THE STRUGGLE BEHIND “I’M ALL RIGHT”

Response shifts and self-presentation
in small-cell lung cancer patients

The study presented in this thesis was performed at the EMGO Institute and the Department of Public and Occupational Health, of the VU University Medical Center (Vumc), Amsterdam, the Netherlands. The EMGO Institute participates in the Netherlands School of Primary Care Research (CaRe), which was re-acknowledged in 2000 by the Royal Netherlands Academy of Arts and Sciences (KNAW).

The study was funded by the Dutch Cancer Society (VU 2000-2339). Financial support for printing of the thesis has been kindly provided by the EMGO Institute and the Dutch Cancer Society.

ISBN: 987 90 6464 161 9

Cover: Frea Lenger, “What to believe” I; collage with gold leaf

Coverdesign: James van der Leden and Marjan Westerman

Lay-out: Ponsen & Looijen B.V

Editorial support: Faith Maddever

Printed by: Ponsen & Looijen B.V., Wageningen, the Netherlands

VRIJE UNIVERSITEIT

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Response shifts and self-presentation in small-cell lung cancer patients

ACADEMISCH PROEFSCHRIFT

ter verkrijging van de graad Doctor aan
de Vrije Universiteit Amsterdam,
op gezag van de rector magnificus
prof.dr. L.M. Bouter,
in het openbaar te verdedigen
ten overstaan van de promotiecommissie
van de faculteit der Geneeskunde
op maandag 29 oktober 2007 om 13.45 uur
in de aula van de universiteit,
De Boelelaan 1105

door

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geboren te Hilversum

promotor: prof.dr. G. van der Wal

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dr.mr. B.A.M. The

Ik heb van het leven vrijwel niets verwacht
't Geluk is nu eenmaal niet te achterhalen
Wat geeft het? – In de koude voorjaarsnacht
Zingen de onsterfelijke nachtegalen

J.C. Bloem

nooit zonder jou, voor jou… Frans

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1

Introduction

"Until treatment is totally effective and side effects non-existent it is important that the quality of life of patients undergoing cancer treatment is properly evaluated" Peter Selby, 1985.[1]

Background

Quality of life (QoL) is considered to be as an important outcome for evaluating the impact of disease, and for assessing the effectiveness of treatment. QoL is particularly relevant for cancer patients who are willing to undergo risky and toxic treatment.[2-5] Indeed, when treatment has no apparent benefits with regard to survival, possible effects on QoL can become the deciding factor. However, even if there are no survival benefits, chemotherapy can provide important palliative benefits.This is especially so in patients with small-cell lung cancer (SCLC), for whom therapy is unlikely to be curative. Therefore, understanding the relative effects of chemotherapy on QoL is important for decision-making, and even more vital in daily clinical practice for optimizing the QoL of these patients throughout the course of their illness.[6]

However, QoL research often yields counter-intuitive and paradoxical findings which raise questions about what QOL measurement instruments actually assess, and how the scores should be interpreted. For example, Groen *et al*. studied patients with inoperable non-SCLC treated by radiation with and without chemotherapy.[7] QoL was measured with the European Organization for Research and Treatment of Cancer Core Quality of Life Questionnaire (EORTC QLQ-C30) [8], which has been designed specifically for use in clinical trials focusing on cancer patients. However, contrary to expectations, they found no significant deterioration in the scores over the 6-week treatment period. Furthermore, patients with a life-threatening disease reported a stable QoL, and the level of QoL of patients with a severe chronic illness was found to be no worse no better than that of less severely ill patients or healthy people.[9-13] Such counter-intuitive findings, labelled by Breetvelt and Van Dam as 'under-reporting of problems', suggest that patients report less distress and dissatisfaction than they actually experience.[12] Several studies show that patients' own evaluations may differ considerably from those made by clinicians and significant others.[14;15] In short, QoL measures do not consistently distinguish known groups, they are often only weakly related to objective criteria, and they show little convergence across measurement perspectives.[16]

It is suggested that patients make the best of their condition by coping, rethinking and reframing their experiences, and that this adaptive self-regulation may explain these discrepancies.[17-19] In recent years, the response shift theory has gained increasing acceptance in explaining paradoxical and counter-intuitive findings. Response shift refers to a change in internal standards, values and conceptualization of QoL, and it is recognized as an important mediator in adaptation to changing health status.[17] However, despite the explanatory power of the response shift theory, our current understanding of phenomena that can complicate the interpretation of QoL scores is still limited.[20] Although the occurrence of response shift might complicate 'objective' evaluation of QoL and treatment [17-22], it could also be seen as a desirable outcome of adaptation. From both perspectives it is therefore necessary to explore response shift and other phenomena that can complicate the interpretation of QoL scores.

Response shift

If a test or measurement is reliable, one usually assumes that the construct that is being assessed is *stable*, for example the assessment of temperature or blood pressure. If one interprets a change in QoL over time (or a difference between groups) as an *actual* change (or difference), one assumes that the concept of QoL itself remains the same over time (or between groups), in the same way as the concepts of temperature and blood pressure remain the same over time. Response shift complicates such interpretations, due to *recalibration*, *change in values* or *reconceptualization*.

The concept of response shift originated in the 1970's during research on educational training interventions (Howard et al. [23]) and organizational change (Golembiewski *et al.*[24]). Howard defined response shift in terms of changes in internal standards of measurement, whereas Golembiewski also introduced the component of reconceptualization in addition to this scale racalibration; changes in values are inherent in Golembiewski reconceptualization. Sprangers & Schwartz introduced reconceptualization as a separate component in their theoretical model of response shift (see Figure 1).[17] Response shift is defined as "a change in the meaning of one's self-evaluation of a target construct as a result of: a) a change in the respondent's internal standards of measurement (scale recalibration, in psychometric terms); b) a change in the respondent's values (i.e. the importance of component domains constituting the target construct); or c) a redefinition of the target construct (i.e. reconceptualization)". Sprangers & Schwartz emphasize that the extent to which the three components are distinct or interconnected is unknown.

The theoretical model has five major components: 1) *Catalysts* refer to a change in the respondent's health status that may or may not result from treatment; 2) *Antecedents* include personal characteristics, such as gender, education, personality, expectations; 3) *Mechanisms* encompass behavioural, cognitive, or affective processes to accommodate changes in catalysts (initiating social comparisons, reordering goals); 4) *Response shift* includes changes in the meaning of one's self-evaluation of QoL, resulting from changes in internal standards, values, or conceptualization; and 5) *Perceived QoL* can be defined as a multidimensional construct incorporating at least the three domains of physical, psychological and social functioning. The model uses a dynamic feedback loop to explain how QoL scores can be stabilized, despite changes in health status.

Measurement of response shift in QoL

Schwartz & Sprangers [22] describe and evaluate a number of different approaches to the measurement of response shift and the identification of recalibration, change in values and reconceptualization. Two categories of approaches appear to be immediately applicable in QoL research, because already existing QoL instruments can be used. One category consists of 'design approaches', of which the 'then-test' is an example. The then-test consists of an additional measurement with the same QoL instrument that has been used for QoL measurement (though with different instructions for the patient). The other category consists of 'individualized methods' which are characterized by the fact that from the outset they are not intended to measure a stable concept, but require that respondents define the concept for the moment of measurement only. The Schedule for the Evaluation of Individual Quality of Life – Direct Weighting (SEIQoL-DW)[25] is such an individualized approach which measures QoL from the unique individual perspective. Patients can choose, rate and weight five domains (cues) that they consider important. At different points in time, patients might nominate different cues, and they might give the same cues a different weight. As Schwartz & Sprangers have indicated, both changes are indicative of a change in values.

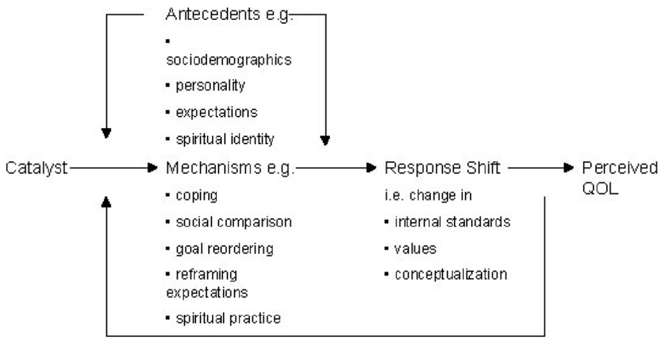

Figure 1. A theoretical model of response shift and quality of life (QOL).[17] "Integrating response shift into health-related quality of life research: a theoretical model" by M.A.Sprangers and C.E. Schwartz, 1999, *Social Science and Medicine*, 48(11), pp.1507-1515.

Why small-cell lung cancer patients?

Worldwide, lung cancer is one of the most frequently occurring forms of cancer, with a high mortality rate in both men and women.[26-28] Lung cancer is usually sub-divided in non-SCLC and SCLC. The first category accounts for about three-quarters of all lung cancers, and has a slower progression rate than the second category. Apart from being a relatively fast-growing form of lung cancer, SCLC is also early spreading, and metastases are almost always present at the time of the initial diagnosis. Standard therapy for SCLC is systematic treatment with chemotherapy (4-6 cycles). If the SCLC is limited, the patients will receive chemotherapy with concurrent radiotherapy. But even after a successful initial course of chemotherapy there is a very high rate of early recurrence, resulting in limited survival. For more than 90% of the patients, extensive SCLC is fatal within 2 years of diagnosis. Nevertheless, compared with supportive care, chemotherapy offers substantial benefits by improving both the quality and the quantity of life within this limited period. Patients with limited disease have a median survival period of 12-16 months, and those with extensive have 7-12 months.[28] The treatment of SCLC patients is not aimed at recovery, but at prolonging and enhancing the quality of the patient's life, and therefore the correct interpretation of QoL scores is of major importance. An important reason for selecting SCLC patients for our study was that the general illness trajectory was already known and described by The [29], and this provided us with essential background knowledge for the design of the study. The average illness trajectory of SCLC patients is relatively short, so complete trajectories could be covered in a four-year study.

Research questions

In this thesis we explore response shift in QoL in the palliative treatment of SCLC patients.

Listen to the patient

This first part of the thesis focuses on response shift and other explanations to account for counter-intuitive findings in the measurement of QoL by means of EORTC questionnaires.

1. Is the then-test a useful approach to determine whether a recalibration response shift has occurred?
2. Can response shift explain counter-intuitive findings sufficiently in the measurement of QoL?

Focus on what really matters

The second part of the thesis addresses the elicitation of QoL domains (cues), change in what really matters to patients, and the measurement of response shift (i.e. reconceptualization and change in values) in individual QoL.

3. Is SEIQoL-DW a reliable instrument in providing all the relevant information that is needed to determine wether a response shift (i.e. reconceptualization and change in values) has occurred?

Understand the struggle behind "I'm all right"

The third part of the thesis describes how patients manage to cope with their incurable disease.

4. What are the factors that result in positive self-reports, despite deteriorating health?

Methods

To answer the research questions a qualitative exploratory longitudinal study was designed, and various methods of data-collection were used. These methods were a mix of techniques and procedures which are derived from different research traditions. The way in which these various methods are integrated, and the way in which the resulting data were analysed, is based on the (longitudinal, multiple) *qualitative case-study*.[30] To fully understand changes in standards, values and concepts and, moreover in QoL, it was necessary to probe beyond quantitative responses to discover what mattered to the patient. In a qualitative, semi-structured interview we encouraged patients to tell us in their own words what they valued (most) in their life (at that specific point in time, in their current circumstances). We were not only interested in collecting qualitative data from the patients about (changes in) standards, values and concepts, but we also wanted to relate such data to actual measurements of QoL obtained with established measurement instruments. We used the EORTC QLQ-C30 and the QLQ-CL13 for this purpose because they are the current European standard for trials involving (lung) cancer patients. A ‘think aloud’ procedure was used (see Method sections of Chapter 2 and 3 for further details). Response shift (i.e. change in internal standards) in EORTC QLQ measurements was assessed by means of the ‘then-test’. We used the SEIQoL to measure individual QoL, because it not only gives patients the opportunity to describe their own perspective, but also because it is one of the few QoL measures that explicitly take reconceptualization and change in values into account. The main reason for using both the EORTC and the SEIQoL (instead of only one measure) was that we wanted to explore actual change in QoL and response shift in QoL from as broad a perspective as possible. By presenting a large variety of QoL stimuli to patients in different contexts (i.e. different QoL measures), we provided them with the opportunity to reflect on a broad range of aspects of QoL.

Outline of this thesis

In this thesis, we investigated adaptation in the illness trajectory of SCLC patients by means of observations made by the QoL researcher and also from the patient’s perspective. Chapters 2 to 6 of this thesis are based on articles which have already been published, accepted or submitted for publication. This implies that various chapters overlap, especially with regard to the Methods sections. Chapters 2 and 3 describe the measurement of QoL with the EORTC questionnaires, the use of the then-test in exploring changes in internal standards, and the investigation of other phenomena complicating the interpretations of QoL scores. Chapters 4 and 5 describe the measurement of individual QoL, and the usefulness of SEIQoL-DW in assessing adaptation to changing health, reconceptualization and change in values. Chapter 6 describes how patients manage to cope with their incurable disease. Finally, Chapter 7 presents an overview of what we have accomplished, implications of the findings are discussed, and we conclude with recommendations for further research.

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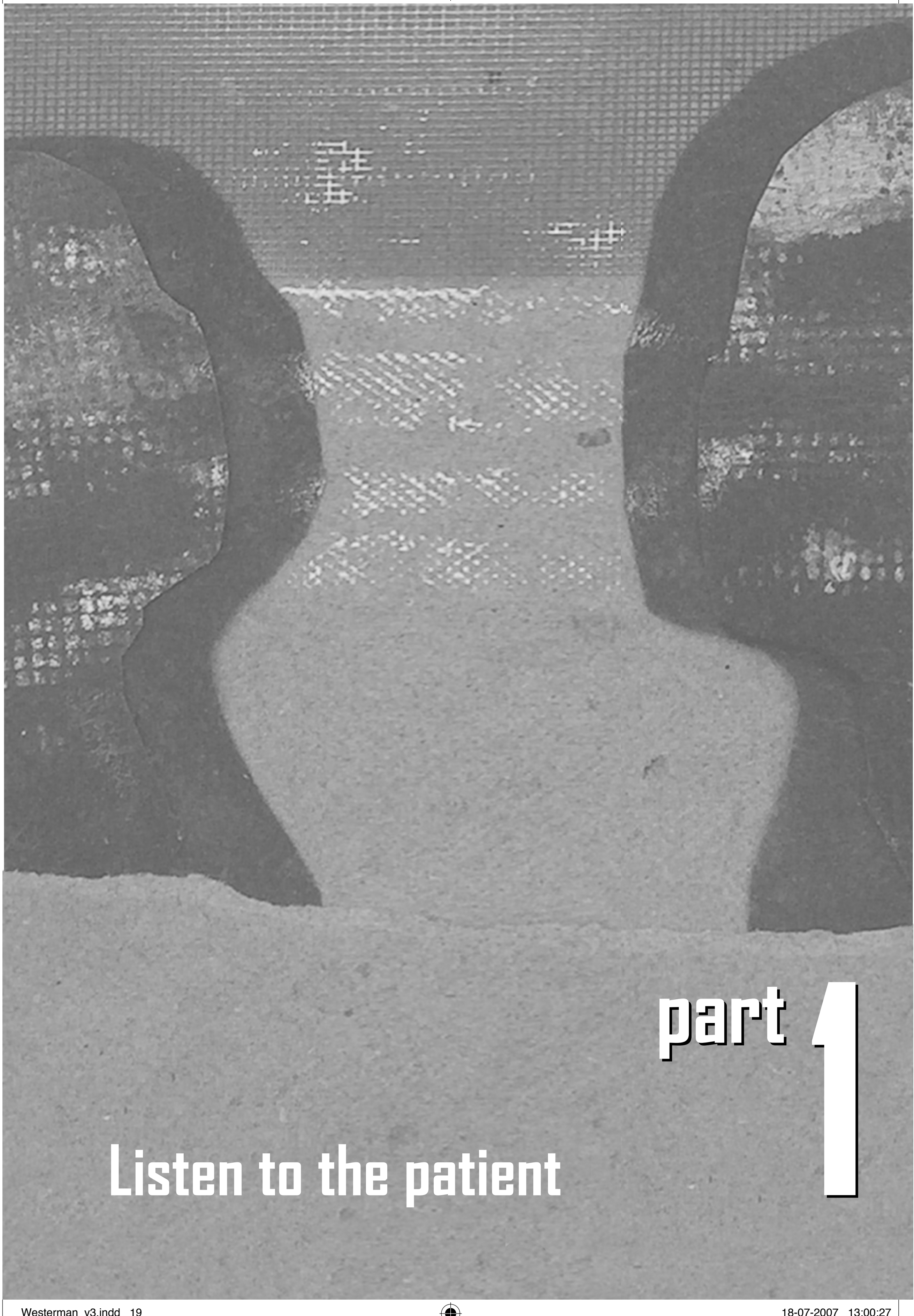

part 1

Listen to the patient

2

Small-cell lung cancer patients are just 'a little bit' tired: response shift and self-presentation in the measurement of fatigue

Marjan Westerman, Anne-Mei The, Mirjam Sprangers
Harry Groen, Gerrit van der Wal, Tony Hak

Quality of Life Research 2007; 16 (5):853 -861

Abstract

Background
Response shift has gained increasing attention in the measurement of health-related quality of life (QoL) as it may explain counter-intuitive findings as a result of adaptation to deteriorating health.

Objective
To search for response shift type explanations to account for counter-intuitive findings in QoL measurement.

Methods
Qualitative investigation of the response behaviour of small-cell lung cancer patients (*n*=23) in the measurement of fatigue with the EORTC QLQ-C30 question ‘were you tired’. Interviews were conducted at four points during 1st line chemotherapy: at the start of chemotherapy, four weeks later, at the end of chemotherapy, and six weeks later. Patients were asked to ‘think aloud’ when filling in the questionnaire.

Results
Fifteen patients showed discrepancies between their answer to the EORTC question ‘were you tired’ and their level of fatigue spontaneously reported during the interview. These patients chose the response options ‘not at all’ or ‘a little’ and explained their answers in various ways. In patients with and without discrepancies, we found indications of recalibration response shift (e.g. using a different comparison standard over time) and of change in perspective (e.g. change towards a more optimistic perspective). Patients in the discrepancy group reported spontaneously how they dealt with diagnosis and treatment, i.e. by adopting protective and assertive behaviour and by fighting the stigma. They distanced themselves from the image of the stereotypical cancer patient and presented themselves as not suffering and accepting fatigue as consequence of treatment.

Conclusion
In addition to response shift, this study suggests that ‘self-presentation’ might be an important mechanism affecting quality of life measurement, particularly during phases when a new equilibrium needs to be found.

Introduction

Quality of life (QoL) is considered an important treatment outcome when the treatment intent is not curative but palliative. However, the expected deterioration in QoL often does not occur, even in cases of serious illness. For example, Groen *et al.* studied patients with inoperable non-small cell lung cancer treated by radiation with and without chemotherapy.[1] QoL was measured with The European Organization for Research and Treatment of Cancer Core Quality of Life Questionnaire(EORTC QLQ-C30) [2], which has been designed specifically for use in clinical trials focusing on cancer patients. However, contrary to expectation, they did not find significant deterioration in the scale scores over the treatment period of 6 weeks. Although cancer patients are willing to undergo risky and toxic treatments[3], it seemed implausible that the side-effects of treatment had not affected their QoL. Other studies also reported counter-intuitive results. For example, patients with a life-threatening disease or disability were found to report stable QoL, and patients with a severe chronic illness reported QoL levels that were not inferior to that of patients with a less severe illness or to healthy patients.[4-6] Such counter-intuitive findings, labelled by Breetvelt and Van Dam as 'underreporting of problems' suggest that patients report less distress and dissatisfaction than they actually feel.[6]

In recent years, response shift theory has gained increasing attention in explaining paradoxical and counter-intuitive findings. Response shift refers to a change in internal standards, values and conceptualization of QoL and is recognized as an important mediator in adaptation to changing health.[7] However, despite the explanatory power of response shift theory, our current understanding of phenomena that can complicate the interpretation of QoL scores is still limited. Therefore, we investigated QoL measurement in small-cell lung cancer (SCLC) patients during 1st line chemotherapy. We were quite surprised when we noticed discrepancies between levels of fatigue measured with the questionnaire and answers spontaneously reported during the interview. We therefore investigated these 'conflicting' findings in the measurement of QoL in more depth.

This paper reports the results of an exploratory longitudinal multiple-case study, in which we focused on how patients responded to the EORTC QLQ-C30 question 'were you tired' at different points in their treatment trajectory. We aimed to describe the patients' explanations when answering the question, and to search for explanations of counter-intuitive findings.

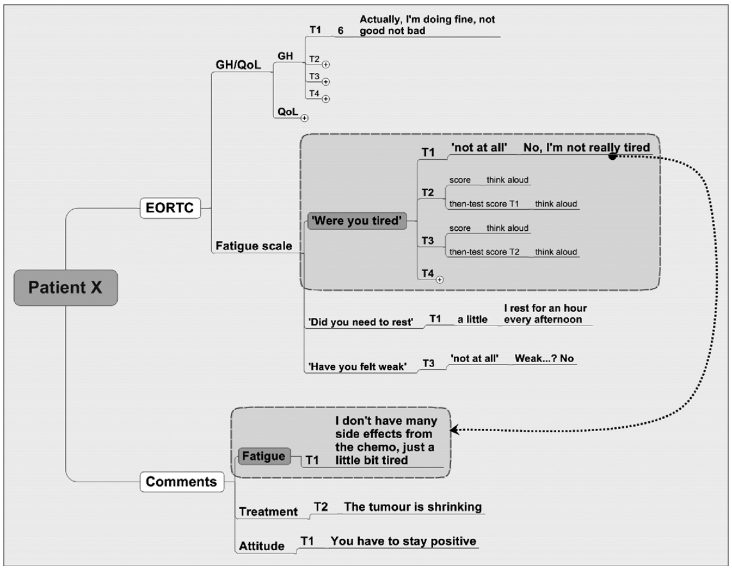

Figure 1 Analysis by means of a mind map. Branch EORTC: scores of GH/QOL and the fatigue scale are organized per interview and complemented by the think aloud data. Branch Comments: core texts of relevant parts of transcripts are organized per code and per interview.
Note: (+) not all branches of the different interview moments are shown.

Methods

Procedures and study sample

Between March 2001 and September 2003, we recruited newly-diagnosed patients with SCLC who were evaluated for 1st line chemotherapy. The patients were attending one of five outpatient clinics for chest diseases in the Netherlands. To maximize the likelihood that we would interview patients from the beginning of their treatment, we were informed about new patients immediately after diagnosis. No restrictions were made with regard to age or treatment (chemotherapy or a combination of chemotherapy and radiotherapy). Participating patients gave written consent and were interviewed at equivalent points in the treatment trajectory. The first interview (T1) was carried out at the start of chemotherapy. In the original plan the second interview was planned after completion of the course of chemotherapy. However, after inclusion and first interviews of 3 patients we made a decision to interview the patients during treatment as well. Therefore the second interview (T2) was conducted four weeks after T1 and the third (T3) 7-10 days after completion of the treatment with chemotherapy and the fourth (T4) six weeks later. Approval for this study was obtained from the Medical Ethics Committees of the research site and the participating hospitals.

During the course of the study, 41 eligible patients were invited to the study. Four patients were unwilling to participate, 3 died before informed consent could be obtained, and 3 were not interviewed because of imminent death. Of the 31 respondents who were interviewed, 8 were excluded from further analysis because their data were incomplete, i.e. they were only interviewed once (six died within a month after T1 and two were too sick at T2 and died before the end of the planned chemotherapy). Consequently, the final study sample consisted of 23 SCLC patients, of whom12 had limited (3 male and 9 female, mean age 55, range 42-69) and 11 had extensive disease (8 male and 3 female, mean age 64, range 39-72). All patients received standard chemotherapy, except for 7 patients whose chemotherapy was combined with local radiation of the tumour. The majority of the patients were married (19, 83%), and had children (17, 74%).

Of the 23 patients in our study sample, 15 were interviewed four times, 7 were interviewed three times, and one patient was only interviewed twice resulting in a total of 83 interviews. The interviews were conducted by MW in the homes of the patients. In three cases the 1st interview was held in the hospital. Interviews averaged 80-110 min.

Materials and qualitative method

In this exploratory, longitudinal multiple-case study, QoL was assessed with EORTC QLQ-C30 (version 3.0)[2] and the lung cancer module QLQ-CL13.[8] The EORTC QLQ-C30 is the most widely used cancer-specific QoL instrument in European clinical trials. The questionnaire is composed of several scales, which measure among others physical function, mental health, general health and global QoL. Furthermore, it measures different symptoms such as pain, dyspnoea, nausea and fatigue. The Fatigue Scale consists of 3 items: ‘did you need to rest’, ‘have you felt weak’ and ‘were you tired’ (respectively questions 10, 12 and 18), for which there are 4 response categories: ‘not at all’, ‘a little’, ‘quite a bit’ and ‘very much’ (respectively scores 1, 2, 3 and 4).

The EORTC QLQ-C30 and CL13 were completed in combination with the Three-Step Test-Interview (TSTI) to investigate how respondents interpreted the items and how they responded to them. The TSTI consists of the following steps[9] : 1) *concurrent think aloud*, aimed at collecting observational data on how a respondent completes the questionnaire, expressing his thoughts aloud; 2) *focused interview*, aimed at clarifying respondents’ previous expressions while completing the questionnaire; 3) *semi-structured interview,* aimed at eliciting respondents’ experiences and opinions with regard to the questionnaire.

Interview protocol

Each interview was conducted in an identical format. At T1, QoL was measured with the EORTC QLQ-C30, followed by the lung cancer module QLQ-CL13. The questionnaire

was conducted in a concurrent think aloud manner and after completion, respondents were asked to clarify previous hesitations, expressions when rating certain items and experiences (i.e. second and third step of the TSTI). Individual QoL was then measured with the Schedule for the Evaluation of Individual Quality of Life – Direct Weighting (SEIQoL-DW).[10-11] Finally, we encouraged patients to talk freely about the impact of diagnosis and treatment. At follow-up (i.e. T2, T3 and T4), after the SEIQoL-DW assessment, the EORTC questionnaires were administered a second time as a so-called 'then-test' [12-14] (i.e. the patients filled out the questionnaire in reference to how they perceived themselves as they were in the previous interview). In these 2nd and following interviews, EORTC assessments were conducted in a concurrent think aloud manner and with the second step of TSTI integrated in the assessment. In fact, we encouraged patients to think aloud and we probed for clarification after each item in the case of extra information was considered useful to understand patients' answer. We used a flexible approach in order not to interrupt the natural flow of both the assessment as well as the patient-interviewer communication.
The interviews were audio-taped and transcribed verbatim. In this article we focus on the EORTC QLQ-C30 question 'were you tired'.

Analysis

Three types of data were collected and used for analysis[15]: 1) completed EORTC questionnaires (T1-T4), 2) observed respondent behaviour recorded in field notes, and 3) transcriptions of the interviews, including 'think aloud'. The analysis was aimed at identifying discrepancies, response strategies and explanations of response behaviour. We used the qualitative computer package Kwalitan 5.0 (http://www.kwalitan.net) to extract relevant parts of the transcriptions: 1) 'think aloud' of the question 'were you tired' and of other items that were useful in understanding the response behaviour to the question on fatigue (i.e. two other items of the fatigue scale, the general health and the global QoL question of the QLQ-C30), and 2) 'comments' related to fatigue symptoms, impact of treatment on perceived QoL, and attitudes towards life. In order to deal with the still remaining large amount of extracted data, two of the authors (MW, AT) condensed extracted transcripts of the 'comments' into core texts. For each patient, the data (i.e. think aloud combined with scores and core texts of comments) were organized per interview in one mind map (see example in Figure 1) by means of the computer package Mindjet Mindmanager Pro 6 (http://www.mindjet.com). Furthermore, a different mind map was made to organize think aloud data related to the question 'were you tired' for all patients per response category per assessment, including then-test. For the analysis, three authors (MW, AT, TH) each independently read the mind maps of each patient. They studied patients' scores, their think aloud responses, and examined whether response shift type explanations would be provided: recalibration (i.e. using different standards of comparison to assess fatigue over time), reprioritization (i.e. changes in the importance attached to fatigue over time) and reconceptualization (i.e. changes in the meaning of fatigue over time). Two researchers (MW, AT) searched for additional explanations in the core texts to account for the response behaviour and the discrepancies. The research team (MW, AT, TH, MS) discussed critically the different response strategies used by the patients and the robustness of the interpretations of response shift.

Patient Mary

Mary was 60 years old and married. She had two sons and two grandchildren. Her answer to the EORTC question was consistently 'not at all', except for the then-test concerning the interview T1. This suggests that she did not suffer from fatigue in the week prior to the interviews and that fatigue due to chemotherapy did not have any impact at all on her energy level. But, during the interview she spontaneously provided information that indicated that chemotherapy had an impact on her life and that she regularly suffered from fatigue.

T1 EORTC score 'not at all'
Think aloud: Were you tired… no not at all, no, no difference compared to the past
Interview: I'm getting tired at the least little thing.

T2 EORTC score 'not at all'
Think aloud T2: Not tired, last week, not at all
Think aloud then-test T1: A little, more than at the moment.
Interview: According to the doctors, the X-rays were very good no. I'm very optimistic, sometimes I'm tired but that's my own fault. I don't have as much energy as I did before I became ill.

T3 EORTC score 'not at all'
Think aloud: Last week I wasn't tired, it's the second week after my chemo, not at all tired
Think aloud then-test for T2: Not at all
Interview: Yes, it was my last cycle of the chemo, I was afraid that I wasn't going to be able to carry on through the treatment. But I managed, okay; I'm tired but apart from that… nothing at all.

T4 EORTC score 'not at all'
Think aloud: No I wasn't tired last week, not at all
Think aloud then-test for T3: Not at all
Interview: Now and then, I'm tired. It's different to before my chemotherapy. Sometimes I'm so tired, so tired, more than in the past. It comes suddenly…in the middle of the day.

Box 1. Example of a patient with discrepancies in reported level of fatigue

Results

Patients with or without discrepancies

Of the 23 patients, 15 (5 male and 10 female, age 46-72) showed discrepancies at least at one measurement point, i.e. differences between their answer to the EORTC question 'were you tired' and their level of fatigue spontaneously reported during the interview. In their answers to the EORTC questionnaire they all presented themselves positively and said that they were not tired. Mary, for example, was 60 years old at the time of the first interview. During all her interviews she reported that she was tired, but she consistently answered 'not at all' to the EORTC question (Box 1). Only once she scored 'a little bit'. This was at T2 when filling in her questionnaire as a then-test for T1 and she commented "*a little, more than at the moment*".

Patient Ann

Ann was 47 years old and living with a partner. She didn't have any children. Her scores suggest that the chemotherapy had a slight impact on her energy level during her treatment and a greater impact six weeks after completion of the treatment. But, the interview and the think aloud provided information that indicated that the chemotherapy had a growing impact on her life, and the score 'quite a bit' at T4 was the result of bad news (i.e. a recurrence of the tumour). Furthermore, her data show examples of different response strategies, comparison with more sick patients and self-presentation.

T1 EORTC score 'not at all'
Think aloud: Were you tired.. in principle I wasn't tired. I was mentally tired, it costs me a lot of energy to talk with my relatives. I think you mean physically tired. You're tired in the sense that you can hardly put one foot in front of the other. That's being tired. No I'm not tired.
Interview: I want to be realistic, think positively. I try not to worry; there is nothing I can do. I don't know when I will die. I get angry when people are surprised…if I say I'm doing fine…it's my decision how I'm feeling.

T2 EORTC score 'a little bit'
Think aloud: I feel it a little bit, compared to other people who are very sick. So, if I have pain or when I'm tired I say to myself don't complain, so everything I feel, I only feel a little. I'm doing fine.
T*hink aloud then-test for T1:* A little
Interview: Yesterday, I worked for three hours and I was exhausted. I went to bed in the middle of the day in order to be able to show my friend that I'm doing fine. She has trouble in coping…me… having cancer.

T3 EORTC score 'a little bit'
Think aloud: A little, yes because I have the feeling that I was able to get over it
Think aloud then-test for T2: I think that I've said a little last time, it felt a little, but I shift my limit.
Interview: It's a kind of tiredness, I don't know. I've never been like that. To allow yourself to be tired. I think that the story they all tell, that the last cycle of chemo has the most impact, I think that's very very true.

T4 EORTC score 'quite a bit'
Think aloud: Yes, I was quite a bit tired, but only mentally tired. I have to adjust to the idea of a new course of treatment and radiation. My health is excellent. I'm able to do everything I like, better than six weeks ago. But, with all the medicine I'm taking to suppress the epileptic fits… I'm scared, just like after the start of chemo.
Think aloud then-test for T3: A little but I am not really sure, the pain which I had in my ankels made me tired.
Interview: The radiation will make me tired, but it has not started yet, so I am not tired yet.

Box 2. Example of a patient using different response strategies, comparison with more sick patients, response shift and self-presentation.

For many of our respondents, every new cycle of chemotherapy had a more severe impact on their energy level. Therefore, the highest level of fatigue was to be expected after the 5th and last cycle of chemotherapy, at T3. However, the 15 patients with discrepancies all answered the question 'were you tired' with 'not at all' or 'a little'.
For example, during her interviews at T2 and T3, Ann reported the growing impact of every cycle of chemotherapy, but her answer to the EORTC question at both interviews was 'a little'(Box 2).

Think aloud about the question 'were you tired'

I'm not tired all the time
I'm only tired in the afternoon
No not at all, I'm not tired at the moment, it comes suddenly
I have to be honest, sometimes I'm tired, I can't say not at all, otherwise I would be lying

I'm not really tired, it's something else
No not tired, it's the flue, that's why I'm tired
It's not being tired you know, it's more like being restless
I'm not physically tired, I'm mentally tired
Actually, I can't be tired because the Hb level in my blood is okay

I've no problems with it
I'm currently doing nothing, so I've no problems, I'm not tired
Of course, you can make yourself tired, but I' don't
I can still cope with it; I don't want to exaggerate

I'm a little bit tired but it's due to something else
A little, but it was my own fault, I did too much
I didn't have a proper meal, that's why I was tired
I didn't have my lady working for me in the house, she went on holiday
It's because I've got problems with my voice caused by the radiation

Box 3. Examples of different response strategies used by patients with discrepancies (n=15)

Patients with discrepancies were identified in both stages of disease and with both treatment regimens, i.e. 12 patients (LD *n*=5, ED *n*=7) treated with chemotherapy and 3 LD patients treated with chemotherapy and radiotherapy. During the course of the treatment (T1-T4, 55 interviews) they answered the question 'were you tired' 20 times with 'not at all, 29 times with 'a little', 4 times with 'quite a bit' and twice with 'very much'(see individual scores per interview in Table 1).

No discrepancies were identified in 8 of the 23 patients (LD *n*=4, ED *n*=4, age 39-72). They answered the question 'were you tired' at the end of chemotherapy (T3) with 'quite a bit' or 'very much'. During the course of treatment (T1-T4, 28 interviews) these patients answered the question twice with 'not at all', 9 times with 'a little', 11 times with 'quite a bit' and 6 times with 'very much' (Table 1).

Then-test scores were dissimilar with scores of the previous assessment in 25 out of 52 cases, with higher then-test scores in 16 cases. Transcripts showed that patients had difficulty remembering either the previous measurement point and/or their fatigue at that time.

Patients' characteristics										
Discrepancies	M/F	Age	LD/ED	T1	T1t	T2	T2t	T3	T3t	T4
P 02	Male	57	ED	3	3	-	-	1	-	-
P 04	Female	50	LD	1	2	2	3	2	3	2
P 08	Female	69	ED	2	2	1	2	1	2	2
P 09	Male	66	ED	3	2	2	2	2	2	2
P 10	Male	46	LD	2	2	2	2	2	2	2
P 12	Female	47	LD	1	2	2	2	2	2	3
P 15	Female	69	LD	1	2	2	3	2	3	2
P 17	Female	64	ED	2	2	2	2	2	-	-
P 18	Male	72	ED	1	1	2	-	1	-	3
P 21	Male	69	ED	2	2	2	3	2	2	2
p 22	Male	55	LD	4	1	1	-	2	-	-
P 24	Female	56	LD	1	1	1	2	1	2	1
P 26	Female	59	LD	4	2	1	2	2	-	-
P 32	Female	60	LD	1	2	1	1	1	1	1
P 34	Female	51	LD	1	1	2	1	2	1	1
No discrepancies										
P 01	Female	42	LD	2	2	-	-	4	4	3
P 03	Female	64	ED	3	1	-	-	3	3	4
P 13	Male	72	ED	3	3	2	3	3		2
P 14	Male	39	LD	4	3	2	2	3	-	-
P 16	Male	68	LD	1	1	3	3	3	3	2
P 20	Female	44	LD	1	-	4	-	4	-	4
P 27	Male	69	LD	3	2	2	-	-	4	2
P 29	Male	63	ED	2	3	3	2	3	3	2

Table 1. Individual (then-test)scores of patients answering the EORTC QLQ-C30 question 'were you tired'. Response categories 1, 2, 3 and 4 are representing respectively the category 'not at all', 'a little', 'quite a bit' and 'very much'. Small-cell lung cancer patients (*n*=23), limited (LD) and extensive (ED) disease receiving 1st line chemotherapy were interviewed at equivalent points in treatment: at start of chemotherapy (T1), 4 weeks later (T2), at end of chemotherapy (T3), and 6 weeks later (T4). T1t, T2t and T3t are representing then-test scores obtained at respectively T2, T3 and T4, when patients are asked to provide a renewed evaluation of their fatigue at the previous assessment. Two groups were identified: patients with (*n*=15) and without (*n*=8) discrepancies between their questionnaire answer and fatigue spontaneously reported in the interview.

Response strategies for the four response options

The think aloud texts for the response categories ‘quite a bit’ and ‘very much’ were minimal in the entire study population. Only a few patients reacted briefly during the think aloud, e.g. “*quite a bit, too tired to keep my eyes open*”, “*yes quite a bit, very tired*” and “*next week it will be better…very much*”. The same pattern was found in all patients for all four response options when filling in the questionnaire as a then-test, e.g. ”*a little bit, I think*”, “*tired then, no*” and “*very much*”. However, patients in the group with discrepancies had much more to say in the conventional QoL measurement when choosing the options ‘not at all’ and ‘a little’. During ‘think aloud’, they seemed to justify the chosen response category. They used various strategies to moderate the impact of fatigue on their life. We summarized their strategies in four categories: 1) I am not tired all the time (e.g. “*only in the afternoon*”); 2) I am not really tired, it’s something else (e.g. ” it‘s the flu”); 3) I have no problems with it (e.g. “*I can still cope with it; I don’t want to exaggerate*”); 4) I am a little bit tired but it is due to something else (e.g. “*I didn’t have a proper meal*”).

Many of our respondents said that they had expected to become very tired as a result of the treatment, but that they were not as sick as they had expected. They were very happy that they were able to cope with the treatment, and had adjusted to the situation. A male patient, for example, had a score of ‘not at all’ at T2, and explained: “*I’m currently doing nothing, so I’m not tired*”. The respondents indicated that they wanted to be honest when filling in the questionnaire and did not want to lie or to exaggerate their fatigue and, because many patients were not tired all the time they considered a score of ‘not at all’ or ‘a little’ to be a suitable score.

Optimism

Most patients told the interviewer regularly that they had adapted to the situation and had changed their attitude towards a more optimistic perspective. They were not hopeful immediately after diagnosis, but optimism about recovery increased when the tumour was shrinking. Of the 23 patients 17 reported spontaneously that they were optimistic: e.g. “*I’ve got good news, I’m as optimistic as can be”, “I’m full of hope because I was diagnosed in an early stage, so I’m good in time*”. Furthermore, they said that they felt better off than expected, compared to patients who were worse off e.g. *“I’m lucky not to be very sick, compared to the patients I saw at the hospital*”. Although they experienced the impact of every new cycle of treatment as more severe, they said to be able to cope with the treatment and to accept the side-effects: e.g. “*it’s part of the package, I’m willing to put up with, knowing the chemo is doing the job properly”*. Some patients were actually feeling better after each cycle and happy that they were still alive: e.g. “*It was much worse than I wanted to admit last time, I’m feeling much better*”, “I’ve *already a couple of months extra*”.

In contrast, 5 of the 23 patients expressed pessimistic feelings: e.g. *"I'm a broken man, hard work all my life and now…I don't think I've much time left"*, *"I'm a bit depressed, when does it stop, if it doesn't stop it would be better if my life was over"*, *"No plans for the future, you never know when the tumour will come back"*. These patients all reported high levels of fatigue during the course of the treatment. One patient did not provide specific comments about optimism or pessimism.

Spontaneously reported coping behaviour

Protective behaviour
I'm trying to avoid or minimize pessimistic thoughts
I don't think about it, otherwise I can't cope with it
We don't talk about it, just follow my every day routine
I'm building a wall around myself

Assertive behaviour / power display
I'll show others that I'm managing all right
You have to be positive
You have to believe in yourself, otherwise you can't manage it anymore

Fighting the stigma
I'm not the cancer patient my neighbour thinks I am
They think I'm lying on my bed all day
People look at me, and give advice that I don't want
I have to admit that I really am a cancer patient… I didn't want to be (see patient Ann, Box 2).

Box 4. Examples of coping strategies used by patients with discrepancies (*n*=15)

Response shift and self presentation

In both groups – with and without discrepancies - we found patients who had reported to have changed their reference point after T1 (i.e. recalibration in contrast to T1). They compared their fatigue at the second and following interviews with that of other patients (e.g. *"I was tired, yes, but compared to the patients I've seen in the hospital, I'm just a little bit tired"*) or, with the period in which they were more tired (e.g. *"Compared to the first week after chemo, it's the second now … I'm not tired "*). In one case, a patient spontaneously re-evaluated her previous measurement: *"I told you that I was really tired then, but compared to how I'm feeling now, it was then just a piece of a cake"*. Another one spoke about a shift of limits which also suggests recalibration: *"I already told you that I would change my standards"*.

We did not find indications of reconceptualization and reprioritization of fatigue. The only exception was Ann who made a distinction between being physically and mentally tired (see Box 2). At T1, she said that she was mentally tired but not physically and her answer was 'not at all': *"Tired means that you can hardly put one foot in front of the other"*. At T4, she said that she had recovered from chemotherapy and was physically able to do anything she wanted but, unfortunately, suffered from sudden epileptic attacks caused by metastases. She was feeling anxious in the same way as at the start of her chemotherapy, and had to consider further treatment options. Just like in the interview at T1, she said that she was mentally tired but not physically. However, this time her score was ‘quite a bit’ instead of ‘not at all’: “*Purely, because I was mentally tired last week. I have to adjust to the idea of a new course of treatment. Actually, I have to admit that I really am the cancer patient I never wanted to be* ”. This response pattern might be interpreted as reprioritization (i.e., changes in the importance attached to mental fatigue over time).

Because the above mentioned response shift type explanations could not adequately explain our conflicting findings in the discrepancy group we questioned: “Why are patients presenting themselves in the questionnaire more positively than in the informal interview”. In our search for an other explanation, we found that 13 of the 15 patients with discrepancies had spontaneously reported how they dealt with having cancer and the perspective of a short life-expectancy. We summarized their comments in three categories (see examples of coping strategies in Box 4): 1) *Protective behaviour* (e.g. protecting themselves from harmful thoughts); 2) *Assertive behaviour/ power display* (e.g. projecting the image of being positive and managing all right); 3) *Fighting the stigma* (e.g. fighting against being stigmatized).

Taking these strategies into account, we concluded that a possible mechanism underlying the discrepancies in this group was ‘self-presentation’. As the questionnaires are explicitly related to cancer and since these patients want to distance themselves from being reduced to only a cancer patient, they want to present themselves as a person who just happened to have cancer. Therefore, they applied various strategies to respond to the question on fatigue in order to produce a score that was as favourable as possible and presented themselves as positive and managing their fatigue.

Discussion

Two third of the patients showed discrepancies in their reported level of fatigue. They reported a gradual decrease in energy at the end of chemotherapy, but they were ‘not at all’ or just ‘a little bit’ tired according to their answer to the EORTC questionnaire, with ‘underreporting’ as a result. They presented a positive image of themselves and used various strategies to explain their choice of response category. A predominant finding was that patients adopted a more optimistic perspective on the treatment. Interestingly, this was not exclusively found in the discrepancy group. The same was true for recalibration and for the only indication of reprioritization. These response shift type explanations did not sufficiently account for the conflicting findings in our discrepancy group.

Self-presentation was found to be an additional (coping) mechanism underlying the discrepancies. Our results suggest that patients are not only concerned about the impression they make on others. They try to protect themselves from negative thoughts and they also feel the need to be positive and to distance themselves from the stereotypical cancer patient. With this strategy they are more capable of coping with a situation that they cannot change.

The suggestion that self-presentation is an underlying mechanism is supported, for example in the case of Ann. She told us that after a recurrence of the tumour she adopted the realistic perspective by admitting that she really was 'the cancer patient', which she did not want to be before. It seems that she gave up her attitude of showing others that everything was all right, and for the first time she did not present her self as more positive than she actually was as she did before.

Self-presentation (also called impression management [16-17]) is a phenomenon described by Leary *et al.* in relation to health behaviour. [18] They discussed its implications for research in health psychology. Our study shows that, in addition to response shift, self-presentation may explain unexpected results, at least in SCLC patients. The question 'were you tired' in the EORTC-QLQ-C30 does not unequivocally measure the impact of chemotherapy on the energy level of patients; in fact, with their responses, patients seem to show how that they are managing the situation. From our results we cannot conclude that in the group without discrepancies 'self-presentation' is not present at all, or that whenever self-presentation occurs discrepancies will also be present. However, our study does show that 'self-presentation' affects QoL measurement. These findings must be taken into account when investigating and interpreting QoL data, also in other study populations. Especially after diagnosis and in the initial phase of treatment, 'self-presentation' might be an important coping strategy. In fact, during each phase in which a new equilibrium and a new identity has to be found (e.g. after a recurrence of the tumour, or metastases) self-presentation might affect QoL measurement.

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3

Listen to their answers! Response behaviour in the measurement of physical and role functioning

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Submitted

Abstract

Background

Quality-of-life (QoL) is considered to be an indispensable outcome measure of curative and palliative treatment, but QoL research frequently seems to produce counter-intuitive findings. Occasionally, the expected deterioration in QoL is not reported, and intra-individual comparisons over time do not seem to be sensitive to change.

Objective

To investigate how patients interpret and respond to questions on the EORTC-QLQ-C30 over time, and to find explanations to account for counter-intuitive findings in QoL measurement.

Methods

Qualitative investigation of the response behaviour of small-cell lung cancer patients (*n*=23) in the measurement of physical and role functioning with the European Organization for Research and Treatment of Cancer Core Quality of Life Questionnaire (EORTC QLQ-C30). Interviews were held at four points during 1st line chemotherapy: at the start of the chemotherapy, 4 weeks later, at the end, and 6 weeks after the end of the chemotherapy. Patients were asked to 'think aloud' when filling in the questionnaire.

Results

The patients used various response strategies when answering questions about problems and limitations in functioning. By taking the wording of questions literally, by guessing their functioning in activities which they did not perform, and by ignoring or excluding certain activities which they could not perform, the patients had scores suggesting that they were less limited than they actually were.

Conclusion

Terminally ill patients evaluate their functioning in terms of what they perceive to be normal under the circumstances. Their answers can be interpreted best in terms of response behaviour, which had been described by Rapkin & Schwartz. Change in the appraisal of QoL explains how levels of physical and role functioning are sustained under deteriorating physical health.

Introduction

Quality-of-life (QoL) is considered to be an important outcome measure of curative treatment, but it is particularly relevant for cancer patients, who receive life-prolonging and ‘palliative’ therapy. Not only the assessment of the burden of symptoms and psychological well-being, but also the assessment of global QoL and physical and role limitations is considered to be important in the evaluation of medical and psychosocial interventions. However, QoL research frequently seems to produce results which are not very consistent. For example, discrepancies have been found between objective and self-rating of health; patients with a chronic disease have been found to report levels of QoL that are equivalent to those of healthy controls, and other patients have rated their QoL better than their caregivers assessment.[1-4] Such findings, explained as ‘underreporting’ by Breetvelt & Van Dam[5], suggest that patients report less emotional distress and dissatisfaction than they *actually* experience, and that intra-individual comparisons over time may not be sensitive to change.

The results of other longitudinal studies also illustrate the problems that are encountered in the interpretation of QoL outcomes. The European Organization for Research and Treatment of Cancer Core Quality of Life Questionnaire EORTC (EORTC QLQ-C30) is designed to measure the physical, psychological and social functioning of cancer patients.[6;7] This instrument was found to be useful, for example, to evaluate the effect of palliative treatment with radiotherapy[8]; but it was found that the role functioning scale was not reliable at all. Groen *et al.*[9] measured QoL with the EORTC questionnaire in patients with inoperable non-small-cell lung cancer treated by radiation with and without chemotherapy. They found no significant change in QoL between pre-treatment and on-treatment, despite an observable increase in the side-effects of treatment with radiotherapy. Furthermore, Cox[10] studied cancer patients who participated in a clinical trial; she found no significant change in the EORTC assessment, but the in-depth interviews led to alternative conclusions about the impact of the trial on these patients.

In recent years the response shift theory[11], which refers to a change in internal standards, values and conceptualization of QoL, has gained increasing attention in explaining counter-intuitive and paradoxical findings. Therefore, we investigated response shift in QoL measurement in small-cell lung cancer (SCLC) patients during 1st line chemotherapy. We were also interested in other explanations for counter-intuitive findings, and we therefore wanted to find out how patients actually dealt with QoL questions. Recently[12] we reported the discrepancies we found between the levels of fatigue measured with the questionnaire and the levels spontaneously reported during the interview. In addition to the discrepancies we found, we were also surprised by the way in which these patients interpreted the questions about global

QoL and physical and role functioning, and produced scores suggesting that they were less limited than they actually were. We therefore investigated their answers to these questions in more depth.

This paper describes the results of an exploratory longitudinal multiple case study in which we investigated how patients interpret and respond to questions on the EORTC-QLQ-C30 over time. We focused on global health and global QoL (GH/QOL) and items on the physical and role functioning scales. We aimed to search for explanations to account for counter-intuitive findings in QoL measurement.

Methods

Procedures and study sample

In 2000, approval was obtained from the Medical Ethics Committee of the VU University Medical Center for our study 'Response shift in quality of life in the palliative treatment of small-cell lung cancer patients'. In March 2001 we started to recruit SCLC patients in five outpatient clinics for chest diseases in the Netherlands. All patients were evaluated for 1st line chemotherapy and no restrictions were made with regard to age or treatment (chemotherapy or a combination of chemotherapy and radiotherapy). The participating patients gave written informed consent and were interviewed during the treatment trajectory. The first interview (T1) was carried out at within 7-10 days after diagnosis at the start of the chemotherapy. In the original study design the second interview was planned after completion of the course of chemotherapy. However, after the inclusion and first interviews with 3 patients we decided that we would also interview the patients during the treatment. Therefore, the second interview (T2) was held four weeks after T1, the third (T3) 7-10 days after completion of the chemotherapy treatment, and the fourth (T4) six weeks later.

Between March 2001 and September 2003, 41 eligible patients were invited to participate the study. Four patients were unwilling to participate and 6 were not interviewed because of their imminent death. Of the 31 patients who were interviewed, 8 were excluded from the analysis because their data were incomplete, i.e. they were only interviewed once (six died within a month after T1 and two were too sick at T2 and died before the end of the planned chemotherapy). Consequently, the final study sample consisted of 23 SCLC patients, 12 of whom were diagnosed with limited disease (3 male and 9 female, mean age 55, range 42-69) and 11 with extensive disease (8 male and 3 female, mean age 64, range 39-72). Except for 7 patients whose chemotherapy was combined with local radiation of the tumour, all patients received standard chemotherapy. The majority of the patients were married (19, 83%), and had children (17, 74%).

Of the 23 patients in our study sample, 15 were interviewed four times, 7 were interviewed three times, and one patient was only interviewed twice resulting in a total of 83 interviews. The interviews were conducted by MW and had an average duration of 80-110 min. Except for the first interview with 3 patients, all interviews were held in the patient's home.

Materials and qualitative method

In this exploratory, longitudinal multiple-case study, QoL was assessed with the EORTC QLQ-C30 (version 3.0)[7] and the lung cancer module QLQ-CL13.[13] The EORTC QLQ-C30 is the cancer-specific QoL measurement instrument that is most widely used in European clinical trials. The questionnaire consists of several functioning scales which measure, among other things, physical and role functioning, mental and general health, and global QoL. Furthermore, it measures different symptoms such as pain, dyspnoea, nausea and fatigue. For the questions about functioning the respondent has four response options. The respondent circles the most appropriate number, i.e. 1) *not at all*, 2) *a little*, 3) *quite a bit* and 4) *very much*. Furthermore, general health and global QoL are rated by circling a number between 1 and 7 (respectively from *very poor* to *excellent*). The QLQ-CL13 module measures the extent to which patients experience symptoms or problems related to their lung cancer, such as coughing and hair-loss.

The EORTC QLQ-C30 and the CL13 were completed in combination with the Three-Step Test-Interview (TSTI) to investigate how respondents interpreted the items and how they responded to them. The TSTI consists of the following steps[14] : 1) *concurrent think aloud*, aimed at collecting observational data on how respondents complete the questionnaire, expressing their thoughts aloud; 2) *focused interview*, aimed at clarifying the respondent's previous expression of thoughts while completing the questionnaire; 3) *semi-structured interview,* aimed at eliciting the respondent's experiences and opinions with regard to the questionnaire.

Interview protocol

Each interview was conducted in an identical format. At T1, QoL was measured with the EORTC QLQ-C30, followed by the lung cancer module QLQ-CL13. The questionnaires were completed in a concurrent think aloud manner, and after completion the respondents were asked to clarify previous hesitations, expressions when rating certain items and experiences (i.e. second and third step of the TSTI). Individual QoL was then measured with the Schedule for the Evaluation of Individual Quality of Life – Direct Weighting (SEIQoL-DW).[15;16] Finally, we encouraged patients to talk freely about their experiences with the treatment. At follow-up (i.e. T2, T3 and T4), after the SEIQoL-DW assessment, EORTC questionnaires were completed again as a so-called 'then-test'[17-19] (i.e. the patients filled in these questionnaires according to how they perceived themselves at the time of the previous interview). In the second and following interviews, EORTC questionnaires were completed in a concurrent think aloud manner, and with the second step of the TSTI integrated in the assessment. We encouraged patients to think aloud and we probed for clarification after each item if extra information was considered to be useful in order to understand the patient's answers. We adopted a flexible approach in order not to interrupt the natural flow of both the assessment and the patient-interviewer communication.

The interviews were audio-taped and transcribed verbatim. In this article we focus on the physical functioning scale (PF, items 1 to 5), the role functioning scale (RF, items 6 and 7) and global health and QoL rating (GH/QOL, items 29 and 30).

Analysis

Four types of data were collected and used for the analysis[20]: 1) completed questionnaires (T1 t/m T4), 2) observed patient behaviour recorded in field notes, 3) transcriptions of the interviews, and 4) memos with background information regarding the illness trajectory of each patient. Firstly, three authors (MW, AT, TH) independently analysed the first two interviews and questionnaires completed by the first patient. This analysis included all questions on both of the EORTC questionnaires, and the aim was to understand why a specific response category was chosen, and to identify change in the response behaviour by comparing the results of the two interviews. The results of our measurement of response shift with the SEIQoL-DW will reported elsewhere.[21]

In our further analyses of all the interviews (*n*=83) concerning GH/QOL, PF and RF, we used the qualitative computer package Kwalitan 5.0 (http://www.kwalitan.net) to extract relevant parts of the transcriptions: 1) think aloud of the GH/QOL questions and the functioning scales (i.e. physical and role) and 2) 'comments' related to the way in which patients were functioning, and the perceived impact of treatment on QoL. Furthermore, in order to deal with the still remaining large amount of extracted data, two of the authors (MW, AT) condensed extracts from transcripts of the 'comments' into core texts. The data were analysed for each patient separately, resulting in 23 case-studies.[22] For each case the data (i.e. think aloud combined with scores and core texts of comments) were organized per interview in one mindmap by means of the computer package Mindjet Mindmanager Pro 6 (http://www.mindjet.com) (see for an example of organizing data by means of a mindmap our article on the EORTC question 'were you tired').[12] For the further analysis, MW read the mindmaps of each patient and investigated how the patient had answered each item (i.e. interpretation of the question, response, and choice of response category). She interpreted the individual score and the think aloud data of that item against the background of the patient's illness trajectory, and assessed whether that item had received a score that one would expect if it truly reflected the limitations the patient experienced. Finally she looked for change in the item appraisal process over time. After completing the individual cases, MW and AT searched for patterns in the response strategies for each item by examining similarities and differences between the cases. The robustness of their interpretations was critically discussed by the research team (MW, AT, TH, MS).

Results

Quality of Life during first-line chemotherapy

For all the patients chemotherapy consisted of 5 cycles. After the first cycle (T2), 13 of the 19 patients said that they were doing well, considering the circumstances. Patients diagnosed with extensive disease who suffered from tumour-related symptoms, such as dyspnoea and coughing, even felt much better than before the treatment. Furthermore, 3 of the 7 patients who had been treated with chemotherapy and radiotherapy, experienced the severe physical burden of the side-effects of radiotherapy (e.g. pain, problems with eating and drinking, and consequent loss of weight). All the patients experienced the impact of every new cycle as more and more severe. Six weeks after the last cycle, 3 patients had died and 2 were confronted with a recurrence of the cancer and further treatment. The others tried to pick up their normal life again.

According to the results of the EORTC-QLQ-C30, QoL was affected very little by the chemotherapy (Table 1). The mean values for GH/QOL at the start (T1) and at the end of the chemotherapy (T3) were both 66. The mean value of the physical functioning scale decreased from 75 at T1 to 72 at T3, but the mean value of role functioning increased from 62 at T1 to 72 at T3; 7 patients had the highest possible rating (100) and only 1 patient had the lowest possible rating (0).

We were not able to interview all 23 patients 4 times, and therefore the mean values presented for T2 and T4 can not be adequately compared with the mean values for T1 and T3. However, despite these differences and the large individual variations, most of the patients perceived their QoL to be at a higher level at T2 than at T1 (start of chemotherapy): 12 of the 19 patients reported a higher level, 2 an equal level, and 5 a lower level of GH/QOL. The patients also reported better QoL at T4 (six weeks after the course) than at T3 (end of the course): 10 of the 18 patients reported a higher level, 4 an equal level, and 4 a lower level of GH/QOL. With respect to the functional scales, the mean values of physical and role functioning also increased six weeks after the course, suggesting that most patients no longer had any limitations: 8 of the 18 patients reported the highest level (100) of role functioning, and only 1 patient the lowest possible rating (0). In order to understand how the QoL outcomes in our study should be interpreted, background knowledge of the response behaviour described below has to be taken into consideration.

Patients' answers at the first QoL assessment

From the questions about physical and role functioning (questions 1 to 7) the researcher can detect the degree of problems or limitations a patient experiences during certain activities. However, the patients responded in unexpected ways: by focusing on one aspect of the question, by taking the wording of the question literally, by ignoring

Table 1. Individual and mean scores of small-cell lung cancer patients (n=23) receiving 1st line chemotherapy and interviewed at equivalent points during treatment: start of chemotherapy (T1), 4 weeks later (T2), end of chemotherapy (T3), and 6 weeks later (T4). Three patients died before T4 (#). Scores range from 0 to 100; higher scores represent a higher level of functioning. Seven patients with limited disease were treated with chemotherapy and radiotherapy (LD X*).

Patient characteristics				T1 (*n*=23)			T2 (*n*=19)			T3 (*n*=23)			T4 (*n*=18)		
No.	M/F	Age	LD/ ED	GH/ QOL	PF	RF	GH/ QOL	PF	RF	GH/ QOL	PF	RF	GH/ QOL	PF	RF
P 12	F	47	LD	83	100	100	100	100	67	66	100	67	83	93	33
P 04	F	50	LD	83	94	67	-	-	-	33	67	83	67	87	83
P 24	F	56	LD	83	100	50	75	93	100	92	93	67	100	100	100
P 17	F	64	ED	83	80	83	83	80	67	83	40	67	#	#	#
P 03	F	64	ED	83	67	33	-	-	-	58	40	0	50	60	33
P 34	F	51	LD	75	100	83	83	100	100	92	100	100	100	100	100
P 15	F	69	LD X*	75	93	83	50	60	67	42	80	67	83	93	100
P 01	F	42	LD X*	66	73	50	-	-	-	50	60	34	83	87	67
P 20	F	44	LD X*	66	87	100	42	73	0	42	80	50	33	47	0
P 32	F	60	LD	66	87	83	100	67	100	83	67	100	83	53	100
P 26	F	59	LD X*	58	42	56	92	93	100	75	92	83	-	-	-
P 08	F	69	ED	50	80	83	33	60	100	75	73	100	83	67	50
P 22	M	55	LD X*	75	73	67	83	75	67	50	60	100	-	-	-
P 21	M	69	ED	75	47	50	83	60	83	83	87	100	83	93	83
P 10	M	46	LD X*	66	87	67	83	87	67	83	93	83	83	93	83
P 09	M	66	ED	66	100	50	83	92	0	92	67	67	75	73	100
P 16	M	68	LD X*	66	67	100	75	73	67	58	67	67	75	87	100
P 18	M	72	ED	66	73	67	50	67	17	50	60	100	66	67	100
P 29	M	63	ED	50	80	50	50	80	67	58	83	33	75	87	100
P 27	M	69	LD	50	53	33	66	53	50	66	67	50	66	80	83
P 13	M	72	ED	50	53	33	66	60	67	66	60	67	42	67	33
P 02	M	57	ED	42	40	33	-	-	-	58	67	100	#	#	#
P 14	M	39	LD	33	53	0	66	73	83	58	60	67	#	#	#
All patients				GH/ QOL	PF	RF	GH/ QOL	PF	RF	GH/ QOL	PF	RF	GH/ QOL	PF	RF
Mean score				66	75	62	72	76	67	66	72	72	68	80	75

or excluding certain activities which they could not perform;the patients produced scores suggesting that they were less limited than they actually were (see Figure 1 for examples). Furthermore, a few patients guessed their level of functioning in activities which they did not perform or used the strategy "*I didn't do it, so I don't have any trouble*" (example see item 2), and other patients compared present with previous experiences (e.g. "*I feel not too bad, compared to last week*"), or with expectations (e.g. "*I feel better as expected*").The following examples illustrate for each question how, during the first interview (T1), a certain strategy resulted in a different score than would objectively be expected.

Item 1: *Do you have any trouble doing strenuous activities, like carrying a heavy shopping bag or suitcase?*

A patient who could hardly walk outside the house answered this question with 'not at all' by arguing that you can't have any trouble if you don't do any shopping*: "I never carry a shopping bag, my wife does the shopping"* [M, age 72, T1]. At T1, 17 out of the 23 patients interpreted this item more or less literally, and focused on just shopping bags or suitcases: "*I can lift the shopping bag but I can't walk with it*" or "*my suitcase has wheels, so I don't have to carry it*". Out of the other 6 patients, 4 were thinking about other streneous activities when evaluating this item, and 2 were just circling the number which corresponded with 'a little' and explained that they had already been limited for a longer time. It seemed that they were therefore suggesting that their limitations were considered as 'normal'.

Item 2: *Do you have any trouble taking a <u>long</u> walk?*

For many patients this question was difficult to answer, because they had not had long walks during the previous weeks. Of the 23 patients, 11 first struggled with the definition of 'long' (which differed from 10 minutes to 2 hours or from 500 metres to 10 kilometres) and then they tried to guess their limitations: *"What is a long walk, 5 kilometres…? I never go for a long walk, I don't like serious walking,, but I think I would be limited…. a little?"* [M, age 68, T1]. The other 12 mentioned a recent walking experience, and most of them tried to guess: *"I haven't had a walk in the past weeks, so I don't know, but I walked with my son through the corridors in the hospital and that went fine. So, I haven't tried …a little?"* [F, age 69, T1], and 2 used the same strategy as mentioned earlier (i.e. I didn't walk, so I don't have any trouble).

Item 3: *Do you have any trouble taking a <u>short</u> walk outside of the house?*

Most patients seemed to have an image of a short walk in their mind, and circled without hesitation a response category. Others remembered a recent experience: *"A little, I've been to the shops at the end of the street and that was enough for me"* [F, age 64, T1].

Item 4: *Do you need to stay in bed or a chair during the day?*
Many patients answered this question by laying the stress on one word in particular. This resulted in higher levels of functioning than would be expected. For example, 7 out of the 23 patients took the word 'need' very seriously and did not report any limitation because *"it's not really necessary to stay in bed or on a chair"*. Another patient interpreted the word 'need' as being prescribed by the doctor. Because he did not have such a prescription, his answer was 'not at all', even though he spent most of the day in his bed [M, age 71, T1]. Another 6 patients interpreted this question with an emphasis on staying in bed, which they did not do during the day. Although these patients said that they took a nap or a rest on a regular basis, they did not take sitting on a chair into account. The other half of the patients did not comment on this item, but just circled the response category of their choice.

Item 5: *Do you need help with eating, dressing, washing yourself or using the toilet?*
One older male patient considered the help which he received with washing and dressing as normal under the circumstances, and said: *"No not at all, I can do it by myself if I want to, I don't really need help, but at the moment I have less energy and help from my wife makes it easier for me"*[M, age 72, T1]. Only two patients needed help with eating, dressing, washing and using the toilet: all the other patients circled the response category of their choice without further comments, or saying that it was self-evident that they were not limited.

Item 6: *Were you limited in doing either your work or other daily activities?*
Most of the retired male patients (*n*=6) answered with 'a little' or 'not at all', arguing that they were not working anymore and therefore not limited. All female patients who did not have a job (*n*=8) reported during the first interview the limitations they experienced in housework. The other 9 who worked had taken sick leave, and only 3 of them (i.e. two male patients with their own business and one female patient with a part-time job) were actually thinking about their job and reported limitations. The other 6 acted differently, and ignored their work and focused on activities in and around the house: *"I can do some work in the house, like sweeping the floor, so I'm not limited"* [M, age 46, T1]. It seems that they considered not working as normal under the circumstances.

Item 7: *Were you limited in pursuing your hobbies or other leisure time activities?*
During the first interview this question was answered with 'very much' in some cases when pursuing hobbies was not possible: *"I always played billiards with my friends. I could play at the moment but I don't, because then I'll drink a couple of beers and that's not a good idea now"* [M, age 39, T1]. Other patients reasoned as mentioned earlier (i.e. I didn't do it, so I'm not limited) : *"I've not played the piano in the past few weeks…no, not at all"* [M, age 68, T1]. The answer suggests that the patient is feeling physically

able to play, but that he does not take into account the fact that his illness might be the reason for not playing.

Item 26: *How would you rate your overall* <u>*health*</u> *during the past week?*
Item 27: *How would you rate your overall* <u>*quality of life*</u> *during the past week?*
Most patients circled a number between 1 and 7 without much thinking aloud, and therefore after they had completed these two questions the interviewer asked them how they interpreted the questions. Most patients defined overall health as how they were actually feeling: “Overall health…, you’re not a healthy person of course. Well it’s simple, you’re terminally ill, but you’re not really feeling sick so … a 5” [M, age 39 at T1]. In evaluating overall quality of life, a few patients had just circled a number which they thought appropriate, without knowing what QoL meant. Most patients defined overall quality of life as being able to do the things they want to do: “I can’t do things as usual, watching television, reading my paper and going to the bookshop. Walking at this moment is not possible … a 4” [M, age 72 at T1].

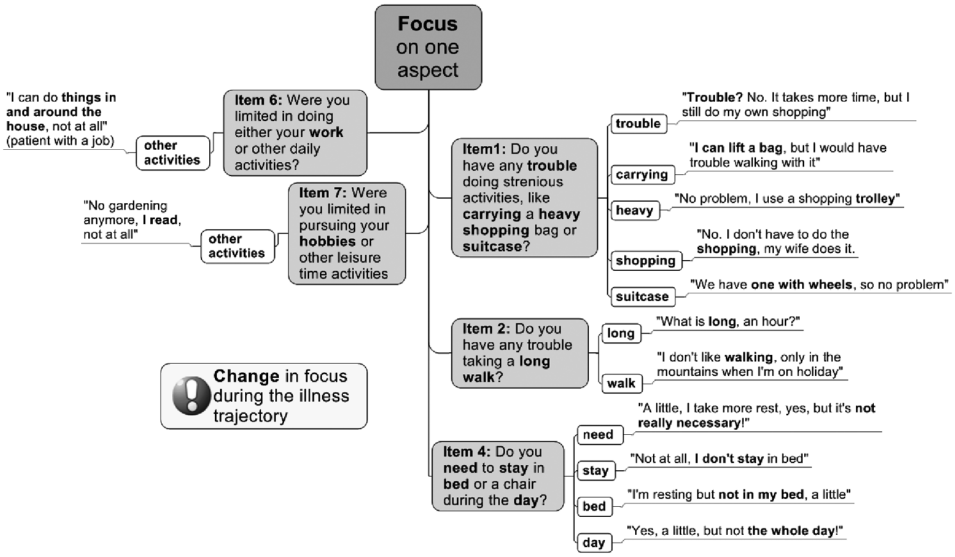

Figure 1. Examples of response strategies used to answer question 1, 2, 4, 6 and 7 of the EORTC-QLQC30 questionnaire. These strategies and change in the use of a certain strategy over time may explain why patients do not report the deterioration in physical and role functioning that would objectively expected.

Change in patients’ answers

The variation in the interpretation and evaluation of the different items described above was not only found between patients, but also in the individual patient over time. Change occurred especially in the sampling of experiences when evaluating problems and limitations concerning items 2, 6 and 7. These changes sometimes resulted in scores suggesting that a patient was functioning better than actually was the case, but we saw the opposite as well. For example, Nina (age 69) changed her interpretation of a long walk (item 2) by adjusting the distance. Her answer can also be interpreted as evidence of response shift, i.e. a change in standards. Her scores reflect an equal level of functioning, but she actually had less trouble in going for a walk at T4 than at T2. This improvement was not reflected in her scores:

[T2] *That's very difficult, to the shopping mall, 450 metres, quite a bit.*
[T4] *A long walk, two kilometres, I walk too fast, it's my own fault, quite a bit.*

Recalling different experiences over time suggested that there was a change in the patients' perspective on "what was considered as normal under the circumstances" was changed. For example, Ralph, age 46, ignored his job (item 6), suggesting that taking sick leave was normal. Six weeks after the course he was actually less limited than during the treatment, but no longer ignored his job. That is why his improvement in role functioning was not reflected in his scores:

[T2] *A little, it depends how I'm feeling. If I have a good day, I can take on the whole world. Vacuum cleaning, my motorbike, my car"*
[T4] *A little, the first day back at work again, the tension having to tell everyone the same story over and over again, but of course I feel much better than I did six weeks ago.*

Another example shows how high levels of functioning are scored, while the patients were actually not functioning any better at all. Like most patients, John (age 69) felt very limited at T1 in pursuing his hobbies, but in his next interview these limitations/ were no longer mentioned.

[T1] *"My hobby is working in the garden, that's very difficult, quite a bit".*
[T2] *"I'm reading at the moment. Gardening is not possible anymore, a little".*

We found no evidence of change in the patients' definition of overall health. Overall health was consistently interpreted as "how I'm feeling" (e.g. *"Yes, you can't miss it. My health is a 7, it feels like a 7"* [F, age 57, T2]. Neither did we find any change in the interpretation of overall quality of life (i.e. "being able to do the things I want to do"), even in the case of a reoccurence of the cancer and brain metastases (e.g. "*it depends on my contacts…that I can do my own things… a 5 is to low … it has to be a 6"* [F, age 47, T4].

Discussion

The mean values of GH/QOL, physical and role functioning scales during the treatment trajectory suggest that the SCLC patients were functioning without many limitations and had a good QOL. Actually these outcomes confirm what was already known from the studies mentioned earlier, and demonstrate the need to investigate more in depth what is actually happening in QOL measurement. A predominant finding was that the patients evaluated the items concerning functioning against a standard of what they perceived as normal under the circumstances. This standard changed when the circumstances and their health changed. Furthermore, our results also suggest that the patients redefined what is important and what is no longer important (e.g. work or hobbies which used to be important before the diagnosis). These findings suggest the occurrence of response shifts during the illness trajectory.

Observing the QoL self-assessment by means of the think aloud procedure has provided us with insight into the black box of what actually happens in repeated QOL measurement. We have lifted the lid just for a little while by listening to the patient who is filling in the questionnaire.The physical and role functioning items appeared to be multi-interpretable, and therefore enabled a patient to give the impression of performing well under the circumstances. Just by ignoring certain activities which are mentioned in a question, or by taking a question literally, patients are able to maintain reasonable levels of functioning, if we are to believe the EORTC data. Through this behaviour looks it seems that, at a subconscious level, the patients are distancing themselves from the meaning behind the question, i.e. measuring the impact of treatment and disease on their functioning. If this is the case, the patients are presenting their situation more positively than it actually is. Previously[12] we found that, in addition to the response shift phenomena, self-presentation is also a coping mechanism that can explain discrepancies in the measurement of fatigue. The aim of this study was not to investigate whether patients present a more positive image of themselves than they experience in their day-to-day life, but was predominantly to observe how patients interpret and answer the questions, and whether they change their way of answering. We found that the patients did not deal with the questions in the way that the researchers had probably intended. At face value, some questions were also found to be unimportant, not applicable to the patients' situation, or not relevant at a certain point in time. These findings are in line with Mallinson's suggestion that problems may arise when response options do not quite fit the questions, because there are no such response options as 'I don't do this' or 'I don't know'.[23] On the other hand, the answers of patients who used the strategy 'I can't do this anymore, so I'm not limited' can also be interpreted as evidence of response shift, i.e. reprioritization. Patients make good and legitimate use of the opportunity the QOL instrument provides to adjust the question to their own situation, and consequently present an image of not being as limited in functioning as one would expect.

Our findings confirm what has been stated by Rapkin and Schwartz[24], who acknowledge that counter-intuitive findings might be explained by change in the appraisal process: "QOL assessment induces a *frame of reference* which depends upon the meanings the individual attaches to questions; in order to respond to any item, individuals necessarily *sample specific experiences* within their frame of reference, and each sampled experience is judged against relevant, subjective *standards of comparison,* and to arrive at a QOL score, individuals must apply some *combinatory algorithm* to summarize their evaluation of relevant experiences and formulate a response". Changes in the perspective of SCLC patients during their treatment trajectory, change in their sampling of experiences when evaluating an item, and in their use of standards of comparison, explain the variance in QOL measurement at the individual level. Rapkin and Schwartz propose a psychometric model of appraisal, which questions the existing methods for establishing the reliability and validity of QOL assessment tools, and they recommend that the assessment of appraisal should be integrated into QOL research and clinical practice. The results of our study show that knowledge about how patients change their interpretation of questions is useful in interpreting QOL data. Therefore, we agree with Rapkin and Schwartz, that research is needed to learn more about the appraisal process.

Conclusion

Terminally ill patients evaluate their QOL under the prevailing circumstances. Changes in the appraisal process, due to differences in the frame of reference, the sampling of specific experiences and the standards of comparison used when rating items, explain how reported levels of physical and role functioning and QOL are sustained under deteriorating physical conditions. Background knowledge about the illness trajectory and appraisal processes is therefore relevant for the interpretation of QOL outcomes.

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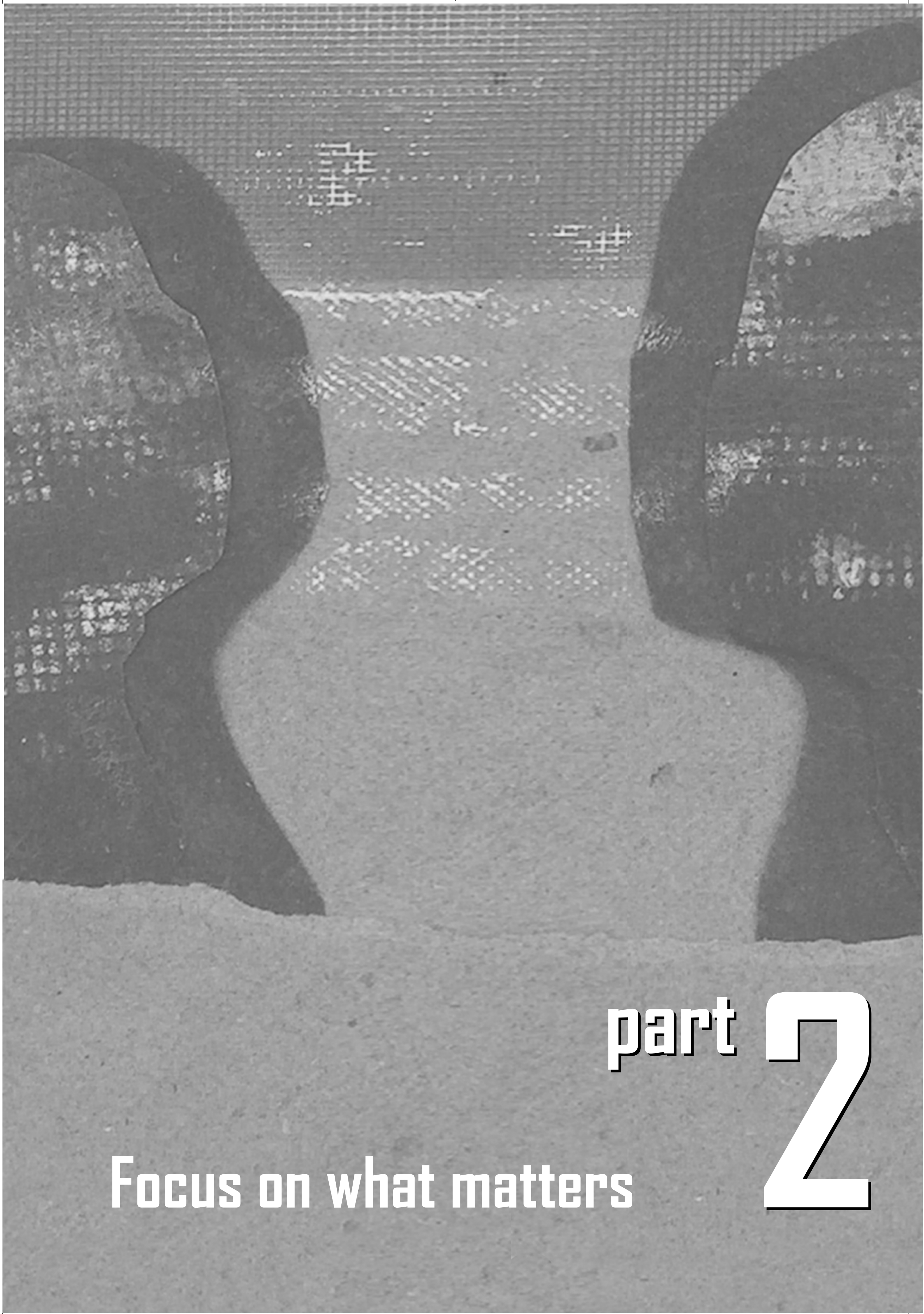

part 2

Focus on what matters

4

Problems eliciting cues in SEIQoL-DW: Quality of life areas in small-cell lung cancer patients

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Harry Groen, Gerrit van der Wal

Quality of Life Research 2006; 15 (3):441 -449

Abstract

The Schedule of Individual Quality of Life – Direct Weighting (SEIQoL-DW) is an individualized approach in the measurement of quality of life in which patients can choose, value and weight five areas that they consider important for their quality of life. Although a number of studies have reported on the feasibility of the administration of the instrument, little is known about how patients choose and define these five areas, the so-called 'cues'. This article describes problems in the elicitation of cues experienced in a qualitative, exploratory study among small-cell lung cancer patients (*n*=31) in the Netherlands. Cues originate from patient-interviewer interaction which is best described as an area of tension between the patient's answers and the instrument instructions. As a result, the interviewer may inadvertently introduce bias while attempting to elicit cues, ultimately affecting patients' SEIQoL-DW measures. In order to prevent possible unnoticed interviewer bias special attention should be paid to the interviewer behaviour. Methods to record the meaning of cues should be considered. More research is needed in order to investigate differences in nominating cues with and without the use of the prompt list.

Introduction

Quality of Life (QoL) is increasingly being incorporated in clinical research and is considered a necessary end point in palliative medicine.[1-3] In traditional approaches to the measurement of QoL, a generic content of relevant domains, criteria and weights is constructed.[4] However, the use of predetermined tools has come under criticism. There are arguments that standard tools contain items that may not be relevant for all individuals and it is assumed that all aspects applied are of equal importance for all respondents.[5] Individual QoL measurements provide the possibility to respondents to represent their individual perspective by indicating and rating the areas that are most important to their QoL. The Schedule for the Evaluation of Individual Quality of Life (SEIQoL) and its shorter Direct Weighting (DW) version is such an individualized approach, in which subjective needs and desires can be identified.The instrument is based on the definition: "quality of life is what the individual determines it to be".[6-9]

The SEIQoL has been validated in healthy subjects and non-healthy populations.[4;6;9-13] The relative strengths and weaknesses of the instrument have been addressed in the literature.[14;15] Moons *et al.*[15] have examined aspects of validity, reliability and responsiveness of the SEIQoL-DW and argue that the instrument measures determinants of QoL rather then QoL itself.

Since the introduction of the SEIQoL and the SEIQoL-DW, many studies have reported on the feasibility of using the instrument to measure QoL in specific patient populations such as patients with amyotrophic lateral sclerosis[16;17], Alzheimer's disease[18], mental illness[19], leukaemia[20], cancer patients participating in phase I clinical trials[21;22] and older medical patients.[23] The SEIQoL-DW has been explored in palliative medicine.[12] Waldron *et al.*[13] report that patients with incurable cancer are very good judges of their own QoL, with very high levels of consistency and validity.

SEIQOL-DW

The SEIQoL is administered in a standardised semi-structured interview format. In the first step the individual is asked to name the 5 areas of life *(cue elicitation)* considered to be important for his/her overall QoL. The second step is to rate the level of functioning on each of the elicited areas. The third step is to weight the relative contribution of each area to overall QoL by means of judgement analysis. To enhance the applicability of this method in routine clinical situations, the quite cumbersome weighting technique of judgement analysis has been replaced with a simpler 'Direct Weighting' (DW) procedure. The relative contribution of each area to the overall QoL is weighted in SEIQoL-DW by means of a set of five coloured disks.[7;10;11] In both the SEIQoL and the SEIQoL-DW, a global QoL score is calculated by multiplying the individual's self rating on each area by the corresponding weight, and summing up the products. The SEIQoL-DW manual describes the procedure which has to be followed in the elicitation, rating and weighting: what to read as an introduction; what to ask and how to rephrase in order to elicit 5 cues and what to say in each further step. Not all cues should be accepted. In case of the respondent volunteers cues which resembles 'quality in life' in meaning (e.g. satisfaction, life quality') the interviewer has to probe for more specific cues such as 'happiness', or 'attitude to life'. It is explicitly advised: *"Elicit areas… NOT individuals, e.g. marriage, not wife"*, and *"Do not give examples"*, and to fully explore and document the meaning of each cue on the 'Cue Definitions Record Form'. Furthermore, potential and most commonly encountered problems in the nomination of cues, the determination of cue levels and the weighting of cues are described. A suggested solution if a respondent cannot think of 5 cues is to use the prompt list with the most commonly elicited cues, i.e. *family, relationships, health, finances, living conditions, work, social life, leisure activities and religion/spiritual life*. This prompt list *"provides for consistency across interviewers where such prompting is absolutely necessary"*. The interviewer must read the list, excluding any cues already mentioned.[7]

Cue elicitation in our Response Shift study

We used the SEIQoL-DW in our study 'Response Shift in Quality of Life in the palliative treatment of small-cell lung cancer patients', a qualitative exploratory longitudinal multiple case-study involving 31 patients. It is suggested that the SEIQoL-DW might be a useful instrument for the measurement of response shift (i.e. a change in the meaning of the self evaluation of quality of life resulting from changes in internal standards, values, or conceptualization).[24] The ability of the SEIQoL-DW to investigate response shift as an important mediator in the accommodation of changes in health status, opens up the possibility to acquire better understanding of how patients make the best of their condition by coping, rethinking and reframing their experiences.[22;25]

When, at different points in time, patients indicate different areas of their life (cues), this could represent a reconceptualization, and a change in their weighting of areas could reflect a change in values. In order to assess these response shifts accurately, cues have to be elicited and recorded in a reliable way.

Most studies mentioned above report on the feasibility of administration, understanding the instrument, the time taken to administer the instrument and the percentage of patients who experienced difficulties in identifying five areas. However, little has been reported about the elicitation and recording of the cues. Campbell & Whyte[21] found that the first two or three cues were nominated spontaneously but thereafter elicitation became more difficult with hesitation from all 15 patients participating in a phase I clinical trial. Only one patient managed to elicit five cues without prompting; all the others required to be read the prompt list. Mountain et al.[23] reported similar findings in their study of older medical patients. Although, both studies reported on difficulties in the elicitation in more detail, they do not report on 'what actually happens' in the elicitation of cues.

In order to get insight into the process of elicitation, i.e. the nomination of reliable and relevant cues, exploration of 'what actually happens' in the interaction between interviewer and patient is necessary. In this article we describe problems in the elicitation of cues. Findings on measuring response shift with SEIQoL-DW will be reported elsewhere.

Methods

Procedures and participants

All patients were recruited from five outpatient clinics for lung diseases in the Netherlands, between March 2001 and September 2003. Patients diagnosed with small-cell lung cancer (limited and extensive disease) and beginning their first-line chemotherapy were reported to the researcher (MW). Of the 41 consecutive patients who were contacted and informed about the study, 34 agreed to participate and gave informed consent; 3 were not interviewed because of imminent death. Consequently, first interviews (T1) were held with 31 patients (16 men and 15 women, aged 39-82 years). Approval for this study was obtained from the ethics committees of the research site and the participating hospitals.

Patient interview

Each first interview was conducted in a format with three consecutive parts. Firstly, QoL was measured with the EORTC QLQ-C30 (general) and the QLQ-CL13 (lung cancer module).[26;27] Secondly, the SEIQoL-DW was explained and administered. Finally, the patients were asked about changes in their perspectives with regard to their life and its quality. The interviews were conducted by the first author (MW). The time taken to complete the SEIQoL-DW was 10–30 minutes. The interviews were audio-taped and fully transcribed, and all but two interviews were held in the home of the patient.

Analysis

Findings are based on three types of data. First, the cues and notes recorded by the interviewer on the Cue Definitions Records Form. Secondly, the audio-tapes and transcriptions of the administration of the SEIQoL-DW. Thirdly, observations of the behaviour of the patient, recorded in field notes made by the interviewer after completion of the interview. These data were analysed for each of the 31 patients separately, resulting in 31 case-studies. The aim in each case-analysis was to investigate the administration process, focusing on the elicitation of cues, the interaction, the role of the interviewer, the ultimate decision on cue labels, the meaning and categorization of the nominated cues, and any possible bias effects. Analyses of interviews in which patients were not able to complete the whole elicitation were also included to illustrate the problems that were encountered. After the individual cases-analyses had been completed, similarities and differences between cases were analysed. The analyses were performed by the interviewer (MW) and checked by two other members of the research team (TH and AT).

Table 1. Areas mentioned as important by patients after diagnosis of small-cell lung cancer

Areas	Cues (n=126) %	Patients (n=26) %
Issues related to family: My husband; my wife; my children, becoming a granny, grandchildren; contact with my grandchildren; support from my family; to sort things out with my wife; ability to enjoy my family and other relations.	30	96
Issues related to health: Fatigue; health; to be cured; feeling physically and mentally well; being able to do what I want to do; becoming healthier; feeling good; not to get too ill; being mobile; getting back to my former daily routine.	14	66
Issues related to social life: Social contacts; social life; contacts in my living environment; friends; relations; support from colleagues; club life; family not directly related.	12	58
Issues related to leisure: Leisure activities; sports; football; playing cards and fishing; sewing; my garden; working as a volunteer at the cemetery.	11	54
Issues related to enjoying life: Having a holiday; to enjoy life; time all to yourself; freedom and happiness; going out everywhere.	10	46
Living conditions: Living conditions; home, garden and pets; housing conditions; a quiet and peaceful well-organized life; norms and values in society.	7	7
Issues related to autonomy: Being independent; my car, my freedom; being physically and mentally independent; doing something on my own; continuing my former independent life.	6	31
Issues related to work: Own shop; moving firm; business; work; working in alternative medicine; my work as baby-sit.	6	23
Issues related to finance: Keeping control of my finances; my wife's budget after my death; not being restricted in budget to enjoy life.	2	12
Attitudes towards life: Positive thinking; putting everything into perspective.	2	8

Findings

Quality of life areas

Out of 31 patients 26 (84%) completed the elicitation procedure and nominated a wide variety of cues (in total 126). Aspects related to *family* were mentioned most frequently: 38 times (30%) by 25 patients; cues in the area of 'health' were nominated

18 times (14%) by 18 patients; 15 cues were nominated in the area of 'social life/other relations' (12%), 14 cues in the area of 'leisure activities' (11%), and 12 cues in the area of 'enjoying life' (10%). Cues and percentages of other areas are presented in Table 1. The content and variety of cues was as expected from reports in the literature. However, unique for our study is that we looked into how these results were produced.

First of all we show how patients responded to the initial question "*What are the five most important areas of your life at present…the things which make your life a relatively happy or sad one at the moment… the things that you feel determine the quality of your life…?*".

Patient

Initial remarks

Out of the 31 patients, there was only one patient (low level of education) who found the initial question too difficult and did not want to continue with the SEIQoL-DW interview. All other patients responded spontaneously and gave a concise description of an area of life which could be labelled as such (*n*=21) or a more elaborate one (*n*=9). Some examples of their replies are the following (cue labels as registered on the Cue Record Form in underline print):

Well, to recover… as clear as daylight... that's it [M, age 46]

That's not so difficult… as soon as I can, walk into town and back again [M, age 83]

Well… my family and my grandchildren, that's really something to fight for [F, age 56]

That's really a difficult question…. my quality of life… well,… to be able to get up at a reasonable time, to sit at the table and to read the morning newspaper… to sit in the sun… to watch the news… [data omitted]… if it's possible just to continue with my everyday life… yes, please [M, age 72]

Five cues

Further nomination of five cues appeared to be a task that was sometimes arduous. Twelve patients had no difficulty in nominating 5 cues spontaneously. One patient decided that 2 cues (*relations* and *independence*) were enough for being her "concept of QoL". Thirteen patients needed to be prompted with the list: 6 patients for 3 cues; 5 patients for 2 cues, 2 patients for 1 cue and 1 patient was not able to nominate the fifth cue even with help of the list.

With 4 patients, who had mentioned 1 or 2 important areas in their initial remarks, the interviewer decided not to continue with the SEIQoL interview. Further elicitation was difficult for various reasons. One patient who had been fired from his job a few years previously was very emotional, and kept on talking about his work. One patient was overtaken by a sudden wave of tiredness. One patient was absorbed by her fears that chemotherapy would harm her eyesight, which was already poor. In a very personal and emotional story, one patient talked about his illness, his fears about the future of his family, and the injustice that he should get this disease after having worked so hard for 45 years. Starting the elicitation process again after this sad story seemed inappropriate.

Response style

The response style differed between patients. Out of 26 patients who completed the SEIQoL-DW, 17 nominated their cues more or less voluntarily, without much explanation. An example of such a concise nomination is the 'emotional' elicitation of the first three cues from a female patient who was living alone and had two adult sons.

R: *<u>Being healthy again</u>… and… <u>the children</u> (silence, crying)*
I: *Don't hurry… what's coming to the surface… worries about them?*
R: *Yes... (silence)*
I: *Do you want to tell me more about it… or… at this moment… You'd rather not?*
R: *No… (silence)*
I: *Anything else... you're thinking about as being important?*
R: *<u>Family</u> (coughing, clearing her throat)* [ID 03, F, age 64]

Nine patients were very eager to talk and presented narratives rather than cues. For example, the male patient (age 72) who started with describing his everyday life (see *initial remarks*) and then elaborated on his favourite operas etcetera.

R: *Quality of life…can't say that I appreciate much company… I must say… I never liked visitors… I've got my hobbies, my classical music and DVDs* […] *I like to watch TV... I' don't know if you want to hear this.. but, I like to watch the "National Geographic Channel", the "Discovery Channel" and "Animal Planet", which are all very interesting programmes* [ID 13, M, age 72]

The more elaborate the patient's narrative, the more "work" was needed to deduce cues from the patient's answer. In such situations the interviewer's contribution to the formulation of a cue was crucial.

Interviewer

Premature categorization

In our first (pilot) interview, the interviewer was focused by the instruction in the manual to elicit <u>areas</u> and started from the assumption that the patient would nominate a more or less abstract area. However, theory and practice differed.

R: *Areas... erm… you mean… ? If I say fatigue… or… what do you mean?*
I: *Well… fatigue…. Does that concern…. your… er... health?*
R: *Yes… but for me… I find it hard to describe… when I'm feeling miserable… I try to enjoy every day…* [..]
I: *Are there other things, which are important… which you need to have… ?*
R: *Yes, of course… everything that's around you, your family…*
I: *… your social relations... ?* [F, age 42]

This interview was analyzed and discussed intensively in the research team. There was agreement that, the interviewer, had reacted in an inappropriate way by immediately labeling the initial remarks *fatigue* and *family* in terms of cues from the prompt list: *health* and *social relations* respectively. A decision was made to avoid such premature categorizations and, in subsequent interviews, to accept the initial remark as cue label and to categorize afterwards in the office.

Accepting concrete aspects e.g. individuals

In subsequent interviews the patients talked in their own way about sensitive issues, and the interviewer responded and adapted the conversation accordingly, taking into account the seriousness of the situation. It was difficult for the interviewer to follow the instruction to "*Elicit areas… NOT individuals, e.g. marriage, not wife*" because of a tendency to name concrete aspects, e.g. individuals. Of twenty-six patients 20 were married. All but one of them mentioned individuals, i.e. *my wife* or *my husband* as being important. None of them mentioned *marriage* as a cue. In some cases patients combined their partner with children and/or grandchildren as *family*. *Family* was mentioned regularly in a context expressing meaning, for example "*the ability to function well in my family as wife and mother*", "*keep on thinking positively for my family*", "*receiving much loving support from my family*" and "*to sort things out with my wife*". Furthermore, patients distuinguished between close and not directly related family.

Probing, deducing and reorganizing cues

The meaning of each cue must be documented on the Cue Definitions Record Form. However, meaning was not expressed in words alone. It could be read between the lines, in the intonation, in the silence, the choice of specific words, etc. Patients were telling complete and different stories in nominating a cue. In general, the interviewer endeavored to establish the meaning of what actually was said, but the method differed depending on the style of patient's response. In patients who named their cues in short descriptions the interviewer probed for further explanation. Because all the interviews were audio-taped, it was not necessary to write down the meaning of cue labels in detail, which made the atmosphere more relaxed.

R: *To live a comfortable life… and… with enough money, that's important…*
I: *Two aspects at the same time… to live a comfortable life… enough money… ?*
R: *Yes, it has to be like that, because if you don't have enough money… you can't live a comfortable life… everything included…* [M, age 66]

In patients with a more elaborate response style the interviewer needed to unravel what mattered to the patients, i.e. to deduce cues from the answer. On occasion, aspects were reorganized such as combining two aspects that were mentioned into one cue.

R: *Yes… financially, I'm trying to look into the future, when I won't be here anymore. I'm busy thinking about her… will she able manage the finances… I need to know (silence).*
I: *Yes, well we talked about the various aspects, health… social relations… classical music…information on TV… being able to continue with your everyday life… we have five already… and at the same time you're saying… finances are important… ?*
If we put together classical music and information on TV… then there is room for finances as well… would that be a good idea… ?
R: *Yes…* [M, age 72]

Actually, together with the respondent the interviewer identified cues from the answers that were given, summarized and asked if any mistakes had been made, and documented cue labels in words chosen by the patient.

Introducing prompt list e.g. health

Thirteen patients were unable to choose 5 cues, therefore the interviewer had to make suggestions from the prompt list. While reading the list, patients weighted the pros and cons and aspects were recognized as important enough to mention in order to bring the total to 5. Provided cues turned out to be very important and/or self-evident, e.g. *health.*

Yes… health… on the top of the list… otherwise you can't enjoy other things [F, age 69]

Out of 18 patients who mentioned *health* as cue, 5 patients nominated *health* with help of the prompt list. Although they did not mention *health* voluntarily, three of them weighted *health* as first and most important. Despite the fact that all the patients were in the process of starting chemotherapy and knew about their poor prognosis, 8 out of 26 did not mention any cues related to *health*. Of these 8 patients 6 nominated their cues without use of the list and the other 2 were not prompted by the list to mention *health*. Although these 8 patients did not mention *health*, aspects related to *health* were self-evident, for example, as a goal to accomplish and/or in descriptions of their cues:

If I'll be cured.. we'll go to Curacao. I'll be brave enough to fly… [F, age 50]

The most important thing is my relationship with my wife… that we have sorted out everything, … when you are diagnosed with cancer… you might think you'll die soon… [M, age 64]

Discussion

Asking people to introspect their life in the context of being recently diagnosed with small-cell lung cancer seems to border closely on the invasive. However, many patients were eager to talk about sensitive issues, and to tell the interviewer what was important for their QoL in the format of five cues. The percentage of patients who were able to complete the SEIQoL interview is in accordance with findings from previous studies in other patient populations.[11;17-21] Reasons for non-completion are in line with findings from Mountain et al.[23], i.e. confusion, distress, fatigue and blindness.

The content and variety of cues was as expected from reports in the literature, with *family* as the most frequently mentioned aspect (96%). Twenty-five patients mentioned 38 cues in the area of *family*, which means that some patients mentioned more than one cue in the same area. Our findings, i.e. that patients named individual members and distinguished between partner, children, grandchildren and other family members were identical to the findings of Campbell & Whyte[21], and explain the nomination of more than one cue in the area of *family*.

The prompt list was needed for half of the patients, which is in contrast to other studies such as Frick *et al.*[28] and Campbell & Whyte.[21] Frick *et al.* reported no need at all (0%) in his research of 79 patients undergoing autologous peripheral blood stem cell transplantation. Campbell & Whyte reported that 14 out of 15 oncology patients (93%) needed the prompt list. The very disparate results between these two studies, as well as compared to our own, suggest that the elicitation of cues is interviewer dependent.
In our study, 2 patients completed the administration with less than five cues. In fact, the interviewer (MW) made a choice to accept less than 5 cues, rather than to elicit cues artificially, and the risk of choosing a "socially desirable" concept of QoL. Nominating less than 5 areas seems to be entirely consistent with what could be expected from people whose "horizons" are shrinking. The inability to nominate 5 cues, and the implicit assumption that QoL is reducible in this way, has also been reported by Macduff.[14] Campbell & Whyte argue that it may be unproductive to encourage the patient to choose a total of 5 cues.

Bowling[29] reports differences between voluntary cues and prompted cues. When respondents in her study selected their own cues from a show card, there were some discrepancies with their (office-coded) verbatim replies (which were recorded by the interviewer before the respondents saw the show card). This discrepancy illustrates a possible bias when prompting cues. In our study *health* was nominated as a cue in 5 patients with help from the prompt list. Interestingly, their weighting of *health* was high. It could be argued that the list helps people to nominate important cues, which are "forgotten" because they are self-evident. This finding raises the question of the 6 patients who nominated 5 cues without help of the list and did not mention *health* as cue. It can be assumed that some of these patients would reconsider their cues and nominate *health* after checking with the prompt list.

Although Bowling's results and the present findings suggest possible influences of bias when using the prompt list, it could be argued in favor of providing cues. The ability of patients to express themselves in an abstract concept of QoL differs, and important information may not be disclosed, valued and weighted if it is not explicitly asked for. Therefore, Wettergren *et al.*[30] used an extended Swedish version of the SEIQoL-DW with a disease-specific module to identify specific issues. More research is needed in order to investigate differences in nominating cues with and without the use of the prompt list.

Our findings show that cues were not just the result of a process of elicitation and subsequent recording. Cues originate from the patient-interviewer interaction, which is best described as an area of tension between the patient's answers and the instrument's instructions, in which the interviewer is trying to balance freedom and control. Eliciting cues implies making decisions about: 1) what to accept as a cue, 2) when to probe for further clarification, 3) which cue to deduce from the answer, 4) when and how to reorganize cues, 5) whether and when to use the promptlist, 6) whether and when to accept less than five cues, 7) when to discontinue further elicitation, 8) how to label and record the meaning of cues and 9) when and how to categorize. It is conceivable, however, that SEIQoL-DW data are vulnerable to unnoticed bias, because of intra- and interinconsistencies in the behavior of the interviewer(s).

Meaning

The quality of SEIQoL-DW is not only to measure QOL, but also to grasp "what really matters to patients". The methodology entails discussing the answers given by respondents in order to make them more concrete, or if needed, less abstract. The aim is to have a good understanding of the meaning of each cue, but without accurate recording this meaning disappears as soon as categorization takes place. It could be argued that in research with a global index score as main outcome the recording of this meaning is of less importance. However, in longitudinal and response shift research, as well as in the clinical setting, where the instrument is used therapeutically, meaning is crucial to reliable categorization and correct interpretation of cues and scores. After all, if *family* can connote for example in the first interview 'staying positive for my *family*' and in the second 'I'm happy with the support of my *family*', then these cues refer to the same area of *family*, but the outcome data has to be interpreted differently. In other words, cues categorized in the same area may have different meanings and cues categorized in different areas may have a similar meaning. Therefore, the question how to record arises. Although it is possible to express meaning in a short description of the label on the Cue Definitions Record Form, it could be questioned whether this can be done accurately and consistently over time and across interviewers.

Conclusion

Since the introduction of the SEIQoL-DW, recommended by Hickey et al.[11] as relatively simple to administer and requiring minimal training in the form of the manual, many studies have reported on the acceptability of the instrument. Our study shows that cues originate from patient-interviewer interaction which is best described as an area of tension between the patient's answers and the instrument instructions. The instructions on how to elicit these cues are not described in sufficient detail in the manual. In order to prevent possible unnoticed interviewer bias in the elicitation of valid and reliable cues special attention should be paid to the interviewer behavior. If correct interpretation of cues and scores is crucial, methods to record the meaning should be considered. More research is needed in order to investigate possible differences in nominating cues with and without the use of the prompt list.

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Change in what matters to palliative patients: Eliciting information about adaptation with SEIQoL-DW

5

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Accepted for publication in Palliative Medicine

Abstract

This study was carried out to investigate the usefulness of the SEIQoL-DW to elicit information about response shifts in palliative patients. The instrument measures individual quality of life and allows respondents to choose, rate and weight important areas of life (cues). We explored patients' reconceptualizations (i.e. change in cues) and their value change (i.e. change of cues weights). Results of twenty-one patients showed what mattered to these patients and how they had adjusted to deteriorating health. There is a risk that repeated measurements do not to provide all the information which is in potential present and relevant to explore response shifts. But clear instructions to interviewers, such as careful listening, probing self-evident cues such as *health* and *family*, and accurate recording of cues on the forms may overcome this risk. Future research is recommended to explore the possibilities of regular assessments to facilitate better adjustment of patients.

Introduction

Quality of life (QoL) is considered an important goal in clinical practice and particularly relevant to palliative medicine.[1-3] However, the expected deterioration in QoL often does not occur, even in cases of serious illness.[4-6] There is ample evidence that the occurrence of response shift complicates "objective" evaluation of treatment and quality of care.[7-10] Alternatively, response shift might be seen as a desirable outcome of adaptation. From both perspectives it is necessary to find out how response shift could be investigated.[11]

Schwartz and Sprangers defined response shift as a change in the meaning of one's self-evaluation of a target construct as a result of a) a change in internal standards of measurement, b) a change in values or c) a redefinition of the target construct (i.e. reconceptualization). They evaluated different approaches to the measurement of response shift.[11] Two categories of approaches appeared to be immediately applicable, because they allow the use of existing QoL instruments: 1) design approaches, e.g "then-test"[12], and 2) repeated use of individualised methods.[13]

The Schedule for the Evaluation of Individual Quality of Life – Direct Weighting (SEIQoL-DW)[14] is such an individualized approach which measures the unique individual perspective on QoL. Patients can choose, rate and weight five areas (cues) that they consider important. O'Boyle *et al.* [13] defined change in cues between first and second interview as reconceptualization, and change in the weighting of identical cues as value change. Two studies used the instrument explicitly to identify reconceptualization and value change. In the study of Echteld *et al.* SEIQoL-DW was assessed in patients admitted to units for terminal care at one, three, and five weeks after admission, to determine the extent to which response shift influenced QoL.[15] Sharpe *et al.* investigated the relationship between response shift and adjustment in patients with metastatic cancer patients at baseline, and three and six months later.[16] However, both studies differ considerably in the level of abstraction of nominated cues. Scharpe *et al.*, for example, reported family without further definition, whereas Echteld *et al.* reported family but defined as maintaining good contacts with family. In a previous study, we investigated the way in which patients choose and define their cues[17], and we found that complete stories are told in the elicitation procedure and that the interviewer makes decisions what to write down on the form as label and definition. Therefore, we questioned whether the measurement of response shift could be prone to error when the meaning of cues (i.e. patients' story told in the elicitation) is not taken into account.

It has been suggested that the SEIQoL-DW is useful for eliciting information about adaptation (i.e. response shift). It has not previously been investigated whether the data of repeated measurements are unambiguous and provide all the relevant information that is needed to determine whether response shift has occurred. We did so in a population of small-cell lung cancer (SCLC) patients during palliative chemotherapy.

Methods

Participants and procedure

Newly-diagnosed SCLC patients, who were evaluated for first-line chemotherapy, were recruited from five outpatient clinics for chest diseases in the Netherlands. Participating patients gave written consent and were interviewed during the treatment trajectory. The project was approved by the Medical Ethics Committee of the VU University Medical Centre. The first interview (T1) was held at the start of chemotherapy, the second (T2) 4 weeks later, the third (T3) after completion of the chemotherapy course, and the fourth (T4) six weeks later. The SEIQoL-DW was assessed, as described in the manual. Assessments were audio-taped and fully transcribed verbatim.

SEIQoL-DW

The SEIQoL-DW is administered in a standardized interview format.[14;18;19] First, five areas of life (cues) that are considered central to the individual's QoL are elicited by asking: *"What are the five most important areas of your life at present… the things which makes your life a relatively happy or sad one at the moment… the things that you feel determine the quality of your life…?"*. If cues are not nominated spontaneously, a list of nine frequently mentioned cues can be proposed[15]. Secondly, the patients' perceived level of functioning within each cue is recorded, using vertical visual analogue scales ranging from *"As bad as could possibly be"* (value 0) to *"As good as could possibly be"* (value 100). Thirdly, the patients are invited to rate their current overall QoL on a horizontal VAS scale with the same anchors as the vertical scales (value 100). Finally, the relative importance (i.e. weight) of each cue is recorded using a disk with five coloured sections, representing each elicited cue. Respondents are asked to change the sizes of the coloured sections by rotating labels attached to the sections until they correspond with the perceived weight of the cues. A total weight score of 100 is distributed over the five cues. An overall score (SEIQoL-index, range 0-100) is then calculated: the sum of cue levels multiplied by cue weights, and divided by 100.

Analysis of response shift

The findings reported in this article are based on two types of data: 1) the Cue Definitions Records Form (form) with written information (i.e. cues, definition, weights, Index-score and notes about procedure), and 2) transcripts of the audio-tapes, which were analysed in Kwalitan 5.0, a software package for ordering qualitative data. Reconceptualization and value change was investigated in each patient (case) per transition, i.e. determining changes in cues and their weighting between T1 and T2, between T2 and T3, and between T3 and T4. Reconceptualization was determined by comparing cue labels and their definition (patient's own words) as written on the forms. These findings were compared with information from the transcripts to determine whether the identification of cue change was correct. Analyses were performed by two members of the team (MW, TH). They categorized per transition each instance of change in cues and each instance of no change in cues. They compared and critically discussed their findings in meetings and consensus was reached on the following categories:

1. Change in cues ⇒ supported by transcripts ⇒ reconceptualization
2. Change in cues ⇒ not supported by transcripts ⇒ false positive reconceptualization
3. No change in cues ⇒ supported by transcripts ⇒ no reconceptualization
4. No change in cues ⇒ transcripts indicate reconceptualization ⇒ false negative reconceptualization.

If no reconceptualization was found, a change in the weighting of identical cues was determined as value change if there was a minimum difference in weight of 10 points.

Results

Study sample

During the course of the study, 41 patients were reported to the interviewer and invited to participate. However, 4 patients were unwilling to participate and 6 were not interviewed because of imminent death. Of the 31 patients who were interviewed, 6 patients were excluded from analysis because they were only interviewed once (they died within a month after T1) and 4 due to incomplete datasets (e.g. confusion, distress, fatigue), resulting in a study population of 21. One patient was only interviewed twice at T1 and T3 and died before T4. Three patients were interviewed three times and died before T4, resulting in a total of 58 transitions in 79 interviews to explore for response shift. The mean age of the 21 participants was 58 years (range 39 – 72), 12 (57%) were female, 18 (86%) were married and 16 (76%) had children**.**

SEIQoL-DW during the treatment trajectory

The frequency of elicited cues (*n*=372) and the mean SEIQoL Index and VAS are presented in Table 1. Cues related to *family* (e.g., partner, children, grandchildren) were nominated most frequently. *Health* was the second most frequently mentioned cue (definition: e.g. hoping chemotherapy would not to be too tiring, being cured, successful chemotherapy, feeling well under the circumstances, hoping that the tumour would not reoccur). Other cues concerned *hobby/leisure* (definition: e.g. fishing, gardening, making puppets), *social life/other relations* (definition: e.g. visiting friends, distant family), *enjoying life/holiday, work, living conditions, autonomy/independence, attitudes towards life,* and *finance.*

Table 1 SEIQoL index, VAS scores and frequency of important life areas (cues) elicited at T1 (start first-line chemotherapy), T2 (four weeks later), T3 (end chemotherapy) and T4 (six weeks later) in small cell lung cancer patients.

	Means (range 1-100)			
Quality of Life	T1 (*n*=21)	T2 (*n*=21)	T3 (*n*=20)[a]	T4 (*n*=17)[b]
SEIQoL-Index	70	83	77	82
SEIQoL VAS	70	75	81	78
	Frequency (%)			
Cue categories	T1	T2	T3	T4
Family	34	34	33	32
Health	16	17	16	23
Hobby/leisure	14	10	12	10
Enjoying life/holiday	7	8	11	13
Social life/other relations	7	10	11	9
Living conditions	6	6	3	2
Work	5	5	4	6
Autonomy/independence	5	5	5	1
Attitudes towards life	3	3	3	3
Finance	3	2	2	1

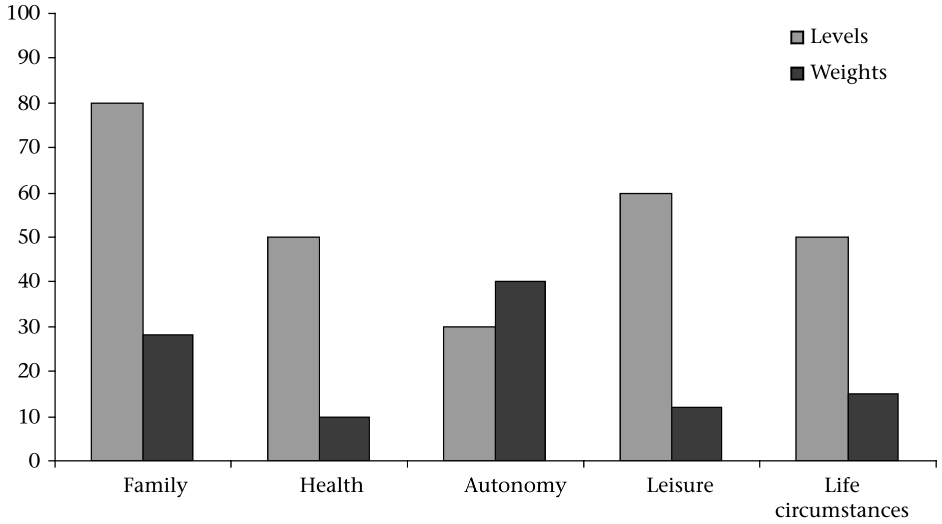
Note: [a] and [b] patients died before (*n*=1) and after (*n*=3) the end of first-line chemotherapy

In 18 out of the 21 patients change in the nominated cues was observed, and mostly concerned a change of 1 to 3 cues. Some cues were important at one specific moment in time only. For example, finances nominated at T1 was not mentioned again at T2 because *"I'm no longer worrying about my husband's finances after my death"*. There were two exceptions: 1) one patient changed all 5 cues at each interview; her cues were very concrete wishes or goals, which differed at each assessment (e.g. *wisdom to accept the situation, strengthening the relationship with my son, searching for new goals in life*), and 2) one patient nominated only two cues in every interview and did not change these cues and their weights (i.e. *relations* (level 100, weight 50) and *my independence* (level 100, weight 50).

In 16 out of the 79 interviews the prompt list was used. At T1, 10 patients nominated 2-3 cues with help of the list. At T2, 5 of them needed to be prompted again, but at T3, only one of these 5 still needed the list. At T4 the list was not needed anymore, but two patients who had been prompted before, asked the interviewer to write down their previously nominated cues again because "nothing has changed". The time taken to complete the SEIQoL-DW ranged between 10 and 30 minutes.

A case study

The data as shown in Figure 1a and 1b are illustrative of the information obtained from repeated measurements with SEIQoL-DW. Patient John (age 57) was not feeling well and suffered, among other things, for dyspnoea at the first interview (Figure 1a).

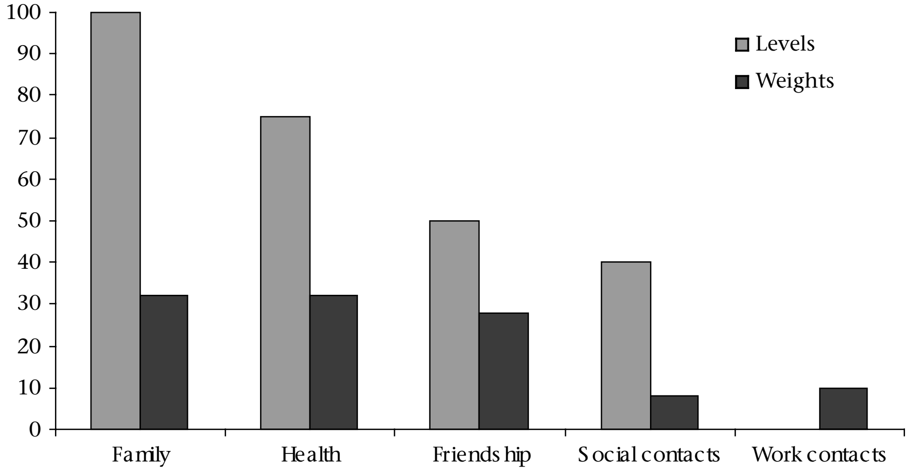


John's cues 10 days after diagnosis: SEIQoL index score 54

Figure 1a. Cues, levels of functioning and cue weights for a 57-year-old male patient with extended small-cell lung cancer, obtained 10 days after diagnosis at the start of 1stline chemotherapy. Family and health were nominated spontaneously; autonomy, leisure and life circumstances were prompted with the list of most frequently mentioned areas.

Spontaneously, he nominated the cues *autonomy* (definition:I don't want to become dependent on others) and *family* (definition: my wife and children, I'm not important but they are).Three cues were prompted with the list, i.e. *life circumstances*, *health*, *leisure* (definition: fishing with friends). The level of *autonomy* further illustrates his worries about dependency. His second interview was conducted after his last treatment cycle. He was feeling very well and eager to talk (Figure 1b). Without help of the list, he nominated *family* (definition: my wife and children, I'm happy with their support), *health* (definition: feeling well), and three new cues *friendship*, *social contacts* and *work contacts*. The low level of *work contacts* illustrates his disappointment that his colleagues had not contacted him. Weights show *family* as the most, *health* as the second and *social contacts* as the least important. Five weeks later he died.

John's SEIQoL -DW data provide information about how he adapted during his treatment trajectory. However, further inspection of the transcripts showed more information, and valuable in understanding how he had adapted. He had changed his definition of *family*. In his first interview he had given a further definition of family, which was not written on the form, while he was rating the level of functioning: "*I'm worrying about whether everything has been arranged properly for my wife after my death, I have to arrange what's necessary*". This definition differed from the one at the second interview and written on the form "*my wife and children, I'm happy with their support*".



John's cues 5 weeks before his death: SEIQoL index score 69

Figure 1b. Cues, levels of functioning and cue weights for a 57-year-old male patient with extended small-cell lung cancer, obtained one week after the last treatment cycle of 1stline chemotherapy and 5 weeks before death. Cues were nominated spontaneously.

Transcripts showed that the experience of feeling supported was new and special for him, and that he trusted his family with the necessary arrangements. Therefore, we concluded that a reconceptualization had occurred: the cue *family* had shifted from "caring for his wife and children" to "feeling supported by his family". After further reading of the transcripts, the same phenomenon was seen with the cue *health*, which had shifted from "getting cured" (transcript: *"that's what we fight for"*) to "feeling well". Both shifts in the meaning of cues were not detected by comparing the forms. Therefore, we categorized these instances of response shift as false negative reconceptualization because the SEIQoL had not detected these reconceptualizations. Furthermore, the transcripts showed a value change: *"I've changed my mind. I've said… health was not important for me but, now I'm feeling well I've experienced how health strongly affects my life and, although not the most important thing, it is certainly the most decisive"*. Because different cues were nominated and the definition of health had shifted, we concluded that nothing could be said about the magnitude of this value change.

Reconceptualization

We analyzed 58 transitions in the way we did John's data (see Figure 2). Reconceptualization was observed in 25 (43%) transitions and no reconceptualization in 16 transitions (28%). In 11 transitions (19%), no change in cues was seen but transcripts indicated that a reconceptualization had occurred (false negative reconceptualization). These reconceptualizations concerned the cues *health* (7 times), *family* (2 times), *leisure* (1 time) and *work* (1 time). In 6 transitions (10%) a conclusion about reconceptualization had to be withdrawn (false positive reconceptualization, i.e. change in cues not supported by transcripts) because differences in cues were a result of different wording, recording and/or response style (examples of false positive reconceptualization are not reported, but are available from the first author).

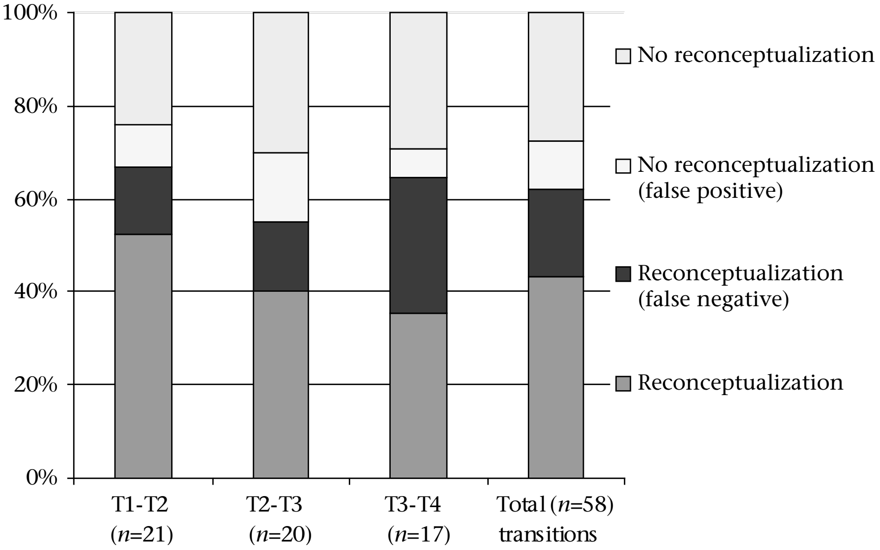


Figure 2. Measurement of reconceptualization with SEIQoL-DW in small-cell lung cancer patients during 1st line chemotherapy. Percentage of patients showing 1) reconceptualization, 2) reconceptualization not measured (false negative), 3) incorrectly measured reconceptualization (false positive) and 4) no reconceptualization.

Value change

In 12 patients (57%) a value change of more than 10 points was observed in at least one transition during treatment. Six patients weighted their *health* as more important at the end of the treatment with a mean change of 28 (range 10-64), and 3 patients weighted it as less important, with a mean change of 18 (range 12-27). *Family* was weighted as more important by two patients (change from 22 to 75, and from 20 to 33). One patient's weighting of *work* fluctuated during the treatment from 10 at the start of her chemotherapy (T1) to 3 four weeks later (T2). After the treatment (T3 and T4) she had plans to start with her work again and gave the area the weight of 16.

Discussion

Individual QoL appeared to improve remarkably within a 4-weeks period from 70 to 83 at T2 and was higher than the SEIQoL-DW scores reported by Waldron *et al.*[20] in advanced cancer patients and even in healthy elderly patients.[21] After a slight decrease at T3, levels of T2 were reached again six weeks after the end of chemotherapy. Results of other studies in SCLC patients confirm this pattern.[23;24]

Repeated measurements with the SEIQoL were generating data which showed immediately why "expected deterioration in QoL doesn't occur". [4] Respondents changed their focus, and emphasized more positive aspects, contributing to Qol in stead of focusing on problems and concerns.[24] The experienced support of the family and the knowledge that necessary issues such as finances had been arranged contributed positively to their QoL. These data showed how patients reconceptualized by nominating other cues that were important to them. The example of patient John showed that, solely by asking at two different points in time "*What are the five most important areas of your life at present… the things which makes your life a relatively happy or sad one at the moment",* it is possible to obtain information about a patient's adaptation.

Although not all reconceptualization was detected by comparing the two forms, the instrument had the potential to elicit important changes in priorities (i.e. the nomination of other cues), as well as changes in perspective (i.e. change within a cue). Shifts in the perception of *health,* in particualar, explained the high levels of functioning in this area and their contribution to good overall QoL. The audio-tapes of the interviews made it possible to assess these kind of reconceptualizations.

In 6 cases the nomination of other cues was not a real response shift (i.e. false positive reconceptualization). This result confirms Westerman's *et al.* suggestions about dependency of the instrument on differences in the elicitation and recording of cues.[17] Although standardization of cue elicitation (e.g. standard use of list) could reduce this problem, it may increase the number of unmeasured (i.e. false negative) reconceptualizations, because it might prevent patients from talking freely about issues that are important to them[15;25] and valuable information about adaptation could be lost.

Figure 2 shows a greater amount of false negative reconceptualization at the T3-T4 transition, compared to T1-T2 and T2-T3. A possible explanation for this might be that at T4 all cues were spontaneously nominated, and because the patients were more experienced the cues were nominated (and recorded on the form) more or less as a matter of routine.

Because SEIQoL generated relative cues that are constrained to unity[11], the measurement of value change has its shortcomings. Only when no change at all occurs in the nomination of cues, the value changes are clear. The nomination of just one new cue might also change the weight of the other 4 cues. In this study we chose to investigate the prevalence and quality of value change and not to measure the quantity of change. However, reconceptualization and value change are actually two interwoven concepts in SEIQoL-DW measurements. Although the nomination of a new cue could be considered as reconceptualiation, it could also be considered as a value change or reprioritization (e.g. nomination of *holiday* instead of *finances* suggests a value change of the cue *finances* to zero). The listing of cues in order of priority might overcome the earlier mentioned problems in measuring value change (see e.g. Sharpe *et al.*[16]). Furthermore, ranking opens up the possibility for the interviewer to ask at T2 for a ranking including the cues that were mentioned at T1, but not mentioned at T2.

Conclusions and recommendations

The SEIQoL -DW was found to be useful in exploring response shifts. But, there is a risk that repeated measurements do not to provide all the relevant information which is necessary to determine whether a response shift has occurred. Audio-taping was beneficial to us in detecting these pitfalls. However, it is not necessary to audiotape each assessment. It is recommended to listen carefully, to probe in particular the cues which are self-evident (*health* and *family*), to record the meaning of cues accurately, and to give clear instructions when other interviewers are involved in the assessments.

In the assessment of SEIQoL-DW valuable information was elicited about what really mattered to patients in the face of adversity, and whether response shifts had occurred. Whereas several studies show that response shifts are associated with favourable QoL, the question that arises is whether response shifts might be induced in patients who have trouble in adjusting to changing health. Especially when a cure is unlikely, clinicians are just as concerned with changes in how a patient feels as with the more strictly physical aspects of the patient's medical condition. An important objective of therapy may be to facilitate changes in the way particular states are experienced. Wilson[26] showed that, from a clinical perspective, response shift is not a new phenomenon, and that facilitating coping processes in ways that improve QoL is part of clinical care. Therefore, our study justifies future research to investigate the possibilities of regular SEIQoL assessments in clinical practice to find out: 1) whether and how patients are able to adjust to changing health, and 2) whether the obtained results can assist clinicians in helping patients to understand, to cope and to rethink and reframe their experiences so that they can make the best of their condition.[26]

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part 3

Understand the struggle

6

The struggle behind "I'm all right": Adaptation in cancer patients

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Submitted

Abstract

Objective

To investigate the factors that result in positive self-reports, despite deteriorating health in small-cell lung cancer patients.

Design

Prospective longitudinal multiple case study with home interviews.

Setting

Five out-patient clinics for chest diseases in the Netherlands.

Participants

31 newly-diagnosed small-cell lung cancer patients who were evaluated for first line chemotherapy.

Results

Patients were firstly and predominantly struggling to manage the side-effects of chemotherapy, to think positively and to fulfil their duties as a family member. Their struggle to maintain a sense of control over their life went hand in hand with giving up goals and expectations which were no longer important or tenable. Despite deteriorating health, most patients managed to keep going and still enjoy life by living it day by day and treasuring the good moments.

Conclusion

Acknowledgement of the struggle behind positive self-reports may assist physicians in their approach to patients with a terminal illness, and to discuss hope as well as end-of- life issues.

Introduction

Many patients with incurable cancer are willing to undergo risky and toxic treatments which can have an impact on their quality of life (QoL).[1-4] Since QoL is considered to be a necessary end-point in palliative medicine, it seems plausible to presume that doctors are happy when patients report that they are "all right". However, there is an awareness that the patients'own evaluations may differ considerably from those made by clinicians and significant others.[5;6] Actually, QoL research often yields counter-intuitive findings. Patients with a life-threatening disease have been found to report a stable QoL, and the QoL of patients with a severe chronic illness was found to be no worse or no better than that of less severely ill patients or healthy people.[7-10] It is suggested that patients make the best of their condition by coping, rethinking and reframing their experiences, and that this adaptive self-regulation may explain these discrepancies.[11;12]

We investigated aspects which contribute to the fact that patients are able to say "I'm doing all right" from the perspective of the terminally ill patient, despite deteriorating health. This topic is important because positive self-reports affect the decision-making concerning treatment and the discussion of end-of life issues.

Methods

Our study was based on observations made by AT (second author), who had carrried out an ethnografic study among small-cell lung cancer patients focusing on the role of collusion in the doctor-patient relationship contributing to "false optimism about recovery".[13] During observations and interviews in the university hospital it became apparent that these patients who were treated with chemotherapy did not report the deterioration in QoL which, objectively seen, would be expected, and that they reported less distress than they actually experienced. We designed a qualitative exploratory longitudinal multiple case study among others to investigate factors that contribute to positive self-reports of well-being and QoL. We draw comfort from the fact that AT had discovered in these patients a common illness trajectory consisting of five stages: i.e. 1) existential crisis after diagnosis, 2) focus on therapy during the initial treatment period, 3) relative peace of mind after treatment, 4) another existential crisis when a recurrence of the cancer was diagnosed, and 5) the final crisis when the patients knew that no further treatment with chemotherapy was available or feasible.

Data were collected through home-interviews and at equivalent points during first-line chemotherapy: at the start, 4 weeks later, after the last cycle and 6 weeks after completing the (first) course. Furthermore, at the start and the end of further chemotherapeutic treatment. Each interview consisted of two consecutive parts. Firstly, QoL was assessed by means of two measurements (the results will be reported elsewhere) Secondly, the interviewer (MW, first author) encouraged the patients to talk freely about their experiences with the treatment, and about "what matters most" with regard to their life and its quality. After each interview MW made field notes about her observations and the informal conversations after the interview. The interviews were audio-taped and transcribed verbatim.

After approval by the Medical Ethics Committee of the research site and the participating hospitals, recruitment commenced in five outpatient clinics for chest diseases in the Netherlands. Forty-one consecutive patients, diagnosed with small-cell lung cancer (limited and extensive disease) and evaluated for first-line chemotherapy, were approached between March 2001 and September 2003. Seven felt unable to participate, and three died after giving written consent. The age of the patients who actually participated (*n*=31) ranged from 39 to 82 years, and 15 (48%) were female. The majority were living with a spouse (25, 81%) and had children (26, 84%). However, 8 patients died during the first course of treatment and 2 immediately afterwards. Six weeks after the course (T4) only 21 patients were still alive and could be interviewed. Six of these patients were interviewed again (in January 2004 data-collection had to be closed), and of these 6, 2 died during the second course and 3 died shortly afterwards. One female patient (age 42) was interviewed again at the start and the end of a third course.

The findings are based on the analysis of three types of data. Firstly, transcripts of 112 formal interviews. Secondly, field notes with a detailed description of the observations and informal conversations with patients and spouses. Thirdly, the log book in which telephone calls were recorded when appointments for interviews were made. The data were analysed per patient, resulting in 31 case studies.[14] The aim of each case-analysis was to investigate adjustment in patients with incurable cancer and the factors that result in positive self-reports, despite deteriorating health. Kwalitan 5.0 was used as software package for the qualitative analysis of the transcripts (www.kwalon.net). After individual case-analyses were completed, similarities and differences between cases were analysed. The analyses were performed by MW and AT, and there were critical discussions about the results with the other members of the research team.

Results

Management of expectations

The patients were informed about the risks and side-effects of chemotherapy and the fact that the treatment would probably exhaust them or make them feel more ill than they already were. However, the expectations differed from their experiences (see Box 1). After the first treatment cycle, most patients were relieved to have suffered less than they had expected. Some patients with extensive cancer were even feeling psychically better than before because of an alleviation of their symptoms (e.g. dyspnoea and coughing). This first experience with the chemotherapy, seemed to lift the patients out of their existential crisis and to give them back a sense of control. This sense of control was enhanced by the 3-weekly routine of check-ups and treatment, with a predictable number of bad days, days of recovery, and days to pick up their normal life again. But, like an uphill struggle, the impact of every new cycle was experienced as more and more severe, and most patients had some very difficult days with considerable suffering caused by the treatment. Six weeks after the course, fatigue and a decline in functioning was obvious, but for most patients it was a relief to have endured the treatment and to be feeling much better than immediately after the course: *"You really feel that you´re quite somebody again, compared with just after the chemo. Of course I´m not as good as before, but sometimes you think that it´s all a mistake, that you haven´t got cancer because you feel so good. But, yes, you are terminally ill and every day can be your last"* [female, age 69].

Expectations and experiences

Not as ill as expected
The doctor said ´you must have chemotherapy, as soon as possible´. So then I started, but I must say that it hasn´t really made me feel ill yet.
[male, age 66, second interview]

Well, they say now that I´m in between the tenth and the fifteenth day, and then I should really be very tired, but I still go shopping in the mornings.
[female, age 56, second interview]

I´ve really been very lucky that the chemo has hardly bothered me at all.
[male, age 65, third interview]

Feeling better all the time
People think it´s strange that you say I don´t feel any different. You hear all these stories about the chemo that it makes you feel really ill. Of course it´s not nice, but it wasn´t too bad and actually I feel better after every course.
[male, age 67, third interview]

Then I heard that I had cancer and, you see, it´s incurable … Yes, then you start thinking about what will happen … it will only get worse … but with me it´s really only got better. So, actually, it was exactly the opposite with me.
[male, age 39, third interview]

Becoming very ill
The last chemo made me very ill. I was so ill that I thought I wasn´t going to make it. I did nothing but vomit and vomit, and on top of that I also got the ´flu. Never again, I thought!
[female, age 44, third interview]

Well, then it begins in the evening – sick, oh terribly. I can´t even tell you how awful. I sometimes wished I was dead. So ill. You can´t tell any man that … sweating and soaking wet right down to my toes.
[male, age 73, third interview]

Box 1

Managing prognosis

Adopting a positive attitude and presenting one selves as “doing all right” seemed to function as an anchor for the patients in the struggle of shifting back and forth between awareness, acceptance and denial of their prognosis. Fifteen patients (out of the 21 who were still alive after the first course) said that they had regained control over their life by forcing themselves to think positively (see Box 2). They tried to put the issue of prognosis out of their mind by suppressing thoughts that arose and by focusing on the positive effects of the treatment. Feeling better than they had expected together with the good news that the tumor was disappearing, strengthened them in coping this way. A positive attitude was perceived as a necessary condition to keep going and to have a life at all. The patients, and especially those with young children, also felt the need to be positive in order not to be a burden on their family.

An act of willpower

If you have negative thoughts yourself, you will only have negative people around you. You just have to think that there´s a life in spite of the lung cancer, it mustn´t rule my life.
[female, age 47, first interview]

Sometimes I can see that I´m not here any longer. My husband here alone. What will happen then? And then I think, no, stop! I´m still here, and we´ve not got that far yet. I´m here now.
[female, age 56, first interview]

I turn the tables and say, ´don´t be so stupid, we´re not going to go on like this, we´re going to fight´, that´s my only chance of survival.
[female, age 51, second interview]

As long as I feel all right I´m able to forget it. I have to, because otherwise I can´t go on.
[male, age 69, second interview]

There´s a big chance that it will come back again. If you keep thinking about that, then you don´t have a life anymore.
[female, age 42, third interview)

Box 2

After the course, i.e. “the period of relative peace of mind”, the patients started a new routine of regular check-ups at the clinic. These visits were welcomed, but feared at the same time. In some patients, fear of a recurrence was so strong that they actually wished to continue with chemotherapy. Others were very doubtful as to whether they would wish to receive more chemotherapy in case of a recurrence. However, when a relapse did occur, all accepted a new course, hoping to gain more time. Amy (age 42) even insisted on having a third course, although she knew that it was pointless: *“I’ll fight till the bitter end, that’s my way”*.

Commitment to the family

Family was perceived to be the most important factor contributing to QOL, but at the same time, especially in the initial period, it was also a focus of concern (see Box 3). All patients worried about their loved ones, and made a special effort to fulfil their established role in the family. Men first checked and discussed finances and living conditions, and tried to do whatever was in their power to support and secure future family life. For example, a recently retired man exchanged his cherished car for a smaller one, which was more convenient for his wife; a young father arranged for a house in a safer area of the town for his family. Women were especially concerned with the continuation of family life and the household in general. Although they had to delegate tasks in the early stage of their illness, as soon as they could they tried to carry out their former tasks again and to restore the normal family life.

However, during the course of the treatment family concerns abated. Patients who had perceived themselves primarily as the provider of support learned to accept and enjoy the support that was given to them. In some cases, this shift from concern to enjoyment seemed to be confusing for their spouses, who actually felt quite alone in their worries, and were anxious to discuss the observed physical deterioration. They did not want to destroy the hope by reacting negatively to remarks such as “*I’m all right and I’m happy*”.

Family

You have to cheer your family up yourself. If I were to cry all day, that would not be good for them. Besides, I still want to experience so many things. That keeps me strong. I really wouldn’t like think about the fact that someone else would have to bring up my children, and furthermore, we would also have a financial problem. There are times when my wife and I discuss things like this.
[male, age 46]

I’ve suffered a lot, especially in the week after the first treatment. I was so tired … my husband had to do everything … cooking. I hope that it will never happen again. I don’t like it at all. I want to do it myself. Ofcourse he did it with love, but even then …. The second treatment was less of a problem. I was able to do just what I wanted to do. As long as I can, I want to do it all by myself.
[female, age 63]

We’ve already arranged a lot. We’ve also taken a family photo with the children. But my husband is still very quiet, he doesn’t talk about it, I can’t reach him, and that’s hard for me. But, I think that’s his way of coping, so I let it go, I find that’s the best way. I’m feeling tense towards him and the children, that’s why I want us all to enjoy ourselves together. Just a shortbreak, together in a holiday home, that would be nice and relaxing.
[female, age 53]

Box 3

The awareness of imminent death could cause a thorough shake-up of relationships. Lisa (age 64) for example, had taken care of her sick husband for seven years, and had expected more support now that she herself was ill. She complained: *"chemotherapy to prolong my burdensome life is quite useless"*. When Lisa's last treatment cycle was cancelled, because the tumor was not responding, the whole family suddenly took notice: her children and grandchildren visited her on a more regular basis and, for the first time, she felt supported by her children. She did not complain anymore and created her own precious moments. Despite her husband's disabilities, she picked up her life again and together they spent many happy times touring in the countryside which they both enjoyed.

Here-and-now: an art of living

During the first course of chemotherapy
You don't think about next year, what you're going … you're enjoying the daffodils that are coming up. Although I don't feel well, I still try to enjoy things.
[female, age 42]

You can't forget, the cancer is there everyday. But I don't do things just because I don't have time left, that's not the way to cope with the limited time. You have to think…If I honestly want to do something … then just do it.
[male, age 68]

After the first course
I usually finished the work in the garden in one day. Now it's different, I rest more, your body is warning you. I've no trouble at all in splitting the work up in more days. What I can't do today I'll do tomorrow.
[male, age 68]

My life seemed to be over when I heard the diagnosis. But after a while, suddenly, I could cope with it, it doesn't matter anymore. We can accept what comes, we're enjoying being together as long as possible.
[male, age 69]

After the second course
I never knew that I was capable of it, and that it was so easy to live life day-by-day.
[male, age 57]

I don't worry anymore, that's all over now. I wake up, it's 6 o'clock, the sun is rising and I think to myself…a new day is coming for me to enjoy … how it will be tonight? We'll see that when the time comes.
[male, age 66]

Box 4

Living in the here-and-now

Regardless of how persistently and emphatically the patients stated that they were "doing all right", the fabric of their positive stories was interwoven with threads of imminent death. Their seemingly random remarks masked their existential concerns and pain of loss. For example, Mark (age 39) said that he was very happy: "*My friend and I bought a boat last week, and we've already had a marvellous trip. Actually I can't* *afford a boat on my salary, but you can't wait till you're retired, can you?"*. Although he was happy and enjoying the moment, he had given up his his life-plan and his remark also expressed this loss. Living in the here-and-now (see Box 4) and enjoying the precious moments was advocated and aimed for, but not easily mastered. Ella (age 47), for example, suffered a lot from nausea during the treatment and protested strongly when this advice was given to her: *"how can it ever be possible to enjoy life when you know that you will die, you feel miserable because of the chemo and you know that you won't be able to see your son growing up"*. Retired patients seemed to master the art of living in the moment more easily: *"we'll all die in the end, whether it's from cancer or from something else. I will die, but not now, and meanwhile I'm going to enjoy the extra time"*. Younger patients, who realized that they had to give up their work, hobbies and goals, such as seeing their children get married and becoming a grandparent, tried to make the best of their situation by redefining what really mattered to them and by changing situational goals. Amy, for example, kept in touch with colleagues and supported them, and changed her hobby of gardening into one of making dolls, which were sold to raise money for local charities. In the last months of her life, walking became difficult and she had to use a wheelchair when leaving the house. Altough her health was severely deteriorating after her three courses of chemo, she still felt that she had a high quality of life: *"I'm still me you know, fighting the cancer, but also enjoying my life. Actually, I think my quality of life is hundred percent. I've nothing to worry about, I enjoy the visits of colleagues, the weekend trips with my husband, the contacts with my children, and I don't feel any pressure…"*

Discussion

An important finding of our study is that the positive image which small-cell lung cancer patients might present to their doctors and relatives is neither just the result of their own evaluation of health, nor just the result of succesful adaptation and "practicing the art of living".[15] Patients seem to be continiously busy maintaining control by focusing on treatment, by anchoring themselves to a positive attitude, and by continuing their commitment to the family. Our results suggest that positive self-reports seem to show that patients are able to survive emotionally. However, it could also be argued that the expression "I'm all right" in itself is a useful strategy which enables patients to protect themselves from their own and people's negative thoughts. Especially in the initial phase of treatment, when patients force themselves to stay positive, this way of presenting themselves might be a useful coping strategy in maintaining a sense of control in a situation which cannot be changed.[16] Our findings also suggest that patients actually know their prognosis, but do not want to think about it and therefore try to focus on positive aspects. Therefore, self-presentation might also partially explain the "false optimism about recovery" in these patients, as described by The *et al.*[13]

Thompson *et al.* found that patients who perceive that they have control over their health are more capable of adjusting to illness.[17] Their results indicate that even patients who were physically or psychosocially worse off were better adjusted if they had higher perceptions of control. Our findings suggest that especially through this sense of control patients are able to let go, to revise goals and expectations, and thus to enjoy life, despite their deteriorating health.[11;18;19] The realistic appraisal of changing circumstances generates a positive affect, and is in line with the revised model of the coping theory[20] in which positive mood and allocating meaning to a stressful situation has been integrated.[21] But, it has to be remembered that the art of living with a terminal illness sometimes requires a daily need for willpower from the patients, and presumably also from their loved ones.

Conclusion

The dual role which physicians have in caring for terminally ill patients is not an easy one. Not all patients are prepared to discuss reality and their imminent death. However, the aknowledgement of a positive self-report as a product of the struggle for emotional survival, might open up the doctor-patient communication about end-of life issues without negating the importance of fostering hope. Explicitly acknowledging the patients' efforts to maintain a sense of control can be a useful tool in the communication with patients who do not easily talk about their concerns,[21-23] and it may promote satisfactory adjustment and quality of life.

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7

General discussion

Discussion

The objective of our study was to obtain more insight into response shift in QoL measurement. During the study it was found that other phenomena were also important in explaining counter-intuitive findings, so we decided to investigate these phenomena as well. In the first part of this chapter we will discuss the main results. In the second part we will answer the research questions listed in the General Introduction and describe the implications for research on quality of life (QoL) and response shift.

Over the past decades, there has been a growing interest in the impact of disease and treatment on a patient's life and functioning. Systematic evaluations of QoL may enable clinicians not only to choose more easily between different treatment options, but also to identify patients who are at risk. For example, QoL was found to be a significant predictor of survival in lung cancer patients.[1] However, when patients are confronted with a life-threatening or chronic disease, they are faced with the necessity to adapt themselves to their deteriorating health. Response shift is considered to be an important mediator in this adaptation process.[2] However, response shift has been found to complicate the interpretation of QoL outcomes. Therefore, integrating response shift into health-related QoL research would enhance the sensitivity and relevance of the research.[3] But the question that arose was: how?

Response shift

When our study was designed (1999), Sprangers and Schwartz had just developed their theoretical model.[2] We therefore aimed to obtain more insight into response shift in QoL measurement by investigating whether the methods suggested by Schwartz and Sprangers[3] were useful for the measurement of the three different aspects of response shift. However, soon after the initial analyses we found that 'measuring' the different aspects by means of the then-test and the SEIQoL-DW was much more complicated than we had expected (Chapter 2 and 5). The problems with the then-test in measuring recalibration were partly due to our longitudinal study design, which made it difficult for patients to remember the previous assessment. The problem of recall bias had been already discussed by Schwartz *et al.*[4] We assumed that a design in which the previous assessment is easier to remember, for example before and after a surgical operation, will produce less recall bias. But even so, it is debateable whether the then-test is applicable in combination with a questionnaire such as the EORTC QLQ-C30. By observing the response behaviour in more depth we found that even when there is no recall problem, most of the questions about functioning in particular, are multi-interpretable and therefore cannot be compared with the then-test questions which evaluate a patient's functioning during the previous assessment (Chapter 3). From a theoretical point of view, the then-test could be questioned as well. An assessment of fatigue or pain here and now, when a patient can still feel the fatigue or pain, or when the patient is feeling happy to have recovered from the last cycle of treatment, is totally different from remembering a certain moment in the past and recalling the feelings of that moment.

In the 'measurement' of response shift with the SEIQoL-DW we were confronted with the fact that the instrument has its risks which threaten its reliability (Chapter 4). Preventive measures to overcome the risk of unnoticed interviewer bias cannot guarantee that in repeated measurements all the response shifts that occur are measured (Chapter 5). The advantage of using this instrument is that in the semi-structured interview the interviewer is able to elicit important information about what matters to the patient. However, this information must be recorded accurately. Because we audio-taped the interview, we were able to investigate response shifts, and this was furthermore productive in explaining the counter-intuitive QoL outcomes. However, audio-taping and analysing the verbatim of each assessment in order to investigate response shifts is time-consuming. Before a decision is made to use the SEIQoL-DW to measure response shift, researchers must think about 1) how to prevent interviewer bias, 2) how to record what the patients say when choosing the cues, and 3) how and when to use the prompt list. However, we see the possibilities of using the instrument in order to find out what really matters to patients and to identify patients who have trouble in adjusting to their deteriorating health in a palliative setting.

Change in appraisal

Our in-depth study of the response behaviour of patients in the assessment of QoL revealed phenomena which could explain counter-intuitive findings in QoL (Chapter 2 and 3). In addition to self-presentation, which phenomenon we will discuss later, we found that QoL evaluations cannot be interpreted without considering the evaluation process. Our findings are most in line with the Rapkin and Schwartz process model for the evaluation of QoL, which has recently been published.[5;6] Four appraisal processes are distinguished: 1) Induction of a frame of reference (referring to kinds of experiences that a person deems relevant), 2) recall and sampling of salient experiences, 3) standards of comparison used to appraise experiences, and 4) the subjective algorithm used to prioritize and combine appraisals to arrive at a QoL rating. Schwartz and Rapkin recommend the use of this model to investigate the cognitive processes underlying the evaluation of QoL. The model can easily explain that a change in appraisal is possible at each assessment and that, in addition to response shift, this change may explain counter-intuitive findings. Using their model in designing a new study might provide us with more insight into the evaluation process. However, asking patients to analyse each step of their evaluation is actually giving them the opportunity to formulate a different answer from that which they would have given without analysis of their evaluation in the proposed four steps, and this does not resemble 'normal' response behaviour. Furthermore, it could be questioned whether the 'why' question can be answered. Why are patients behave the way they do when answering the questionnaire? Why do we find discrepancies between answers to the questionnaire and the information given in the informal interview (Chapter 2)?

Think aloud method

In answering the "why' question, we found he think aloud method useful. One advantage of this method is that it closely resembles the normal process of evaluation, because observation has priority over the cognitive interview. Actually, the way in which we used the think aloud method was similar to a participant observation study in the measurement of QoL. With participant observation, phenomena are observed in their natural setting, which not only increases the validity of the data[7], but also enables the researcher to uncover routines (i.e. response behaviour) of which participants themselves may be unaware.[8] However, the think aloud method also has it limitations. The participants are conscious of the researcher's presence and may therefore be tempted to adapt their response behaviour (social desirability). Furthermore, thinking aloud while filling in a questionnaire is not a 'normal' activity. However, by interviewing the patients with SCLC several times, as we did, it is likely that they got used to the interviewer (MW) and the think aloud method, and probably reverted to their normal behaviour (if it had changed at all). What we did find was that some patients said that they were more motivated to fill in the questionnaire because: 1) someone was listening, 2) they had the opportunity to explain nuances in their answers, and 3) they felt that they were able to explain more clearly how they felt. Furthermore, the results of other studies of SCLC patients[9] confirm the pattern we found in the QoL outcomes of both the EORTC and the SEIQoL-DW (Chapter 3 and 5): improvement at four weeks after the start of the chemotherapy, a slight decrease at the end, and an increase in the level of QoL six weeks after the chemotherapy.Taking this all into account, it is not likely that we have observed adapted behaviour which would have resulted in different QoL outcomes than we would have found without using the think aloud method.

Self-presentation

The think aloud method uncovered response behaviour, of which the patients themselves might have been unaware. They tried to distance themselves from the image of the stereotype cancer patient, and presented themselves as not suffering and as accepting fatigue as a consequence of treatment and not only when filling in the questionnaire (Chapter 2). Even in the informal part of the interview they presented themselves as positive and not suffering, but at the same time they also talked about their sometimes daily struggle to cope with the diagnosis and treatment (Chapter 6). Especially during the first course of chemotherapy the patients tend to anchor themselves to a positive attitude, and herewith to distance themselves from the prognosis. These findings seem to be in line with the results of the study in which The *et al.* investigated the 'false optimism' about recovery in SCLC patients.[10] However, taking into consideration the fact that the patients made preparations for their imminent death, and said that they knew that their cancer was incurable, it could be questioned whether the term 'false optimism' is accurate. Our study (Chapter 2) suggests that 'self-presentation' might be an important mechanism which explains the optimism and the positive reports in QoL measurement. Self-presentation has been found to be used by patients as coping strategy which helps them to keep control over their life (Chapter 6). The patients tried to live as normally as possible, and to adopt and maintain a positive attitude. This was sometimes a daily struggle, but one which they considered to be necessary. They did not give up goals which were no longer tenable from one day to the other; control and adjustment went hand-in-hand. Feeling physically well seemed to be helpful in maintaining a positive attitude, but this does not necessarily mean that the patients did not know about their prognosis. They knew, but did not want to know, and tried to enjoy life as much as possible by living it as normally as possible. When we argue that self-presentation is a coping strategy (Chapter 2), it would actually fit into the theoretical model of response shift as one of the mechanisms.[2] In an attempt to specify this type of coping it seems to resemble a combination of 'redefining' and 'confronting', or in other words 'accept but find something favourable' and 'take firm action based on present understanding', respectively, as described by Weisman in his book "Coping with cancer".[11] In Weisman's words: "The effect of redefining is to put a bad situation into a better, more benign, and acceptable light one, which must not be mistaken for denial. This reinterpretation is an excellent way of rationalizing illness into smaller complaints". Weisman presents an example of a women who also minimized her complaints by blaming them on something other than her illness (see our Chapter 2)[12], and he argues that this coping strategy works best when combined with confronting. Without it, patients are reduced to avoidance and passivity. In our study we also observed the firm action patients take to keep control over their life. Although self-presentation, as coping mechanism, seems to fit into the model of the response shift theory, the results of our study have also shown that self-presentation is not identical to recalibration response shift, and complicates the investigation of response shift in a yet unknown way.

Answers to the research questions

Q 1. *Is the then-test a useful approach to determine whether a recalibration response shift has occurred?*

In the qualitative investigation of the the response behaviour in the measurement of fatigue with the EORTC QLQ-C30 question 'were you tired', we found indications of recalibration response shift (e.g. using a different standard for comparison over time). However, it was difficult to demonstrate recalibration by means of the then-test (Chapter 2). The patients had difficulty in remembering either the previous measurement point and/or their fatigue at that time. Identical problems were encountered in the physical and role functioning questions (1-7). In addition to recall bias, change in the way which patients sampled their experiences when interpreting and responding to the questions made the interpretation of the then-test data complicated. The then-test, in combination with the EORTC questionnaire, is not a reliable method of demonstrating recalibration during a treatment trajectory.

Q 2. *Can response shift sufficiently explain counter-intuitive findings in the measurement of QoL?*

There were discrepancies between the answers of 15 out of the 23 patients to the EORTC question 'were you tired' and the level of fatigue they spontaneously reported during the interview (Chapter 2). Although response shift occurred (i.e. a different standard of comparison over time, and a change towards a more optimistic perspective), this type of response shift was not only found in the discrepancy group. Patients in the discrepancy group reported spontaneously how they dealt with the diagnosis and treatment (i.e. by adopting protective and assertive behaviour and by fighting the stigma). They distanced themselves from the image of the stereotype cancer patient and presented themselves as not suffering and accepting fatigue as a consequence of the treatment. Our study suggests that 'self-presentation' might be an important additional (coping) mechanism that affects QoL measurement and can explain counter-intuitive findings. Furthermore, the patients did not report the deterioration in physical and role functioning that would objectively have been expected (Chapter 3). The various response strategies (e.g. taking the wording of questions literally, guessing, and excluding activities which could not be performed) and the change in the patients' appraisal process explained how levels of physical and role functioning were sustained under worsening physical conditions. In addition to response shift, self-presentation and change in the patients' appraisal process were found to be additional mechanisms explaining counter-intuitive findings in QoL measurement.

Q 3. *Is SEIQoL-DW a reliable instrument in providing all the relevant information that is needed to determine whether a response shift (i.e. reconceptualization and change in values) has occurred?*

The SEIQoL-DW is an individualized approach to the measurement of QoL in which patients choose and define their own so-called 'cues'. Our study showed not only the usefulness of the SEIQoL-DW in eliciting what matters to patients, but also the problems that are encountered when using this instrument (Chapter 4). Because cues originate from patient-interviewer interaction, the interviewer may inadvertently introduce bias while attempting to elicit cues, ultimately affecting the patients' SEIQoL-DW score**.** There is a risk that repeated measurements do not provide all the information that is potentially available and relevant for investigating response shifts (i.e. reconceptualization and change in values)(Chapter 5).

Q 4. *What are the factors that result in positive self-reports despite deteriorating health.*

Adaptation through the eyes of the SCLC patient is sometimes a daily struggle (Chapter 6). Although these patients make preparations for their imminent death, they tend to anchor themselves to a positive attitude, and herewith to distance themselves from their prognosis. They try keep control over their life by living it as normally as possible, by letting go those expectations and goals which are no longer tenable, and by simply enjoying life. Feeling physically well seemed to be helpful in maintaining a positive attitude, which was perceived to be 'a necessary condition to have a life at all', and resulted in positive self-reports despite deteriorating health.

Implications for research on QoL and response shift

Despite our study, the answer to the question of how to integrate response shift in QoL research is still not clear. But, by lifting the lid of the black box just a little while by listening to the patient who is filling in the questionnaire we have learnt more about how SCLC patients cope with their illness and how this coping affects the QoL measurement. An important issue which, to our knowledge, has never been discussed is the fact that when we present a patient with a cancer-specific QoL questionnaire, what we are communicating is that the patients has cancer. The patient probably feels stigmatized, and tries to distance him/herself from the image of the stereotype cancer patient. We have also learnt that it is possible to focus on what really matters in the palliative treatment of SCLC patients, and that the SEIQoL-DW is a useful instrument in communicating this type of information. But, above all, we know now that despite high levels of QoL, these patients have a lot of work to do, and we have to acknowledge their struggle behind words such as "I'm feeling fine". More research is certainly necessary and useful. However, we have to be aware that QoL outcomes may mainly reflect how patients are coping, in stead of measuring only the actual impact of cancer treatment.

Final recommendations

1. Future research is needed to investigate the pros and cons of using of the prompt list in the SEIQoL-DW assessment
2. We recommend further research on the possibilities of regular SEIQoL assessments in clinical practice to investigate: 1) whether and how patients are able to adjust to changing health, and 2) whether the obtained results can assist clinicians in identifying patients who need help in adjusting to their changing health.
3. Because each illness trajectory has its own characteristics, we recommend qualitative research to investigate background knowledge about the illness trajectory and appraisal processes. This is not only relevant for the design of quantitative studies, but also for the interpretation of QoL outcomes.
4. It is important to investigate in more depth the phenomenon of self-presentation in relation to QoL measurement and its place in the model of response shift theory.

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Summary

Quality of life (QoL) is considered to be an important goal in clinical practice and particularly relevant to palliative medicine. However, there is an awareness that the patients' own evaluations may differ considerably from those made by clinicians and significant others. Actually, QoL research often yields counter-intuitive findings. The expected deterioration in QoL often does not occur, even in the case of serious illness. It is suggested that the occurrence of response shift (i.e. a change in the meaning of one's self-evaluation as a result of a) a change in internal standards, b) a change in values or c) a reconceptualization) complicates the measurement of QoL, and may explain these discrepancies. Alternatively, response shift might be seen as a desirable outcome of adaptation. The aim of this thesis was to investigate: 1) the measurement of response shifts, 2) other phenomena to account for counter-intuitive findings in QoL measurement, and 3) adaptation from the perspective of the patient.

In this study the following research questions were addressed (Chapter 1):

1. Is the then-test a useful approach to determine whether a recalibration response shift has occurred?
2. Can response shift sufficiently explain counter-intuitive findings in the measurement of QoL?
3. Is SEIQoL-DW a reliable instrument in providing all the relevant information that is needed to determine whether a response shift (i.e. reconceptualization and change in values) has occurred?
4. What are the factors that result in positive self-reports despite deteriorating health.

We followed 31 patients with small-cell lung cancer during their illness trajectory. Patients who were evaluated for 1st line chemotherapy were informed about the study, invited to participate, and asked to give written consent. The patients were interviewed personally at home, they filled in written questionnaires (EORTC QLQ-C30 and QLQ-LC13) according to a 'think aloud' and a 'then-test'protocol, and individual QoL was assessed by means of the SEIQoL-DW. Our qualitative study included 4 measurements points during the first course of chemotherapy (i.e. T1 at the start of the chemotherapy; T2 4 weeks after T1; T3 at the end of the chemotherapy; and T4 6 weeks after the end of the chemotherapy), and 2 measurements during a second or third course (i.e. at the start and at the end of these courses, respectively).

Listen to the patient

Chapter 2 reports on the response behaviour in the measurement of fatigue with the EORTC QLQ-C30 question 'were you tired'. For 15 out of 23 patients there were discrepancies between their answer to the EORTC question 'were you tired' and the level of fatigue they spontaneously reported during the interview. These patients chose the response options 'not at all' or 'a little', and explained their answers in various ways. Patients in the discrepancy group reported spontaneously how they dealt with the diagnosis and treatment. They distanced themselves from the image of the stereotype cancer patient presenting themselves as not suffering and accepting fatigue as a consequence of the treatment. The results of our study show that the question 'were you tired' in the EORTC-QLQ-C30 does not unequivocally measure the impact of chemotherapy on the energy level of a patient. Patients seem to give the impression that they are managing the situation, and this self-presentation may explain unexpected results. In patients with and without discrepancies, we found indications of recalibration response shift (i.e. using a different standard of comparison over time) and change in perspective (i.e. change towards a more optimistic perspective). However, it was difficult to demonstrate recalibration by means of the then-test. The patients had difficulty in remembering either the previous measurement point and/or their fatigue at that time.

Chapter 3 describes the response behaviour in the measurement of physical and role functioning. The terminally ill patients evaluated their functioning in terms of what they perceived to be normal under the circumstances. Various response strategies when answering questions about problems and limitations in functioning explained why the patients had scores suggesting that QoL was affected very little by the chemotherapy. For example, the patients focused on one aspect of the question, took the wording of the question literally, ignored or excluded certain activities which they could not perform, and guessed their level of functioning in activities which they did not perform. These strategies gave the impression that the patients were less limited than they actually were. Their answers could be interpreted best in terms of response behaviour, which has been described by Rapkin & Schwartz, and the change in their appraisal of QOL explained how levels of physical and role functioning were sustained under deteriorating physical health.

Focus on what matters

Chapters 4 and 5 address the usefulness of the SEIQoL-DW (i.e. an individualized approach in the measurement of QoL in which patients can choose, value and weight five areas that they consider important for their quality of life) in the measurement of response shift. Chapter 4 reports on how patients choose and define the five areas, the so-called 'cues' and describes the problems that were encountered in the elicitation of cues. The instrument was found to be useful in eliciting what mattered to the patients. Many patients were eager to talk about sensitive issues. *Family* was the most frequently mentioned cue (i.e. patients named their partner, children, grandchildren and other family members) and some patients nominated more than one cue in this area. However, our study also demonstrated that eliciting cues implies that the interviewer makes decisions (e.g. about what to accept as a cue, whether and when to use the prompt list, and how to label and record the meaning of cues). SEIQoL-DW data are therefore vulnerable for unnoticed bias, because of intra- and inter-inconsistencies in the behaviour of the interviewer(s).

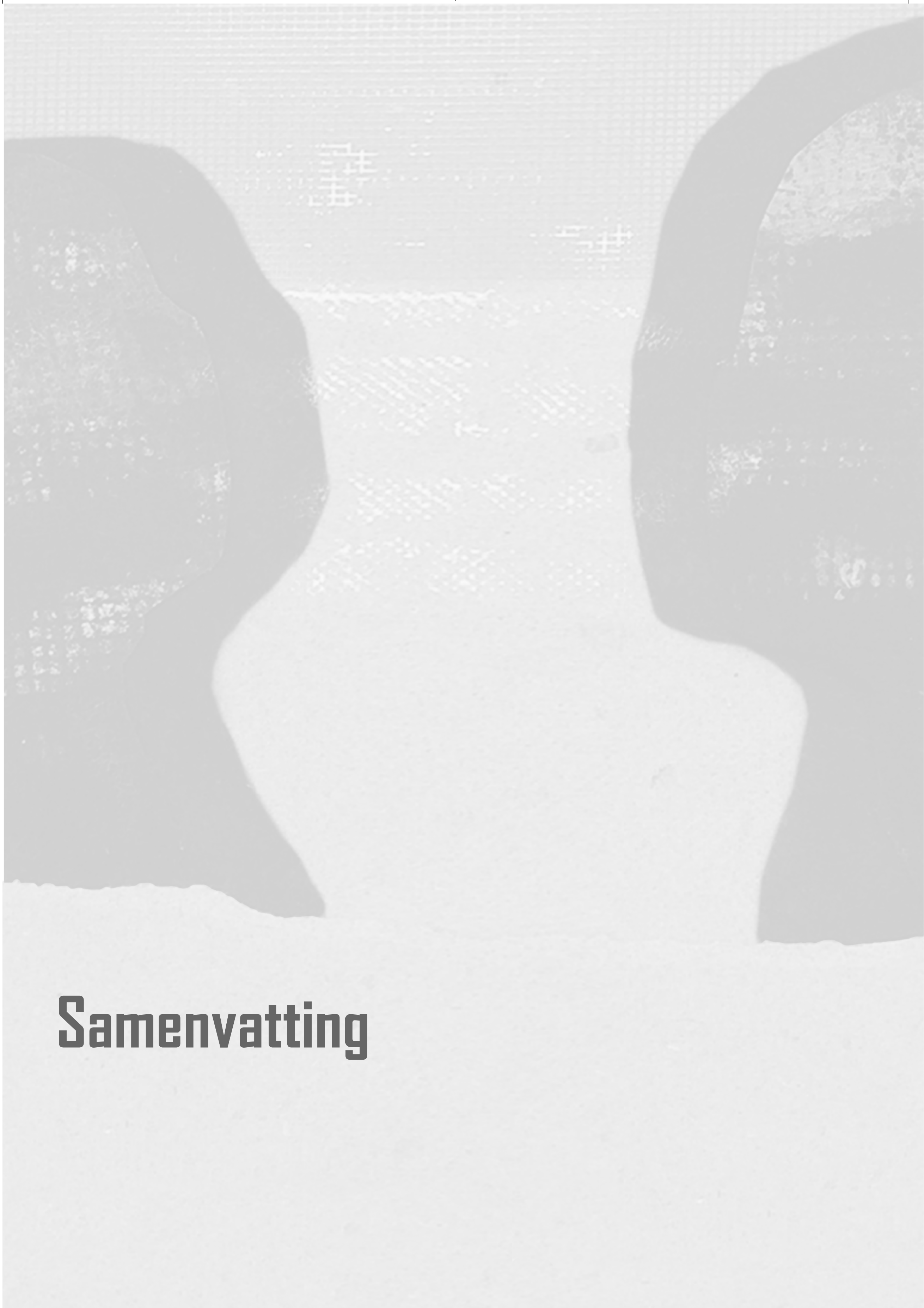
Chapter 5 describes the measurement of response shift with the SEIQoL-DW. Repeated measurements with the SEIQoL-DW generated data which showed that the instrument had the potential to elicit important changes in priorities (i.e. the nomination of other cues), changes in perspective (i.e. change within a cue), and changes in values. Shifts in the perception of *health*, in particular, explained the high levels of functioning in this area and their contribution to good overall QoL. The audio-tapes of the interviews made it possible to assess these response shifts. There is a risk that repeated measurements do not provide all the information which is potentially available and relevant for the investigation of response shifts.

Understand the struggle

Chapter 6 describes adaptation through the eyes of the patient**.** The positive image which the patients presented was neither just the result of their own evaluation of health, nor just the result of successful adaptation and 'practicing the art of living'. The patients actually knew their prognosis, but did not want to think about it, and tried therefore to anchor themselves to a positive attitude in order to survive emotionally. Especially through this sense of control the patients were able to let go, to revise goals and expectations, and thus to enjoy life, despite their deteriorating health. But, the art of living with a terminal illness sometimes required a daily need for willpower from the patients, and presumably also from their loved ones.

Discussion

Chapter 7 contains a general discussion of the findings presented in this thesis, and addresses implications for QoL and response shift research. Firstly, by listening to the patient who is filling in the questionnaire we discovered that not only response shifts, but also that self-presentation affects the QoL measurement. It will be a challenge for QoL and response shift researchers to investige more in depth QoL appraisal, the phenomenon of self-presentation, and its place in the model of the response shift theory. Secondly, patients are eager to talk about sensitive issues in the SEIQoL-DW assessment, and the instrument is useful for investigating response shifts. We recommend that clear instructions are given to the interviewers, and accurate recording of the cues is essential to overcome the risk of interviewer bias. New research should be encouraged to investigate more in depth: 1) the use of the prompt list, and 2) the possibilities of using the instrument in clinical practice to identify patients who need help in adjusting to deteriorating health. Finally, our findings indicate that the positive self-reports in the measurement of QoL may cover up the efforts that patients have to make in order to survive physically, emotionally and socially, to keep going and still enjoy life. QoL and response shift research will benefit from investigating the struggle behind "I'm allright", by observing QoL measurement in the real life context during the illness trajectory. Because each illness trajectory has its own characteristics, we recommend qualitative research in order to enhance the interpretation of QOL outcomes.



Samenvatting

In de klinische praktijk en met name in de palliatieve geneeskunde wordt kwaliteit van leven als een belangrijke uitkomstmaat beschouwd. Toch is men er zich ook van bewust dat er aanzienlijke verschillen kunnen zijn tussen het oordeel met betrekking tot de kwaliteit van leven zoals een patient die zelf maakt, en het oordeel zoals artsen en partners die maken. In feite zien we in kwaliteit van leven onderzoek dikwijls uitkomsten die tegen de intuitie indruisen. De verwachte verslechtering in kwaliteit van leven wordt soms niet gezien, zelfs niet in het geval van een ernstige ziekte. Er wordt verondersteld dat het fenomeen *response shift* hierbij een rol speelt. *Response shift* wordt gedefinieerd als een verandering in de zelfrapportage als gevolg van: a) een verandering in interne standaarden (*recalibratie*), b) een verandering in waarden (*herprioritering*), of c) een verandering in de betekenis van kwaliteit van leven zelf (*reconceptualisatie*). *Response shifts* kunnen worden gezien als een gewenst resultaat van het vermogen van de mens om zich aan te passen aan veranderende omstandigheden om zo het leven draaglijk te maken. Echter, *response shift* bemoeilijkt het meten van kwaliteit van leven en het interpreteren van veranderingen hierin. Dit proefschrift beschrijft de resultaten van het onderzoek 'Response shift in kwaliteit van leven in de palliatieve behandeling van patienten met kleincellig long kanker' dat als doel had te exploreren: 1) het meten van response shifts, 2) andere fenomenen die de tegen-intuïtieve resultaten in kwaliteit van leven onderzoek zouden kunnen verklaren, en 3) aanpassing aan een veranderende gezondheidstoestand gezien vanuit het perspectief van de patient.

De volgende vragen zijn in dit onderzoek beantwoord (hoofdstuk 1):

1. Is de toen-test een bruikbare methode om vast te stellen of er een recalibratie response shift heeft plaatgevonden?
2. Kunnen de uitkomsten in kwaliteit van leven metingen die tegen de intuitie indruisen voldoende toereikend verklaard worden door response shift?
3. Is de SEIQoL-DW een voldoende betrouwbaar instrument in het verschaffen van de relevante informatie die nodig is om te bepalen of er response shift (i.e. reconceptualisatie en herprioritering) heeft plaats gevonden?
4. Welke factoren dragen bij aan de beleving van het hebben van een goede kwaliteit van leven ondanks een afnemende gezondheid?

We hebben 31 patienten met kleincellig long kanker tijdens hun ziekte traject gevolgd. Patienten die met een eerste lijn chemokuur zouden beginnen werden geïnformeerd over de studie en vervolgens uitgenodigd om deel te nemen en hun schriftelijke toestemming te geven. In onze kwalitatieve studie werden bij iedere patient 4 interviews rond de eerste chemokuur afgenomen (i.e. bij aanvang van de kuur, 4 weken later, na de laatste behandeling en 6 weeks na afloop van de kuur) en 2 interviews bij een eventuele volgende chemokuurkuur (i.e. bij aanvang en na afloop). De interviews werden bijna altijd bij de patient thuis gehouden. Ondere andere werden kwaliteit van leven vragenlijsten (EORTC QLQ-C30 and QLQ-LC13) ingevuld. Patienten werd gevraagd hierbij hardop te denken aan alles wat er tijdens de beantwoording van de vragen door hen heen ging. Daarnaast werd de individule kwaliteit van leven gemeten met behulp van de SEIQoL-DW. Vanaf het tweede interview werd bovendien een zogenaamde 'toen-test' afgenomen met gebruik van de EORTC vragenlijsten. Hierbij werd gevraagd om terug te denken naar het moment van het vorige interview en de vragen te beantwoorden voor dat 'toen' moment.

Luister naar de patiënt

Hoofstuk 2 gaat over hoe patiënten de vraag 'was u moe' van de EORTC QLQ-C30 vragenlijst beantwoorden. Bij 15 van de 23 patiënten vonden we stelselmatig tegenstrijdigheden tussen hun antwoord zoals gegeven bij het invullen van de vragenlijst en dat wat deze patiënten spontaan vertelden over hun vermoeidheid op een ander meer informeel moment in het zelfde interview. Deze patiënten kozen de antwoordcategorie 'helemaal niet' of 'een beetje' en legden op allerlei manieren uit waarom dit zo was. In deze groep van patiënten zagen we ook dat spontaan verteld werd hoe men om ging met de diagnose en de behandeling. Deze patiënten leken zich af te zetten tegen het beeld van de stereotype kankerpatiënt en zichzelf te presenteren als niet lijdend en de vermoeidheid accepterend als behorend bij de behandeling. De resultaten van onze studie laten zien dat de vraag 'was u moe' van de EORTC-QLQ-C30 vragenlijst niet een helder en duidelijk antwoord kan geven op de vraag wat de invloed van de chemokuur is met betrekking tot vemoeidheid. Patiënten lijken het beeld te willen afgeven dat ze in staat zijn om om te gaan met de situatie, en deze 'zelf-presentatie' zou mede kunnen verklaren waarom de resultaten van kwaliteit van leven onderzoek soms anders zijn als verwacht. We vonden indicaties van recalibratie response shift (i.e het gebruik van verschillende standaarden in de tijd) en een verandering van een pessimistisch in een meer optimistisch perspectief. Maar het was moeilijk om recalibratie daadwerkelijk aan te tonen met gebruik van de toen-test. Dit omdat de patiënten het niet alleen moeilijk vonden om zich het vorige interviewmoment te herinneren maar ook niet meer precies wisten hoe moe ze zich toen hadden gevoeld.

Hoofstuk 3 beschrijft het antwoordgedrag bij het meten van het fysieke en het rol functioneren. De ongeneeslijk zieke patiënten beoordeelden hun functioneren van uit hun idee van wat normaal was gezien de omstandigheden. De verschillende manieren waarop de patiënten de vragen beantwoordden over de ervaren problemen en beperkingen verklaarden waarom scoren kunnen suggereren dat de kwaliteit van leven weinig door de chemokuren is aangetast. De patiënten namen bijvoorbeeld de betekenis van woorden uit de vraag van de vragenlijst soms heel letterlijk, negeerden activiteiten die genoemd werden, namen activiteiten die ze niet meer konden doen niet meer mee in hun beoordeling, of probeerden te raden hoe beperkt ze waren in hun functioneren. Door op deze manieren de vragen te beantwoorden wekten de patiënten via hun scoren de indruk dat zij minder beperkt waren dan zij feitelijk waren. Hun antwoordgedrag past binnen het verklaringsmodel zoals beschreven door Rapkin & Schwartz. Door de verandering in hun 'appraisal' zijn de kwaliteit van leven uitkomsten beter te begrijpen en is het te verklaren hoe het komt dat volgens de scoren het fysieke en rol functioneren nauwelijks achteruitgaat of gelijk blijft ondanks de afnemende fysieke gezondheidstoestand.

Focus op wat er echt toe doet

In de Hoofdstukken 4 en 5 wordt het meten van response shift met de SEIQoL-DW beschreven. Dit instrument beoogt de individuele kwaliteit van leven te meten doordat de patiënt niet alleen zelf gebieden kan noemen die hij/zij van belang vindt bij de beoordeling van kwaliteit van leven, maar ook kan aangeven welke gebieden het meest van belang zijn. Hoofdstuk 4 laat zien hoe de vijf te kiezen gebieden, de zogenaamde 'cues' gekozen worden, en beschrijft de problemen tijdens het kiezen van deze cues. Door dit instrument te gebruiken bleek het mogelijk om in beperkte tijd belangrijke informatie te verkrijgen over wat er echt toe doet voor de individuele patiënt. *Familie* werd het meest genoemd , ze noemden hun man, vrouw, kinderen, kleinkinderen of andere familieleden. Sommigen noemden meer dan één cue die te maken had met familie. Onze studie heeft echter ook laten zien dat het kiezen van de cues impliceert dat de interviewer keuzes maakt tijdens dit proces (bijvoorbeeld wat te accepteren als cue, of en wanneer er hulp wordt geboden door middel van het aanbieden van een lijst met topics, de zogenaamde 'prompt' lijst, en hoe dat wat is gezegd van een label wordt voorzien en de betekenis daarvan wordt opgeschreven). De SEIQoL-DW blijkt gevoelig te zijn voor bias, omdat zowel inconsistenties in het gedrag van de interviewer zelf als ook tussen interviewers onopgemerkt kan blijven.

Het meten van response shift met de SEIQoL-DW komt aan bod in Hoofdstuk 5. Het herhaald afnemen van de SEIQoL-DW leverde data op die lieten zien dat het instrument het potentieel heeft om de response shifts vast te kunnen leggen. Door het noemen van andere cues en ook door een verandering in de betekenis en de weging van een cue werd zichtbaar hoe de patient zich had aangepast aan zijn veranderde omstandigheden. Met name een verandering in de betekenis van *gezondheid* kon de hoge kwaliteit van leven score verklaren. Deze response shifts konden we vooral goed onderzoeken omdat het gesprek tussen de interviewer en patiënt tijdens de afname van het instrument was opgenomen op de band. Als het gesprek en de cues met hun betekenis niet goed worden opgeschreven of op band wordt opgenomen bestaat er het risico dat informatie die relevant is om te bepalen of er response shifts zijn opgetreden wordt gemist.

Begrijp de strijd

Hoofdstuk 6 laat 'aanpassing' zien vanuit het perspectief van de patiënt. "I'm allright" of wel "het gaat goed met mij" is iets dat patiënten niet alleen kunnen zeggen omdat het goed met ze gaat, of omdat ze zich goed hebben aangepast, en zich de kunst eigen hebben gemaakt om te leven in het hier en nu. In feite waren ze op de hoogte van hun prognose, maar ze wilden er niet aan denken, en ze probeerden zich volledig te richten op het hebben van een positieve houding om emotineel te kunnen overleven. Door deze gerichtheid op het positieve en het uitspreken van "het gaat goed met mij" hield de patiënt een gevoel van controle en juist daardoor was hij/zij in staat om los te laten, doelen en verwachtingen bij te stellen en te genieten van het leven ondanks een afnemende gezondheid. Maar deze kunst om te leven met een ongeneeslijke ziekte vereiste ook wilskracht en ging soms gepaard met een dagelijkse strijd, niet alleen voor deze patiënten maar waarschijnlijk ook voor de mensen dicht om hen heen.

Discussie

Hoofstuk 7 bevat de algemene discussie met betrekking tot de resultaten zoals gepresenteerd in dit proefschrift en bespreekt de implicaties hiervan voor kwaliteit van leven en response shift onderzoek. Door allereerst te luisteren naar de patiënt die de vragenlijst invult ontdekten we dat niet alleen response shifts, maar ook zelf-presentatie van invloed is op de kwaliteit van leven meting. Allereerst zal het een uitdaging zijn voor kwaliteit van leven en response shift onderzoekers om niet alleen 'appraisal' verder te onderzoeken maar ook het fenomeen zelf-presentatie en haar plaats binnen de response shift theorie. Daarnaast is ook het meten van response shifts met de SEIQoL-DW interessant om verder te onderzoeken, juist omdat er zoveel informatie door de patient wordt gegeven over wat er (voor hem of haar) echt toe doet. We raden daarbij wel aan om de interviewers heldere instructies mee te geven om bias te voorkomen en ook de cues en hun betekenis goed vast te leggen. Verder onderzoek is nodig naar het gebruik van de 'prompt' lijst bij het kiezen van de cues en de mogelijkheden van gebruik van dit instrument in de klinische praktijk bij het signaleren van problemen in de aanpassing aan een afnemende gezondheid. Tot slot laten onze bevindingen zien dat positieve uitkomsten in kwaliteit van leven onderzoek vaak het werk versluieren dat deze patiënten moeten doen om überhaupt fysiek, emotioneel en sociaal op de been te blijven, om door te gaan met hun leven en daar ook nog van te genieten. Kwaliteit van leven en response shift onderzoek zal baat bij hebben het onderzoeken van de strijd achter "I'm allright", door de patiënt te observeren bij de kwaliteit van leven metingen gedurende het ziektetraject. Omdat ieder ziektetraject zijn eigen specifieke karakteristieken heeft kan kwalitatief onderzoek waardevol zijn om de kwaliteit van leven uitkomsten beter te kunnen interpreteren.

Dankwoord

Nabeschouwing

In dit proefschrift heb ik verslag gedaan van ons onderzoek naar de zogenaamde 'response shift in kwaliteit van leven' bij mensen die wisten dat hun longkanker niet meer te genezen was. Uit de gesprekken kwam naar voren dat 'aanpassen' aan het feit dat je een ongeneeslijke ziekte hebt… aan het feit dat je fysiek achteruitgaat… aan het feit dat je levensplan anders verloopt als verwacht… niet gaat zonder slag of stoot, ook al blijkt dit niet altijd af te lezen uit de kwaliteit van leven zoals gescoord op vragenlijsten. Omdat de vragenlijsten niet werden toegezonden per post, maar door mijzelf werden afgenomen bij de mensen thuis heb ik beter kunnen begrijpen wat het betekent om een ongeneeslijke ziekte te hebben en daarvoor chemokuren te ondergaan. Daarom ook "The struggle behind I'm allright" als titel van mijn proefschrift. Niet alleen uit respect en bewondering, maar bovenal ook uit dankbaarheid voor de openheid waarmee mensen met mij spraken en mij leerden over hun worsteling om er toch te zijn, er te zijn voor hun familie, er te zijn om te leven…in het hier en nu en dat in de wetenschap dat hun tijd beperkt was…zoals iemand zei: *"Het gaat goed met mij, maar het leven is transparant geworden, de dood is steeds op de achtergrond aanwezig".*

Ik heb het hele land afgereisd, heel wat kilometers gemaakt met mijn oude Volvo, de koffie stond altijd klaar en in Brabant soms met het burenbezoek erbij. Afspraken maken was niet altijd makkelijk. Is er ooit een goed moment om af te spreken voor een interview vlak na de diagnose kanker…als je hele leven overhoop ligt? Wat zeg je als je graag weer wilt afspreken voor een interview als het moment daarvoor nog onbekend is… als de kanker weer terug is gekomen… bij de start van een 2e kuur? Kan je… mag je dat eigenlijk wel vragen? Ook afscheid nemen hoorde er bij. Soms heel onverwacht, zoals bij een patiënt die ik bezocht 6 weken na de kuur voor het 4e interview. Hij lag op het moment van mijn aanbellen op sterven. Zijn vrouw had er niet meer aan gedacht dat ik zou komen. Op zo'n moment was ik blij dat ik niet meer zo jong was… dat ik geleerd had in mijn leven om niet te schrikken van mijn emoties en van die van de ander… dat ik kon zijn... in het hier… in het nu. Vaak stopte ik mijn auto na een interview op een rustig plekje… weg van de snelweg. Even alles laten bezinken, wat aantekeningen maken en alles los laten. Loslaten om weer thuis te kunnen komen en er ook daar weer te zijn. Niet alleen het leven van de patiënten en hun familie, ook mijn leven is in die tijd meer transparant geworden… geboorte, ziekte, dood … blijdschap en verdriet…. het was er allemaal… soms heel indringend… in één en hetzelfde moment…het moment van het leven zelf. Kwaliteit van leven hebben? Toen ik startte met het onderzoek heb ik eens gezegd dat kwaliteit van leven niet is af te meten aan alles wat in dat leven aanwezig is… aan wat je hebt, aan wat je fysiek kan etc.! De patiënten met wie ik heb mogen spreken hebben me laten ervaren dat kwaliteit van leven alles te maken heeft met *hoe* je er bent… met een kwaliteit van *zijn* die zich moeilijk laat meten.

Dankbetuiging

Promoveren doe je niet alleen en daarom een woord van dank aan allen die direct of indirect een belangrijke rol hebben gespeeld bij de totstandkoming van dit proefschrift. Allereerst nogmaals de patiënten die de tijd hebben genomen om met mij vragenlijsten in te vullen en met mij zo openhartig wilden spreken. Dank ook aan hun familie die vaak klaar stond om mij te ontvangen, om zich vervolgens weer discreet terug te trekken voor het interview begon. Ook dank aan de longartsen van de diverse ziekenhuizen die deelnamen in het onderzoek. Zij hebben zich enorm ingezet bij de inclusie van de patiënten. Het zal vast niet makkelijk zijn geweest om mensen die slecht nieuws te verwerken kregen te vragen om aan onderzoek deel te nemen.

Terugblikkend op de jaren waarin dit proefschrift vorm kreeg wil ik alle collega's van het EMGO en met name van de afdeling Sociale Geneeskunde van de VU bedanken voor de samenwerking, de vriendschappen en de gezellige momenten. Ook buiten de VU waren er mensen die mij stimuleerden en die er toe hebben bijgedragen dat dit proefschrift afkwam. Een aantal wil ik hier persoonlijk bedanken.

Allereerst Tony, mijn projectleider en co-promotor. Tony, je vroeg een schaap met vijf poten toen ik solliciteerde op deze 'baan'. Of ik dat ook voor je ben geweest? Ik hoor het graag nog eens van je. Je ging weg bij de VU, vertrok naar Rotterdam, toen ik net begon met het onderzoek. Toch was je er altijd als het nodig was en ik heb ongelofelijk veel van je mogen leren deze jaren.

Dan Anne-Mei The, mijn tweede co-promotor en waanemend projectleider. Door mijn verhalen over de patiënten met jou te delen, en dit soms onder het genot van een heerlijke lunch, groeide mijn inzicht. Jij hielp me altijd weer op weg en niet alleen met je grote vertrouwen in mij dat het wel zou lukken. Dank je daarvoor en voor de smsjes om mij te vragen hoe het ging, of om even in het weekend te melden dat je blij was met wat ik had geschreven. Ik hoop ook in de toekomst nog veel van je te leren en met je samen te werken.

Gerrit van der Wal, jij was mijn promotor gedurende het gehele traject, zelfs nu je alweer bijna een jaar een andere baan hebt. Ik dank je voor het vertrouwen dat je bleef houden in mij ondanks een fors uitlopend traject. Ook dank voor de snelle en adequate reacties en het opbouwend commentaar tot en met de komma's en puntjes op de i.

Als 4^{e} lid van de projectgroep wil ik ook Harry Groen bedanken. Harry, je stond aan de wieg van ons project, want je was er al bij betrokken nog voor je wist wie het onderzoek zou gaan uitvoeren. Uiteindelijk kwam de gehele groep maar zelden bij elkaar. Maar je kwam dan toch helemaal over uit Groningen. Vooral wil ik je bedanken voor de wijze waarop je me hebt 'ingewijd' in alles wat er rond de SCLC patiënt gebeurt in het ziekenhuis en op de polikliniek.

Mirjam Sprangers, je hoorde niet officieel tot onze projectgroep, maar was er wel op de achtergrond. Je adviezen, jouw kritisch commentaar, de wijze waarop je de tijd voor me nam die je eigenlijk niet had, een artikel met mij doorspreken per telefoon vlak voor je weer afreisde naar het hoge Noorden…ik ben er trots op dat jij met mij wilde meedenken. Bedankt!

Michael Echteld, jij leerde mij het gebruik van de SEIQoL-DW. Wat was ik blij dat je na een kort uitstapje (ook al naar Rotterdam) toch weer bij de VU kwam werken en mij wilde adviseren bij het schrijven van één van mijn artikelen.

Interviews moeten worden uitgetypt. Minou Adami, de meeste van de ruim honderd interviews zijn door jou uitgetypt. Ook Esther Marcus en Juun Vogel hebben heel wat banden afgeluisterd en uurtjes getypt. Bedankt jullie!

Artikelen die ingediend gaan worden bij een tijdschrift moeten ook nagekeken worden op correct gebruik van de Engelse taal. Faith Maddever, je was daarvoor onmisbaar!

Graag bedank ik ook de leden van de promotiecommissie: Prof.dr. N.K. Aaronson, Prof.dr. D.L. Willems, Prof.dr. P.E. Postmus, Dr. F. van Zuuren en Dr. M. Echteld.

De collega's van het secretariaat wil ik ook niet onvermeld laten. Rita, jij was in mijn begintijd als secretaresse van Gerrit altijd een eerste praatpaal en vraagbaak voor mij. Je betrokkenheid bij de AIO's vond ik bijzonder. Ik weet niet of ik nu de laatste AIO ben uit 'jouw tijd', maar ik weet wel zeker dat je je best zult doen om ook bij mijn verdediging te zijn. Manigeh en Inge, ook jullie bedankt voor de kleine klusjes en de adviezen!

Dan ook nog speciaal 3 collega's met wie het goed was om de kamer te delen en sloten koffie weg te werken. Annemieke, jij was al gepromoveerd, daarom konden we veel van je leren. Harm-Jan en Sander, wat konden jullie kletsen af en toe… en tafeltennissen omdat de computer aangaf dat het weer tijd was. Dank voor jullie steun!

Buiten de VU zijn er ook mensen om te bedanken, mensen die er altijd waren voor mij en mijn gezin. Ted, samen hebben we gezorgd voor een stevige basis, in mijn ogen onmisbaar bij het doen van dit soort van onderzoek. Rob, je woont soms te ver weg naar mijn zin, maar bedankt dat je mij weer het juiste perspectief kan laten zien als dat nodig is. Ria, niet alleen een heel bijzondere vriendin, maar ook steun en toeverlaat. Je weet zelf waarom jouw werk er toe heeft bijgedragen dat ik (weer even) zonder zorgen aan mijn proefschrift kon werken. Joep en Ineke, fijn om mensen in de buurt te hebben bij wie je bij nacht en ontij terecht kan.

Dan nog mijn familie, mijn ouders en in het bijzonder mijn kinderen. Jullie hebben nauwelijks kunnen bevatten waar ik allemaal mee bezig was en al evenmin waarom het nog steeds maar niet klaar was. Dat je om te kunnen promoveren erg veel doorzettingsvermogen nodig hebt en heel hard moet werken alvorens de klus te kunnen klaren is jullie nu wel duidelijk. Jan en Bep, het is fijn om te voelen dat je ouders zo met je meeleven! Jan, Harm en Lise, fantastisch om zulke zelfstandige kinderen te hebben die er mee konden leven dat ik al heel vroeg in de ochtend vertrok naar de VU en soms ook weer veel te laat in de avond thuis kwam. Dat al het aanwezige lekkers ondertussen soms op raadselachtige manier uit huis verdween… dat waren vast de kaboutertjes… want over jullie had ik niets te klagen. Eindexamen doen, overgaan, alles ging thuis gewoon door en dat zonder al te veel hulp van mijn kant! Beatrijs en Gijs (en natuurlijk ook Peter en Diana), jullie zijn er altijd voor mij en voor Jan, Harm en Lise als dat nodig is. Ik ben heel blij en trots dat jullie mijn paranimfen zullen zijn.

Als laatste… als eerste ben jij al genoemd… Frans! Jij was er altijd… jij bent er nu… zonder jou was er geen proefschrift. Jij en ik kennen de strijd en het werk dat nodig is om in alle eerlijkheid en met respect voor elkaar afscheid te kunnen nemen. Het is het mooiste geschenk dat we elkaar nog hebben kunnen geven aan het einde van jouw leven….I'm allright!

Curriculum Vitae

Marjan Westerman werd geboren op 15 juli 1951 in Hilversum. Zij behaalde in 1969 haar Gymnasium B diploma aan het Christelijk Lyceum Hilversum en studeerde in 1973 af als fysiotherapeut bij “Leffelaar” Amsterdam. In de jaren daarna volgde zij verschillende specialisatie cursussen waaronder de Bobath opleiding bij het Goois Kinder Ziekenhuis te Huizen en de opleiding tot haptotherapeut (A en B) aan de Academie voor Haptonomie te Doorn. Zij was eerst als kinderfysiotherapeut 6 jaar verbonden aan de Rooms Katholieke Ziekenverpleging Hilversum en startte vervolgens in 1979 haar eigen fysiotherapie praktijk in het bejaardenhuis St. Carolus. In 1986 opende zij ook een praktijk voor haptotherapie. Naast het werk in haar praktijken was zij in Hilversum vele jaren werkzaam als docent pre- en postnatale educatie bij de Kruisvereniging en als gastdocent bij de verpleegkunde opleidingen van de drie ziekenhuizen. Zij was verder betrokken bij de ontwikkeling en de uitvoering van de cursus “Pre- en Postnatale Educatie” van de Stichting Wetenschap en Scholing Fysiotherapie in Amersfoort. Rond 1990 begon zij met haar universitaire studie en vanaf dat moment werkte zij alleen nog (tot 1998) als haptotherapeut. Zij behaalde haar doctoraal bul in 1999 aan de Faculteit der Wijsbegeerte van de Universiteit van Utrecht. Haar afstudeerrichting betrof de Praktische Filosofie met het accent op Bio-ethiek en Beleid & Organisatie van de gezondheidszorg. In september 2000 kwam zij in dienst bij het EMGO op de afdeling Sociale Geneeskunde van de Vrije Universiteit te Amsterdam en startte daar met haar onderzoek zoals beschreven in dit proefschrift. In de jaren 2005 en 2006 werkte zij op zeer kleine schaal weer als haptotherapeut. Sinds 2006 werkt zij bij de Faculteit der Aard- en Levenswetenschappen van de VU. Zij is als bachelorcoördinator, stagecoördinator en docent kwalitatieve methoden verbonden aan de afdeling Methodologie en Biostatistiek voor de opleiding Gezondheidswetenschappen. Marjan is zeker ook de trotse moeder van vijf kinderen, en oma van twee kleinkinderen.