher et al. 1993; Jones and Hunter 1995). In the Netherlands, since 2003 various disease-specific charity funding agencies and governmental funding agencies have included the patient perspective in research agenda-setting to design their research policy. To this end, the Dialogue Model has been developed, which is used in most cases (Abma and Broerse 2010; Elberse 2012; Schipper 2012). However, experiences so far seem to indicate that patient involvement in research agenda-setting is not sufficient in itself to bring about a more needs-oriented health research system, since patient

involvement in subsequent phases of programming and programme implementation seems limited (Elberse 2012).

This paper has two objectives. First, we aim to gain insights into the extent to which research agendas set by multiple stakeholders, including patients, are translated into the policies of funding agencies and their programmes, and are subsequently implemented. We specifically assess whether and how research topics identified and prioritized by patients receive attention from researchers. Second, we assess the extent to which patient involvement, after setting a research agenda, is sustained in the subsequent phases of programming and implementation. This insight will contribute to more effective patient involvement in research agenda-setting and implementation. To meet these objectives, a qualitative evaluation of nine Dutch research agenda-setting projects, which involved patients and used the Dialogue Model, was conducted. Based on this evaluation, we identify factors which facilitate and impede patient involvement in programming and implementation of research agendas.

2. Methodology

The Dialogue Model was developed in the Netherlands (Abma and Broerse 2010) and is based on participatory and interactive methodologies and the Interactive Learning and Action approach (Abma 2006a; Abma et al. 2009; Brorerse 1998; Broerse and Bunders 2000; Caron-Flinterman et al. 2005b; Roelofsen et al. 2008). It comprises six phases (see Box 1):

- initiation and preparation
- consultation
- prioritization
- integration

- programming
- implementation

The first four phases have often been executed by an academic institute and have been validated and improved extensively in several settings (see Table 1). The subsequent phases of programming and implementation of the research agendas were mainly the responsibility of the bodies commissioning the research.

The research agenda-setting projects were selected based on the criteria that: first, they involved patients; second, they used the Dialogue Model; and third, were disease-specific. The nine research agendas were focused on the following diseases: spinal cord injuries; asthma/chronical obstructive pulmonary disease (COPD); update and extension of asthma/COPD and rare lung disease; renal failure; intellectual disabilities; diabetes; burns; neuromuscular disease; and congenital heart disease (see Table 1). Six of the nine projects were commissioned by disease-specific charities. The remaining three projects were initiated and financed by ZonMw (The Netherlands Organization for Health Research and Development), a governmental research funding agency. The evaluation took place between November 2010 and April 2012. A research team of five researchers (the authors) was established, as well as an advisory committee including three members of patient organizations and three policy-makers from funding agencies. The responsive evaluation methodology was employed (Abma 2006b; Greene 2001), comprising four phases: exploration; in-depth evaluation; validation and action planning; and integration and implementation. This paper focuses on the outcomes of the first two phases.

2.1 Phase 1: Exploration (November 2010–March 2011)

This phase provided an overview of the nine projects and initial insights into programming and implementation. Document analysis was performed involving research reports; documents relating to follow-up of the research agendas, either publically available or provided by key respondents; and published articles. The analysis focused on both process (stakeholder involvement, including patients) and outcomes (incorporation of input of different stakeholders, with a special focus on patients).

Eight key informants were identified from the research reports and approached for exploratory interviews: six were policy-makers and two were affiliated with patient organizations (spinal cord injuries, neuromuscular disease). All informants consented to be interviewed, with interviews lasting 60–90 minutes. The interviews were semi-structured and an interview guide was used with two main themes: research agenda-setting (first four phases) and programming and implementation activities. Attention was paid to patient involvement and how patients' input was implemented.

2.2 Phase 2: In-depth evaluation (April 2011–September 2011)

This phase covered stakeholders' experiences of programming and implementation, employing semi-structured interviews. Informants were identified during the desk study and exploratory interviews. All those who were approached consented to be interviewed, with one exception who felt unable to provide additional information.

In total, 46 in-depth interviews were held with 12 policy-makers, 12 representatives of patient organizations, 12 health-specific researchers, four qualitative researchers (involved in the execution of the research agenda-setting), and six project leaders. 43 interviews focused specifically on the agenda-setting projects. Three interviews were with informants who were involved at a more general level in the agenda-setting projects. The average duration was 90 minutes. The interview guide, developed for the exploratory interviews, was used as a basis and further specified for each research agenda and respondent, and as more insights were acquired. For instance, for asthma/COPD specific assessment criteria were drawn up to assist patient reviewers. In interviews extra attention was paid to how these criteria were developed.

2.3 Analyses

The findings of the document analysis were described in a research log. Interviews were audio-taped after consent and transcribed. A summary was sent to informants for each to check their own contribution. The following five questions were used to analyse the verbatim transcripts and the research log:

- What actions or decisions were taken in response to the research agenda?
- How are the topics of patients programmed and implemented?
- Which factors influenced the programming and implementation?
- To what extent did patients have an active role in programming and implementation?
- How did the stakeholders experience the phases of programming and implementation?

The data analysis was iterative, that is, the data were analysed during the process and the outcomes steered the rest of the process. The content analysis was open and oriented towards the above five questions. The transcripts were read line by line, labels were attached to text fragments, and subsequently clustered. Two researchers each analysed half of the data. The initial analysis was cross-checked. To check the accuracy of the coding, the coding matrix was discussed with the project team and further improved.

Table 1. Overview of research agendas

Disease domain	Commissioner	Period	Consulted	Programmed	Patient involvement	References
Spinal cord injuries	ZonMw	April 2003–April 2004	Patients	No. Though, one research topic on agenda received funding	– No research agenda related patient involvement activities have taken place – Based on experience with this project, patient involvement activities have been organized within ZonMw (e.g. patient reviewers, patients on advisory committees)	Abma 2005, Abma et al. (2004)
Asthma/COPD	Lung Foundation and ZonMw	August 2003–June 2004	Patients, health care professionals and researchers	Yes, one-on-one translation, resulting in funding programme active 2005–8	– Reviewing of research proposals by patient reviewers – Based on experience with this project, possibilities for patient involvement within Lung Foundation have been explored	Caron-Flinterman et al. (2006) Teerling, Caron-Flinterman and Broerse 2004, Elberse, Boer and Broerse (in press)
Revision and extension asthma/COPD and rare lung disease	Lung Foundation	March 2009–September 2009	Patients, health care professionals and researchers	Yes, one-on-one translation, resulting in a new funding programme active 2009–13	– A patient representative was part of programming committee – Reviewing of research proposals by patient reviewers	Elberse et al. (2012)
Renal failure	Kidney Fund	December 2005–May 2006	Patients	Yes, integration of research topics in their policy and programme structure	– None	Schipper and Abma 2011, Abma et al. (2007), Nierse et al. 2011 and Schipper et al. 2010
Intellectual disabilities	ZonMw	December 2005–September 2006	Patients	No	– No research agenda related patient involvement activities have been implemented – Based on experience in this project, patient involvement activities have been organized within ZonMw, e.g. patient reviewers, patients on advisory committees	[Nierse et al. 2011, Nierse et al. 2006 Nierse et al. 2007]
Diabetes	Dutch Diabetes Foundation	March 2006–July 2006	Patients	Yes, one-on-one translation, resulting in funding programme 'Voice of the patient' (*Stem van de patient*)	– A 'reviewing committee' (in which patient representatives are included) assesses relevance of submitted research for patients	[Broerse et al. 2006]
Burns	Dutch Burns Foundation	February 2006–February 2007	Patients, health care professionals and researchers	Yes, one-on-one translation in foundation's funding programme	– None	[Rensen et al. 2007, Broerse et al. 2010 and Broerse et al. 2010]
Neuromuscular diseases	ZonMw	April 2006–December 2007	Patients and researchers	No. Though, one research topic from agenda received funding	– No research agenda related patient involvement activities have been implemented – Based on experience in this project, patient involvement activities have been organized within ZonMw, e.g. patient reviewers, patients in advisory committees	[Nierse et al. 2007, Nierse et al. 2012]
Congenital heart disease	Heart Foundation	June 2007–October 2007	Patients, health care professionals and researchers	Yes. In 2008 a call for research proposals was financed on research topics on agenda. A new call was organized in 2012.	– A 'selection committee' in which one patient representative is included assesses research proposals	Elberse, Caron-Flinterman and De Cock-Buning (2007), Elberse et al. 2011]

2.4 Ethical considerations

The study aimed for anonymity and respect for privacy/confidentiality. The study did not need the approval of an accredited Dutch medical research ethics committee, since the study did not concern medical research or any form of invasion of the participants' integrity, and anonymity was guaranteed.

3. Results

First, the different strategies employed to translate research agendas into research programmes are identified. Second, factors which facilitate or impede programming and implementation of patients' priorities are presented. Third, we describe how patient involvement has been realized in the phases of programming and implementation.

3.1 Strategies for translation of research agendas

The organizations commissioning the agenda-setting projects dealt differently with the research agendas which resulted from the first four phases of the Dialogue Model. Three main strategies could be identified:

- Translation of the research topics of the research agenda one-on-one into a research programme or action plan. Research topics identified by patients were described in the research programme and in the call for proposals. The Burns Foundation, the Lung Foundation, the Heart Foundation and the Diabetes Foundation followed this strategy. Topics identified and prioritized by patients and researchers were directly recognizable by these stakeholders in the research programme and calls for proposals.

- The outcomes of the research agenda were used to adapt general policies and internal structures. For instance, research themes linked to critical moments in the daily lives of renal patients were used to change the programme structure of the Kidney Foundation. The new research agenda was integrated into the existing programmes 'Donation and transplantation' and 'Education', and has led to the creation of a new programme on 'Self-management'.
- The outcomes of the agenda-setting projects were not implemented in research programmes. For example, ZonMw did not programme and implement the findings from three of the agenda-setting projects they had financed. However, they funded one research topic identified by patients in the projects on spinal cord injuries and neuromuscular disease. Researchers involved in the agenda-setting process considered these topics important and they were able to include them in a research programme.

Most funding agencies did not consider beforehand which strategy to use. By establishing a research agenda, funding agencies followed a new, unfamiliar process in which patients' perspectives had a central place. Usually, the outcomes determined the strategy to be used. Table 1 provides an overview of the nine agenda-setting projects.

3.2 Factors that facilitate or impede

A variety of factors were identified that facilitate or impede the translation of research priorities identified by patients into a research programme or action plan and programme implementation (see Table 2). Factors are related to the context (the environment) or the process (execution) of programming and implementation.

Table 2. Factors that facilitate or impede programming and implementation of research agendas

Context			
Attitude	*Relations between stakeholders*	*Characteristics of organization*	*Time*
• *Willingness* to cooperate and *openness* to each others' perspectives • *Intention* to use topics from patients/*Ownership of agenda* • Presence of *change agents* • *Sense of urgency*	• Presence of existing (informal) *relations* • *Tension* between stakeholders • Representation	• Available *resources* for translation of patients' topics into a funding programme • Presence of *expertise* • -*Structure and procedures* of funding agency	• *Habitation* • *Timing* (e.g. available time between agenda setting and call for proposals)

Process		
Translation of research agenda	*Research proposal assessment*	*Evaluation of funded research*
• *Formulation research topics and themes* • *Current research trends* • *Shared perspectives* • Scientific *validation* research agenda • *Communication and dissemination* research agenda	• *Patient reviewer* • *Assessment criteria*	• *Check* on match funded research with research topics formulated by patients • *Check* on quality of funded research

3.2.1 Context.

Four clusters of factors related to context can be distinguished: attitude, stakeholders and relations, characteristics of the organization, and time.

3.2.1.1 Attitude.

The attitude of all stakeholders (policy-makers, researchers, carers, and patients' representatives) is crucial: support for the outcomes of the research agenda facilitates programming and implementation. There needs to be willingness among all stakeholders to collaborate and share decision-making power, and openness to each others' perspectives and, in particular, the perspectives of patients. Patients provide input from a different perspective than that of researchers who are more likely to focus on known research topics. Policy-makers and researchers who were directly involved in the agenda-setting project generally had a more positive attitude towards the perspectives of patients than those who were only involved in programming and implementation. In the project for intellectual disabilities, the attitude of researchers was predominantly negative towards patient involvement because they considered that people with intellectual disabilities could not provide valuable input for a research agenda. In this case, it was therefore decided to not include researchers in the project and to only focus on patient involvement. In the case of spinal cord injuries, the patients were the sceptics because, initially, they thought a research agenda would not have value. As one patient argued:

> A spinal cord injury can never be cured. We will never experience that.

Ideally all stakeholders should have the intention to programme and implement the research agenda. If the parties involved thought in advance about the implications for the research agenda, programming and implementation was easier. In the case of the agenda for spinal cord injuries, there was no concrete plan beforehand and the outcomes were not translated into a research programme or used in any other way. The Burns Foundation started the agenda-setting project with the clear purpose of directly translating the outcomes into their research programme with resources allocated in advance to make this happen.

At least one stakeholder group needs to feels ownership of the research agenda if the outcomes are to be implemented, although ownership by multiple stakeholder groups is preferred. In the case of research agendas commissioned by ZonMw, lack of ownership contributed to the fact the outcomes were not programmed. In addition, patient organizations should try to find ways to implement specific issues on the agenda that they consider important or relevant. For instance, the patient organization for people with burns developed a so-called buddy system, in which people with recent burns could share their experiences with other people with burns. The agenda-setting process revealed there was a need for such system but, since it was not considered a research topic, it did not become part of the research agenda. However, the patient organization decided to take up this issue itself. The patient organization for neuromuscular diseases did not have financial resources to programme the research agenda or to fund research. However, they used the outcomes to adapt their policy and to focus on projects and collaborations that address research topics prioritized by patients. However, as a patient also emphasized:

> You should know that patient organizations do not receive any financing. They do not have money. A lot of things have to be organized within projects.

Programming and implementation is most likely to be unsuccessful when there is no so-called change agent or ambassador. Change agents facilitate collaboration by the different stakeholders and aim to embed patients' perspectives. The following quote from a policy-maker illustrates how a change agent can positively stimulate patient involvement in programming and implementation:

> You should lose the thought that patients have nothing to tell. Also professors could be patients. They are not stupid!

A change agent (together with a patient representative) was clearly visible in the asthma/COPD project. The policy-maker of the Lung Foundation motivated stakeholders, convinced the scientific advisory board of the importance of patient involvement, and took the lead in implementing the research agenda. In the case of neuromuscular disease, a professor acted as the change agent. His involvement in the project team of the agenda-setting project was responsible for a more positive attitude among other researchers. A disadvantage is that the success of programming and implementation of research agendas is dependent on such an individual. When he or she leaves the organization, the perspectives of patients and the multi-stakeholder process are at risk. Many informants emphasized that successful patient involvement is closely linked to individual policy-makers as change agents. As a researcher stated:

> It [successful patient involvement] is completely dependent on individuals. For me, this [patient involvement] is essential. I do molecular research, and in addition I also do this [involving patients in research agenda setting] work. So, I want to combine, and continue to combine.

Another important factor relates to a sense of urgency which is needed if changes are to be made to research agendas. The interviews revealed that the input of patients was not always considered important by the scientific advisory boards of funding agencies. They had developed many research programmes without paying attention to specific (patient) topics, and did not have any reason to change this. Therefore, some scientific advisory boards were not motivated to actively programme patients' topics or stimulate the funding of research on these topics.

3.2.1.2 Relations between stakeholders. In most projects, there were no existing (in)formal relations between stakeholder groups (patient organizations, funding agencies, and research institutes) or there was tension between different stakeholder groups (such as disagreement on objectives) as a result of negative experiences from earlier collaborations. The different stakeholder groups were brought together during the agenda-setting projects but this did not automatically lead to partnership: when the joint project was finished and an agenda was delivered, each stakeholder group went back to its day-to-day activities. This was the case for the research agendas for renal failure, spinal cord injuries and intellectual disabilities. Relationships between stakeholder groups with a sense of shared responsibility contribute to a more greater level of progress in the agenda setting, and increase the chance of successful programming and implementation because groups have more insights into, and value, each other's perspectives and knowledge. For example, the Lung Foundation was originally a patient organization and is now both patient organization and funding agency. This contributed to cooperation between the patient group and policy-makers of the funding agency during programming and implementation. The research agenda has been programmed to pay attention to the topics selected by patients and patient involvement.

Another factor relates to the representation of the stakeholders who are involved. If the stakeholders who are involved represent an organization (instead of providing individual experiences), the research agenda was considered to be more valid and objective since the agenda-setting process and its outcomes are supported by a larger group (the organizations). This is particularly important for patient organizations. They are taken more seriously by the other stakeholders, when representatives represent a patient group and do not simply provide individual opinions. However, during the interviews several respondents questioned the extent to which representativeness is possible. This could be illustrated by a quote from a researcher:

> We just heard a group of patients. To what extent do they represent The patient? That is not possible.

3.2.1.3 Characteristics of organizations. Adapting internal structures and procedures was often necessary for successful programming and implementation. Some commissioning organizations adapted their reviewing process, the formation of research programmes, the division of funding, and external communication. For some organizations, adaptation of structures and procedures took little effort while, for others, it involved drastic adjustments. For example, the Diabetes Foundation created a new research programme which included research topics suggested by patients. This required little change because it was in parallel with existing programmes. The integration

of research agendas commissioned by ZonMw required more adjustments of procedures and structures because ZonMw is a large governmental funding agency with many hierarchical layers and with research programmes for different domains. In addition, the policy-makers who commissioned the research agenda were not necessarily involved in the decisions concerning the related research programmes. As a consequence, implementation and programming were hampered, with few research topics selected by patients being reflected in the research programmes.

Resources are needed if organizations want to adapt internal structures and procedures or pay specific attention to patients' topics. Resources (finance, time and employees) concern the internal capacity of both funding agencies and patient organizations. Finding a match between the topics chosen by the patients and potential research departments is time consuming. Also, funding agencies receive research proposals that could possibly, with some adjustments, correspond with the research topics selected by patients. However, assisting in these adjustments is labour intensive and expensive, and many organizations do not have these resources available. As one policy-maker from a funding agency noted:

> We try to involve a minimum of employees in the agenda setting project. But if you enter such a process, it is very labour intensive.

The resources of patient organizations are usually even more limited. As one patient representative said:

> You should keep in mind that patient organizations do not receive subsidies. They do not have money. So, a whole bunch of stuff is only possible on project basis.

Finally, some commissioning organizations wanted to give priority to patients' topics and involvement but did not have sufficient expertise, and, as a consequence, their involvements were limited. Some organizations, for instance the Lung Foundation and the Diabetes Foundation, experimented with involvement activities and developed expertise over time by trial and error.

3.2.1.4 Time. Time is required for the translation of the research topics into a research programme. Time is needed to think about an appropriate strategy for the translation of the research topics proposed by patients into a funding programme (and what role patients could have in this process). Furthermore, other stakeholders (researchers, policy-makers, and scientific advisory boards) need time to become accustomed to the new research programmes (habituation). The Burns Foundation, for instance, noticed that it took a few years before researchers started submitting research proposals on topics specifically identified by patients.

Another important factor is timing. For the research agenda on intellectual disabilities, the research programme

was established before the agenda setting was finished. As a result, only one research topic identified by patients was included. As a policy-maker explained:

> I think it [programming of the research agenda] did not happen because the staging [of the research programme and the agenda setting process] was not equal to each other. The agenda setting process was somewhat delayed and the research programme needed to be finished earlier.

3.2.2 Process. Three clusters of factors related to the process of programming and implementation are distinguished: the translation of the research agenda, the assessment of the research proposal, and the evaluation of the funded research.

3.2.2.1 Translation of research agenda. Some funding agencies mainly focus on biomedical and clinical research, while others focus on care research or the quality of life of patients. If the topics identified and prioritized by patients do not fit within the focus of the commissioning organization, programming and implementation is difficult. For instance, the research agenda for intellectual disabilities focused predominantly on social participation, care research, and quality of life. This was not consistent with the research programme which focused on biomedical research.

In a funding programme, research themes can be described very broadly (such as 'progress of disease') or very narrowly (such as 'relationship between regulation of blood sugar and mood swings'). A broad formulation of research themes and topics provides researchers with better opportunities to subsume their research within the research programme, although research may then addresses the needs of patients to a lesser extent. When research topics become more specific, research will be more needs-oriented, specifically addressing the articulated needs of patients and giving researchers less room for manoeuvre to address their favoured concerns. A policy-maker explains why they consider it important to formulate broad themes:

> When you formulate detailed themes, things [research topics] become very distant from each other. The question is whether you want that. We have a policy plan for four years. We have a horizon that is far away, and that is where we are heading. All we are doing should contribute to that [reaching the horizon].

If the identified research topics are in line with current research trends, it is more likely that research proposals will be submitted. For instance, researchers for neuromuscular disease pointed out that patients placed new topics (concerning quality of life and rehabilitation) on the research agenda. Although they agree that these topics are important, they also admit that, as scientists, they give them less priority because they have a lower status in the scientific community.

Stakeholder support for a research agenda increases when it represents a shared perspective of patients, researchers and health care professionals. Those involved in the agenda-setting process could share their own perspective and personally experience the added value of patients' perspectives to the research agenda. As a consequence, they were more likely to accept the research topics identified by patients. For instance, the research agendas for burns and asthma/COPD were accepted by researchers because they recognized their own priorities in the programme. The research agenda for diabetes has been programmed into a new separate research programme 'Voice of the patient' (*Stem van de patient*) and topics from patients have not been integrated with those of researchers. Although the research programme has no shared perspective, the topics from patients are clearly visible, and a call for proposals is specifically aimed at patients' topics. Due to this central positioning, researchers are specifically invited to send in proposals on patients' topics.

Another factor relates to scientific validity. Research agenda-setting is seen as more valid when it is conducted by an academic institute using a validated methodology and when the process is described in a scientific paper. A funding agency, therefore, often chooses an academic research team to conduct the research agenda-setting project. Scientific papers have been published for many agenda-setting projects (see Table 1). During the interviews, it was regularly indicated the papers contributed to acceptance of patients' topics on the agenda. As a researcher stated:

> Because a [scientific] publication is one of the few aspects, one of the few tools, by which people [other researchers] are persuaded.

Finally, the way in which a call for research proposals is communicated and disseminated to researchers influences its implementation. For instance, a few policy-makers of funding agencies who programmed the research agenda (e.g. Burns Foundation, Heart Foundation and Lung Foundation) indicated that it is difficult to write a call that makes researchers aware that research should correspond with the needs and preferences of patients, and that they should take into account the fact that patient reviewers also assess their proposals. Furthermore, the call should, in some cases, be disseminated using different channels to connect to other researchers who could potentially investigate patients' topics.

3.2.2.2 Research proposal assessment. The implementation of research topics considered to be a priority by patients could be realized when research proposals are reviewed from patients' perspectives, and ideally by patient reviewers (see next paragraph) by assessment criteria specifically drawn up for this purpose. These criteria assist patient reviewers in assessing research proposals and concentrate on societal relevance and patient

friendliness. A patient reviewer of the Lung Foundation explained how researchers could highlight the societal relevance in their research proposal better:

> To write clearly down what the [societal] implications are of the research over a three year time period. Or five year, whatsoever.

The Lung Foundation and the Diabetes Foundation make the distinction between scientific and societal quality of research, embedding the patients' perspective in the latter.

3.2.2.3 Evaluation of funded research. At the end of a subsidy round, most funding agencies do not check which research topics from the programme are covered. As a consequence, it is difficult to determine if the research topics identified by patients are researched. An exception to this is the Burns Foundation which checked the distribution of the approved applications and noticed that, although 'itching of scar tissue and donor places' was highly prioritized by patients and recognized as important by researchers, they received no proposals on this topic. Therefore, they actively lobbied for research groups on this topic. The Lung Foundation actively tried to match patients' topics to appropriate stakeholder groups when these topics were not appropriate for the research programme. For instance, patients gave a high priority to the issue of side-effects caused by medications and the Lung Foundation brought this to the attention of the pharmaceutical industry.

Furthermore, the quality of the execution of subsidized research has an influence on the success of the implementation because this determined whether the research contributed to more insights or solved the problem. A few funding agencies indicated they receive research proposals from non-scientists (such as patients). Most often, it concerns a patient topic with a high societal relevance, but the funding agencies experience that the scientific quality is often limited. For instance, the Burns Foundation decided to assist in increasing the scientific quality of the proposal.

3.3 Sustained patient involvement

In general, patient involvement in programming and implementation was marginal. In only two projects were patients involved in programming. For the revision and extension of the research agenda for asthma/COPD, a patient representative was part of the programming committee. Her specific task was to ensure the inclusion of patients' perspectives in the translation of the research agenda. For the other five research agendas, which have been programmed, the policy-makers of the funding agencies were responsible for programming.

In the implementation phase, several funding agencies organized participatory activities related to the assessment of research proposals from a patient perspective (see Table 1). For instance, the Lung Foundation appointed patient reviewers to join a scientific review committee to examine research proposals using assessment criteria specifically drawn up to review proposals from a patients' perspective. Researchers were required to provide an understandable Dutch summary written in lay terms for patient reviewers. The Heart Foundation has a review committee which includes a patient representative. No specific assessment criteria were developed for them at the time of this evaluation. The review committee of the Diabetes Foundation is composed of three patient representatives and two researchers in order to prevent patient representatives being overshadowed by researchers. The interviews revealed that the patient reviewers encountered several problems when reviewing research proposals. A researcher stated:

> Reviewing research proposals could sometimes result in problems. Especially when it concerns biomedical research. Researchers often use jargon which is too complicated for patients. As a result, the quality of patient reviews is variable.

The quality of patient reviews is also negatively affected by insufficient time for review and too many proposals.

In addition to participation in agenda setting, most commissioning organizations involved patients in other activities. For example, ZonMw appointed patient reviewers to assess research proposals, developed specific assessment criteria for review from a patients' perspective, established an advisory committee of patient representatives and included patient representatives on some programme committees.

4. Discussion and conclusions

As far as we know, this is the first detailed evaluation study of programming and implementation activities of research agendas, which have been set by involving patients in the decision-making process. It contributes to the growing need for systematic evaluation of patient involvement initiatives (Abelson et al. 2003; Abma and Broerse 2010; Elberse et al. 2011; Mitton et al. 2009; Stewart et al. 2011). Research agenda-setting projects can be considered effective when:

- the agenda is translated into the research policies of funding agencies
- the resulting policies are implemented
- specific attention is paid to patients' research topics
- patient involvement is sustained during programming and implementation

Our findings reveal that effectiveness for most projects was limited to the translation of the research agenda into a research programme and that there were few participatory activities during programme implementation. After the agenda was established in phase four of the Dialogue

Model, most collaboration between patients, researchers and funding agencies stopped. As a consequence, the patients' perspective was not sustained.

Six out of the nine evaluated research agendas have been programmed and included in research policies of funding agencies. Several research agendas contained both topics identified by patients and researchers. The agendas were, as a whole, translated into a research programme, whereby no specific attention was paid to patients' topics. Except for diabetes, the topics from patients and researchers were integrated in one funding programme. As a result, patients' topics were not clearly visible, which did not invite researchers to specifically submit proposals on these topics. Since researchers were not stimulated to specifically submit research proposals addressing patients' topics, it can be expected that researchers submit on familiar topics. Except for the research agendas for burns and asthma/COPD, no additional funding or allocated funding was provided. During programme implementation, three charity funding agencies and ZonMw organized patient involvement, particularly the reviewing of research proposals from a patient's perspective or acting in an advisory capacity to make adjustments to research proposals. To this end, two funding agencies drew up specific assessment criteria which could be used by patients to judge proposals (Teunissen et al. 2011).

Our findings indicate that commissioning organizations that carry out current programming and implementation activities stimulate research on patients' priorities, but have not yet been successful in including patients' perspectives in programming and implementation. To enhance and sustain the inclusion of patients' perspectives in these phases, more patient involvement activities seem to be needed in the follow-up of research agendas. These findings are consistent with those of the James Lind Alliance which established that funding agencies are still dominated by the culture of basic laboratory science (Staley and Hanley 2008). However, the James Lind Alliance has also observed a shift in the way research funding is allocated (Elwyn et al. 2010) with increased attention being paid to topics chosen by patients (Buckley et al. 2010; Lloyd et al. 2012).

Since the organizations commissioning the evaluated projects were initially not familiar with patient involvement in agenda setting, they usually did not consider in advance what they wanted to do with the outcomes and how patient involvement could be sustained. This may be inherent in their learning process. In addition, the funding agencies were not aware of the potential added value of patient involvement in programming and implementation. The scientific literature shows that patient involvement in programming and implementation provides embedding and sustainability of the perspectives of patients. Due to the involvement of the patients, their research topics are neither lost in translation nor become the victim of faulty

interpretation by other stakeholders (Abma and Broerse 2010; Stewart et al. 2011).

Differences in programming and implementation between disease-specific charities and ZonMw were observed. The ZonMw policy-makers who commissioned the research agendas were not necessarily involved in the related research programmes. As a result, these agendas were barely used as an input for research programming, either because they did not fit within the scope of the research programme or because the programme had already been developed. The disease-specific charities are smaller and have fewer research programmes and policy layers. Processes could more easily be adjusted which made it easier to translate the research agendas into a research programme. Programming and implementation appeared to be even more successful when the funding agency is also patient organization (such as the Lung Foundation), because this supported cooperation between the patient groups and policy-makers.

A number of factors were found to influence programming and implementation. O'Donnell and Entwistle (2004) found that patient involvement within funding agencies was affected by resources, types of research projects, and the extent of patient involvement at other stages of the research enterprise. We found that the context appeared to be most crucial for effective programming and implementation, including: positive attitudes to patient involvement, good relations between stakeholder groups and supportive characteristics of organizations (especially within funding agencies).

Currently, funding agencies are further expanding their patient involvement with patient reviewers and the inclusion of patient representatives on scientific advisory boards (Elberse et al. 2012). Patients could also fulfil other roles during programming and implementation, as can be seen from a structured overview of patient involvement by funding agencies in the UK, including patient involvement during the development of research proposals prior to submission, review of research proposals by patients, and the inclusion of patients on project advisory or steering groups, and in dissemination activities (O'Donnell and Entwistle 2004). Staniszewska et al. (2007) described the involvement of patients in the development of a research proposal, influencing aims, methods and ethical considerations.

4.1 Limitations of this study

First, the evaluation covered the period 2003–9 while the research was undertaken in 2010–11. This field is dynamic and further improvements and developments in patient involvement have taken place since then. For instance, several more funding agencies have developed assessment criteria from a patient perspective, and some funding agencies are exploring the possibilities of extending involvement activities to the monitoring of funded research and providing recommendations for the implementation of

the research. Second, patient organizations and funding agencies have seen changes in employees, which meant that some of our informants were not always conversant with the agenda-setting projects and the subsequent programming and implementation activities. To overcome this limitation, we undertook additional interviews and document analysis.

4.2 Recommendations

This study provides valuable insights which could contribute to the improvement of the Dialogue Model and more effective programming and implementation of shared research agendas by commissioning organizations and other stakeholders. For instance, in the exploratory phase, stakeholders need to prepare their strategy to translate the research topics into a research programme or action plan; their plan to formulate research themes and topics (broad or narrow) and research directions (e.g. biomedical research and care research); and their approach to sustain patient involvement. Concrete guidelines might be useful to bring about multi-stakeholder processes in the phases of programming and implementation phases (Abma et al. submitted). These guidelines should describe specifically what patient involvement activities are possible in which step of programming and implementation, what options there are to shape these activities (in line with the context of the specific funding agencies) and who should organize these activities.

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