

Patient involvement in agenda-setting processes in health research policy: A boundary work perspective

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1. Introduction

The involvement of patients in agenda setting in health research policy is gaining ground on the basis of three arguments (Telford et al. 2002). First, patients have acquired experiential knowledge through their daily encounter with their condition and its consequences. This is a unique source of knowledge, which could complement scientific or biomedical knowledge. This is called the *substantial* argument for patient involvement (Caron-Flinterman et al. 2005; Chalmers 1995; Faulkner and Nicholls 2001). Second, patients have a *moral right* to be involved in research policy as they are affected by the outcomes of scientific experimentation (Goodare and Smith 1995; Popay and Williams 1996). Third, there is a *political* argument to patient involvement: the chances of successful implementation of innovations are enhanced when patients are involved at an early stage of research and research policy (Collins and Evans 2002; Williamson 2001). It aims to increase the quality and legitimacy of research policies of, for instance, funding agencies and to stimulate research considered important by patients.

Various methodologies have been developed to guide the introduction of patient perspectives in agenda setting in health research policy. These methodologies vary in structure, frequency and level of involvement. Examples are the James Lind Alliance (JLA) in the UK, which facilitates priority setting partnerships (PSPs) and other collaborations in which patients, caregivers, and clinicians aim to list research priorities regarding a particular disease (Priority Setting Partnerships 2017). In the USA, the Patient-Centered Outcomes Research Institute (PCORI) funds research that has been proposed by patients and other stakeholders (PCORI 2015). Other methods include the Cochrane Agenda and Priority Setting Methods Group (Nasser et al. 2013) and demand articulation processes (Boon et al. 2011). Compared to these approaches, the Dutch Dialogue Model developed by Abma and Broerse (2010) is more oriented toward mapping the context of patients' research wishes by explicating their disease experiences, everyday problems, and concerns. An acknowledged merit of the Dialogue Model is its explicit attention to differences in discourse and attitude between patients, researchers, and

healthcare professionals (Abma 2006; Abma et al. 2014; Broerse et al. 2010).

The complementary nature of experiential knowledge implies that there is heterogeneity between different stakeholders: they may have different demands, ideas, or judgments regarding (biomedical) research (Caron-Flinterman et al. 2005). Differences in discourse, attitudes, power and status of stakeholders in health research decision-making have been studied in-depth both empirically and conceptually (Abma 2006; Abma et al. 2014; McKie 2003; Wehrens 2014). This recognition of the different perspectives of stakeholders encourages a knowledge co-production process in which patients' and researchers' knowledge is articulated and integrated to develop robust and reliable knowledge regarding a condition (which is better than the visions of separate perspectives) (Pittens 2013). A certain level of homogeneity in perspectives is assumed within stakeholder groups. However, it may be that a stakeholder group is more divided than initially assumed by outsiders. We consider a stakeholder group divided when members or representatives of a subgroup consider their perspectives as so different from other subgroups that they define themselves as a separate stakeholder group and defend their own interests rather than unite forces with other subgroups.

Agenda setting in health research policy from the patients' perspectives has often taken place in contexts with well-organized and relatively homogenous patient communities (Broerse et al. 2010a; Deane et al. 2014; Elwyn et al. 2010; Gadsby et al. 2012). A more heterogeneous patient community was involved in a number of research agenda-setting studies employing the Dialogue Model (Elberse et al. 2012; Nierse et al. 2013). In these cases, however, the heterogeneity in the patient community was not associated with explicit and/or articulated dividedness, and the challenges associated with these patient communities were not the primary object of study. In addition, how to set up a PSP in a divided patient community is not addressed in the JLA Guidebook (Cowan and Oliver 2016). Hence, the challenges associated with a research agenda-setting process in a divided patient community have received relatively little scholarly attention. Although some scholars have explored the link between social identities and patient involvement

(Allsop et al. 2004; Renedo and Marston 2011), their focus was on the effects of social network formation on the discourse with professionals and less on the existence or management of tensions within patient communities.

The organization of research funding differs between countries. In the Netherlands, health research agendas are primarily set by the publicly funded research organizations NWO [Netherlands Research Organisation] and ZonMW [The Netherlands Organisation for Health Research and Development]. Research funding takes place through competitive calls for proposals, which can be either aimed at a specific condition, be open to any condition, or promote (multidisciplinary) research across disease domains. In addition, privately funded research funding agencies have similar funding schemes but are generally more focused on a certain condition. In this competitive funding system, groups with different conditions may compete for research resources, but ideally the system promotes research aimed at multiple closely related diseases. This latter argument is often used by research groups and patient representatives alike to join forces.

This article focuses on the methodology of research agenda setting from the patients' perspectives for patient communities that can be considered divided (by outsiders and by themselves). Academic attention to this issue is needed because the dividedness of a patient community challenges the development of a widely supported research agenda amongst its stakeholders. Subsequently, the implementation success of such a research agenda can come under pressure. For example, Pittens et al. (2014) have shown that the translation of research agendas into research policies of funding agencies and their programs requires good relations between stakeholders.

It is thus important to gain insight into the underlying reasons of dividedness of patient communities and possible strategies to enhance alignment in research agenda-setting processes. To this end, a research agenda-setting process involving people with a visual impairment was analyzed using the concept of boundary work, as introduced by Gieryn (1983). Boundary work describes the discursive practices of actors that demarcate the boundaries of their field of expertise. Since the studied case applied the Dialogue Model to agenda setting, the analysis allows us to assess the Dialogue Model in relation to its strategies to align a divided patient community effectively and draw lessons to guide agenda-setting processes in health research policy with such patient communities in the future.

2. The Dialogue Model: an established approach to bridge heterogeneity between stakeholders

The Dialogue Model acknowledges the heterogeneity of perspectives between patients, researchers, and healthcare professionals, and takes it as a starting point for knowledge co-production. The Dialogue Model can be initiated by for instance research institutes, patient organizations or funding agencies (often in collaboration with each other). The initiators are often also considered the 'owners' of the agenda. However, the real power and control generally lie with the funding agencies who decide whether or not a research project gets funded. However, based on a research agenda, researchers/research institutes and/or patient organizations can decide to only adhere to research which is in line with the research agenda, thereby indirectly steering (and controlling) the focus of research.

The approach is based on the premise that after specifying and explicating all stakeholders' perspectives, integration of the

perspectives can take place (Pittens 2013). As described by Abma et al. (2014: 13), 'the Dialogue Model was originally grounded in the notion of partnership and dialogue'. Its six guiding principles reflect the following position: (1) *the active engagement of patients* implies that extra attention should be paid to incorporate this stakeholder group in the decision-making process; (2) *conducting social conditions*, such as openness, trust, and respect is needed to realize a genuine dialogue between the stakeholders; (3) *respect for experiential knowledge* means that one should aim at understanding patients' questions and concerns to incorporate them in the decisions; (4) a genuine *dialogue* provides an opportunity for stakeholders to listen to each other and learn about their own and each other's perspectives and experiences, which may lead to an adjustment of participants' opinions; (5) *emergent and flexible design* allows unexpected issues to be addressed, by adjusting the research plan; and (6) *process facilitation* executed by an independent party, with no stake in the outcome, can create the above-required conditions for successful participation and dialogue (Broerse et al. 2010).

The Dialogue Model has been employed involving a heterogeneous patient community in a number of cases, of which two examples will be elaborated. First, people with various neuromuscular diseases (NMDs) were involved in setting a joint research agenda (Nierse et al. 2013). In this case, 'intense discussions' (406) resulted in the inclusion of a limited number of diagnoses. This methodological choice was not contested afterwards and the research agenda for NMDs was supported amongst members of the Dutch Patient Association for NMDs. Second, in the establishment of a research agenda for medical products, the heterogeneity of the involved patient population was considered challenging. In this case, input from patient groups of 15 types of diseases was sought, ranging from people with burns to anxiety disorders (Elberse et al. 2012). Little overlap between the patient groups was encountered on specific medical needs; however, shared research themes could be identified. Abma et al. (2014: 10) describe how 'balancing between a general and specific patient agenda appeared to be difficult' in various cases employing the Dialogue Model. Indeed, Elberse et al. (2012) experienced a dilemma between creating a research agenda relevant for many patient communities and specifying data for distinct patient communities.

The choice to employ the Dialogue Model in a divided patient community was justified, considering its conceptual emphasis on and empirical experience to deal with heterogeneity. However, the responsiveness of the approach to the challenges associated with a divided patient community, such as people with a visual impairment in the Netherlands, is unknown.

3. Methods

To gain insight into the responsiveness of the Dialogue Model in relation to a divided patient community, we conducted a case study. To this end, we studied a research agenda-setting process for people with visual impairments employing the analytical framework of boundary work.

3.1 Case description

The research agenda was initiated in 2013 by two organizations acting for people with a visual impairment in the Netherlands. An umbrella organization represents people with any ophthalmological disease with approximately 7,000 members in 2014 and sixteen employed staff members. This organization initiated the research

agenda. The second organization, not part of this umbrella organization, represents people with one specific type of ophthalmological disease and has approximately 4,000 members and one paid employee in 2014.

People with visual impairments can be characterized as a heterogeneous patient population. It has been shown that a variety of factors can impact people's health-related quality of life, including the severity of their visual impairment and an anxiety for potential worsening of their condition (Williams et al. 1998). Also, a variety of visual impairments have been grouped together under low vision—visual acuity under normal and low luminance, contrast and glare sensitivity, stereo-acuity, and visual fields all affect people's self-reported visual disability (Rubin et al. 2001). This heterogeneity within the patient population and a fragmented social landscape of patient representation in the Netherlands are intertwined, illustrating the dividedness of the patient community. For example, patient communities pursuant to a specific ophthalmological disease fund scientific research specifically aimed at their own disease. Also, a number of patient communities are organized in separate organizations not linked to the umbrella organization as they feel there is too little common ground with those of other ophthalmological conditions.

3.2 Research-setting activities

See Box 1 for an overview of the research agenda-setting activities. In the exploration phase, the social landscape was mapped out. In the consultation phase, people's daily life problems and concerns were investigated and translated into research needs. Subsequently, the research needs were ranked in the prioritization phase. Lastly, in the agenda-setting phase, the research agenda was discussed with other stakeholders such as ophthalmological researchers, healthcare professionals, and research funders. Three independent academic researchers (authors of this study), whose primary concern was to safeguard and monitor the program fidelity, facilitated the process. A more detailed description of the research agenda-setting activities and its outcomes can be found in (Scholvinck et al. 2017).

3.3 Data collection

During the research agenda-setting process, all interventions directly contributed to the establishment of the research agenda following the Dialogue Model (see Box 1). In addition, data were collected post-hoc to reflect upon the research agenda-setting process and its implementation. Data collection for this study consisted of the following elements:

- During the exploration phase, six formal semi-structured interviews were conducted with representatives of various patient communities, such as people with macular degeneration, retinitis pigmentosa, glaucoma, and parents of children with a visual impairment. The interviews were aimed at gaining insight into the relationships between the patient communities; hence, the dividedness of the patient community was discussed explicitly.
- During the consultation phase, the authors AS and CP employed participant observation to analyze the agenda-setting process. Observations were related to participants' attitudes, behaviors, and input toward the subject and toward each other.
- During all the research agenda-setting activities, numerous informal conversations took place with participants and members of the project team regarding the process of the study.

Box 1. Activities to set the research agenda for people with a visual impairment (September 2013–October 2014).

Phase 1: Exploration (3 months): To gain insight into the patient community, six preparatory interviews were held with patient representatives on behalf of different patient communities. In addition, a project team consisting of eight patient representatives and three facilitators (the authors of this article) was established.

Phase 2: Consultation (3 months): To explicate daily life problems and concerns and to subsequently identify research needs of people with a visual impairment, eight focus group discussions (FGDs) were organized ($n = 82$). The aim was for homogeneity among the participants in which either ophthalmological disease, target group, or severity of the visual impairment was considered. In addition, semi-structured interviews ($n = 7$) were held with members from underrepresented patient groups. Reflection upon the results of each FGD took place with two to three patient representatives of the corresponding patient community (feedback meetings). In a qualitative analysis, people's daily life problems and concerns were mapped, and research needs were categorized as medical or socio-psychological. Research needs were organized in seven medical and six socio-psychological themes under which specific research topics were included.

Phase 3: Priority setting (6 months): Based on the data gathered during the consultation phase, two surveys (focusing on medical and socio-psychological research) were developed to prioritize the research topics. Respondents ($n = 850$) were asked to allocate priorities to the most important topics within each theme and across the themes. Personal data were collected. A quantitative analysis stratified the survey results according to age, gender, ophthalmological disease, severity of visual impairment, and membership of a patient association.

Phase 4: Agenda setting (2 months): To create support for the preliminary results of the research agenda and to take the first steps toward implementation, a dialogue meeting was organized which was attended by ophthalmological researchers and/or medical doctors in various ophthalmological disciplines ($n = 7$), staff members of research funding agencies and/or revalidation institutes ($n = 9$), and patient-representatives ($n = 19$).

- Approximately 1 year after the completion of the research agenda, four key informants were interviewed (representatives from the two involved patient organizations, a research funding agency, and a program council) to gain insight into the interviewees' views regarding the research agenda-setting process. Also, respondents were asked about their organization's plans to implement the research agenda.
- Post hoc, a document analysis was performed on: (1) summaries and transcripts of the interviews, FGDs, feedback meetings, and project team meetings; (2) written and electronic correspondence regarding the research agenda-setting process; and (3) comments provided by respondents of the questionnaire. The document analysis was aimed at analyzing the interactions of the involved stakeholders.
- Throughout the research agenda-setting activities and post-hoc, the facilitators regularly discussed their own roles and responsibilities regarding the management of the research agenda-setting

process. This form of self-reflection aided in evaluating the effectiveness of the employed strategies of the facilitators.

3.4 Data analysis

After consent, the interviews and FGDs conducted during the consultation phase and the post-hoc reflection interviews were audio-taped and transcribed verbatim. All formal interviews, FGDs, feedback meetings, and the dialogue meeting were summarized. The reports were sent to the participants for member-check. Observations, conversations, and discussions within the project team as well as others involved, either in person or through e-mail, were recorded in the researchers' logbooks and frequently discussed among the authors of the study.

The transcripts and logbook were analyzed through a thematic content analysis (Green and Thorogood 2004), applying the analytical framework of boundary work. This theory describes the discursive practices by actors within a field to mark the boundaries of that field (Gieryn 1983). Although the theory was originally developed to study scientists demarcating science from non-science, the theory has been extended outside the realm of science, for example in other (health-related) contexts (Burri 2008; Hall 2005) and to examine tensions within a community (Albert et al. 2009). Zietsma and Lawrence (2010) have extended Gieryn's (1983) definition of boundary *creation*. Actions aimed at bridging the gap between boundaries are described as the *management* of cross-boundary connections. The *disruption* of boundaries, for example by integration of different patient communities, is identified as the third type of boundary work. Following Zietsma and Lawrence (2010), we conceptually distinguished observed boundary work into the (1) creation, (2) management, and (3) disruption of boundaries. The coding process was done in an iterative manner, in which the authors discussed the coding regularly and switched from the initial open coding to the thematic coding scheme. All citations in this article were translated from Dutch.

3.5 Ethical considerations

The privacy of participants and confidentiality of the data were assured through careful data management. As the study did not concern medical research or any form of invasion of the participant's integrity, approval of an accredited Dutch medical research ethics committee was not required. Instead, the authors were guided by the ethical framework provided by Richards and Schwartz (2002), who have considered potential risks to participants in qualitative health research and offered suggestions to minimize these risks. Three issues particularly concerned this study. First, the possibility of misrepresentation of the socially constructed truth was considered, as the analysis of the data was inevitably influenced by the theoretical framework applied and preconceptions of the involved researchers. To avoid such misrepresentation of the narrative, the manuscript has been presented to the involved stakeholders prior to submission. Also, data triangulation and self-reflection of the authors were aimed at contextualizing the data correctly. Second, avoiding the identification of the participants involved a precarious balancing act between ensuring the anonymity of the participants (e.g. by not revealing the involved patient organizations) and describing the situation with enough specificity to make sense. Although the participants may recognize themselves in this article, the (inter)actions of the involved actors are untraceable to anyone who has not participated in the project team. Third, we treated informed consent as an on-going process, as this study's research objective arose

during the research agenda-setting process and gained clarity post-hoc. In addition to the agreements that were made prior to study about the use of data generated in the study, participants were informed about the authors' interests in their perception of the agenda-setting process and the implementation plans when interviewed post-hoc. Also, participants' consents to publication of the manuscript were especially important in this respect.

4. Results

In this section, we describe the research agenda-setting process with a divided patient community following Zietsma and Lawrence (2010), who conceptually divide boundary work in actions aimed at (1) creating, (2) managing, and (3) disrupting boundaries. The research agenda from the perspective of individuals with a visual impairment can be found in Scholvinck et al. (2017).

4.1 Creation of boundaries

The reinforcement of boundaries between the patient communities was prominent throughout this study. It pervaded each phase of the research agenda-setting activities and was brought up repeatedly in the post-hoc reflection interviews. However, individual patients consulted in the FGDs and results of surveys were shown generally to be less outspoken, the patient representatives often strongly emphasized the difference in daily life problems, concerns, and research wishes between people with various ophthalmological diseases and varying levels of severity of visual impairments. Such discussions occurred between and within representatives of the participating patient organizations. The distrust of patient representatives toward each other and mutual claims of being discriminated by other patient communities did not seem to diminish throughout the course of the research-setting activities.

In the explorative interviews, for example, an interviewee pointed out that the problems experienced by societally blind people (with a residual vision of <5%) are substantially different from the problems of completely blind people (with a residual vision of 0%). According to the interviewee, people with limited residual vision often do not consider themselves as blind, and they have substantially different coping strategies compared to those who are completely blind. Yet, by counting societally blind people to the 'faction of the blind', the entire group of blind people appears to be larger. This enhances their political and societal importance. Also, according to this interviewee, society feels sorry for blind people than for people with low vision. Finally, the interviewee believed that blind people were more assertive in patient advocacy than people with low vision. All these factors would fallaciously enhance the legitimacy of the 'lobby of the blind' at the expense of people with low vision. This example demonstrates how this patient representative emphasized a perceived heterogeneity that strengthened the dividedness between the patient communities thereby creating boundaries. It also illustrates how a substantive issue fueled the feelings of being disadvantaged; this patient representative did not just consider the factual differences and similarities between people with varying severity of visual impairment, (s)he interpreted the blind people's 'lobby' as harmful to the interests of people with low vision.

In response to these sentiments, the project team appointed five out of eight FGDs in the consultation phase to a specific ophthalmological disease to increase the mutual recognition of the FGD participants' experiences. For example, people with glaucoma were invited to a different FGD than people suffering from retinal disorders. The

remaining three FGDs were organized for a mixed-patient community. The project team asked a patient organization, which did not take part in the project, to invite their members to an FGD to have their members' issues covered as well. However, the board members of this organization seemed to consider the participating organizations as not giving them enough acknowledgment in the process. While they were granted simultaneous access to the (publicly available) end report, they rejected the request to invite their members to the FGD.

To enhance the support for the outcomes of the consultation phase, seven feedback meetings were additionally organized with patient representatives in which the FGDs were discussed. The composition of these feedback groups was a cause for fierce debates; both patient organizations wanted to take seat to endorse the results and accused each other of partiality or having conflicting interests. Nevertheless, considering the outcomes of the FGDs, quite extensive overlap on the issues between the patient communities can be revealed: out of 101 everyday life problems and concerns mentioned in the consultation phase, eleven problems (10.9%) were specifically relevant for only one patient community. The heterogeneity or dividedness between patients was neither an explicit topic of discussion brought up by the participants in the FGDs. This dichotomy suggests the dividedness amongst patient representatives was, at least, partly caused by feelings of distrust.

In the priority-setting phase, patient representatives and members of the project team repeatedly scrutinized the scientific legitimacy of the employed methodology. Especially the recruitment strategies of the questionnaire were criticized, and the representativeness of the respondents was questioned regarding the etiology and severity of people's visual impairment. The facilitators tried to strengthen the support for the research agenda by stratifying the survey results with respect to type of ophthalmological disease, severity of the visual impairment, age of onset of the visual impairment, gender, age, and membership in a patient association (see Box 2 for the stratification results). This analysis revealed respondents' prioritized research themes quite homogeneously. A few differences were detected on specific research topics, especially regarding medical topics. In addition, respondents were asked to provide feedback on the questionnaire; eighty-two out of 850 respondents of the questionnaire (9.6%) indicated they had not seen questions regarding their specific ophthalmological disease or that the entire questionnaire was insufficiently tailored to their medical situation. However, some of the disorder-specific topics brought to the attention in these comments were indeed queried in the questionnaire. Presumably, in these cases, respondents used the open question as an opportunity to emphasize the weight they wanted to give to this particular topic.

In the post-hoc reflection interviews, almost all interviewees pointed out that the divided patient community hinders the joint allocation of research resources. People with a specific ophthalmological disease support several Dutch funding agencies; because of this fragmentation, they have little financial power to have a substantial impact on their research field of interest. Interviewees considered the lack of coordination between the priorities of these funds a substantial barrier for the implementation of the research agenda. One of the involved patient associations made an attempt to coordinate this endeavor by taking the lead in an implementation program. However, a patient representative of a different organization continued to express doubts regarding the possibility and desirability of shared action. Despite the relatively homogenous research agenda, this illustrates that the deep-grained perceived boundaries

Box 2. Stratification of prioritized research topics based on type of ophthalmological disease, severity of the visual impairment, age at onset of the visual impairment, gender, age, and membership in a patient association

Stratification on type of ophthalmological disease: analysis revealed that people with glaucoma favored research that investigated ways to improve duration of action of medication, the relation between eye-ball-pressure and damage to the optic nerve, and means to develop a method to detect normal-pressure glaucoma. Respondents with MD sought research that looked into differences in severity of ophthalmological diseases, the efficacy of familial screening to detect a disease at an early stage, and at the development of an alternative method of application of eye medication. People with a retinal disorder were particularly interested in the risks and preventive measures regarding retinal detachment. People who were diagnosed deaf-blind prioritized research into diagnostics to detect this specific combination. People with a corneal disorder and people with acquired brain damage did not prefer particular research topics.

Stratification of age at the onset of the visual impairment: respondents who experienced visual impairment at a younger age tended favor research themes regarding revalidation and reintegration schemes and employment opportunities. Also, they preferred research regarding genetic factors influencing the onset and progression of their ophthalmological disorder. People with a latter age of onset sought research retaining social support structures for people who became visually impaired at a later age, and they were interested in the influence of an early diagnosis on the progression of the disease. This latter topic was also highly prioritized by sighted people who had an increased risk of developing visual impairments.

Stratification on gender, severity of the visual impairment, membership of a patient organization, and age: no significant differences were found regarding stratification of the results on these characteristics. According to the aging patient population, a relatively older age group filled out the survey.

are not easily disrupted and patient communities are not readily integrated.

It's an ideology: the eye. In its entirety. [...] However, corneal disorders are a very specific thing. So to tell people with a corneal disorder: you need to be involved with the eye, the entire eye, that is forced. It's the same for people with eye-prosthetics, also a small patient organization. And it counts for us too. People want information about their own disorder. [...]. We have our doubts about cooperating [...]. [patient representative during post-hoc reflection interview]

4.2 Management of boundaries

Several patient representatives, of whom some were part of the project team, considered the process of setting a research agenda jointly as an opportunity to manage the boundaries between the patient communities. The facilitators employed two main ways to manage the boundaries strategically. These means primarily focused on resolving substantive issues through responsive co-design and joint reflection on research outcomes, as well as neutral facilitation.

First, the facilitators aimed to prevent conflicts between patient groups by facilitating ample opportunities of reconciliation, in order to maintain support for the process and outcomes of the research agenda. For example, the composition of the advisory board was adjusted in the exploration phase; in addition to representatives of

ophthalmologists, ophthalmological researchers and the two major revalidation institutes in the Netherlands, and the optometrists' association were also invited to the advisory board to accommodate concerns of one member of the project team. Similarly, following the FGDs in the consultation phase, feedback meetings were organized with patient representatives to verify the results. After difficulties to determine the composition of these groups (see Section 4.1), patient representatives in the feedback meetings considered the outcomes of the FGDs recognizable. This contributed to the support of this step in the research agenda-setting process. In the priority-setting phase, the facilitators answered all questions regarding the recruitment strategies and validity of the questionnaire from a substantive perspective: doubts were refuted with scientific theory and research findings. This was a time-consuming process, which delayed the launch of the questionnaire by weeks; yet, the facilitators considered such discussions essential to maintain the support of all members of the project team.

Second, by presenting themselves as neutral and independent throughout the process, the facilitators aimed at depoliticizing the study. Although, at times, patient representatives attempted to draw the facilitators into the political force field, the facilitators considered creating and nurturing trust between all the stakeholders and themselves an important aspect of guiding the study. The facilitators made an effort to achieve this by answering all questions and combatting all concerns on substantive grounds, instead of drawing on their academic authority or addressing patient representatives' voice tones, which was sometimes agitated. The members of the project team were generally open toward the facilitators regarding their interests and stakes. Since the financial support for the project was issued through the largest patient organization, at times, this caused distrust amongst the members of the project team. For instance, there were fierce differences of opinion concerning the external communication of the study; the patient representatives did not agree on what the collaboration had to be called.

4.3 Disruption of boundaries

In the challenging sociocultural landscape in which this study took place, one can consider the initiative to set a joint research agenda as a deliberate attempt to disrupt the boundaries between the different patient communities. The initiating patient organization suggested establishing a project team in which patient representatives from the smaller patient organization would also have a seat. This could be considered an indication of good faith. At the same time, not all members of the project team supported this gesture.

During the research agenda-setting process, the facilitators encouraged this joining of forces, whilst acknowledging the potential difficulties to maintain support amongst all involved stakeholders. From this position, three out of eight FGDs in the consultation phase had an open character. These included an FGD organized for people with a range of severity of low vision, another intended for blind people resulting from any etiology, and one that consisted of parents of children with any type of visual impairment. Participants for all FGDs were recruited through various communication channels of the participating patient organizations and were invited to the session they would feel most at home. During the mixed FGDs, participants with diverging medical conditions met and shared experiences. In general, they recognized and supported each other's everyday problems and concerns to the same extent as the participants in the more specified FGDs. The research topics they contributed to the research agenda were sometimes diverging when

concerning medically oriented topics about the etiology or treatment of their condition. For example, in the FGD for blind people, a participant suggested research to repair the optical nerve. However, to people who are born blind, this is not directly a beneficial solution as their visual cortex is underdeveloped. Research topics of a socio-psychological nature were commonly recognizable for all participants. Participants to the mixed FGDs treated other attendees with respect, even when they did not share certain experiences. The atmosphere during the mixed FGDs was no less pleasant than during the specific FGDs. This indicates that patients with different ophthalmological diseases were less concerned with the heterogeneity of their conditions than the dividedness of the patient representatives would suggest.

As the fragmented interests among patient representatives were a concern to many involved stakeholders, several people attending the dialogue meeting emphasized the need for joint action and a shared organizational responsibility to implement the research agenda. One participant mentioned the possibility and value of emphasizing the public interest of some of the socio-technological issues on the agenda, thereby questioning the boundaries between people with a visual impairment and the general public.

I would greatly appreciate it if we can clarify that these measures are not just helpful for people with a visual impairment, but for everybody. It gets accepted more easily and you'll get funding for it. [...] It's the public interest of accessibility for the visually impaired, I think that's good for everyone. [ophthalmological researcher during dialogue meeting]

The first step toward such a joint action was initiated during the dialogue meeting, as the topics on the research agenda were discussed in subsets of participants. These groups consisted of ophthalmological researchers, policy makers, and patient representatives deliberately brought together to encourage collaborations. The attendees were positive about the value of these sessions, but emphasized that more in-depth analysis of the research topics was needed to establish sustainable collaborations.

One of the involved patient organizations took the initiative to execute this recommendation. In a subsequent project, patient representatives with varying ophthalmological disorders, experts from the field of ophthalmology and rehabilitation, and other relevant healthcare professionals were brought together to investigate the state-of-the-art of scientific research regarding the topics on the research agenda and provide routes for implementation of the research. Healthcare professionals and ophthalmology researchers welcomed this initiative with enthusiasm. Representatives from other relevant patient organizations were invited to join this follow-up project, but refrained, as they did not believe the outcomes would benefit them sufficiently.

5. Discussion

The objective of this article was to evaluate the responsiveness of the Dialogue Model in setting a joint research agenda for and with a divided patient community from the perspective of boundary work, in order to stimulate successful agenda-setting processes in health research policy.

Analyzing the research agenda-setting process, we observed that the perceived heterogeneity of involved patient representatives regarding the nature of their visual impairment and its accompanying disabilities was one of the causes of their dividedness. Already at the very start of the research agenda-setting process, some patient

representatives emphasized the heterogeneity of their condition to fuel the dividedness between the patient communities. Turning to the literature on organizational conflict, this ‘social category diversity’ can be viewed as a source of affective (interpersonal) conflict (Jehn et al. 1999). Rahim (2002: 210) defines such affective conflict as ‘inconsistency in interpersonal relationships, which occurs when organizational members become aware that their feelings and emotions regarding some of the issues are incompatible’. The analysis of patients’ problems and concerns and the prioritization of their research needs in the research agenda-setting process indicated that patient representatives overestimated the differences between the patient communities. The dividedness did not become apparent during the FDGs in which individual patients were consulted. In the two FDGs with a heterogenic composition—patients with different visual impairments—there was even much shared recognition in experienced barriers. Patients were able to convert their individual ‘I-voice’ into a ‘we-voice’ (Pittens 2013). However, the patient representatives did not fully endorse this shared recognition between the several sub-groups of patients, meaning that the boundaries arising from this affective conflict did not seem to diminish throughout the study. This marks how deeply rooted the divide was, and how some stakeholders seemed to lack the intrinsic motivation to disrupt the boundaries. As such, affective conflict originating from a perceived heterogeneity appears to be a root of the boundaries created and reinforced during the study.

We further argue that one of the origins of this dividedness was the perceived difference in power between the involved patient organizations; representatives from the smaller patient organization seemed to distrust the intentions of the larger patient organization. Drawing on Pierre Bourdieu’s idea of science as a site of ‘competitive struggle’, Vuolanto (2015) describes how the exertion of power can be an important factor in boundary work between and within communities. As Vuolanto (2015) reports, novel power dynamics within a community are revealed in times of controversy. Our case study similarly revealed power dynamics taking place during the agenda-setting process. The patient organizations had, apart from differences regarding substantial matters (i.e. which patient community they aimed to represent), a reason to compete with each other on the basis of power (e.g. over the size of the patient community they represented and the funds they acquired). This power struggle was, however, not reflected in the consultation of individual patients in the FDGs.

5.1 Strategies employed in this research agenda-setting process

This study manifested that the research agenda-setting process was, at times, the arena of the reinforcement of existing boundaries between the patient communities. The complicated socio-cultural landscape was discussed prior to the commencement of the research agenda-setting activities. The facilitators employed several strategies to guide the process based on this knowledge. On the one hand, deliberate strategies were applied to acknowledge and cooperate with existing boundaries to a certain extent, with the explicit goal to maintain broad support for the research agenda-setting process amongst the involved stakeholders. Following the theory of Rahim (2002) on organizational conflict, this can be regarded as an effective conflict management strategy as the interventions were aimed to manage intra-group substantive conflict. According to the definition provided by Rahim (2002: 210), ‘substantive conflict occurs when

two or more organizational members disagree on their task or content issues. [...] A moderate level of substantive conflict is beneficial as it stimulates discussion and debate, which help groups to attain higher level of performance’.

On the other hand, in the shared pursuing of the research agenda, the disruption of existing boundaries was one of the aims of the study. Although this goal was endorsed by all stakeholders at the start of the process, deducing from the retrospective interviews we can conclude that this endeavor was only partly successful. The follow-up implementation project to stimulate the translation of the agenda into health research policy illustrates a dichotomy. While this implementation project was enthusiastically endorsed by professional ophthalmological healthcare and research organizations, one patient organization initiated and executed this project without other relevant organizations joining. This illustrates that the dividedness between the patient communities was rooted too deeply. In concurrence with Rahim (2002), adverse affective conflict continued to exist, and we can conclude that there was too little support for a continued collaboration.

5.2 An underlying presumption of the dialogue model: Conflicts can be resolved by reflection on content matters

As described earlier, the Dialogue Model is based on the assumption that after specifying and explicating stakeholders’ perspectives, integration of the perspectives can take place (Pittens 2013). Thereby, the model is grounded in the notion of partnership and dialogue. However, this evaluation reveals that this underlying and implicit presumption—that the clarification of, and reflection on, differences and similarities is sufficient to result in better understanding and more collaboration between divided communities—does not sufficiently address certain forms of dividedness. By explicating and discussing the overlap and differences in research needs between the patient communities in both the qualitative and quantitative data, it was assumed affective conflicts could be solved. With the point of departure of the Dialogue Model not being rivalry, as is evident from its guiding principles (Broerse et al. 2010a), this study reveals that the employed strategies insufficiently address the adverse effects of affective conflict, resulting from social category diversity, on group loyalty and group performance (Amason 1996; Jehn et al. 1999).

5.3 Recommendations to set a research agenda more effectively in a divided patient community

Our analysis has explicated a condition of the Dialogue Model, which has thus far remained implicit: that stakeholders are willing and able to bridge the boundaries within a patient community by understanding and reflecting on similarities and differences in perspectives. This statement allows us to ask the following two questions: (1) Is this condition absolute? and (2) What additional strategies could have been used to involve a divided patient community more effectively in setting a shared research agenda?

Regarding the first question, this case study raises questions regarding the feasibility of a joint research agenda in a divided patient community. As Pittens et al. (2014) emphasize the need for good relations between stakeholders for the successful implementation of agendas into health research policy, this case study illustrates the importance of this criterion as a means to set a joint research agenda in the first place. Mapping the social landscape—the

characteristics of the patient community, their mutual relationships and expectations—in which the research agenda will operate, should be an important aim in the exploration phase of the Dialogue Model (Abma and Broerse 2010).

However, this condition does not imperatively imply that dividedness between patient communities is incommensurable. In the case of a challenging social landscape, there is a need to enrich the cognitive-oriented Dialogue Model with approaches enhancing the reflexivity of the involved stakeholders with affective conflicts. Participants need to show commitment to listen to each other and adjust their presumptions accordingly, regarding both substantive and affective conflicts. Strategies encouraging such reflexivity in multi-stakeholder situations have been described in the research methodology reflexive monitoring in action (RMA) (van Mierlo et al. 2010). In RMA, an independent facilitator helps stakeholders to shape the change process themselves, adapt to the new reality by monitoring the situation, and initiate and support interventions when needed. As such, stakeholders become aware of the (substantial and affective) challenges that are influencing the change process and how to overcome them. Similarly, scholars in deliberative dialogue emphasize the importance of a knowledgeable, yet independent facilitator to manage conflict. Cuppen (2012) warns that conflicts risk intertwining at the cognitive and affective level. To avoid detrimental affective conflicts, facilitators should:

contribute to an atmosphere in which participants [feel] open to explore divergent, even conflicting, points of view. [...] Importantly, [the facilitators should] be able to bridge disparate perspectives or ideas when necessary; for example to serve as a mediator between stakeholders with different perspectives or conflicting ideas. (Cuppen 2012: 39)

Facilitators who are active listeners, empathetic and who present themselves as an authority on the process (rather than content) may contribute to a conversational context which help understand the socially shared meanings emerging from group discussions (Kupper et al. 2007).

In addition to strategies enhancing stakeholders' reflexivity on the affective conflict, and facilitators providing an open and conducive atmosphere, development of a shared vision and commitment is needed to overcome boundaries between patient communities. In this regard, Hall's (2005) account on interprofessional teamwork within healthcare settings is insightful as it describes the strain between healthcare professionals as boundary work and provides avenues for more successful collaborative practice. Following Petrie (1976), Hall (2005: 194) emphasizes that

a clear and recognizable idea or goal must serve as the focus for team members in order for teamwork to succeed. [Such] 'idea dominance' allows each member to shift from his/her specific professional focus to one requiring understanding of another's observations and interpretations.

As we have seen, in our case study, it was attempted to accomplish a shared vision (namely, a shared research agenda) using predominantly strategies aimed at resolving substantial conflict. However, to achieve 'idea dominance' more successfully, McCallin (2001) claims that aside from cognitive strategies, strengthening personal connections is essential, for example, by organizing multi-day residential team-building workshops. Also, insights into mediation may be insightful in this regard. However, it is beyond the scope of this article

to go into full depth in the extensive literature regarding this topic (Lewicki et al. 2016).

5.4 Methodological considerations

This study aimed at reflexively and retrospectively evaluating the process to set a joint research agenda for a divided patient community using a boundary work perspective. We triangulated our data using interviews, participant observation, document analysis, and self-reflection. This allowed the authors to examine the process and outcome of this case study in depth, yielding insights in the sometimes unforeseen effects of the employed strategies. Triangulation was particularly important to reduce researchers' biases in this case study, as the research agenda-setting process was regularly accompanied by passionate discussions via email or telephone, which required some professional distance to evaluate the process.

Analyzing the data from a boundary work perspective proved useful, as it conceptually clarified the causes and consequences of stakeholders' actions during the research agenda-setting activities and enriched our understanding of the presumptions or conditions underlying the Dialogue Model. This clarification subsequently provided several avenues to explore regarding professional conflict management.

As this research is to be considered a case study, it is challenging to separate context-bound contingencies from more general factors affecting divided patient communities. For example, issues brought up by a few patient representatives, which contributed to a strong feeling of dividedness, could be related to recent developments between patient organizations regarding their position. They could therefore be considered primarily context-bound contingencies, but that was not confirmed. To the best of our knowledge, similar analyses in patient involvement have not been described in the literature, making comparisons difficult. In addition, due to its retrospective nature, this analysis could not inform the employed strategies during the research agenda-setting activities. Although the authors discussed the progress of the study and its accompanying challenges regularly, decisions were based on the available knowledge and expertise regarding the Dialogue Model. The potential efficacy of strategies enhancing reflexivity and reducing affective conflict has therefore not been tested.

6. Conclusion

We conclude that the Dialogue Model has been recurrently validated to set agendas for health research policy from the patients' perspectives. However, our case study shows it provided insufficiently effective directions to guide this process in a divided patient community and delimited the successful translation into policy. The authors balanced between acknowledging and reinforcing existing boundaries to maintain support for the research agenda-setting process and managing or disrupting them to produce a joint end product. It can be concluded retrospectively that managing moderate substantive conflict through understanding of, and reflection on content matters was the prime focus of the facilitators. The research agenda is endorsed by some of the participating patient organizations and the implementation project explicitly addresses the joining of forces in the ophthalmological research field. The research agenda contributed to the dialogue of the patient organizations with societal stakeholders but did not result in a shared agenda and ultimately a health research policy. However, to involve a divided patient

community more effectively in agenda setting in health research policy, we argue that management and disruption of boundaries is necessary by actively enhancing stakeholders' reflexivity regarding affective conflict, acknowledging the importance of strengthening personal connections, and containing the adverse effects of affective conflict.

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