VALUES AND CRITERIA OF PEOPLE WITH A CHRONIC ILLNESS OR DISABILITY

Strengthening the voice of their representatives in the health debate and the decision making process.

Truus Teunissen
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This thesis is dedicated to my mother † who has always stimulated me to fight against injustice and inequality by letting my voice be heard.

Dit proefschrift is opgedragen aan mijn moeder † die mij altijd gestimuleerd heeft om tegen onrecht en ongelijkheid te strijden door mijn stem te laten horen.
COLOFON

-About the cover
Oenothera - English: Evening Primrose
This plant will grow virtually anywhere, even on very poor soil along the road. It is unbelievably sturdy yet tall and exuberantly blossoming the entire summer and fall seasons with its bright lemon-yellow flowers. My garden back home is full of them. In Dutch it is called “Teunisbloem”, like my family name “Teunissen”.

-Intermezzo pages
Preceding each chapter there is an intermezzo page. The front of these successive intermezzo pages shows a different sculpture for each chapter. All sculptures were made by Truus Teunissen, the author. They highlight the struggle between strength and vulnerability in her own life with illness and disability. The sculptures are made of a variety of materials like bronze, cherry wood, cast concrete and various kinds of stone: marl, steatite, Belgian freestone, serpentine. They were not given a name or title to allow everyone to freely observe and interpret their meaning. The rear of these intermezzo pages progressively shows the 6 criteria from the perspective of people with an illness or disability. The last intermezzo page shows pictures taken at some of the many meetings held among these people and among professionals during the research being presented in this thesis.

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ACADEMISCH PROEFSCHRIFT

ter verkrijging van de graad Doctor aan de Vrije Universiteit Amsterdam, op gezag van de rector magnificus prof.dr. F.A. van der Duyn Schouten, in het openbaar te verdedigen ten overstaan van de promotiecommissie van de Faculteit der Geneeskunde op dinsdag 17 juni 2014 om 11.45 uur in de aula van de universiteit, De Boelelaan 1105

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PREFACE

“A little while ago my husband and I were visiting friends in France for a couple of days. During our stay we were looking around town and enjoying its pleasantries. I noticed that there were no people around with an obvious and visible disability e.g. using a wheel chair, a red-white cane or a guide dog. There were no special measures to improve access of streets, side-walks and shop entrances. In other words, even I had to watch my own steps to avoid stumbling over something. Of course it was rural, romantic and typical ‘a la campagne’, and all you could want when visiting France.

When I asked our friends where all the people with disabilities would be, they were not sure “. . . in a care institution perhaps, or at home because they were well taken care of and would not need to go out in the streets . . .” (!). I then wondered, if all the people with a visible disability are at home ‘well taken care of’, how then would people with a chronic illness - not visible but with impairments nonetheless - be dealt with ? And how would they be full citizens participating in society ? As this thought struck me, I tripped and fell because the sidewalk suddenly stopped and was followed by a drop of 10 cm.” (personal story from Truus Teunissen, the author, 2010)

At first glance, the possibilities to move about for people with illness or disability seem to better taken care of in comparison with Dutch cities. Indeed, the physical space often accommodates for this. Also social, mental and societal impairments often play a role with illness and disabilities though. Is freely moving about in physical space enough, or is it just a first step for people with an illness or disability in the direction of full citizenship in society ? Just like many others I was raised to believe that taking well care of oneself prevents illness and misery, brushing your teeth avoids the dentist and a scarf avoids catching a cold. Never ever (!) go outdoors with your hair still wet. And if you do get ill, eat oranges and take your time to get better. It sounded logical and reasonable back then and it still does now. You are either healthy or ill, you can either walk or you cannot walk. Meanwhile I have found out this logic is wrong.

As I acquired several chronical diseases over the years myself I started looking for the reasons why the chronic aspect of it appears to be so elusive and unreal. As time went on my search focused on what it takes for people with an illness or disability to express and voice their concerns and needs.

Also that their voice is taken seriously and in full when it comes to the health dialogue and to decision making in practice. Window dressing and placation aren’t good enough. True involvement is what it is all about.

People with an illness or disability are a new and growing group with their own voice, issues and concerns. They move in between ill and healthy conditions. They are both strong and vulnerable at the same time. These are the new partners in the health research and health care dialogue. This stands, solid as a rock, at least for myself.
Truus Teunissen
Material: cast concrete
1-GENERAL INTRODUCTION

1.1 BACKGROUND
The number of people acquiring another chronic illness on top of the chronic illness or disability they already have, is growing. Extended life expectancy increases the number of elderly people with multiple chronic conditions (Lucht & Polder, 2010). These developments and the widening range of medical technical treatment possibilities lead to a growing number of people with a chronic illness or disability, more susceptibility to co- and multi-morbidity and age related chronic illness (WHO, 2011). The demand for health care is equally growing.

Co- and multi morbidity complicate health care since different chronic illnesses and disabilities require different treatment and different care providers. This may lead to conflicting advice and treatment and in some cases even unsafe situations (RIVM, 2013). The people concerned not only experience medical problems associated with their multiple diseases or disabilities, they also have social and societal problems (Rijken et al, 2012). Their health is more vulnerable due to secondary chronic illnesses such as medication side effects, decubitus or infections of the urinary tract, but also due to low income and social exclusion. There are indications that people with a disability experience more obstacles when accessing the health care and revalidation facilities they need (WHO, 2011). This implies that chronic illness and disability are not merely a health problem. The interaction between a persons’ physical limitations and the persons’ societal environment makes this a complex phenomenon. The WHO refers to this: ‘. . . the term disability is a general term which includes limitations and disturbances in activities and societal participation’.

All this changes our mind set when looking at chronic illness, disability and health. The current WHO definition states that “health is a condition of full physical, mental and social well-being and not merely the absence of chronic illness or disability”. This definition was created shortly after the Second World War and at the time was considered innovative and accurate. Confidence in science and idealism reigned. During that time, infectious chronic illnesses were very important and the discovery of antibiotics supported the hope that chronic illness could be eradicated all together (WHO, 2011).

Today, other ideas about a better definition of health are being explored to underline the flexibility and resilience of people. One of the latest definitions of health is: “Health is the ability to adapt and be in control in view of the physical, emotional and social challenges of life” (Huber et al., 2011). This is based on the idea that people, living with an chronic illness or disability, learn to cope with it and still lead a fairly healthy life. The goal is no longer to survive but merely to live one’s life. People with chronic illness or disability are already for quite a while convinced that the focal point ought to be shifted to ‘what you can’, to self-management and being in control next to their vulnerability. Although the latter often gets priority, this shift toward a capabilities approach makes life much more pleasant than when only looking at chronic illness and disability.
Current scientific research supports this view. For example in health care people with a chronic illness or disability are more and more placed at the centre, get more control over and more responsibility for the care they receive (Ursun et al, 2011). The currently dominant discourse is based on the concept of the empowered person with chronic illness or disability, being in control, being supported by an informal network, being self-sufficient and independent, making informed choices and taking responsibility for the consequences of those choices. This discourse is founded on several assumptions. One of them is that people with a chronic illness or disability are actually able to be in control over the main part of the care provisions they receive, referred to as “the patient in the drivers’ seat” (Gerads, 2010). One may wonder whether ideals fit with reality; a considerable amount of people cannot live up to this assumption that they are ‘in control’.

Research has demonstrated that the position of people with chronic illness or disability in health care does not concur with the consumer model since decision-making in medical care practice is more complicated than assumed in the dominant discourse. People with a chronic illness or disability also experience vulnerabilities and dependencies (Rijken et al, 2012). They wish to be regarded as a whole person rather than a mere chronic illness or disability case.

1.2 INVOLVEMENT of people with a chronic illness or disability

The last years, groups of people with a chronic illness or disability emerged as a new party in health discourse, health care and health research. Active involvement occurs in a wide variety of contexts by a great amount of them and their organizations. Many new involvement roles have been developed, among them the research partner (Schipper, 2012; de Wit, 2013). This thesis focuses on the role of the representatives of people with a chronic illness or disability in involvement practices.

The term involvement can be looked at in a broad and narrow sense. The broad sense addresses the participation in society in all its manifestations and the more passive ways of engagement like keeping oneself informed about the latest developments and using facilities like a library (RMO, 2008; Bijl, 2011). The narrow sense is about actively taking part in a specific activity (Levasseur et al., 2010). This thesis deals with the narrow involvement concept where people with an chronic illness or disability actively deal with processes in health research and quality of health care.

Involvement illness: Fundamental Right or Obligation?

The last years, involvement of people with an chronic illness or disability has been encouraged by the Dutch government, but in the near or more distant future involvement activities of people with chronic illness or disability can be affected by budget cuts or even a cease of funding all together. Economic developments may lead to new and different political ideas at the government. Such changes can have a significant impact on the concepts of citizens’ own responsibility, own strength, self-sufficiency and social involvement. Active citizens involvement is increasingly being called for and since recently,
formalized into the Dutch Social Support Act (Dutch: Wet Maatschappelijke Ondersteuning). Various national laws and directives on international levels must ensure and guarantee citizens involvement including that of people with a chronic illness or disability. This also applies to their representatives and advocates dealing with health policy, health research and health care practice.

The European Commission not only declared the right on information about one’s own medical condition, having a say in health care is also regarded as a fundamental right. Governments must enable involvement as an integral part of care, create proper pre-conditions, legislation, and facilitate and safeguard cooperation (EC, 2000). Citizens are expected to embrace these concepts. There is a forceful emotional calling by governments to be involved. Citizens feel governments want them to want it: being involved and helpful. The Dutch government stimulates broad participation of its citizens in society. The terms ‘own strength’ and ‘responsibility’ are used to seduce citizens to control their own lives and enforces them to be caring for one another (Verhoeven et al., 2013). Citizens in general and people with a chronic illness or disability are confronted with these ideas and concepts on responsibility, control and self-sufficiency. This is being referred to as ‘good patient ship’ (Knibbe &Verkerk, 2012). Although currently this consists merely of a set of stories and notes about what being a good patient ought to mean, professionals often indirectly use it as a frame of reference.

In the Netherlands, the health care system is based on solidarity. It is a system for all and by all. The solidarity requires reciprocity of all parties involved: citizens, care professionals, health insurers and care institutions (RVZ, 2013). It assumes that citizens with a chronic illness or disability are aware of their responsibility as a co-producer in the system and that the systems’ continuity is at stake. Ideally, the collective health insurance policy most people in the Netherlands use becomes an instrument or vehicle to move health forward. The question is whether the collective voice of people with a chronic illness or disability is audible and heard.

**Areas of Involvement illness**

The voice of people with a chronic illness or disability is important in the relationship between the individual patient and general practitioner (Guadagnoli et al, 1998;Bensing, 2000; Gattellari et al, 2002). This thesis focuses on the collective voice of people with a chronic illness or disability in health research, health policy and quality in institutions and governmental organisations (Leys et al, 2007) (WHO, 2011).

People with an chronic illness or disability are directly involved in health care and health research. Giving them a voice in decision-making is therefore ideologically seen as a right thing to do. Involvement enhances the empowerment of people with chronic illness or disability. Engagement can, just like the other parties in the health domain, reinforce their sense of equality. It can contribute to a culture change in health care towards a more equal relation between care professionals and people with a chronic illness or disability (Elberse et al., 2012). Involvement can create a new active role in society, building up social capital (Caron-Flinterman, 2005; Trappenburg, 2008). Involvement of the third party has the potential to contribute to health of citizens and thereby to their self-sufficiency. Patient
Involvement in decision-making is being promoted on all levels, both in the Netherlands and internationally (Bovenkamp et al., 2008).

**Involvement in Quality of Care**

Health care could be improved by using the contributions of people with a chronic illness or disability. Their life experiences provide a basis for that. These personal experiences are extended in the case of patient representatives of patient groups and patient organisations. Representatives join the experiences of their fellow people and can therefore exceed the individual experience level and bring the collective point of view to the table in decision making.

Involvement of people with a chronic illness or disability can increase commitment among members of their group and thereby increase the chances on successful implementation of the decisions taken. New guidelines, developed together with them and also laid out in a lay version, could be distributed among them. These patients in turn can then confront their care provider with the new guideline and in doing so speed up the implementation of the guideline (Veenendaal et al, 2004).

Finally, there is the increase of efficiency and efficacy of the health system to consider. Involvement is required to achieve the political ambitions of the government in its attempts to curtail the health budget by introduction of changes in the health system. The prime importance is to create a counter force coming from people in need of care (i.e. health consumers) against the health care providers and insurance companies, and making care more demand oriented (Bovenkamp et al, 2008).

Involvement may help to generate a wider spread of ideas and experiences to increase the chances for new, original and innovative solutions to problems. An example of this effect is the 2007 patient consultation carried out by the Health Research Council (RGO) to obtain advice for its Medical Biotechnology agenda (RGO, 2007).

Involvement can also be used to achieve other parties’ goals. Here, involvement is not being used for its originally intended purpose. In such cases it is reduced to pseudo-involvement or placation, minimising the impact of ideas and experience expressed by a group (Harrison & Mort, 1998) (Bovenkamp et al, 2008) (Broerse et al, 2010).

**Involvement in Health Research**

In scientific medical research, involvement has been the subject of scientists and (para) medic professionals for a long time. Research projects are often initiated from a scientific perspective by researchers (bottom-up) or by institutes or foundations. Occasionally, a patient organisation funds a research project (top-down). The choice of research themes on the agenda and the appraisal and priority setting of themes is done predominantly from a scientific point of view. Scientific quality and relevance are traditionally the decisive criteria when appraising and evaluating health research. Recently though, societal relevance originating from the perspective of people with a chronic illness or disability emerged as a evaluation criterion.
Representation in Involvement Practices

Representatives act on behalf of their peer group, the group of people with chronic illness or disability they belong to, and express widely supported opinions of their group in the process or debate at hand. In the Netherlands and many other European countries, people with a chronic illness or disability are united in organisations that look after their common interests. In current practices, representatives often sit at the table to discuss issues and concerns of their peer groups in relation to health research and health care guidelines. These representatives usually are patients or clients. They have individual experiential knowledge about living with a chronic illness or disability. In their role as a representative they share experiential knowledge of their peer group. Representatives usually experienced the process of acquiring a chronic illness or getting a disability themselves, being confronted with its chronic character and having to live with a chronic illness. This illness or disability process may evoke feelings of anger, grief, denial and loss. Only after processing these feelings and experience these initial phases of the illness process, a patient or client can or wants to act as a representative for a group.

Representatives of people with a chronic illness or disability may assume different roles and tasks when taking part in an involvement process in research and health care, like when they appraise research proposals or projects to develop guidelines. They can provide information about issues experienced by their patient or client group in their life with chronic illness or disability. Professionals may either use or not use this. Representatives can advise professionals. Representatives may even become partners and cooperate with professionals. Below we present a next example from an appraisal practice. The example illustrates that, although it states that involvement is useful, necessary and sometimes obligatory, legislation is not sufficient to realize it in an effective and practicable way for all parties.

“As a lung patient representative, I was asked by a governmental research funding institution to appraise a series of research proposals from the patients’ perspective. I felt proud that we, the patients, were finally asked for our opinion and we could participate in the process. A few days later the doorbell rang and an impressive package was delivered: the research proposals . . . There were some forty of them, each at least twenty pages in size. In the weekend that followed I sat down to have a look at them. After two hours I finished thoroughly reading the first one. The researchers described a fantastic and beautiful research proposal. What could I possibly add to that from a patients’ perspective? I wondered: What is the meaning of the patients’ perspective here anyway? Reading a bit better once again might help, I reasoned. By noon I had read two proposals and had written down three remarks about what the patients’ perspective ought to be on a sheet of paper. It was frustrating to see so little result an I felt more and more uncertain. If these few remarks were the only thing to put forward in the discussion between professionals exchanging a stream of arguments and opinions I would clearly blunder . . . I felt even more uncertain. So, deciding to spend the afternoon on it as well, I felt I had to perform well as a patients’ representative taking part in the discussions in a
committee with professionals. At the end of the afternoon I had reviewed six proposals, but the number of remarks was still very small. Again I was puzzled: what is the patients’ perspective, where is it in the proposal, and how on earth could I ever finalize so many research proposals in a single weekend? Each proposal was so well described and so positively formulated . . . HELP . . . !” (Truus Teunissen, the author, 2009)

This example clearly illustrates that being involved as a representative is in itself not enough to be able to bring in the voice and perspectives of patients. Several conditions are needed for meaningful involvement. In the example above, Truus, the first author and initiator of the study, lacked time to review the proposals, but she also felt she lacked guidance on how to review the proposals from a patients’ perspective and she missed input from fellow patients. She wondered what really mattered to patients and which issues she as a representative could bring in in the review process. Also, she wondered how to act in the committee, and how to be taken seriously.

In the literature conditions regarding the process of involvement are described including required skills of people with a chronic illness or disability (Oliver et al, 2004), the way information is gathered among the represented diverse and sometimes vulnerable patient group (Nilsen et al, 2006), collaboration with patient research partners (Schipper, 2012), inclusion and exclusion strategies (Elberse et al, 2012) and situations that foster partnership and dialogue with other stakeholders (Baur et al, 2010). Little is known about the issues and criteria that patient representatives use in involvement practices. The importance of patient perspectives and their issues is widely acknowledged. These issues determine how they lead their life and what makes them choose, appraise and decide when it comes to quality of provided care and research projects (Nierse & Abma, 2010; Caron-Flinterman et al, 2005). Neither the groups of people with chronic illness or disability themselves, nor the care providers, nor the health researchers, nor the policy makers have a good understanding of what these issues might be though. There are studies that pay attention to existential and moral issues and values of people with a chronic illness or disability (Schipper, 2012; Visse, 2012), but there is no systematic insight in what matters to people with a chronic illness or disability as a collective. In other words: there is no inventory of issues that matter to groups of people with a chronic illness or disability and how these are and can be used by representatives in involvement practices to ensure that their perspective is articulated.

1.3 MAIN RESEARCH QUESTION

In order to investigate what constitutes experiential knowledge of people with a chronic illness or a disability, and how their issues are and can be used in involvement practices by representatives three aspects need to be addressed.

First it is important to investigate what matters to the people, and what are shared issues of concern. Second, it is crucial to investigate what criteria representatives use to appraise care and research from a patient perspective. Third in which way representatives can put
these matters on the table in the interaction with professionals. All this does not relate to the individual patient-caregiver interaction nor to individual experiential knowledge but to the widely shared experiential experiences and opinions of people with a chronic illness or disability on a collective level. This leads to the main research question for this thesis:

*What constitutes the body of experiential knowledge of people with a chronic illness or disability in the context of appraising quality of care and research?*

The sub research questions are:

1. *What matters to people with a chronic illness or disability concerning research and quality of care?*
2. *According to which criteria do representatives of people with a chronic illness or disability appraise research and quality of care?*
3. *How do representatives of people with a chronic illness or disability and professionals interact and relate to each other during involvement activities?*

### 1.4 THEORETICAL FRAMEWORK

The theoretical framework used in this thesis consists of two concepts: experiential knowledge and partnership.

**Experiential knowledge**

Involvement of people with a chronic illness or disability is largely based on their experiential knowledge. The concept behind experiential knowledge is that ideas, experience and knowledge, coming from the environment where people lead their life, can contribute to better research and better quality of care. Involvement in for instance guideline development is justified because issues from this personal life-world environment are very different from issues important in the scientific and professional environment. Additionally the concept assumes that interaction between the two environments is both possible and necessary.

There is no clear and accurate description of what experiential knowledge might contain. The relation between experiential knowledge and expertise is subject of scientific research and debate (Haaster et al, 2005; Karbouniaris et al, 2009).

Scientific research as a process is based on ‘evidence based’ starting points and criteria. The term experience hardly plays a role in traditional scientific and knowledge theories. The term ‘experience based’ as compared to ‘evidence based’ is not often used. Certainly there is, as yet, no equality when comparing scientific knowledge and experiential knowledge. In spite of this, several methods were developed to test experiential knowledge. These methods use the following criteria (Depraz et al, 2003; Haaster, 2007):

- Practical relevance and applicability
- Recognition and acknowledgement
- Reflection
- Mutuality
Ideas and insights on health and disease are often worded in terms like ‘lay conviction’ and ‘expert knowledge’ (Lupton, 1994). Knowledge (also referred to as *wisdom*) is not restricted to wise people, it is also considered as an integration of cognitive, reflective and affective personality characteristics. Experiential knowledge fits in with this model (Ardelt, 2004; Bury, 1997, 1998).

Increasingly often people with a chronic illness or disability are expected to express their views and take a stance about the condition they are in (Kangas, 2002; Prior, 2003). Three levels can be distinguished: experience, experiential knowledge and experiential expertise (Deth et al, 2012):

- *Experience* is about what happens to people related to their chronic illness or disability. Every experience is unique and has a knowledge aspect and an experience aspect (Erp et al, 2011). Attribution of a meaning to the experience leads to the individual knowing something. This may become transferable knowledge when others could use it.

- *Experiential knowledge* requires individuals to analyse and reflect upon their own experiences. New insights can emerge which could be valid for or useful to others. This type of knowledge arises from experiences in practice.

- While exchanging these experiences someone may find out that others recognize the experiences and whether the insights are working for them too. The common element is a key property of *experiential expert knowledge*. Personal experiences can be connected to that of others. This leads to collective experiential expert knowledge. By increasingly sharing experiential knowledge this starts to spread out and develops itself (Rip, 2005). If done in a systematic way a knowledge domain, often referred to as ‘body of knowledge’ will be established. This is a knowledge area belonging to a scientific domain with clear boundaries, in this case experiential knowledge integrated to experiential expert knowledge. Currently however, the recognition, identification, storing and consulting of experiential knowledge is in its infancy.

The terms ‘experiential knowledge’ and ‘experiential expert knowledge’ are frequently mixed and are defined and used in different ways in practice. People live through unpleasant experiences and process them into their own lifestyle and set of values. Afterwards they may look back and rise above their individual experiences level. The insights obtained may give them the strength to participate in society and support others. It is as yet unclear whether everyone has such capability to participate, nor is it clear what it takes to get people to look at themselves and rise above their individual level.
All this implies that experiential knowledge does not always emerge automatically and sufficiently. Initiatives to obtain and build up expert knowledge and skills in a controlled way are therefore necessary (Simons et al, 2013).

Creating a sufficiently large experiential experts group (capacity development) to ensure continuity of the experiential expert’s voice is important. Building up the skills and knowledge levels of experiential experts about subjects relevant to the entire peer group is a pre-requisite for effective involvement (Involve, 2011). Individual people with a chronic illness or disability may develop themselves in involvement practices (Schipper, 2012).

Where people would be ill or disabled for a rather short time in the past, medical technical developments currently result much more often in a lifelong situation with chronic illness or disability. People with chronic illness or disorder are not always considered as ‘sick’ or even as ‘patients’. Chronic illnesses or disabilities never heal.

A chronic illness or disability can roughly be described by three phases:

* the chronic illness or disability is acquired (diagnosis, care)
* the chronic illness or disability stays (quality of care, quality of life, being in control, self-management) leading to **Experience**
* learning to live with the chronic illness or disability (quality of care, quality of life, being in control, self-management, social participation) leading to **Experiential knowledge and - expert knowledge**.

**Partnership**

The perspective of people with a chronic illness or disability, founded on the broad experience among individuals in the group is the first starting point in this thesis. This implies that they are taken seriously as a source of experience and knowledge and that their knowledge is being translated and used in health related processes (Abma & Broerse, 2010).

In this way, the uncertainties, questions and themes, originating from personal experience become a factor in the control of research, policy and quality of care. The experiences join in with the vision of professionals such as care providers, financial experts, policy makers and health researchers. The experience of people with chronic illness or disability can lead to experience based knowledge and to expert knowledge if this knowledge is being shared and connected to experiences of others in a similar situation. Experiential knowledge is an important source for survival, for the art of life, for problem solving and for questions and support.

In the past, too often experiential knowledge (lay knowledge) about health was labelled by health professionals and policy makers as a set of insignificant, poorly underpinned and subjective ideas. This in spite of the essential importance of understanding their constructs and interpretations in their dialogue with health professionals (Yuill et al, 2012).

A second starting point for involvement of people with a chronic illness or disability is to make them partners in the process of producing knowledge. This means they are actively and equally involved in the design, execution and evaluation of research, quality of
care projects and policy definition. They are also one of the parties in the negotiations between for example the health care providers and the health insurance companies. Arnsteins’ citizens participation ladder (Arnstein, 1969) was the basis for today’s involvement ladder indicating the levels of involvement people with chronic illness or disability may have with increasing influence from having no say whatsoever, via being consulting, to advising and cooperation towards full control, see Figure 1 (Teunissen & Abma, 2010; Baur, 2012).

**Involvement ladder**

![Involvement ladder](image)

**Control**
Organisations of people with a disease or disability are leading and in control of the contents and processes.

**Partnership**
These people have a partnership as equal partners with other parties and take part in the joint decision making process.

**Advising**
These people provide advice (upon request) but do not take part in the decision making.

**Being consulted**
These people are asked for their opinion.

**No involvement**
These people do not ask anything and are not being asked.

Notes:
This involvement ladder design is based on Arnstein’s 1969 citizen participation ladder. The above figure was first published in Dutch in TSG, 2010 by G.J. Teunissen. The illustration copyright resides with the author G.J. Teunissen from 2009.

Figure 1 People with a chronic illness or disability involvement ladder with increasing levels of influence.

With this thesis’ vision in mind, regarding involvement as a dialogue, the partnership or cooperation level would ideally be the way to act for people with a chronic illness or disability. This is a higher level than the usual consultation (they provide information) and
advising (they do not take decisions) and differs from full control (all decision power resides with them). In this thesis, the latter is considered not being beneficial for people with a chronic illness or disability. Full control is expected to lead to resistance, to tensions and to less opportunity for experiential knowledge to contribute to enrichment of professional and scientific knowledge. This is based on two involvement goals: 1) increasing democracy and 2) contributing to the quality of processes and their outcome. Involvement must not be allowed to deteriorate to obligatory walking down a preconceived path to a preconceived outcome (Harrison, 2012). People with a chronic illness or disability need to be regarded by their care providers in their entirety, as full citizens with a broad diversity of available knowledge and skills. Communicating with them via protocol like recipes in a cookbook does not reflect that they are being taken seriously (Entwistle et al., 2012).

Involvement is related to groups that somehow are lagging behind in society. Involvement and emancipation are therefore associated terms (ECAS, 2008). For proper involvement it is not sufficient to know that people are officially free to join in with an activity or not. If in practice a specific group is hardly involved in an important activity, this is a sign of their disempowerment. Ideas about involvement, equality, equal rights, equal opportunities and being heard are closely interrelated (Calman, 1997). It is not enough for a group or an institution to simply state that people with a chronic illness or disability can voice their concerns. Partnership means that the parties in the dialogue might have both differences and agreements in their views at the same time. In the continuing, permanent dialogue both sides mutually develop understanding for each other’s views and new possibilities open up (Widdershoven, 2001).

1.5 RESEARCH DESIGN AND METHODS
In this thesis a responsive evaluation design was followed. Responsive evaluation supports mutual understanding of multiple participants and includes their concerns and issues into the research process itself. This offers the opportunity to participate in gathering issues and addressing values that matter to people while appraising and defining criteria for evaluation of research and quality of care activities.

Responsive evaluation focuses on stakeholders’ perspectives. Its purpose is to facilitate a dialogue between these stakeholders (Stake, 1978; Guba & Lincoln 1989; Abma, 1999, 2005, 2006; Abma & Widdershoven, 2008). The responsive evaluation process begins by initiating a dialogue among people with chronic illness or disability and subsequently a dialogue between them and researchers, care providers and others taking part in e.g. a project. People exchange their perspectives and redefine those while the process is going on. The researcher investigating this process, attempts to grasp common ideas (shared constructions) from different opinions, expectations and perspectives among the participants (Stake, 2004). This is done at first among people with a chronic illness or disability and later also among other stakeholders. Within this responsive design, several qualitative research methods have been used, like interviewing, focus groups, observations and literature review. During a series of interactive meetings (focus groups) over a period
of three years with a rich variety of participants having a chronic illness or disability, proposed criteria were discussed, adjusted, modified, extended and in some cases -in part-deleted.

The actual research was not conducted in the sequence presented in the chapters in this thesis and in the articles published. Some of the parts of this research work were conducted in parallel, leading to insights and results not in all cases being taken along to the next chapter. This is due to the many years that the research lasted and that research in practice is more unruly than theory.

In chapter 2 sub question 1 is being addressed. To this end a narrative analysis is being used to understand the lived experiences, feelings and events of an individual patient with a chronic illness or disability (in this case the 1st author) as she passes through several phases of coping and living with her illness. The auto ethnographical method is used in constructing the narrative of the first author (Ellis & Bochner, 2003). The narrative, the story telling itself is the subject of analysis. In (auto) ethnography a researcher either focuses on a specific theme or searches for characteristic common ideas among members of a social group or culture. In the case described in chapter 2 the theme is the 1st authors’ life with her chronic illness or disability in the context of her family, her work and in more broader terms: the society she lives in. This research is not neutral or objective but inter-subjective, normative ethical and culture and context dependent (Schipper et al., 2010). The narrative approach enabled the execution of an integral story reconstruction and analysis.

In chapter 3 we investigate sub questions 2 and 3. For this we used a descriptive qualitative method to gain understanding of which criteria and values representatives of people with a chronic illness or disability use to appraise or to evaluate. This resulted in a set of criteria. These criteria are now being used in Dutch and European projects by patient representatives. With the qualitative approach, the realms outside traditional medical science areas can be explored to find issues that matter to people with chronic illness or disability as well (Britten, 1995). Content analyses on literature survey, interviews and participant observations were used in this case.

Chapter 4 attempts to find the answer on sub questions 2 and 3 from existing knowledge. The method here is a literature search. International scientific literature was searched and the resulting data were processed via content analysis methods. Primary, secondary and tertiary sources (non-scientific publications, reports and patient information databases) were gathered for this purpose. The study aimed to further develop and validate the content of criteria found from the representatives in practice. This led to a comprehensive list of values and issues.

In chapter 5 further answers to sub questions 2 and 3 are being searched for in practice. As a start a qualitative and responsive research method was used to further develop and validate criteria for use in practice. This resulted in a priority structure of values and criteria. In a responsive research method or approach the research is regarded as a common search strategy with stakeholders to find value and meaning in current practice (Abma & Widdershoven 2006). Issues that matter to people with a chronic illness or disability were gathered and analysed via open coding, clustering and data-synthesis and
combined into a set of criteria. Open coding is a qualitative analysis method consisting of marking non-structured bits and pieces in the data (e.g. text fragments from interviews) and labelling these with a name (a code) to store and analyse them (Boeije, 2005; Strauss et al, 1998;) (Yin, 2003) (Moghaddam, 2006). A combination of thematic analysis, meta-aggregation and data synthesis methods was used for the analysis. A draft set of criteria was proposed to a group of people with chronic illness or disability in a series of focus groups. This was done repeatedly to evaluate and adjust this criteria list design, priority of issues and usability from their perspective.

In chapter 6 the answer to sub question 3 is being addressed in three parts. In chapter 6.1 a longitudinal case study research method was used to investigate a seven-year lung patient involvement process at Lung Foundation Netherlands (LFN). The following aspects were investigated over time:

- Research basis; What basis initiates and chooses themes for scientific research?
- Funding criteria; What appraisal criteria are leading in LFN research?
- Patients say; What is the basis for LFN patients’ say in research?
- Scope; What is the scope of LFN appraisal of patients’ involvement?
- Advocacy; How is LFN patient advocacy in research organized?
- Commitment; What is the level of commitment for LFN patient involvement?
- Context: What contextual and paradigm considerations play a role?

Five significant policy changes at LFN and its activities in society were implemented during this time and included in this study:

- Research cycles up to 2005: Introduction societal research agenda, vision and system
- 2005-2009 research cycles: Update societal agenda
- 2010 research cycle: Patient expert group
- 2011 research cycle: Societal relevance criteria
- 2012 situation planning: Present situation

In chapter 6.2 a qualitative study is used with interviews, focus groups and questionnaires with lung patients to gain insight in the practice of research agenda setting.

In chapter 6.3 we present a qualitative study to reflect on shared responsibilities in involvement practice with people with Chronic Obstructive Pulmonary Disease (COPD) and their care professionals. In this study, in-depth interviews were held to explore the meaning and substance of these shared responsibilities and involvement in practice.

Table 1 shows the methods of this thesis.
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1.6 READING GUIDE

There are different ways to read this thesis. If you prefer a short summary then we advise you to read the preface at the start and the summary at the end. This thesis is written in logical order, although the articles it consists of, are not published in a corresponding time order.

If you like to know how it all began, you can start with reading chapter 2. That chapter describes the struggle of a person with a newly acquired chronic chronic illness. It is presented as a ‘counter story’ because it sheds another light on what matters to people with a chronic illness or disability in practice and why. This chapter offers valuable insight in the strength and vulnerability and in disability and possibility of the first author.

In chapter 3 involvement of people with chronic illness or disability in the Netherlands is subject of research. This study was done first to explore the possibilities for the research in this thesis. Using the findings, it became possible to find the problem areas, to enhance the research questions, and to decide which methods would be most suitable to get the answers.

Chapter 4 presents the first results. An international literature search on what problems are reported originating from people with a chronic illness or a disability about health research and quality of care. This chapter attempts to identify what problems and issues these people have and whether these are actually being reflected in scientific literature. This is an important step towards an inventory of problems and issues in this area.

If you are interested in problems relevant to people with chronic illness or disability in practice, you need to read chapter 5. There is a striking difference between how these people currently appraise research and quality of care and what appear as the most important aspects when further questioned. Surprisingly enough a number of problem areas reported by people with a chronic illness or disability is not reflected in scientific studies. This chapter integrates the findings into a structured appraisal and evaluation method for research proposals and quality of care. This method was further developed together with them into an evaluation instrument and this method is currently being used by patient organisations and health funding organisations in the Netherlands.

Chapter 6 describes several stages of the implementation of lung patient involvement in the Lung Foundation Netherlands (LFN) research funding approval process cycle. The structured appraisal with the evaluation instrument was evaluated in practice with this case study. The 7 years longitudinal case study provides useful details on pitfalls, complications and practical solutions. Insight in the practice of research agenda setting together with lung patients is provided in a second qualitative study, included in the same chapter. A third, also qualitative study reflects on shared responsibilities in involvement practice with Chronic Obstructive Pulmonary Disease, COPD.
Chapter 7 first lists the main findings and then subsequently addresses the interpretation of the main findings, reviews the methodology and provides recommendations for further research and for practice. Finally it presents the conclusion. Here the discussion points coming from the separate chapters are reviewed in the perspective of the thesis objectives as a whole. They are brought together in order to allow evaluation of the research design and the uncertainties in its outcome.

A general remark about referencing is important here. Applicable references are included with each chapter at its end. After chapter 8 there is also a “references compiled” list for convenience. This list merges all references in this thesis, including the ones only used in chapters 1 and 7 but not elsewhere.
The referencing system in this thesis is based on the references’ first authors last names being placed in alphabetical order. If authors are unknown institution names are used in a similar manner. Chapters that were published as articles in scientific journals with another referencing system were converted to comply with this. Punctuation in referencing is kept to a minimum.
In order to make this thesis usable, accessible and readable for a wide range of people with a chronic illness or disability, an illustrated Dutch summary in lay language is available: an info-graphic. Please feel free to contact the author for more information.

REFERENCES


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Wet Wmo Wet op de maatschappelijke ondersteuning


Truus Teunissen
Material: cherry wood
2 - A COUNTER STORY

This chapter contains the work published in the following article:

Teunissen GJ, Visse MA, Abma TA (2013). Struggling between strength and vulnerability, a patients’ counter story. Health Care Analysis. (Accepted for publication 29 april 2013)

The article describes a narrative study about a patients’ story of 27 years elapsed time between acquiring a chronic lung disease in 1984 and the first ever presentation held by a patient at the 21st European Respiratory Society (ERS) Annual Congress held in Amsterdam in 2011.
Struggling between strength and vulnerability, a patients’ counter story.

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\textit{Health Care Analysis}, Accepted for publication 29 april 2013.

\textbf{ABSTRACT}
Currently, patients are expected to take control over their health and their life and act as independent users and consumers. Simultaneously, health care policy demands patients are expected to self manage their disease. This article critically questions whether this is a realistic expectation.

The paper presents the auto-ethnographic narrative of the first author, which spans a period of 27 years, from 1985 to 2012. In total nine episodes were extracted from various notes, conversations and discussions in an iterative process. Each of these episodes was condensed around a ‘critical moment’ as perceived by the “self”.

The critical moments in the illness process vary between newly encountered problems with basic needs and mourning, to renewed strength and the desire to grow, embracing new situations. Being confronted with and living with a chronic illness involves periods of anxiety and self centredness alternating with strength and advocating the interests of peer-patients. These episodes of emotion, confusion and refinding a balance have a cyclic pattern. The narrative illustrates the vulnerability and dependency of a patient with a chronic disease. The discussion relates this to mainstream dominant views on patients ‘in control of their own life’.

The narrative illustrates, that the vulnerability and dependency of the patient are key factors to take into account in health care policy. The narrative provides a counter story, challenging current thinking in terms of strength, self-management, patients’ own control and independent role.

\textbf{Keywords}
Narrative, patient involvement, counter story, recursive, vulnerability, autho-ethnography.
INTRODUCTION

An astounding 4.5 million people in the Netherlands suffer from a chronic disease (Hoeymans et al, 2008). Worldwide between 40% and 60% of the population has one or more chronic diseases. The number of people with a disability estimated worldwide is about 15% or 1 billion (WHO, 2011). Acquiring and having a chronic disease or disability has a major impact on life and well-being of both those that have it and on their family and friends. This may concern education, participation in society and quality of life (de Klerk, 2000) (van den BrinkMuinen et al, 2009) (Meulenkamp et al, 2011).

From a historic point of view we are used to seeing doctors as being in charge of the disease; today many responsibilities are being transferred to patients and clients. In current thinking patients are being perceived as informed and autonomous experts, directing their life and care, and have progressed from being passive recipients of health care as they were regarded in the past.

While doctors and other health care professionals are expected to act as providers of treatment, care and objective and scientific information, patients are now expected to act like consumers making their own decision. The shifting of responsibilities is reflected in new organizational arrangements of care. These arrangements aim to maximize the autonomy of patients.

The notion of autonomy is widely accepted in Western countries. It has gained the status of one of the core principle’s within the field of bioethics. The concept of patient consumerism is also reflected in healthcare policies and legislation in Western countries. In the area of medical sociology, doubts have been raised about the consumerist ethos in healthcare. First of all, patients are not always adequately informed and fully aware of the value or relevance of care as a ‘product’ (Badcott, 2005) (Cox et al, 2003).

Also power relations should be taken into account. Patients often find it hard to articulate their needs, and many patients feel that their voice is overridden or silenced, or stripped of personal meaning in medical encounters (EC, 2012) (Barry et al, 2000) (Toombs, 1990) (Charon, 2001).

Healthcare professionals often lack the required communication skills to elicit patients’ preferences and involve them in treatment decisions (Say et al, 2003).

Also healthcare professionals are nowadays expected to be more a coach and a motivator rather than a person who knows it all and cures the disease, which is of course not possible with a chronic disease (Trimbos, 2011) (Newman et al, 2004).

Incorporation of the individual patient or client perspective, in various ways, by people known to have experiential knowledge about a chronic disease or disability, has become an everyday phenomenon. Patients are recognized as contributors of experiential knowledge, preferably ‘acquired by personal or even bodily experience’ (Toombs, 1990) (Svenaeus, 2000) (Caron-Flinterman, 2005). In spite of this widely accepted practice, still only a few people with illness or disability are able to carry out this expectation (Williamson, 2010).

The few patients that take this role upon them face an overload and feel inadequately equipped with information about their illness or disability (Teunissen & Abma, 2010). In the current health discourse patients are expected to act as users and consumers and make
choices within the services provided by the health system. In this way patients are assumed to be able to take control over their health, self manage their disease and manage their path through life (Alakeson, 2011) (Boivin et al, 2009). This raises the question whether current health care and patient involvement policy that focuses on self-management and self-control are realistic and viable. This paper aims to enhance our understanding on this. It finds answers by analyzing 27 years of personal life experiences of a patient with a chronic lung disease.

METHOD

Autho-ethnographic approach
An authoethnographic approach aims to interpret the experience of a life history or episode in life through a personal narrative. The personal experience is critically analyzed via introspection. An authoethnography can serve to produce descriptions of: (a) the researcher’s own culture, (b) a culture in which she has been accepted completely, (c) the culture of the self – personal authoethnography and (d) the culture of others as they relate to the self and the researcher – reflexive autoethnography (Ellis & Bochner, 2003). This article uses a mix of a, c and d because the first author applies an ethnographic inquiry to a personal experience (c), relates them to the context of health care settings in which she participates (a and c) and subsequently uses the insights to understand her position as a patient-representative within settings of participation (d). This article therefore uses a combination of personal and reflexive authoethnography. Personal, because it aims to learn from the personal experiences of the first author with her chronic disease, her role as a representative and the implications for her involvement in health care settings. Reflexive, because she reflects on underlying values and expectations (obligations) of patients, her own intrinsic willingness to regain a meaningful role, and the consequences of user involvement on the way she experiences her illness. Truus, the first author, whose experiences are the focus of this study (the ethno) is the one who describes and interprets her experiences (the autography).

Narrative method: collection & analysis in one
Truus’ experiences and interpretations are represented by an episodic and reflexive self-narrative. Truus’ narrative is written primarily by herself during an iterative and reflexive process. During this process, besides writing the article, the second and third authors asked clarifying and reflexive questions that supported Truus to build her narrative. The co-authors helped to interpret the personal narrative in the context of Western society, health care policies and cultural shifts in thinking about the role of patients. Truus’ narrative is presented by several narrative episodes which inform the reader about significant events during her illness history. Literature reports on the meanings people assign to important events with ‘episodes’ and ‘turning points’(Taylor, 1983) (Tedeschi & Calhoun, 1995) (Vaillant, 1977) (McAdams, 2001). People who experience critical events and episodes in their lives often tell that these events and episodes made them see

Experiencing turning episodes influences several dimensions. Firstly, the perceived identity of the narrator and her way of coping with her disease. Secondly, her relations with others in general (Sperry, 2011) (Goldsmith et al, 2006) and her relations with others during involvement in particular. Lastly, Truus’ experiences of several episodes, transformed her needs and values of life in general (Tedeschi & Calhoun, 1995) (Walker, 2007).

In the literature on narratives a distinction is made between, on the one hand, dominant or standard stories, that reflect mainstream culture, and on the other, counter-stories that represent alternative plot-lines (Polletta, 1998). The dominant narrative about patients today is that they are resolute and robust and in control over their disease and their life. The narrative in this article qualifies as a counter story since it clearly reveals differences with respect to the dominant narrative.

**Quality criteria**

An autho-ethnographic narrative should provide an in-depth and honest narrative that function as a vicarious experience (Stake, 2000). In our case, this concerns a narrative of how someone who lives with a chronic illness develops experiential knowledge and what this means for being a patient-representative. Within auto-ethnography, specific criteria to judge the quality of the account are used, and they differ from traditional positivistic criteria. An important criterion concerns credibility, which refers to the match between the autho-ethnographic account and the reality as experienced by the narrator (the first author). Several techniques, to enhance this match, have been used, like peer debriefing with the second and third author and progressive subjectivity (Guba & Lincoln, 1989). The latter means that the development of the first author’s ethnography has been closely monitored and prejudices have been questioned by the second and third author. Another criterium to judge the quality of an autho-ethnographic study concerns its transferability to other than the studied context. This article does not aim to generalize insights. Instead, by providing a complete as possible account it is on the part of the reader to determine which insights are transferable to his/her context and which are not. The narrative therefore needs to ‘evoke in readers a feeling that the experience described is life-like, believable, and possible’ (Ellis et al, 2011). Lastly, by reflecting upon her experiences theoretically and together with the two co-authors, Truus develops a deeper understanding of her experience, which contributes to her individual experiential knowledge. This interpretive process, where interpretations were discussed can be seen as a quality control procedure in its own (Guba & Lincoln, 1989).

**RESULTS**

The first result of this study is the auto-ethnographic story itself, spanning a period of 27 years, from 1985 to 2012. The first author is the “self” in the narrative. In the spring and summer of 2012, a series of episodes was constructed from various notes and discussions
in an iterative process. Each of these episodes was condensed around a ‘critical moment’ as perceived by the self. The episodes are included below in time order.

**Episode I - Being ill and scared**

*Spring of 1985: ‘One day early in the spring I had ‘flu’. The ‘flu’ did’nt go away but developed into bronchitis and pneumonia. After treatment also the pneumonia did’nt go away an I had to go to hospital for a while. Tests resulted in a diagnosis I could not get my head around. Overwhelmed, dazzled and numb, I could not even think of any question to ask. Deep inside I felt anxiety, insecure about what was going to happen to me, unsafe in my own body. I only wanted one thing: to get away from that frightening place, to the safety of my own home! So there I was, just moved to the other side of the country, with two little kids, feeling ill, sick and exhausted. All this was frightening and –quite literally – breathtaking. There was one thing among all the doctors had said that was clear: the disease would probably come back. A very scary thought, 30 years old and stuck with a disease that suddenly popped up. Now what ? I recovered and decided to regard the time I had been ill as a stroke of bad luck, an incident. I did not know anything about patient organisations, expectations of resuming control of my own life and selfmanagement, nor was I interested in any of this. Full of ambition, taking care of a young family, moving into a new house in a new town, I just looked ahead, thinking positively, so saw no problem . . .”*

**Episode II - Help, it doesn’t go away**

*Summer of 1985: “As I got better I enjoyed the summer, healthy food and the sunshine. Life felt as it had been before. It had been an incident and I went on with my life as if nothing had happened. And that went well for a while. Until I got ill again at the end of 1985, but I recovered again, deciding not to complain and fight my way through it. Not visiting the GP too soon and certainly not using all medication if I felt just a little better. The mere thought of getting used to medication or even becoming addicted . . .

Though a feeling of panic often encroached on me increasingly during illness periods in 1986 I wondered: My body doesn’t let me down, or does it ? I started to feel unsafe in my own body. But then again, I was convinced nothing was going to stop me to organise my life as I wanted it, I simply ignored the disease, assuming that living healthy would prevent me from becoming ill again.
I reasoned it was amazing that an infection could not be cured. Surgeons could transplant whole hearts and lungs. Baby’s were born from a test-tube in a lab, why was my disease not curable? I felt angry at the doctors. I definitely was not open to people telling me that it might be a hereditary disease or that it might be chronic. After yet another period of illnesses I decided in 1989 to take up a study I had been interested in for a long time, and not return to my old job. Being forced to work on fixed times and distant places would only lead to problems. As a student any damage would
only affect myself and studying in my own time would be easier to manage. So that was how I rationally coped: if you have a problem look for a solution. It made me feel proud.”

Episode III - Don’t leave me behind

Winter of 1990: “Then I saw a documentary on TV about elephants living in an African nature reserve. A herd travels around, searching for food and water. There is continuous danger of exhaustion, dehydration and predators. One of the animals is ill and can’t keep up. The herd comes to a halt to allow it to recover. Yet, it doesn’t, so it takes a day, and another day . . . The herd suffers from lack of water and is now an easy prey. Then the ill animal can no longer walk and is left behind. The herd moves on.

This story precisely described the threat I felt myself: I will be left behind, losing everyone around me. Even my husband, worried about my health as he was and still is, is not only loving me but also starts to provide care, which is not good in an equal relationship. I was not brave enough to say to everyone presenting ‘good advice: ‘mind your own business, I will do this my way . . .’. My way included taking garlic on an empty stomach, drinking strange black herbs extracts and visiting a magician. I even visited a Greek monastery having an icon with healing power for my chronic disease. They offered me a stay and said that many prayers would heal me for certain, if I would surrender to it that is. My husband pulled me away from them but the seed of false hope was planted in me. Perhaps I could solve the problem and be back to normal.

I wondered who I was, having problems with showing up at work, doing a hobby not wisely chosen (sculpting, causes a lot of dust), attending social events like a birthday party with people smoking thus making me ill for days. I felt as a burden to everyone around me and wanted to run away and not come back before it was over and done with . . .”

Episode IV - Going down

Autumn 1994: “Loss was becoming the most important feeling. I was losing work, friends, hobby, grip. I was getting off the track. I gave up on buying theatre tickets at the beginning of the season since I could hardly ever make it to a show. Financially wise but a pity since going out and having fun with a couple of friends was lost too.

I remembered one evening where I went to the theatre with husband and friends. My lungs had been giving me a lot of problems already for weeks but I did not want to cancel yet again just before the show. To avoid coughing during the show I took medication in advance and brought a little steel flask with drinking water. It was the type of flask hunters use to have for a snort after the deer is shot dead. I brought all sorts of medication too, and honey-candy, peppermints, salty liquorish and a few packs of handkerchiefs. I felt both happy and frightened to go. During the show my husband handed me the flask on request and I sipped every time I felt like coughing. Now I was not only ill but also a secret drinker . . .

My biggest problem was the erratic way the illness behaves. I had become an unreliable person, missing appointments, hesitating to make plans. I felt unreliable, unpredictable, a
disturbing factor and an unwanted confrontation with the limits of being in control of one's life. I have to acknowledge that the ideal - or rather the illusion - of good health based on leading a good and healthy life is not working for me. I felt disappointment in my body and my life in general.

Being chronically ill I had become a modern day pariah. A bit like in the middle-ages when people with the plague had to live outside the towns fortifications to avoid contamination of other citizens. I sensed, people did not like to be confronted with erratic diseases and loss of control. And sure I would rather be in control and plan ahead for nice summer holidays in the mountains and stay over for Christmas at my sisters’ house. All the interruptions caused my work to be lagging behind more and more. Every time a recovery was followed by a frantic attempt to catch up at work, avoiding to face my being undependable. It became a vicious cycle of getting ill, recovery at home, starting to work too quick, a ‘tour de force’ to catch up and – perhaps – for a moment, things being normal for a while before getting ill again. I was slowly drowning . . .

I got depressed. How to give direction and meaning to my life? Whatever evasive manoeuvre, there seemed to be no escape from my illness. I got stuck, a prisoner in my own body. A body that made me feel ugly, affected by prednisolone and extra kilos due to fluid retention. Looking at my face, puffy and spotted red, thanks to a bit of fever, inspired people to say to me: ‘I heard you were ill, but you are looking healthy with that blush on your cheeks’. I wondered, am I still credible?”

Episode V - I should accept it

Spring 1997: “I started to accept the idea of having a chronic disease, and being part of a group of patients. A while later, I found the lung patients’ organization magazine in the mailbox, read one sad story after another and threw it in the waste paper basket. This was precisely why I hesitated to become a member: too much talking about illness and complaining.

Nonetheless I contacted the organization, realizing I had to live with the disease and had to learn how to ‘manage’ my ‘new’ life. There wasn’t much else than peer group meetings but I decided to try.

After a long time I added some fun and meaning to my life: I found out that my own sculptures could be cast in bronze if I used wax rather than clay to make a model. Bronze, that was it: making something strong and solid and not vulnerable like myself. I felt my erratic and unpredictable disease as something I cannot adjust to, unlike a disability that does not change. So it all started with a very small sculpture of a person squatting with the head resting on the arms. It looked a bit sad but it made me feel very happy when I picked it up at the foundry. Finishing and polishing it made me feel proud, I created a thing of beauty. It was a relief to notice that artistic part of me was still there. Friends and family were surprised and enthusiastic and said: ‘wow, wonderful, keep going’. That felt very good indeed!

Before I had felt only deficiencies in basic needs, recognition, safety and meaning, now I gathered there was actually a new future for me as a person with a chronic illness. So, perhaps a new beginning, how would I go about it?”
Episode VI - I'd better do something with it

Winter of 1998: “Then, during one of my quick glances in the lung patients magazine – you never know – my eyes caught a call for a patient representative in a nationwide patient advisory council. Working as a consultant and involved in policy studies, this would be a good match with my skills and interests.”

Episode VII - Are we being taken serious?

Spring 2003: “Some time later I joined that patient advisory council. Providing advice to management and board of the lung patient organisation, both asked for and on councils’ own initiative, was the task at hand. After the first period of euphoria I realized that the council was rather a cosmetic tool: the patient organization could claim that it listened to the patients voice and society appreciated that. Surely being a member of the council was pleasant too, travel expenses, tasty lunches, lots of new pencils and note pads and interesting reports to read and discussing patients’ opinions. Most council members were happy to be able to contribute something but I slowly started to feel anger because I felt not taken seriously at all. My presence was used as an excuse to not give patients a right to have a say. Councils’ advice was never followed by information about what happened with it. Asking for such information often made things worse: delays, cancellations, temporary suspensions and so on. I started to see that the impact an advisory council can have is limited. Its members are often confronted with medical or political questions they cannot handle. How can members know what the interests of the whole patient group is? Can the council represent the entire group? How to involve all these patients in the process? There were more questions than answers.

Just as this was becoming clear to me I was asked to come to work for the lung patient organization as a professional. Perhaps the council did have an impact after all. Of course my self esteem got a boost. So the illness payed off, being a patient was now my job . . .”

Episode VIII - Nothing more to loose

Summer 2005: “And then, on a sunny summer day, I heard I had a second disease, probably lethal within a few years and all. The world stopped around me. I started back at the beginning again, being afraid, fighting, despair, having no control and living by the day. The process was more chaotic and taking less time than before though. I felt there was nothing left to loose. It was very frightening and also changed my perspective: I moved unimportant things aside and focussed on what really mattered. Carpe diem! Life, shortened as it is, became more intense and I became more conscious that there is an end to it.

This made my last bit of hesitation disappear. I felt that I wanted to fight for people with an illness or disability. Dealing with the two diseases together changed my balance. Life went on and my job became a burden I could no longer carry. Having to take sick leave more and more often I found myself in a two year battle against the clock. I tried to keep my job, not wanting to receive social security. After eighteen months I had to give
up. Tired and without any energy left I had to conclude this was no way to live, something had to change. I had to face it: finally I was incapacitated and it was official. The first thing I felt was that there was no stigma. People didn’t look at me differently. The pressure was off and slowly I got some energy back. I cheered up again and was more surprised myself about this turn than family and friends around me."

**Episode IX - Becoming a patient representative**

**Autumn 2008:** “I was meeting many patient representatives like myself and recognized the difficulties they were having. Many had a problem with advocating in practice because of difficulties with attending meetings and vulnerability issues. My own vulnerability caused by repeated periods of involuntary absence due to illness made it difficult to ensure continuity as a patient representative, although I tried hard, of course. Working together with other patient representatives, in pairs, reduced this vulnerability a lot. It is helpful and encouraging to talk to each other also about this vulnerability and about periods of more dependence. Most other patient representatives recognized this too. In 2011, being a member of an appraisal committee, I realized that I had reached Arnsteins’ citizens participation ladder partnership level (Arnstein, 1969). My lung disease can still unexpectedly hit me very hard. Spring 2012 for instance I became so ill that I was taken to hospital in a sunny town in the south of Spain. No control, no choices, no escape, back to square one. I was scared again and fully occupied with the loss, the grief and recovery – some instances all at the same time – for months. Looking back, the process I went through came in cycles and I learned that more people experienced this. I also found out that the key moments in the development of the illness and the path of life do not always come sequentially but may happen in a mixed way at the same time. Over the years another illness joined the lung disease and the entire process repeated itself. Parts of the process repeat themselves at different turns of the path too. I have had little control over it. I feel strong and vulnerable at the same time. This continues until today.”

**Reflection**

Truus’ auto etnography demonstrates the dynamics of her experiences as she encounters: (1) being vulnerable (2), being dependent, (3) experiencing bereavement and mourning, (4) being subjected to a recursive pattern of relapses and falling back, (5) changing needs and finding a new meaning in life.

**Vulnerability**

**Observations from story**

In episodes I to VIII the first disease gradually turns Truus’ life upside down. It gets to the point where she feels unreliable, her appearance deteriorates, can no longer do her work properly, is feeling no longer credible to people around her and she is finally hit by a second life-threatening disease in episode VIII.
**Analysis**

Despite the enormous will to live up to the ideal of the ‘successful patient’, Truus’ story shows that living with a chronic disease is capricious and messy. Truus cannot be autonomous in the sense of directing her life without the support of others. Vulnerability and interdependency are key values in her story. Although Truus’ main vulnerability is caused by the diseases and associated irregular relapses, there are other medical vulnerability factors to be considered. These vary from e.g. good personal relations with the GP, to frequent changes in hospital staff, medication being altered for cost reasons and non availability of treatment at home. Becoming chronically ill also means there is a major threat to financial income and to self-esteem. The risky transition from having a job to only receiving social security clearly underlines this. Deciding to go on holidays is a gamble each time.

**Theoretical context**

The dominant policy and theory about patients today is to assume that they are willing and able to act as consumers or users who choose health services (Alakeson, 2011) (Boivin et al, 9). Truus’ story clearly indicates that she is willing, but not always able to do this. Part of her is not under her control as the illnesses cause erratic disruptions of her life and a feeling of insecurity in her own body. Also the quality of the relation with care professionals is mentioned (Toombs, 1990) (Svenaeus, 2000) (Charon, 2001).

**Interdependency**

**Observations from story**

Truus describes dependency of diagnosis, treatment, GP and medical specialists and hospital facilities in episode I. Dependency and risks of medication are touched upon in episode II. Her dependency of the erratic disease itself emerges too. In episode III and VIII she mentions the importance of people around her: family and friends.

**Analysis**

When confronted with her diagnosis, Truus experiences her newly acquired disease or disability as something she was not in any way prepared for. It took years before a capability to do some form of self-management emerged. Although there are enormous differences between patients, only few patients can be left completely without treatment or other support. Many diseases and disabilities create an unwanted dependency of medication, treatment or counselling that is being offered at a single location and strict regulation. This leaves little room for patients exerting their own control.

**Theoretical context**

In current thinking it is expected that patients also act as representatives who are involved in the design and provision of care services and decision-making processes. The autonomy myth underlying this dominant narrative can be questioned. Truus’ story shows it is neither realistic for her to see herself as fully self-sufficient and in control, nor is it to be regarded
as fully autonomous and independent. The illness is experienced and often felt as *Unheimlichkeit*, meaning uncanny unhomeliness (Svenaeus, 2000).

**Bereavement and mourning**

**Observations from story**

The narrative story episodes explicitly describe different emotional responses to events: anxiety (I), denial and anger (II, III), resistance (IV, V, VI) and acceptance (V). Not only does this response repeat after each major event like in episode VIII, it also repeats after minor events like the one mentioned in episode IX. The notion at the very end of the narrative story part IX suggests that mourning doesn’t stop but continues in ever-smaller cycles.

**Analysis**

Early in the narrative story the phases take considerable time, are explicit, come in sequence and are separate. Later in the story they repeat, clutter and are experienced in a more chaotic manner and have increasingly shorter durations. Both the unpredictable relapses of the first disease and the regular hospital visits to investigate recurrence of the second disease, lead to frequent critical events. Every time Truus experiences a critical event, she experiences chaotic emotional responses of varying intensity and magnitude like anxiety, fighting and depression. Finally there are sudden bursts of emotions in between events without a directly observable trigger.

**Theoretical context**

Truus experiences disruption of her body functions, her self-worth and the world around her (Toombs, 1990). The loss of health, career and way of life certainly qualifies as ‘bereavement’. We contend that the Kübler-Ross *five phases of mourning theory* describes the sequence and nature of the emotions in the mourning process experienced by the ‘self’ in this narrative quite well. The five phases are all discernible: 1-Denial, 2-Anger, 3-Fighting, 4-Depression and 5-Acceptance (Kübler-Ross, 1970). In the beginning the different emotions come sequentially and they closely follow the five mourning phases. The story shows several mourning phases unfolding again after major events. The first time the full five phases mourning process appears to last almost 12 years. After the second major event the five phases are recognized too but they go by in a shorter time of about one year. During and after each unexpected stay in hospital, a shorter, more chaotic periods occurs with the ‘self’ experiencing many or all of the associated mourning emotions at the same time. During such repeats, mourning phases may appear at the same time rather than sequentially and not all phases might be present in each repeat. Also the repeats have a shorter duration with time.

**Repeating pattern**

**Observations from story**

Several structures can be distinguished in Truus’ narrative story. Episodes I up to VI describe a full mourning process taking a decade, episode VIII a shorter repeat lasting a
year, episode IX mentions an example of several shorter mixed and incomplete repeats, all linked to a trigger event. Episode IX suggests an even shorter and more frequent repeat of emotions without an identifiable trigger.

**Analysis**
The ‘self’ experienced many Kübler-Ross phased or chaotic mixed emotional responses of decreasing duration and intensity over time. We observe that mourning emotions apparently never completely stop and may have different types of triggers.

**Theoretical context**
This response pattern cannot simply be characterized by any of the terms ‘repeats’, ‘repetitive’, ‘cyclic’ or ‘recurring’. The best fit in our view is a recursive pattern (Gouyet, 1996). Although recursion is, in this case, merely descriptive and not explanatory, we contend that such a complex recursive pattern in the mourning process - when observed generally - would require further attention of professionals during e.g. psychological treatment. Also the pattern affects the ability to do proper self-management.

**Human needs**

**Observations from story**
Truus’ story describes nine critical moments. Episodes I to IX are each leading to a change in perspective and a turn in her path through life. There is a period of several years with a downward trend in episodes I to VI followed by a ‘turning moment’ in episodes V and VI and an upward trend in episodes VII to IX. In the downward period Truus worries about physical safety (II) and being accepted in the group of people around her (III, IV). After the changeover she refinds herself and her ambitions (VI, VII, IX). The long 1985 - 1997 period before the changeover Truus was merely a patient being busy with her own health. Only after the episode VI changeover Truus started to do increasingly more active patient advocacy work.

**Analysis**
Truus’ story indicates that ‘needs’ might be associated with this changeover. The need to ‘feel safe’ and of ‘being loved’ and ‘belonging to a group’ are clearly mentioned in the downward period. The ‘self esteem’ and ‘self realisation’ needs are emerging in the upward period. In between is the turning moment, leading to a changeover in orientation from basic needs towards higher needs. The personal development process towards becoming a ‘patient advocate’ coincides with the moment the Kübler-Ross ‘acceptance’ phase was reached.

**Theoretical context**
A range of ‘human needs’ and a theory, claiming that lower physiological needs preceed higher psychological needs, were published in the 1940s. Maslows *Hierarchy of Needs-theory* distinguishes five levels of needs: 1-Physiology, 2-Safety, 3-Love and belong, 4A/B-Self-esteem, 5A/B-Self-realisation. Levels 4 and 5 are growth oriented needs and lie beyond
a ‘transgression point’ (Maslow, 1943) (Maslow, 1954) (Maslow, 1970). The narrative story compares rather well with the original hierarchy of needs theory. In episode VIII also the ‘human needs’ show a repeating process: physiological needs, safety needs, love and belonging needs and self-esteem needs recur back from the start. Here, the trigger appears to be a relapse event where physiological and safety needs are endangered.

Truus’ story mentions she moved along Arnstein’s ladder of citizen participation. We contend that her path in fact closely follows its 8 upward steps: 1-Manipulation, 2-Therapy, 3-Informing, 4-Consultation, 5-Placation, 6-Partnership, 7-Delegate power, 8-Citizen control. Steps 6, 7 and 8 are regarded as citizen control (Arnstein, 1969). Most striking is the alignment of the changeover with the first mourning acceptance phase, with the needs transgression point and with reaching citizen control in her advocacy role. Figure 1 illustrates this.

![Figure 1 Alignment between theoretical frameworks in Truus’ narrative.](image)

**DISCUSSION**

So how does this complex recursive pattern fit within the dominant thinking in health policy? The current paradigm depicts patients as lay-citizens being subjected to treatment by medical professionals in institutions. Recent economic set-backs have resulted in budget cuts. These were accompanied in the political debate by the image of patients making their own choices and being user and consumer of services offered up by the health system.

The Dutch government aims at chronic disease patients dealing with the problems themselves without professionals for a longer time (GR/RVZ, 2012). Self-management is
not only out of reach for the illiterate, drug addicts or mental health patients. This counter story shows that a much larger group can not comply to the self-management model. Too easy relying on the patients’ voice being heard in current patient involvement presents a risk. Diverse patients will express diverse opinions and not an averaged out mainstream-mix that can simply be compared to a standard (Barnes & Cotterell, 2012).

**The resolute and robust patient**

In practice patients often have little real influence on health policy, health research and quality of care (Bovenkamp, 2010). People with a disease or disability are often fully occupied with managing their own lives even up to the point of being overwhelmed. Society, asking patients to participate in health research or in projects on quality of care, might be overloading them (Trappenburg, 2008). There are other explanations for the difficulties with getting patient involvement afloat. Patients’ organisations, especially the smaller ones, have only limited possibilities and resources to select and support their representatives (Elberse et al, 2011). At the same time, in spite of the above, a number of patients, well motivated and highly driven, is being observed to take up this ‘work’ as a representative (Kapitein, 2011). Hence, both scientific literature and the current health discourse are incomplete and paradoxical about patients living with a disease or disability and their level of control over life.

In literature no scientific in-depth analysis was found on experiences by patients who became patient representatives. This is to be expected since many people are reluctant to share their experiences of disease, pain and loss. In the current discourse on public health the concept of the ‘successful patient’ sits at the core. Such patients are portrayed to be resolute and robust and therefore not being vulnerable or in need of care.

**Patient consumerism, a flawed concept**

The story of Truus illuminates that the dominant story hides many aspects of learning to live with a chronic disease. Health policy and discourse assumes independent patients acting as users and consumers. Truus underlines she was not able to do this when confronted with a chronic disease and a life threatening cancer in her thirties with two kids. Self-management and user-involvement have been completely outside her experienced reality for many years. She was dependent, vulnerable and needed care and support. Also in general, citizens are by far not all self-sufficient or autonomous, and neither are patients. Their demographic characteristics are roughly the same as those of the general population. This implies that between 10 and 25% have insufficient skills to shape their life independently in today’s complex society, even if they would be healthy (Lindhout et al, 2012).

It is logical to assume that there is also a group of patients that otherwise cannot be self sufficient due to the very nature of their psychological or physical illness, disease or disability.
As a general pre-condition for the design of the health system, the concept of self-sufficient patients is therefore not viable for all patients all the time and is hence unethical.

**Main new insights**
We derived three main insights from this narrative story:
- Before a patient may become able to contribute to any active patient involvement, there must be some acceptance of the disease and basic needs must be satisfied.
- Taking into account vulnerability and dependency
Recognition of the relation between mourning phases and human need aspects, in the process a person with a new chronic disease faces, is a valuable asset in the work of both professionals and patient organisations. Understanding the vulnerability and dependency of patients will enlarge the chance to properly address and take into account all this in the way patient representatives operate and require support.
- Recursive pattern
Recognizing and acknowledging a recursive pattern in a mourning process provides additional insight to patients trying to manage and accept the disease adds to quality of care.
Patient organisations and medical professionals can translate this in to better adapted services and better taking into account of the mourning process.

**Uncertainties and limitations**
Critics may argue this narrative over 27 years lacks completeness and depth. Indeed several other diseases and several other aspects like work, family, relation and way of life were left out for - among others- privacy reasons. These aspects cannot be isolated from the experiences with the diseases. On the other hand true completeness is not practically possible. Also, writing this narrative story is a highly subjective thing. Examples of this are found when looking at the triggers of mourning phase emotional responses. For the first author, the ‘self’, a specific odour or a minor event in the hospital reception area would be such a trigger. Other patients may show completely different reactions.
So, can this story told by one person, incompletely told as it is here, be of value to other patients, their organisations and to professionals? Is the story transferable to a more general level and relevant to others?
We contend that our observations are indeed of practical relevance to the discourse of patient involvement and to patient representatives.
Most importantly this counter story generates a different sound in the health discourse about patients dealing with returning episodes of vulnerability and dependency as an important issue in their life.

**CONCLUSION**
The research question can now be answered. Is the current healthcare and patient involvement policy that focuses on self-management and self-control realistic and viable?
This story shows that patients are not all resolute and robust. Nor are they in control of their life with a disease or disability. In reality it is much more complex and subtle. It is of great importance to not deny but acknowledge their vulnerability and dependency, both when merely being a patient and when acting as a patient representative. Besides strength, selfmanagement and patients’ own control in life as guiding concepts, also the vulnerability and dependency of patients are factors to take into account in health policy discourse, professional health practice and patient involvement. A more realistic view will do justice to both the patients and to patient involvement. To start with, vulnerability can be compensated for by a supporting network. Working together with ‘joint-sufficiency’ as a basis rather than the idea of ‘self-sufficiency’ and being in control of ones own life will help. Acquiring of-, and living with, a chronic disease is associated with a sense of grief, loss and bereavement. In fact a complex recursive process, combining mourning emotions and deficiency of needs, appears to take place. Knowing this is a valuable asset for patients and their representatives (recognition, insight, compassion), for health care and research professionals (compassion, recognise the vulnerability and more or less dependence), and for organisations promoting patient involvement.

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Truus Teunissen
Materials: marl, bronze and steatite
3 - EXPLORING CRITERIA OF PEOPLE WITH A CHRONIC ILLNESS OR DISABILITY IN THE NETHERLANDS

This chapter contains the work published in the following article:


A first article supporting the start of the research work in this thesis was published in the Dutch in 2010: “Teunissen GJ, Abma TA (2010). Derde partij: tussen droom en daad. Een verkennend onderzoek naar de patiëntenantij en -criteria voor onderzoek, beleid en kwaliteit bij overheDEN en zorginstellingen. *Tijdschrift voor Sociale Gezondheidszorg*, 2010;4(88):182-9.” This first article was used in the Netherlands to prepare the research and to generate interest in the field.

The journals *Patient Education & Counseling* and *The Patient* both successively processed and peer-reviewed an English translation in 2011 but decided not to publish it in view of the content in the first Dutch version.

A newly written second English article about the work as it was progressed by 2012 was submitted to the *International Journal of Person Centered Medicine* (IJPCM) and was reviewed and accepted soon after on September 17, 2012. Regrettably the IJPCM journal ceased to exist end of April 2013, resulting in unexpected delay and making publication uncertain once again. Fortunately, the activities of IJPCM were continued under a new name: the *European Journal for Person Centered Healthcare*, EJPCH. There the article was again accepted for publication on October 15, 2012.

Publication was then re-scheduled for June 2013 but setting up the new journal caused further delay. Next the publication was planned by the editors for August 2013. After an additional month delay the article was finally published on October 1st 2013 in this new journal, EJPCH, Volume 1 Issue 1 on pages 232-239.
Patients at the negotiating table: exploring appraisal criteria of health research and quality of care used by patient advocacy groups in The Netherlands.

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ABSTRACT

Background: Western Governments and the public at large acknowledge the importance of strong patient advocacy groups. A new type of involvement has emerged: patient representatives at the negotiating table, the patient group negotiating as a collective with other stakeholders. However, patient representatives feel inadequately equipped. This study was designed to identify ‘issues that matter’ to patient groups in The Netherlands and whether these issues are brought to the healthcare and research negotiating table between healthcare providers and health researchers.

Methods: Using a qualitative approach, the extent to which patients are involved in the assessment of health research and quality of Dutch healthcare from a patients’ perspective was explored and also which criteria they use. A literature search, participant observation and interviews were carried out.

Results: The results demonstrate that patients are mainly consulted on an individual basis, but are to a much lesser extent involved as a group. There are patient criteria and guidelines in use for assessment of the quality of care, but there is virtually none for assessment of health policy and research. Many patient criteria are poorly operationalized, vague and abstract and are difficult to apply in practice.

Discussion and Conclusion: Based on these results the authors propose and discuss a new concept: a list of patient criteria for evaluating health research, policy and quality of care.
These should be developed in dialogue with patient groups. A list of such criteria is expected to be of practical use to many patient advocates in many countries.

**Keywords**
Appraisal, commitment, decision-making, experiential knowledge, health policy, lay expertise, lung patient, medical research, participation, patient advocates, patient involvement, patient preference, person-centered care, quality of life.

**INTRODUCTION**
Recognition of the importance of the patient’s voice being heard – and not only in the individual patient-doctor setting – is increasing. The political impact of patients as a collective is also equally important for health-related research, policy, and quality in Government institutions and private organizations. Patient involvement in the decision-making process is currently a subject of great interest and is being advocated at many levels (European Commission, 2012) (Leys et al, 2007) (Williamson, 2010) (WHO, 2007) (Garrido et al, 2008) (IAPO, 2008). Theoretically the patient collective can now act at the healthcare and research negotiating table between healthcare professionals, providers and researchers; offer advice; engage in cooperative efforts; and criticize project proposals. However, even though patient groups are endorsed as a negotiating party, this does not imply that they are adequately equipped to identify relevant opinions among their own constituents, gather and oversee experience data, and formulate a related patient vision. Patient groups, therefore, not only struggle with the question whether they are perceived as a fully equal participant in negotiation, but also with how to usefully perform their role as a negotiating party.

This article concentrates on two questions:
- To what extent are patients being involved as a group in health research, quality of care, and related policy within the Netherlands?
- Which criteria can be used to advance the patient perspective in a way that it is complementary to the professional perspective within the Netherlands?

When we speak of patient criteria, we refer to criteria that can be used by patient groups to bring in and assess quality from the perspective of patients. Starting from the patient’s perspective implies, firstly, that patients are regarded as a useful source of experience and knowledge and, secondly, that this experience-based knowledge must be integrated into the policy-setting and decision-making process (Herxheimer, 1988). Any uncertainty, question, or issue originating from patient experience then impacts the research, quality, and policy process. Patient experience thus adds to the vision of professionals in these fields. Personal experiences can lead to experience-based knowledge via reflection and in turn to experiential expertise when this knowledge is tied to that of fellow patients. Experience-based knowledge is an important source for survival, ‘joie de vivre,’ problem solving, and pleas for support (Herxheimer et al, 2000). Patient participation means that the patient is accepted as a partner in the knowledge acquisition process. This implies that patients are actively involved on an equal footing in the planning,
execution, and evaluation of research, quality, and policy. The patient is also a player in the negotiations between healthcare suppliers and health insurance companies. The ‘participation ladder,’ based on Arnstein’s ladder of citizen participation, (Arnstein, 1969) indicates upward steps in the level of control that patients can have: from no say whatsoever to consultation, advice and cooperation, and ultimately full control (see table 1).

Table 1. Patient involvement level and increasing influence.

<table>
<thead>
<tr>
<th>Level</th>
<th>Influence</th>
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<tbody>
<tr>
<td>Patient control</td>
<td>Patient party is the principal and has full control of content</td>
</tr>
<tr>
<td>Cooperation</td>
<td>Patients cooperate as equal partners with other parties and take decisions jointly</td>
</tr>
<tr>
<td>Advice</td>
<td>Patients offer advice when asked but take no group decisions</td>
</tr>
<tr>
<td>Consultation</td>
<td>Patients are asked for their opinion</td>
</tr>
<tr>
<td>No participation</td>
<td>Patients do not ask and are not asked</td>
</tr>
</tbody>
</table>

Based on our vision of patient involvement as a dialogue (Abma & Broerse, 2010), patients would ideally be collaborative partners. This goes beyond the usual consultation (patients provide information) and advice (patients have no decision power) but does not constitute full control (all decision power is delegated to patients). In between is ‘equal partner,’ the role of patients as a partner in the dialogue based on equality and the value of integrated, experiential, and professional or scientific knowledge.

The purpose of patient involvement is twofold: (i) more democracy and (ii) as a contribution to the quality of process and outcome. Five arguments in favor of patient involvement are mentioned in the literature. The first argument, which is most often heard, is the added content that patients may bring to the dialogue (Epstein, 2008). Their contribution can improve the quality of processes and results. A second argument is the legitimacy of the decisions taken. This underlines the importance of more democracy. Since patients are direct stakeholders in healthcare, it is ideologically proper to take their voice into account. Moreover, this ensures transparency of the decision-making process (Baker, 2007). A third argument is that participation in decision making impacts the empowerment of patients in a positive way (Boote et al, 2002)(Caron-Flinterman, 2005). A fourth category of arguments revolves around the likelihood of implementation of decisions taken. It is assumed that patient involvement leads to more commitment in the patient group and thus to increased probability that decisions are actually implemented. The fifth argument relates to the efficiency and effectiveness of the healthcare system. Patient involvement provides care consumers with a counterforce versus care suppliers and insurance companies (WHO, 2007).

In brief, patient involvement becomes a goal in itself when considering democratic decision making and empowerment. In addition, it can be seen as a means towards substantive improvement, better implementation, and greater efficiency.
There is also need for concern. Patient participation must remain a means towards substantive improvement, better implementation, and greater efficiency, rather than to become a goal in itself. There is a risk of patient involvement degenerating into ‘pseudo-involvement,’ thus becoming a means to achieve the goals of other parties (Harrison & Mort, 1998).

In theory, patient involvement in research, quality of care, and policy is logical. Patients with a chronic illness and ‘people with disabilities’ and their organizations, however, emit signals that suggest the opposite. To explore the actual situation, we employed two methods: a literature search and interviews. The data resulting from these different methods were cross-checked, and the issues found in the literature were presented to respondents to elicit comments.

**METHODS**

Firstly, a literature search was conducted. The search strategy comprised the following. To begin with, relevant search terms were established. These are: ‘patient involvement’, ‘influence and participation’, ‘citizenship’, ‘emancipation’, ‘right to have a say’, ‘patient movement’, ‘empowerment’, ‘patient perspective’, ‘patient and public involvement’ (PPI).

The literature in journals on care, research, policy, and well-being was then searched systematically using the search terms. In the sources identified, the references and internet links were searched until no further new relevant sources were found (data saturation). Because of the exploratory purpose of this study, the literature search was limited to the situation in the Netherlands and in the UK. It was focused on non-profit organizations and Government institutions. Business organizations, including the pharmaceutical industry, were excluded from the search.

In total, eleven scientific and patient organization journals, and six international internet sites over the last 10 years were searched using the search terms. (see Box 1 and Box 2). Secondly, following the literature search, some 18 semi-structured interviews, each lasting about 1 hour, were conducted. Respondents were people from various patient and disability groups in their role of advisor and cooperation partner in the time period 2008–9. The respondents all were representatives, participating on behalf of their respective patient group in health research, policy, and quality matters. The selection of respondents took place as follows. Members of the Dutch Asthma Foundation, taking part in national committees, were asked to provide the names of patient representatives. The lead author also approached people in her own network who have been active as patient representatives for many years in various committees. After interviews with nine respondents from seven different patient and disability organizations, no further new information was obtained (data saturation). All interviews were recorded, and typed out. On the basis of quotes from the interviews, a ‘member check’ was conducted to obtain feedback from the respondents as to the correctness of the quotes. The collected data were thematically analyzed by content in an inductive way. The respondents were asked for their claims and concerns and any issues encountered during participation activities.
The interview questions (both the original Dutch version and the English translation) are available at the authors.

**Box 1 Journals searched using key terms**
1. Medisch Contact;
2. Zorgvisie;
3. Zorg en Welzijn;
4. Nederlands Tijdschrift voor Geneeskunde;
5. Vraag in Beeld, Federation of Patients/Consumer Organisations in the Netherlands (NPCF);
6. Chronisch zieken en Gehandicapten Raad Nieuws;
7. Vilans, Kenniscentrum voor langdurige zorg, Nieuws;
8. Nieuwsbrief Participatie min Volksgezondheid, Welzijn en Sport (VWS);
9. International Alliance of Patients’ Organizations (IAPO) bulletin;
10. Health Policy;

**Box 2 Internet sites searched using key terms**
1. Patients Like Me ([www.patientslikeme.com](http://www.patientslikeme.com));
2. Involve ([www.involve.org.uk](http://www.involve.org.uk));
3. People in Research ([www.peopleinresearch.org](http://www.peopleinresearch.org));
4. The James Lind Alliance ([www.lindalliance.org](http://www.lindalliance.org));
6. National Institute for Health and Clinical Excellence (NICE) ([www.nice.org.uk](http://www.nice.org.uk)).

**RESULTS**

**Quality of Care**

There are many ways for patients to be involved in the decision-making process in the healthcare sector. The patient’s voice is increasingly heard in the development of medical guidelines, for example by becoming involved in development working groups. Opinions of patients are used more often to ensure greater transparency and quality of care. Examples of this include quantitative data collection for consumer input in a database of patients’ experiences (Originally called DIPEx, now available from Healthtalkonline) and publication of European Health Consumer Index (EHCI) ratings (Herxheimer et al, 2000) (Hjertqvist, 2008) (DIPEx, 2012). Health care institutions also use qualitative methods such as focus groups, mirror conversation, and shadowing (Involve, 2006). The European Patients’ Forum (EPF) has gathered these and other instruments for patients’ healthcare involvement in a ‘toolkit’ (EPF, 2009). Both DIPEx and EHCI have Dutch derivatives: the Consumer Quality (CQ) Index and the former Institute for Healthcare Improvement (CBO) ‘toolbox.’ Also a set of methods for consumer involvement in health policy, health care and health research is available (Nilsen et al, 2006). In the Netherlands, the introduction of the Social Support
The Wmo Act has increased the possibilities for local patient involvement significantly. Municipalities are now required to investigate customer satisfaction among people requiring care (consultation). This involves asking individual patients their opinion, which thereby provide information. As can be seen from table 1, they are not involved in the analysis of the data obtained. Involvement in the role of advisor is possible via the client councils.

Patient organizations and federations of patient organizations mainly concentrate on the development and use of criteria for appraisal of the quality of care within improvement projects. General quality criteria for care of hospitals, aiming at professional competence, information, client treatment, supportiveness, independence, organization, and accommodation have become subject of scientific study (Bovenkamp et al, 2010). Also health research priorities as seen by both patients and by researchers is being studied (Stewart & Oliver, 2008).

In summary, we contend that instruments for transparency and quality improvement, such as DIPEX and the EPF toolkit, do not focus on collective involvement of patients but rather on individual *ad hoc* patient consultation. Advice by and cooperation with any patient group on a regular and structural basis often comes about due to coincidence, for example as a result of someone happening to know a member of the patient group. The patient collectives and their national umbrella organizations, on the other hand, are busy with development of patient criteria for quality improvement that, aside from individual consultation, provide for possibilities of cooperation.

**Health Research and Policy**

Patients can exert influence on institutional policy in the Netherlands through client councils in health institutions. The introduction in the Netherlands of the Wmcz (Care Institution Clients Involvement Act) gives them extensive advisory rights, although it is subject to criticism. Patients can also influence Government policy. Patient representatives take part in various Dutch Government advisory councils. Furthermore, patient organizations are invited on an *ad hoc* basis to participate in parliamentary committees and express their views on policy decisions at the Ministry of Health. Finally, patients and their organizations have the possibility to change policy in a direction they wish via lobbying activities.

Hardly any criteria are available to assist the patient group with the appraisal of policy. Two criteria for health-related policy appraisal were found: joint development of policy and the policy document evaluation.

Upon request and on commercial basis, experts offer advice to organizations on the client perspective in research projects and research policy. They also provide information on education. Criteria are described to evaluate research policy from a patient perspective, but these have as yet not been tested in practice.

There are also possibilities for involvement in health research. Patients can discuss and take decisions in medical scientific research and increasingly do so, although their role is often limited to that of information provider or advisor (Caron-Flinterman, 2005). An example of cooperation in research by a patient group is the set-up of an integrated societal research
agenda for and with people suffering from asthma and chronic obstructive pulmonary disease (COPD), on behalf of the Dutch Asthma Foundation. Since then, this foundation has continued to work with criteria from a patient perspective for the appraisal of research project proposals. These criteria are (Caron-Flinterman et al, 2005):
- how and to what extent the results contribute to the needs of the target group;
- is there an improvement of health and quality of life of the target group;
- what is the level of influence on the research proposal;
- can the target group have an impact on the execution of the project;
- are results being fed back to the target group.

Several health foundations apply the same criteria as the Dutch Asthma Foundation. The Netherlands Organization for Health Research and Development (ZonMw), a Government health research finance organization, also compiled a list of patient criteria for the appraisal of research projects. This list takes into account the relevance and importance of the research project for the target patient group, and whether (and if so, to what extent) the patient perspective is integrated into the planning and execution of the research (Keizer, 2012).

In summary, we can state that involvement in health research and policy making is mainly at the information and advisory level, and that the involvement is hardly structural and systematic. Cooperation with patients as a collective group is rare, apart from a few positive exceptions. Criteria for the appraisal of policy are scarce. Criteria for the appraisal of research are available at ZonMw and several health foundations. Whether they are fully operationalized and practicable for most patient representatives in their advice or partner roles remains a question. An answer to this question is presented in the next section.

The results of the literature search are shown in table 2. Patient criteria found with different organizations and Government institutions are grouped by type of criterion (relevance, right to have a say, and ethics and safety) and by domain (research, quality, and policy).

Table 2. Patient criteria found in literature, in use at public and private organizations, by type of criterion and domain.

<table>
<thead>
<tr>
<th>Criterion types</th>
<th>Patients, disabled and elderly organizations</th>
<th>Healthcare institutions</th>
<th>Health foundations</th>
<th>Health research and development organization</th>
<th>Government organizations</th>
</tr>
</thead>
<tbody>
<tr>
<td>Relevance to patients</td>
<td>Q, P</td>
<td>Q</td>
<td>R</td>
<td>R</td>
<td>P</td>
</tr>
<tr>
<td>Right to have a say</td>
<td>Q</td>
<td>n/a</td>
<td>R</td>
<td>R</td>
<td>n/a</td>
</tr>
<tr>
<td>Ethics and safety</td>
<td>n/a</td>
<td>n/a</td>
<td>R</td>
<td>n/a</td>
<td>n/a</td>
</tr>
</tbody>
</table>

Criterion types:
Q = Quality of care,  R = Health Research,  P = Health policy,  n/a = Not available
**Interviews**

Our literature search shows that criteria for quality are fairly well operationalized. However, the patient and disability group respondents indicated that they do not use them in practice. Either the criteria are not readily available or, if they are, then the respondents are only vaguely familiar with them. The following quote illustrates this:

“No, there are no appraisal criteria; at least not that I know of. I use my intuition.” (interview 3, male, visually impaired)

Patient representatives who are involved in development of clinical guidelines find it even more difficult. Several quotes illustrate this:

“More and more professionals act on behalf of the patient group in platforms and committees. As a patient, it is no longer clear on what basis an appraisal is done. That is a pity, and it is quite different from how it used to be; then we sat at the table ourselves as patients.” (interview 6, male, Parkinson’s disease)

“I needed to search a lot on the internet just to understand the gist of the proposals.” (interview 4, female, lung disease)

The problems expressed here relate to the fact that the patient group needs a minimum level of professional expertise to be able to join the discussion on the often complex process of developing a guideline (EPF, 2009). On the other hand, too much professionalism has the disadvantage of being too close to the specialists and thus not being authentic enough. This also applies to representatives speaking on behalf of others. Patient criteria for the appraisal of policy and research are, in the eyes of the interviewees, hardly operationalized or not known. Remarks about patient perspective tend to be abstract and vague. Criteria are difficult to apply in practice by the average patient or patient group. The following quotes clarify this:

“There is much demand for patient criteria among professional associations. They ask us: ‘Tell us how to approach patients. What do you consider good care?’” (interview 3, male, visually impaired)

“Also my client group would like to do more appraisal of research, but they don’t know how.” (interview 1, male, rheumatism)

Also some of the issues that matter to patients are not found in the criteria, such as quality of life. Another quote to underscore this follows:

“Not the quality of care but rather the quality of life is the most important goal for patients. That ought to be the starting point from which research and policy need to operate.” (interview 4, female, lung disease)
Respondents also explicitly mentioned several criteria on ethics to be missing. This concerns the availability of insurance for patients who are test subjects in clinical trials covering effects on illness, work, or social commitments. Also they mention access to written information in understandable language about the choice whether to participate, plus sufficient time to consider and discuss before deciding to accept or not. Finally they consider feedback of the intermediate or final results of the project to test subjects and patient groups as important in relation to human dignity.

Again, a quote to illustrate this follows:

“It is so easy to say that the test you are subjected to is harmless and that there is medical support, but if your ailment gets out of control it will take more than just some antibiotics. You may end up being on sick leave for weeks or even months, and that you don’t want.” (interview 5, female, lung disease)

The respondents furthermore missed criteria on patient empowerment such as whether the patient group is a partner in the project committee, whether the patient group is involved in project definition and evaluation and whether the project sufficiently considers the diversity within the patient group in terms of gender, ethnic background, age, socio-economic circumstances, and other diversity aspects.

One patient representative says about this,

“The input that patients provide is entered into the process but does not really have an effect. It’s like getting a bit of mustard with the last bite of the meal.” (interview 2, female, chronic headache)

Finally, criteria on relevance were felt to be missing. The criterion ‘whether the project objective is relevant for the patient group’ was too general, vague, and abstract for the respondents to be able to say anything specific about it. The respondents preferred to concentrate the relevance criterion on the question: ‘can the project upon reaching its goal improve the quality of life for the patient group in relation to their physical, social, and mental limitations?’

In summary, we contend that there are hardly any patient criteria for appraisal known to patient representatives and their patient groups. The few that exist are difficult to use, vague, abstractly written, and poorly recognized. In other words, they are hardly available or operationalized.

**ANALYSIS**

The literature search on quality of care, policy, and health research resulted in a set of largely common issues. The results from the respondent interviews underline and confirm these results. Also, the lack of a systematic appraisal method, available to patient groups, was confirmed. Both the literature search and the interviews indicated that the issues
found show three major areas of concern for patients and people with a disability: (i) relevance, (ii) right to have a say, and (iii) ethics and safety. Hence, these are the three criteria categories by which patients judge both quality of care and health research, so a new concept is proposed: a patient criteria list. The criteria were incorporated in the patient appraisal criteria list proposed in table 3. These criteria must be sufficiently met in order to realize patient involvement.

Table 3. Summary of patient criteria for evaluating health research, quality of care, and health policy

<table>
<thead>
<tr>
<th>Criterion</th>
<th>Description</th>
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</thead>
<tbody>
<tr>
<td>1. Relevance</td>
<td>-Improves the quality of life of patients (physical, social, mental)</td>
</tr>
<tr>
<td></td>
<td>-Leads to better health</td>
</tr>
<tr>
<td></td>
<td>-Takes other diseases into account (co-morbidity)</td>
</tr>
<tr>
<td></td>
<td>-Improves the care for patients</td>
</tr>
<tr>
<td></td>
<td>-Improves social participation</td>
</tr>
<tr>
<td></td>
<td>-Leads to useful results for the patient group as it takes patient group properties into account, e.g. ethnicity, gender, age, and socio-economic situation</td>
</tr>
<tr>
<td></td>
<td>-There is a plan on how to implement the result in practice</td>
</tr>
<tr>
<td>2. Right to have a say</td>
<td>-The patient is a source of knowledge</td>
</tr>
<tr>
<td></td>
<td>-Patients are equal negotiation partners, with a voice in research projects (definition, execution, and evaluation), guidelines, quality requirements, choice assists with alternative treatments in care, and policy</td>
</tr>
<tr>
<td></td>
<td>The patient group is recognized as citizens that must be listened to</td>
</tr>
<tr>
<td>3. Ethics and safety</td>
<td>-The least cumbersome means to achieve the goal</td>
</tr>
<tr>
<td></td>
<td>-Safety of trial subjects</td>
</tr>
<tr>
<td></td>
<td>-Explicit freedom of choice</td>
</tr>
<tr>
<td></td>
<td>-Comply with and notify patients of the rules, codes of conduct, standards, and values in medical trials on patients</td>
</tr>
<tr>
<td></td>
<td>-Human dignity</td>
</tr>
<tr>
<td></td>
<td>-Responsible and conscientious handling of complaints and informing the patient’s own family doctor</td>
</tr>
<tr>
<td></td>
<td>-Information in understandable language on results, side effects, aftermath, discomfort, and absence from school, work, or the social setting</td>
</tr>
</tbody>
</table>

DISCUSSION

The research on involvement of patient representatives at the negotiating table shows that patients are mainly consulted on an individual and ad hoc basis in the Netherlands. Advising by patients and cooperating with patients as a collective group is coincidental at best.
There is little awareness among active patient representatives of the few existing appraisal criteria from the patient’s point of view. This requires – on first analysis – better information and communication. The few currently available criteria are found to be unclear and unpractical, however. Hence, patient participation is not effectively supported, and non-indigenous patients with language issues cannot even use the criteria. The patient’s voice is therefore not sufficiently heard in the healthcare quality and health research areas. So, more effective tools for appraisal by patients are also required, the proposed concept for a criteria list being one of these. To better equip patients in their role as a negotiating party, it is desirable to arrive at a more refined and recognizable list of patient criteria for quality, health policy, and research. The initial list of patient criteria as developed in this study comprises three main sections: relevance, right to have a say, and ethics and safety. This list needs further refinement and validation in future studies. The list may be compared to internationally available patient criteria for health research such as the IAPO toolkit for patient organizations (IAPO, 2008). Besides addressing patient safety, this includes criteria for patients subjected to medical trials, advocacy, and partnership. After the comparison, the criteria list may be adapted and re-scoped to also envelope specific areas and groups, in close consultation with patients and clients.

Apart from developing criteria, it is also important to increase the skills of patients in addressing their new task. This empowerment is necessary since patients often feel insecure about their contribution and added value, especially when new in their role. Moreover, research and policy are complex domains for the average patient and patient representative. The Dutch organization Tools2use, a foundation for empowerment through expertise of the chronically ill or people with disabilities, works on advice, support, and training of experience experts (Tools2Use, 2012). Similar initiatives in the UK (Involve, NICE) have resulted in guidelines for patient groups and members of the public to appraise research grant applications, healthcare policy, and guidelines on quality of care (Involve, 2009) (NICE, 2009) (Involve, 2006). Describing examples that illustrate the problems can be of help to inform patients and other parties and thereby contribute to patient empowerment.

Finally, it is not just a sound democratic principle to use patient criteria so the voice of patients is heard loud and clear. Patient groups have a profound knowledge of their own situation and have no other interest than in defending but their own the quality of life of patients. This constitutes an argument to employ the patient group itself in the negotiation process rather than having outside organizations act on behalf of patients, as these tend to mix their own interests with those of patients.

**CONCLUSION**

Between the ideal of patient groups as equal partners at the negotiating table and the actual reality, there still is a wide gap in the Netherlands. Although patients increasingly attempt to negotiate health with other stakeholders, in practice virtually no systematic assessment method from the patient’s perspective is
available. The few criteria that exist for patients are poorly operationalized and abstract, and it is unclear what significance they have. They are therefore difficult to apply and offer little sense of direction to the patient representative or council.

To satisfy the expectations of patients, more support and greater effort are required. The gap can be reduced by systematic development and validation of patient criteria, increasing the skills of patients via increased awareness and training, and building on successes such as good examples of cooperation. Patients need a set of criteria that are recognizable, workable, and complete. By developing and validating criteria systematically with patients, the gap between current practice and their desired role as an equal negotiation party is being reduced. An instrument, such as a criteria list, makes negotiation more doable for patients and it enhances the quality of the outcome of their participation.

The proposed concept of a patients’ criteria list is expected to be of practical use to many patients in many countries.

Acknowledgements and conflicts of interest
Drs. G.J. Teunissen conducted the research study and wrote the article. Prof. Dr. T.A. Abma, authority in the field of patient participation, acted as guarantor for the scientific content of the article.

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Truus Teunissen
Material: bronze
4 - INVENTORY OF ISSUES THAT MATTER FROM LITERATURE

This chapter contains the work published in the following article:

ABSTRACT

Aim The purpose of this review is to generate an inventory of issues that matter from a patient perspective in health research and quality of care. From these issues, criteria will be elicited to support patient(s) (groups) in their role as advisor or advocate when appraising health research, health policy and quality of health care.

Background Literature shows that patients are beginning to develop their own voice and agendas with issues in order to be prepared for the collaboration with professionals. Yet, patient issues have not been investigated systematically. This review addresses what patients find important and help to derive patient criteria for appraising research and quality of care.

Methods/search strategy Information was gathered from Western countries with similar economic, societal and health-care situations. We searched (from January 2000 to March 2010) for primary sources, secondary sources and tertiary sources; non-scientific publications were also included.

Results The international inventory of issues that were defined by patients is covering a large array of domains. In total, 35 issue clusters further referred to as criteria were found ranging from dignity to cost effectiveness and family involvement. Issues from a patient perspective reveal patient values and appear to be adding to professional issues.

Conclusions Patient issues cover a broad domain, including fundamental values, quality of life, quality of care and personal development. Quite a few issues do not find its reflection in the scientific literature in spite of their clear and obvious appearance from tertiary sources. This may indicate a gap between the scientific research community and patient networks.
**INTRODUCTION**

In most Western countries, patient participation is increasingly acknowledged and accepted. Patients are involved in health-care services (Santvoort, 2009) health-care quality, such as the development of guidelines (Broerse et al, 2009) (Stewart & Oliver, 2008) and health-care research, such as agenda setting (Abma, 2005) (Abma, 2006) (Abma et al, 2009) (Abma & Broerse, 2010) (Herxheimer, 1988) or in studies concerning juridical and ethical aspects of the position of patients in research. The level of participation differs according to the context and can be assessed by the ‘participation ladder’ model (Arnstein, 1969) (Herxheimer, 1988) (Bovenkamp et al, 2010). In this review, we refer to ‘patient groups’, indicating the patient collective rather than individual patients. Patient representatives, patient organizations or patient advocates are all acting on behalf of a ‘patient group’.

Although the aim of participation is to make patient organizations an equal party in health-care decision making, this goal is not reached in practice (Bovenkamp et al, 2010). There is a lack of formal knowledge among patients when negotiating with well trained professionals. Other limitations relate to politicized and asymmetrical contexts where it is difficult for patients to become an equal partner in morally sensitive and strategic issues (Abma & Broerse, 2010).

Empowerment of patient groups and consumers is therefore a recurring issue in the literature on patient involvement Nierse and Abma (2011). show that ‘enclave deliberation’ among groups with converging interests is a necessary step towards development of a political voice, especially when it concerns vulnerable groups. A process of appreciation and raising awareness is required to develop a shared agenda, and only thereafter, negotiations with professionals are feasible (Baur et al, 2010a). Oliver et al (2004) concluded that successful involvement requires appropriate skills, resources and time and provides consumers with information, resources and support to empower them in key roles for consulting their peers and prioritizing topics.

In attempts to answer the question how the dialogue between patients, researchers and health-care professionals can be improved, quite a few studies focused primarily on the methodology and process: they describe what conditions are required and how these can be created (Abma, 2005) (Caron-Flinterman et al, 2005) (Baur et al, 2010) (Schipper et al, 2010). In a systematic review that investigates best ways of involving consumers in health-care decisions at population level, Nilsen et al (2006) distinguishes two basic forms of generating patient issues. Patient issues can be achieved either through consultations or through collaborative processes. Consultations can be single events, or repeated events, either on a
large or on a small scale (Nilsen et al, 2006). Consultation happens on an individual or group level to stimulate a dialogue. The dialogue model for research agenda setting developed by Abma and Broerse (2010), which is based on interactive policy models and responsive evaluation, combines consultation and collaboration. The purpose of this review is an inventory of issues that matter to patients before they start negotiating with professionals about health-care research and quality of care. Its added value lies in the fairly wide international coverage and in the comprehensive number of key issues it identifies, compared to specific studies. This review also aims to contribute to the political power of patients but concentrates mainly on issues of content in an attempt to make an inventory of the issues patients bring forward when negotiating with professionals about health research and quality of care. The review intends to derive a set of patient issues that reveal the patient perspective and can be used to develop ‘criteria’ for appraisal of health research and quality of care activities and policy. Patients experience specific challenges when participating in these processes, because there is no appraisal tool from a patient perspective. At the same time, the number of scientific studies and non-scientific projects wherein patients raise their issues increases gradually. From these studies and projects, issues can be identified that were raised by patient and patient representatives when they responded to health research, quality of care activities and policy. We assume that in general, these issues differ from the issues raised by health-care professionals and researchers as patient issues originate from life world experiences and experiences are colouring one’s world view and values (Nierse & Abma, 2011) (Caron-Flinterman et al, 2005).

**METHOD**

This inventory and synthesis of data started from a focused and selective review of scientific literature using guidance provided by the Centre for Reviews and Dissemination (2008). Soon, however, it became clear that issues from a patients perspective are not only mentioned in scientific literature but more so in a variety of other information sources (Teunissen & Abma, 2010). Therefore, the authors agreed to conduct a data synthesis as described by the Joanna Briggs Institute (2008). A data synthesis has the aim to assemble conclusions, to categorize these into groups on the basis of similarity in meaning and next to aggregate these to generate a set of statements that represent the aggregation (Joanna Briggs Institute, 2008). The issues found are extracted as full text parts, tabulated and finally clustered to descriptive themes, in this study referred to as ‘clusters of issues’, based on similarity of meaning.

**Inclusion/exclusion criteria**

In this review, inclusion criteria are used to focus on patient issues in state-of-the-art health-care systems in Western countries with a similar socio-economic situation and health-care level. Inclusion and exclusion criteria are set on population, language, geographical area, quality of information sources, keywords and search strategy. The inventory and synthesis uses English and Dutch language sources only. In this review,
people can have multiple roles, such as advocate, adviser or provider of information. In this review, we focus on patients as advisor or advocate. They may also be healthcare consumers. In our point of view, patients, as advocates, speak on behalf of the patient group and their organization. The patient organizations have collected data about issues, claims and concerns in an early stage from individual patients/users within the health-care system. Professionals are excluded.

Integration of primary, secondary and tertiary sources
We focus on empirical scientific studies with a method section (primary sources) and other highly relevant scientific studies or articles, either with or without a method section (secondary sources). The limited quantity of available primary and secondary sources necessitates the use of a tertiary source group: non-scientific publications, reports and patient information databases. The authors assumed that this indicates that patient group issues might not be sufficiently explored on a scientific basis. Hence, we included these three types of sources because of their special interest for the main research question and aim of the study. Tertiary sources, originated by patient groups, are assumed to reflect genuine patient issues rather than issues attributed to them by, e.g. social scientists. This further defines the special character of this study: the integration between primary, secondary and tertiary sources.

Search terms
The search strings we use consist of terms being used to describe the role of patients when it comes to their involvement in quality of care and health research. Where in some countries the term ‘participation’ is being used either for ‘right to say’ or for ‘taking part in society’ as opposed to social exclusion, the European and North American literature uses ‘empowerment’ and ‘involvement’ in relation to ‘patient rights’ and ‘decision making’.

Our central search string is: patient involvement. Terms used in conjunction are the following: public, patient advisors, expert patient, patient participation, criteria, peer reviewers, client councils, research clinic guidelines and agenda setting. Furthermore, the –currently fashionable– terms: patient rights, patient advisor, patient empowerment and patient centeredness are used to verify completeness of the search.

The search strategy to locate primary, secondary and tertiary information sources comprises use of the electronic databases Cochrane Library, Pubmed (Medline), Cinahl, Dipex, Patient and Public Involvement Programme (PPIP) and James Lind Alliance sources. Furthermore, the search includes patient organization information exclusively published on the internet and reference list tracking on author, conducted by keywords or by implied content. The search is conducted in information published within the time period between 2000 and March 2010. Patient groups were critical about their influence in research and health care and wrote about this also before this time period. Literature search further back in time would distort the image of ‘current’ patient issues however. The search was therefore limited this time period.
The identification of relevant articles and publications took several steps. The first database search in PubMed and Cochrane Library on our central search string (patient involvement) on the complete text of articles provided us with more than 6000 hits in PubMed and >1000 in Cochrane. When we limited our string to ‘public patient involvement’, it resulted in less hits (e.g. about 650 hits in PubMed). Searches in conjunction with other terms mentioned above (e.g. patients advisor) provided us with less hits of which a selection has been made via screening of titles, abstracts and keywords and a screening of a selection of full texts. Some 301 articles were found by database search and a further 48 by reference tracking and internet search, resulting in 349 sources in total. No comparable synthesis was found in the Cochrane Library.

After a check of the quality of the research methods, duplicates and articles without a method section were eliminated as a primary source. We decided to eliminate articles that were not providing information from a collective patient perspective but, for example, from a professional point of view. The sources were then allocated to one of three categories: primary – empirical studies having a method section; secondary – other highly relevant scientific studies; and tertiary – reports and publications that originate from patient organizations and governmental institutions. Two sources originated outside the time interval, but we included them for special interest: Herxheimer (1988) and Lithuania (1996). Next, we included a publication of the WHO (2009e).

![Flowchart](image)

**Figure 1** Flowchart indicating the results of the search and data synthesis.
This source indicates that emphasis is primarily concentrated on development of tools for advocacy such as ‘position statements’, ‘fact sheets’ and ‘example letters’. These tools enable patients or citizens to discuss disease-specific health-related matters in lay language with those in charge and professionally involved. (Fig. 1).

RESULTS
Of the 349 sources identified, some 296 sources were deemed not relevant to this study as they have no bearing on collective issues of patients. The selected 53 sources do contain a variety of information on issues that matter and their context from a collective patient perspective. Below, we review relevant sources, we summarize the extracted issues from these sources and we analyse them in depth.


The search further resulted in 39 tertiary information sources originating from patient organizations, government institutions and private organizations. Tertiary sources on both the quality of care and the research domain issues are WHO (2008a), AF(2009) and EPF(2009a).


Other tertiary sources are relevant to this study although they are not providing issues from a patient perspective: Vilans (2009), WHO (2009e), LHSC (2009), PatientView (2008) and Hjertqvist (2009).

Quality of care
Herxheimer et al (2000) introduce a database of UK patients experiences called The Database of Personal Experiences of Health and Illness (DIPEx). One of its purposes is to identify ‘questions that matter’ for people who are ill and their families when dealing with investigations, prognosis, lifestyle and treatment choices. Four main issues are identified: (i) finding information when confronted with a new diagnosis or choice, (ii) how to discuss difficult subjects related to a disease, (iii) positive experience stories at times when
negative stories are highlighted by the media, and (iv) stories reinforcing solidarity with others.

Bal & Lindeloof (2005) analyse the policymaking process around the allocation of limited health-care system budgets in different countries: USA, Canada, Sweden, UK, New Zealand and Israel. They mention patient criteria being used in Oregon (USA) and report the use of a set of 13 criteria from patient perspective. Among these were ‘quality of life’, ‘prevention’ and ‘cost effectiveness’. According to their study, Canada shows a variety of ‘patient involvement’ methods between provinces. Sweden organized a discussion in society around three ethical principles and their priority that became part of the current law: (i) ‘Human dignity’, (ii) ‘Need for care and solidarity’ and (iii) ‘Cost effectiveness’. In the UK scientific and social value, judgments on policy have separate paths. Social judgments, based on ‘standards, values and preferences prevalent in society’, come from an –ideally– representative Citizens Council. Both types of judgments are used by the National Institute for Health and Clinical Excellence (NICE) to evaluate healthcare, guidelines and research. Next, Bal & Lindeloof (2005) describe that in New Zealand, the National Advisory Committee on Health and Disability (NHC) uses questionnaires in an evaluation by stakeholders in the report or proposal submission process and health-care priority trade-off studies. Four criteria are being used: ‘efficacy’, ‘efficiency’, ‘fair distribution’ and ‘consistency with social values’. In the Health Parliament in Israel, citizens deliberate on ‘ethical and cost issues’ related to health-care services. Grit et al (2008) stipulate the specific needs of foreign patients using health-care systems in two countries. Lindert et al (2000) investigate the needs of the four biggest non-indigenous groups in the Netherlands resulting in some fifty important issues. This list contains many issues that would normally be considered equally relevant by any patient. However, for this study, it provides three points of specific interest for the foreign patients group: first, the complexity of also ‘receiving treatment and prescription drugs in another country’; secondly, the need for ‘medical information in another language’, both verbally and in writing; and thirdly, there are ‘cultural issues, e.g. a preference for a female doctor’.

Differences between countries and health-care systems result in differences in issues that patients value of importance. Teunissen & Abma (2010) point out that immigrants are using both the Dutch health-care system and systems in other countries. Uiters et al (2006) identify ‘compliance with prescribed medication in the Moroccan and Turkish ethnic groups [in the Netherlands] as non-optimal’. From this, we elicit the issue: intercultural sensitivity. The private and public supported publishers of PatientView (2008) present 172 entries in their European Patient Groups directory ‘with an interest in some element of health advocacy’. Three organizations were found to list issues relevant to this study in their publications: HFE (2007), EPECS (2007) and EIWH (2006). These mention a wide variety of issues, dealing with information, quality, self-care and intercultural sensitivity problems. Santvoort (2009) investigates the relation between policy and disability in nine European countries and how this translates into participation in society and subjective well-being. Key policy issues for people with a disability are ‘coherence in legislation and adequate budget’ for implementation of countermeasures. A risk is also identified: new
‘fragmentation owing to increased autonomy’ of the local communities in adopting their own policy on execution of health-care activities. Schalock & Alonso (2002) describe the individual perception of quality of life. Their inventory of different ways to express, measure and describe quality of life in English-speaking countries highlights commonly felt aspects such as ‘well-being, social inclusion, freedom of choice, positive self-image, future perspective, opportunities for self-expression’. Their model is being used in the Netherlands among Disabled Care Institutions according to Vilans.nl(2009). The Schalock & Alonso (2002) model mentions aspects in relation to quality of life: ‘happiness, lifestyle, physical, psychological and social impairment, living conditions in institutions, family contacts’.

Hjertqvist (2009) provides a series of source document references on European country level when it comes to ‘patient empowerment’. Lithuania, Hungary, Belgium, Estonia, Poland, Slovakia, Germany and the Netherlands were further investigated because these show the highest rankings in the EPEI (European Patients Empowerment Index). The wide variety of issues found comprises e.g. access, choice, information, consent, complaint, medical file, privacy and damage compensation.


The Canadian Association of Genetic Counsellors compiled a directory of support groups on a wide variety of – in some cases rare – genetic diseases. The London Health Sciences Centre (LHSC, 2009) publishes this directory on the internet. The wide variety of diseases, each organized in separate patient groups, did not result in a common set of patient perspective issues. To explore patient issue diversity, we decided to further investigate four groups: heart diseases, alcoholism, diabetes and the rare neurodegenerative genetic disease Tay–Sachs. A large number of issues were found in HSF (2009), Al-Anon (2009), CDA (2009) and Catsad (2009), varying from privacy and information needs to access and information requirements to the health-care institutions.

Voice4Patients.com (2010) presents on the internet links within the USA to some 75 disease-specific patient support groups. To identify the commonality of issues shared between large patient groups, the internet information of four groups known to represent diseases with large number of patients were explored. Publications covering arthritis, lung, Alzheimer and liver diseases were searched. The main issues, found in AF (2009), ALA
Table 1 Sources for key CCI’s / issues in quality of care (A) and research (B) domains

<table>
<thead>
<tr>
<th>Key issue</th>
<th>A</th>
<th>B</th>
<th>A</th>
<th>B</th>
</tr>
</thead>
<tbody>
<tr>
<td>access</td>
<td>43,51,20,48,53,40</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>age</td>
<td>35,52</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>alternatives</td>
<td>56</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>buildings</td>
<td>28</td>
<td>41,55,40,54</td>
<td></td>
<td></td>
</tr>
<tr>
<td>choice</td>
<td>28</td>
<td>20,36,37,38,39,48,54</td>
<td>8</td>
<td></td>
</tr>
<tr>
<td>communication</td>
<td>35,38,31</td>
<td></td>
<td>31,59</td>
<td></td>
</tr>
<tr>
<td>compensation</td>
<td>20,39</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>complaints</td>
<td>32,20,36,37,38</td>
<td></td>
<td>31</td>
<td></td>
</tr>
<tr>
<td>consent</td>
<td>20,37,38,50</td>
<td></td>
<td>31,58</td>
<td></td>
</tr>
<tr>
<td>cost</td>
<td>23,27</td>
<td>50,31,33</td>
<td>23,3</td>
<td>31,59,56,60,61</td>
</tr>
<tr>
<td>cross border</td>
<td>24,25,26</td>
<td>29,51,48,53</td>
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</tr>
<tr>
<td>dignity</td>
<td>23</td>
<td>36,37,50,31,40</td>
<td></td>
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<td>disability</td>
<td>28</td>
<td>35,49,50,31</td>
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<tr>
<td>diversity</td>
<td>24,25</td>
<td>35,45,49,31</td>
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<td>57,56</td>
</tr>
<tr>
<td>education</td>
<td>35,44,40</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>effectiveness</td>
<td>23</td>
<td>50</td>
<td>23,3</td>
<td>31,59,56,60,61</td>
</tr>
<tr>
<td>empowerment</td>
<td>23</td>
<td>34,35,43,32,38,50,31,48</td>
<td>8</td>
<td>57,59,56,58,60,61</td>
</tr>
<tr>
<td>ethics</td>
<td>23</td>
<td>38,50</td>
<td>23</td>
<td>31,60</td>
</tr>
<tr>
<td>family, friends</td>
<td>28</td>
<td>44,47,36,50,48,53,40,54</td>
<td></td>
<td></td>
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<tr>
<td>fear</td>
<td>48,53</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>gender</td>
<td>35,52,49</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>health system</td>
<td>23,1</td>
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<td>23</td>
<td></td>
</tr>
<tr>
<td>information</td>
<td>24,22,25</td>
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<td>8</td>
<td>31,58</td>
</tr>
<tr>
<td>lifestyle</td>
<td>23,27</td>
<td>35,43,41,30,46,33</td>
<td></td>
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<tr>
<td>medical file</td>
<td>27</td>
<td>32,20,36,37,39,50</td>
<td>8</td>
<td>31</td>
</tr>
<tr>
<td>method</td>
<td>29,41,54</td>
<td></td>
<td>30,31,56,60</td>
<td></td>
</tr>
<tr>
<td>pain</td>
<td>30,32,37</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>privacy</td>
<td>35,42,20,36,37,39,50,55</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>quality</td>
<td>43,51,37,38,39</td>
<td>3</td>
<td>31,60</td>
<td></td>
</tr>
<tr>
<td>quality of life</td>
<td>23</td>
<td>36,53,55,40,54</td>
<td></td>
<td></td>
</tr>
<tr>
<td>relevance</td>
<td>30,46,47,50</td>
<td>8</td>
<td>57,31,59,60</td>
<td></td>
</tr>
<tr>
<td>safety</td>
<td>27</td>
<td>52,48</td>
<td></td>
<td></td>
</tr>
<tr>
<td>self care</td>
<td>28</td>
<td>50,40</td>
<td></td>
<td>31</td>
</tr>
<tr>
<td>social security</td>
<td>23</td>
<td>48,53</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

KEY to table 1

1 Santvoort, 2009
2 Stewart & Oliver, 2008
3 Herxheimer, 1988
4 Nierse & Abma, 2011
5 20 (Lithuania, 1996)
7 23 Bal & Lindeloof (2005)
11 27 HFE(2007)
13 29 WHO (2008a)
14 30 AF(2009)
15 31 EPF(2009a)32 PA (2009)
16 33 Patient UK (2009)
17 34 WHO (2007b)
18 35 WHO (2001c)
20 37 Belgium (2002/2004)
22 39 Deutschland (2002)
24 41 HSF(2009)
25 42 Al-Anon (2009)
26 43 CDA (2009)
27 44 Catsad (2009)
28 45 ALA (2004)
29 46 NIA (2009) 47
30 48 IAPO (2007)
31 49 EPF (2009b)
32 50 EPF (2009c)
33 51 EPECS (2007)
34 52 EIWH (2006)
35 53 Picker (2009)
36 54 Planetree (2009)
37 55 Shaller (2007)
38 56 CC (2009)
39 57 WHO (2008d)
40 58 Kelson (2009)
41 59 IAPO (2008)
42 60 Involve (2009)
43 61 JLA (2009)
(2004), NIA (2009) and ALF (2009), are related to safety, lifestyle and the health system. WHO (2008a), IAPO (2007), EPF (2009b), EPF (2009c), Picker (2009), Planetree (2009) and Shaller (2007) provide a wide variety of issues from patients in health-care institutions. Most frequently mentioned issues relate to information and to contacts with family and friends. (Table 1)

Health research
Herxheimer (1988) lists the six rights of patients in clinical research as used in the Primary healthcare department at the University of Oxford, UK: (i) ‘Know what his/her rights are, (ii) The right to adequate information, (iii) The right not to be worried or hurt by information, (iv) The right to withdraw from trial, (v) The right on confidentiality, (vi) Post-study results should be communicated to patient or next of kin’. These can be translated into the following issues: information, choice, ethics and privacy.

Nierse et al (2007) conducted a research project where patients and their organization engaged in a dialogue with researchers about an agenda for scientific research. In this study, patients ‘asked attention for the daily, short term problems outside the medical realm’ (ibid). In another study, Nierse and Abma (2011) ranked discrimination and friendship as top priorities for research. Bal & Lindeloof (2005) address both quality of care and health research. Main issues are related to cost, ethics, values and the health system. Stewart and Oliver (2008) conducted a literature survey, on behalf of James Lind Alliance JLA, to assess patient experience input in setting research priorities. This literature survey resulted in patients contributing in various ways in 43 of the 258 Cochrane library studies explored. This group of studies addressed ‘services’, ‘interventions’ and ‘health conditions’ as issues of importance.


Critical analysis
The objective of this study is to identify the international usage of collective patient issues in order to develop criteria for appraisal. The 357 extracted issue texts found in the search were tabulated and – based on the available information from the various primary, secondary and tertiary sources – assigned to either the quality of care (Q) or research (R) domain. In seven exceptional cases, an extracted issue text had to be allocated to both domains on a fifty-to-fifty percentage basis. Each of the 357 issues was subsequently allocated to a geographical area and to a specific disease, as applicable, both based on the contents of the source document.

This study looks for non-specific patient criteria applicable to a wide variety of diseases and a large geographical area. For each issue related to a single country (or a smaller geographical area) or related to a single disease / impairment (rather than multiple diseases or impairments), markers for verification purpose were set.
The first observation is that many of the 357 issues are almost identical in their linguistic meaning or show significant overlap. This calls for clustering in order to find key issues. These key issues are the starting point for defining patient ‘criteria’ in the future of our research. The clustering process begins with the first issue text extract. Any overlapping other issue texts are searched for and a common denominator is defined. This results in the first cluster. All other issue texts are processed in a similar manner until all texts have been allocated to a cluster and all clusters together constitute an envelope around the content of all issue texts, being defined by detailed cluster descriptions. Accordingly, each of the 357 issue texts was assigned to one of in total 35 clusters based on equality, similarity and linguistic best match. Then, the total frequency of occurrence per cluster was counted by simply adding up the number of extracted texts allocated to each individual cluster. These clusters are presented in table 2. The descriptions provide detail on cluster attributes found in the extracted issues.

Table 2 Patient criteria found in international literature

<table>
<thead>
<tr>
<th>Nr</th>
<th>Criteria</th>
<th>Description</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>information</td>
<td>Information about disease, medicines, treatment, positive and negative experiences, difficulties and project results in simple, jargon free, own language</td>
</tr>
<tr>
<td>2</td>
<td>health system</td>
<td>Health system provides medical advice when needed, a suitable range of therapies, coordinated, integrated and continuous care, assigns adequate means and enough professional care providing staff, arranges transport, nutrition and prevention activities.</td>
</tr>
<tr>
<td>3</td>
<td>empowerment</td>
<td>Patients are involved and/or represented in health care policy, quality and research and have a say in how the providers and health authorities are held accountable. The patients voice differs from professionals' voice. Patients have an independent and equal say in priority setting and appraisal.</td>
</tr>
<tr>
<td>4</td>
<td>safety</td>
<td>Approved, tested, appropriate, hygienic and safe methods, medication and equipment are used while providing care and/or conducting clinical trials. Risks are identified and explained. Continuous and responsible care and follow-up are provided. Availability of experimental drugs after trial is known.</td>
</tr>
<tr>
<td>5</td>
<td>lifestyle</td>
<td>The patients lifestyle, weight control, physical exercise and addiction aspects are taken into account</td>
</tr>
<tr>
<td>6</td>
<td>choice</td>
<td>Patients choose doctor, nurse, treatment and institution. Patients may withdraw from treatment or trial, leave an institution and have self-determination up to the end of their life.</td>
</tr>
<tr>
<td>7</td>
<td>effectiveness</td>
<td>Medical intervention outcomes for patients are positive, effective, are beneficial to- or an improvement of -the patient groups health and well-being as experienced in daily life and are well balanced against negative effects. Equity.</td>
</tr>
<tr>
<td>8</td>
<td>quality of life</td>
<td>Quality of life experienced while staying in health care institutions is ensured by comfort, human contacts, nutritional and nurturing food, opportunities for self expression, arts, culture and entertainment, spirituality and religious services and enhancing each individuals life journey.</td>
</tr>
<tr>
<td>9</td>
<td>method</td>
<td>The best methods, technologies, therapies and techniques are used. Innovation, early diagnosis and prevention are of prime importance. Researchers are skilled and experienced, use the best international evidence to conduct trials. Peer review of experiment design.</td>
</tr>
<tr>
<td>10</td>
<td>cost</td>
<td>Cost is in balance with the value of the outcome. Patients are informed about funding, about cost for their participation, about financial support and about cost reimbursement. Duplication of resources is avoided.</td>
</tr>
<tr>
<td>11</td>
<td>disability</td>
<td>Disabilities of patients are taken into account in health care provided. This includes disfigurement, reduced performance, requiring assistance, physical fitness, health condition, the severity of impairment either physical or psychological. Mental/intellectual capacity and transportation needs.</td>
</tr>
<tr>
<td>12</td>
<td>medical file</td>
<td>Medical records are confidential, secure, accurate and accessible for patients.</td>
</tr>
</tbody>
</table>
The second observation is that a significant part of the clusters is not unique to a single domain. Figure 2 shows the clusters, listed in count frequency ranking order, distributed over the domains quality of care (Q) and research (R). This demonstrates that 18 of the 35 clusters are associated with both Q and R domains. These are empowerment, information, safety, health system, cost, choice, effectiveness, method, diversity, medical file, quality, ethics, complaints, consent, social security, communication, lifestyle and values. The Q domain contains 33 of the 35 clusters. The R domain contains 20 of the 35 clusters, so in total 15 clusters are not found to be associated with R. In contrast, only two clusters are unique to the R domain: relevance and alternatives.

The third observation concerns the insensitivity of this analysis for disease- and geographical area–specific issues. The 357 issues are found to be 73.7% quality of care-
oriented and 26.3% research-oriented. Of 357 issues analysed, 39 are disease specific. In total, 180 of the 357 issues are originating from various single countries. This raises the question whether clusters have common ground for use by a wide variety of patient groups. When two or more single disease–originated issues support a cluster, the cluster itself is not disease specific. The same applies to single country–originated issues. All 35 clusters pass these two checks. Addition of any further single disease– or single country–originated issue to the 357 issues would not be likely to necessitate addition of a new cluster to the 35 clusters. This implies data saturation within the search limitations set for this review.

Figure 3 illustrates the clusters non-specificity for single disease / impairment and single country.
The fourth observation is an underrepresentation of key issues in the primary and secondary sources. There is a striking lack of presence of high frequency Q and R key issues in the scientific sources information. The clusters: empowerment, safety, lifestyle and choice are found in relatively small proportion compared to their presence in tertiary information sources. Some 13 clusters are not found in scientific sources, in spite of their clear and obvious appearance from tertiary sources. These are privacy, relevance, access, complaints, consent, communication, education, gender, pain, age, compensation, fear and alternatives. This may indicate a gap in, or rather a lack of presence of, scientific research activity in a significant part of the field of patient involvement.

Figure 3 Criteria and their non-specificity for disease and country
The fifth observation is that a substantial number of key issues extend beyond the biomedical realm of health research and health-care institutions. Some eight of the 35 clusters identified in this study qualify as mostly society- or well-being-oriented issues. These are quality of life, family/friends, lifestyle, diversity, fear, dignity, self-care and social security. The other 27 clusters deal with the relevance of treatment or research, the role and right to say of patients and ethics/safety issues.

**DISCUSSION**

This study has some limitations. It focusses solely on Western, mostly English and Dutch language countries. Further research on this could amend our results. This is not a conventional systematic review, as mentioned in the methods paragraph, but focuses on secondary and tertiary sources as well. We include secondary and tertiary sources in order to study experiential knowledge on patient issues. Without the use of experiential knowledge, found predominantly in secondary and tertiary sources, we would not have been able to elicit the issues as described. Thus, what could explain this underrepresentation gap between issues found in primary and secondary scientific sources and tertiary sources?

First, patient groups appear to be fighting predominantly on an issue level for better health care and research performance. Patient groups often use fact sheets and standard letters to equip their representatives and advocates for negotiations. Although many issues can be derived from the sources found, patient organizations have not synthesized these and translated them into, e.g. a systematic appraisal method or preset levels of acceptance per issue. This may explain why only some relevant information was found in primary sources. Secondly, agenda of patients are often characterized by a broad range of subjects whereas agendas of professionals are more focused on specific areas. Researchers tend to focus primarily on physical functioning and medical issues like effectiveness of medical interventions or improving diagnostic possibilities. Patients, however, mention a plethora of issues, including daily, also work-related problems, quality of life, emotions (fear and anxiety) and issues concerning the relationship with health-care professionals (Baur et al, 2010a).

This attention for a broad context on patient issues relates with a need for a more integrated perspective on health and illness and an integral vision of how health care should be organized. This perspective includes more existential issues as well as psychological, social, spiritual and cultural issues when looking at well-being in addition to illness (Dahlberg et al, 2009) (Visse et al, 2010). From this perspective, issues such as ‘vitality’ and ‘movement’, ‘being able to’, ‘freedom’ and ‘peace’ could be of importance.

**CONCLUSION**

This article describes the first data inventory and synthesis conducted on key patient issues in health research and quality of health care in Western countries. Patients are beginning
to develop their own voice and agendas. This is done not only to enhance collaboration with professionals but also to empower the patient groups and it raises their awareness of issues, concerns and claims, their autonomy and self-support. Often, they are involved in the appraisal of research, quality of care and policy on health care, but without a clear and systematic view on issues that matter from a collective point of view.

We conclude that the primary sources that resulted from our search seem to focus on the biomedical and methodological aspects of patient issues. Therefore, we searched for issues from experiences of patients in secondary and tertiary sources that cover a much wider range of experiential knowledge. This experiential knowledge includes issues originating from fundamental values (relevance, right to say and safety), quality of care and society and wellbeing-related values, e.g. quality of life, lifestyle and psychological and social impairment. Most of these issues, especially the issues related to daily life, do not find any reflection in the primary, scientific literature. Williamson (2010) addresses patient activist issues from an emancipation point of view and presents 10 mainly UK-oriented ‘principles’.

Our review differs in that it collects issues international, includes chronic illness, disabilities and mental illness and considers both the health care and the health research domains. We gathered patient issues that matter before entering any negotiation. Therefore, the ‘criteria’ we set out to find extend beyond these ‘principles’.

The key patient issues found, appear to be interlinked among the two domains quality of care and health research. They are uniquely associated with neither specific diseases nor geographical areas, nor – for a significant part – with the separate domains. Patient organizations cannot always cope with the participation possibilities attributed to them (Nierse & Abma, 2011). They do not have sufficient tools to be a professional partner in dialogue with health-care professionals and researchers. Being invited to participate, does not automatically mean, one is genuinely included as a partner in appraising research and quality of care. Asymmetric power relations may hinder that (Baur et al, 2010b).

Specific inclusion strategies should be developed (Elberse et al, 2010). This article is a first step towards a better equipment of patient representatives. One possibility is to provide them with an appraisal tool. Available tools are reported to be poorly operationalized, to be incomplete and to have unclear boundaries and overlaps (Teunissen & Abma, 2010). In order to support patients when appraising quality of health care, research activities and policy, we intend to take a next step and create a generalized appraisal tool: a patient ‘criteria’ list.

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Truus Teunissen
Material: bronze
5 - STRUCTURED APPRAISAL AND EVALUATION IN PRACTICE

This chapter contains the work published in the following article:


This article originated from two article concepts. The first concept was about patient issues in practice as derived from interviews. It was submitted to Health Affairs, Health Policy, INTQHC, PEC, QIX, QoLR and QHR and was rejected, reviewed and commented several times but not accepted.

The second concept article was about structuring patient appraisal using the issues found in practice from a series of meetings and focus groups. This second article concept has not yet been submitted.

The original plan to publish in two stages had to be abandoned and the two concepts were merged into one article presenting both stages:

1-gathering issues that matter to patients in the practice of patient involvement from interviews and 2-establishing a structure and sequence in this content using a series of meetings and focus groups.
Structuring patient advocates’ appraisal and evaluation of health research and quality of care.

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\textbf{ABSTRACT}

\textbf{Background:} In the Western world governments and the public at large acknowledge the importance of a strong patient group as a collective partner. Patients are increasingly involved in the appraisal, evaluation and decision-making concerning health research projects and quality of care guidelines.

\textbf{Objective:} Previous work suggests that not all ‘issues that matter’ to patient groups in practice are addressed in scientific studies. This study was designed to obtain a valid inventory of patient issues in order to close this gap.

\textbf{Methods:} Patient organisations were approached and interviews were held with their patient advocates. The issues inventory was then structured into a patient criteria list via open coding, clustering and data synthesis. The criteria list was evaluated and adapted based on priority, sequence and usability from a patient perspective in a series of focusgroup meetings.

\textbf{Results:} Six common patient appraisal criteria in health care and health research were identified: (1) Relevance, (2) Quality of Life, (3) Quality of Care, (4) Ethics and Safety, (5) Information & Communication, and (6) Involvement. The criteria “Quality of Life” and “Information & Communication” are presently being ignored in scientific studies. Only after basic values, such as respect, being recognized and acknowledged, and being accepted, are assured does a dialogue about relevant aspects for Quality of Life become possible. Participants in this study expect that the criteria will increase patient advocates’ awareness of the issues that matter and that using the patient criteria list will make their appraisals more complete, better structured, and more efficient.

\textbf{Conclusion:} Patient criteria for appraisal and evaluation of health research and quality of care, have a priority structure, starting from Fundamental values, then proceeding to
Quality of life, continuing via Quality of care and, finally reaching Right of say. This structured set of patient criteria is expected to be of practical use, primarily to patient representatives but also to health researchers and other professionals.

Keywords
Appraisal, empowerment, patient involvement, decision-making, quality of life, patient-preference, issues, value, social inclusion, human dignity.

INTRODUCTION
Patients and their organizations desire to have a voice in the debate on health related subjects and want to have a say in the decision making process on health care and health research (Teunissen & Abma, 2010). Increasingly, patients take part in the debate on behalf of their patient-, disability- or elderly organization or client group. A patients’ advocacy role is related to the provision of information to large groups of fellow patients and also to take the patient issues into the debate. In research and health care policy patient advocacy has gained a broad acceptance (Involve, 2009) (Kelson, 2010) (Wersch & Eccles, 2001). Despite relatively few studies that do not ascribe positive effects of patient involvement, it is generally accepted that patient advocacy groups are considered as important stakeholders in the improvement of health care research and policies (Nierse & Abma, 2011) (Abma & Broerse, 2010) (O’Donnel & Entwisle, 2004) (Nilsen et al, 2006) (Royle & Oliver, 2004). They provide researchers and policymakers with an insiders’ perspective on their disease and on the meaning of living with a disease, and with important themes that research and policy should address. In that way patient involvement contributes to the quality of decision making processes and health practice. It enhances the content, processes and outcome of health care and increases legitimacy of decisions taken (Epstein, 2008) (Baker, 2007) (Boote et al, 2002) (Caron-Flinterman, 2005) (WHO, 2007).

The international literature describes several ways in which patients participate in research and quality of care. For example the inclusion of the patients as partners in guideline-development or research agenda setting (Abma & Broerse, 2010) (O’Donnel & Entwisle, 2004) (Krahn & Naglie, 2008). Mostly, when participating in research and policy, patient organisations work by persuasion and exert influence via patient representatives (Williamson, 2010). Patient representatives face several challenges when they participate in an advisory board or committee among professionals. It is hard for them to articulate their voice and viewpoints towards professionals due to asymmetric power relations (Witte et al, 2011). Often committee’s use a voting system that undermines the patient’s perspective (Broerse et al, 2010). Furthermore, like anyone starting a new job who has doubts about what they can contribute, may not be clear about informal rules, and are not familiar with technical terms, inclusion of the patient’s perspective as a committee member is often hindered (Hewlett et al, 2006). In order to enable patient involvement specific conditions are necessary. Occasionally, special procedures are designed to guarantee the genuine inclusion of patient perspectives in research (Elberse et al, 2011).
representatives need to know about values, needs and issues of patients in their group in order to identify relevant opinions among their own constituents, gather and oversee experience data and formulate a related patient vision (Schipper, 2012). Patient groups therefore, not only struggle with the question whether they are perceived as a fully equal participant in the decision making process, but also with how to usefully perform their role as a knowledgeable party. Given these challenges, patients lack instruments that will strengthen their role and improve their working relationships with professionals (Wit et al, 2011).

An explorative study in the Netherlands indicated that patient groups are being asked to participate more often, but that a knowledge gap remains between professionals’ issues and those introduced by patient representatives (Teunissen & Abma, 2010). A review of international scientific literature on patients issues was carried out in an attempt to complete an “issues inventory” (Teunissen et al, 2011). This revealed that such issues are better reflected in publications from government institutions and patient organizations than in scientific studies. It also demonstrated that the issues covered in the scientific literature do not accurately reflect what patients consider important in everyday practice (Teunissen & Abma, 2010) (Teunissen et al, 2011).

In earlier studies only a relatively small difference in ‘issues that matter’ to patients was found between the health research and quality of care domains. Apart from disease specific medical details, also little difference was found in patient issues between countries and in comparing different diseases (Teunissen et al, 2011). The issues that matter to patients can be compared to the theory of human needs by Maslow. He defined several needs levels in his ‘pyramid of needs’. The lowest need level comprises physiological and survival needs such as hunger and thirst. Higher need levels include belonging and love, self-esteem, and self-actualization (Maslow, 1970). Intertwined with these human needs are the values that drive human behaviour. Patients’ issues are thus directly related to human needs and values. In order for patient representatives to be effective in their role, they must have an opportunity to provide insight about these issues that really matter to patients. This leads to the following research questions:

- What are the issues and values that matter to patients in quality of care and health research?
- How can these issues and values be prioritized and structured into criteria for general use?

**METHOD**

This qualitative study comprises the inventory of issues that matter to patients to enhance our understanding of these phenomena (Tong et al, 2007). This was achieved via interviews with patient advocates, analysis and condensation of these issues into a criteria list and establishing a values based structure in these criteria by means of a series of focus group meetings.
Defining issues and values
The term ‘issues’ is used in this study for all claims, concerns and issues patients might have concerning their disease and its consequences (Meyer, 2000). The term ‘value’ reflects in this study the ideals and for rules for good conduct. A classification of values with the central value ‘wisdom’, surrounded by some 43 other values, segmented into groups, called ‘dimensions’ was developed around 1990 by Schwartz (Schwartz, 1992).

Inclusion/exclusion
Respondents for interviews and focusgroups were selected from four categories: 1- chronic illness, 2-disabled, 3-elderly and 4-healthcare client councils. We included respondents with one or more chronic illnesses or disabilities who are members of organizations or councils dedicated to their illness or disability. (Table 1) Respondents all spoke on behalf of their organization at the negotiation table, participated themselves in debates and were providing advice on research and quality of care. Mentally ill or mentally disabled people, e.g. with dementia, were indirectly included via spokespersons. All personal identifiers have been removed or anonymized.

Sample
This study was carried out in The Netherlands. Currently, there are an estimated 200 patient-, disabled- and elderly organizations and 560 client councils in institutionalised health care (CG-raad, 2010). A sample of 16 interview respondents representing all of the four different organisation types was considered to be sufficient to provide an initial indication of relevant issues in the practice of patient and client involvement (Guest et al, 2006). The respondents represent 14 different organizations of patients, disabled and elderly people. (See table 1)
A further 52 respondents participated in three preparatory meetings and a series of three focusgroups, converting the initial issues inventory into a condensed and structured set of criteria. (See table 2)
After each focusgroup meeting, an analysis was performed. The criterion for data saturation was that the last focus group meeting did not add any new insights. After three focusgroups this criterion was met.

Interview method
This study followed a responsive approach (Meyer, 2000) (Abma & Widdershoven, 2006) (Guba & Lincoln, 1989), in which the researcher acts as a facilitator to map stakeholders’ points of view. Issues were gathered using two types of interviews: semi-structured, in-depth face-to-face interviews and semi-structured telephone interviews for several chronically ill participants. Interviews typically lasted one hour face-to-face and 30 minutes by phone. The first two authors conducted the interviews (Britten, 1995). The transcripts were sent back to the respondents for review.
### Table 1  Interview respondents

<table>
<thead>
<tr>
<th>Nr</th>
<th>Date</th>
<th>Organisation of patient/client organisation</th>
<th>Organis - sation</th>
<th>Interview type</th>
<th>Male/female</th>
<th>Age</th>
<th>Active advocacy years</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>03022010</td>
<td>Organisation of Elderly Migrants (NOOM)</td>
<td>1</td>
<td>Face to face</td>
<td>male &amp; female</td>
<td>31-50, 65+</td>
<td>5</td>
</tr>
<tr>
<td>2</td>
<td>01062010</td>
<td>Client Association for Mental Health</td>
<td>2</td>
<td>Face to face</td>
<td>male</td>
<td>31-50</td>
<td>10</td>
</tr>
<tr>
<td>3</td>
<td>22032010</td>
<td>Dutch Organisation of Blind and visually impaired people (NVBS)</td>
<td>3</td>
<td>Face to face</td>
<td>male</td>
<td>31-50</td>
<td>25</td>
</tr>
<tr>
<td>4</td>
<td>12082010</td>
<td>Elderly Organisation (SBO-NH)</td>
<td>4</td>
<td>Face to face</td>
<td>male</td>
<td>65+</td>
<td>10</td>
</tr>
<tr>
<td>5</td>
<td>18022010</td>
<td>Organisation of Migrants with Cancer (SAK)</td>
<td>5</td>
<td>Face to face</td>
<td>male</td>
<td>31-50</td>
<td>5</td>
</tr>
<tr>
<td>6</td>
<td>20072009</td>
<td>Organisation for Patients with Headache (NVvHP)</td>
<td>6</td>
<td>By phone</td>
<td>female</td>
<td>50-65</td>
<td>20</td>
</tr>
<tr>
<td>7</td>
<td>07072009</td>
<td>Parkinson Patient Organisation</td>
<td>7</td>
<td>By phone</td>
<td>male</td>
<td>50-65</td>
<td>10</td>
</tr>
<tr>
<td>8</td>
<td>27072009</td>
<td>Parkinson Patient Organisation</td>
<td>7</td>
<td>By phone</td>
<td>male</td>
<td>50-65</td>
<td>10</td>
</tr>
<tr>
<td>9</td>
<td>01072019</td>
<td>Platform Patient and Industry (Platform PI)</td>
<td>8</td>
<td>By phone</td>
<td>male</td>
<td>5</td>
<td></td>
</tr>
<tr>
<td>10</td>
<td>07092009</td>
<td>Lung Patient Organisation</td>
<td>9</td>
<td>By phone</td>
<td>female</td>
<td>50-65</td>
<td>10</td>
</tr>
<tr>
<td>11</td>
<td>14072009</td>
<td>Retinal Patient Organisation</td>
<td>10</td>
<td>By phone</td>
<td>male</td>
<td>50-65</td>
<td>20</td>
</tr>
<tr>
<td>12</td>
<td>22072009</td>
<td>Platform Clients in Mental Healthcare (LPGGz)</td>
<td>11</td>
<td>By phone</td>
<td>male</td>
<td>50-65</td>
<td>10</td>
</tr>
<tr>
<td>13</td>
<td>23072009</td>
<td>Rheumatism Patient Association</td>
<td>12</td>
<td>By phone</td>
<td>male</td>
<td>31-50</td>
<td>10</td>
</tr>
<tr>
<td>14</td>
<td>20082009</td>
<td>Organisation for Patients with Backproblems (NVRWervelkolom)</td>
<td>13</td>
<td>By phone</td>
<td>female</td>
<td>31-50</td>
<td>20</td>
</tr>
<tr>
<td>15</td>
<td>15072009</td>
<td>Lung Patient Organisation</td>
<td>9</td>
<td>By phone</td>
<td>female</td>
<td>18-30</td>
<td>10</td>
</tr>
<tr>
<td>16</td>
<td>19122009</td>
<td>Glaucoma Patient Organisation</td>
<td>14</td>
<td>By phone</td>
<td>female</td>
<td>30-50</td>
<td>10</td>
</tr>
</tbody>
</table>

### Table 2  Focusgroups respondents

<table>
<thead>
<tr>
<th>Date</th>
<th>Participants</th>
<th>Gender</th>
<th>Age</th>
<th>Location</th>
<th>Subject</th>
</tr>
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<tr>
<td>Preparatory meetings (18 different participants in total)</td>
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<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>PM-1</td>
<td>07-09-2009</td>
<td>14</td>
<td>F 8</td>
<td>Leusden</td>
<td>Patient Issues and values in research</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>M 4</td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td>25-75</td>
</tr>
<tr>
<td>PM-2</td>
<td>13-4-2010</td>
<td>10</td>
<td>F 6</td>
<td>Leusden</td>
<td>Patient Issues and values in quality of health guidelines</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>M 4</td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td>25-75</td>
</tr>
<tr>
<td>PM-3</td>
<td>25-10-2011</td>
<td>11</td>
<td>F 6</td>
<td>Amersfoort</td>
<td>Patient Issues and values in research</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>M 5</td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td>18-75</td>
</tr>
<tr>
<td>Focus groups (34 different participants in total)</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>FM-1</td>
<td>14-4-2011</td>
<td>15</td>
<td>M 4</td>
<td>Utrecht</td>
<td>Patients issues and values in quality of care guidelines</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>F 11</td>
<td></td>
<td></td>
</tr>
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<td></td>
<td></td>
<td></td>
<td></td>
<td>28-68</td>
</tr>
<tr>
<td>FM-2</td>
<td>23-8-2011</td>
<td>7</td>
<td>F 7</td>
<td>The Hague</td>
<td>Patients issues and values in quality of care/guidelines</td>
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<tr>
<td></td>
<td></td>
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<td></td>
<td>25-55</td>
</tr>
<tr>
<td>FM-3</td>
<td>25-11-2011</td>
<td>12</td>
<td>M 7</td>
<td>Utrecht</td>
<td>Patients issues and values in research</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>F 5</td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td>28-55</td>
</tr>
</tbody>
</table>

**Focusgroup method**

Three preparatory meetings with patient advocates groups (in total 18 participants) were held by the first author. The issue clusters from the interviews were reviewed and
participants were asked to express their views on issues, on linking them to criteria and to propose descriptions. The preparatory meeting participants were also invited to submit written remarks or suggestions.

The resulting concept criteria list was used for a series of focusgroup meetings, conducted in accordance with the Helsinki Declaration (WMA, 2008) and guidelines (EC, 2005) (CIOMS, 2002). Only new participants, 34 in total, were admitted for each new focusgroup. At least one representative from each of the four group categories was present in each meeting. Each focusgroup lasted about 2 to 3 hours and was conducted by the first two authors. Participants were first asked to review the inventory of issues, as condensed by the researchers into a concept criteria list, and associate these with values listed in the Schwartz classification (Schwartz, 1992). Secondly, participants were asked to apply the concept criteria in two example cases (a research proposal and a care guideline development). Subsequently, subgroups were asked to inform the other groups whether they could benefit from the criteria in practice; whether they recognized them; whether they had suggestions for any additional issues and values or whether the concept criteria should be renamed; and whether they agreed with the sequence, priorities, and clustering the researchers had proposed. The researcher facilitated dialogue and asked clarifying questions. All focus groups were recorded and transcribed. Afterwards, a report of the process and outcome of the focus group was written and sent to the participants for review.

Interview data analysis method
We used a thematic approach to analyse our data recorded in interview transcripts, using a content analysis focused on issues of patients (Glaser & Strauss, 1967). This analysis was done according to a combination of techniques: ‘thematic analysis (Thomas & Harden, 2008), ‘meta aggregation’ and ‘data synthesis’ (Lockwood, 2008). Data gathering was conducted by manual extraction. Records were kept of the details associated with the extracted text parts and their source. After five transcripts, no new issue clusters emerged in the following transcripts, implying data saturation.

Credibility and reliability
Credibility ‘refers to the extent to which conclusions and inferences make sense to the people in the context studied’ (Greene, 2011). Safeguarding the credibility of our study also entailed conducting member-checks after each interview and focus group meeting. The combination of methods (triangulation) added to the breadth of the study and helped to verify data. The research team discussed whether or not saturation was reached, and the process of data collection was ended when repetition of findings was found. To ensure inter-rater reliability the first and second authors compared each others notes and recordings after the interviews and focusgroup meetings. Only consistent and common observations were used in transcripts, member checks and further analysis.
RESULTS

Patients’ issues from involvement practice

The transcripts of face-to-face interviews were analysed first. Subsequently, the transcripts of telephone interviews were analysed. The first transcript was searched for issues encountered while participating in health care quality and health research. Some 420 issue text parts containing reference to a patient issue were selected and coded. Logical relations between the issue text parts were found. For example “gender” and “diversity” are logically related because gender is an element to be considered within diversity. Such logical relations suggest that several issue texts may be combined into issue clusters. In order to cluster closely related or common issues in an inductive way, the coded text parts, representing all issues and activities mentioned in the first transcript, were subjected to open coding (Berg & Lune, 2011). The resulting issue clusters were used as a start, and led to additional clusters or to subdividing previously defined clusters. Coding of each next transcript started with the accumulated issue clusters from the preceding transcripts. In total 33 issue clusters were derived.

Patient criteria

Next we looked at ways to condense these issue clusters into groups. We used three steps in this analysis:

1) First a detailed comparison was done between 35 issue clusters found in our previous literature study (Teunissen et al., 2011) and the 33 practice issue clusters found in this study. We observed that, in the literature, several “quality of life” and “fundamental human values” issues were not mentioned (share experience, recognition, independent voice, trust, be taken seriously, social participation, support, vulnerability, identity preserved). The authors believe the results from practice reflect the importance of these issues for patients.

2) Then we looked at logical relations. In our previous exploratory study on patient issues in the Netherlands, three key terms were found: ‘Relevance’, ‘Right to say’ and ‘Ethics and Safety’ (Teunissen & Abma, 2010). The key term ‘Right to say’ has a logical relation with ‘Empowerment’, hence they were joined under the more general term ‘Involvement’. The range of ‘fundamental human values’ issues is fully covered by the ‘Ethics and Safety’ and ‘Quality of life’ key terms. Issues about information to patients and communication with patients were both allocated to the key term ‘Information & Communication’. In this way we arrived at a core of six issue cluster groups, further called criteria.

3) Finally we observed during the interviews that patients preferred a sequential order between specific kinds of issues within these clusters, which we think requires further research.
Establishing the criteria sequence
In order to validate the six criteria and to establish their relation and structure, homogenous and heterogenous (in terms of diseases) patient meetings were held. First, three preparatory meetings were held within a homogenous group of 18 participants, all members of the lung patient advocates group at the Lung Fund Netherlands (LFN, formerly called Astma Fonds). In each meeting between 10 and 14 lung patients expressed their views on the above proposed six groups of issues clusters into patient criteria. They could change, add and re-sequence them. This resulted in: ‘Relevance’, ‘Quality of life’, ‘Quality of care’, ‘Ethics and Safety’, ‘Information & Communication’, and ‘Involvement’. In the debate during these meetings it became clear that the issue content itself was adequate but within the contents of the six criteria a new, value related, sequential structure emerged. Participants were invited to reflect on this, verify the criteria description content and submit written comments about their view on criteria, values and this sequential structure.

Validate the criteria list contents
Next, three heterogeneous focusgroup meetings, were held, each time with different participants. A total of 34 respondents participated in these meetings. They reviewed the concept ‘six criteria list’, leading to the following observations:
- No new issue clusters emerged. This validates the issues content of the six criteria as obtained via the interviews among patient advocates and the preparatory meetings.
- The descriptions of the six criteria were elaborated and questions were formulated to operationalize them as an appraisal tool.
- The ‘Involvement’ criterion was felt to be both a precondition and an appraisal criterion.

Establishing a sequential structure
Then participants were asked to assess which patient values are involved in the appraisal of research proposals and quality of health care from a patient perspective. Starting with 14 values listed from the preparatory meetings and written responses received afterwards, no new values emerged in the first focusgroup meeting. The second focusgroup meeting identified one new value, namely ‘accessibility’. In the third focusgroup meeting no new values or other insights emerged.
Subsequently, participants were asked to describe the barriers they encountered during their involvement efforts. Their first concern was ‘attitude’ as an external factor limiting them in their activities in society (WHO, 2001). Basic human values, like ‘being respected’, ‘recognized’ and ‘accepted’ and ‘not being excluded’, primarily determine the way participants think and act. (Step I) ‘Trust’ and feeling ‘safe’ in spite of their ‘vulnerability’ are important. Patients in their diversity want to do their ‘own appraisal’ and expect to be treated ‘equal’ and with ‘fairness’ and ‘dignity’. Hence, the more effectively fundamental human values are addressed the more the “quality of life” issues can become a concern.
Physical, social, and mental functioning constitutes a key factor in respondents’ lives. This means ‘well being’, ‘belonging’, ‘live a fulfilling life’, ‘self-actualization’, ‘compassion’,
‘freedom’, ‘autonomy’, and ‘self care’ values are in order. They want to ‘be loved’, experience ‘hospitality’, feel good, and have work and a social environment with ‘family and friends’ to experience a good quality of life. (Step II).

After these fundamental and personal values are addressed, other subjects might become an issue. Access, freedom of choice, an agreed upon treatment plan, clear and complete patient information, effective communication on an equal basis between patient and care provider, an up to date medical file, and quick and adequate diagnosis are important when it comes to quality of care (Step III) (Teunissen & Abma, 2010) (Teunissen et al, 2011).

Finally, a patient group has a right to speak when it comes to negotiation, advice, decision making, and guidance in quality of health care and health research matters. They consider “purpose and means,” and “what is right” and “responsible” as key issues. Empowered patients have found the strength and the ways to use experiential knowledge about their disease in an effective way. These are involvement issues (Step IV).

The respondents thus indicated a clear preference to deal with issues in four sequential steps. The six patient criteria, descriptions and the related issue clusters with their sequential step numbers (I to IV) are shown in table 3.

Table 3  Patient appraisal criteria list originating from involvement practice

<table>
<thead>
<tr>
<th>Patient criterion description</th>
<th>Related patient issues*) and step numbers</th>
</tr>
</thead>
<tbody>
<tr>
<td>1   Relevance</td>
<td>I-Diversity (gender, disabilities, education, life-style, social, cultural, ethnic, skills, comorbidity) &lt;br&gt; I-Age &lt;br&gt; IV-Practical relevance to patients &lt;br&gt; IV-Theme choice in research</td>
</tr>
<tr>
<td>2   Quality of life</td>
<td>I-Recognition, being heard &lt;br&gt; I-Trust &lt;br&gt; I-Taken seriously, being respected &lt;br&gt; I-Vulnerability &lt;br&gt; I-Identity preserved &lt;br&gt; I-Privacy &lt;br&gt; II-Best possible life (physical, mental, social) &lt;br&gt; II-Share experience &lt;br&gt; II-Social participation &lt;br&gt; II-Support &lt;br&gt; II-Self care &lt;br&gt; III-Access to facilities and medication &lt;br&gt; III-Care environment hospitality, buildings, medical file &lt;br&gt; IV-Practical relevance to patients</td>
</tr>
<tr>
<td>3   Quality of care</td>
<td>I-Diversity (gender, disabilities, education, life-style, social, cultural, ethnic, skills, comorbidity) &lt;br&gt; I-Age</td>
</tr>
</tbody>
</table>

*) Notes: These criteria and issues are intended to highlight the importance of patient involvement in healthcare decision-making.

Structuring appraisal and evaluation 110
| Perception of a diverse group of patients in their social environment. | I-Independent voice  
II-Family and friends  
II-Support  
III-Care environment, hospitality, buildings, medical file  
III-Cross border care |
|---|---|
| **4** Ethics and safety  
Is the least objectionable way to achieve the goal chosen. Does it comply with regulation and codes of conduct. Are patients protected against harm and damage during and after the medical intervention. Is there freedom of choice and respect for fundamental needs. Are unnecessary repeats avoided. | I-Proper medical care (valid methods & techniques, following codes of conduct, alternative methods, relevance to other diseases)  
I-Diversity (gender, disabilities, education, life-style, social, cultural, ethnic, skills, comorbidity)  
I-Dignity  
I-Social security  
I-Fear & Pain avoidance  
I-Patient values respected  
I-Compensation of costs/damage, complaints procedure, insurance.  
I-Privacy  
III-Patients being informed (correct, complete, informed consent)  
III-Access to facilities and medication  
III-Freedom of choice |
| **5** Information and communication  
Is correct and clear lay information ensured. Are patients made aware of potential risks to allow informed decisions about any medical procedure to be performed on them. Are results shared with patients in lay terms. | III-Patients being informed (correct, complete, informed consent, project results)  
III-Contact with carers (attitude, empathy, language) |
| **6** Involvement (Precondition)  
Are patients interests included, are patients treated as equal, knowledgeable partner, are patients trained, enabled and supported with their involvement activities. | I-Recognition, being heard  
I-Equivalency  
I-Education support, training & facility  
I-Independent voice  
III-Patients being informed (correct, complete, informed consent)  
IV-Advocacy  
IV-Right to say  
IV-Conditions (patient representation)  
IV-Theme choice in research  
IV-Knowledgeable partner |

*) Several issues are related to two or more criteria.
DISCUSSION AND CONCLUSION

Discussion
Because of practical funding and time limitations, some groups were under-represented in our study: patients with mental illness and patients belonging to various immigrant groups. Language and cultural issues, as well as non-membership of patient organizations, resulted in failure to include these groups (Nierse & Abma, 2011).

The differences in issues found between health research and health care domains, between diseases and between countries were small (Teunissen & Abma, 2010) (Teunissen et al, 2011). Hence, the geographical limitation of conducting the study in the Netherlands is expected to have little impact. This qualifies the current study as a first exploratory inventory of issues and values that matter to patients. Further study might provide more insight in, for example, disease specific issues and the need for special criteria lists.

The findings in this study underline the importance of fundamental human values grouped under the criterion 'Quality of life' (WMA, 2008) (EC, 2005) (CIOMS, 2002). Since the application of ethics and human rights in health research is not guaranteed, our findings could support patient organizations who attempt to monitor these important issues.

These patient criteria enable patient organisations to more clearly express concerns and share their experiential knowledge (Tong et al, 2012) (Kuper et al, 2008) with health care- and health research professionals. They may also help increase the skills of patient advocates.

Several European projects within the Innovative Medicines Initiative (IMI) are already (IMI, 2013) using the patient criteria list presented in this study.

Conclusion
This study resulted in a criteria structure linking issues and values that matter to patients. These appraisal and evaluation criteria will help patient advocates to become more aware of their patient groups’ issues, to more effectively engage in collective decision making, and to accomplish appraisal and evaluation in a more complete, structured, and efficient way. Relevance, Quality of Life, Quality of Care, Ethics and Safety, Information & Communication, and Involvement constitute the six generally applicable appraisal criteria from a patient perspective. The preferred sequence to deal with these criteria was determined by patients.

We expect that this instrument will make patient involvement in research projects and development of guidelines more workable and effective. As a result the quality of research and guidelines will improve because the patient perspective is properly included. The proposed criteria list is expected to be of practical use to many patient groups in many countries.
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Truus Teunissen
Material: serpentine
6 - IMPLEMENTING ISSUES AND VALUES OF PEOPLE WITH A CHRONIC LUNG ILLNESS

This chapter contains the work published in the following articles:

The main research effort on implementation of patient involvement presented in this chapter was spent during a 7 years case study at the Lung Foundation Netherlands (LFN), previously called Asthma Foundation.

This work is described in section 6.1 and published as:


Two additional studies were conducted in parallel. Both have particular relevance to the implementation process described in the above article. The first of these two studies is about research agenda setting.

It is described in section 6.2 and published as: 1)


The second of these two studies is about the dialogue on the Chronic Obstructive Pulmonary Disease (COPD). It is described in section 6.3 and published as:


Note:
1) Both “Teunissen G.J.” and “Teunissen T” author names were used in different instances.
6.1-Implementing patient involvement in LFN research

Patient involvement in Lung Foundation research: a seven year longitudinal case study.

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ABSTRACT

Patient involvement in health research is getting more accepted over the years. Until recently scientists and medical professionals were the sole assessors of quality and relevance of research proposals. In the Netherlands, as in other European and North American countries, emancipatory, political and democratic developments stimulated the emergence of patient involvement as a new ‘voice’ in the appraisal of research. A time-series cross sectional longitudinal case study was used to describe and analyse a seven year period since the introduction of the patients’ perspective in the Longfonds research cycle. Longfonds, the Lung Foundation in the Netherlands (LFN) was formerly called “Astma Fonds”. The study was conducted using an actors-interaction model against the background of the dynamics in society. The introduction of patient involvement resulted in a paradigm shift. The scientific and societal relevance of research proposals are now being reviewed by all parties in a more effective and efficient way. Patients, now involved in the review procedure of research funding, are trained and equipped with an appraisal tool for societal relevance from a patients perspective. Scientific relevance and societal relevance are evaluated separately and balanced in the research funding application approval process. Societal relevance is being evaluated by a patient advocates group. The results show how a government initiative and an approach by a patient organisation have led to more patient involvement in lung research. It requires ‘believers’ both to initiate and continue the work and to promote the lessons learned inside and outside the patient organisation. As this depends on devoted individuals, the continuity of patient
involvement remains vulnerable. This seven year study offers valuable insight in patient involvement against the background of the changing health discourse.

**Keywords:** Appraisal, lungpatient, participation, patient involvement, lay expertise, medical research, quality of life, commitment.

**INTRODUCTION**

Scientific medical research has been the exclusive domain of (bio-or para)medical scientists doing scientific research for a long time. Research is often initiated by researchers from a scientific perspective (bottom-up) or by institutes, funding organisations or patient organisations funding research (top-down). Theme choice, appraisal and prioritizing are usually done only from the scientific perspective. In Western societies, a general trend towards more emancipation and exerting democratic rights by citizens with disabilities is being observed (WHO, 2011). Literature search shows that patient involvement at a health fund has thus far not been subject of a long term case study. Several years ago LFN started to look at possibilities for ‘patient involvement’ in theme choice and appraisal in LFN research. Currently, the LFN scientific advisory board (WAC) not only consists of scientists but also has three permanent lung patient members with experiential knowledge (Astma Fonds, 2012). This article reports a study on the introduction and evaluation of patient involvement in the LFN cycle of research funding. The study is carried out against the background of a model of actors and interactions and looks at five transition phases over a seven year period. These phases lead to five comparable situation descriptions over time from evidence gathered. This result is analysed and discussed to assess lessons learned.

**METHOD**

**Design of the study**

An in-depth study of the development, effects and background of patient involvement in the research cycle at LFN, can best be carried out with the case study method, following the basic principles and methodological rigor as laid out by Yin et al (2006). Field-work for data gathering requires a ‘blueprint’ theory and suitable data collection instruments (Yin, 1994). This case is the ‘carrier’ of the subject phenomenon ‘patient involvement’ at LFN (Eisenhardt, 1989).

**Selection of type and instruments**

Eisenhardt (1989) presents a general 7 steps case study approach and underlines the importance of methodological rigor. For this study a variant is used, adapted and regrouped into 4 main steps: design, conduct, analyse and, finally, formulation of the evidence outcome.
Kohn (1997) focuses on case study analysis and links it to the need for a logic model covering process/context system, identification of actors, their roles and behaviour, expected changes, actor strategy and function in the system, driving forces, interactions among actors. The 7 year investigation period necessitates a longitudinal approach rather than a latitudinal one. The main challenge is the thematic analysis over time. Such studies are referred to as ‘time-series cross sectional’ (TSCS). Several discussion points must be noted here, e.g. whether the perspective (or ability) of the researcher is changing (Pettigrew, 1990; Stein et al, 2005).

**Data gathering**
Based on the above considerations, the LFN case study design comprises 5 cross-sections over time. The inductive analysis technique (Bowen, 2006) was used to constitute 7 themes. Each of these situation descriptions addresses the same themes. Collection of evidence took place via:

- Documents (minutes of meeting; interviews; policy reports). Several documents used in this case study were confidential. They are not listed in the references.
- ‘Open’ interviews, (questions like: how, why) In total 10 open interviews were held by the first author with patient representatives participating inside and outside the LFN organisation in the period 2009-2012. Interviews typically lasted 45 min and a topic list was used to ensure theme completeness. A further 4 open interviews with research professionals and external research scientists with a role in the LFN research funding process were held. New interview questions were raised until the themes in the list were all discussed. After each of these 14 interviews a member check was performed.
- Direct observation and observation by participants (in focusgroups). A scientific advisory board meeting with 14 (out of 23) members was held in 2011 while being monitored via direct observation. The board members were research professionals, medical professionals, care providing professionals and two -high educated- patient representatives. The observation by 2 researchers was set up to explore their behaviour in the discussions: topics, tone, respect, formal or informal, interruptions and discussion lines. A series of 4 focusgroups were held in 2009 (20 participants), 2010 (18), 2011(17) and 2012(18) to gather experiences, issues and concerns emerging from the patient involvement activities, based on a topic list. These meetings typically lasted 2 hours and were held at a neutral location. Written summaries were made and member checked with all participants. The participants were highly educated people between 18 and 76 years of age, men and women. All participants have a chronic lung disease, are patient representatives and participate in the LFN research proposals societal relevance appraisal process. Further direct observation by the first author as a policy advisor during 3 appraisal cycles in 2010, 2011 and 2012 added insight into the practical problems of funding proposal appraisal. Participants in interviews and focusgroups were treated according to the declaration of Helsinki (WMA, 2008).
Data analysis
Interview transcripts and notes taken during focusgroups were used to make descriptions at 5 transition phases over time. These time-crossectional situation descriptions have directed content using a 7 themes template. All gathered data were then tabulated in summarized form in a simple transcript matrix with rows according to table 1 and columns according to table 2. The notes made during the direct observations were used for reference in this process. Then 7 histories by theme were composed from this matrix.

Quality criteria
Based on quality considerations like validity and reliability (Devers, 1999) general criticism on the case study method points at sensitivity for ethical problems, mixed interests of the researchers, sensitivity to pre-disposition and uncontrolled change of direction during the study. These dangers were avoided by the above choice of methods, use of well proven techniques and accurate description of the analysis. The first author conducted the research over the entire period.

CASE
The case study requires a model, a ‘blue print’ describing the LFN patient involvement actors and interactions. The actors are the different stakeholders (see figure 1). The model is based on the current LFN research funding process design. The definition of patient involvement as presently used by the Dutch Government determines how we look at patients representatives and their role (Bijsterveldt & Dekker, 2006).

Definition of Patient involvement
About half the people with a chronic disease have some sort of physical disability degrading their self-sufficiency and participation in society. Personal factors (e.g. knowledge and skills) and environmental factors (e.g. access to care) determine the level of problems they experience with this (Ursum et al, 2011). Patients try to arrange their lives around the disease and mainly focus on quality of life, whereas the medical professionals mainly look at diagnosis and treatment possibilities (Teunissen et al, 2011). There are many reasons to increase the patients perspective in the health research cycle: legitimacy (democratic approach), contribution of experiential knowledge, increased likelyhood of implementation in practice and, most importantly, better quality of the decision making and more commitment for the results (Teunissen & Abma, 2010). In this study the following definition of patient involvement is used: (Bijsterveldt & Dekker, 2006)

"The intention of patients and their organisation(s) contributing experiential knowledge is to exert more influence on patient issues, for example in health research. It is expected that usage of patients experience improves the relevance, quality and outcome of research. . . . The level of patients’ involvement may range from test subject via information provider or advisor to equal partner in research or quality of care."
This is based on the notion that patients know better about their interests, needs and issues than the other parties in the health system. Their collective experience constitutes a
unique and specific perspective. So, patient involvement is about shared interests within the entire patient group and not about individual patient-care provider contacts.

**Setting, actors and interactions**

This study investigates the development process of patient involvement in LFN during a seven year period from 2005 till 2012. The LFN organisation is positioned between health care and health research professionals and lung disease patients in the Netherlands. The LFN organisation participates in mixed project teams among scientific institutions, care providers, insurance companies, government, industry and patient organisation representatives. In this study a simplified actors-interactions model of lung disease related health research activities is used with a societal ‘context’ that surrounds the following actors: (see: figure 1)

1-Lungfoundation (LFN).

The LFN organisation consists of two main parts: the LFN Lungfoundation and the LFN lung patients organisation. The LFN organisation funds scientific research for over 50 years, primarily on asthma and COPD but in recent years also on rare lung diseases. The WAC advises the LFN board on quality and priority of research proposals. At present both scientific quality and scientific and societal relevance are being actively considered.

2-Scientific advisory committee LFN (WAC)

Choosing health research funding themes on the ‘scientific research agenda’ has mainly been the responsibility of the members of the WAC for a number of years. The WAC members are professionals in the biomedical, clinical and health care research fields, contributing on basis of their personal expertise. From 2005 patient representatives are present in the WAC too.

3-Health research and health care professionals

The medical research professionals at universities, health institutes, care providers and expert centres submit project proposals after an LFN funding research call for fundamental, clinical and applied research proposals on subjects related to lung diseases and living with a lung disease.

4-Project teams

Mixed professionals project teams are used in various settings in the Netherlands and Europe to carry out research.

5-Patient advocates

Some 20 to 25 patients with a lung disease, capable of forwarding experiential knowledge, inside and outside the LFN organisation, both in the Netherlands and on an international level, are organised in a ‘patient advocates’ group. They contribute with patients involvement activities to the research cycle. They have provided patient issues for the societal research agenda in 2005 and for its update in 2009. In the WAC at first 1, later 3 patients were involved in decision making about research proposal funding.

6-The (lung disease) patient group

The group of 1 million patients with a lung disease in the Netherlands is the source of information for the LFN patient advocates. It is also a main target group, both for the LFN organisation and for the research professionals.
These actors have the following interactions: (see: figure 1)

A – The Health research and health care professionals groups interact with patients in the Netherlands during care and research activities.

B – The LFN organisation interacts with health research professionals via the WAC. This deals with appraisal of research proposals, policy discussions, planning the health research agenda and patients involvement issues.

C – The LFN organisation interacts with project teams on health care and health research, both in the Netherlands and abroad. This is being done by LFN professionals and patient advocates and takes place outside the research funding process.

D – The ‘WAC’ and ‘Patient Advocates’ are part of the LFN organisation which has several interacting internal organs. WAC evaluates and prioritizes scientific research study proposals. The ‘Patients advocates’ transform their experiential knowledge into different products: advocacy statements, processing forms, patient criteria, systematic appraisal of research calls and input to advice by the WAC.

Figure 1. LFN patient involvement actors and interactions model. (See: setting, actors and interactions chapter)
E – The Patients advocates have a range of activities. Besides the product oriented activities they interact with lung patients in the Netherlands to gather experiential knowledge and ‘sense’ the issues that matter among patients. They also defend patients’ needs and interests, both on national and on local level, using data from the annual patient(member) monitoring and other monitoring instruments about chronic lung patients done by external parties.

F – The LFN organisation provides disease related information to lung patients in the Netherlands via publicity, provides support via an information service desk, and has local presence via 22 regional support workers. LFN also raises funds from the general public and receives minor contributions from industry and the government.

The LFN research cycle and patients’ perspective anno 2012
Patients’ perspective is considered to be a crucial element in the entire research cycle according to the management, executive board and member council. Patients donate their experiential knowledge, their time, and their, due to their chronic lung disease, sometimes very limited energy to health research, quality of health care and quality of life. Patients take part in ethics committees, in research and care project groups and in development teams for health care guidance.

The LFN organisation manages the research cycle as a process having several stages. Both scientific and societal aspects are reviewed in a series of stages. The stages and the role of involved patients’ are:

I – Establishing the research agenda. Using interviews and focusgroups the patient group is consulted about issues that matter and about subjects for future research.

II – Reviewing research proposals. Representatives from the patients advocates group evaluate submitted proposals on societal relevance from a patients’ perspective in small groups of three patient advocates. LFN uses a standard review procedure with 5 criteria from patients’ perspective.

III – Prioritizing proposals based on the scores on the 5 criteria. This is carried out within the WAC by patient advocates sharing their experiential knowledge with the other (scientist) members. A general WAC meeting is being convened to review the results and compose the acceptability and priority advice listing to the LFN management.

IV – After funding approval, stage four contains the start and execution of the research projects. Patients’ involvement is not arranged via the LFN organisation in this stage.

V – Monitoring progress and scientific quality of research projects during execution and finalization is evaluated by the LFN research staff and the WAC as well. Evaluation from scientific and societal perspective is prepared by WAC members and LFN research staff. Patient involvement is not present in this stage.

Context and expected changes
In the field of lung disease related research and care activities in Western societies several changes were being observed during this study. In Europe and North America research proposals are no longer only appraised on their scientific quality and relevance. In the UK patients are involved in health care issues via the NHS-Involve for over a decade (Involve,
Emancipatory and democratic developments within groups of people with disabilities brings a new party to the health system negotiating table (WHO, 2011). Instead of just looking at a disease from a medical point of view, the whole patient is currently at the focal point. These issues are especially felt by the growing group of patients with a chronic disease, and get more important in all aspects of their lives than before. Rather than just one, three phases are now to be dealt with in the health system:

i  contracting a disease – requiring treatment, facility and quality of care

ii  keeping the disease – requiring self-management and quality of care

iii learning to live with the disease – requiring social participation and quality of care

Chronic patients build up experiential knowledge over the years, a new type of knowledge, represented by a new party during negotiations in health research and health care. Being new at this however, patients face practical problems affecting their efficient and effective contribution. Their disease makes it in many cases difficult to always attend at meetings. There was no consistency in the quality of their contribution and there was no instrument for appraisal of proposals from patients perspective (Teunissen & Abma, 2010).

Patient needs shift towards quality of life related societal issues, leading to a shift in the research agenda (Caron-Flinterman, 2005). Governments have started to support patients taking control over their own lives by self-management and a stronger advocacy role in the health care system policy, health cost and health insurance debate (Keizer, 2012). Increased individuality and autonomy of people in general also affects (chronic) patients. A newly acquired chronic disease is a dramatic change and it is a daunting task to create a new, equally individual, way of life. The importance of self-management is expected to increase due to increasing numbers of chronic disease patients (Heijmans, 2010). The traditional medical approach via diagnosis and therapy needs to be extended to a partnership between a well informed patient and a care provider.

Governments stimulate patient organisations to participate in the development of guidance and research agenda setting (Involve, 2009). Respondent researchers and care providers did not regard patients as experts since all relevant knowledge is assumed to reside with professionals. Patient groups on the other hand were very positive about being involved. In their view increasingly important chronic illnesses and the trend from ‘only cure’ to ‘also care’ would lead to a new setting: medical professionals ‘coaching’ the patients in deciding and managing quality of life issues via ‘selfmanagement’. Patient respondents also recognize a danger: patient involvement could be abused to achieve the goals of the other parties, leading to pseudo-involvement or ‘placation’.

In spite of countries having different health systems, different legislation regarding health insurance and different ways patients are organised as a collective, issues that matter to patients remain uniform (Teunissen et al, 2011). Efficient and effective involvement via patient advocates is troubled by poor contact with patients in the groups represented. In various countries patients are involved on personal capacity only. In order to be able to gather up to date information in a patient group, the advocates need to be adequately equipped. Due to usage of complicated professional language patient advocates need a large amount of time to evaluate reports and documents (Trappenburg & Bovenkamp,

**Interventions**
During this case study there was one single major intervention: the introduction of patient involvement in the LFN cycle of research funding.

**RESULTS**
In order to allow comparison and analysis a data collection theme list according to table 1 was used. This list has seven aspects and was derived from critical analysis of the patient involvement as it was at LFN before 2005 (Caron-Flinterman et al, 2005). Based upon 5 key changes in policy and practic at LFN 5 time crossectional descriptions were made according to table 2.

Then, using the situation descriptions gathered from documents, interviews and focusgroup observations, the development histories for each of the seven aspects were described including a critical reflection by each of the seven aspects.

Table 1. Template for the LFN research cycle patient involvement case study.

<table>
<thead>
<tr>
<th>Nr</th>
<th>Aspect</th>
<th>Description</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Research basis</td>
<td>What basis initiates and chooses themes for scientific research</td>
</tr>
<tr>
<td>2</td>
<td>Funding criteria</td>
<td>What appraisal criteria are leading in LFN research</td>
</tr>
<tr>
<td>3</td>
<td>Patients say</td>
<td>What is the basis for LFN patients’ say in research</td>
</tr>
<tr>
<td>4</td>
<td>Scope</td>
<td>What is the scope of LFN appraisal of patients’ involvement</td>
</tr>
<tr>
<td>5</td>
<td>Advocacy</td>
<td>How is LFN patient advocacy in research organised</td>
</tr>
<tr>
<td>6</td>
<td>Commitment</td>
<td>What is the level of commitment for LFN patient involvement</td>
</tr>
<tr>
<td>7</td>
<td>Context</td>
<td>What contextual and paradigm considerations play a role</td>
</tr>
</tbody>
</table>

Table 2 Situation descriptions at 5 discrete moments during the study period.

<table>
<thead>
<tr>
<th>Phase</th>
<th>Time cross section</th>
<th>Description</th>
</tr>
</thead>
<tbody>
<tr>
<td>I</td>
<td>Research cycles up to 2005</td>
<td>Introduction societal research agenda, vision and system</td>
</tr>
<tr>
<td>II</td>
<td>2005-2009 research cycles</td>
<td>Update societal agenda</td>
</tr>
<tr>
<td>III</td>
<td>2010 research cycle</td>
<td>Patient expert group</td>
</tr>
<tr>
<td>IV</td>
<td>2011 research cycle</td>
<td>Societal relevance criteria</td>
</tr>
<tr>
<td>V</td>
<td>2012 situation planning</td>
<td>Present situation</td>
</tr>
</tbody>
</table>
Research basis
The scientific research agenda is initiated and composed by scientific and health care professionals in 1999. It is still in use in 2005. In 2004 a scientific study on a societal agenda was started.
As a first change towards patient involvement in the LFN research cycle in 2006 a societal research agenda was composed in cooperation between researchers, health care professionals, patients and other relevant stakeholders for the first time. In 2010, 2011 and 2012 the basis is an updated Societal research agenda, jointly prepared with researchers, care professionals, lung patients and other relevant stakeholders (Elberse et al, 2012).

Critical reflection
At the start of the case study period generally, scientists were suspicious about the added value of patient involvement for a long time. Some of them even considered appraisal by patients as a threat to their position in the research field. Traditionally researchers and professionals minimise the influence by third parties on the contents of their work (Bovenkamp et al, 2008). Researchers often want to protect their power/status and promote the authenticity and supremacy of their knowledge over consumer- or patient lay knowledge (Boote, 2009). Patient involvement brings ‘different’ perspectives to research decision-making spaces, based on what has been referred to as ‘experiential expertise (Thompson et al, 2012). The separation of scientific quality and relevance from societal relevance as introduced by LFN was therefore crucial. Separated appraisal allowed the point of view to change and mutually acknowledge the potential added value of each others’ appraisal. Currently research scientists still not all wholeheartedly commit to it. Since it requires a ‘culture’ change this takes some more time and effort.
It is also crucial for researchers to be aware of other distinctive patient voices rather than the ‘average’ as that is mostly based upon dominant methodological or policy assumptions. Both in general and in this study it is a concern how the vulnerable group within the total patient group can be heard. Diverse patients will express diverse opinions and the patient perspective can become more than an averaged out ‘mainstream-mix’ that can simply be compared to a standard (Barnes & Cotterell, 2012).

Funding criteria
Upto 2009 scientific quality was the leading primary criterion in the research funding process. Scientific and societal relevance were handled as a single secondary criterion. From 2010 scientific and societal relevance were handled as two separate criteria, remaining of secondary importance. This split-up of ‘relevance’ was an important change. In the 2010 LFN research cycle, the patients’ perspective was introduced as a new conditional requirement into the call for proposals. If not properly addressed these additional patient criteria would lead to exclusion from the funding cycle. In 2011 the LFN funding application and appraisal procedure was changed to accommodate patient involvement requirements and to clarify patient criteria to the research community. By 2012 scientific and societal parts had equal weights.
Critical reflection

In theory, societal and scientific relevance criteria are equally important and must be allocated equal weight in health research. In practice however, scientific quality reigns. It is decisive for a project proposal to pass through the first phase, whatever its societal relevance. The question is whether this does justice to all parties involved, especially the patients. Recently, in both the scientific and the societal policy discourse, besides relevance, also working with societal quality equal to scientific quality has become more accepted. The social quality of research, also called valorisation, means making a result of research valuable for society. This more comprehensive research evaluation, including questions about ‘societal quality’ and ‘valorisation’, requires a broad discussion and an approach with a wide participation of disciplines and other stakeholders (Spaapen et al, 2007). Patients are much in favour of the addition of societal relevance as an appraisal criterion but this has no bearing on the commitment for this with other stakeholders in the LFN process. The decision making process in the WAC about the advice to reject or approve and prioritize proposals is carried out by ranking on individual votes against a total funding budget constraint. This means that rejection does not imply the proposal is substandard in all cases.

Patients involvement and patient perspective

From the year 1998 patients were consulted by LFN. In 2004 a Patient Advisory Group was used to assess issues that matter to patients. Patients provided advice when asked and when patients felt advice was necessary and appropriate on a wide range of subjects (Caron-Flinterman et al, 2005). Before 2005 LFN strategy and policy documents did not mention patient involvement. A study on societal agenda setting was published in 2005 (Caron-Flinterman, 2005). Patient involvement was implicitly mentioned in LFN research policy by 2009. Patient involvement was explicitly mentioned in LFN research policy by 2011. Patient involvement is explicitly mentioned both in the LFN strategic plan and in the LFN research policy in 2012.

Critical reflection

Working with budget allocation and dedicated support for patient involvement activities still is in its infancy. Although a specialist policy advisor has been working on this for 3 years and a group of patient advocacy volunteers was built 2 years ago, involvement is still far from being structural and sustainable.

Scope

In 2005 there was only 1 tick-box: ‘is patient involvement sufficiently being addressed?’ on the funding application form. By 2010 a detailed description of how patient perspective is implemented in the proposed research project was required. The Societal relevance requirement contained ‘patient criteria’. Working with patient criteria was a key change in the process (Teunissen & Abma, 2010) (Teunissen et al, 2011). Patients in health care, the test subjects in health research and the research professionals had identified the need for a set of appraisal criteria. In 2011 societal relevance was extended to 5 criteria: ‘relevance’,
'quality of life', 'quality of care', 'information/communication’ and ‘right to say’. A requirement on a lay-summary description with research questions, workplan and schedule, in Dutch was introduced, to assist the appraisal by patient advocates.

**Critical reflection**
Using 5 patient criteria turned out to be very helpful for the patient advocates. Unfortunately the lay-summary in the local and ‘plain’ language still often misses in proposal packages to be appraised. Researchers appear to favour writing in English. Both the right depth in the description of the project contents in the lay-summary, and its readability in the local language, require further attention. It is this lay-summary that avoids social exclusion and enables patient advocates to adequately fulfill their role.

**Advocacy**
Chronic lung patients did not have a group of patient advocates capable of defending their interests on a national and international level before 2005. In 2005 the WAC effectively had one patient member. Between 2005 and 2009 two lung patients participated in the WAC, based on their personal capacity. These two became also member of a newly established group of lung patients with experiential knowledge. This was a key change in the process. The group was advocating on (inter)national level, and was being facilitated by an LFN staff professional from 2010. In 2010 the two patient WAC members had difficulties to consult with the chronic lung patients in the country and getting some more support. The group of lung patients with experiential knowledge, then engaged in appraisal of 71 research proposals, using 3 ‘patient criteria’. This was done in small appraisal teams of minimum 3 expert patients for continuity reasons. Participants were compensated for travel expenses. By 2011 three lung patients participated in the WAC as equal partners. The patient advocates group handled the appraisal of 90 research proposals, on societal relevance using a 5 ‘patient criteria’ list as a tool. Two additional weeks throughput time were allocated to the review process for appraisal of the societal elements in the large quantity of research proposals.

**Critical reflection**
The 3 patient WAC members are still a minority (20%) when it comes to voting on rejection or acceptance. Also the 2 weeks throughput time they originally had for proposal appraisal was rather short. Many of the patient representatives have a job, a household or simply themselves and their disease to manage and have limited capacity to do advocacy work on the side. This was not in line with LFN organisation research application procedures, geared to professionals that appraise project proposals as a part of their day-jobs.

**Commitment**
Patient involvement was not considered to be an important factor before 2005. Commitment for patient involvement was poor with professionals, both inside and outside the LFN up to 2009. Although LFN policy mentioned patient involvement, practice showed very little evidence. The patient involvement in the research cycle was further intensified
every year based on experience and discussions with the patient members in the WAC. There was a fair to adequate level of commitment by 2010. By 2011 commitment was good considering the policy and its implementation in practice. By 2012 patient involvement was widely accepted in the LFN organisation and it was mentioned in the LFN strategy, in the research policy and in the annual budget allocation for training and information exchange. It was implemented in practice via facilitation of the patient advocates group.

**Critical reflection**
The commitment of LFN organization, WAC members and researchers in the funding cycle has increased during the 7 years of this case study. Continuity strongly depends on highly motivated professionals in the WAC, especially the chairmen, and in the LFN organization, so-called ‘believers’ however. The appointment of a new chairperson of the WAC in 2012 will also be crucial.

**Context**
In 2005 patient involvement, equal partnership, full citizenship and own responsibility were emerging phenomena in society. They were being discussed between government, health insurance and health providers as potential subjects for new policy. Between 2005 and 2009 government institution ZonMw started a research programme on patient involvement in health research and quality of care in order to speed up patient involvement and make it more efficient and effective. The LFN organisation was the first health fund to set up a societal research agenda. In 2012 government budget cuts caused the withdrawal of part of the financial support for advocacy to patient organisations. Emphasis was placed on patient organisations’ own responsibility and cooperation with each other.

**Critical reflection**

**ANALYSIS**
The effect of the intervention on the quality, efficiency and effectiveness of patient involvement was derived from observations, interviews and focusgroup findings. An important change was the introduction of a lay language summary in Dutch. An observation during the many appraisals was that proposals that had such summaries, were often also addressing readability of information for test subjects in their project. Although much improvement was observed, research proposals often still lack information about the expected results on short or long term and the impact on the quality of life for the patient group. Equal partnerships include involvement in all research activities from beginning to end. But also a focus on experiential knowledge, mutual learning, openness, and respect are needed. The idea of a mutual learning process helps to close the gap between ideas of
'real value' professional knowledge and 'lesser worth' lay knowledge, it might even overcome these stereotypes. The collaboration can have an extra value for the research process and for those involved (Abma et al, 2009). It may be useful in the nearby future to differentiate the value of participation by the levels of the individual patient, the patient group, the patient association, the researchers and the Health Fund platform (Denis & Teller, 2011). Finally, there might be a beneficial insight for the research community in the Netherlands and abroad. Patient criteria on societal relevance were not seen before in the health research area so explicitly stated as in this case study. A rise in quality level of proposals and implementation of results may be expected since now all parties involved (care- and research professionals and patients) contribute their expert knowledge.

**DISCUSSION**

This study is about getting lung patients involved in the LFN health research funding process. Arguably the observed shift in vision and commitment for patient involvement may depend partly on general societal and political trends and a change of thinking among the general population over time. On the other hand one can say that if LFN had not consciously chosen for an intervention - to implement patient involvement in the research cycle - this would not have translated into a new and better balanced approach in health research funding. Quality improvement was achieved since all parties are now being involved. A critical success factor is well motivated expert patients group. At the same time it is also the main risk factor for the process. A sufficient number of qualified patients, available and able to carry out the work needed for effective patient involvement, is crucial. Patient advocates at LFN do this as a volunteer, often on the side of other activities. The financial support for patient involvement activities is easily prioritized when budget cuts are needed, making it vulnerable.

**Limitations to this study**

There is always the question whether the analysis of qualitative data is sound and meaningful (Kohn, 1997). To this end we gathered, described and analysed in a completely transparent way. The research cycle, subject to intervention in this study, exists not only in the LFN organisation but in many similar patient organisations and fund raising organisations in many countries. Findings might be influenced by changes on different contextual levels (Pettigrew, 1990). On the one hand the perspective of neither the researcher, nor the LFN organisations’ main strategy have changed during this study. As a consequence of the intervention the patient involvement related activities became more structured, more explicit as societal relevance and embedded as a requirement, hence more important. The socio-economic context on the other hand has changed considerably. After three research cycles in the new situation it is highly likely that true effects are observed, not affected by a wrongly chosen start or stop moment in time or duration of observation. The immediate future may be affected by the reduction or even withdrawal of government activities in this field. This may require new ways and new conditions for cooperation with industry. The effects of the new LFN research funding cycle review
procedure require further monitoring to see if patterns evolve in the subjects addressed in proposals approved versus proposals rejected. This monitoring step was not yet implemented at the time the case study was ended in 2012.

**Recommendations**

This study shows that creating commitment among all stakeholders took quite a while. Although expectations are favourable, proving the added value of patient involvement in the research cycle is an important subject for further study. As a result patient organisations will gain specific expertise in acting as a partner in research, and researchers will involve patient representatives or advocates in more or even all aspects of their research. From a patients’ point of view it is difficult to accept that it may happen that a typical ‘patient research subject’ is not approved for funding by LFN. Further pursuit of funding may require new liaisons and cooperation outside the own patient organisation, either on national level or on international level, e.g. in the European Commission environment. Doing this appraisal process properly requires ‘believers’: highly motivated and capable expert patients and dedicated staff for support, as well as researchers. This crucial contribution also identifies a weakness: what happens if they leave? In this case study a practical solution was mentioned: a group of 25 expert patients, working in small teams (3 or more members) dividing the work in many small portions. If one of the small team members cannot make it to a meeting the other team members can still complete the task at hand. The experiences during the period 2007-2012 at LFN have attracted the attention of other patient organisations and health funding organisations. Some are working now on an adapted form for their own use (U-Biopred, 2012) (PGO, 2012). More cooperation between disease specific patient organisations on general aspects like fatigue, work, education access will strengthen the voice of the patient in health research. Research projects should ensure the allocation of a budget to patient involvement. Foundations and government should take up patient involvement in their policies and practice. The knowledge associated with the practice of patient involvement needs to be managed and made accessible.

**CONCLUSION**

Based on this seven years case study we conclude that patients’ involvement in health research is practicable, both in applied research and in fundamental/clinical research. The LFN review procedure was changed and all stakeholders are now participating in its execution. It proved to be workable for patient representatives, provided they can make use of an appraisal criteria instrument. Also they need support by professionals for their training, and for creating and keeping together a peer-group of expert patients. Patient involvement at LFN has become more effective and more efficient. Still patient involvement can be further improved, in process and outcome but also in representing the
'silent voice' among the patients. Patient involvement remains dependent on individuals and 'believers'. Commitment is not yet supported by hard evidence of effectiveness. The vulnerability of individuals in the peer-group of expert patients requires - besides compassion - also management of experiential knowledge and its continuity. It is not realistic to assume that the voices of all kinds of patients are equally well heard, the way patient involvement is currently organized. It is a serious point of concern that the the 'silent voices' of vulnerable patients are not likely to be heard among the patients representatives, although they deserve to be heard in the research process. This seven year study offers valuable insight in patient involvement against the background of the changing health discourse. It is an example with successes, failures and pitfalls during the introduction and implementation of patient involvement in lung disease related health research. There are do’s and don’t’s which can be useful for other patient organisations and fund raising organisations in other countries. Commitment for patient involvement, well equipped patient advocates and systematic gathering of up to date patient group information, are key conditions for structural and sustainable patient involvement. Besides this an allocated budget, suitable tools for proposal evaluation and agreed funding application procedure details about patient involvement and criteria, are necessities in the process. Funding of lung disease related health research may now be more directed towards issues relevant to chronic lung patients than before. Both patient advocates and health research professionals will need to adapt to this shifting balance.

ACKNOWLEDGEMENTS
This case study was made possible thanks to the cooperation between the Lungfoundation (LFN) in Amersfoort and the VU University Medical Center, EMGO+, Dept. Medical Humanities in Amsterdam. The authors of both institutions thank all those involved in the 7 years period for their continued support to the worthy cause of developing patient involvement in health research.

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6.2 – Patient involvement in LFN agenda setting

Patient involvement in agenda setting for respiratory research in the Netherlands

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Letter to the Editors:

Would it be beneficial to actively involve people with a respiratory condition in identifying and setting respiratory research priorities? Research priorities are traditionally set by medical and scientific experts, and it is often argued that this should remain unchanged since it gave rise to the development of high-quality knowledge, medical innovations and the improvement of quality of life. Patients are often considered subjective, knowing little about health research. Furthermore, patient involvement would cost additional time and money (Caron-Flinterman et al, 2007). So why would one consider giving patients a voice in setting research priorities? Different arguments are described in a growing body of literature addressing patient participation. First, there are indications that research priorities from experts differ from those of patients (Entwistle et al, 1998) (Tallon et al, 2000) (Telford et al, 2002). To become more responsive to patients’ needs, it would be vital to involve patients in identifying priorities. Patients’ “experiential knowledge”, can complement scientific or medical knowledge (Tallon et al, 2000) (Lloyd & White, 2011) (Stewart et al, 2011). Secondly, the process itself becomes more democratic (Entwistle et al, 1998) (Lloyd & White, 2011) . Furthermore, patient involvement in decision making can lead to better acceptance of these decisions and outcomes. These arguments have inspired a growing number of funding agencies, including the European Union, to involve patients in research.

A pioneer in involving patients research is the LungFund Netherlands (LFN) organisation, formerly called Netherlands Asthma Foundation. To be more democratic, responsive to
patients’ needs and to improve societal relevance of their research agenda, in 2004 LFN involved, besides scientific and medical experts, people with asthma and/or COPD in setting their research agenda (Caron-Flinterman et al, 2005) (Caron-Flinterman et al, 2006). In a facilitated process, patients articulated needs and prioritised research topics in a well-argued way. Patients did not, as was expected by some experts, just prioritise care research, social research or focus only on their own problems, they also prioritised (bio)medical research topics and thought of future generations (Caron-Flinterman et al, 2005). In 2009, the LFN research agenda was updated and extended, including rare lung diseases: pulmonary fibrosis (IPF), pulmonary arterial hypertension (PAH) and the respiratory aspects of cystic fibrosis (CF) and sarcoidosis. Patients and medical and scientific experts were again involved because new diseases were included for which the patients’ perspectives were not known and perspectives might have shifted. A project team was established, responsible for agenda setting, consisting of two employees of the LFN scientific department, a patient, an external researcher and an external advisor for the participatory process. Based on the “dialogue model”, which describes a methodological design for patient participation in research agenda setting (Abma & Broerse, 2010) a concise process was developed. The process had to meet the following criteria: 1) patients’ and experts’ perspectives should be recognisable in the agenda; 2) participation should be realised on the level of consultation (Cornwall & Jewkes, 1995) whereby patients’ and experts’ input should be equally weighted; 3) the process should not exceed available resources (time, budget); and 4) the agenda should highlight state-of-the-art research.

The agenda-setting process comprised three phases: consultation, prioritisation and integration. Experts and patients were consulted separately. Patients were invited by patient organisations via advertisements on their websites, email and oral invitation by regional consultants. Patients all differed from those involved in 2004 and none was refused. Patients were consulted in three heterogeneous focus groups (n=31) to identify research themes and topics. Issues encountered in daily life due to their diseases were identified. Some issues, e.g. regarding organisation of care and communication, were discarded being beyond the “research” focus. The identified issues were ranked and discussed by patients to gain insight in which issues were considered important and why. Differences and similarities between the people with different respiratory conditions were discussed. A broad range of issues were identified, e.g. the relationship between aetiology and genetic, environmental or lifestyle factors, interaction of multiple drugs and psychosocial effects. Noteworthy was the attention for children with a respiratory condition while children were not consulted. After three focus groups, saturation was reached. Data regarding CF was retrieved from the Dutch Cystic Fibrosis Foundation who had already established their own research agenda in 2007.

Experts were consulted using two routes. Members of the scientific advisory board (SAB) of LFN (n=20) indicated in a survey which topics of the former agenda should be kept, refined, removed or added. Secondly, research topics for rare lung diseases were obtained by seven semi-structured interviews. Respondents were selected on the basis of maximum variation sampling with respect to disciplinary background, disease and affiliation. Important research developments in their field, relevant knowledge gaps and often heard but
unaddressed complaints from patients were discussed. Focus groups and interviews were transcribed and summaries were sent back for member check. Data were analysed to gain insight in important themes and topics according to patients and experts.

A session with SAB resulted in a list of research priorities from experts. For patients, a questionnaire was developed to validate and prioritise the identified 30 research topics, clustered in seven research themes. The link to the questionnaire was sent by email to members of relevant patient organisations. A paper version was provided for those without e-mail. From 201 returned questionnaires, 169 were filled in correctly and analysed using SPSS-10. This resulted in a list of research priorities from patients. No new topics were brought up in the questionnaire. Patients prioritised research on (genetic) origins, improvement of diagnostic tools and co-morbidity highly. Patients prioritised research topics specifically relevant for their condition as well as general topics like disease causes and development. For example, people suffering from IPF prioritised reduction of side-effects of prednisone highly, while people suffering from PAH prioritised the improvement of the method for administering medication.

The project team integrated priorities. On the main research themes there was broad consensus, but priorities differed in the details (table 1). Issues highly prioritised by both patients and experts were taken up. Unique topics of patients or experts, which were highly ranked, were also included in the agenda. The overlap between priorities of experts and patients was substantial with respect to basic, translational and applied care research. For example, both identified genetic and environmental factors as well as early diagnosis as important research topics. This indicates that patients and experts have similar thoughts on which issues are important to address in respiratory research. Differences were also noted. Comorbidities and drug interactions were prioritised by patients (not experts), while smoking interventions were prioritised by experts (not patients). Topics exclusively introduced by patients were side-effects of drugs, unpleasant administering techniques and specific drugs for children. Two research agendas, with considerable overlap, were formulated; one for asthma/COPD (Chronic Obstructive Pulmonary Disease) and one for the defined rare lung diseases (table 1, parts I and II).

Development and execution of the approach was realised in 5 months with limited resources. The approach for updating and extending the research agenda proved useful for involvement of experts and patients and elicit their priorities. According to SAB, the research agendas were feasible, based on patients’ perspectives and state-of-the-art science. Patients were satisfied with this approach and considered focus groups a useful method to gain insight in their experiential knowledge.

Recruitment of patients took place by convenience sampling. This entails the risk of a non-representative (biased) sample. However, we have no indication that there is a serious bias apart from the fact that, in focus groups, children and severely ill patients did not take part. Though, using two approaches (focus groups, questionnaire) increases the opportunity to become involved. The fact that no new topics arrived from the questionnaire indicated that saturation had been achieved. The facilitators were competent, using non-steering inquiry methods and had a neutral position (Abma & Broerse, 2010).
### Table 1 (I) - Themes in the research agenda for asthma and COPD.

<table>
<thead>
<tr>
<th>Development and mechanisms</th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>Genetic etiological factors of asthma and COPD</td>
<td>E/P</td>
</tr>
<tr>
<td>The mechanism of increase or decrease of symptoms of asthma and COPD</td>
<td>E/P</td>
</tr>
<tr>
<td>The processes of asthma and COPD during life and their mechanisms.</td>
<td>E</td>
</tr>
<tr>
<td>Comorbidity/ multimorbidity</td>
<td>P</td>
</tr>
<tr>
<td>Fatigue</td>
<td>P</td>
</tr>
<tr>
<td>Factors (environmental and psychosocial) and lifestyles affecting the development asthma and COPD)</td>
<td>E/P</td>
</tr>
<tr>
<td>Air pollution, climate</td>
<td>E/P</td>
</tr>
<tr>
<td>Fear, depression, physical stress</td>
<td>P</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Care &amp; Treatment</th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>The earliest stages of asthma and COPD and methods to ascertain these stages</td>
<td>E/P</td>
</tr>
<tr>
<td>Possibilities to improve the treatment of asthma and COPD based on individual disease characteristics</td>
<td>P</td>
</tr>
<tr>
<td>Compliance to treatment</td>
<td>E</td>
</tr>
<tr>
<td>Self-management</td>
<td>P</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Prevention</th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>Interventions to prevent of the development or progression of asthma and COPD</td>
<td>E/P</td>
</tr>
<tr>
<td>Physical activity/ physical therapy</td>
<td>E/P</td>
</tr>
<tr>
<td>Human smoking behaviour</td>
<td>E</td>
</tr>
<tr>
<td>Intervention to affect starting or stopping smoking</td>
<td>E</td>
</tr>
</tbody>
</table>

### (II) - Themes in the research agenda for rare lung diseases.

<table>
<thead>
<tr>
<th>Development and mechanisms</th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>Etiological factors</td>
<td>E/P</td>
</tr>
<tr>
<td>Genetic factors</td>
<td>E/P</td>
</tr>
<tr>
<td>The process of chronic lung disease during life and their mechanisms</td>
<td>E/P</td>
</tr>
<tr>
<td>Prognosis</td>
<td>P</td>
</tr>
<tr>
<td>Fatigue</td>
<td>P</td>
</tr>
<tr>
<td>Airway infection in cystic fibrosis</td>
<td>E/P</td>
</tr>
<tr>
<td>Biomarkers for prognoses and progression</td>
<td>E/P</td>
</tr>
<tr>
<td>Factors (environmental and psychosocial) and lifestyles affecting the development of chronic lung diseases</td>
<td>E/P</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Care &amp; Treatment</th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>The earliest stages of lung disease and methods to ascertain these stages</td>
<td>P</td>
</tr>
<tr>
<td>Possibilities to improve the treatment of lung disease</td>
<td>E/P</td>
</tr>
<tr>
<td>Compliance to treatment</td>
<td>E/P</td>
</tr>
<tr>
<td>Target finding for therapeutic interventions</td>
<td>E</td>
</tr>
<tr>
<td>Airway infections in cystic fibrosis</td>
<td>E/P</td>
</tr>
<tr>
<td>Physical activity/ physical therapy</td>
<td>E/P</td>
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</tbody>
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<table>
<thead>
<tr>
<th>Prevention</th>
<th></th>
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<tbody>
<tr>
<td>Prevention of development or progression of chronic lung diseases</td>
<td>E/P</td>
</tr>
</tbody>
</table>

*Note: In the second column it is indicated which stakeholder groups indicated the research topic as priority (E = experts, P = patients)*
Patients identified and prioritised research topics that were previously not the research focus in the Netherlands, such as co-morbidity, fatigue and effects of psychological problems (e.g. stress, depression) on the development of asthma or COPD. The input of patients largely reproduced the previous research agenda from 2004 (Caron-Flinterman et al, 2006) (Abma & Broerse, 2010), though “fatigue” which was newly introduced. This issue is currently getting more attention in several disease domains. It indicates that patients are becoming aware of fatigue as a symptom of their disease.

Although there is much overlap between priorities of experts and patients on broader themes, in details they differ and bring different and challenging perspective and issues to the table. Ideally, a dialogue would have taken place between experts and patients to discuss differences and to increase mutual understanding (Abma & Broerse, 2010). However, different perspectives do not have to be reconciled when the topics are complementary and not contradictory. Nevertheless, if topics are only considered important by patients, not by experts, it entails the risk that experts will not submit research proposals on these topics. This shows once more, the importance to consider patient participation in research agenda setting as a mutual learning process for patients, experts and policymakers.

**Acknowledgment**
The authors are grateful for the willingness of the patients and experts who were involved in the process. Their contribution has been highly valued. Also the collaboration with the Asthma Foundation lung patients association, Dutch Sarcoidosis interest group, Dutch PHA Foundation, Dutch Cystic Fibrosis Foundation and the Dutch Pulmonary Fibrosis organisation was of great value to us.

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6.3 – *Shared responsibilities in COPD practice*

**Dialogue for Air, Air for Dialogue: Towards Shared Responsibilities in COPD Practice**

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**ABSTRACT**

For the past several years patients have been expected to play a key role in their recovery. Self management and disease management have reached a hype status. Considering these recent trends what does this mean for the division of responsibilities between doctors and patients? What kind of role should healthcare providers play? With findings based on a qualitative research project of an innovative practice for people with Chronic Obstructive Pulmonary Disease (COPD) we reflect on these questions. In-depth interviews conducted with people with COPD, physiotherapists and a pulmonologist show that shifting responsibilities require a supportive attitude from healthcare providers and a dialogical communication between patients and professionals. Our findings show more is needed in order to motivate people with COPD to take responsibility and become co-owners in a process of recovery. The case example illustrates that people with COPD need support from fellow patients to learn to accept their disabilities. Awareness that COPD is more than just a lack of air, that mind and body interact, is a first step to investigate other potential problems and to enhance one’s quality of life.

**Keywords:** Autonomy, COPD, Self-management, Mind–body, Deliberation, Dialogue, Interdisciplinary, Responsibility.
INTRODUCTION

Responsibilities between doctors and patients with a chronic disease have dramatically shifted over the past decades (May et al, 2004) (Thorne & Paterson, 1998). Examples are everywhere. Consider an advertisement for healthcare professionals containing a photograph of a mentally disabled boy with Down Syndrome, wearing a suit, sitting behind a large desk in an office (1). The boy is dressed as a director, and the advertisement mentions him as ‘the employer’. The suggestion is clear: this institution pictures their clients as being in control. Historically we are used to seeing doctors as being in charge of the disease; today many responsibilities are transferred to patients/clients. Patients are no longer passive recipients of care, but are perceived as informed, autonomous experts, directing their life and care. While patients are expected to act as consumers making their own decisions, doctors and other healthcare professionals are expected to operate as providers of objective and scientific information (versus judges of the patients’ interests). The shifting of responsibilities are reflected in new organizational arrangements of care. These arrangements, whether called disease management, self-management, integrated care or otherwise, all aim to maximize the autonomy of patients.

The notion of autonomy is widely accepted in our Western culture. It has gained the status of a core principle within the field of bioethics (besides the principles of doing no harm, benefiting and doing justice). In bioethics an autonomous person is defined as someone who is independent of others and free to make his own choices without interference of others (Beauchamp & Childress, 1994). The idea of the patient as consumer is also reflected in healthcare policies and legislation, for instance, in the Dutch Medical Treatment Agreement Act (WGBO). According to this Act healthcare professionals have the duty to inform patients (VWS, 2001) (RGZ, 2003). The trend towards consumerism is not unique for the Netherlands and can also be seen in other Western countries. In the UK, for example, governmental policy documents spanning the last decade clearly envision the patient as a consumer of healthcare (Bissell et al, 2004) (DH, 2001) (Rycroft-Malone et al, 2001) (Tyreman, 2005).

In the field of medical sociology serious doubts have been raised about the consumerist ethos in healthcare. First of all, patients are not always adequately informed and fully aware of the value or relevance of care as a ‘product’ (Badcott, 2005) (Cox et al, 2003) (Rycroft-Malone et al, 2001). Furthermore, unequal power relations should be taken into account. Patients often find it hard to articulate their needs, and many patients feel that their voice is overridden, silenced, or stripped of personal meaning and social context in medical encounters (Barry et al, 2000) (Bissell et al, 2004) (Britten et al, 2000a/2000b) (Cox et al, 2003) (Mckinley & Middleton, 1999) (Rycroft-Malone et al, 2001). Healthcare professionals often lack the required communication skills to elicit patients’ preferences and involve them in treatment decisions (Cox et al, 2003) (Say & Thompson, 2003). Patients may well expect more than just information from their doctor, such as an advice from an expert (Goldsteen et al, 2007) or support and a listening ear to deliberate their situation with an empathic caregiver (Oeseburg & Abma, 2006). If mutual expectations are not
discussed, this may lead to misunderstandings, which ultimately have a negative effect on the quality of care (Abma et al 2009) (Bissell et al, 2004) (McKinley & Middleton, 1999). The aim of this article is to show that the idea of self-management is indeed much more complicated than just giving information to the patient as consumer. It also entails relational, narrative and communicative work since relationships and responsibilities shift. While most studies either promote or critique the concept of self management in chronic care, we aim to examine the possibilities to enrich the notion of self-management in a dialectical circle between practical understandings and theoretical insights from ethics (Widdershoven & Abma, 2007). Using a practical case example, a Dutch centre for people with Chronic Obstructive Pulmonary Disease (COPD), we will investigate the changing division of responsibilities (Walker, 1998). We will argue that this calls for a dialogical approach to healthcare.

METHODOLOGY

The first two authors of this article were asked to evaluate two programs in which healthcare for chronically ill people is improved by care innovations stemming from dialogical interactions in the triad of patient, nurse and doctor. The evaluation would focus on values such as autonomy, self management, enthusiasm and genuine involvement. The researchers were also asked to develop a method that reflects their unique character. The aim was to disseminate guiding values and instructions for actions to other contexts, so that other healthcare professionals and patients could learn to apply them to their situation.

Criteria for selection of the two programs included the level of experience (programs that functioned several years) and quality and robustness (identifiable programs grounded in the values of the movement). Furthermore, the programs had to be developed for different patient groups. The selection of the two programs was negotiated with key stakeholders of the movement, and resulted in the choice of a COPD program and Fibromyalgia program. In this article we focus on one program, the Dutch centre for COPD. This practice is appropriate as a case example for exploring the notion of self management from the perspective of shifting expectations and responsibilities.

The evaluation followed a responsive approach which implies that human beings are considered as active interpreters of their world and that those with a stake in the program (the stakeholders) are regarded as research partners (Abma & Widdershoven, 2005a). The research project wanted to answer several questions. What made the evaluated programs so unique according to stakeholders (patients and professionals)? How could we describe the interaction between the patient and health professional and in what way did this interaction change from the past? Which working routines and underlying values characterized the programs? By answering these questions we aimed to develop transferable working routines grounded in a dialogical view of care and to describe what conditions would be necessary to implement such a working routine in different contexts (e.g. what competencies).
To evaluate the COPD program, we conducted in-depth interviews with relevant stakeholder groups; patients with COPD, medical doctors, nurses, therapists and managers. A criterion for selection of participants was variety: we wanted to gain as many as different experiences with the programs as possible. Furthermore, the participants were sampled according to their (professional of personal) involvement in the program. Everyone we approached was willing to cooperate; we had no negative responses. One time it was difficult to find the correct way of conducting a member-check to validate our analysis of interview data; the patient we interviewed had trouble talking on the phone because he was very short of breath, so we decided to make an extra appointment for a face-to-face meeting to talk about our interpretation of the interview.

In the COPD program interviews were held with two lung physiotherapists (four interviews in total), and a pulmonologist (interviewed twice). Furthermore interviews were conducted with two persons who suffer from COPD. All interviews were tape-recorded and entirely transcribed. We validated the interpretations of the interviews by doing individual member-checks and by following a hermeneutical-dialogical process (Guba & Lincoln, 1989). This meant that the interpretations of earlier interviews were used as input during next interviews to develop ownership and a shared understanding of the program. When we started interviewing the style was open. Because our knowledge of the program grew as we talked to more respondents the interviews gradually became semi-structured; we learned about the issues that mattered and were better equipped to focus on certain topics.

The inductive analysis focused on recurring values, communication styles and relationships, and the data were related to theoretical insights from the field of ethics. The research team paid special attention to differences in opinion and perception between stakeholders; the aim was not to diminish differences, but to make them explicit and facilitate a dialogue. An example of the hermeneutical dialogical process was a series of conversations about taking medicine. Whereas the pulmonologist told that his patients did not experience difficulties in taking medicine, someone with COPD noted that the way the lung specialist explained the use of medication, could be improved. He found the physiotherapists more clear in this regard. In a subsequent conversation with the pulmonologist, we told him the persons’ view, and discussed why this patient experienced shortcomings. Before introducing this experience of the patient at the pulmonologist, we asked the patient for permission. The patient agreed with us talking to the pulmonologist, because he already shared his opinion with the pulmonologist himself. By letting us talk to the pulmonologist again, his viewpoint could be introduced again, with the expectation that the pulmonologist would take action to improve his skills. A final step was a conversation between the medical specialist, respiratory therapists and the person with COPD to see what exactly could be improved.

The positive involvement of all the respondents of the program was striking. The research resulted in a mutual learning process. The health professionals and persons with COPD wrote lengthy remarks in response to our interpretations of the interviews and we had several talks about these interpretations. The actual research report included both programs and was published as a Dutch book (Visse & Abma, 2008).
THE CONTEXT: DUTCH COPD CENTRE

In 2003 a group of Dutch healthcare professionals and people with COPD discussed the need to improve the quality of the communication with each other. The purpose was to create more equal, dialogical relationships in which values as equality and self management played a prominent role. These professionals and people with COPD gained support from various larger organizations like the Royal Dutch Medical Association (KNMG), Dutch Patient and Consumer Federation (NPCF) and the Foundation for Nursing and Care professionals (VV&V). This was the beginning of a movement called Changing by Connecting (Verbindend Vernieuwen). This movement functions as an umbrella for several programs in which healthcare for the chronically ill is improved.

Chronic Obstructive Pulmonary Disease (COPD) is a disease of the lungs in which the airways have become constricted. It is a deficiency in ventilating. People who suffer from COPD have a short of breath and coughing is often a first sign of it. Diseases under the umbrella-term COPD are chronic bronchitis and emphysema. Although (one of) the primary cause(s) is tobacco smoke, according to the World Health Organization, COPD is not simply a ‘smoker’s cough’, but an underdiagnosed, life-threatening lung disease. COPD is not reversible, but it can be managed, controlled and slowed down. Two percent of the Dutch population is diagnosed with COPD and 17 percent of the people above the age of 80 suffer from this chronic disease. Worldwide it is estimated that 210 million people suffer from COPD. This percentage is expected to rise because of pollution, unhealthy diets and physical inactivity (2).

People with COPD are short of breath. In the beginning mostly when being physically active, like cycling, running or doing hard physical labor. After a while, when the disease progresses, simple activities like walking the stairs, getting dressed and doing the dishes will also cause a shortness of breath. This shortness of breath results in increased fatigue and inactivity which have a negative effect on the patients’ mental health also, thus creating a vicious circle. The common way to treat patients with COPD is to provide a mix of medication and physical therapy. COPD is a chronic disease and the development of this disease has to be seen within the historical context of chronic illnesses in general. In the past, it was very common for people with chronic illnesses to take rest and become less active. Nowadays, the effectiveness of rest is questioned and replaced by training and re-activation programs.

In the early 1980s in the Netherlands an alternative COPD program was developed by two lung physiotherapists and a pulmonologist who noticed that a growing amount of patients consulting them returned after a while with the same health concerns. The doctor advised rest, physical therapy and medication, but realized that for some patients, this was not sufficient. A centre for COPD was established. The centre offers a multidisciplinary group-program that consists of physical movement, exchanging experiences with other fellow patients in a group and education about physical and mental health by the use of insights from cognitive behavioral therapy.
The centre is embedded in a network of healthcare institutions, health insurance companies, regional professional associations and the patient association for COPD. Besides providing treatment, the network focuses on the development of guidelines and protocols for treatment. The quality of care is constantly monitored and improved through the exchange of experiences and knowledge. The participants of the network are patients, general practitioners, pulmonologists, (lung) physical therapists, families of the patients, pharmacists, nurses and psychologists. One of the characteristics of the network is its non-hierarchal structure. This means that the patient is really an equal ‘partner in care’, which is unique in the Dutch healthcare system. The participants of the network decided that in order to become an equal partner, being able to participate in a dialogue, the patient needs to be educated in a specific way. On an organizational level, the patient should learn how to participate in the network as a representative of a patient organization and on the individual level he should develop capacities to actively participate during the treatment. The patient needs to be trained, know what he could do when there are physical problems and complaints. In turn, the professionals need to understand and answer the questions of the patient and should be able to enter into a dialogue.

On a yearly basis, the COPD program consists of sixteen groups of eight patients, so in total about a hundred patients per year join the program, of which 75 percent or more suffers from severe COPD. The program is offered when the person with COPD consults the pulmonologist. The pulmonologist informs the patient about the program and together with the lung physiotherapist they decide whether to join the program or not. Most of the times, a referral to the program happens when the patient keeps having problems with accepting his disease, or is having problems with the amount of activities he can undertake during daily life.

The first step in the program is an interview with the lung physiotherapist. Preferably the partner of the patient joins this intake conversation. By using a special anamnesis form, the lung physiotherapist discusses the medical history of the patient, the way the patient and his partner experience the disease and the way it influences daily life. Since COPD is considered not to be just a lack of air, but rather a disability grounded in the life history and daily experiences of the patient, the intake also focuses on the narrative of the patient (Who is the patient? What are important values in life?).

The next step is a test to determine how much strain a patient can endure. With the results, an individual training program is compiled. The actual training is carried out in a group of patients, where the lung physiotherapists supervise the exercises and ask lots of questions during the training. As indicated, the Dutch program tries to make patients more aware of their total state of being and way of living. The activation is dosed; the term ‘graded activity’ is used, which means that the degree of activation starts at a low level, and is slowly progressing to more intensive levels. It is, however, the patient who decides which level is feasible. During the training the physiotherapists ask how the patient experiences the work out. Another underlying notion is that the physical activity needs not to be dependent on the level of pain or fatigue. Yet, it is very important that patients themselves discover how valuable the physical movement is for their total well-being.
The program teaches patients self-management. Patients have to learn to deal with their limitations and they learn this through physical exercise, but also by sharing experiences with a group of fellow patients.

The COPD program is successfully evaluated from a professional perspective in terms of a reduction of hospital admissions (15% between October 2003–2004), admissions after three months (15% same year), exacerbations (30% ibid) and quality of life (10% ibid) [30]. The patients evaluate the Dutch program also as successful. A patient: The program supports the activities I can do at home. Very simple exercises, walking, home trainer and household jobs, like washing the dishes. They state that overall doctors are less sensitive to the mental aspects of the disease, and have less ability to explain practical advices. They do notice, however, that the communication between the physiotherapists and the doctors is short: The lung physiotherapists have direct contact with the doctors. I do notice that the doctors don’t talk a lot with each other, but they do talk with the therapists a lot, they seem to have a very easy accessible contact. The professionals of the program are very involved with the patients and know the balance between taking care and empowering patients. A patient: Here in the therapy they are much better at explaining (how to take my medicine). One patient even says that the program prolonged his life: They saved my life, because they taught me how to breathe. The patients also emphasize that the route that has to be walked to reach that result is difficult. One of the patients is very clear: I do not want to do it over again, all the physical training. It was a very, very tough year.

UNDERLYING THEMES

The evaluation gave insight in a variety of themes which helped answering the research questions as stated above.

Patients’ Life Story

In the COPD program, the physiotherapist listens to the patients’ life story and at the same time learns about and questions the values of the patient. The physiotherapist does that through introducing new insights and information about COPD. This often induces an increased understanding among the patient and partner about the way the disease influences their lives. The lung physiotherapist explains:

During the intake, when the atmosphere is right, patients realize they don’t suffer from a shortage of breath only, but it’s also ‘I’m not feeling well in general’. When the patient thinks about that, talks and comes to the conclusion that he needs to work on that, than you are a step further in improving the quality of life.

Another example is the female patient who enters the centre remarking that following the program and the physical activities is impossible. After the intake with the physiotherapist she decides to stay; it was clear that the cause was her fatigue. That bothered her even more than her breathing problems. During the talk she cries and expresses she doesn’t feel understood; ‘working part-time and doing the household, it is too much for me’. Talking to the physiotherapist during the intake, she starts to understand the relation between
smoking and her fatigue. She accepts the need to find a balance between her activities and quit smoking and decides to join the program. From the start of the program it is important to understand the development of the patient’s life; what he values, the important moments he experienced and how he coped with that. In the Dutch COPD centre, the telling of the illness narrative helps to shape and create experiences. Besides reflection, eliciting, articulating and questioning values, it also contributes to the direct experience of symptoms and suffering (Kleinman, 1988). The telling of the life or illness story helps the therapist and patient to determine what specific treatment is needed and most of all: who the person is (Abma et al, 2005b) (Goldsteen et al, 2007). A life story is a narrative of someone’s life in order to give meaning to the things he experienced (Ricoeur, 1983). In the COPD program, the lung physiotherapist is trained in conversation techniques similar to those used in cognitive behavioral therapy to elicit and listen to the life story of the patient. In this context, ‘listening’ has to do with paying attention and showing a genuine involvement. The sincere ‘presence’ of the professional facilitates a natural conversation. It is not about accountability, fulfilling one’s obligations; the professional adequately responds to the patient as a person. Listening has also to with summarizing and giving feedback.

**Integral View on Mind–Body Connection**

One purpose of the program is to increase patients’ awareness of their own body and mind and their integration. Patients learn that it is not only shortness of breath they are bothered by, but also other aspects of their life like their awareness of their boundaries and the amount of physical activity they can handle during the day. The switch to this more integrated approach started in the early 1980s when the lung physiotherapists noticed that there is a very strong connection between someone’s physical and mental or emotional state. A lung physiotherapist illustrates the problem:

*Somebody doesn’t want to get up anymore because he is short of breath. The other person (who tries to help) treats him very carefully, but that leads to inactivity. The condition diminishes because the patient is tight in the chest, becomes more tired, so he just wants to be left alone, he becomes afraid, his medical consumption increases and social desolation may follow.*

Social desolation worsens the patients’ situation. In the Dutch program it is important to stimulate the patient to stay socially active, continue to work and meet family and friends. The conventional way of treatment included rest, but the physiotherapists realized that had an opposite effect: rest leads to inactivity. The Dutch program illustrates that social functioning benefits the integral experience of the person with COPD. In order to strengthen the physical state and stay socially active, patients need to learn to think differently. Just realizing that inactivity increases health problems is not enough to change their behavior. Behavior that has been developed for years, changes step by step through training and increased awareness. Together with the physiotherapists and peers, patients reflect on their behavior and thought patterns. Often, this has to do with recurring themes, like the ability of patients to notice their limitations and accept when to take a rest:
The most difficult was to learn how to stop. Accept my boundaries. Since I was born, I am used stretching my boundaries (patient I).

After some exacerbations the hardest thing to do, was deceleration of the pace of my life and work, and to accept my limitations and this new, lower pace. When I cross my limit, my breathing increases and I’m snapping for breath.

During the training the therapists helped me to regain my balance, but I lost other functions forever, unfortunately (patient II).

Whereas in the past physical issues were stressed one-sidedly, in the COPD centre observations of the emotional and mental state of the patient are of equal importance. This requires training in and openness towards mental and emotional areas. The physiotherapists of the Dutch COPD-program joined a short training in cognitive therapy in order to learn how to approach patients.

Decartes defined body and mind in terms of mutual exlusivity. The body is pure res extensa—unconscious, material—whilst the mind is res cogitans—mental, without location, bodyless (Bordo, 1999). Descartes described that the mind could ‘control’ the body, but mind and body would be two distinct substances. This Cartesian dualism with the mind with its consciousness and self-awareness and the brain with its intelligence is confirmed by studies by neuroscientists. This material monist view reduces mental phenomena to brain processes (Boscarino & Chang, 1999) (Gupta, 2006) (North, 2002) (Kolk et al, 1996). It does not need explanation that this dualism influenced medical science and its focus. We can see developments however, that illustrate that the acceptance of a non-material, nondualistic relation between mind and body grows. This is especially the case among some psychologists and psychotherapists. In the other program we evaluated, the treatment of people with fibromyalgia, professionals used insights from Mindfullness Based Cognitive Therapy (MBCT). MBCT is proved to be an effective treatment for several personality disorders (Evan et al, 2008). Research about the effectiveness of cognitive therapy for people with physical chronic illnesses is still in its infancy (Al-Obaidi et al,2000) (Vlaeyen et al, 2002) but the assumptions about the mind–body connection already percolate in these kinds of multidisciplinary programs.

More philosophically, these assumptions deal with an integral philosophical approach to the mind–body problem. In general, this has been one that acknowledges the essential unity of body and mind while emphasizing consciousness, or interior subjective awareness, as primary. Indirectly, one refers to this unity when stating: ‘I am a body versus I have a body’ (Abma, 2002) (Slatman, 2009a) (Slatman, 2009b). This relates to the work of Merleau-Ponty. According to Merleau-Ponty(1968) the human body is not just a domicile of the mind but, the ‘higher’ functions, including thought itself, should be regarded as bodily functions referring to the whole body in its relational being-in-the-world. We could find this relational view in the case example where both patients and therapists need to take an appropriate view of the relation between mind, body and its ‘being in the world’ in order to enable effective dialogue and therapy.
Self-Management
Taking care of one’s boundaries is closely related to the concept of self management. Patients have to learn to deal with their limitations. If they experience difficulties, they need to know where to get help and how to ask for it. They need to change the belief and thought that ‘asking’ is ‘not a sign of weakness, but of strength’ (physiotherapist). Self management also means: setting limits and knowing what decisions to make in one’s own interest. The pulmonologist gives an example:

A patient consulted me when he was unable to cycle with his friends. I examined him, explained the cause of his problems and prescribed medicine. After a while, this patient consulted me again and said: ‘But doctor, I am still unable to cycle with my friends!’ I could have answered something like ‘then you have to find another group of friends or ask them to slow down’. Instead, I told him: ‘We can also find another way to solve this. Let’s look if physiotherapy has an effect. Let’s see if we can find out what your limits are.’ His physical condition was fine, but he had to learn how to set limits (pulmonologist).

Self management includes the ability to make adequate decisions. Several factors influence the ability to make the right decisions, like the existence of comorbidity. People with COPD have an increased risk to get other health problems, like a lung infection (Heijmans et al, 2005). Fear for drawbacks and additional problems may prevent people to become active. Therefore, an important part of the program consists of education. One example is a card for patient with an SOSplan created by the health professionals in cooperation with the patients. The card carries advices that match the symptoms and feelings of the patient at a specific moment. Patients can consult the card in case of problems and decide what to do.

Learning from Fellow Patients
Still another example of how self management is implemented is the interaction in the larger group. Patients need this support of a group of fellow patients. It helps them to learn to accept their disabilities, because of the increased awareness through listening to each other’s stories. The lung physiotherapist:

We talk about our observations during the training and we ask whether other patients recognize them. Sometimes there are tears, but that is possible because the people trust each other. People recognize the other persons’ story. They all feel the same. And then you see that they support each other. Help is offered by fellow patients.

Patients recognize each other’s stories and find them comforting in an empowering way: they hear different perspectives on the meaning of the disease, and may discover that their own understanding is quite limited. The sharing of experiences also happens during informal meetings, like a coffee break. A patient:

During the coffee-break we talk and exchange ideas and that’s how we learn from each other. You’re among colleagues, there is also a lot of laughter, besides sadness, we’re also having fun a lot of the time!

While joining the group, patients learn practical methods from peers to cope with COPD. And they come to see their situation in a new light by humor and fun.
Professional Competences
To be able to work with and in groups like this, professionals need specific competences, from both a professional and a personal point of view. The approach of the therapists in the group-work varies from keeping a professional distance to openly expressing emotional involvement; they facilitate the dialogue by sharing their observations of the training and their own, more personal experiences. In that way they induce a conversation about specific subjects and create an atmosphere of trust. The professional needs to be able to think and act beyond the limits of their discipline and workplace. An example is the lung physiotherapist who works in the hospital, but often visits patients when they are unable to attend the training because of a relapse. The therapist advises on what the patient can do at home, to help the patient to become active again and resume the training. Another skill a professional needs is the ability to act in an independent way. Where new, innovative programs are developed, they will meet resistance of more conventionally oriented professionals. For example, during the development of the Dutch COPD centre, other professionals doubted the design of the program and especially the duration of it. The lung physiotherapist:

*We experienced a lot of resistance. They thought a patient cannot be a manager of his own disease. We were convinced, at the time, that patients were equipped enough to continue on their own (to leave the program) after six to 8 weeks, whilst scientific research stated that a minimum of three to 6 months was required to treat the patient.*

Another competence that is mentioned is empathy. The pulmonologist tells:

*Empathy. To be in sympathy with. If you do not have empathy, the conversation with the patient will be very rational. You will be able to help someone, but only till a certain degree. You must be able to talk about emotions and experiences.*

Narrative, Dialogical and Relational Work
In the Dutch COPD centre the COPD patient is no longer a passive recipient of medical treatment delivered by a medical specialist. He becomes a partner in the program, and takes on responsibility for his health. This new division of responsibilities is, however, not something that comes about automatically, or can be planned and organized by clinical guidelines and protocols. The ethicist Margaret Walker (1998) explains that from an ethical (versus juridical) perspective responsibilities are actively negotiated among people by exchanging normative expectations. In their interactions people develop shared understandings over what they expect of themselves and others, and vice versa. This entails moral issues like the interdependences between people and the risks of a certain division of responsibilities. When people begin redefining responsibilities they redefine themselves, their roles and their relationships. This requires a lot of narrative, dialogical and relational work.

First of all the doctor needs to redefine his traditional medical expert role. In the paternalistic patient-doctor relationship the medical specialist is unlikely to have much interest in discussing patients’ concerns (Emanuel & Emanuel, 1992) (Oeseburg & Abma,
The professional decides, acting in the best interest of the patient without having to explore the patients’ values and concerns. The professional acts as the guardian of the patient. This paternalistic relationship is perhaps appropriate in situations where there is a life threatening or an acute illness; in the case of chronic COPD, it is highly unlikely to work. So, the medical specialist needs to develop a new role towards the patient. In the COPD centre, the pulmonologist shifted his role to that of a teacher, educating the patient to recognize his limits and to find new ways of dealing with the disease. Likewise patients had to become more active in both defining their problems and in determining the appropriate treatment. One of the hardest things for a person with COPD is that he and others initially see him as a patient. This is the effect of COPD being a chronic disease; having an illness still means, for most of us, going to bed, taking medicine, to stop and quit doing what we used to do. We still associate an illness, even if it is chronic, with an acute disease. So, one of the biggest challenges is to redefine COPD patients as persons with COPD. Once the person with COPD begins to define himself as such, he is able to see that he is not the disease, that he is able to carry on his life despite limitations and that he can take on responsibilities for the quality of his life. This is not a one way process; both parties (patients and professionals) have to develop a new understanding of their own and the other’s role.

Secondly, both parties in the COPD program need to redefine their relationship. The clinical distance common for the paternalistic relationships is no longer suitable. In the COPD program the patient and professionals developed an engaged and empathic relation. In each instance a lot of energy was invested, mainly by the physiotherapists, in helping to understand the patient and the story of the patient. The story is seen as a way of making sense and giving meaning to experiences. The lung physiotherapists attended a specific course to learn how to question patients. The story is not only about the physical disease, but also about the emotional and social impact of it. Think of the female patient who was initially not at all motivated to join the program. The therapists had to elicit the story and listen to it, and then had to interpret what was going on in the life of this woman. Why did not she want to quit smoking at first, why was she so stubborn? The therapists began to develop a sense of what was going on, not by asking what she needed or wanted, but by focusing on her personal life-story. They discovered that the fatigue was more urgent than the breathing problems. The fatigue had to do with her problems combining a job and doing the household and the high expectations of herself. Furthermore, her husband seemed quite insensitive to her problems. By telling her story to the therapists and by reflecting on it together, the woman was able to give meaning to her experiences. The therapists had to interpret what she valued in life and which treatment could help her. In this case, the therapists acted as counselors or advisors assisting the patient to elucidate and articulate her values. It became less important for her to meet everyone’s needs and more important to treat her own mind and body in a healthy way. That’s why she decided to join the program. Another example is the patient who needed the conversations from the physiotherapists to help him to accept a new balance in his life. He could not do this on his own, but needed the dialogue with the professionals to discover what was right in his life at that moment in time.
This corresponds with the purpose of the COPD program as mentioned: to develop a new lifestyle in a dialogic way through the introduction of self management. The relationship between the professional and the patient is a means to reach that goal. We see here that the traditional, one way of communication is replaced by a more dialogical conversation in which the patient as well as the partner and the professional are engaged (Emanuel & Emanuel, 1992). Whereas in the past healthcare professionals gave primacy to the cure of the physical disease, in the COPD centre, the emphasis is on the meaning of the disease and finding a way to deal with the disease by the patient himself. Therefore, empathy and listening are important skills that need to be developed among professionals. Our findings illustrate that one of the patients experienced the conversation with the pulmonologist as a ‘one-way’ conversation. Unlike his relation with the physiotherapists, he felt less understood by the pulmonologist. Hence, concerning ‘empathy’, the professional needs to be trained to show a genuine involvement with the patient, without losing himself. During listening, three things are important: hearing, understanding and exchanging (Bolsenbroek, 2007). ‘Hearing’ implies that the professional is able to rephrase what the patient is telling him. Understanding is about the ability to hear the meaning in the words and exchanging deals with an advice, remark or observation that arose while hearing and understanding. These are three aspects of listening that are important in forming relationships with patients in the COPD centre. Communication does not only take place between professionals and patients, but also among fellow patients. The safety of a group is an important component to assist patients in their empowerment. The group helps them to recognize that they are not alone with their problems; they need not to be ashamed of themselves. Furthermore, the stories exchanged and responses are often more universal. The group sessions stimulate participants to socialize, to make contact and to stimulate and support each other. Together the patients can write their we-story; the story of how they gained back control over their lives. Finally, the new division of responsibilities entails relational and communicative work among professionals. The multidisciplinary focus of the program requires personal skills from all the professionals. Old values like hierarchy and control are replaced by values as equality and autonomy. Old structures between doctor, nurse, therapist and also organizational structures (like first en second echelons) vanish and are replaced by a network organization with a dialogical way of communication. Direct and easy accessible communication between professionals, including new participants like lung physiotherapists, psychologists, general practitioners and welfare workers, requires constant attention. Different vantage points should be respected and if necessary, negotiated. To overcome feelings of insecurity and resistance courage, entrepreneurship and enthusiasm are needed. Also between professionals, the importance of listening and building an atmosphere of trust is important.
CONCLUSIONS

The case example illuminates that people with COPD (3) need the surrounding of a larger group of fellows to learn to accept their disabilities. Awareness that COPD is more than just a lack of air is just a start. According to people with COPD it is a first step towards a better quality of life while health care professionals regard it as a first step to investigate other problems in life and to break through the circle of inactivity. In a reflection on the case we argued that a chronically ill person is not just a consumer; he is also a conversation partner, learning from as well as teaching his professionals and fellow patients. Likewise the healthcare professional is more than just an information provider; he is often also a coach and Socratic guide who challenges life styles taken for granted, who motivates patients to change their behavior and who gives support and practical feedback. Doctors as well as patients have to grow into these new roles and establish new relationships. Empathy, support, listening to the patient’s story and dialogical interaction are as important as giving information and asking for consent.

Self management is often regarded as a panacea for patients with a chronic disease. Patients have to actively deal with their situation in order to regain autonomy. Self management is at the same time highly contested. Critics state that the patient has to take up a role which he does not want to play, and often is not able to, and that professionals leave patients alone in their suffering. Although these views of self management are opposed, they share the presupposition that self management is equal to being independent and in control. In this article, we have shown that the practice of self management in a Dutch COPD centre does not focus on independency and control. It rather involves a new division of responsibilities, in which patients and professionals develop new roles and relationships. Patients and professionals have to become partners in care. This implies a lot of relational, narrative and communicative work. Both parties have to grow into the new roles, in which values like equality, autonomy, and genuine involvement are important. The professional needs to develop skills like empathy, giving support and listening. The patient needs to develop an awareness of limits, learn how to share experiences with fellow patients in a group and how to use self management to change his life style for the better. An underlying assumption is that mind and body are connected in a subtle way and that patients can only improve their quality of life when they work on both aspects. Another assumption is that patients function in a social system and that in order to learn how to cope with their disease, they should play with their position in the system and the limits they meet. Patients of the COPD centre have learned how to do that and have also learned that it is a fragile process that never stops; self management needs continuous attention and relational support.
Notes
(1) Volkskrant, 6/7 December 2008.
(2) This is based on information provided by the website of the World Health Organization http://www.who.int/mediacentre/factsheets/fs315/en/index.html
(3) In scientific discourse, the term ‘participant’ is widely used to describe the subject. In this article we wittingly use the terms ‘patient’ and ‘person with COPD’. People with COPD prefer the last term because they regard the disease as something they have. They are not the disease itself. In the context of the case of the COPD centre, we decided to talk about ‘person or people with COPD’. In the more analytical paragraphs, we use the term patient.

REFERENCES


Implementing issues and values


Truus Teunissen
Material: bronze
Pictures taken during the research, interactive sessions in focus groups, discussion groups, expert meetings, round table meeting and a conference presentation.
7 – General Discussion

7.1. INTRODUCTION

This thesis explores the body of experiential knowledge of people with a chronic illness or disability. It investigates how this unique kind of knowledge can be included in health care and health research practices. The thesis started from the increasing acknowledgement of inclusion of patient perspectives in medicine and health care services (Crawford et al, 2002). Inclusive practices either begin with the invitation by researchers and care professionals to people with a chronic disease or handicap to join processes of decision-making on quality of care and research, or people actively seeking ways for participation and partnership themselves. In both cases, they act as a representative and advocate. Hence, people with chronic illness or disability themselves work as representatives on behalf of their advocacy groups and organizations during involvement activities.

In order to conduct their role accurately, representatives need to know what matters to the ones they represent. Only then there can be a genuine representation of perspectives of people with a disability or chronic disease. Experiential knowledge of these peoples’ values and concerns may provide building stones for advocates to voice perspectives. One of the areas where this occurs is during the appraisal of care and research: the main focus of this thesis.

This chapter integrates the insights of the previous chapters and answers the research questions. The main research question is:

What constitutes the body of experiential knowledge of people with a chronic illness or disability in the context of appraising quality of care and research?

Sub questions are:

1. What matters to people with a chronic illness or disability concerning research and quality of care?
2. According to which criteria do people with a chronic illness or disability appraise research and quality of care?
3. How do people with a chronic illness or disability and professionals interact and relate to each other during involvement activities?

The chapter is divided as follows. The first section presents the main findings per sub question (7.2), reviews the methodology (7.3), provides recommendations for further research (7.4) and for practice (7.5). The general conclusion, answering the main research question, is presented at the end (7.6).
7.2 MAIN FINDINGS
The studies of this thesis result in three main findings. These main findings are presented below.

7.2.1. Issues and values that matter
The first finding concerns the issues and values that matter to people with a chronic illness or disability. Our research shows that people with a chronic illness or disability ascribe major importance to certain issues and values in their lives (Chapters 2 and 3). These values and issues are grounded in the experiential knowledge of people with chronic illness or disability.

Experiential knowledge is experience gained by people while living with a chronic illness or disability and closely related to the practical knowledge used in the clinic (Pols, 2012). Both types of knowledge are characterized by processes of adjusting and tinkering of emerging insights. The experience and knowledge coming from the environment where they lead their life, can contribute to better research and better quality of care (Mol & Law, 2004) (Mol et al, 2010) (Mol, 2008) (Moser, 2000).

Clinical knowledge derives authority and reliability from the experience of the clinician and build on many observations of patients’ reactions to treatment and advice, as well as the ongoing process of observing, intervening and evaluating this particular case (‘mimicking’ and ‘tinkering’). It is about improving rather than proving.

This thesis illustrates that patients travel a similar path when they relate knowledge their doctor or other care professional tells them about, to their own situation. The ‘tinkering’ also occurs in daily life settings where people juggle and improvise what they can or cannot do depending on their physical condition (Chapter 2). People with a chronic illness or disability use practical knowledge, or know-how that was needed to travel from one situation to another, providing footholds for any new kind of problem that arises within a particular situation. The experiential knowledge of people with a chronic illness or disability emerges through a tinkering process within a multitude of areas: e.g. education, work, care, income, living, family and mobility. By experiencing and interpreting daily events, people learn what they value and what not.

Set of 35 values in two domains: quality of care and research
Figure 1 shows a set of 35 values and issues of people with a chronic illness or disability in the domains quality of care (Q) and health research (R) as found from the systematic literature review (chapter 4). These 35 values and issues were initially ranked by their frequency of occurrence. People with a chronic illness or disability indicated in various focus group meetings that these are the right issues. This thesis shows that the right sequence of importance turned out to be another matter (Chapter 5). The frequency of occurrence was not the right indicator of their importance.
Figure 1  Values and issues of people with a chronic illness or disability

Chapters 3 and 5 report on a certain sequence in *What matters* to people with a chronic illness or disability. According to the participants of our study, some values are more important than others. For example, for participants, feeling safe is more important than good communication with the professional. This leads to a value based priority structure in appraisal and evaluation of health research and quality of care. We have called this the “involvement iceberg”, which is visualized in figure 2.

According to the participants of this study, the most important values are the *fundamental values*. A dialogue about relevant aspects of quality of life with them is only possible after their fundamental values are ensured. (Chapter 3 and 5). A value is called ‘fundamental’ when it is of the utmost importance to people with a chronic illness or disability. These fundamental human values need to be addressed before attention for quality of care and research is possible: Fear, Vulnerability, Being taken seriously, being Respected, Accepted, being Heard, being Recognized, Identity preserved, Values respected, Dignity, Privacy, Trust, ‘experiencing Hospitality’ and ‘Not being excluded’.
For the participants of this study, it is only possible to deal with values associated with quality of life, after fundamental values have been met. Values associated with quality of life concern: Support, Independent voice, Self-care, Share experience with peers, Being able to move about and have Social environment and contact with Family and friends. When those are met, quality of care related values become more important: Access, Choice, Treatment plan, Patient Information, Effective communication on an equal basis between patient and care provider, Up to date medical file, Quick and adequate diagnosis, and Freedom of choice. Finally, values related to involvement and participation itself start to matter: Advocacy, Partnership, Acknowledgment and recognition of the value of experiential knowledge and Conditions for participation are items that matter to people with a chronic illness or disability.
**Representation of values and issues**

People with a chronic illness or disability, are usually quite busy with and absorbed by how to meet their fundamental values and quality of their life (Chapter 5). They are, for instance, dealing with a lack of trust of persons in their environment, or do not feel acknowledged by their neighbours. Participants who are mainly focused on activities that meet fundamental values like being seen, getting respect and trying to improve their quality of life e.g. to fit in with family and friends often do not aim to fill out inquiry forms, to join focus groups or participate in debates. Their values of concern are predominantly ‘under water’ in the iceberg, and may therefore remain invisible for society around them. In addition, values and issues of people with chronic illness and disability are not seen in primary and secondary scientific literature. Chapter 4 reports on a data synthesis that consisted of a literature search in primary literature (scientific literature) but also in secondary and tertiary literature (‘grey’ literature). The selected ‘grey’ literature sources show that 13 out of the 35 issues that matter to people with chronic illness or disability are not found in the primary sources but only in the secondary and tertiary sources (Chapter 4). The information obtained from secondary and tertiary literature, e.g. proceedings, reports or ego-stories, is often closer to the experiences and experiential knowledge of people with chronic illness or disability than subjects of primary scientific studies.

**7.2.2. From issues and values to criteria**

The second question concerns the criteria that representatives use to appraise care and research. In practice, the extensive inventory of values and issues presented above is not suitable for representatives. Chapter 3 describes that a lack of clearly defined criteria adversely influences the motivation of people with chronic illness or disability to contribute to the appraisal of research and quality of care. Chapter 3 shows that often vague, too abstract and poorly operationalized criteria are being used. Involvement becomes unattractive for representatives and its continuity and quality is troublesome. In order to be able to critically review, to advise or to participate in discussions and to provide information, representatives of people with chronic illness or disability not only need skills, but also knowledge and tools that may guide them. Until recently, criteria were not available.

Chapters 3-6 highlight the importance of a structured set of criteria for use by people with chronic illness or disability when taking part in discussion, appraisal and evaluation activities. Not only as a vehicle for dialogue with other parties at the table, but also to enhance the quality of the appraisal process by the inclusion of systematically investigated and validated values and issues. Therefore, the inventory list of values and issues has been condensed and structured on behalf of- and together with- patient and client representatives, into a set of six main appraisal criteria by a responsive evaluation process (Chapter 3). The six appraisal criteria provide a structured appraisal approach from their perspective that matches the current health involvement practices. Table 1 presents these six criteria.
Table 1 People with chronic illness or disability appraisal and evaluation criteria for involvement practice

<table>
<thead>
<tr>
<th>People with chronic illness or disability criteria</th>
<th>Description</th>
</tr>
</thead>
<tbody>
<tr>
<td>1 Relevance</td>
<td>Does it deal with effective health improvement to be obtained for people with chronic illness or disability? Are the practical importance for society as a whole and the diversity aspects taken into consideration?</td>
</tr>
<tr>
<td>2 Quality of life</td>
<td>Is it of practical use, is it increasing their independence, their societal participation and is it improving their everyday quality of life? Is it respecting their basic values and needs?</td>
</tr>
<tr>
<td>3 Quality of care</td>
<td>Is care being improved and does it reflect the many facets of quality perception of a diverse group of these people in their social environment?</td>
</tr>
<tr>
<td>4 Ethics and safety</td>
<td>Is the least objectionable way to achieve the goal being chosen? Does it comply with regulation and codes of conduct? Are these people protected against harm and damage during and after the medical intervention? Is there freedom of choice and respect for fundamental needs? Are unnecessary repeats avoided?</td>
</tr>
<tr>
<td>5 Information and communication</td>
<td>Is correct and clear lay information ensured? Are these people made aware of potential risks to allow informed decisions about any medical procedure to be performed on them? Are results shared with them in lay terms?</td>
</tr>
<tr>
<td>6 Involvement (Prerequisite)</td>
<td>Are the interests of these people included? Are they being treated as equal, knowledgeable partner? Is the experiential knowledge recognized and acknowledged? Are advocates and representatives trained, enabled and supported with their involvement activities?</td>
</tr>
</tbody>
</table>

The six criteria can be regarded as an expression of the current body of experiential knowledge of people with a chronic illness or disability when appraising research and quality of care. Experiential knowledge is not limited to health care (Ardelt, 2004; Kangas, 2002; Hoppe, 2010). It stretches out to all areas of life such as education, work, care, income, living and mobility. The criteria that ‘embody’ experiential knowledge may empower people with a chronic illness or disability and their organisations, because the
criteria provide a validated hold and offer a structure for appraisal and evaluation and clearly identify what key issues matter to people with chronic illness or disability (Chapters 5 and 6). Subjects are not likely to be forgotten and overemphasis of a single subject at the expense of others can be avoided. There are however, some risks, that we will address in a following section.

Example: patient involvement in appraising health research
Chapter 6 describes how patient involvement in lung research is being carried out. The Chapter describes this from the perspective of lung patients in The Netherlands over the period 2005-2012. The seven year longitudinal case study looks at involvement of people with a lung disease in the research funding approval cycle at several time cross sections. Over these seven years, the criterion for approval and funding changed. These criteria developed from scientific quality, being used as the only key criterion, towards the implementation of both scientific relevance and societal relevance as equally important additional criteria. The latter two are being used since 2010.

7.2.3. Relational dynamics in involvement practices
The third question concerns the dynamics between representatives and professionals while appraising quality of care and research. The studies of this thesis show that people with chronic illness or disability can be involved in the appraisal of research and quality of care from several roles. What our studies show is that representatives often function in an advisory role in a committee or health debate. In the role of advisor one is present and included, but this is not to say that one is heard and taken seriously. Quite often the advisor is the only person with a chronic illness or disability in a group with professionals. Over the last years this is changing. The example below illustrates this gradual change in the involvement practice in a charity fund. It concerns a summary of the seven-year longitudinal case study (2005-2012) on the involvement of people with a lung disease in research funding approval is presented (Chapter 6).

Example: patient involvement in appraising health research
Over seven years, the criterion for approval and funding changed. These criteria developed from scientific quality, being used as the only key criterion, towards the implementation of both scientific relevance and societal relevance as equally important additional criteria. The latter two are being used since 2010.

Chronic lung patients did not have a group of patient representatives who were capable of introducing their interests on a national and international level before 2005. In 2005 the scientific advisory committee (WAC) effectively included one patient member. Between 2005 and 2009 two lung patients participated in the WAC, based on their personal capacity. These two persons were assigned members of a newly established group of lung patients with experiential knowledge as well. This was a key change in the process. The group represented lung patients on an
(inter) national level, and was being facilitated by an LFN staff professional from
2010. In 2010 the two patient WAC members had difficulties to consult with the
chronic lung patients in the country and getting some more support. By 2011 three
lung patients participated in the WAC as equal partners.

At first, it lacked commitment of professionals to patient involvement, both
inside and outside the LFN up to 2009. The patient involvement in the research
cycle was further intensified every year, based on experience and discussions with
the patient members in the scientific advisory committee. By 2012 patient
involvement was widely accepted in the LFN organization and it was mentioned in
the LFN strategy, in the research policy and in the annual budget allocation for
training and information exchange.

Representatives of people with chronic illness or disability can be advisors and they may
also act as an equal partner or have control over processes in care and research. The role
primarily indicates the status of the information or activity provided by the representative.
This determines the crucial difference between being present and included and having a
right to say and influence. Involvement roles match with Arnsteins’ ladder of citizens
participation (Arnstein, 1969). These roles are shown in figure 3. Higher steps on the ladder
indicate a higher level of involvement.

Figure 3 Involvement roles of people with a chronic illness or disability

Chapter 2 of this thesis describes how Truus, the author, acted herself as advisor and
partner. The thesis itself is a manifestation of the principal/control role.
**Situated and relational roles**

Our studies show—see chapter 5 and 6.1—contrary to the Arnstein model people with chronic illness or disability representatives not merely aim for the highest level of involvement (control), but consider what level is the best, the most effective and the most feasible to achieve their goal. This also depends on the individual person’s history, identity and situation. Many representatives indicate that conditions for involvement like support, financial compensation, and having a replacement available are often not met, thus limiting their involvement (Kool, 2013; Dedding & Slager, 2013). No need to say that without proper conditions for involvement and experiences of being treated as an—in Dutch—“excuse Truus” the motivation to be involved will decrease.

While Arnstein’s model is static, in practice people with a chronic illness or disability experience a dynamic process of trial-and-error finding out what fits their ambitions and dreams when it comes to involvement. Chapter 2 of this thesis highlights that representatives may have been exposed to a difficult and emotional individual process of coping with mourning, loss, acceptance and finally awareness and growth after becoming a person with a chronic illness or disability, a ‘patient’ or a ‘client’. Being confronted with and living with a chronic illness or disability sometimes involves periods of anxiety and self-centeredness alternating with periods of strength and acting on behalf their groups. Hence, people who function in their role as representatives, are both strong and articulated as well as vulnerable and dependent. This finding places the notion of the robust, resolute and articulate person with a chronic illness or a disability, being in control and doing self-management in perspective.

Chapter 2 demonstrates this concept is flawed, since people with chronic illness or disability are not all able to live up to this ‘sturdy’ image all the time for reasons like having a mental illness, being fully occupied with survival or coping with a disease, being in the early phases of a mourning process, being part of an ethnic or cultural minority group or having poor literacy skills. When actively involved in quality of care and research activities, they face difficulties with keeping up the continuity of their activities due to uncertainties associated with their chronic illness or disability and improper conditions for involvement, such as the lack of fresh air when having a lung disease or lack of assistance to go to the toilet when wheelchair bound (Kool, 2013). In addition, it is hard to appraise research proposals from their perspective because existing research forms and protocols practice a language that is often unfamiliar to them and not matching up with their experiences (Chapter 5).

To bridge this gap between the daily muddiness of their situation and high notions of involvement, representatives of people with a chronic illness or disability attempt to act according to the dominant thinking, model and discourse. They experience feelings of shame and guilt as they are neither meeting the high demands nor want to be excluded from involvement at the same time.

Proper conditions need to be created to prevent this dysfunctional relational dynamic. Inclusion strategies include good circumstances, (the setting), behavior (i.e. the opportunity to speak) and verbal communication (lay terminology) (Elberse et.al., 2012) and mutual
support and relational empowerment (Baur, 2012; Schipper, 2012). Our study indicates that one of the most important factors is ‘support’. In order to contribute in involvement practices, people need support to properly fulfill their roles. Fulfilling their roles is never a solitary activity, but a relational activity (Chapter 6.2). Ideally, people with a chronic illness who act as representatives, function in the context of a partnership with researchers, policymakers or any other person who is involved in the appraisal practice.

Example: patient involvement in agenda setting

Another example of how a partnership emerged between people with a chronic illness or disability and professionals when appraising quality of care and research, concerns a partnership in research agenda setting. Chapter 6 describes, in 2004, the lung foundation invited experts and people with asthma and/or COPD in developing a shared research agenda. In 2009, the LFN research agenda was updated and extended including rare lung diseases. Again, patients and experts were actively involved as partners. This resulted in two new societal lung research agendas. The approach for updating and extending research priorities proved useful for eliciting research priorities from patients and experts. Although research priorities of patients and experts are largely comparable, they differ in details and patients prioritized research topics in chronic lung diseases that were previously not included in the research focus in The Netherlands, such as fatigue and comorbidity.

Conditions: Support and communicative space

There are some risks that involvement in health research and quality of care becomes too instrumental by using the appraisal and evaluation criteria that this study developed (Chapter 3). Professionals might use the criteria as a tool or a checklist and consider involvement to be ready as soon as the questions are asked or the boxes are ticked. True involvement consists of more than ticking boxes, it requires genuine cooperation, sincere dialogue and partnership (Elberse et al, 2010; Baur, 2012; Schipper, 2012). Moreover, this thesis and other studies show it requires space for people with chronic illness or disability to express their concerns, vulnerabilities and negative experiences with research and care (Todres et al, 2007; Visse, 2012). Too often, there is no space for these parts of experiential knowledge. The communication style of people with a chronic illness or disability not always matches with the scientific and professional discourse, and can easily be dismissed as being anecdotal and thus not credible and relevant.

7.3 REVIEW OF METHODOLOGY

In this section the role of the secondary – and tertiary literature is discussed, also called grey literature, followed by a reflection on the methodology of this thesis.
Secondary and tertiary literature
Scientific research is based on the recognition of only scientific studies and literature (primary sources) as a valid basis for further work. This thesis required the use of so-called ‘grey literature’ as well. This consists of secondary literature (e.g. government, scientific and international institutions originated, without a method-section) and tertiary literature (e.g. national and international chronic disease or disability oriented organization publications. The rationale for this has been described in chapter 4.

Did the research reach the people with chronic illness or disability?
For practical reasons and limited time available several groups are underrepresented in this work. These are: people with mental disease, mentally disabled and migrant patients as well as people with lower levels of education, living from social benefits in poor housing conditions or a low social and economic status. They are not likely to have had a voice in this study. Other sources indicate that language issues, cultural differences and non-membership of these groups among chronic disease or disability oriented organizations causes underrepresentation at the negotiation table and among representatives (Dorgelo, et al 2013).

Our findings might not reflect their values and issues, and be too positive about the involvement practice.

7.3.1 Quality procedures
Several methodological strategies were adopted to increase the quality of the study. The most significant criteria are credibility, confirmability, dependability and transferability (Guba & Lincoln, 1989). For responsive evaluation there are additional criteria related to authenticity (Abma, 1996). The following procedures were used in this thesis to meet these criteria.

Credibility
Credibility refers to the degree of correspondence between the data (like perceptions of respondents) and the interpretations of the researchers. In qualitative and responsive research approaches, a major procedure to enhance the study’s credibility concerns the conduction of member checks (Barbour, 2001). Member checks are carried out to receive feedback on interpretations. By conducting formal member checks, we have asked whether the respondents agreed with our interpretation. They received the interpretations by mail or email and were asked: does our interpretation match your experiences? Did we leave out important information? Several respondents reacted with a letter with comments and corrections. Often this concerned the tone and the wording. The changes thus made the text match better with the respondent’s intentions. In this way, the outcomes of our data collection and analysis represent the experiences of the respondents more precisely. Another procedure to enhance credibility is triangulation. This means using different sources of data, different methods, different analyses. Looking for evidence indicating the
contrary, rather than ignoring it, and incorporating such data and thereby eliminating outliers, strengthens the interpretations of the results. Data triangulation has been carried out by multiple data sources like interviews, focus groups, observations and occasionally journals written by respondents. Moreover, the studies took at least 12 months and this prolonged engagement made it possible to better determine the significance of perceptions of participants. Care was taken to build up rapport and interviews and focus groups were conducted in participants’ own ‘natural’ environments, under conditions and on a time of day selected to their preference.

**Authenticity**

Authenticity criteria include fairness, ontological authenticity, educative authenticity and catalytic authenticity. This study primarily focuses on enhancing ‘fairness’ and ‘ontological authenticity’. Fairness refers to the task of the evaluation team to genuinely include various voices, values and interests, and expose and explicate several, possibly conflicting, constructions and value structures (Lincoln & Guba, 1986). Fairness is reached when there is a ‘balanced view that presents all constructions and the values that undergird them’. This has been accomplished by critically reflecting on the inclusion in the research process – both with respondents as with co-researchers – and by discussing the researchers’ interpretations with supervisors and peers. As indicated before some voices have not been included in this study. Those who did participate, approved with how their voices and interests have been represented in both the research process and findings.

Educational and catalytic authenticity refers to the enhanced understanding of the study in their own situation. This was clearly the case with the auto-ethnography written by the main author. The writing process in cooperation with the supervisors was experienced as a process of growing personal insight and thereby control over her situation. Next, my research contributed to increasing the participant’s insight in their involvement setting. The issues that matter to people with a disease or disability are clarified and condensed into six criteria (educational authenticity). This provides them with a firm hold during their involvement activities. It also enables them to better express themselves in terms used by the other partners in the more general debate (catalytic authenticity). They experience being better heard and being taken seriously (catalytic authenticity). This strengthens them in their involvement roles and believes. Likely, this has also led to better understanding of the position and ideas of professionals as well as for the people with a disease or disability. Since the issues of people with a disease or disability are brought forward in a structured manner – the societal relevance – professionals can now focus on scientific relevance and quality of research projects or guidance and leave the appraisal of the societal relevance to the people with chronic illness or disability (educational authenticity).

### 7.3.2 Transferability

The transferability of qualitative data refers to the extent to which findings from those data can be transferred to other settings. Qualitative researchers talk about possible and legitimate relations or patterns between concepts or factors without saying anything about
frequencies and statistical correlations (Boeije, 2005; Fisher et al, 2010). In qualitative research transferability and naturalistic generalization are not guaranteed by the researcher but by the reader. Readers can judge whether or not findings apply in similar settings on the basis of the details and ‘thick descriptions’ provided (Abma & Stake, 2001). The primary strategy is to provide a detailed description of the meaning and context.

7.3.3 Dependability and confirmability

If the study were repeated, in the same context, with the same methods and with the same participants, similar results should be obtained. Dependability informs us about the degree to which results are independent of the propositions of the researcher, time and instruments. This closely relates to confirmability which concerns the extent to which constructions, assertions, facts and data can be traced to their sources, the inspection being done by an external auditor (Guba & Lincoln, 1989). The main strategy for reducing dependability is co-checking by multiple researchers; this strategy has been used in the various sub-studies. Each researcher analyses the data, before they compare and discuss their analysis. Another strategy is peer debriefing by an independent expert to ask critical questions on the methodological decisions and eventual bias. This role was performed by peer reviewers and the supervisor. Keeping a notebook reflecting on one’s own filters during data collection and analysis is used to reduce unreliability. The main researcher described methodological decisions and reflected on the influence of personal perspectives on the data collection and analyses. ‘Auditing’ data is another way of enhancing the study’s dependability. The analysis process and coding methods need to be detailed described in order to allow detailed assessment of the support these data provide for the findings by an auditor (e.g. EMGO+ Quality Committee). The detailed description should also provide the reader with sufficient information to repeat the research process. All analysis procedures were described in detail and the outcomes of analysis have been archived by the authors. The data were stored in digital form, and kept until the completion of the project. At this point the project report was stored together with the transcriptions but e.g. audio records were destroyed according to the quality procedures of the VU University medical center/EMGO+ research institute.

Confirmability has been enhanced by providing the reader of the research reports and publications with information on how the ‘raw products’ have been processed into interpretations and results. Reflexive journaling and detailed reporting of each step and decision has increased our study’s confirmability.


7.4 RECOMMENDATIONS FOR FURTHER RESEARCH

7.4.1 Involvement by disruption
Space for own personal experience from people with a chronic illness or disability is needed in involvement practice. This includes openness for different ways of communication, different from the usual, traditionally professionally oriented, way in the current discourse and codes. Offering space and opportunity to express stories and experiences in their own way leads to uncovering the real concerns and questions that people with chronic illness or disability might have (Ghorashi et al, 2010) (Ryan et al, 2012) (Boevink, 2006). It is not clear whether their involvement disrupts the existing balance of power and processes, and if so, to what extent. What happens if these people do not adapt to the dominant discourse and stay with their own views? Are they then placing themselves outside the group, outside of the debate and have no influence whatsoever? Or is the balance of power slowly shifting and are they becoming really equal partners in the discussion, in other words towards true partnership. It would be interesting in this context to study experience and methods being deployed by research by different universities like currently in use by Patient and Public Involvement (PPI) in the UK (Kelson, 2009). This is recommended for further research.

7.4.2 Exclusion effect
Issues, concerns, priorities and themes of professionals and people with a chronic illness or disability in general, overlap significantly but differ in details and operationalization’s. Ideally, a dialogue between experts would be used to assess these differences and to increase mutual understanding (Abma & Broerse, 2010). Different views do not necessarily aim for subjects to be discussed as complementary rather than as mutually excluding subjects (Elberse, 2010). There is a risk associated with this. If subjects on the table are only perceived to be important to (groups of) people with chronic illness or disability and not to professionals, professionals may not submit research proposals on these subjects. This is an undesirable situation. We recommend further research on how to enhance the meaning of these peoples’ issues as subjects on the research agendas of scientific researchers.

7.4.3 Involving silent and marginalized people with chronic illness or disability
Above, we have addressed that people need to have met fundamental values before they are ready to join processes of involvement. This raises the question how to involve vulnerable, silent people with chronic illness or disability, whose fundamental values have not been met. An example is the group of non-indigenous people:

Quote “Currently, mainly ‘white’ patient organisations say that ‘non- indigenous people do not want to join the discussions’. Organisations should ask themselves ‘what do we need to change?’” (Interview 2010, male patient representative of migrants with cancer)
Further research is recommended on this marginalized group and involvement in the appraisal of research and care. How can their experiential knowledge be introduced and weighed in the health debate or decision-making process?

7.4.4 Alternative ways to get grip, recognition and perspective
Chapter 2 describes the critical moments described in the personal struggle of the author with chronic illness and disability while passing through phases of mourning, needs, shortage and growth. Writing down this story, a narrative writing, illuminated the situation for the author and reflecting on it, together with the two co-authors, provided insight in what these phases and its events meant to the author. According to her, this resulted in an increased grip on recurring experiences. A chronic illness or disability can also create space for a hitherto not utilized possibility or not yet discovered talent. This leads to a new sense of perspective in life. Could this approach of writing down one’s critical moments and by a reflexive process enhance acceptance also mean something for other people with chronic illness or disability? Would they also increase their understanding, recognition and perspective by exchanging views on each other’s critical moments in the story of their life in a small group? Alternative ways exist too: narrative poetry and artistic impression can be used as a tool to gain insight in life with a chronic illness or disability (Owton, 2013). Further research to explore whether these alternative methods have positive effects is highly recommended.

7.4.5 Western countries
This thesis and its constituting studies, focus on the voice of people with a chronic illness or disability in Western countries producing Dutch, English (original or translation) and German publications. This is in fact a limitation, which might leave a part of the issues that matter to people with chronic illness or disability in other countries as yet, unexplored. Further research on how these criteria relate to one another and whether this would enhance their usability and the insights obtained by the users is likely to have merit. One preliminary observation is already that ‘involvement’ is about the process. It is in fact an enabling pre-condition. For the criterion ‘Communication & Information’ this is similar. Without satisfaction of these criteria any active involvement would be pointless. They can be labelled as process criteria.
The criteria ‘Relevance’, ‘Quality of life’, Quality of care’ and ‘Ethics and safety’ are about what is actually contained, they can be labelled as content criteria.
7.5 RECOMMENDATIONS FOR PRACTICE

7.5.1. Values, issues and criteria

7.5.1.1 Fundamental values and quality of life
The findings in this thesis underline the importance of fundamental human values. This implies that people, whose fundamental human values are not being respected or recognized, are not present in the health dialogue. People associate fundamental human values mostly with the criterion ‘quality of life’. This points at ethics and human rights (EC, 2000). On a European level, national governments ought to create an environment that stimulates citizens’ involvement and specifically the involvement of those with chronic illness or disability in health care decision-making. The goals of ‘patient empowerment’ and ‘involvement of citizens’ can only be achieved when the fundamental rights of people with a chronic illness or disability are being implemented and protected.

Involvement of these people that includes the criteria that have been developed in the context of this thesis, can be used as a tool to realise full implementation of these rights in daily practices. Creating and reinforcing organisational structures for involvement is a part of this. Listening to patients and citizens’ voices, and including their perspectives, must be a constant aim of the health system at all levels of control and all geographical branches of health institutions. This is pointed out in the WHO Health 21 programme for the European region in its health policy documents about citizen rights and patient involvement (WHO, 1994).

Several generally accepted documents address ethics and human rights but they are not legally binding (WMA, 2008) (EC, 2005) (CIOMS, 2002). Therefore the implementation of ethics and human rights in health research is not necessarily guaranteed. It is recommended that organisations of people with a chronic illness or disability monitor this well.

7.5.1.2 Values and human behaviour
Values are the most important drive behind human behaviour. Issues that matter to people with chronic illness or disability are directly related to their value. The advocates of people with a chronic illness or disability need to understand the priority, sequence and urgency of problems experienced by the group they represent. Hierarchy of values and needs is in itself subject of scientific debate. Both extremes in this debate, values without any hierarchy and values fully embedded in a hierarchical structure, are of interest here (Tay & Diener, 2011). Values and their hierarchy appear to play an important role in appraisal and evaluation of research and quality of care by people with chronic illness or disability however. Further research is necessary to develop our understanding on this.

7.5.1.3 Vulnerable and Silent people
Chapter 2 presented an auto-ethnography on strength and vulnerability. It indicates that people can only start involvement activities that are meaningful to others after they have in
some way come to an acceptance of their condition and situation. As long as basic needs are not met in the first phase after acquiring the chronic illness or disability and during (recursive) relapses they cannot take part in any involvement activity or let their voice be heard. This point of attention ought to be taken into account in health research in general. So, how to reach out to these people? Ignoring them until they join in? Can organisations of people with a chronic illness or disability, the government or care providers somehow contact them? In any case, this group of people is subject part of the health care system, both in policy and practice.

The concept of robust and outspoken people with chronic illness or disability is being challenged. They are not merely care consumers managing their own chronic illness or disability. There is room and reason other stories, ‘counter stories’ (Knibbe & Verkerk, 2012). Such stories ought to be taken into account in the health discourse and health policy. Research on how the voices of people can be connected to health policy, is required.

7.5.1.4 Visual communication of criteria by symbols and pictograms

People with a mental disability or a mental disease, such as Alzheimer’s disease, and their spokespersons require a special version of our criteria (Chapter 5). Language, graphics and pictograms needs special attention. A pictogram is a symbol or picture representing an object or a concept (Braich et al, 2011). While understanding information on research and care, illiteracy indicates that written information alone is not enough to support people with chronic illness or disability for involvement (Knapp et al. 2005). For some people, recognizing a pictogram or illustration is easier than reading a text. It appears that pictograms are more accurately interpreted than words (Tijus et al, 2005). Symbols and pictograms can thus be used as tools to better assess issues and values. Additionally, our operationalized criteria may be presented with a pictogram, symbol or illustration or any other adaptation of the contents so that they match the group of intended users. Extension of the criteria with other specific issues and concerns related to these peoples’ daily life like choice of meals, time to get out of bed, personal hygiene and so on are possible.

7.5.2. Involvement roles

7.5.2.1 Pseudo involvement

Both governmental and other institutions require an increasingly active role by people with a chronic illness or disability in decision making rather than just be spectators (Bovenkamp, 2010). Involvement assumes that all parties are willing to join and recognize its added value. Unfortunately, this is a too bright picture. Often, people with a chronic illness or disability are still seen as a threat to the existing state of affairs in a project or a committee (chapter 6.1) . A new voice at the table is not seen as a positive development by all parties around the table (Chapter 6.2). One of the unwanted forms of involvement is to involve people as a fig leaf, the so-called pseudo participation or placation (Broerse, Ham, et al, 2010). In such situations, people with chronic illness or disability are actually present at the
table to discuss on behalf of their group but their voice and ideas have no bearing on any decisions taken.

“Patients do provide input at different times in the research process, but that does not really have any effect. It feels like getting a bit of mustard after the last bite of the meal.” (Interview 2009, female, chronic headache)

This type of involvement needs to be avoided as much as possible since it ignores chronically ill or disabled persons’ expertise (including enough support), their values and their political choices. Thus it is therefore recommended that the people with a chronic illness or disability who are advocates for their group, engage only under the pre condition of being equal partners in the debate or decision-making process.

7.5.2.2 The chronically ill or disabled person as citizen, consumer and self-manager

In political debate and in policy documents, people with chronic illness or disability are often depicted in their consumer, self-manager and citizen roles. The ill or disabled citizen is addressed when it comes to responsibility and dividing the limited budget and resources in health care. The same ill or disabled person as a consumer is very much influenced by an economic discourse and the market model in health care. The ill or disabled person as self-manager points at self-care and self-sufficiency in society. These terms and their meaning are often being mixed (Knibbe & Verkerk, 2012). As ‘good patients’, persons with a chronic illness or disability being citizens, consumers and self-managers, are assumed to contribute to keeping health care affordable. Putting the emphasis on an economic perspective on people with a chronic illness moves our attention away from other important questions about the implications of disease and health. In order to increase attention for people with a chronic illness or disability and their issues and concerns in political debates it is recommended to inform policy makers and politicians on the narratives and ‘counter stories’ from them. We recommend more attention for existential and lifeworld-dimensions of people with a chronic illness.

7.5.3. Partnership

7.5.3.1 Something to hold on to

Feedback from chronically ill or disabled peoples’ advocates, shows that they appreciate our criteria. The criteria are found convenient and practical to hold on to (Chapter 5). This set of criteria is expected to be of practical use to representatives and advocates. To know whether criteria sufficiently help them to act as equal partner, requires more research. The possibility and desirability to weigh the contributions of process criteria (Involvement and Information & communication) versus content criteria (Relevance, Quality of life, Quality of care and Ethics and safety) would require further research.
Furthermore, the criteria could be of practical use to health researchers and health care professionals. We recommend to explore the meaning of our criteria for other groups as well.

7.5.3.2 Capacity and capability building

People with a chronic illness or disability are strong and vulnerable at the same time (Chapter 6.1). As stated above, current policies and health discourse are centred around the image of a robust, resolute and articulate ill or disabled person, being in control and self-managing his or her life. Our study showed, this concept is flawed. People with chronic illness or disability are not only strong and articulate but also vulnerable and dependent. Involved ill or disabled persons face difficulties with keeping up the continuity of their activities due to uncertainties associated with their chronic illness or disability. A solution to this would be to use capability and capacity building in their health related involvement activities (Involve, 2013). This ensures continuity as two representatives jointly might take up an activity and can mutually replace each other. It would help if stakeholders, especially governmental agencies and departments, take this vulnerability into account. To realize expectations of both people with chronic illness or disability and health professionals in research and care domains, more support and training is required for the individuals being involved. Apart from financial possibilities there is capacity and capability building to do (Involve, 2013).

The organisations of people with a chronic illness or disability need financial resources to do this. By building a sufficiently large group of willing individuals with general skills and knowledge important for their involvement activities, the problem of ‘sick leave’ or ‘absence’ can be solved. Continuity can then be ensured via e.g. working in pairs to accommodate for absence. Regular training and creating experience with application of appraisal and evaluation criteria can be used to build up the group members’ capability. The voice of people with a chronic illness or disability representatives will be better heard and expectations, also from professionals in the research and care domains, will be better matched when they are well prepared, trained and supported with involvement activities. A bigger group of expert representatives with chronic illness or disability, reduces the gaps in the health related dialogue.

7.5.3.3 Believers in involvement

It requires ‘believers’ both to initiate and continue the work and to promote the lessons learned inside and outside the organisation of people with chronic illness or disability. It is important to develop sustainable involvement of people with chronic illness or disability structures. As this depends on devoted individuals, the continuity of the involvement of people with a chronic illness or disability remains vulnerable and uncertain. This is something to keep in mind as an organisation.

7.5.3.4 Societal relevance

The outcome of involvement by people with chronic illness or disability in research and care remains strongly dependent on the commitment by the different stakeholders. Ideally,
all stakeholders need to respect each other’s unique view and mutually allow and recognize their ideas. The views of people with chronic illness or disability are not replacing existing ideas of researchers, care providers and policy makers, but add different value. People with a chronic illness or disability address societal relevance of a subject while scientific quality and relevance are addressed and determined by professionals. By separating these two evaluative perspectives – scientific and societal – the competition on each other’s area of knowledge and experience is avoided. This enables the additional perspective to become an enrichment and as an added value for all parties.

7.5.3.5 *Integral vision on health*
People with a chronic illness or disability and their organisations strongly feel the need for a broad and a more integrated vision on sickness and health. They argue for an integral vision on well-being and articulate a need for a health care system that is organised accordingly. Policy makers in health care and health research professionals increasingly support this notion in, for example multidisciplinary integrative programs. This new perspective envelopes also psychological, spiritual and cultural matters besides disease. Underlying awareness that health, self-sufficiency and wellbeing are intertwined, and that this is affected by many different factors, already existed for many years (GR, 2010). This broader view on health and well-being matches with the experiences and impact of chronic illness and disability as experienced by the people with chronic illness or disability.

**7.6-CONCLUDING REMARKS**

The main research question is:

> What constitutes the body of experiential knowledge of people with a chronic illness or disability in the context of appraising quality of care and research?

This thesis investigates the content of experiential knowledge of people with a chronic illness or disability in in the context of appraising quality of care and research and how and under which conditions this may be included in the health debate and in the decision making process. In conclusion, the answers found to the three sub questions are:

1. *What matters to people with a chronic illness or disability concerning research and quality of care?*

The research in its entirety shows that their experiential knowledge consists of issues and values in an area, not just limited to health care or research, but reaching all aspects of life: from living circumstances to education, (paid or unpaid) from work to mobility. Truus, the author, considered this to be of such importance that she wanted to explore this and share
the results with other people, that she herself as a person with more than one chronic illness initiated this research. The results of this thesis show that there is a sequence in values and issues to be taken into account when involving people and organizing involvement. Currently, for some groups involvement is challenging: people whose basic values have not been met are not likely to participate in appraisal activities: participation in focus groups, fill out questionnaires and express opinions. In order to include the voices of people with a chronic illness or disability a dedicated and tailor made research approach is required.

2. According to which criteria do representatives of people with a chronic illness or disability appraise research and quality of care?

A set of six criteria was developed together with people having a chronic illness or disability for them to use when appraising or evaluating research and quality of care projects. This includes two process criteria that constitute a pre-condition for involvement and four content criteria: Relevance, Quality of life, Quality of care and Ethics & Safety. The set of 6 criteria which is developed in this research is -for the moment- suitable and workable for people with a chronic illness or disability who are advocates for their group. Practice experience so far has indicated that these six criteria provide something to hold on to, both literally (the criteria fan) and figuratively (a shortlist of relevant subjects to formulate their point of view on behalf of their group in a brief and structured way) In the future, actions are required to strengthen and support people with a chronic illness or disability speaking on behalf of a group as a representative. This thesis recommends a more structured way to advise, appraise or evaluate guidelines, activities, plans and research proposals.

3. How do representatives of people with a chronic illness or disability and professionals interact and relate to each other during involvement activities?

The results in this thesis show the necessity of further culture changes in governmental-, health research- and health care institutions. This may be directed towards increased acceptance and commitment among professionals so that the voices of people with chronic illness or disability are actually translated into research and quality of care. A tailored approach also includes taking into consideration that persons with a chronic illness or disability can be, on the one hand, strong and outspoken and on the other hand vulnerable and dependent. Involvement and partnership implies a two-way communication and respect for each other’s challenges and capabilities. It is the only way that complies with the current democratic and social vision on the collective health care and research system. All stakeholders -including weak and strong- must have the opportunity to bring their issues and concerns to the table and to be heard and their views being taken seriously. It is also the decent and respectful way to get well-balanced complex decisions made and make tough choices in a legitimate manner. It is not the involvement but it is the lack of involvement that leads to frustration and feeling ignored. It leads to a gap between people
with a chronic illness or disability and their advocates and health institutions, to conflicting positions and to legal action within health care relationships. All this may happen on individual micro-, but also on collective meso- and national macro-scale levels.

During the course of our research, we were fortunate to receive a confirmation of the value of our work by a Dutch organization that supports organisations of people with a chronic illness or disability in The Netherlands - PGO Support. This organization actively contributed to the dissemination of our criteria in the form of a ‘criteria fan’. This has made it possible for many people with a disease or disability and their organisations to benefit from this set of criteria. (Figure 4)

![Image of the criteria 'fan' as it is being used in practice.](image)

Several organisations adopted our ‘criteria fan’ either completely or in part for their own specific situation and members. For example, the Lung Foundation Netherlands (LFN) printed a criteria fan in their own house-style and distributed it among lung patients for their use. In two projects within the European Innovative Medicines Initiative (IMI) the set of criteria for appraisal and evaluation was translated into the English language and used by lung patients within the project. By using a set of criteria from their perspective these people could enhance involvement outcome and make involvement more feasible. We are proud we have been able to support this and will continue to strengthen chronically ill or disabled peoples’ involvement in research and quality of care.
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LIST OF ABBREVIATIONS & EXPLANATION OF TERMS

Abbreviations

CCI  Claims, Concerns and Issues
COPD  Chronic Obstructive Pulmonary Disease
CQi  Consumer Quality index
EC  European Commission
EPEI  European Patients Empowerment Index
GP  General Practitioner
GR  Gezondheidsraad (Health council)
ICF  International Classification of Functioning, Disability and Health
IMI  Innovative Medicines Initiative
LFN  Long Fonds (Dutch Lung Foundation Netherlands)
Long Fonds  a charitable organisation (health foundation) and lung disease patient organisation in the Netherlands
NHS  National Health Service (in the UK)
PGO Support  Independent network in the Netherlands for people with illness and disability and their organizations. They recommend these organizations and support initiatives that strengthen their position.
PPI  Patient and public involvement
PPP  Patient Public Partnership
PROM  Patient Reported Outcome Measure
Qaly  quality-adjusted life year, a measure of disease burden, including both the quality and the quantity of life lived
RGO  Raad voor Gezondheids Onderzoek (Health research council)
RMO  Raad voor Maatschappelijke Ontwikkeling (Societal development council)
RVZ  Raad voor Volksgezondheid en Zorg (Health care council)
TOOL2USE  Foundation dedicated to improve the expertise of people with a chronic disease or disability in the field of patient advocacy
VSB fonds  Funding organisation promoting active societal participation for all.
VU  Vrije Universiteit
WAC  Wetenschappelijke Advies Commissie (Scientific advisory committee)
WHO  World Health Organisation
ZonMw  Government organisation financing scientific research and health oriented development programmes.
Explanation of terms

Health - The ability of people to adapt and be in control when confronted with the physical, emotional and social challenges of life. Illness must be treated where possible but in addition to that the resilience, self-management and health literacy requires attention.

Disease/Disorder/Illness – Pathological condition of a part, organ or system in the body, identifiable by symptoms leading to an unhealthy condition of body or mind.

Chronic disease – a disease lasting for a long time that does not heal.

Co-/Multi morbidity – the presence of more than one disease.

Handicap – General term identifying disabilities and disorders in activities and participation.

Disability – a physical or mental impairment leading to the inability to perform specific tasks or functions.

Experiential knowledge – experience about living with an illness or disability of individual people with an illness or disability that can be shared with others.

Experiential expertise – Experiential knowledge gathered from many people that exceeds the level of individual interactions between patients, clients, treating medical experts, and health researchers and that exceeds the level of a single disease or disability.

Patient involvement – people with an illness or disability are engaged as participants in decision making about health research and health care. They are advocates or representatives for their organisations and group. This is done on a collective level between the advocates or representatives and health care institutions, research centres, health insurance companies and the government.

Participation ladder – citizens and therefore also patients and clients, can take part in the health care system in different roles. They can be passive, can provide information, be an advisor, be a decision partner or take full control.

Mourning phases – according Kübler-Ross model, referred to as the "five stages of grief": Denial, 2-Anger, 3-Fighting, 4-Depression, 5-Acceptance

Recursive – A process which is repeated within itself while getting smaller.

Relevance – Issues, concerns or feelings that matter for a person or group.

Right to say – To have the right to voice one’s opinion.

Ethics – Principles of right conduct and a system of moral values.

Safety – feeling or being free of danger, risk or injury.

Instrument – a means by which something is done, a tool, an equipment or method.

Issues – The term issues is an abbreviation of ‘claims, concerns and issues’ (cci’s) which is commonly used in scientific literature.
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The research was carried out with program funding of ZONMw/VSBB “Patient involvement in research, quality and policy”.
Both PGO Support and the Lung Foundation used one of the results of this thesis’ research - the appraisal and evaluating criteria from the perspective of people with illness or disability - and took this into production within their own organisation and with their identity and distributed it among their own members.
The VU University Metamedica/EMGO+ and the foundation TOOLS printed a summary of this thesis in lay language made by Truus and made that available to people with a chronic illness or disability and their member organisations.
Dankwoord


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SUMMARY

VALUES AND CRITERIA OF PEOPLE WITH A CHRONIC ILLNESS OR DISABILITY

Strengthening the voice of their representatives in the health debate and the decision making process.

INTRODUCTION

The number of people in the Netherlands with an illness or disability is on the rise, and the same trend is also observed in many other European and non-Western countries. This expanding group of people has experience and knowledge about what it means to live with chronic illness or disability and about the consequences for themselves, their immediate environment and for society. This knowledge and experience, usually accumulated over the years, and in many cases in interaction with professional and even scientific knowledge, is generally referred to as experiential knowledge.

With their experience and knowledge of their fellow members, representatives of people with chronic illness or disability are increasingly serving as partners in the health debate and in negotiations on health care and research. The idea behind this is that the quality of health projects improves, e.g. on guidelines or scientific research, is a better reflection of their voice, and what these people contribute leads to a much more complete picture. Moreover, this is a normative ideal involvement in quality and research and gives voice to those to whom it applies: people with a chronic illness or disability.

To date little is known about how representatives of people with a chronic illness or disability experience their involvement activities. Criteria are not existing or incomplete and people are not fully aware of the issues that matter to their peer group. Not clear, for example, are the criteria on which their appraisals of research and care should be based.

The purpose of this study is to investigate what constitutes experiential knowledge of people with a chronic illness or a disability, and how their issues are and can be used in involvement practices by their representatives. Three aspects need to be addressed. First it is important to investigate what matters to the people, and what are shared issues of concern. Second, it is crucial to investigate what criteria representatives use to appraise care and research from a patient perspective. Third in which way representatives can put these matters on the table in the interaction with professionals. All this does not relate to the individual patient-caregiver interaction nor to individual experiential knowledge but to
the widely shared experiences and opinions of people with a chronic illness or disability on a collective level. This leads to the main research question for this thesis:

What constitutes the body of experiential knowledge of people with a chronic illness or disability in the context of appraising quality of care and research?

Together with these people, and based on this assessment and knowledge, we developed appraisal criteria to be used by their representatives who assess and evaluate on their behalf. This will give them something to hold on to and hence more support when they are at the table with scientists, policymakers, managers, care professionals and the many other discussion partners they meet during review processes and in the debate in general. This means that the values and issues of the individual members of their peer group can be put forward more effectively.

To this end we examine the following in this study:

1. What matters to people with a chronic illness or disability concerning research and quality of care?
2. According to which criteria do representatives of people with a chronic illness or disability appraise research and quality of care?
3. How do representatives of people with a chronic illness or disability and professionals interact and relate to each other during involvement activities?

This study examines the involvement of advocates and involvement of groups, and the widely shared experiential knowledge of people with a chronic illness or disability that are put forward in the health debate and decision-making processes in research for them, or on their behalf.

THEORETICAL FRAMEWORK

We assume in this study a theoretical conceptual framework based on the following concepts:

Experiential knowledge: ideas, experience and knowledge from the environment in which people live their lives, can contribute to better research and improve the quality of care. Scientific research and guideline development are processes derived from science-based principles and criteria, and are ‘evidence based’.

Experiences or principles based on practical experience; usually referred to as ‘practice-based’, hardly ever play a role in traditional scientific knowledge and guideline development. There is in fact disparity when comparing or appreciating the two fields of scientific knowledge and experiential knowledge.
The main characteristic of experiential knowledge is that it is practically relevant and applicable. Experiences become experiential knowledge as soon as they are recognized by a larger group of people with a chronic illness or disability. Experiential experts would also have the capacity or ability to learn to reflect on their experiences and knowledge, and to be able to view them from multiple angles and identify with stories of fellow peer group members.

Involvement of people with a chronic illness or disability is largely based on their experiential knowledge. The concept behind experiential knowledge is that ideas, experience and knowledge of those with an illness or disability covers all aspects of their life, is intertwined with the illness or disability concerned and comes from the environment where people lead their life. Involvement in for instance guideline development is justified because issues from this personal life-world environment are very different from issues important in the scientific and professional environment. Additionally the concept assumes that interaction between the two environments is both possible and necessary.

**Partnership:** the experiences of people with a chronic illness or disability are a source of experiential knowledge and are used in decision-making processes. A second prerequisite is that these people are seen as equal partners in the knowledge acquisition process. They are actively and equally involved in the development, implementation and evaluation of the research, the quality of care projects and policies. There are several parties in the dialogue all with differences and similarities in their views and perspectives. The idea is that all parties in an on-going dialogue develop mutual understanding of each other's positions, thereby enabling new opportunities and ideas. When a specific group of people are barely involved at all in participation practice, or do not actively participate or truly join in with discussions on a major activity, then what we are looking at is non- or pseudo-participation. Ideas about commitment, equality, equal rights, equal opportunities and being heard are all closely linked. Involvement and participation are often cited as the opposite of exclusion, which is about more than just a lack of participation in decision-making processes. Whether or not to participate in society is directly related to groups of people who, in one way or another, are behind or disadvantaged and marginalized in society. Participation and empowerment are closely linked.

**Responsive research design and research methods:** This thesis follows a responsive research design approach in which an inventory of the issues of different stakeholders (people both interested in the study and important to the study) were assembled and compared for comparison. An attempt was made to establish the issues and their value and meaning, by means of a common search strategy together with stakeholders. The idea is that mutual understanding between stakeholders is, in itself, a requirement for achieving improvements in practice. Various research methods were used to identify, elaborate and exchange issues and values among stakeholders and in a dialogue with researchers.
Firstly, an auto-ethnographic research type based the story of Truus, the first author, who lives with multiple chronic diseases, is reported on in Chapter 2. The research method used is a narrative data collection and analysis and was conducted in order to record and analyse the experiences, feelings and events in her life with multiple diseases.

In Chapter 3 we present a qualitative exploratory research type to get an idea of the experience, important issues and values of people with chronic illness or disability in practice. As method we drew up a content analysis of relevant documents, and interviews were held and analysed and participant observations performed. Areas outside traditional medical science were also examined to find those issues relevant to people with chronic illness or disability.

Chapter 4 describes a literature review type study to identify subjects and values of people with chronic illness or disability as they are currently identified by science. International scientific literature and content analysis was carried out as research method. In addition to the search in primary (scientific sources) literature, information was also collected to this end from secondary and tertiary sources (databases of organisations of people with chronic illness or disability, non-scientific publications, reports and information).

The research type we used in chapter 5 is a qualitative type study and as a research method we used in-depth interviews, telephone interviews and focus groups. Also we used content analysis thematic analysis en data syntheses. Focus groups were held to find out which topics are important for people with chronic illness or disability. These issues were analysed and then combined into a set of criteria that was discussed, revised and prioritized by the people themselves in a series of focus groups, roundtable discussions and interviews. This was done repeatedly with different participants each time.

In chapter 6 we use three different research methods, all of the qualitative research type. The chapter presents three studies that describe the participation of people with chronic lung disease. Section 6.1 presents a longitudinal case study with document search, interviews and observations and a time cross section analysis. We investigate how and to what extent lung patients participated over the past 7 years in Lung Foundation Netherlands (LFN) funded lung research. Section 6.2 we use a qualitative study as a research method and we use questionnaires, interviews and focus groups for data collection and content analysis for the data analysis. We examine how the LFN sets the research agenda together with people with a lung disease. Section 6.3 uses also a qualitative study as a research method with in-depth interviews for data collection and content analysis to analyse the data. We investigate shared responsibilities and involvement in the practice of people with Chronic Obstructive Pulmonary Disease (COPD) and their caregivers, via an interactive method.
A COUNTER STORY

The author’s search and the questions that gave rise to this research and thesis are presented in Chapter 2 about “struggling between strength and vulnerability, a patient’s counter-story”. Living with multiple chronic diseases and disabilities, she describes her 27 year’ struggle from the moment she became ill to the present. This led to a counter-story based on a number of critical moments in her life. Her struggle with strength and vulnerability exposed another side to people with illness or disability, which contrasts with the articulate, independent patient that is increasingly talked of in health care. The prevailing image is that of sturdy citizens whose own choices and control of their life are firmly in their own hands.

This story shows that things are not that simple: the patient will experience everything between strength and vulnerability, and between constraints and opportunities. Recurrent episodes of loss and mourning, and development and growth alternate with each other. People with a chronic illness or disability can be fully occupied merely by their basic needs and physical survival. This may involve work, attending courses or school, having to stop a hobby, cancelling an appointment with friends yet again, or constraints to the support they can give to their peer group – and all have major implications for them to have their say in processes and participation in society.

At any given time a reversal may need to be recognized in the life of someone with an illness or disability: renewed growth, and an opportunity to focus on the immediate environment. Particularly important is that strength and vulnerability i.e. both sides that are part of ‘being someone with a chronic illness or disability’, are recognized. This does justice to their abilities and their occasionally limited capabilities.

By linking various theories to this story it becomes clear that not all people with a chronic illness or disability can be empowered and self-reliant throughout their lives. The image that depicts people with a chronic illness or disability as ‘healthcare consumers' with control over their own lives may sometimes be true, but more often than not it is not the case.

Living with a chronic illness or disability may be associated with feelings of sadness and loss, but also with unmet basic needs, such as a sense of security. Once the chronic illness or disability has been accepted, feelings of grief and loss can return again in full force, triggered by an event or by deteriorating chronic illness or disability. These stages of grief and unmet needs can crop up, recursively, several times throughout the life course.

When living with a chronic illness or disability, it is not important whether someone is a ‘mature healthcare consumer' or a ‘dependent insecure patient’. But it is a fact that he or she regularly experiences both states, and needs to learn to cope, together with others in the vicinity, with both aspects simultaneously. A welcoming environment for participation of people with a chronic illness or disability would take account of both their strength and their vulnerability.
EXPLORING CRITERIA OF PEOPLE WITH A CHRONIC ILLNESS AND DISABILITY IN THE NETHERLANDS

The representatives of people with a chronic illness or disability participate in projects on research and quality of care in the Netherlands. The problems they encounter while doing so are investigated in Chapter 3 about “Patients at the Negotiating Table. Exploring appraisal criteria of Health Research and Quality of Care used by Patient Advocacy Groups in the Netherlands”. This study is a preliminary exploration in support of the definition of the research described in this thesis.

It investigates the current practice of assessment and appraisal conducted by people with an illness or disability used in the evaluation of scientific research and quality of care. The results of this study enabled us to ascertain where the problems lie, and to clarify the research questions.

Three initial criteria in the areas of policy, research and quality of care, emerged from this initial exploratory study. These three criteria were found to be important to representatives of people with chronic illness or disability. These criteria were: relevance, empowerment, and ethics & safety. The demand for criteria and for practical instruments or methods to be used by representatives of people with a chronic disease or disability was loud and clear. These people need more support and something to hold on to during their involvement. This would have to be in the form of a tool or reference document. A need was also felt for more structured appraisal using a limited number of subjects important to their peer group. With these subjects and several pre-prepared questions they would be able to appraise a document more quickly and find arguments in a discussion more easily. Finally the study identified the need for training and support before and during appraisal activities.

INVENTORY OF ISSUES THAT MATTER FROM LITERATURE

Chapter 4 is about “Patient issues in health research and quality of care: an inventory and data synthesis”. It describes an international literature study into the issues that matter to people with a chronic disease or disability in the domains health research and quality of health care. The results provide an inventory of these issues as far as they are known and acknowledged in the scientific literature. It soon became clear that there is very little about such issues in primary scientific sources. Publications from governments, international organisations and information provided by groups of people with a chronic disease or disability themselves, were needed in order to get a more complete picture. These publications are usually referred to as secondary and tertiary or ‘grey’ literature.

A total of 35 issues that matter to people with a chronic disease or disability were established. Some 13 of these issues were only found in tertiary or ‘grey’ literature. These 13 ‘missing’ issues (privacy, relevance, access, gender, pain, age, fear, consent,
communication, education, complaints, compensation and alternative methods) reveal a gap in the scientific literature, and mainly involve fundamental and personal values and quality of life issues (Table 1).

Table 1 Issues that matter to people with chronic illness or disability

<table>
<thead>
<tr>
<th>No.</th>
<th>Issue</th>
<th>Description</th>
<th>Found only in grey Literature</th>
<th>No.</th>
<th>Issue</th>
<th>Description</th>
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<tr>
<td>1</td>
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<td>22</td>
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<td>23</td>
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<td></td>
<td></td>
<td>28</td>
<td>education</td>
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<td></td>
<td>29</td>
<td>gender</td>
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<td>10</td>
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<td></td>
<td></td>
<td>30</td>
<td>pain</td>
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<td>32</td>
<td>age</td>
<td></td>
<td>x</td>
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<td>compensation</td>
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<td>14</td>
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<td></td>
<td></td>
<td>34</td>
<td>fear</td>
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<tr>
<td>15</td>
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<td></td>
<td></td>
<td>35</td>
<td>alternatives</td>
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</tr>
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<td></td>
<td>x</td>
<td>17</td>
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<td></td>
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<td>21</td>
<td>access</td>
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</tr>
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STRUCTURED APPRAISAL AND EVALUATION IN PRACTICE

Chapter 5 is about “Structuring patients advocates’ appraisal and evaluation of health research and quality of care”. The issues, values and criteria that are relevant to people with a chronic illness or disability in practice were further investigated in this chapter. In a series of focus groups, round table discussions and interviews, a draft set of criteria was debated, adjusted and prioritized by people with a chronic illness or disability. The initial aim of the research was to find values behind the issues that are important to these people in their daily life. This resulted in 4 different value group categories: fundamental, quality of life, quality of care, and empowerment / involvement.
The term ‘fundamental’ value in this thesis is used to identify those values that are of the utmost importance to people. These values relate to basic conditions to be able to function socially and psychologically. The participants in this study identify these fundamental values as being the most important. Only after these issues have been taken care of is a discussion about the aspects of quality of life possible. Only after the first two values have been secured, can the issues relating to the third and fourth value groups, i.e. quality of care and involvement respectively, be the subject of discussion and activities. For example, participants clearly state that feeling safe and respected is more important than good communication with a care professional or a say in the treatment process. This sequential order of importance can be depicted as an ‘involvement iceberg’ (Figure 1). Only the tip of the iceberg is above water level. This represents the people who are involved, take part in discussions, whose voices are heard, and who have secured the fundamental and quality of life value related issues in their life.

Figure 1 The involvement iceberg showing value-related issues
The major part of the iceberg is below water level and is largely invisible. This represents issues related to the fundamental and quality of life value levels that remain unseen in society. Society has therefore a biased view of people who are sturdy and on top of things in their life. The question is how can society listen to those who have not secured fundamental and quality of life related values, since they are ‘below the water line’. For the time being they remain largely invisible and unheard.

The 35 issues were used to generate a set of criteria with a view to making the issues and values useful and practicable for involvement practice. This work resulted in a ‘criteria fan’. Representatives of people with a chronic illness or disability have evaluated this set of criteria as a sound reflection of their perspectives and as a useful instrument and tool in their involvement activities. The ‘criteria fan’ would appear to be seen as a practical tool for structured appraisal of research proposals and for the evaluation of guidelines or as a reference in health-oriented debate. The criteria are about relevance to people with a chronic illness or disability, about quality of life, quality of care, ethics and safety aspects, information and communication and finally about involvement. The ‘criteria fan’ is currently used in the field by representatives of people with a chronic illness or disability, and also by professionals at various health foundations and even in two European lung research projects (Figure 2).

Figure 2 Examples of the ‘criteria fan’ currently in use
IMPLEMENTING ISSUES AND VALUES OF PEOPLE WITH A CHRONIC LUNG ILLNESS

Chapter 6.1 about involvement of people with chronic lung illness, presents a seven-year case study that highlights the path to the current situation with people having a chronic lung illness being involved in the appraisal of scientific research proposals at Lung Foundation Netherlands (LFN). Implementation and practice of lung patient involvement practice in the LFN research cycle were described and analysed. This study investigated the basis for selection of research themes and which funding criteria were leading within LFN. The involvement of lung patients was then studied.

The scientific and societal relevance of research proposals are currently evaluated by all stakeholders. This was made possible by implementing an involvement method including a set of criteria from the point of view of people with lung disease. Implementing this involved training a group of people with lung disease and providing them with the developed appraisal and evaluating tool to evaluate the societal relevance from the patients’ perspective. Scientific relevance and quality are evaluated and traded off separately in the research proposal approval or rejection process. This seven year case study provides an insight into the commitment of people with a chronic lung illness in the research cycle and into the pitfalls and complications that may arise.

Two related studies are included in this thesis since they elaborate on several points described in section 6.1. The study in section 6.2 describes the increasing level of involvement of people with chronic lung illnesses in LFN agenda setting since 2004. Although research priorities set by professionals and by patients are quite similar, the details tend to differ. People with a chronic lung illness give different priorities to specific subjects in lung research, for example aspects of fatigue, co-morbidity, and side-effects.

Shared responsibilities between people with Chronic Obstructive Pulmonary Disease (COPD) and health care professionals in practice are dealt with in section 6.3.

On one hand the dialogue between peer lung patients is very important. The safety of a group and sharing experiences among peers turns out to be an important means to give new meaning to the illness in the lives of people with COPD. On the other hand people with chronic illness or disability are expected to play an important role in their own recovery. This has consequences for the division of responsibilities among medical professionals and the people themselves. In-depth interviews with people with COPD, physiotherapists and a lung practitioner demonstrate that a shift in responsibilities requires a supporting attitude on the part of care providers and dialogue between patients and care professionals. People with COPD become co-owners of the recovery process. The notion that COPD is more than just a lack of air, and that mind and body mutually affect each other, is a first step towards investigating other problems in their life, and towards improving their quality of life. Exchanging ideas with respect for each other’s views is important here.
GENERAL DISCUSSION

The research questions are answered in Chapter 7 and recommendations are made for further research and for involvement practice. The three main findings in this thesis’ research are presented here.

The first main finding is the formulation of issues and values that matter to people with a chronic illness or disability. They constitute part of their experiential knowledge. The research resulted in 35 issues (see Table 1) that are important to these people in their life with illness or disability since they affect their functioning on societal and mental levels and their ability to join in with involvement activities. With the help of the values it was possible to divide the issues up into four different sequential groups: fundamental human values, quality of life, quality of care, and involvement.

The second main finding is the set of appraisal and evaluation criteria coming from and to be used by representatives of people with a chronic illness or disability during the evaluation of health research and quality of care. This set of six criteria, derived from the 35 issues, includes all the issues that matter to people with a chronic illness or disability. This set of criteria can be used during involvement activities, e.g. on appraisal of research proposals and on guideline development.

The set of criteria is about the relevance of quality of care or health research activities as seen from the perspective of these people. The criteria are also about whether the diagnostic or treatment activity improves their quality of life.

Further criteria involve ethics and safety aspects and quality of health care and health research activities. Furthermore the criteria are about information that can be well understood, e.g. using lay language, and about careful communication, e.g. the feedback of results to the group of people with the illness or disability concerned, and their organisation. Currently 1 set of criteria is being used for appraisal of research proposals and evaluation of e.g. health standards or guidelines. Comments from the field indicate separate sets of criteria might be useful for the research and care domains. Specific terms and context might be used. Further development of these is being planned for in 2014.

Finally the criteria are about involvement itself and the question as to whether a project or research are also based on ideas and needs expressed by people with a chronic illness or disability. The criterion about ‘involvement’ includes the issue ‘right of say’ which identifies it more as a process criterion than a content criterion. This criterion also addresses the point of these people being an equal partner in the project or activity and whether they are not only allowed to have a voice but may also be truly involved in making decisions.

The third and final main finding is about the relationship between professionals and the involved people with a chronic illness or disability. This relationship can facilitate a kind of
partnership which prevents parties from opposing one another. This kind of partnership requires commitment and support by all participants about the usefulness and added value of including the perspective of people with illness or disability. Linking the different perspectives of these people and the perspectives of professionals leads to a qualitatively better outcome that enhances current practice. This leads to research and guidelines that are both scientifically sound and scientifically and societally relevant in the eyes of people with a chronic illness or disability.

**RECOMMENDATION FOR FURTHER RESEARCH**

*Changing balance*: the first recommendation is the question as to whether the presence of advocates of people with a chronic illness or disability and their voice actually disturbs the existing power balance, and if so, to what extent. What happens when people do not adjust to the dominant discourse in health care or research and introduce their own counter-story, as did the author of this thesis? What happens when their views are fundamentally different from those of other participants? Are they placing themselves outside the group or the debate altogether, or can this counter-story have some influence? Or is the balance of power between people with a chronic illness and professionals slowly shifting towards more equal roles in the debate? Are they, in other words, moving towards true partnership? It would be interesting to investigate these questions since they could shed light on how the voice of people with a chronic illness or disability could be more empowered within existing power relations and processes.

*Vulnerable people*: further research is also recommended into the question of how the experiential knowledge of vulnerable people who are not heard or who do not voice or express their views, can be used and utilized in their own interest for the appraisal of research and care activities. How can their experiential knowledge be included and seriously considered in health debate or decision making?

*Stories as an instrument to gain insight*: it is finally recommended that alternative ways for people with a chronic illness or disability be further investigated with a view to understanding their situation and their life with its constraints and limitations. In Chapter 2 the author (Truus) wrote her story following the path between several critical moments in her life surrounded by illness and disability. She developed her story and reflected on it together with two others. This increased her insight into, understanding of, grip on and perspective in her life. Research could clarify whether this approach, or other modes of creative expression such as poetry or other artistic representations, also have a positive effect on other people with a chronic illness or disability.
RECOMMENDATIONS FOR PRACTICE

**Monitoring:** listening to people with an illness or disability should be permanent practice in health care on all levels. This is what the WHO states in its health policy on citizens’ rights and patient involvement (Health 21) for the European region. Ethics and human rights are mentioned in several generally accepted documents but they are not legally binding and are therefore not compulsory. There is therefore no guarantee that they are applied. It is recommended that organisations of people with a chronic illness or disability monitor this closely.

**Value structure:** values are a main driver behind human behaviour. Issues that matter to people with a chronic illness or disability are closely linked to values. A recommendation is to further implement the value structured criteria (iceberg), resulting from the research in this thesis, in practice.

**Counter-stories:** the dominant concept of resilient, sturdy and empowered people with a chronic illness or disability is under debate. These people are not mere health consumers with control over their own illness or disability. Other views, described in alternative or ‘counter-stories’ need to be taken up and considered in the health debate.

**Pictograms and symbols:** people with a mental disability or illness, e.g. Alzheimer’s, but also illiterate people, require a special version of the criteria developed in this thesis’ research. The choice of words, illustrations and pictograms needs to be adapted to their needs. Some people recognize a pictogram much more easily than they can read a text. Pictograms are also interpreted more accurately than words. The ‘criteria fan’ is a living document. Extending the criteria with other specific subjects and problems related to the daily lives of specific groups of people is both desirable and possible.

**Partnership:** people with a chronic illness or disability are still regarded as a threat to existing power relations in a project or a committee. Accordingly, their relatively new voice at the negotiation table is not seen as a positive development but as a formality that needs to be accepted but which is, in fact, not taken seriously. A clearly undesirable kind of involvement is apparent when people are invited merely for their formal presence, which is referred to as ‘pseudo-involvement’. It is recommended that people with a chronic illness or disability only agree to be involved if it is on an equal footing with others occupied with discussion and decision making in a project, committee or debate.

**Capacity building:** people with a chronic illness or disability often find it difficult to ensure continuous involvement due to absence because of illness. A practical solution to this would be to be involved and work in duo-teams. This requires the availability of a sufficiently large number of people with a chronic illness or disability active in involvement on a regular basis. This calls for these people to be trained and educated in involvement activities, in relevant developments, and they need to know the facts about their peer
group. The larger the group of capable people available, the fewer problems with absence and interruption of the health dialogue and decision-making processes.

It is recommended that extra effort be put into reinforcing and supporting people with a chronic illness or disability who represent their peer group and who are involved in discussions and decision making. All stakeholders, both vulnerable and strong, must be given an opportunity to table their issues, to be heard and to be taken seriously.
CRITERIA FAN

1. **RELEVANCE**
   1.1 Does this improve the health of the people with illness or disability?
   1.2 Is their diversity being taken into account?
   1.3 Is this based on their questions and needs?
   1.4 Can the project results be used in practice?

2. **QUALITY OF LIFE**
   2.1 Does this improve the health related quality of life of the people with illness or disability?
   2.2 Is their societal participation being improved?
   2.3 Are their fundamental values being respected and being used as guiding principle?
   2.4 Are the project results beneficial to them in practice?

3. **QUALITY OF CARE**
   3.1 Does this improve health care for the people with illness or disability?
   3.2 Is their diversity being taken into account?
   3.3 Are their social network and environment being taken into account?

4. **ETHICS AND SAFETY**
   4.1 Are the means (e.g., pain, absence of work or school) justified by the goals (e.g., better medication)?
   4.2 Is the safety of participants, patients and test persons being ensured?
   4.3 Are people concerned having explicit freedom of choice?
   4.4 Are their social norms, values and dignity being respected?
   4.5 Are the health professionals respecting rules, codes of practice and guidance?

5. **INFORMATION AND COMMUNICATION**
   5.1 Can the information be properly understood by the people with illness or disability?
   5.2 Are these people and their organisations being informed about project results?
   5.3 Is the information provided correct and complete, also on possible complications?
   5.4 Are these people having the possibility to explicitly agree or refuse (informed consent)?

6. **IN卷VOLVEMENT (precondition)**
   6.1 Are the interests of people with illness or disability taken into account?
   6.2 Are they being included as equal discussion partners?
   6.3 Are they being regarded as full citizens?
   6.4 Is their experience included as a source of experiential knowledge in project, guidance or research?
   6.5 Are they being supported in their involvement effort?
SAMENVATTING

WAARDEN EN CRITERIA VAN MENSEN MET EEN CHRONISCHE ZIEKTE OF BEPERKING

Het versterken van de stem van hun vertegenwoordigers in het gezondheidsdebat en het besluitvormingsproces.

INLEIDING

In Nederland neemt het aantal mensen met een chronische ziekte of een beperking toe. Dat geldt ook voor veel andere Europese en niet-Westere landen. Deze steeds groter wordende groep mensen heeft ervaring met en kennis over wat het betekent te leven met een chronische ziekte of beperking. Zij hebben aan den lijve ervaren welke gevolgen dat heeft voor henzelf, de naaste omgeving en de maatschappij. Deze kennis en ervaring, die vaak gedurende jaren is opgebouwd, al dan niet in interactie met professionele en wetenschappelijke kennis, wordt ook wel ervaringskennis genoemd. Met hun eigen ervaringskennis en die van de achterban zijn vertegenwoordigers van mensen met een chronische ziekte of beperking steeds vaker een gesprekspartner in het zorgdebat en bij onderhandelingen over de zorg en gezondheidsonderzoek. Het idee hierachter is dat de kwaliteit van gezondheidsprojecten, zoals richtlijnen en wetenschappelijk onderzoek, beter en completer wordt van de inbreng door mensen met een chronische aandoening. Maar bovenal is dit een normatief ideaal: inspraak in kwaliteit en onderzoek geeft diegenen een stem, om wie het uiteindelijk gaat: mensen met een chronische aandoening of een beperking.

Tot nu toe is er onvoldoende bekend over de ervaringen van vertegenwoordigers van mensen met chronische ziekte en beperking met participatiepraktijken. Criteria ontbreken of zijn niet compleet en mensen zijn zich niet bewust van de onderwerpen en waarden die voor hun groep belangrijk zijn. Onduidelijk is bijvoorbeeld op grond van welke criteria zij onderzoek en kwaliteit van zorg zouden moeten beoordelen.

Het doel van dit onderzoek is om te bepalen wat de samenstellende delen van de ervaringskennis van mensen met een chronische ziekte of beperking zijn, en hoe hun onderwerpen in de praktijk bij participatie kunnen worden gebruikt door hun vertegenwoordigers. Drie aspecten moeten hierbij een rol spelen.
Ten eerste is het van belang om te onderzoeken welke onderwerpen deze groepen mensen belangrijk vinden, welke onderwerpen aanleiding tot bezorgdheid geven en breed gedeeld worden.

Ten tweede is het cruciaal om vast te stellen welke criteria vertegenwoordigers van deze groepen gebruiken om vanuit patiënt perspectief onderzoek en zorg te beoordelen.

Ten derde is de manier van belang waarop de vertegenwoordigers de onderwerpen in de interactie met professionals aan de onderhandelingstafel in brengen.

Het gaat hier niet om de individuele patiënt-behandelaar of zorgverlener relatie en ook niet over individuele ervaringskennis, maar het gaat over de breed gedeelde ervaringen en meningen van mensen met een chronische ziekte of een beperking op collectief niveau.

Dit leidt tot de centrale onderzoeksvraag voor dit proefschrift:

_ Waaruit bestaat het geheel van ervaringskennis van mensen met een chronische ziekte of een beperking in de context van hun beoordelen van kwaliteit van zorg en van onderzoek ?_

Samen met deze mensen, en gebaseerd op deze taxatie en kennis ontwikkelden wij beoordelings-criteria bedoeld voor gebruik door hun vertegenwoordigers bij het namens de groep beoordelen en evalueren. Deze criteria geven houvast en dus steun aan de tafel waar zij wetenschappers, beleidsmakers, managers, zorg professionals en vele andere discussiepartners ontmoeten bij evaluatie, beoordelen en debat in het algemeen. Dit betekent dat zij de onderwerpen en waarden die bij hun groepsgenoten leven, meer effectief naar voren kunnen brengen.

Met dit doel voor ogen is in dit proefschrift het volgende onderzocht:

1. _Wat is belangrijk voor mensen met een chronische ziekte of een beperking als het gaat over onderzoek en kwaliteit van zorg ?_
2. _Met welke criteria beoordelen vertegenwoordigers van mensen met een chronische ziekte of een beperking onderzoek en kwaliteit van zorg ?_
3. _Hoe is de onderlinge interactie en relatie tussen vertegenwoordigers van mensen met een chronische ziekte of een beperking en professionals tijdens participatie activiteiten ?_

Deze studie onderzoekt de participatie van vertegenwoordigers en van groepen van mensen met een chronische ziekte of een beperking. Het gaat daarbij om hun breed gedeelde ervaringskennis, die in het gezondheidsdebat en in besluitvormingsprocessen bij onderzoek naar voren wordt gebracht voor hen of namens hen.
THEORETISCH KADER

In dit onderzoek gaan we uit van een van een theoretische denkkader dat gebaseerd is op de volgende ideeën:

**Ervaringskennis:** Ideeën, ervaring en kennis, afkomstig uit de omgeving waar mensen hun leven leiden, kunnen bijdragen aan beter onderzoek en het verbeteren van de kwaliteit van zorg. Wetenschappelijk onderzoek en richtlijn ontwikkeling is een proces dat is gebaseerd op wetenschappelijke gebaseerde uitgangspunten en criteria, het is 'evidence based'.

‘Ervaringen’ oftewel op de ervaringspraktijk gebaseerde uitgangspunten, ook wel ‘practice based’ genoemd, spelen nog nauwelijks een rol in de traditionele wetenschappelijke kennis en richtlijnontwikkeling. Vooralsnog is er geen sprake van gelijkheid bij het vergelijken of het waarderen van de twee gebieden van kennis, wetenschappelijke kennis en ervaringskennis.

Kenmerk van ervaringskennis zijn dat deze vorm van kennis praktisch relevant en toepasbaar is. Ervaringskennis wordt ervaringsdeskundigheid als de kennis wordt herkend door een grotere groep mensen met een chronische ziekte of beperking. Tevens zou een ervaringsdeskundige het vermogen hebben of kunnen aanleren om te reflecteren op diens ervaringskennis en het van meerdere kanten kunnen bekijken en verbinden met verhalen van lotgenoten.

De participatie van mensen met een chronische ziekte of beperking is grotendeels gebaseerd op hun ervaringskennis. Het idee achter ervaringskennis is dat ideeën, ervaringen en kennis van diegenen met chronische ziekte of beperking met heel hun leven van doen hebben, nauw verwikkeld zijn met de ziekte of beperking zelf en dat die afkomstig is uit de omgeving waar deze mensen hun leven leiden. Participatie in bijvoorbeeld richtlijn ontwikkeling is gerechtvaardigd omdat onderwerpen die uit de persoonlijke leefwereld komen zeer verschillend zijn van de onderwerpen die uit de wetenschappelijke en professionele omgeving komen. Daarbij veronderstelt het participatie concept dat de interactie tussen de twee omgevingen zowel mogelijk als noodzakelijk is.

**Partnerschap.** Ervaringen van mensen met een chronische ziekte of beperking zijn een *bron van ervaring* en kennis en worden gebruikt in besluitvormingsprocessen. Een tweede uitgangspunt is dat deze mensen *gelijkwaardige partners* in het proces van het verwerven van kennis. Ze zijn actief en gelijkwaardig betrokken bij de ontwikkeling, uitvoering en evaluatie van het onderzoek, de kwaliteit van de zorg projecten en beleid. Verschillende partijen in de dialoog hebben zowel verschillen als overeenkomsten in hun opvattingen en perspectieven. Het idee is dat in een voortdurende dialoog alle partijen wederzijds begrip ontwikkelen voor elkaars standpunten waardoor nieuwe mogelijkheden en ideeën mogelijk zijn.
Responsief onderzoeksdesign en onderzoeksmethoden: Dit proefschrift volgt een responsieve onderzoekbenadering waarbij de issues van verschillende stakeholders (mensen die zowel van belang zijn voor als belang hebben bij het onderzoek) zijn geïnventariseerd en in dialoog zijn gebracht. Door middel van een gemeenschappelijke zoekstrategie wordt wederzijds begrip tussen stakeholders gezocht: een voorwaarde om verbeteringen in praktijk te bereiken. Diverse methoden van onderzoek zijn ingezet om stakeholderissues te inventariseren, te verdiepen en in dialoog uit te wisselen.

Allereerst wordt in hoofdstuk 2 middels een auto-etnografisch onderzoek type verslag gedaan van een onderzoek verricht aan de hand van het verhaal van Truus, de eerste auteur, die leeft met meerdere chronische ziekten, een zogenoemde auto-etnografie. Via narratieve analyse zijn haar ervaringen, gevoelens en gebeurtenissen rondom het leven met meerdere chronische ziekten opgetekend en geduid.

In hoofdstuk 3 presenteren we een via een kwalitatief verkennend onderzoek type verkregen analyse om zicht te krijgen op de ervaringen, belangrijke onderwerpen en waarden van mensen met een chronische ziekte of beperking in de praktijk. Ook gebieden buiten de traditionele medische wetenschap zijn onderzocht om zaken die voor mensen met een chronische ziekte of beperking van belang zijn te vinden. We hebben gebruik gemaakt van een inhoudsanalyse van relevante documenten, er zijn interviews gehouden en geanalyseerd en participerende observaties verricht.

Hoofdstuk 4 beschrijft een onderzoek van het systematische literatuurstudie type, naar door de wetenschap benoemde onderwerpen en waarden van mensen met een chronische ziekte of beperking. Internationale wetenschappelijke literatuur werd systematisch doorzocht en de resultaten werden verwerkt via de methode van een inhoudsanalyse met data synthese. Naast de primaire (wetenschappelijke bronnen) literatuur is ook in de secundaire - tertiaire bronnen (niet-wetenschappelijke publicaties, rapporten en informatie databases van organisaties van mensen met een chronische ziekte of beperking) informatie verzameld voor dit doel.

In hoofdstuk 5 werden, in een kwalitatief type onderzoek, focusgroepen gebruikt om te achterhalen welke onderwerpen belangrijk zijn voor mensen met een chronische ziekte of beperking. Deze onderwerpen zijn geanalyseerd en vervolgens gecombineerd in een set van criteria. Deze gecombineerde set van criteria werd besproken, bijgesteld en geproriteerd door de mensen zelf in een reeks van focus groepen, ronde tafel gesprekken en interviews. Dit werd herhaaldelijk gedaan met steeds andere deelnemers.

In hoofdstuk 6 zijn drie deel onderzoek beschreven over het participeren van mensen met een chronische longziekte, alle van het kwalitatieve type. In hoofdstuk 6.1 wordt een longitudinale case studie methode gebruikt om te onderzoeken hoe en in welke mate long patiënten participeren in door het Longfonds Nederland (LFN) gefinancierd longonderzoek
gedurende de afgelopen 7 jaar. In hoofdstuk 6.2 is de praktijk van de agenda setting van onderzoek door het Long fonds samen met mensen met een longziekte onderzocht. Er is daarbij gebruik gemaakt van een interactieve onderzoeksmethode met vragenlijsten, interviews en focusgroepen. In hoofdstuk 6.3 zijn via een interactieve methode de gedeelde verantwoordelijkheden en betrokkenheid in de praktijk van mensen met de chronische longziekte Chronic Obstructive Pulmonary Disease (COPD) en hun zorgverleners onderzocht.

EEN TEUGENDRAADS VERHAAL

De zoektocht van Truus, de auteur, en de vragen die tot dit onderzoek en proefschrift hebben geleid zijn te lezen in hoofdstuk 2, over de worsteling tussen kracht en kwetsbaarheid, het eigenzinnige verhaal van een patiënt. Zij beschrijft haar 27 jaren lange strijd vanaf het krijgen van tot het leven met meerdere chronische ziekten en beperkingen. Het is een eigenzinnig verhaal geworden aan de hand van een aantal kritische momenten in haar leven. Deze worsteling met kracht en kwetsbaarheid belicht een andere kant dan die van de mondige, onafhankelijke patiënt waarover in de gezondheidzorg steeds vaker wordt gesproken. Het heersende beeld gaat over de sterke mondige burger die eigen keuzes maakt en de eigen regie stevig in handen heeft en houdt in zijn leven.

Dit verhaal laat zien dat het niet zo simpel ligt, de patiënt beleefde alles tussen kracht en kwetsbaarheid en tussen beperkingen en mogelijkheden. Terugkerende periodes van verlies en rouw en van ontwikkeling en groei wisselen elkaar af. Mensen met een chronische ziekte of beperking kunnen volledig in beslag genomen worden door fysiek overleven en door primaire levensbehoeften. Dit heeft grote gevolgen voor de zeggenschap in processen en het meedoen in de samenleving, de maatschappelijke participatie. Of het nu gaat om problemen met werken, volgen van een opleiding, je hobby moeten stoppen, een afspraak met vrienden al weer moeten afbellen of om het je al of niet kunnen inzetten voor jouw lotgenotengroep.

Op een bepaald moment kan er in het leven van iemand met een ziekte of beperking een ommekeer te herkennen zijn: groei en hernieuwd maken het mogelijk zich te richten op de omgeving. Erkennen van kracht en kwetsbaarheid en dat deze beide kanten onderdeel zijn van het ‘zijn van iemand met een chronische ziekte of beperking’ is van groot belang. Dit doet recht aan hun capaciteiten en de soms beperkte mogelijkheden.

Door verschillende theorieën met dit verhaal te verbinden wordt duidelijk dat niet alle mensen met een chronische ziekte of beperking op ieder moment in hun leven mondig en zelfredzaam kunnen zijn. Het beeld dat mensen met een chronische ziekte of beperking als ‘zorgconsument’ onafhankelijk van anderen de regie over hun eigen leven kunnen houden, klopt soms, maar vaker niet.

Het leven met een chronische ziekte of beperking kan samen gaan met gevoelens van verdriet en verlies maar ook met onvervulde basisbehoeften zoals een gevoel van
veiligheid. Na de acceptatie van de chronische ziekte of beperking kunnen deze gevoelens van verdriet en gemis door een gebeurtenis of een verergering van de chronische ziekte of beperking weer opnieuw in alle hevigheid terug komen. Deze fases van rouw en onvervulde behoeften kunnen in een mens leven verschillende keren opnieuw opvlammen (recursief).

In het leven met een chronische ziekte of beperking is het niet van belang of iemand een ‘mondige zorg consument’ is of een ‘afhankelijke onzekere patiënt’. Maar het feit dat hij of zij met regelmaat en, en, de persoon zelf en diens omgeving met beide aspecten tegelijk moet leren omgaan. Een omgeving die de participatie van mensen met een chronische ziekte of beperking wil verwelkomen houdt rekening met kracht en kwetsbaarheid.

VERKENNING VAN CRITERIA VAN MENSEN MET EEN CHRONISCHE ZIEKTE OF BEPERKING IN NEDERLAND

Hoe vertegenwoordigers van mensen met een chronische ziekte of beperking participeren in onderzoeksprojecten en in kwaliteit van zorg in Nederland en welke problemen zij daarbij ervaren is onderzocht in hoofdstuk 3. Het gaat over patiënten in Nederland en hun vertegenwoordigers aan de onderhandelingstafel en met welke criteria die onderzoek en kwaliteit van zorg beoordelen. Deze studie is een eerste verkenning voor het uiteindelijke onderzoek in het proefschrift.

Het is een verkenning van de beoordelingscriteria die tot voor kort door vertegenwoordigers van mensen met een chronische ziekte of beperking werden gebruikt bij het beoordelen van wetenschappelijk onderzoek en kwaliteit van zorg. Met de resultaten uit dit onderzoek werd het mogelijk om erachter te komen waar de problemen lagen en konden we de onderzoeksvragen verduidelijken.

In de gebieden beleid, onderzoek en kwaliteit van zorg kwamen uit deze eerste verkenning aanvankelijk drie criteria naar voren. Deze drie criteria waren voor vertegenwoordigers van mensen met een chronische ziekte of beperking van belang. Deze criteria zijn: relevantie, zeggenschap en ethiek & veiligheid. De vraag naar criteria en praktisch bruikbare instrumenten voor vertegenwoordigers van mensen met een chronische ziekte of beperking die namens hun groep participeren was duidelijk hoorbaar. Zij hadden behoefte aan meer houvast tijdens het participeren in de vorm van een werkdocument of hulpmiddel om het participeren beter te doen te maken in termen van tijd en belastbaarheid. Ook was er behoefte aan een meer structureerde manier van beoordelen aan de hand van een beperkt aantal relevante onderwerpen van hun achterban. Met deze relevante onderwerpen en vragen in de hand konden zij sneller en eenvoudiger een document beoordelen of argumenten vinden voor in het debat. Evenals de vraag naar meer training en ondersteuning bij het beoordelen van onderzoek en kwaliteit van zorg.
INVENTARISATIE VAN BELANGRIJKE ONDERWERPEN UIT DE LITERATUUR

Hoofdstuk 4 gaat over “patiënt onderwerpen in gezondheidsonderzoek en kwaliteit van zorg, een inventarisatie en gegevens analyse”. Het beschrijft een internationale literatuurstudie naar onderwerpen die mensen met een chronische ziekte of beperking relevant vinden in gezondheidsonderzoek en kwaliteit van de zorg projecten. Het resultaat geeft een overzicht van wat er in de wetenschappelijke literatuur bekend is over problemen en onderwerpen die bij deze mensen leven. Al snel bleek dat over de onderwerpen van mensen met een chronische ziekte of beperking niet veel te vinden is in primaire bronnen uit de wetenschappelijke literatuur. Publicaties en rapporten van overheden, internationale organisaties en van groepen van mensen met een chronische ziekte of beperking waren nodig om een completer beeld te krijgen. Deze publicaties en rapporten worden ook wel secundaire literatuur of grijze literatuur genoemd.

Tabel 1 Onderwerpen die mensen met een chronische ziekte of beperking belangrijk vinden

| Nr | Onderwerp Beschrijving | Alleen gevonden in de grijze literatuur | | Nr | Onderwerp Beschrijving | Alleen gevonden in de grijze literatuur |
|---|------------------------|----------------------------------------|---|------------------------|----------------------------------------|
| 1  | Participatie           |                                        | 21 | Toegankelijkheid       | x                                      |
| 2  | Informatie             |                                        | 22 | Klachtbehandeling      | x                                      |
| 3  | Veiligheid             |                                        | 23 | Instemming             | x                                      |
| 4  | Zorg systeem          |                                        | 24 | Sociale zekerheid      |                                        |
| 5  | Kosten                |                                        | 25 | Gebouwen               |                                        |
| 6  | Leefstijl              |                                        | 26 | Communicatie           | x                                      |
| 7  | Keuze                 |                                        | 27 | Waarden                |                                        |
| 8  | Beperking              |                                        | 28 | Opleidingsniveau       | x                                      |
| 9  | Effectiviteit          |                                        | 29 | Geslacht               | x                                      |
| 10 | Methode                |                                        | 30 | Pijn                   | x                                      |
| 11 | Kwaliteit van leven    |                                        | 31 | Zelf zorg              | x                                      |
| 12 | Diversiteit            |                                        | 32 | Leeftijd               | x                                      |
| 13 | Medisch dossier        |                                        | 33 | Compensatie            | x                                      |
| 14 | Kwaliteit              |                                        | 34 | Angst                  | x                                      |
| 15 | Familie en vrienden   |                                        | 35 | Alternatieve           | x                                      |
| 16 | Privacy                |                                        |    |                        |                                        |
| 17 | Relevantie             |                                        |    |                        |                                        |
| 18 | Buitenlandse zorg      |                                        |    |                        |                                        |
| 19 | Waardigheid            |                                        |    |                        |                                        |
| 20 | Ethiek                 |                                        |    |                        |                                        |
Er werden totaal 35 relevante onderwerpen van mensen met een chronische ziekte of beperking gevonden. Hiervan werden er 13 uitsluitend in de secundaire of ‘grijze’ literatuur gevonden. Deze 13 ‘ontbrekende’ onderwerpen waren: privacy, relevantie, toegankelijkheid, geslacht, pijn, leeftijd, angst, instemming, communicatie, opleidingsniveau, klachten, schadevergoeding, en alternatieve methoden. Deze onderwerpen vormen een lacune in de wetenschappelijke literatuur en houden met name verband met fundamentele en persoonlijke waarden en met kwaliteit van leven onderwerpen. (Tabel 1)

GESTRUCTUREERDE BEOORDELING EN EVALUATIE IN PRAKTIJK

Hoofdstuk 5 gaat over gestructureerd beoordelen door patiënt vertegenwoordigers in gezondheidsonderzoek en kwaliteit van zorg. De onderwerpen, waarden en criteria die relevant zijn voor mensen met een chronische ziekte of beperking in de praktijk zijn in dit hoofdstuk nader onderzocht. In een reeks van focusgroepen, ronde tafel gesprekken en interviews, is onderzocht hoe de nieuw samengestelde set van criteria wordt beoordeeld, bijgesteld en geoprioriteerd door mensen met een chronische ziekte of beperking.

Allereerst is er onderzocht welke waarden achter de onderwerpen van mensen met een chronische ziekte of beperking van groot belang zijn voor en in hun leven. Hier kwam uit dat de onderwerpen verdeeld kunnen worden over 4 waarden groepen; fundamenteel, kwaliteit van leven, kwaliteit van zorg en zeggenschap/participatie.

Een waarde noemen we in dit proefschrift 'fundamenteel' als deze van het grootste belang voor mensen is. Daarnaast is een dergelijke fundamentele waarde verbonden met basisvoorwaarden om sociaal, maatschappelijk en, psychologisch te kunnen functioneren. Volgens de deelnemers in deze studie zijn de belangrijkste waarden de fundamentele menselijke waarden. Pas nadat aan de fundamentele waarden is voldaan, is een gesprek over aspecten van kwaliteit van leven mogelijk en komt participatie in onderzoek en kwaliteit van zorg langzamerhand in beeld. En pas als deze twee waarden zijn veilig gesteld kan er gepraat en meegedaan worden over onderwerpen die met kwaliteit van zorg of vormen van zeggenschap te maken hebben. Bijvoorbeeld, voor de deelnemers, is het zich veilig en gerespecteerd voelen belangrijker dan een goede communicatie met een professional of mee kunnen praten over een behandeling. We kunnen deze volgorde in belangrijkheid verbeelden in een "participatie ijsberg". (Figuur 1).

Hierbij steekt alleen topje van de ijsberg boven water uit. Dit zijn de mensen die participeren, die mee praten, die hun stem laten horen en die hun fundamentele waarden en kwaliteit van leven waarden al hebben gerealiseerd of veilig gesteld. Het grootste deel van de ijsberg, die grotendeels onzichtbaar is, ligt onder water. In dit onder water gedeelte zijn de fundamentele waarden en kwaliteit van leven waarden die onzichtbaar blijven. De samenleving ziet en hoort dus in deze vergelijking alleen de mensen die hun stem laten horen en die hun leven al aardig op orde hebben. De vraag blijft hoe we als samenleving de mensen horen die hun fundamentele waarden en kwaliteit van leven
waarden nog niet hebben gerealiseerd of veilig hebben gesteld, zij komen immers niet boven de ‘waterspiegel’ uit. Zij zijn vooralsnog vaak niet zichtbaar en hoorbaar.

Figuur 1 De participatie ijsberg met onderwerpen ingedeeld naar waarden

Om de onderwerpen en waarden bruikbaar en toepasbaar te maken voor de participatie praktijk is uit de 35 onderwerpen en waarden een set van criteria voor de participatie praktijk samengesteld. Dit mondde uit in een ‘criteriaawaaier’. Vertegenwoordigers van mensen met een chronische ziekte of beperking geven aan dat zij deze set van criteria vanuit hun perspectief zien en gebruiken als een instrument of hulpmiddel bij het participeren. De criteriaawaaier wordt ervaren als een praktisch hulpmiddel om op een gestructureerde manier onderzoeksvoorstellen of richtlijnen te beoordelen of te evalueren.
of te gebruiken tijdens een debat. De set criteria gaan over wat relevant is voor mensen met een chronische ziekte of beperking, over kwaliteit van leven, kwaliteit van zorg, ethische-veiligheid aspecten, informatie-communicatie en tenslotte over mede zeggenschap.

De criteriawaaier wordt in het veld gebruikt door vertegenwoordigers mensen met een chronische ziekte of beperking maar ook door professionals bij diverse gezondheidsfondsen en zelfs in twee Europese longonderzoek projecten. (Figuur 2)

Figuur 2 Voorbeelden van de criteria waaier zoals die nu in gebruik zijn

IMPLEMENTATIE VAN ONDERWERPEN EN WAARDEN VAN MENSEN MET CHRONISCHE LONGZIEKTE

Hoofdstuk 6.1 over participatie van mensen met een chronische longziekte, laat het pad naar de situatie dat mensen met een chronische longziekte mede beoordelaar zijn van wetenschappelijk onderzoek bij Long Fonds Nederland (LFN) onderzoek zijn, zien in een 7 jaar durende longitudinale case studie. We beschrijven en analyseren de invoering en uitvoering van de participatie praktijk van longpatiënten die hun perspectief inbrengen in de onderzoek cyclus van het fonds. Hierbij is gekeken op welke basis onderzoeksthema’s worden bepaald en welke subsidie beoordelingscriteria leidend zijn binnen LFN. Vervolgens is gekeken naar de zeggenschap van longpatiënten in LFN onderzoek.
De wetenschappelijke en maatschappelijke relevantie van onderzoeksvoorstellen wordt momenteel beoordeeld door alle partijen. Dit is onder andere het gevolg van de implementatie van een participatie methode met inbegrip van het gebruik van de set criteria vanuit het perspectief van mensen met een longziekte. Een groep mensen met een longziekte wordt getraind en voorzien van een beoordelingsinstrument om de maatschappelijke relevantie te beoordelen vanuit patiënten perspectief. Wetenschappelijke relevantie en kwaliteit worden afzonderlijk beoordeeld en afgewogen voor het proces van goed keuring of afwijzing van onderzoek. Deze zeven jaar durende studie biedt een inzicht in de betrokkenheid van mensen met een longziekte in de onderzoek cyclus en het wijst op valkuilen en complicaties die zich kunnen voordoen.

Twee gerelateerde onderzoeken gaan nader in op enkele onderdelen die genoemd zijn in hoofdstuk 6.1. Hoofdstuk 6.2 belicht de manier waarop vanaf 2004 mensen met een chronische longziekte in toenemende mate mede zijn gaan bepalen wat op de agenda voor Longfonds longonderzoek terecht komt. Hoewel onderzoeks prioriteiten van hen en de professioneel deskundigen grotendeels vergelijkbaar zijn, verschillen ze in details. Mensen met een longziekte geven andere prioriteiten aan bepaalde onderwerpen voor onderzoek naar chronische longziekten die voorheen minder aandacht kregen in longonderzoek, zoals bijvoorbeeld het onderwerp vermoeidheid, co morbiditeit of bijwerkingen.

Hoofdstuk 6.3 beschrijft de gedeelde verantwoordelijkheden bij belangrijke onderwerpen voor mensen met Chronic Obstructive Pulmonary Disease (COPD) en de professionals in de zorgpraktijk.

Enerzijds is de dialoog tussen longpatiënten als lotgenoten daarin heel belangrijk. De veiligheid van een groep en het delen van ervaringen blijkt een belangrijk onderdeel om mensen met COPD te helpen betekenis te geven aan hun aandoening. Anderzijds wordt tegenwoordig verwacht dat mensen met een chronische ziekte of beperking een belangrijke rol spelen in hun herstel. Dit heeft gevolgen voor de taakverdeling tussen artsen en de mensen zelf. Diepe-interviews met mensen met COPD, fysiotherapeuten en een longarts laten zien dat het verschuiven verantwoordelijkheden vereisen een ondersteunende houding van zorgverleners en een dialogische communicatie tussen patiënten en professionals. Mensen met de chronische longziekte COPD worden mede-eigenaar in een proces van herstel. Het besef dat COPD is meer dan alleen een gebrek aan lucht, en dat lichaam en geest op elkaar inwerken, is een eerste stap om andere problemen in het leven te onderzoeken en om iemands kwaliteit van leven te verbeteren. Het uitwisselen van ideeën met respect voor elkaars invalshoeken is hierbij belangrijk.

ALGEMENE DISCUSSIE

Hoofdstuk 7 beantwoordt de onderzoeksvragen en geeft een aantal aanbevelingen voor vervolgonderzoek en de participatie praktijk.
De drie belangrijkste bevindingen van dit proefschrift zijn hier opgenomen.

Allereerst zijn waarden en onderwerpen van mensen met een chronische ziekte of beperking geformuleerd. Deze zijn onderdeel van hun ervaringskennis. In ons onderzoek zijn 35 waarden gevonden die voor mensen met een chronische ziekte of beperking van groot belang zijn voor en in hun leven om sociaal –maatschappelijk en geestelijk te kunnen functioneren en hun mogelijkheden voor deelname aan participatie activiteiten. Met behulp van de waarden zijn de onderwerpen ingedeeld in vier volgordeelijk te rangschikken groepen: fundamenteel menselijk waarden, kwaliteit van leven, kwaliteit van zorg en participatie.

De tweede belangrijke bevinding is de set van beoordeling en evaluatie criteria vanuit en voor vertegenwoordigers van mensen met een ziekte of beperking om onderzoek en kwaliteit van zorg te beoordelen. Deze set van zes beoordeling- en evaluatie criteria die vanuit de 35 in de praktijk en in de literatuur gevonden ‘issues’ is samengesteld, bevat de onderwerpen die relevant zijn voor mensen met een ziekte of beperking. De set van criteria kan ingezet worden tijdens het participeren in onderzoeksprojecten of kwaliteit van zorg projecten zoals richtlijnen.

De set van criteria gaat over wat relevant is vanuit het perspectief van mensen met een chronische ziekte of beperking bij de kwaliteit van zorg of in gezondheidsonderzoek. Het gaat ook over de kwaliteit van leven van mensen zal verbeteren zoals de diagnostiek en behandeling.


Ten slotte gaan de criteria over (mede) zeggenschap en de vraag of een project of onderzoek mede gebaseerd is op de ideeën en behoeften van mensen met een chronische ziekte of beperking.

Het criterium over ‘participatie’ omvat ook het onderwerp ‘zeggenschap’ waardoor het meer een proces-criterium dan een inhoud-criterium is. Ook gaat dit criterium over de vraag of zij gelijkwaardige gesprekspartner zijn in het project en niet alleen kunnen mee praten maar ook mee beslissen.
De derde en laatste belangrijke bevinding gaat over het belang van de relatie tussen de participerende mensen met een chronische ziekte of beperking en professionals tijdens participatie activiteiten. Hierdoor kan een vorm van partnerschap ontstaan in plaats van dat partijen tegenover elkaar staan. Hiervoor is commitment en draagvlak nodig bij alle deelnemers over het nut en de meerwaarde van het meenemen en mee wegen van perspectief van mensen met een ziekte of beperking. Door de verschillende perspectieven van zowel zorgverleners als onderzoekers als mensen met ziekten of beperking met elkaar te verbinden kan er een kwalitatief beter eind resultaat komen dat aansluit op de praktijk. Zo ontstaat er een onderzoek of een richtlijn die zowel wetenschappelijk relevant en voldoende kwaliteit heeft als maatschappelijk relevant vanuit het perspectief van mensen met een ziekte of beperking.

AANBEVELINGEN VOOR VERDER ONDERZOEK

Veranderende verhoudingen: Eerste aanbeveling voor verder onderzoek gaat over de vraag of de aanwezigheid van vertegenwoordigers en de stem van mensen met een ziekte of beperking de bestaande macht-verhoudingen en processen verstoren, en zo ja, in welke mate. Wat gebeurt er als vertegenwoordigers van mensen met een ziekte of beperking zich niet aanpassen aan het dominante discours en een eigenzinnig verhaal inbrengen zoals dat van de eerste auteur? Wat gebeurt er als hun opvatting fundamenteel afwijkt van andere deelnemers? Plaatsen ze zich dan buiten de groep en buiten het debat of kan dit tegengeluid invloed hebben? Of wordt het machtsevenwicht tussen professionals en mensen met een chronische ziekte of beperking langzaam verschoven en zijn ze daarbij steeds echt meer gelijkwaardige partners in de discussie, en, met andere woorden, onderweg naar een echt partnerschap? Het zou interessant zijn deze vragen nader te onderzoeken en tevens na te gaan hoe de stem van mensen met een ziekte of beperking versterkt kan worden binnen de bestaande machtsprocessen en -verhoudingen.

Kwetsbare mensen: Ook wordt verder onderzoek aanbevolen naar de vraag hoe de ervaringskennis van de meest kwetsbare mensen die hun stem niet laten horen, toch gehoord, gebruikt en in het belang van hen ingezet kan worden bij de beoordeling van onderzoek en zorg. Hoe kan hun ervaringskennis worden ingebracht en gewogen in de gezondheidszorg debat of besluitvormingsproces?

Verhalen als instrument voor meer inzicht: Tot slot is verder onderzoek aan te bevelen naar alternatieve manieren voor mensen met een ziekte of beperking om meer grip en inzicht te krijgen in hun situatie en het leven met beperkingen. In hoofdstuk 2 schreef Truus, de auteur, haar verhaal op aan de hand van enkele kritieke momenten in haar leven rondom ziekte en beperking. Ze ontwikkelde haar verhaal en reflecteerde hierop met twee andere mensen. Dit gaf haar meer inzicht, begrijpen, grip en perspectief in haar leven. Onderzoekt zou kunnen worden of deze aanpak of andere creatieve uitingsvormen zoals poëzie of andere artistieke impressies ook voor andere mensen met een chronische ziekte of beperking positieve betekenis heeft.
**AANBEVELINGEN VOOR DE PRAKTIJK**

**Monitoring:** Luisteren naar mensen met een ziekte of beperking moet een permanente gewoonte in de gezondheidszorg zijn op alle niveaus. Dit communiceert de Wereld Gezondheid Organisatie in haar gezondheidsbeleid over de rechten van burgers en de betrokkenheid van de patiënt ‘Gezondheid 21’ voor de Europese regio. In verschillende algemeen aanvaarde documenten worden ethiek en mensenrechten genoemd maar deze zijn niet juridisch bindend. Daarom is de toepassing niet gegarandeerd. Aanbevolen wordt dat organisaties van mensen met een chronische ziekte of beperking dit goed in de gaten houden en monitoren.

**Waarden structuur:** Waarden zijn een belangrijke motor achter menselijk gedrag. Onderwerpen die belangrijk zijn voor mensen met een chronische ziekte of beperking zijn nauw verbonden met waarden. We bevelen aan om de in dit onderzoek gevonden criteria en de criteriawaaier te implementeren in de praktijk.

**Eigenzinnige verhalen:** Het dominante idee van robuuste, uitgesproken en mondige mensen met een chronische ziekte of beperking staat ter discussie. Mensen zijn niet alleen zorgconsumenten met regie over hun eigen chronische ziekte of beperking. Andere geluiden, beschreven in alternatieve en eigenzinnige verhalen moeten in het gezondheidsdiscours en beleid worden mee genomen en meegewogen.

**Pictogrammen en symbolen:** Mensen met een verstandelijke beperking of een psychische aandoening, zoals de ziekte van Alzheimer, maar ook laaggeletterden vereisen een speciale versie van de criteria die in dit onderzoek zijn ontwikkeld. Woordgebruik, afbeeldingen en pictogrammen dienen op hun behoeften te worden afgestemd. Voor sommige mensen blijkt het herkennen van een pictogram of afbeelding makkelijker dan het lezen van een tekst. Het blijkt ook dat pictogrammen nauwkeuriger geïnterpreteerd worden dan woorden. De criteriawaaier is een levend document. Uitbreiding van de criteria met andere specifieke onderwerpen en problemen die verband houden met het dagelijks leven van bepaalde groepen mensen zijn wenselijk en mogelijk.

**Partnerschap:** Nog steeds worden mensen met een chronische ziekte of beperking regelmatig gezien als een bedreiging voor gevestigde machtsverhoudingen in een project of een commissie. Deze relatief nieuwe stem aan de onderhandelingstafel wordt dan niet gezien als een positieve ontwikkeling, maar als een formaliteit waaraan voldaan moet worden maar die in feite niet serieus wordt genomen. Een van de ongewenste vormen van betrokkenheid is om mensen te betrekken voor de vorm, de zogenaamde pseudo-participatie of schijn participatie. Aangeraden wordt dat mensen met een chronische ziekte of beperking alleen onder vooraf geaccepteerde randvoorwaarden van gelijkwaardige partnerschap in de discussie of besluitvorming deelnemen in een project, commissie of in een debat.
**Capaciteit opbouw:** Mensen met een ziekte of beperking die participeren, ondervinden vaak moeilijkheden om de continuïteit van hun activiteiten als gevolg van ziekte uitval te waarborgen. Een oplossing hiervoor zou zijn om in duo verband te participeren. Hiervoor is het nodig dat er voldoende mensen met een ziekte of beperking beschikbaar zijn, die regelmatig kunnen en willen participeren. Deze mensen zouden dan ook getraind moeten worden in het participeren en geschoold en geïnformeerd dienen te worden over relevante ontwikkelingen en feiten over hun achterban. Een grotere groep van vertegenwoordigers van mensen met een chronische ziekte of beperking vermindert de lacunes in de dialoog en tijdens beslissingsprocedures. Aanbevolen wordt om extra aandacht te besteden aan het versterken en ondersteunen van mensen met een chronische ziekte of beperking die namens een groep als vertegenwoordiger mee praten en mee beslissen. Alle betrokken partijen – zowel zwakke als sterke – moeten in de gelegenheid zijn om hun problemen en zorgen ter tafel te brengen, te worden gehoord en serieus genomen te worden.
Samenvatting

1. **RELEVANTIE**
   1.1 Verbetert dit de gezondheid van mensen met een chronische ziekte of beperking?
   1.2 Houdt het rekening met hun diversiteit?
   1.3 Is het gebaseerd op hun wensen en behoeften?
   1.4 Zijn de resultaten in de praktijk bruikbaar?

2. **Kwaliteit van Leven**
   2.1 Verbetert dit de gezondheid gerelateerde kwaliteit van leven voor mensen met een chronische ziekte of beperking?
   2.2 Verbetert dit hun maatschappelijke deelname?
   2.3 Respecteert het de fundamentele weder en zijn die als leidend principie gebruikt?
   2.4 Is het projectresultaat in praktijk gunstig voor hen?

3. **Kwaliteit van Zorg**
   3.1 Verbetert dit de gezondheidszorg voor mensen met een chronische ziekte of beperking?
   3.2 Houdt het rekening met hun diversiteit?
   3.3 Houdt het rekening met hun sociale network en omgeving?

4. **Ethiek en Veiligheid**
   4.1 Rechtvaardigt het doel (bijv. betere medicijnen) of middelen (bijv. pijn, uitval op werk, school)?
   4.2 Is de veiligheid van deelnemers, patiënten en proefpersonen gewaarborgd?
   4.3 Hebben de betrokkenen expliciete keuze vrijheid?
   4.4 Worden hun sociale normen, waarden en waardigheid gerespecteerd?
   4.5 Volgen de gezondheids professionals de regels, de beroepscodes en de praktijkrichtlijnen?

5. **Informatie en Communicatie**
   5.1 Is de informative begrijpelijk voor de mensen met een chronische ziekte of beperking?
   5.2 Worden deze mensen en hun organisaties over het project resultaat geïnformeerd?
   5.3 Is de verstrekte informatie correct en compleet, ook over mogelijke complicaties?
   5.4 Hebben deze mensen de mogelijkheid expliciet mee te doen of te weigeren (geïnformeerd instemmen)?

6. **Participatie (Randvoorwaarde)**
   6.1 Is rekening gehouden met de belangen van mensen met chronische ziekte of beperking?
   6.2 Worden zij als gelijkwaardige gesprekspartners betrokken?
   6.3 Worden zij als volwaardig burger gezien?
   6.4 Wordt hun ervaring in project, richtlijn of onderzoek als bron van kennis meegenomen?
   6.5 Krigen zij ondersteuning bij hun participatie inspanningen?
Truus Teunissen was born November 3, 1955 in Heerlen, the Netherlands. She grew up in the coal mining district in the south of the Netherlands and became home care coordinator at Groot Rijnland in Alphen a/d Rijn in 1979. She graduated as a social scientist in 1995 at the Utrecht University. She was employed at B&A Group in The Hague as a social researcher, and at the Dutch Asthma Foundation (now Lung Foundation Netherlands) as programme manager on patient involvement. Over the years she was secretary of the Dutch Asthma Foundation patient advisory council, member of the National Academic Hospitals clients committee CRAZ, member in the AGIS health insurance clients council, board member of the National chronic disease and disabilities council IEDERIN (former CG-Raad), board member of the revalidation institute IRV and patient member of the EU-IMI-U-Biopred Ethics committee. Since 2009 she is a member of the Program Patient Participation committee of ZonMw and the VSB Fund, of the National ZonMw scientific research funding review expert patients committee and of the VU/Athena Societal advice council VU-MPA. She has been working on patient involvement since 1996. Having a chronic lung disease since 1985, keeping up her work and family life was increasingly difficult. This chronic disease made her unable to work on a regular basis. In 2005 she was officially incapacitated. In 2007 she also got eye cancer with all the treatments and side effects. She then set out to explore how her own personal experience with patient involvement could benefit all people with a chronic illness or disability, involved in health research and quality of health care. The Lung Foundation adhered to her expert advice role and she continued coaching and coordinating the Lung Foundation patient expert group. In order to carry out scientific research in her own time and place in this field, she started in 2008 as a guest PhD researcher at the VU Free University Metamedica EMGO+ in Amsterdam. There are two results. The first is a newly structured involvement appraisal instrument, primarily intended for use by people with an illness or disability, but it can also be used by professionals. The second result is this thesis which provides valuable insight into issues that matter to people with a chronic illness or disability and constitutes a valid basis for the instrument.
PUBLICATIONS

Publications for this thesis:


1) “Teunissen GJ”, “Teunissen T” and “Truus Teunissen” author names were used in different instances.
Other publications:


People with a chronic illness or disability are the new partners in the health research and health care dialogue. This new and growing group expresses their own voice, issues and concerns. Moving in between ill and healthy conditions, they are both strong and vulnerable at the same time.

This thesis investigates what is required to articulate and voice their needs and concerns in health related discussions and decision making in practice. This work resulted a.o. in a set of criteria for appraisal and evaluation by their advocates.

Truus Teunissen is a social scientist having more than one chronic illness. She combined her scientific knowledge and background with her experiential knowledge in order to write this thesis.