Consequences of brain tumours from the perspective of the patients and of their next of kin
Consequences of brain tumours from the perspective of the patients and of their next of kin
© Tanja Edvardsson, 2008

Title: Consequences of brain tumours from the perspective of the patients and of their next of kin

Publisher: Örebro University 2008
www.oru.se

Editor: Maria Alsbjer
maria.alsbjer@oru.se

Printer: Intellecta DocuSys, V Frölunda 02/2008

ISSN 1650-1128
Abstract


A disease has consequences not only for the afflicted person but also for those who interact with him or her. A low-grade glioma is a brain tumour whose regarding its psychosocial implications for adult patients and their next of kin has received little attention in the literature. In the light of this the overall aim of the present thesis was to provide increased knowledge about how patients with low-grade glioma and their next of kin experience and deal with everyday life.

The methods of the studies were mainly qualitative. Thirty-nine patients and 28 next of kin were interviewed and all except one next of kin completed a quality of life questionnaire.

The onset of low-grade glioma was described from the patients’ perspective as a process, either rapid (up to a few months) or prolonged over several years. This phase of low-grade glioma encompassed repeated visits to physicians and care institutions. The onset of low-grade glioma was accompanied by stress, anxiety and uncertainty in the case of both the patients and those nearest. The symptoms and problems the patients experienced covered a broad range of consequences, physical, psychological and social. The patients presented a wide range of ways to cope with illness-related problems.

The next of kin were often deeply involved in the patients’ situation and many of them experienced extremely stressful emotions mainly in the early period of the illness. They had experience of positive encounters in health care but more often they had had a sense both of powerlessness and of being invisible and neglected. Relations and roles changed in ways that mostly were experienced as negative.

Enabling strength in everyday life had to do with alleviation of strain and having a positive outlook upon life. By means of the questionnaire Subjective estimation of Quality of Life (SQoL) the patients and those nearest estimated their quality of life as being comparatively high. Only one variable, among the patients the absence of work/meaningful occupation and among the next of kin the absence of own children, being estimated at below 60% of the maximum score.

Keywords: brain tumour, low-grade glioma, cancer, patient’s perspective, next of kin’s perspective, duration of disease onset, coping, subjective quality of life, content analysis.
ORIGINAL STUDIES

This thesis is based on the following studies that will be referred to as Study I, II, III and IV in the thesis.


IV. Edvardsson, T. & Ahlström, G. 2007, Quality of Life among persons with low-grade glioma and their next of kin. (Submitted)

Study I, II and III are reproduced with permission from the publisher.
# CONTENTS

<table>
<thead>
<tr>
<th>Section</th>
<th>Page</th>
</tr>
</thead>
<tbody>
<tr>
<td>INTRODUCTION</td>
<td>11</td>
</tr>
<tr>
<td>BACKGROUND</td>
<td>13</td>
</tr>
<tr>
<td>1. Psychosocial aspects of brain tumours</td>
<td>13</td>
</tr>
<tr>
<td>2. Medical and epidemiological frame of reference</td>
<td>14</td>
</tr>
<tr>
<td>3. Coping</td>
<td>17</td>
</tr>
<tr>
<td>4. Next of kin</td>
<td>19</td>
</tr>
<tr>
<td>5. Quality of Life</td>
<td>20</td>
</tr>
<tr>
<td>RATIONALE FOR THIS THESIS</td>
<td>23</td>
</tr>
<tr>
<td>1. Aims of the thesis</td>
<td>23</td>
</tr>
<tr>
<td>MATERIAL AND METHODS</td>
<td>25</td>
</tr>
<tr>
<td>1. Design</td>
<td>25</td>
</tr>
<tr>
<td>2. Participants</td>
<td>25</td>
</tr>
<tr>
<td>3. Data collection</td>
<td>28</td>
</tr>
<tr>
<td>4. Data analysis</td>
<td>30</td>
</tr>
<tr>
<td>5. Ethical considerations</td>
<td>35</td>
</tr>
<tr>
<td>RESULTS</td>
<td>37</td>
</tr>
<tr>
<td>1. Experience of onset of low-grade glioma: the patients’ and the next of kin’s perspectives</td>
<td>37</td>
</tr>
<tr>
<td>2. Illness-related problems and coping: the patients’ perspective</td>
<td>39</td>
</tr>
<tr>
<td>3. The experience of being the next of kin</td>
<td>43</td>
</tr>
<tr>
<td>4. Experience of quality of life: the patients’ and the next of kin’s perspectives</td>
<td>47</td>
</tr>
<tr>
<td>5. Brief summary of the prominent results</td>
<td>48</td>
</tr>
<tr>
<td>DISCUSSION</td>
<td>51</td>
</tr>
<tr>
<td>1. Result discussion</td>
<td>51</td>
</tr>
<tr>
<td>2. Method discussion</td>
<td>57</td>
</tr>
<tr>
<td>3. Clinical implications</td>
<td>60</td>
</tr>
<tr>
<td>4. Implications for further research</td>
<td>62</td>
</tr>
<tr>
<td>SAMMANFATTNING PÅ SVENSKA/SUMMARY IN SWEDISH</td>
<td>65</td>
</tr>
<tr>
<td>ACKNOWLEDGEMENTS</td>
<td>69</td>
</tr>
<tr>
<td>REFERENCES</td>
<td>71</td>
</tr>
</tbody>
</table>
INTRODUCTION

In my work as a psychologist at the Centre for Adult Habilitation run by Örebro County Council I meet patients with considerable and enduring disabilities, the patients next of kin and staff/assistants. I was aware that patients with disabilities resulting from brain tumours or persons around them, sometimes asked for support. Whilst patients with high grades brain tumours often have a shorter survival the patients who came to the Centre for Adult Habilitation were in general people with low-grade brain tumours living with long-lasting disability. My knowledge about these patients and their next of kin was limited. Also in the literature there was little to be found about what it means to live with low-grade brain tumour. As a consequence of this health-care staff do not know much about these patients, the next of kin and their need of psychosocial support. There is therefore every reason to explore and seek to understand the experiences of the patients and their next of kin from onset of the tumour and on. Understanding this specific life-situation also constitutes a basis from which to provide support for those who need it.

Therefore this thesis takes an interest in exploring the situation of the patients who had a diagnosis of low-grade brain tumour and those nearest. The thesis has its focus on the psychosocial aspects of having a low-grade brain tumour and what it means from the perspectives of the patients and their next of kin. This perspective has an emphasis on the study participants’ experiences of the illnesses and the implications for everyday life and on contacts in care. In addition, there is a concern with how the participants cope with the situation, also their estimations of quality of life.

It was good news to note that in spring 2006, the Swedish Brain Tumour Association, Svenska hjärntumörföreningen, was founded. This thesis indicates that a brain tumour disease shows specific characteristics that differ from those of other cancers. Therefore it gives me satisfaction that a Swedish association has been founded.
BACKGROUND

Psychosocial aspects of brain tumours

The psychosocial issues when a brain tumour afflicts a person include the psychological and social aspects of his/her world as well as that of his/her next of kin. Having a brain tumour is a complex condition including threat to life by a tumour located in the most important part of the body. Remaining impairment of cognition and memory, as well as personality changes, can cause an accumulation of distress that in turn has consequences for those around and in different social arenas.

Patients with brain tumours and their family experience extraordinary stress. They have to cope with a threatening diagnosis, changes in brain function and a disease course that is often unpredictable and problematical (Barr, 2003). An example of the diversity of brain tumours in relation to other cancers is that time since diagnosis and treatment do not mitigate the stress effect in brain tumour patients, as is the case among other types of cancer (Keir, Swartz, & Friedman, 2007). Comparisons between high-grade and low-grade brain tumours have also shown the low-grade brain tumour patients report at least the same amount of stress as the high-grade brain tumour patients (Keir, Guill, Carter, & Friedman, 2006).

The threat of a serious illness is a major source of stress and cancer is often viewed as a unitary stressor, without interest as to its different and specific stressors (Wasteson, 2007). In a study on patients with brain tumours the sources of stress were found to be familial and emotional rather than physical concerns (Keir et al., 2007). In addition to the patient with cancer, those nearest also experience stress.

Low-grade gliomas are not always separated from high-grade gliomas in research literature. Within the reviewed research literature malignant typically refers to high-grade, but in some studies it includes low-grade. The research interest in malignant glioma has mainly been focused on medical aspects, and radiology therapy has had a prominent position (Keir et al., 2007; Kilbride, Smith, & Grant, 2007). The rare occurrence of studies investigating the impact of a brain tumour on the patient’s and their family’s everyday life in the case of highly malignant gliomas holds true to an even higher extent for low-grade...
gliomas. How these patients experience and handle their altered psychosocial situation is rarely investigated.

The experience of the next of kin when a family member falls ill with a low-grade brain tumour is also sparsely reported in scientific literature. There is as well a gap in knowledge about carers of cancer patients who are not spouses/partners (Grinyer, 2004; Pitceathly & Maguire, 2003). Therefore the unique experience of being the next of kin of a patient with the diagnosis of low-grade glioma needs to be explored.

**Medical and epidemiological frame of reference**

**Histology**

Brain tumours can be primary (originating from the structures of the brain) or secondary (metastases originating from a pre-existing tumour) and they are also categorised as either malignant or benign (DeAngelis, 2001).

Primary brain tumours are classified according to the presumed cell of origin (Armstrong & Gilbert, 1996). The glioma arises from supportive tissues of the brain, the glial cells (Bautista, 2004). A majority of brain tumours originate from the glia 70%-77% (Berg, Blomquist, & Cavallin-Ståhl, 2003; Schwartzbaum, Fisher, Aldape, & Wrench, 2006). Brain tumours are also classified according to their growth and the most rapidly growing ones are assigned to grades III and IV, highly malignant. Grades I and II refer to low-malignant/low-grade (Grier & Batchelor, 2006; Papagikos, Shaw, & Stieber, 2005). Low-grade gliomas do not remain low-grade forever as they have the ability to transform into high-malignant tumours (Ashby & Shapiro, 2004; Papagikos et al., 2005; Walker & Kaye, 2001). Also these tumours have the ability to infiltrate surrounding areas of the brain (Duffau, 2006; Papagikos et al., 2005). Low-grade malignant glioma is a brain tumour category that gathers biologically varied neoplasms (Ashby & Shapiro, 2004; Papagikos et al., 2005), and the most common histological characteristic of low-grade glioma is astrocytoma (Papagikos et al., 2005). Oligodendroglioma is the next most common form in adults (Walker & Kaye, 2001). The most common mixed gliomas (more than a single glial cell type) are oligoastrocytomas (Walker & Kaye, 2001). This histological diagnosis and its rate of occurrence holds also for the patients in the present thesis. The neuro–imaging techniques in the case of
primary brain tumours are continuing to be developed and researchers believe that this will be beneficial for brain tumour treatment (Smits, Savitcheva, & Ribom, 2007), surgery (Duffau, 2006) and that the clinical care will continue to improve (Hoffman, 2001).

**Incidence**
The incidence of gliomas varies across patients’ ages. In a review the incidence of low-grade astrocytoma was about 1/100,000 a year, with a peak incidence at 30-40 years, and these tumours predominate in males (Walker & Kaye, 2001). Oligodendroglioma has a peak incidence around 40 years in adults and also exhibits male preponderance (Walker & Kaye, 2001). Male preponderance was the case also in the present thesis, where 65% were men. Low-grade gliomas are more common among white people (Cavaliere, Lopes, & Schiff, 2005). Low-grade astrocytoma arises approximately in proportion to the relative mass of brain lobes, hence the frontal lobe is the most common location, followed by the temporal lobe (Walker & Kaye, 2001).

In Sweden the National Board of Health and Welfare registered the incidence of all types of brain tumours in 2005 to 1166 individuals. Brain tumours account for 1166 of the total of 42,589 cases of cancer incidence in Sweden in 2005 (National Board of Health and Welfare, 2007).

**Causes**
There have been many speculations about the causes of brain tumours. Studies have shown conflicting results about for instance head trauma when exposed to high-tension wires (DeAngelis, 2001) and cellular phones (Christensen et al., 2005; Hardell, Carlberg, Söderqvist, Mild, & Morgan, 2007; Kan, Simonsen, Lyon, & Kestle, 2007; Ohgaki & Kleihues, 2005; Schüéz et al., 2006). More than 900 exposures of various kinds (chemicals, complex mixtures, occupations and infectious agent) have been assessed, where only 9 of these exposures show weak associations with nervous system tumours in humans (Ohgaki & Kleihues, 2005). One established environmental cause of higher risk of brain tumours is therapeutic or high-dose radiation (Schwartzbaum et al., 2006), particularly prophylactic treatment of children for acute lymphoblastic leukaemia (Ohgaki & Kleihues, 2005).
Symptoms and dysfunctions

Headache is the most common symptom related to brain tumours. Epileptic seizures and changes in cognition or functional ability are also frequently reported (Remer & Murphy, 2004). Widespread headaches can result from the tumour blocking the flow of cerebrospinal fluid (Armstrong & Gilbert, 1996).

Long-term cognitive dysfunctions affect up to 90% of low-grade glioma patients and are the result of the brain tumour itself, tumour-related epilepsy, treatment and psychological stress, or a combination of these factors (Taphoorn, 2003). However, the focal radiotherapy is not the main reason for cognitive dysfunctions, instead the tumour itself and other medical treatments contribute largely to the cognitive dysfunction (Taphoorn & Klein, 2004). Extensive cognitive dysfunctions have been shown when the tumour was found in the dominant hemisphere (Klein et al., 2002). Among low-grade gliomas, cognitive dysfunction has been reported (Pählson, Ek, Ahlström, & Smits, 2003), with a preponderance of memory problems (Hahn et al., 2003), slower information processing (Ek, Smits, Pählson, & Almkvist, 2005) and reduced attention functions (Giovagnoli, 1999). Other consequences reported from studies on patients with low-grade glioma are for instance depressive symptoms (Hahn et al., 2003) and fatigue (Lovely, 2004). A low-grade glioma involves a complex of symptoms and consequences and the disease concerns both the oncological and neurological domains (Armstrong & Gilbert, 1996). The patients’ own perception and experience of symptoms and changed function as a result of their brain tumour is an important issue investigated in the present thesis.

Treatment

The treatment of brain tumours is designed individually and depends on the type of tumour, its localisation and size, and the patient’s age and health (Bautista, 2004). Surgery, radiation therapy and chemotherapy are the traditional treatments of brain tumours, either separate or in combination (Armstrong & Gilbert, 1996; Bautista, 2004). It has not been confirmed that radiotherapy prolongs life for patients with low-grade gliomas (Berg et al., 2003; van den Bent et al., 2005). In low-grade gliomas higher radiation doses may give poorer quality of life without treatment benefits (Kiebert et al., 1998). Complications of therapy may cause more impairment than the tumour itself (Byrne, 2005) and the survival time in
the case of low-grade gliomas is a decade or more and therefore there is a higher risk of treatment-related complications (Grier & Batchelor, 2006).

There have been an increased variety of gene therapies in clinical trials for brain tumours in recent years. Patients with brain tumours have not been cured though, but the development of gene therapies is still considered to be in an early stage (Engelhard, 2000).

Survival
The prospect of survival in the case of low-grade gliomas can be decades (Påhlson et al., 2003). There has been reported an increased survival among people with malignant gliomas (Remer & Murphy, 2004), astrocytoma and oligodendroglioma (Davis, Freels, Grutsch, Barlas, & Brem, 1998). However, no increased survival rate has been shown among patients with low-grade gliomas (Oertel, von Buttlar, Schroeder, & Gaab, 2005). Survival rate depends on the patient’s age, tumour histology and size, tumour expanding (above midline) and neurological dysfunction before surgery (Papagikos et al., 2005). Poorer prognosis in the case of low-grade glioma is represented by a patient ≥ 40 years, with astrocytoma, presence of neurological dysfunction before surgery, tumour size > 6 cm, and tumour crossing the midline; the more of these characteristics, the poorer the survival prognosis (Pignatti et al., 2002). Lower grades of brain tumours have better prognosis than high-grade brain tumours and, oligodendrogliomas better than astrocytomas (Davis et al., 1998). The possibility of a long-term survival in the case of low-grade gliomas has implications for the everyday life of both the patients and the next of kin and is an essential issue to explore.

Coping
Living with long-term disease does not inevitably lead to a deterioration of the afflicted individual’s quality of life. This observation prompts questions about the importance of coping and today there is a considerable body of research based on this theoretical concept. Much of the research in the literature is based on the cognitive interaction model of stress and coping set forth by Lazarus and Folkman (1984). The model includes the person and the situation in which the person acts. The theory is also built upon the individual subjective interpretation
of a transaction and hence is a cognitive phenomenological theory. The definition of coping offered by Lazarus and Folkman is that it implies “constantly changing cognitive and behavioural efforts to manage specific external and/or internal demands that are appraised as taxing or exceeding the resources of the person” (Lazarus & Folkman, 1984, p. 141). The definition contains a certain amount of endeavour and a conscious striving to handle an extraordinary event/situation.

The process of coping includes three aspects of cognitive appraisal, primary, secondary and re-appraisal. The primary cognitive appraisal is the perception of something as irrelevant, benign-positive or stressful (Lazarus & Folkman, 1984). The stressful appraisal includes harm/loss, threat and challenge. The secondary cognitive appraisal is the reflection upon what can be done or not, and what the consequences might be of the strategies in mind. Re-appraisal is the process of changing appraisal according to new information.

There are several personal and environmental factors that influence the cognitive appraisal. Personal factors include belief, for instance about personal control (Lazarus & Folkman, 1984). Commitment is another person factor relevant to stress theory and this factor signifies what is considered important in life, what the person dearly values. Environmental factors that influence cognitive appraisal are for example the novelty and predictability of situations or events. In addition the timing of an event, the events uncertainty and the duration of a stressful event are of interest in the context of diseases (Lazarus, 2000).

According to Lazarus and Folkman coping has two main functions. The problem-focused coping is about handling the problems that cause the stress, and the emotion-focused coping relates to lessening the negatively experienced emotions. Lazarus emphasises that the problem-focused and emotion-focused coping are not separate, because “both type of strategies are interdependent and work together” (Lazarus, 2000, p. 669).

Sometimes coping is considered to be the same as an adaptive successful solution, a notion that Lazarus and Folkman object to. Coping is not to be regarded as a mastery of the situation either, because there are situations that do not permit being mastered. Coping is the effort to meet stressful situations irrespective of the outcome (Lazarus, 1993; Lazarus & Folkman, 1984). Coping depends in part on what inherent resources the person possesses for instance the ability to solve problems, health and energy (Lazarus, 2000).
Coping used by cancer patients is seldom studied in clinical subtypes, and studies often include broad questions such as “How do you cope with your cancer?” Therefore more specific investigations of coping behaviours for separate subtypes of cancer are needed (Livneh, 2000; Somerfield & Curbow, 1992). In the present thesis the study of coping among the patients was to be related to problems due to the low-grade glioma and the coping had to be related to a current situation.

Next of kin

It is well established that when a person gets a diagnosis of cancer their next of kin become deeply involved (Blanchard, Albrecht, & Ruckdeschel, 1997; Germino, Fife, & Funk, 1995; Nijboer et al., 1998). In studies on adults and cancer the next of kin as a rule are spouses and little attention has been paid to parents of young adults (Grinyer, 2004). There has been an increasing reliance on informal carers to provide support for their ‘patient relative’ with cancer (Pitceathly & Maguire, 2003). The carers’ role has also proved to be of more importance and interest when the time as in–patient shortens (Laizner, Yost, Barg, & McCorkle, 1993; Lim & Zebrack, 2004). In a study it was shown that patients with brain tumours had a significantly shorter rehabilitation length of stay and a greater discharge to the community than patients with traumatic brain injuries (Huang, Cifu, & Keyser-Marcus, 2000). The physical, economic, emotional, practical and administrative burden on the healthy family member increases and can lead to worse physical, psychological and social health for him or her (Jensen & Given, 1991; Lim & Zebrack, 2004; Pitceathly & Maguire, 2003). Family members in the case of cancer patients have reported as many psychosocial problems as the patients with cancer, or even more such problems, (Baider, Koch, Esacson, & De-Nour, 1998; Gaugler, Davey, Pearl, & Zarit, 2000).

In a study from Norway encompassing 12 of the most common cancer diagnoses, the relatives’ experience of received help and support from the public health services was measured. A majority of the relatives were quite dissatisfied with the help and support they had received. They were less satisfied than the patients with all aspects of the public health services (Isaksen, Thuen, & Hanestad, 2003). In addition family members’ adjustment and satisfaction proved
to be better among those who received information adjusted to their own specific needs (Pitceathly & Maguire, 2003). Studies have found that female partners of stroke patients are over-represented when it comes to distress (Franzén-Dahlin, 2007). In a cancer study it was shown that females experienced higher degrees of psychological distress (irrespective of patient or partner role) and female partners had lower quality of life than male partners (Hagedoorn, Buunk, Kuijer, Wobbes, & Sanderman, 2000). Though carers of cancer patients develop high emotional distress they are unlikely to request help (Pitceathly & Maguire, 2003).

The family’s experience when one of the family falls ill with a brain tumour is unique, and such a tumour has more psychosocial effect on the family than other types of cancer (Leboeuf, 2000). It causes a radical change in the routines of daily life and the whole family is affected by the tumour (Fox & Lantz, 1998). Roles and relations within the family are greatly affected when patients fail to fulfil their former roles and many patients retire (Salander et al., 2000; Wideheim, Edvardsson, Pålsson, & Ahlström, 2002). The uncertainty cancer elicits is a source of stress both for the patient and for the relatives (Newton & Mateo, 1994). It is the special focus in this thesis to explore the experience of being the next of kin of a patient diagnosed with a low-grade glioma.

**Quality of Life**

The concept of quality of life (QoL) can be derived from diverse traditions. According to Dijkers (1997) the concept of QoL can be derived from three different traditions. The first is the socio-economic tradition (measuring things like life expectancy and consumption), the second is the subjective tradition (measuring experienced congruence or disparity between people’s aspirations and accomplishments), the third is the objective QoL tradition (measuring things like income level and number of television sets in the home) (Dijkers, 1997).

The World Health Organisation (WHO) has suggested a definition of QoL as “individuals’ perception of their position in life in the context of the culture and value system in which they live and in relation to their goals, expectations and standards” (WHO, 1998, p. 1570). However, no consensus exists on what QoL is. Most researchers in the field agree upon QoL as a multidimensional or multifaceted construct (Cummins, 2005; MacDonald, 2001; Meeberg, 1993; Murrell, 1999; Pain, Dunn, Andersson, Darrah, & Kratochvil, 1998; Smith, Avis,
& Assmann, 1999; Verdugo, Schalock, Keith, & Stancliffe, 2005; WHO, 1995, 1998). QoL seems to have a minimum base of three dimensions, physical, psychological and social (Lovely, 1998; WHO, 1995). The WHO’s definition also points to the largely agreed upon subjective and individual nature of QoL (Ganz, 1994; WHO, 1995, 1998). The subjective definition implies that the evaluation of QoL is dependent on the person who experiences it (Bowling, 1997).

Health–Related Quality of Life
The term Health-related Quality of Life (HRQoL) concerns the impact of a disease and its medical treatment on other aspects of the patient’s life (Murrell, 1999). In a review it was found that many investigators saw quality of life as something like health status or functional status, showing that this concept in the medical context had no unique meaning (Gill & Feinstein, 1994).

HRQoL issues in the case of cancer patients seem to be of increased interest as long-term survival increases (Carver, Smith, Petronis, & Antoni, 2006; Gotay & Muraoka, 1998). HRQoL variables in the case of brain tumour patients often include symptoms like fatigue, nausea and vomiting, as in the European Organization for Research and Treatment of Cancer (EORTC) questionnaire (Gustafsson, Edvardsson, & Ahlström, 2006; Heimans & Taphoorn, 2002; Taphoorn et al., 2005). Also neuro-cognitive functions/neuro-psychological functions (Efficace & Bottomley, 2002; Hütter, Spetzger, Bertalanffy, & Gilsbach, 1997; Weitzner, 1999; Weitzner & Meyers, 1996) and depression (Giovagnoli, Silvani, Colombo, & Boiardi, 2005; Pelletier, Verhoeef, Khatri, & Hagen, 2002) have been of interest in the case of brain tumour patients’ HRQoL. In the case of low-grade brain tumours the effect of treatment such as radiotherapy (Kiebert et al., 1998) and use of anti-epileptic drugs (Klein et al., 2003) has been measured.

In a parallel study the patient study group involved in this thesis answered the EORTC questionnaire and the patients’ function was assessed by means of the WHO performance status scale (Gustafsson, Edvardsson, & Ahlström, 2006). Nearly all patients were capable of self-care but less than half were able to carry out normal activities without restriction. Problems with fatigue, sleep
disturbances and pain were most frequent, and nearly half of the patient group had scores indicating low HRQoL

*General Quality of Life*

Besides HRQoL there is what in this thesis is called general QoL to be seen in subjective estimations of psychological wellbeing, quality of relationships, work and housing. The family wellbeing is important both for the patient and the next of kin (Gotay & Muraoka, 1998) and therefore QoL issues among next of kin of patients with various diseases are of interest. In QoL studies of persons not afflicted by a disease it is adequate to ask for their subjective estimates in general QoL. QoL dimensions that have been identified as important from a family perspective have focused on emotional health, relationships and an enjoyable/meaningful life (Pain et al., 1998).

In Sweden a general QoL instrument has been developed, the Subjective estimation of Quality of Life (SQoL) (Kajandi, 1994, 2006). The aim was to make it applicable to all groups of adolescents and adults (Kajandi, 1994, 2006). This implies that healthy persons and those with various diseases can use the same instrument and it also makes comparisons between groups possible. This questionnaire includes variables measuring external life conditions, interpersonal relations and internal psychological states (Kajandi, 1994, 2006). The significance of emotional experiences with regard to QoL has been emphasised by Naess (Naess, 1987) and has influenced the development of the SQoL (Kajandi, 1994, 2006).

The estimation of QoL may help in planning support and as a feedback on intervention programmes (Verdugo et al., 2005). In a study of Muscular Dystrophy, the patients and the next of kin estimated their own QoL with the same instrument (the SQoL) (Boström & Ahlström, 2005). Nothing has been found in literature on brain tumour or cancer studies that measures subjective QoL in both patients and their next of kin in that way.
RATIONALE FOR THIS THESIS

The everyday life of low-grade glioma patients is seldom described in research literature. These patients constitute a small group as compared to the groups of for instance prostate and breast cancer patients. There are similarities with other diagnostic groups, nevertheless some interesting combinations of features in the case of low-grade glioma patients need to be explored. These patients have the prospect of many years of survival combined with the commonly remaining negatively changed physical and neuro-cognitive functions. There is a risk that these brain tumours will develop into more malignant tumours. A question to pose is how patients and next of kin deal with their lives under these circumstances.

It can probably be said that there is risk of strong impact on the patients and their next of kin in terms of changes in everyday life and distress. Informal carers often provide support for the patient with cancer (Pitceathly & Maguire, 2003). Many years of living in a distressing situation might lead to the next of kin’s own poorer health. Therefore it is important to acquire knowledge about how the next of kin experience the situation of being close to their ‘patient’.

Aims of the thesis

The overall aim of this thesis is to contribute to increased knowledge about how patients with low-grade brain tumour and their next of kin experience and deal with their everyday life. The thesis is presented in four studies about the following specific questions:

- How do the patients and the next of kin experience the onset of low-grade glioma? (Studies I and III)
- How do the patients and the next of kin experience the encounters in care? (Study I and III)
- How do the patients experience the implications of the low-grade glioma illness? What kinds of problems appear? (Study II)
- How do the patients cope with the problems related to the low-grade glioma illness? (Study II)
-What does the low-grade glioma illness imply for the next of kin in terms of experience and emotions? (Study III)

- Are the experiences (following the previous question) differently distributed in the various groups of next of kin? If so, are the differences related to sex, kind of relationship, age or the patients’ disease duration? (Study III)

- How do the patients and the next of kin estimate their own quality of life? (Study IV)

- Are there differences between and/or within the patient group and the next of kin group in quality of life estimates? If so, are the differences related to sex, age or the patients’ disease duration? (Study IV)
MATERIAL AND METHODS

Design
This thesis comprises empirical cross-sectional studies employing qualitative method (Studies I and II), mixed qualitative and quantitative (Study III) and quantitative method (Study IV). An overview of the studies is presented in Table 1.

The qualitative approach was considered appropriate due to the limited knowledge in this specific research area and when openly searching for people’s life experiences (Lieblich, Tuval-Mashiach, & Zilber, 1998). The content analysis used in Studies I, II and III involved an inductive endeavour, involving being open to whatever the participants said, though within the limitations of the study’s research question.

The mixed method approach in Study III consisted of a main analysis within the core of inductive qualitative data supplemented with a quantitative analysis. If the purpose of a study is to describe, discover or find meaning, the theoretical drive (the main direction or thrust) will be inductive. In that case most often a qualitative method is used (Morse, 2003). Thereafter, a second, quantitative method was used to explore whether the themes (appearing from the content analysis) were differently frequent among subgroups of next of kin. The mixed method in this study was inspired by Morse (2003) and the purpose was one of discovery, and the quantitative data analysis added information that otherwise would not have been obtainable. The added information was how the content within each theme was distributed in different groups of next of kin.

The method in Study IV was quantitative, with a descriptive statistics and a non-parametric comparative analysis of differences between the group of patients and the group of next of kin concerning their subjective estimations of their own quality of life.

Participants
The search for patients to be included in this research was performed within a geographical area in the central of Sweden with the use of the Regional Cancer Register, where all cancer cases have to be registered.
Table 1 Overview of the studies in the thesis

<table>
<thead>
<tr>
<th>Study</th>
<th>Design</th>
<th>Method</th>
<th>Data collection</th>
<th>Data analysis</th>
</tr>
</thead>
<tbody>
<tr>
<td>I</td>
<td>Cross-sectional</td>
<td>Descriptive Qualitative</td>
<td>Individual semi-structured interviews</td>
<td>Content analysis</td>
</tr>
<tr>
<td>II</td>
<td>Cross-sectional</td>
<td>Descriptive Qualitative</td>
<td>Individual semi-structured interviews</td>
<td>Content analysis</td>
</tr>
<tr>
<td>III</td>
<td>Cross-sectional</td>
<td>Descriptive Mixed qualitative and quantitative</td>
<td>Individual semi-structured interviews</td>
<td>Content analysis Descriptive statistics</td>
</tr>
<tr>
<td>IV</td>
<td>Cross-sectional</td>
<td>Descriptive Quantitative Comparative</td>
<td>The questionnaire Subjective estimation of Quality of Life</td>
<td>Descriptive and inference statistics</td>
</tr>
</tbody>
</table>

An initial inventory of persons diagnosed with low-grade glioma living in the County of Örebro was conducted with support from a neurologist with access to the register. The inclusion criteria were a diagnosis within low-grade gliomas, age at least 18 years and living in the County of Örebro on the prevalence date, 15th November 1999. One criterion for accepting a patient was a histologically verified diagnosis of low-grade glioma (grades I and II) according to WHO 2002 or the previously used Kernohan classification for tumours diagnosed before 1993 (Kleihues et al., 2002). Another inclusion criterion was that the patient had to have the ability to manage an interview in conversational form. Every patient’s treating neurologist made this judgement. The inventory ended up with 49 patients matching the inclusion criteria and it was considered realistic to carry out the intended research with all of them.

Out of 49 identified patients matching the inclusion criteria, 10 refrained from participating, corresponding to 80% participation. The group of 39 patients with predominantly low-grade gliomas comprised 27 men (69%) and 12 women (31%), mean age 46 years (SD 14), range 21–79 years. The mean duration since diagnosis was 16 years (SD 13), range from less than a year to 47 years. The
histological diagnosis was dominated by astrocytoma grades I-III (n=23), followed by oligodendroglioma grades I-III (n=8), oligoastrocytoma (mixed gliomas) grades I-II (n=4), ependymoma grade II (n=3) and low-grade glioma without further description (n=1). Two persons with glioma WHO grade III with a clinical picture like a low-grade glioma were also judged appropriate to be included, by the two neurologists who reviewed the medical records.

Of the 39 participating patients, 28 were ready to include a next of kin. There was one next of kin that was unwilling to participate, whilst in the case of one patient there were two next of kin ready to participate. Thus this research included 28 adult next of kin connected to 27 patients. Of the next of kin, 15 (54%) were spouses or co-habitants. The 13 next of kin that lived separate from the patients were: 3 live-apart partners, 8 parents, 1 sibling and 1 adult child. There were 8 men and 20 women, ranging in age from 25 years to 77 years (mean = 52.5 years).

In Study I only those patients who remembered the occasion when they got their (explicitly given) diagnosis were included. Twelve patients were thus excluded because they could not remember the specific occasion when they received information about the diagnosis. Since the aim was to describe the phase of falling ill it was important to differentiate experiences before the diagnosis. A total of 27 patients, 18 men (67%) and nine women (33%), were included in this study. One patient had experience of two different brain tumour onsets and hence 28 onsets were investigated. The mean age was 47 years (range 23-79 years), and the mean duration since diagnosis was 15 years (range 1-47 years). Five patients had experienced onset before the age of 18. Seventeen patients were either married or cohabitating, while 10 were living on their own.

In Study II 39 patients who consented to participate were included and in Study III there were 28 next of kin participating.

In Study IV there were 39 patients participating. Out of the 28 next of kin there was one who because of own disease and at own request was interviewed on the telephone. This next of kin therefore never filled in the questionnaire and hence 27 next of kin participated in Study IV. In summary, Study IV comprised 66 participants. The distribution of participants in the different studies can be viewed in Figure 1.
### Participants included or excluded

<table>
<thead>
<tr>
<th>Participants</th>
<th>Study</th>
</tr>
</thead>
<tbody>
<tr>
<td>49 patients</td>
<td>matched the inclusion criteria</td>
</tr>
<tr>
<td></td>
<td>10 patients refrained from participation</td>
</tr>
<tr>
<td>39 patients</td>
<td>participated</td>
</tr>
<tr>
<td></td>
<td>27 patients who remembered when the diagnosis was given were included in Study I</td>
</tr>
<tr>
<td></td>
<td>39 patients included in Study II</td>
</tr>
<tr>
<td></td>
<td>28 patients willing to include a next of kin</td>
</tr>
<tr>
<td>28 next of kin</td>
<td>were asked about participation</td>
</tr>
<tr>
<td></td>
<td>one next of kin refrained from participation</td>
</tr>
<tr>
<td></td>
<td>an extra wanted to participate</td>
</tr>
<tr>
<td>39 patients and 28 next of kin</td>
<td>66 participants</td>
</tr>
<tr>
<td>39 patients</td>
<td>28 next of kin was unable to participate</td>
</tr>
<tr>
<td>28 next of kin</td>
<td>included in Study IV</td>
</tr>
</tbody>
</table>

**Figure 1** Flow-chart of participants in the different studies

---

**Data collection**

From the inventory the participant patients (and later on those nearest) were contacted through a personal letter describing the research and its aims. After about a week I made a follow-up contact by phone. At this time they were given answers to any questions and complementary information about the study. Those willing to participate gave their oral informed consent. We came to an agreement on where to meet for the interview, and about half of the participants wanted it to
take place at my workplace, whilst the other half invited me to their homes. There were two participants (one patient and one next of kin) who chose to do the interview over the telephone. Two patients and one next of kin wished the interview to take place at the time they were at the university hospital. For this purpose I made a reservation for a secluded room in the hospital. The data derived from an interview and a questionnaire was carried out on the same occasion.

**Interview**

The data in Studies I, II and III was based on individual, semi-structured interviews with five main themes formulated in an interview guide: Onset of illness, Life before illness, Current life situation, Experiences of encounters with professionals in care and Thoughts about the future. Care was understood in a broad sense and included what the participants told of their hospital stay, outpatient settings, rehabilitation or habilitation.

Participants sometimes spontaneously started by selecting a topic that was of importance to them. I was striving to make the interview into a conversation and the participants were encouraged to share their experience. The character of the interviews varied, as there were patients with cognitive and communicative difficulties and other patients and next of kin that with ease presented rich stories on the topic in question. Occasionally when the conversation did not correspond with the interview guide I smoothly and easily brought it back to the subject field. The participants’ statements regarding particular areas of experience were followed up with additional questions in order to capture description of thoughts, emotions and actions. This strategy allowed the interviewees to give more comprehensive descriptions of the experiences. I assumed a non-judgmental position towards whatever contents emerged (Patton, 1990). I tried to inspire the participants to tell as much as possible within the themes and gave confirming verbal and non-verbal encouragement.

The interview duration was from slightly less than one hour up to two hours and usually encompassed approximately 70 pages of double space transcribed text. All the interviews and the filled-in questionnaires were collected before the data analysis began.
**Quality of Life questionnaire**

A questionnaire measuring general QoL was used in Study IV (Kajandi, 1994) and is termed the Subjective estimation of Quality of Life questionnaire (SQoL). This questionnaire was developed in Sweden and is intended both for healthy persons and for different patient groups. It is built up of eighteen variables referring to three life-domains: external life conditions, interpersonal relationships and internal psychological states, and one variable concerning QoL as a whole.

The external life conditions consisted of, Housing quality, Work/occupation quality and Quality of personal economy (Study IV). The interpersonal relationships domain included Relationship to a partner, Relationship to friends, Relationship to mother and father, and Relationship to own children. The domain of internal psychological states included Engagement, Energy, Self-actualisation, Freedom, Self-assuredness, Self-acceptance, Emotional experiences, Personal security and General mood. Finally there was the eighteenth variable Quality of life as a whole.

The questionnaire has a nine-point Likert scale where the lowest value is 1 and the highest is 5 and with 0.5 increments. Four variables result in mutually exclusive answers: Work/meaningful occupation (or Absence of work/meaningful occupation), Relationship to partner (or Absence of partner), Relationship to friends (or Absence of friends) and Relationship to own children (or Absence of own children) (Kajandi, 2006). A given score on one alternative thus excludes scoring on the other. The participants estimated the level of SQoL in their current life situations for each variable and filled in the questionnaire in connection with the interview (Study IV).

**Data analysis**

**Qualitative analysis**

In this thesis the method of qualitative data analysis used was content analysis. This is a systematic approach where the researcher moves back and forth between the whole and the parts of the texts in a process of identifying, coding and labelling the condensed data (Downe-Wamboldt, 1992). Content analysis is an appropriate choice in studies exploring meaning, interpretations, consequences and contexts (Downe-Wamboldt, 1992) and real-life problems.
(Lieblich et al., 1998). Graneheim and Lundman state that to understand the theoretical assumption underlying qualitative content analysis it is necessary to relate the method to communication theory (Graneheim & Lundman, 2004). The communication theory recommended is that of Watzlawick, Beavin and Jackson (1967). This theory postulates five axioms starting with “the impossibility of not communicating” (Watzlawick et al., 1967, p. 48). Some of the other axioms' relevance for content analysis concerns the content and relationship level of communication. The words (content) are also carrying a message about for instance attitudes or intentions (relationship aspect). This is often without the full awareness of either sender or receiver. The axiom of digital and analogic communication deals with verbal and non-verbal communication. The authors declare that “the content aspect is likely to be conveyed digitally whereas the relationship aspect will be predominantly analogic in nature” (Watzlawick et al., 1967), exemplifying in what way these aspects differ from and are related to each other.

The content analysis procedure in Studies I, II and III was much the same. The main differences were in the use of concepts, because of the different sources of inspiration and the different focus of the studies. The content analysis all began with an open-minded listening to the audiotapes while scrutinising the interview texts to get an impression of the whole. The interview texts in their entirety served as a reference to be returned to throughout the process of analysis. The procedure was characterised by comparing the evolving content back and forth in order to attain a higher order of abstraction. From the interview (the text and audio tape) forward to a central and concluding theme or category the procedure was characterised by successively making the content more condensed while retaining its essence.

In addition to the question of procedure there is the question of at what content level the analysis is to be performed. In content analysis the content level may be manifest or latent. The manifest level is described as an explicit/superficial level, or close to the text, while the latent level is a more in-depth level (and therefore more interpretation of an underlying meaning is required) (Downe-Wamboldt, 1992; Graneheim & Lundman, 2004). This means that a different depth of interpretation was required depending on the content
level. In Studies I, II and III content analysis was applied both on the manifest and the latent content level.

The audio-recorded and typed-out verbatim interview texts included pauses and expressions of emotion. These non-verbal indications were considered important for understanding and analysing the content. The non-verbal indications in the form of for instance heavy sighs gave an indeterminate statement an accompanying worth that was helpful when interpreting the content.

In addition to the patients’ description of symptoms, the distinctive aspect of the content analysis in Study I was the search for experiences of onset of low-grade glioma. The content analysis was of manifest and latent content, describing the onset (Baxter, 1991; Downe-Wamboldt, 1992; Lieblich et al., 1998). The selected texts, now in the form of meaning units were then formulated in preliminary categories, relying on meaning-bearing words and central topics in the text. The evolving analysis of one interview text at a time led to categories being increasingly filled in and varied. All the categories from all the individual interviews were then brought together, and comparisons were again drawn between the categories of the whole group. The evolving categories were delineated on the basis of differences and similarities in their content and meaning. The categories formed the result, expressed in the themes. The co-author (GA) independently read and reflected on the statements as well as discussed the interpretations in the analysis to establish trustworthiness in Studies I and II.

In Study II the content analysis was focused on manifest and latent content regarding illness-related problems and coping strategies. The analysis was separated as the first procedure concerned the problems and the following procedure concerned coping. The search for, and understanding of, problems and coping had to be related to a current troublesome experience of having the low-grade glioma. From the gradual consolidation of accounts of specific problems and coping by coding, sorting and naming them and putting them into higher-order categories, more complete descriptions emerged. Later in the analysis of problems, memory problems that emerged from the interview texts proved to be notable and they were regarded as important and not to be concealed within ‘cognitive problems’. Therefore memory problems were analysed and described on their own, separated from cognitive problems. A generous understanding of
the effort and consciousness in the definition of coping given by Lazarus and Folkman (1984 p 141) guided the search for statements about coping. The generosity in defining coping meant that there had to be at least some attempt that could be considered an effort and the person had to be aware of handling the problem as an indication of the consciousness aspect (Study II).

The main source of inspiration regarding the content analysis in Study III was the description by Graneheim and Lundman (2004). In Study III the content analysis was of manifest and latent content but more often concerned the latent content since the underlying content I was mainly in search of was the next of kin’s emotions. Meaning units that reflected the experience of being someone close to a person with a low-grade glioma were identified and given a code. The codes were examined and interpreted more deeply by asking, “What does this reflect?” By comparing all the codes it was possible to put them into different categories in accordance with the underlying subject matter they represented (Graneheim & Lundman, 2004). On the occasions where the interview texts were difficult to interpret I turned to the co-author (GA) and a discussion took place until consensus on the most reasonable interpretation was reached. The categories of similar experiences and emotions were then gathered into sub-themes, which in turn constituted the components structuring the overarching themes. Themes were developed through mutual agreement between the authors about key contents (Graneheim & Lundman, 2004). Categories, sub-themes and themes are not necessarily mutually exclusive, the assumption being that “owing to the intertwined nature of human experience, it is not always possible to create mutually exclusive categories when a text deals with experiences” (Graneheim & Lundman, 2004 p 107).

Content analysis can be carried out with or without computerised facilities such as NVivo (Richards, 2002). In Studies II and III the software program NVivo 2.0 (Richards, 2002) was used to facilitate sorting and arranging the content of the interview texts. In this program any text content can always be traced to its source, put in new constellations and connected to other data put into the program.
Quantitative analysis

The quantitative statistical method in Study III applied with the aid of NVivo was designed to explore whether the themes (that appeared from the previously performed content analysis) were differently frequent among subgroups of next of kin. The averages of accounts in the themes were counted in relation to the next of kin in groups of sex, type of relation, age and disease duration. This complementary method gave results that would not have been achievable with qualitative analysis only. The two methods, the main qualitative and supplementary quantitative, were also sequential, meaning that issues first uncovered qualitatively got a subsequent extension (Morse, 2003) (Study III).

In Study IV a QoL questionnaire with Likert scale was used. The quantitative analysis involved ordinal data and consequently non-parametric statistical tests (Bland, 2000). The Kruskal-Wallis test was used to compare groups of three or more for differences and the Mann-Whitney test was used to determine and inspect the differences between two groups. The participant background factors used in this study were sex, age and the disease duration group. These factors were compared with the eighteen SQoL variables. Results of p < 0.05 were considered statistically significant. In order to compare the results of the SQoL with those of previous studies the descriptive data was presented in mean and standard deviation values (SD), above the median values. The computer program Statistical Package for the Social Sciences for Windows (SPSS) 14.0 was used for statistical analyses (Study IV).

Author perspective

My perspective, pre-understanding, in this thesis is essentially related to my professional knowledge and experience as a working psychologist in the area of disability and habilitation. During my many years of encounters with persons having various disabilities, their family members and staff, I have taken the position that every person’ is worth listening to. Every person also has resources within themselves and there are also resources from family, friends and professionals to make use of, and this holds for everybody with or without dysfunctions. I have confidence that there are always possibilities of improvement in one way or another.
Ethical considerations

The Swedish Research Council’s research ethics principles in the humanistic-social scientific field were followed (Swedish Research Council, 2007) and this research has been reviewed and approved by the Research Ethics Committee at Örebro University Hospital, Sweden.

Participation in the research study was clearly formulated as being voluntary, with the right to withdraw at any time without giving reason for it. Also it was pointed out that participating in the research did not influence the patients’ ongoing care contact or treatment. The participants had the possibility of contacting me by telephone or letter. They were informed about how data were to be handled and reported and that confidentiality would be preserved.

Asking the participants where to meet and how to arrange the interview was a question of showing respect for their special requirements. There was no time limit for the interviews and these were with the permission of the individual participants audio-taped. The unlimited time allowed for pauses if signs of distress appear. Despite some participants’ displayed distress, sorrow or excitement they clearly stated this was not a hindrance to the ongoing conversation. On the contrary the conversation seemed to provide an opportunity to alleviate discomfort. Sometimes when discomfort appeared and I asked if we should pause or stop altogether, the participants said it was no problem or even that it was good for them. In the group of next of kin several participants were pleasantly surprised and expressed gratitude for being asked about their experiences.

It was known that I was not employed at the hospital and was independent of healthcare departments where the patients were involved. It was also possible to refer participants to suitable treatment if the interview aroused unbearable reactions, although this was not found use for.

Being allowed to hear another person’s story is a privilege and is in research connected with special obligations. Being an outsider sharing in another individual’s life in a critical phase of life calls for special cautiousness with what is supplied but also an obligation to convey the results.

In April 2005 all participants were invited to a meeting when the results up till then were presented. This meeting was well attended by the patients and the next of kin.
RESULTS

Experience of onset of low-grade glioma: the patients’ and the next of kin’s perspectives

The onset of low-grade glioma proved to have specific implications for both patients and next of kin (Studies I and III). Patients described the onset of the brain tumour as a sequence of events, beginning with symptom experiences that occasioned several health-care contacts and ending with a definitive diagnosis. More patients (71%) described a rapid than a prolonged onset (29%).

Rapid or prolonged onset

A rapid onset is in this thesis [taken to mean that about six months] elapsed from the first symptom to the diagnosis. Some of the patients experienced the onset as frightening and dramatic, with in many cases sudden severe headaches, epileptic seizures, vomiting and vision changes. Muscle pain and fear as to the potential causes followed the epileptic seizures. When the seizure occurred in a public place, the patients experienced shame and fear that onlookers would believe it had been caused by alcohol. Also fear that one’s own children would be frightened by the seizure was expressed. Most of the patients in the rapid-onset group suspected the presence of a brain tumour during the onset period. Several of next of kin or friends initiated the health-care contact because the person was unconscious. Most sought health care immediately; only a few put it off. Roughly one-third sought health care several times before a brain tumour was confirmed (Study I).

A more prolonged onset with successively increasing symptoms was described by eight of the 27 patients (Study I). As among the patients in the rapid-onset group, the most frequently mentioned symptoms were headaches and epileptic seizures. Several symptom experiences were reported only in the group that experienced prolonged onset: difficulty orienting oneself, personality changes, sensory losses, memory problems, racing thoughts and tinnitus. Among those with a prolonged onset it was often several years before a correct diagnosis was made. The symptoms worsened over the years, occasioning numerous health-care contacts in most cases. The onset period in the prolonged-onset group ended with an acute culmination in half of the cases. The patients in the
prolonged-onset group took their initial contact with health-care themselves (Study I).

Patients’ negative and positive experiences in care contacts
In addition to symptoms, the patients described mainly negative but also positive experiences of encounters and treatment when they sought care in the onset phase. Negative experience in health care were for instance disrespectful encounters, lack of medical competence, lack of participation, multiple physician contacts and treatment that did not help. The patients felt abandoned by health-care staff when they in spite of saying that something was wrong, probably “something in the brain”, didn’t get any attention or were taken seriously. On the other hand positive experiences were to be found, like respectful encounters, medical competence among the health-care staff being allowed participate and physician continuity (Study I).

Patients’ negative and positive experiences in everyday life
The patients also described how their life and their relationships were affected by the illness onset. Their usual life was affected in negative terms like lack of social support, encountering ‘bad’ attitudes on the part of other people, limitations in social roles, negatively affected education and work and the necessity to give up driving the car. On the positive flank the patients appreciated being met with involved next of kin and friends who gave practical and emotional support (Study I).

The next of kin’s negative and positive experiences in care contacts
The next of kin experienced extremely stressful emotions with respect to the patient’s illness onset period, including being frightened and being in a chaotic state (Study III). They described the onset period as a terrible time, with uncertainty as to whether surgery would be successful, and some of them wondered if the patient would ever be able to talk and walk again. The onset period was characterised by a strong focus on the patient. Though the next of kin had understanding for this, many of them felt invisible and neglected by health-care staff. Some of them felt powerlessness and perceived themselves as being left outside in their striving for participation on behalf of the patient. The next of
kin had needs of their own that sometimes were not recognised by health-care staff, and they were left on their own to seek professional support. However, they felt confidence when health-care staff in some cases expressed their awareness of the situation concerning the whole family. When health-care staff invited the next of kin to participate in decisions about patient care, clearly indicated to whom they could put questions, gave explanations and all the requisite information, they felt secure and allowed to participate (Study III).

**Illness-related problems and coping: the patients’ perspective**

Every patient described their symptoms, changed functions and illness-related problems and they spoke about how they were handling the problematic situations (Study II).

**Perceived illness-related symptoms and problems**

The patients reported experiences of physical changes and when they compared their present physical condition prior to the brain tumour disease they had worse balance, their muscles were weaker and their stamina was worse. After surgery some patients recounted deformations of the skull, scars or weak spots that caused difficulties. They for instance had difficulty in combing their hair or they had to take care not to fall or hurt themselves at the weak spot on their head (Study II).

Perceived problems regarding memory and cognitive abilities were frequently spoken by from the patients. Memory problems of recently learned information were most commonly recounted but also the prospective memory was worse. Cognitive abilities that were perceived being worse were the ability to concentrate and maintain attention, the process of cognitive handling of information and the taking of initiatives. The patients’ ability to spatially locate themselves, dependent on cognition and memory, was worse. The ability to communicate was negatively affected in different ways, finding the right words in speech or the right letters in spelling had become difficult, speech had become less fluent and they had trouble in putting together words in speech. The patients had become worried, frightened and anxious. Sadness and depressed states and emotions of shame and lowered self-esteem were reported. Some patients were angry about the situation they had been put into while a few others revealed
indifference. Sensory perception was afflicted – eyesight, hearing, tactility and taste (Study II).

The problems the patients described were also extended to and had an influence on every social and environmental area where these problems were constituents. Therefore life with family and friends, work or education, leisure, financial matters and the physical environment were problematic for many of the patients. There were individual patients recounting an almost unaffected life while others were more seriously affected by their tumour disease (Study II).

*Coping with the illness-related problems*

The problems related to the illness required the patients to handle troublesome situations, to cope, and they did this in many ways. A patient could recount one or different ways to cope. The similar ways to cope were gathered under the heading of a coping strategy. The coping strategies that emerged from the patients’ accounts were labelled in close agreement with their descriptions. In Table 2 the descriptions begin with the most usual coping strategy told of first and then goes on to the others in descending order (Study II).

The most commonly used coping was within the strategy of ‘Searching for a solution’ (Table 2), which was constituted of activity arrangements in order to make problematic events in daily life more manageable. Persons described alternative actions to accomplish mobility by changing body movements, making better use of the wheelchair and using the transportation service for the disabled. An alternative way to keep up a conversation was to use neologisms. Patients told of new ways to remember or compensate for memory problems (Study II).

A coping strategy labelled ‘Refraining from and avoiding’ (Table 2) was to not tell others about one’s own condition. The patients refrained from giving or receiving information and help and refrained from or avoided former activities and habits. Patients also coped by ‘Laughing and joking’. They joked about the tumour and/or its consequences. Sometimes joking was followed by laughter. Laughter was used when the patients could not follow the ongoing talk or showed signs of dysphasia. It also came into use when they spoke about serious problems, negative consequences and frightful experiences in connection with the tumour.
**Table 2** The patients’ coping strategies

<table>
<thead>
<tr>
<th>Coping strategy</th>
<th>Examples of ways to cope</th>
</tr>
</thead>
<tbody>
<tr>
<td>Searching for a solution</td>
<td>Move in another way, use transportation service, take down notes, make neologisms to accomplish flow in the dialogue</td>
</tr>
<tr>
<td>Refraining from and avoiding</td>
<td>Don’t inform others, don’t drink coffee any longer, don’t drive the car, avoid showing feelings, don’t think ahead</td>
</tr>
<tr>
<td>Laughing and joking</td>
<td>Laughed when speech was dysphasic, or when recounting frightful experiences, joked when feeling embarrassed</td>
</tr>
<tr>
<td>Caring about self</td>
<td>Lower demands, saving own resources, allowed to take advantage of things that enhance pleasure</td>
</tr>
<tr>
<td>Re-evaluating</td>
<td>Reminding self of positive things despite disease, more gratitude towards life, even depreciate self after disease</td>
</tr>
<tr>
<td>Giving and seeking information and help</td>
<td>Disseminate illness experience to others, read about the disease, telephone people to get information</td>
</tr>
<tr>
<td>Expressing emotions and thoughts</td>
<td>Being open and expressing fear, sadness and anger, reflections on own condition</td>
</tr>
<tr>
<td>Comparing</td>
<td>Comparing own situation with that of other persons, other times, other outcome of the disease</td>
</tr>
<tr>
<td>Struggling</td>
<td>Physically or mentally fighting against problems (e.g. authorities’ decisions), establishing goal to strive for</td>
</tr>
<tr>
<td>Maintaining hope</td>
<td>Positive beliefs and dreams preserved, trust in the future, the disease regarded as a parenthesis in life</td>
</tr>
<tr>
<td>Accepting</td>
<td>Accept that life is not what it used to be, a restricted life is thought of as acceptable</td>
</tr>
<tr>
<td>Changing line of action</td>
<td>Work, education, interests, life-style were changed or given new goals</td>
</tr>
<tr>
<td>Seeking social affinity</td>
<td>A wish to be close to others, breaking isolation, sharing the illness experience with others</td>
</tr>
<tr>
<td>Reducing</td>
<td>Reducing what was required to attain a former goal meant that life could be lived almost as before</td>
</tr>
<tr>
<td>Anticipating</td>
<td>Being prepared for things that might happen in the future. Think of a tumour recurrence, prepare for own death</td>
</tr>
</tbody>
</table>
The coping strategy ‘Caring about self’ focused on the patients’ own needs, wants and pleasures and it dealt with how to allow yourself to take it easy and lower former demands, ambitions and goals in life (Table 2). Coping by ‘Re-evaluating’ included attention to or reminding oneself of positive aspects of life despite illness. It also encompassed positive side-effects of illness, like being glad about losing weight, as this was needed anyway. ‘Re-evaluating’ was a changed view on things and increased gratitude of life after the diagnosis. Also things appreciated earlier could be depreciated, and even depreciation of self could occur within this coping strategy (Study II).

‘Giving and seeking information and help’ (Table 2) is a coping strategy that could be on the one hand a question of dissemination of knowledge about the illness experience, which might be of importance in specific situations to teachers, neighbours and employers, and on the other hand a question of searching for information. ‘Expressing emotions and thoughts’ was a strategy to overtly express feelings or reflections about the brain tumour and its consequences. ‘Comparing’ was a coping strategy where patients compared their predicament with the situation of other persons, with other times, other situations or other possible outcomes. Mental or physical struggling to stand up to illness-related problems was a coping strategy named ‘Struggling’. The maintenance of positive thoughts, beliefs, hopes and dreams, together with thinking of the brain tumour illness as a parenthesis in life, was illustrative of the coping strategy ‘Maintaining hope’ (Study II).

‘Accepting’ was a coping strategy that involved the acceptance of the reality of life circumscribed by illness. ‘Changing line of action’ was a coping strategy consisting of changing interests, work, educational plans or lifestyle. A coping strategy labelled ‘Seeking social affinity’ concerned a wish to be close to others and break one’s social isolation. ‘Reducing’ was a coping strategy where patients were able to maintain former activities in some mode if they reduced time, space and effort needed. ‘Anticipating’ concerned seeking out, by way of activities, emotions and thoughts, whatever illness-related problems or dangers might lurk in the future (Study II).
**The experience of being the next of kin**

The experiences the next of kin had of their situation in relation to the patients were the special focus in Study III. Most of the next of kin were at the time of the interview living in a situation characterised by the patients’ having neurocognitive, physical and psychological symptoms, and some patients had ongoing medical treatment.

The analysis of the interviews gave rise to four themes underlying the next of kin’s experiences. These themes were: Extremely stressful emotions, Being invisible and neglected, Changed relations and roles, and Enabling strength in everyday life (Table 3).

**Extremely stressful emotions** was a theme that included being ‘Frightened and in a chaotic state’ (Table 3). When next of kin thought about the tumour development or knew that the state of the tumour was unstable and there was a risk of its growing, their emotions were characterised by uncertainty. They also felt ‘Uncertainty’ about negative radiation effects, what the future might be or when the patient should see the physician. There was a sense of being on an ‘Emotional roller-coaster’ because of the terrible time from illness onset replaced by the relief of the diagnosis or finding out the patient’s stable condition, instantly demolished when an epileptic seizure occurred. Being ‘Fragile and susceptible’ had to do with the next of kin’s sense of being at the end of their tether. They were worn-out, suffering from insomnia, reporting sick, and were extraordinarily sensitive to anything that could be associated with brain tumour or its possible poor development. There was a new sense of life’s brittleness, a sense that anything could happen at any time (Study III).

**Being invisible and neglected** was a question of experience that had origin in the next of kin’s encounters in the health-care area and in social life and had to do with its perceived unsupportive relations, unsatisfied needs and the connected feelings of helplessness (Table 3). A ‘Feeling of being left outside’ was expressed in terms of disappointment and sadness that in some cases others failed to provide support. In care contacts the next of kin sometimes felt ignored and insignificant. ‘Unsatisfied needs and feeling of powerlessness’ were experienced in connection with wishes or requests in care. Several next of kin spoke of their
own need for emotional support. When this was not forthcoming they felt disappointment and anger. They also told of the unequal balance of power and their feeling of being the underdog in relation to care institutions. Access to information was spoken of as being insufficient and questions were left unanswered, particularly when it came to the consequences of surgery and the life together with the patient, rehabilitation and the issue of continuous support (Study III).

*Changed relations and roles* was a theme encompassing the closest family but also more distant family, friends and fellow-workers (Table 3). The next of kin’s experience of ‘Support and affinity’ in the form of other people’s practical and emotional support evoked great gratitude in them. There were next of kin that had a strong feeling of being closer to other relatives and friends or fellow-workers, people that now had increased in social importance. The relation between next of kin and patient was in some cases experienced as an increase in physical and emotional closeness.

The patient’s negatively changed behaviour and personality, decreased memory and cognitive skills, fear, inability to perform daily tasks and worsened medical condition were perceived, causing ‘Dependency and restricted freedom’. For some next of kin the dependency elicited resistance to be the patient’s carer and experiences of the patient’s being a burden caused next of kin to perceive themselves as trapped. In some cases the dependency was accompanied by a need to carry out supervision of the patient and also some next of kin spoke positively of the dependency as underpinning a sense of being needed (Study III).

The next of kin’s experience of ‘Changed roles and loss of reciprocity’ had to do with new roles in relation to the patient. There were some next of kin who perceived themselves as single parent (though they were one of two parents) or as a mother (though they were partners) taking care of the patient’s personal tasks like dressing and hygiene. Such changes were described in negative terms, and some female partners who also experienced loss of reciprocity in intimate situations found this so distressing that thoughts of separation appeared. Loss of reciprocity was experienced when those nearest had to be attentive to the patient’s fear and other negative emotions, which restrained their own emotions and behaviours (Study III).
Table 3 Experience of being the next of kin of a person with a low-grade glioma (n=28)

<table>
<thead>
<tr>
<th>Themes</th>
<th>Sub-themes</th>
</tr>
</thead>
<tbody>
<tr>
<td>Extremely stressful emotions</td>
<td>Frightening and Chaotic</td>
</tr>
<tr>
<td></td>
<td>Uncertainty</td>
</tr>
<tr>
<td></td>
<td>Emotional roller-coaster</td>
</tr>
<tr>
<td></td>
<td>Fragile and susceptible</td>
</tr>
<tr>
<td>Being invisible and neglected</td>
<td>Feeling of being left outside</td>
</tr>
<tr>
<td></td>
<td>Unsatisfied needs and feeling of powerlessness</td>
</tr>
<tr>
<td>Changed relations and roles</td>
<td>Support and affinity</td>
</tr>
<tr>
<td></td>
<td>Dependency and restricted freedom</td>
</tr>
<tr>
<td></td>
<td>Changed roles and loss of reciprocity</td>
</tr>
<tr>
<td>Enabling strength in everyday life</td>
<td>Being allowed to participate in care</td>
</tr>
<tr>
<td></td>
<td>Opportunity to suggest improvement in care</td>
</tr>
<tr>
<td></td>
<td>Handling threats and everyday strain</td>
</tr>
<tr>
<td></td>
<td>Positive life values</td>
</tr>
</tbody>
</table>

*Enabling strength in everyday life* had to do with the next of kin’s means of alleviation of strain and the receiving of comfort (Table 3). This theme emanated from experience of the possibilities of participating in health-care situations, proposing improvements for future patients and next of kin, different ways of coping and their positive outlook upon life. ‘Being allowed to participate in care’ concerned the positive encounters when next of kin were treated with respect, invited to participate and enough time was given. ‘Opportunity to suggest improvement in care’ gave next of kin a chance to contribute to improved care and support for future patients and their families. The two main areas for improvement concerned a need for emotional and psychological support and a need for information. In addition there was a request for broader professional teams in care, support after discharge and a key health-care employee to be easily accessible to the patients and their families.

‘Handling threats and everyday strain’ had to do with the next of kin’s coping in stressful situations, especially during the critical time around illness.
onset and diagnosis. The next of kin used a broad repertoire of coping, commonly involving repression of distressing thoughts and emotions. However, they also allowed themselves to overtly express strong emotions of anger, sadness and grief. ‘Positive life values’ had to do with next of kin’s having a positive outlook on life and the ability to be empathetic. Having children and grandchildren, enriching interests and the ability to fulfil dreams — these counterbalanced negative emotions and strain. There were also existential views that made life less burdensome. Faith in the human being own strength to heal, in the future and in God, contributed to preserving the next of kin’s hope (Study III).

Themes related to the next of kin’s sex, the kind of relationship, age and the patients disease duration

The quantitative method was used to explore whether the themes (appearing from the content analysis) were differently frequent among subgroups of next of kin (Study III). The results of the comparisons between the sexes showed that there was a predominance of reported experiences among female next of kin within all themes. Within the theme of Extremely stressful emotions, the parents’ experiences predominated. Other next of kin (live-apart partners, sibling and adult child) generally reported fewer experiences and emotions within all themes (Study III).

The next of kin in the 25–39 age group told of more experiences within the themes of Extremely stressful emotions and Enabling strength in everyday life, whilst those in the 40–59 age group related more within the themes of Being invisible and neglected and Changed relations and roles.

The next of kin were also grouped in accordance with the duration of the patient’s disease. Those with the shortest duration (0–3 years) had more accounts within the themes of Extremely stressful emotions and Enabling strength in everyday life. Those next of kin with a patient’s disease duration of 4–9 years told more of experiences and emotions related to Changed relations and roles and those with a patient’s disease duration of 10–19 years told least within that theme. Across all next of kin groupings, the theme Enabling strength in everyday life corresponded to most statements in the interviews (Study III).
Experience of quality of life: the patients’ and the next of kin’s perspectives

The patients and the next of kin estimated QoL by means of the same questionnaire, the Subjective estimated Quality of Life (SQoL). The next of kin estimated more variables higher than the patients, though the difference was small (Study IV). Both among the patients and the next of kin there were high estimations of SQoL, with only one of the variables (among the patients the absence of work/meaningful occupation and among the next of kin the absence of own children) being estimated below 60% of the maximum score. Even higher values, at least 80% (median 4–5) of the maximum score of 5, among both the patients and the next of kin’ concerned: Housing, Work/meaningful occupation, Relationship to partner, Relationship to friends, Relationship to mother and father, Relationship to own children, Emotional experiences and the variable QoL as a whole. Absence of work/meaningful occupation was the only statistically significant lower estimation in patients’ as compared with next of kin’s (Study IV).

The patient group, SQoL variables in relation to background factors

Within the patient group there were statistically significant differences in SQoL variables as compared with the background factor of sex as male patients estimated Emotional experiences lower than female patients, and female patients estimated Absence of work lower than male patients. The oldest patient group (60–79 years) estimated their Energy and Self-assuredness lowest. Patients with the disease duration 10–19 years estimated Relationship to friends significantly lower than did patients with 4–9 and 20–47 years of disease duration. General mood received a low rating among patients with disease duration 20–47 years and was significantly lower than that of patients with 4–9 years of disease duration (Study IV).

The next of kin group, SQoL variables in relation to background factors

Statistically significant differences in SQoL variables in relation to background factors among the next of kin were that female next of kin estimated their General mood lower than male next of kin, and General mood was also estimated lower by those 40–59 years old than by the oldest (60–79 years) next of kin. The next of kin in the 21–39 years age group estimated Housing quality significantly lower than those in the 60–79 years age group. The next of kin of patients with a
disease duration of 0–3 years estimated Self-assuredness significantly lower than those of patients with a disease duration 4–9 years. Personal security was estimated significantly lower by the next of kin of patients with disease duration 0–3 years than by the next of kin of those with disease duration 4–9 and 20–47 years. Relationship to friends was estimated significantly lower by the next of kin of patients with a disease duration of 10–19 years than by those of patients with a disease duration 20–47 years, a result also found in the patient group (Study IV).

**Brief summary of the prominent results**

- The onset of low-grade glioma was described as a process, either rapid (a few months) or prolonged over several years and encompassed repeated visits to physicians and care institutions.
- The onset phase involved patients’ negatively perceived health-care experience like lack of respectful encounters, lack of medical competence, treatment that did not help and unpleasant examinations, but some perceived positive healthcare experience with respectful encounters, medical competence and physician continuity.
- In care contacts there were common that next of kin perceived themselves powerless, as standing beside and not being respected. However, there were next of kin that had experience of positive encounters in health-care: enough time was given, they were involved in decisions, they felt respected and they got the needed information.
- At illness onset the patient’s everyday life was affected negatively in the form of lack of social support, limitations in social roles, negatively affected education and work, but there was also positive experience like encountering the deep involvement of next of kin and friends.
- The onset of low-grade glioma was accompanied by distress, uncertainty and mortal anxiety for the patients and the next of kin, being characterised by chaos, with fear and worry and a feeling of being worn-out.
The symptoms and problems the patients experienced encompassed a broad range of consequences physical, psychological and social. Among those seldom or never reported previously were changed perception of taste and tactility and skull cavities/deformation and scars after surgery.

The patients presented a wide range of ways to cope with tumour-related problems. Some coping corresponded to previously known coping, other coping had seldom been described before like give information, reducing, anticipating and comparing.

The next of kin’s experience in relation to the patients’ disease varied and among the groups of next of kin there was a predominance of recounted experiences and emotions among females. In addition the parents’ stories predominated within the theme of Extremely stressful emotions.

Relations and roles changed in ways that were experienced as highly negative by the next of kin, being exemplified by the patient’s strong dependency on the next of kin and earlier equal partnership being lost. There were next of kin that were not prepared to take on the role of carer in relation to the patient.

The patients and the next of kin estimated their Quality of Life comparatively high, only one variable (among the patients the absence of work/meaningful occupation and among the relatives the absence of own children) being estimated below 60% of the maximum score.

Significant differences between the patient and the next of kin group in Quality of Life estimations in relation to background variables were more often related to the patients’ disease duration than the variables of sex and age.
DISCUSSION

Result discussion

The term ‘low’ in low-grade or low-malignant is connected to the fact that the tumour cells grow more slowly than in the case of high-grade/malignant tumours and has nothing to do with a possibly subjective connotation of the tumour as being less unfavourable. The results from this thesis show that there was a strong negative psychosocial impact on many of the patients and those close to them. Thus the results verify that the term low-grade glioma may give a deceptive impression of the tumours being harmless, but it is not synonymous with benign (Ashby & Shapiro, 2004). It should be borne in mind, though, that a few patients were scarcely affected by their brain tumour disease.

The early illness period

One might assume that falling ill with cancer is psychologically associated with the horror of receiving the diagnosis. In this thesis it was found that a more expansive illness onset was more often described and the distress encompassed much more than the time of getting an undesirable and frightening diagnosis.

The onset period before the low-grade glioma diagnosis was described from the perspective of the patients and their next of kin. The frightening experience, especially in the case of acute and rapid illness, is comprehensible. On the other hand the prolonged onset with vague symptoms could be going on for years may be more difficult to conceive. In this thesis an initial picture emerged that it is a critical period accompanied by strain, distress, anxiety and the threat of potential loss of life (Studies I and III).

In Study I the patients described repeated visits to the hospital. The time span between symptoms and therapy has been described as fraught with uncertainty and despair in a brain tumour study (Salander, Bergenheim, Hamberg, & Henriksson, 1999), and waiting for test results when cancer is suspected has been described in the literature as a stressful experience (Gurevich, Devins, & Rodin, 2002). A short onset period is the most desired, but there may be circumstances that give cause for waiting for the diagnosis. This may be the case when the symptoms are vague and/or point to alternative causes (Salander et al., 1999). In a cancer study including the pre-diagnostic phase there was
evidence of the spouses’ involvement in taking contact with health-care institutions (Vaartio, Kiviniemi, & Suominen, 2003). Study III in this thesis showed that the low-grade glioma illness appeared to have caused especial distress and turbulence among the next of kin in the case of patients’ earlier illness. Therefore the need for health-care professionals to communicate with patients and those nearest in a respectful way is required already in the pre-diagnostic phase.

**Symptoms, problems and dysfunction among the patients**

Most of the symptoms, problems and dysfunction the patients told of have been reported in other brain tumour studies. Among those seldom or never reported previously were changed perception of taste and tactility and skull cavities/deformation and scars after surgery (Study II). Tinnitus is a rare symptom found in this thesis and has been described in a case study (Alleyne, Hunter, Olson, & Barrow, 1998). Nor has the phenomenon of ‘racing thoughts’ that emerged in connection with illness-onset been reported in brain tumour studies. Racing thoughts, however, has been described in connection with depression (Benazzi, 2003), and several patients in this thesis told of depression, lowered self-esteem or similar emotions that could be an indication of co-morbidity with depression. Interest in depression in brain tumour patients seems to have increased in recent years. Studies have shown that patients with low-grade gliomas with a preoperative depression have worse survival (Mainio et al., 2005) and decreased QoL (Mainio et al., 2006). If a patient is afflicted with a clinical depression it might be confounded with the symptoms of brain tumour disease.

Previous cancer studies have revealed that long-term survivorship (> 5 years holds for 74% of the patients in this thesis) (Bowman, Deimling, Smerglia, Sage, & Kahana, 2003; Gotay & Muraoka, 1998) is associated with more problems than generally expected (Welch-McCaffrey, Hoffman, Leigh, Loescher, & Meyskens, 1989). The results from this thesis confirm that problems of a physical, cognitive, emotional and/or social nature remain several years after diagnosis in the condition of low-grade brain tumours. Therefore we can expect that manifold support during long-term brain tumour survival will be required.
Coping
Study II encompassed exploration of coping among patients with low-grade glioma with an inductive qualitative method. The naming of the different types of coping were held close to the patients’ own wordings and the activity they recounted, and this is one of the reasons that there are differences in naming/labelling in relation to terms on coping scales like the Ways of Coping Questionnaire (Folkman & Lazarus, 1988) and the Mental Adjustment to Cancer (Greer, Moorey, & Watson, 1989).

Some findings not or seldom found in other coping studies were the creation of new words to carry out communication when patients lacked the right word, the use of laughing and joking and anticipating. Coping that included uncommon elements was also found: in ‘Giving and seeking information and help’ included, for example giving information to others about one’s illness. Most often the type of coping described in the literature is the search for information (Case, 2005; Leydon et al., 2000). In addition ‘Seeking social affinity’ was a striving to feel a sense of solidarity (Study II) and not the more commonly search for social support to be found in previous studies (Dunkel-Schetter, Feinstein, Taylor, & Falke, 1992; Hupcey, 1998). The coping strategies ‘Caring about self’, ‘Comparing’, ‘Reducing’ and ‘Anticipating’ encompass content making them difficult to compare with other strategies.

When inspecting the patients’ coping in this thesis it was evident that there were both coping that was directed towards controlling single problems (as can be seen when thinking life is OK living with restrictions) and coping that involved chains of ways of coping (as in ‘Changing line of action’ which requires planning and changing several things for a new goal). Use of the inductive qualitative method to explore peoples coping provides the possibility of producing other kinds of result than those of studies relying on deductive approaches with for instance coping scales.

Coping that was typically forthcoming among next of kin in the early phase was predominantly of repressive type (Study III). It is noteworthy that though these patterns of coping are often referred to as maladaptive their objective depend on the specific situation. Studies have shown it to be an important means to manage if there is no other way out (Case, 2005; Lazarus, 1993; Lazarus & Folkman, 1984).
Much of the fear and stress the patients and the next of kin described in this thesis can be understood by means of the tools Lazarus and Folkman provide in their theory concerning personal and environmental factors influencing appraisal (Lazarus & Folkman, 1984). When you are suffering from a malignant brain tumour it is likely that you will convince yourself that you have no personal control. At thirty-forty years of age (common ages of incidence in low-grade glioma) you generally have a partner, a family with own children and maybe parents still alive, and commonly you have working capacity and maybe are in the middle of a career. There are thus numerous dearly valued factors, commitments, which are at stake for many patients and next of kin.

Environmental factors that are relevant for the understanding of stress among participants in this thesis are events which involve novelty and unpredictability. It is impossible to know whether a new tumour-related problem will occur or to predict the next epileptic seizure. Also the uncertainty concerning disease stability and possible recurrence of the tumour, involving the question of the patient’s ability to perform as before, can explain the high distress. The duration of a stressful situation is important to consider since a brain tumour cannot be cured today and there is an omnipresent risk of being reminded of the disease coming back. The special features of the omnipresent risk of the tumour’s developing into the highly malignant tumour and with this the more years of survival and possibly the accumulation of dysfunctions might be one explanation of the result showing that the low-grade brain tumour patients report at least the same amount of stress as the high-grade brain tumour patients have shown (Keir et al., 2006).

In addition to the problem- and emotion-focused coping functions described by Lazarus and Folkman (1984) and especially in the case of further stress appraisal of loss or threat of a serious disease, the theory of meaning-based coping is put forward (Folkman & Greer, 2000). This coping is found to appear when an unsatisfactory outcome is likely, as in the case of a chronic disease. The meaning-based coping includes forsaking untenable goals and formulating new ones, making sense of/creating a meaning for what is happening, and appraising benefit where possible (Folkman & Greer, 2000). The similarities with the patients’ coping strategies in Study II in this thesis are apparent. Within the coping strategies among the patients can be seen different coping functions,
coping directed towards daily hardships and coping directed towards the omnipresent threat of the incurable disease. The meaning-based coping is the result of an altered point of view in relation to an unavoidable stressor, and keeping a positive outlook on life/maintaining psychological wellbeing is an essential feature of the theory (Folkman & Greer, 2000). Within the patients’ coping strategies (Study II) elements of meaning-based coping were forthcoming in for example ‘Caring about self’, ‘Changing line of action’ and ‘Reducing’, in that they included both appraising the benefit of what is possible and forsaking untenable goals and formulating new ones. ‘Re-evaluating’ and ‘Maintaining hope’ are illustrative of making sense/creating a meaning of what is happening and also appraising benefit where possible (Study II). Study III did not have the main focus on coping, however among the next of kin their recounted experience within the theme of ‘Enabling strength in everyday life’ shows agreement with the meaning-based coping, especially when it comes to handling distress by means of Positive life values (Study III).

The next of kin

The next of kin’s extremely stressful emotions in the patients’ early illness phase need to be recognised as many of the next of kin felt as if they were standing to one side, forgotten and alone in this turbulence (Study III). The shortest disease duration, 0–3 years, was associated with significantly lower SQoL values in Personal security and Self-assuredness in next of kin (Study IV). This finding may be related to the fact that brain tumours have been reported to elicit feelings of uncertainty, the sense of a threat to life and extraordinary stress (Kirshblum, O'Dell, Ho, & Barr, 2001; Newton & Mateo, 1994; Salander, 1996). It can also be discussed whether the Extremely stressful emotions in the early illness phase indicate a crisis. The crisis theory is in the literature described as a process with several phases (Cullberg, 2006). Since this thesis has a cross-sectional design it is beyond its scope to explore the process of crisis but there are similarities between reactions among the participants and traumatic crisis theory (Cullberg, 2006).

There were next of kin that experienced an undesired partner relationship and loss of reciprocity in intimate relations and communication in Study III, and carers of patients with brain tumours have reported significantly greater loss of intimacy as compared to other types of cancer (Gaugler et al., 2005). Partner
relationships were sometimes perceived as being so hurtful that next of kin thought of separation as a solution. The next of kin’s readiness to support the patient may be low in such cases.

The next of kin had a lot of experience of encounters in health-care situations and several of them took the opportunity to talk about this in the interview (Study III). Both from negative and positive experience, it was possible to learn about wishes and requests with clinical implications. The next of kin’s own psychological support needs and need for information were the predominant matter for concern which also has been recognised in other brain tumour studies (Janda, Eakin, Bailey, Walker, & Troy, 2006). In addition they had suggestions as to better care for their patient – the patient should be respectfully met and offered (better) rehabilitation, and the community should assume responsibility for offering further help (Study III).

**Quality of Life**

The comparison of subjective estimated QoL by means of the questionnaire SQoL showed that between patients and their next of kin only Absence of work/meaningful occupation represented a statistically significantly difference, being estimated more negatively by the patients (Study IV). Though this result is based on few participant’ estimates, one can speculate as to wether low estimates in Absence of work mean that having a job or meaningful occupation is especially highly valued among these patients. Work has been identified as a significant factor in QoL among patients with brain tumours and is of great concern in rehabilitation (Bell, O'Dell, Barr, & Yablon, 1998). The SQoL has been used in studies on patients with Muscular Dystrophy (MD) and their family members (Boström & Ahlström, 2005) and patients with Multiple Sclerosis (MS) (Isaksson, Ahlström, & Gunnarsson, 2005). When comparing the low-grade glioma patients’ estimates on the variable Absence of work/meaningful occupation one finds similar values to those of patient groups with chronic diseases. The median value both in MD patients and low-grade glioma patients was 3.0 and in the group of immunology treated MS patients the median value was 2.5. This suggests that more patient groups with long-term or chronic diseases would probably appreciate having a meaningful occupation.
The patients’ disease duration was connected to more significant differences in SQoL variables than factors of sex and age. A plausible explanation is that the brain tumour disease influences subjective QoL in the patients and those nearest (Study IV). The patient disease duration 10–19 years was associated with significantly lower SQoL values in Relationship to friends in the case of both the patients and the next of kin. Those participants were less satisfied with their relationship to friends than were those with shorter or longer disease duration. A possible explanation of this result is that the participants perceived that their friends were at the end of their tether in their supportive, consolatory or pleasing function. In the case of longer disease duration Relationship to friends was estimated higher, maybe indicating that relationship of perceived poorer quality was broken off (Study IV).

One finding was that the patients and the next of kin accorded all except one SQoL variable at least 60% of maximum score. Among the patients four variables and among the next of kin five variables were estimated at ≥ 80% (mean ≥ 4) of maximum score (Study IV). The high estimates are known from other QoL studies (Cummins, 2005; Schmidinger et al., 2003) and SQoL studies (Hugosson et al., 1997; Kajandi, 2006). The patient’s and the next of kin’s comparatively high estimates in SQoL may be explained by changed frames of reference after experience of a disease that may result in a more positive view (response shift) of their QoL (Sharpe, Butow, Smith, McConnell, & Clarke, 2005).

**Method discussion**

There are special ethical issues that have to be considered when interviewing persons in a vulnerable situation (Davies, Hall, Clarke, Bannon, & Hopkins, 1998). Some of the patients participating in this research exhibited varying degrees of expressive aphasia and cognitive problems but still chose to participate to share their experiences. Giving these persons a possibility of speaking can be viewed as particularly important from a research ethics and disability science standpoint. Persons with cognitive impairment are seldom included in qualitative research, a possible reason being that they do not fit “the traditional interviewee profile (e.g. articulate, reflective)” (Paterson & Scott-Findlay, 2002 p 399). Conducting an interview or conversation with people with
this impairment is without doubt possible if the interview or conversation is adapted to the communicative, cognitive or memory-related obstacles.

Another ethical consideration was to let the patient decide whether and which of their next of kin to get into contact with to ask about participation and where to carry out the interview. Whether the different places for the interviews, about half in the participant’s home and half at the authors workplace, had an influence on the result was not specifically studied. The own choice probably means that the participants feel confidence and comfortable independently of the place for the interviews. Qualitative methods and analysis provide insight into experiential aspects of the lives of the patients diagnosed with a low-grade glioma and their next of kin. The participants’ unique stories provided vivid descriptions that brought about a deep understanding of their illness experience. Therefore these studies have an important place in enhancing the understanding of this condition.

In the qualitative studies an endeavour was made to ensure trustworthiness by providing sufficient descriptions of data and procedures for the credibility and transferability of the results to be clear. The data and research process was also continuously scrutinised and supervised by an experienced senior researcher. The growing result was saved at several development stages making inspections possible to meet the trustworthiness criteria of dependability and confirmability (Lincoln & Guba, 1985). The reason for using content analysis was the selection of all eligible patients with low-grade glioma within a well-defined geographical area. This meant a larger number of participants than are usually included in qualitative method studies. In this thesis content analysis was also decided upon for keeping the possibilities open for interpretation at various content levels. Some of the patients gave comprehensive and rich stories and others had more difficulty in expressing their meaning. This means that a method that allowed analysis of both manifest and latent content had the merit of including all participants’ voices in the results. This was considered a matter of trustworthiness (Lincoln & Guba, 1985).

Two of the patients included had glioma WHO grade III were judged by the neurologists analysing the medical records to be appropriate to include. On the follow up on 17 September 2004 these patients were both still alive, 9 and 14
years (respectively) after their diagnosis and in spite of their ascribed grade III. The clinical picture like that of a low-grade can therefore be said to be correct.

In Study I twelve patients were excluded who could not remember the specific occasion when they received their diagnosis. The reason why those twelve didn’t remember the occasion is uncertain. The inclusion and exclusion groups were equal in age and time since diagnosis (the mean difference was two years). Some of them were young at the time of diagnosis and some got their diagnosis a long time ago, both circumstances that make it possible that the patients never experienced an explicit diagnosis occasion. Two differences existed that might suggest an explanation since the included group involved more women and more patients were married/cohabiting. Women have been found to have better autobiographical memory of emotional events (Davis, 1999), and couples have the possibility of sharing their experiences and thereby keeping them in mind.

In Study III there was one difference between patients who included a next of kin and those who did not. In the case of the former there was a shorter duration since diagnosis. This could have biased the results towards the occurrence of more negative emotions among the next of kin, which were mostly found in the early phase of illness. As two next of kin represented one patient there might be a skew dependency. It would most likely have been troublesome to exclude one of these next of kin since both wanted to participate and were equal with regard to their connection to the patient. Including both was judged to be of minor significance, as the data mainly concern a qualitative description of a group of next of kin (Study III). The mix of methods in Study III with the added quantitative analysis led to findings that revealed the differences in the next of kin groups’ amount of statements within the various themes. The emotional distress shown by those nearest might be explained by the predominance of women. This phenomenon has been reported in other cancer studies (Baider et al., 1998; Blanchard et al., 1997; Morse & Fife, 1998; Nijboer et al., 1998), in studies of dementia (Gerdner, Buckwalter, & Reed, 2002) and stroke (Franzén-Dahlin, 2007; Hartke & King, 2002; van den Heuvel, de Witte, Schure, Sanderman, & Meyboom-de Jong, 2001; White, Mayo, Hanley, & Wood-Dauphinee, 2003). Whether the women in Study III actually gave more time to
care-giving tasks (a circumstance that possibly influences distress perception) was not investigated.

In Study IV the choice of the questionnaire SQoL was based on its suitability for both healthy people and patients, as well as on its being available in Swedish. The importance of language and concordant culture in collecting QoL data has been pointed out (Guyatt, 1993; Verdugo et al., 2005). The SQoL had undergone psychometric testing with satisfactory results (Kajandi, 1994). In Study IV the SQoL questionnaire’s internal consistency was measured with Chronbach’s alpha and was 0.81 for the patient group and 0.85 for the next of kin group (Study IV).

The mutually exclusive variables were kept separated because of their incomparable QoL conditions. The estimation of for instance an existent job and its qualities and on the other hand a situation of unemployment and its qualities are measures of two different situations. Keeping those variables separate was also for making comparisons with other study results possible. The finding of significantly lower SQoL estimations on Absence of work/meaningful occupation among the patients as compared to the next of kin would not have been demonstrated with the variable merged. The small number of participants was a shortcoming of Study IV, which has to be taken into account when interpreting the results and which restricts the external validity.

The differences between the patients included in the research and those who declined to participate were that the latter were younger and to a higher percentage women. The mean age of the 10 dropouts, five men and five women, was 37 years (SD 14), range 20–59 years. This needs to be taken into consideration especially in interpreting the result in Study IV where the patients’ sexes and ages were variables related to SQoL.

**Clinical implications**

It is a disheartening result that many patients and next of kin found encounters with professionals in care so negative. The experienced lack of or inappropriate information is not in any way a new finding (Davies & Higginson, 2003; Eriksson & Lauri, 2000; Kent et al., 1996). This does not automatically imply that professionals have not improved their skill in this respect. Being a patient or the patient’ next of kin nowadays is different than in earlier years. The available
information has expanded and is easy of access on the Internet for most people in the Western countries and maybe more patients and next of kin nowadays ask to be on an equal footing with health-care staff. Therefore if the professionals in care are to be able to meet these new challenges there is a need for improving methods. Some experiences and suggestions in this thesis may serve as an inspiring source of development in the direction of communication (built upon established relations, being mutual and equal, and bearing on what information is needed at the moment).

Waiting for test results when cancer is suspected is a stressful experience; nevertheless there might be circumstances that give cause for waiting for the diagnosis. Physicians can explain why the waiting is necessary and thereby in the best lower the patients’ and the next of kin’s level of distress. The special nurses at some hospitals in Sweden that uphold function as key coordinators in the care for brain tumour patients and their next of kin serve a purpose and have been reported welcome by brain tumour patients and next of kin in this thesis and other studies (Janda et al., 2006; Spetz, Henriksson, Bergenheim, & Salander, 2005).

There are several similarities between high-grade glioma and low-grade glioma. However, there are two differences that are of special interest from a psychosocial perspective, the greater prospect of longer survival and the risk of recurrence bringing a more highly malignant brain tumour. The omnipresent risk that the tumour will recur and become highly malignant one is something which the patient and those nearby have to live with and that denotes long-term distress, great uncertainty and an uncontrollable situation that makes people vulnerable (Lazarus & Folkman, 1984). Many years of survival with remaining disabilities may well put a burden on next of kin. Rehabilitation and habilitation to support the patient and the next of kin might be needed. A long survival might require different support at different times and also from different professionals in care and the community.

Support after the in-patient period could be for instance rehabilitation and compensatory aids, even so it is especially complex in neurorehabilitation (van den Broek, 2005) and brain tumours (Mukand, Guilmette, & Tran, 2003).

The importance of offering support to the patients and the next of kin through a multidisciplinary team including cognitive, vocational and
psychological rehabilitation has previously been suggested (Kibler, 1998; Meyers & Boake, 1993). Compensatory strategies and aids, re-learning and training provide different methods of rehabilitation for brain tumour patients in the outpatient post-medical phase and can be guided by professionals like nurses, occupational therapists, physiotherapists, psychologists, social workers and speech therapists. Educative and psychosocial support should be offered within rehabilitation to both the patients and the next of kin. Minimising the patient’s disability includes that rehabilitation has to reach beyond the patient and take the environment into account.

The feeling of isolation and missing work displayed among the patients (Study II) and low estimates in Absence of work (Study IV) most likely be prevented by plans to return to the workplace. Even if the patient’s ability to regain their former work lacking, there are important psychosocial gains in the contact. Also work may encompass a variety of duties and along with vocational rehabilitation the return to a workplace could enhance QoL for these patients. Inability to work has shown negative effects on QoL in brain tumour patients (Bell et al., 1998).

There is also a need to identify if - and if so what kind of - family support is available at discharge (Bohan, 2002), a consideration reflecting the social and familial difficulties displayed in this thesis. Though most of the patients in this thesis were cohabiting, those living on their own may not have familial support available, and this needs particular notice. Special attention also has to be paid to the next of kin in early phases of the brain tumour illness, who experience a distressed relationship and a high level of patient dependency. In addition a distinction has to be made between male and female next of kin’s need of support. Not forgetting adult patients’ parents who recounted deep involvement but also experience great distress.

**Implications for further research**

Patterns of communication between patients and family members, patients and health-care staff and families and health-care staff need to be further investigated. There are examples of intervention studies in communication training and programmes for health-care staff that can improve the quality of encounters in care (Maguire & Pitceathly, 2002; Tiernan, 2003).
The shortage of psychosocial perspectives in brain tumour studies force one to get references in the much larger field of cancer literature and research. Reliance on cancer studies can in some cases be problematic. The large groups of prostate, breasts and lung cancer commonly characterise cancer studies. The specific additional characteristics in brain tumours like influence on cognition, communication and behaviour may (with references from these studies) run the risk of being overlooked. Therefore psychological and psychosocial research concerning patients with a brain tumour diagnosis and their next of kin might count more upon kindred studies in for instance diagnosis of stroke, traumatic brain injury and intellectual disability.

Work rehabilitation seems to be a matter of concern for patients’ QoL; however the issue of returning to work requires more research (Bell et al., 1998; Huang, Wartella, Kreutzer, Broaddus, & Lyckholm, 2001). In studies on traumatic brain injury prominent obstacles to returning to work were for instance inattention and impaired memory (Shames, Treger, Ring, & Giaquinto, 2007), also dysfunctions that may remain in brain tumour patients.
SAMMANFATTNING PÅ SVENSKA/SUMMARY IN SWEDISH

När någon drabbas av en svår sjukdom så innebär det konsekvenser både för den som drabbas och dennes närståendes vardagsliv. Psykosociala aspekter är sparsamt beskrivna i vetenskaplig litteratur när det gäller lågmaligna hjärntumörer. Studier som rör hur patienter som drabbas av lågmaligna hjärntumörer och deras närstående upplever sin situation och vad sjukdomen får för psykologisk och social betydelse i det fortsatta livet saknas nästan helt. Syftet med den här avhandlingen var därför att bidra med ökade kunskaper om vad det innebär att få och leva vidare efter diagnosen lågmalign hjärntumör utifrån de erfarenheter och upplevelser som patienterna som drabbats och deras närstående förmedlar.


Studierna som ingår i den här avhandlingen genomfördes med kvalitativa och kvantitativa forskningsmetoder. Studie I och II har en kvalitativ analysmetod, Studie III både kvalitativ och kvantitativ analysmetod och Studie


Symtomen och problemen som av patienterna beskrevs som en konsekvens av hjärntumören var mycket varierande och rörde fysiska, kognitiva, emotionella och beteendeapecter och påverkan på flertalet livsområden. Företrädesvis påverkades familjen, arbetet och utbildningsmöjligheterna, ekonomin, vänskapsrelationerna samt fritiden. Vanliga fysiska symtom som patienterna upplevde var balanssvårigheter och muskelsvaghet. Kognitiva problem som ofta beskrevs var koncentrationssvårigheter och ökad uttömbarhet samt minnesproblem. Mycket framträdande var svårigheter att minnas det som nyligen hänt eller lärt in, men även att minnas tider, platser och att minnas något som ska ske eller göras längre fram i tiden beskrevs som påverkat. Patienterna

Avhandlingen visar en rik variation av att hantera (coping) de problemsituationer som uppkom som ett resultat av hjärntumörsjukdomen. För patienterna var de vanligaste sätten att söka efter en praktisk lösning och att avstå ifrån/undvika. Att använda humor och skratt var också ett vanligt hanteringssätt. Mer sällan eller inte alls beskrivna copingsätt var till exempel att jämföra eller föregripa händelser. Hög stress som de närstående upplevde särskilt i sjukdomens tidiga skeden hanterades vanligtvis med undertryckande av de starka negativa känslorna.


De positiva krafterna kunde också ses i deltagarnas höga livskvalitet. Endast en av 18 variabler värderades under 60 % av maximalt värde, för patienterna var det ‘Avsaknad av arbete/meningsfull sysselsättning’ och för de närstående var
det ‘Avsaknad av egna barn’ som uppvisade de lägsta värdena. Det var inga påtagliga skillnader mellan hur patienterna och de närstående värderade sin livskvalitet. Endast en livskvalitetsvariabel visade på signifikant skillnad, patienterna värderade Avsaknad av arbete/meningsfull sysselsättning lägre än de närstående. För både patienter och närstående var de högst värderade livskvalitetsvariablerna; boendet, arbetet, relationerna till partner, vänner, föräldrar och till egna barn, förmåga till känslomässiga upplevelser samt variabeln livskvalitet som helhet.


ACKNOWLEDGEMENTS

I owe my greatest thanks to all the participants in this research. Without your willingness to share your experiences of good and bad in respect of what the brain tumour diagnosis entails, I would have been badly off.

I should like to thank you, my main supervisor, Professor Gerd Ahlström, for your guidance, competent advice and deep involvement throughout these years. My meeting with you and the neurologist Anneli Pählson who planned a research project on brain tumour patients and the next of kin was the origin of my doctoral journey. The project concerned these groups’ psychosocial situation and perspective suited me well, and I joined the project.

Without the financial support from the Centre for Rehabilitation Research, Örebro County Council, I wouldn’t have accomplished this thesis. The Centre has partly been my work place and I thank all of you working there and sharing coffee breaks, lunches and of course research discussions with me.

Thanks to Anneli Pählson at Örebro University Hospital, co-author and who together with Anja Smits at the Department of Neurology, Uppsala University Hospital processed the selection of patients and scrutinised the medical records, which has been of vital importance for my work with this thesis. I also thank Lena Ek, part of a low-grade glioma research group, for our joint lunches and interesting conversations.

Thank you, colleagues, doctoral students and friends at the Swedish Institute for Disability Research, the School of Health and Medical Sciences at Örebro University and also the Psychiatric Research Centre and the Centre for Health Care Science at Örebro University Hospital for interesting as well as relaxing discussions. I cannot mention all of you from these institutions but I have to call attention to my assistant supervisor Jerker Rönnberg, my collaborators Ann-Kristin Wideheim and Margareta Gustafsson.

Over the years there have been seminars and involved people have inspected my manuscripts, encouraged me and shared their knowledge with me, and I offer my thanks to Marianne Omne-Pontén, Birgitta Andershed, Birgitta Sjöqvist-Nätterlund, Katrin Boström, Stig Wenneberg, Lars-Olov Lundqvist, Ann-Kristin Isaksson and Madeleine Abrandt-Dahlgren.

Thank you all who have supported me with administration of various kinds: Agneta Klintenäs, Birgitte Kolsung and Katarina Perälä at Örebro University,
Helén Johansson at Linköping University, Karin Lobenius and Leena Pettersson (Leena for transcribing my interviews) at the Centre for Rehabilitation Research. Thank you Malcolm Forbes for making improvements to my poor English writing. Thank you Margareta Landin, chief librarian at the Medical Library, Örebro University Hospital, for supporting me with reference styles and programs and to you and your colleagues for finding books and articles I hunted for.

Thank you my former chiefs at the Centre for Adult Habilitation Ingvar G Wahlström and Tomas Ring, for your support in my strivings to make the first move towards the doctoral studies, and thank you my present chiefs Karin Greve and Marie Johansson for approving my being on leave of absence for continuing the doctoral studies.

To my team colleagues at the Centre for Adult Habilitation I offer thanks for tolerance of my being absent and sometimes an obstacle in the customary work schedule.

I finally express my gratitude to my parents Aslaug and Bengt and my children Linda and Lars for being the greatest part of my private life and reminding me what’s most important in my life.
REFERENCES


Davies, E. A., Hall, S. M., Clarke, C. R., Bannon, M. P., & Hopkins, A. P. (1998). Do research interviews cause distress or interfere in...


associated with use for >10 years. *Occupational & Environmental Medicine, 64*(9), 626-632.


Pelletier, G., Verhoef, M. J., Khatri, N., & Hagen, N. (2002). Quality of life in brain tumor patients: the relative contributions of depression, fatigue,
emotional distress, and existential issues. *Journal of Neuro-Oncology, 57*(1), 41-49.


25. Edvardsson, Tanja (2008). *Consequences of brain tumours from the perspective of the patients and of their next of kin*.