Chronic sorrow and quality of life in patients with multiple sclerosis
To my beloved family
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Ann-Kristin Isaksson

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Abstract


The overall aim of this thesis was to increase our understanding and knowledge of patients’ experiences of living with multiple sclerosis (MS). A mixed-method design was used, including both qualitative and quantitative methods. Thirty-one immunologically treated MS patients were randomly selected and matched with patients without immunological treatment. Matching criteria were gender, impairment, time since diagnosis and age. One patient dropped out and therefore the final sample consisted of 61 patients. All 61 were interviewed (Studies I, III and IV) and completed the 36-item Short Form health survey questionnaire (SF-36), the Subjective estimation of Quality of Life questionnaire (SQoL), the Self-reported Impairment Check-list (SIC) (Study II) and the Montgomery–Asberg Depression Rating Scale questionnaire (MADRS) (Study III). The interviews were subjected to content analysis.

Initial symptoms and being diagnosed with MS were described in terms of becoming vulnerable and remaining in that vulnerability long after the diagnosis. Eventually the patients were able to manage this emotional distress and acquired strength in their illness situation (Study I). In the matched analysis, 29 pairs of patients were included after internal drop-out. There were no statistically significant differences between the treatment and control groups. Concerning the total group of 61 patients, the self-reported impairment check-list showed that they had various problems of impairment, most evident in balance and walking. The score on health-related quality of life (SF-36) was reduced, disclosing a negative influence on vitality, physical role and function. However, subjective quality of life (SQoL) was not reduced, showing that the patients estimated their well-being to be quite unaffected (Study II). Thirty-eight of the 61 patients (62%) experienced chronic sorrow (Study III). The group were not depressed in general, only four having mild symptoms of depression, revealing that chronic sorrow is a particular form of emotional distress in MS. Chronic sorrow was experienced in terms of loss of hope, loss of control over the body and loss of integrity and dignity. When the patients could not adequately manage their sorrow and lacked support, they were struggling with their vulnerability. In the successful management of MS, losses and emotional distress are managed in such a way as to enhance personal growth, appreciation and trust in life. The theoretical model of chronic sorrow facilitated the sorting of the empirical data and the linking of these data to theory, showing the usefulness of the model (Study IV).

Key words: Multiple sclerosis, illness experience, diagnosis experience, impairment, health-related quality of life, subjective quality of life, immunological treatment, chronic sorrow, depression, managing chronic illness, content analysis, well-being.

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Original Publications

The present thesis is based on the following four studies, which will be referred to in the text by their Roman numerals:


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**Abbreviations**

- **MS**: Multiple Sclerosis
- **EDSS**: Expanded Disability Status Scale
- **QoL**: Quality of life
- **HRQoL**: Health-related Quality of Life
- **SF-36**: The 36-item Short Form health survey
- **SQoL**: Subjective estimation of Quality of Life
- **SIC**: Self-reported Impairment Check-list
- **MADRS**: Montgomery–Asberg Depression Rating Scale
- **NCRCS**: Interview-guide of the Nursing Consortium for Research on Chronic Sorrow
- **MRI**: Magnetic Resonance Imaging
Introduction

In a historical perspective, the knowledge of multiple sclerosis (MS) started with the observations of the French neurologist Charcot (1825-1893). He observed his housekeeper becoming more and more disabled. He described her disease-development and the finding of several hardened indurations in the central nervous system after her death. Such development was also found among patients with similar symptoms and gave rise to the term multiple sclerosis.

There is no curative treatment available for MS. During the last 20 years new medical treatments have been developed, but the prospect of cure still lies in the distant future. Most treatments reduce the progression of the disease, although the patient still has to live with symptoms and — in more severe cases — a reduction of physical capacity.

There has long been recognition in Sweden of the importance of support from the providers of health care with regard to rehabilitation after a bout of MS or its progression (Fagius & Aquilonius, 2006). Patients are being supported through different interventions performed by multidisciplinary MS team comprising nurses, physiotherapists, occupational therapists, social workers and doctors. However, there are local differences with respect to gaining access to these interventions. Swedish neurologists established an expert group in the mid 90s for the purpose acquiring greater knowledge of MS treatments, keeping up to date with international developments and to reducing the inequality of access to care across the country. Nurses were invited to participate in this, and did so; and national guidelines regarding MS care have been drawn up (MS-Association, 2006).

In the mid 80s many patients were hospitalized during medical investigation and periods of rehabilitation were offered at the hospital in-patient clinic. Nowadays investigation and diagnosis is conducted at the out-patient neurological clinic and rehabilitation is often conducted at the out-patient physiotherapeutic clinic. With the availability of immunological treatment in the mid 90s, finally there was a treatment to directly reduce disease development and not only treat symptoms. MS nurses were educated to teach patients how to self-inject since the treatment is given by injection. Treatments were reducing the disease activity but had side-effects that were sometimes
troublesome for the patients. Thus these treatment options have been positive for the patients, offering a chance to slow down the progression of the disease, but have also given rise to new problems. Working as an MS nurse, my curiosity as to the benefits of these immunological treatments in terms of quality of life was awakened. Living with a chronic progressive disease can give rise to distress at the loss of physical function and at the prospect of such loss. This distress was revealed in my encounters with MS patients. Many were sad and worried about their situation.

This experience gave me an insight into the importance of having increased knowledge of the entire illness trajectory: from the patient’s falling ill, to his or her receiving the diagnosis, to his or her subsequently having to live with and manage MS. A wish for more knowledge and a curiosity about the patients’ own experiences has led to this research project that hopefully will improve MS nursing.
Background

Disease description of multiple sclerosis

Multiple sclerosis (MS) is a neurological disease with inflammatory episodes in the central nervous system. This inflammation causes a variety of symptoms. Initial episodes of relapses lead, in general, to full recovery. Later relapses can lead to lasting impairment. In addition, MS can develop a secondary progression disease pattern years later. However, 20% of patients experience a primary progressive form of MS with progression starting from the initial symptoms (Compston & Coles, 2002). The disease usually debuts when the person is 20-40 years (ibid) and it is the leading cause of disability in young adults (Olek & Dawsson, 2000). The cause of MS is not fully determined, though it is acknowledged to be an interplay between genetic and environmental elements — factors such as viruses been discussed, for example, since genetics alone cannot explain the occurrence (Compston & Coles, 2002). There is a hereditary component involved: there is higher risk for monozygotic twins (35%) than for siblings (3%), and higher risk for children of married couples both with MS (20%) than for children just one parent is affected (2%) (Compston & Coles, 2002). MS is assumed to be an autoimmune disease since it affects twice as many women as men (ibid). The prevalence of MS in Sweden is estimated to be 100 individuals per 100,000 (Fagius & Aquilonius, 2006) but the geographical distribution is uneven (Landtblom, Riise, & Kurtzke, 2005). The neurologists arrive at the diagnosis by means of a predetermined criterion: a minimum of two events with symptoms affecting different anatomical sites in the central nervous system, confirmed by magnetic resonance imaging (MRI) and spinal fluid analyses (Compston & Coles, 2002; Poser et al., 1983).

The inflammatory episodes in the central nervous system give rise to symptoms. The location of the inflammation can vary and consequently the symptoms of the disease can vary too. Sensory disturbance, fatigue and reduced physical function are common impairments associated with MS (Olek & Dawsson, 2000). Other symptoms can be optical neuritis, bladder dysfunction, heat sensitivity, sexual dysfunction, cognitive problems, balance problems and co-ordination problems (ibid.). In the disease development neurologists follow the patients with ratings of disability through the employment of the Kurtz Expanded Disability Status Scale (EDSS) (Kurtzke, 1983). EDSS gives an estimate of disability for MS patients in terms of 20
possible outcomes. It scores from 0=normal neurological status, to 10=death from MS. MS can cause physical losses of different kinds, but also cognitive impairment and fatigue, which all can reduce working capacity as well as influencing the patients’ possibilities of functioning in their family and social environment. The progressive nature of MS does not just imply having to adapt to an impairment or disability resulting from the disease, it also implies having to live with uncertainty and the threat of increased disability brought about by a new bout or progression (Compston & Coles, 2002). Consequently the occurrence of depression in MS patients is higher than in other chronically ill patients (Siegert & Abernethy, 2005; Olek & Dawsson, 2000). Recent research found moderate to severe depression in 28% of MS patients (McGuigan & Hutchinson, 2006), and lifetime prevalence is reported to be 40-60% (Wallin, Wilken, Turner, Williams, & Kane, 2006). Clinical depression includes psychological deficits as well as the manifestation of psychosomatic symptoms such as cognitive impairment, changes in sleeping patterns and fatigue, which also can occur as symptoms of MS (Mohr & Cox, 2001; Ptiton-Vouyovitch et al., 2006; Williamson, 2000).

Disease-modifying therapies regarding MS to reduce relapse have been used since the mid 90s (Goodin et al., 2002). The goal of all immunological treatments is to reduce the inflammatory process in the central nervous system, leading to fewer relapses as well as a reduced amount of axonal injury as shown by MRI (Dhib-Jalbut, 2003; Narayanan et al., 2001). The most common medical treatments are based on the beta interferons, interferon β-1a (PRISMS, 1998) and interferon β –1b (Kappos, 1998; Paty & Li, 1993). These interferons have been shown to reduce the relapse rate by 30–37% in placebo-controlled studies (Compston & Coles, 2002). Side-effects at the beginning of the treatment can be flu-like symptoms such as headache, muscular pain and fever, and also redness at the injection site (Arnoldus et al., 2000). Glatiramer acetate also has immuno-suppressive properties in the same range as interferon, the side-effects being indurations and erythema at the injection site but no flu-like symptoms (Dhib-Jalbut, 2003). Treatment with mitoxantrone is available for patients with rapidly deteriorating MS but as a chemotherapeutical agent it can have severe side-effects, even though it has been well tolerated by MS patients (Myers, 2001). There are also other immunological therapies less frequently used and with varying
evidence (Bryant, Clegg, & Milne, 2001), and steroids can be used in cases with severe exacerbations (Goodin et al., 2002).

Pharmacological treatment for symptom management can be of good effect. Many of the neurological deficits caused by MS can be treated, with subsequent beneficial effects such as the reduction of pain or spasticity (Compston & Coles, 2002). Depending upon the type of neurological disorder, bladder dysfunction can be treated with pharmacological agents or through self-catheterization. Treatment of depression can be with either antidepressants and/or psychotherapies (Siebert & Abernethy, 2005; Wallin, Wilken, Turner, Williams, & Kane, 2006). It is also important to include a multidisciplinary management of the symptoms through physiotherapy, occupational therapy and social and nursing interventions (Compston & Coles, 2002). Many patients need support during the process of adjusting to their illness and possible disability. Nurses should provide support for the patient and the family. Nurses can also mediate contact with a counselor or other professional in the multidisciplinary team (Barker, 2002). The patients can benefit from intense rehabilitation after a bout or disease progression (Freeman, Langdon, Hobart, & Thompson, 1999). As described below, living with a chronic progressive disease may affect the patients in many different ways.

Living with MS

Living with MS is living with adjustment to both physical and psychological difficulties brought on by the disease. The first symptoms can be very scary and the afflicted persons may believe them to be those of a life-threatening disease like a brain tumour (Johnson, 2003). Receiving the diagnosis can be a relief but also a negative emotional experience. The emotional burden of anxiety and distress has been found to be high in the early phase of the disease, also among those with minimal disability, and occurring in both patients and their partners (Janssens, van Doorn, de Boer, Kalkers et al., 2003). MS being a progressive disease with an unpredictable prognosis, experiences of uncertainty are often reported by MS patients as being a major source of emotional distress (Williamson, 2000; Gulick, 2001). The elements of uncertainty have a negative influence on the psychological adaptation to the disease (McNulty, Livneh, & Wilson, 2004). Consequently depression is recognized as a common psychological disturbance in MS (Janssens, van Doorn, de Boer, Kalkers et al., 2003), though anxiety is more
common than depression (Feinstein, O’Connor, Gray, & Feinstein, 1999), alone or in combination with depression. The psychological impact, including anxiety and anger, should be recognized by health-care staff (Mohr & Cox, 2001; Mohr et al., 1999). Investigation of the psychological impact of MS showed that patients reported increased distress. However, there has also been a report of MS patients’ positive reappraisal of their situation in the form of deepening relationships and increased appreciation of life (Mohr et al., 1999).

The patient can live a rather normal life between periods of relapses. Nevertheless, living with MS requires coping with symptoms during relapses and the lasting impairment and progression towards increasing disability. Sensory disturbance may not affect performance but is sometimes connected with pain which can be bothersome for the patient if not successfully treated pharmacologically (Henze, 2005; Olek & Dawsson, 2000). Patients commonly experience physical weakness and/or intense muscular strain in the legs and problems with unsteadiness (Olek & Dawsson, 2000), which interferes with walking. Decreased mobility can affect family life, leisure activities, social commitments and working capacity. Cognitive impairment can also interfere with the ability to continue to work (Mohr & Cox, 2001). One other reason for not being able to continue working is fatigue, one of the most troublesome symptoms of MS and interfering with daily activities (Flensner, Ek, & Soderhamn, 2003). Fatigue is a loss of energy, both physical and mental, and has a negative effect on the patient. The draining of energy makes it periodically impossible for the patient to do anything, he or she simply requiring absolute rest (Flensner, 2005).

MS can cause a variety of impairments. Some impairment is visible (e.g. ataxia), other impairment less visible (e.g. visual disturbance, bladder dysfunction or sexual dysfunction). Physical disabilities and change of bodily image (function), involving for example walking with a stagger or using an assistive device, require psychological adjustment. Symptoms reveal the illness to people and the patient cannot chose to inform or not inform others. In Western society appearance is important and visible signs of disability can lead to stigmatization (Joachim & Acorn, 2000). MS can have an impact on family relations, roles can change and with progression of the disease the care-giving burden can increase (Williamson, 2000). Nurses and other health-care staff should support the patient and the family in the adjustment process and encourage them to seek knowledge of MS and its effects. It is important to give
information to the patient and the next of kin about MS so that they can understand the illness and remain in control over their lives and deal with uncertainty (ibid).

Nurses and other health-care staff should provide the latest available information about interventions or treatments directed towards symptom management as well about immunological treatment. A multidisciplinary team with different health-care professionals will be able to optimally match the treatment needs of the patient (Compston & Coles, 2002). Rehabilitation is important to restore function after a bout of MS or progression of the disease. In-patient rehabilitation has been shown to improve physical function as well as mental health, and benefits lasted up to nine months, showing the need of continuity in rehabilitation (Freeman, Langdon, Hobart, & Thompson, 1999).

Quality of life

Quality of life is a multidimensional concept where the meaning depends on the theoretical perspective and the context in which it is used. Health-related quality of life includes aspects of health and quality of life which are influenced by disease. Areas generally included are physical function, emotional health and social interaction (Murrell, 1999). The concept of health-related quality of life captures aspects of function that influence health status and are useful in interventions to improve care, research, clinical practice or decision-making in health policy when it is important to identify poor physical or mental health (Benito-Leon, Morales, Rivera-Navarro, & Mitchell, 2003). Health-related quality of life has been investigated in MS patients in many studies since 1992 (ibid). Research has shown that patients with MS experience lower health-related quality of life than the general population and other groups of patients such as those with epilepsy or diabetes (Hermann et al., 1996). A frequently used generic health-related scale when it comes to patients with MS is the MOS 36-item Short-Form health survey (SF-36) (Nortvedt & Riise, 2003). The SF-36 scale is for self-administration and assesses eight health concepts (Ware & Sherbourne, 1992). Physical health and role function, social function and mental health are included. There is also ongoing development of MS-specific health-related scales that include such factors as cognitive problems or sexual function (Nicholl, Lincoln, Francis, & Stephan, 2001). Health-related quality of life is mostly used in research but it can also be used to evaluate medical care, rehabilitation and nursing interventions for MS patients.
The impact of MS on quality of life may go beyond the impairment and functional capacities (Kilian, Matschinger, & Angermeyer, 2001), and it is therefore also important to investigate the effects on the psychosocial and emotional domains in the form of experienced well-being (Murrell, 1999). There are indeed quality of life instruments that focus on the subjective sense of well-being (Benito-Leon, Morales, Rivera-Navarro, & Mitchell, 2003). Quality of life in the form of well-being is a person-centred approach with subjective interpretation (Murrell, 1999). Subjective quality of life contains all aspects of well-being, including social, emotional, economic and cultural values (Benito-Leon, Morales, Rivera-Navarro, & Mitchell, 2003). Subjective quality of life can also be seen within an interactionistic perspective including the psychological and interpersonal aspects of well-being (Kajandi, 1994). Here the patients estimate their experience of well-being on the basis of their satisfaction with their own situation and without focusing on illness or disability (Kajandi, 1994; Kajandi, 2006). During the process of adaptation to disability the patient’s value system is likely to change and he or she regards the consequences as less important to well-being (Murrell, 1999). Furthermore, mood can also affect patients’ subjective quality of life and it can be negatively influenced by depression and should thus be estimated by means of a depression rating scale (Siegert & Abernethy, 2005; Wallin, Wilken, Turner, Williams, & Kane, 2006). Another manifestation of emotional distress connected with MS and described in the literature is chronic sorrow.

Chronic sorrow
Psychological distress has been described in the case of MS patients, one form such distress being chronic sorrow (Mohr & Cox, 2001). Olshansky first described chronic sorrow in 1962 in the case of parents of mentally deficient children. He recognized that the parents were suffering from sorrow that varied from time to time. He concluded that chronic sorrow is a natural, rather than neurotic, response to a tragic fact. If health-care providers can accept this they may be more effective in helping the parents of a mentally deficient child to cope (Olshansky, 1962). Mary L. Burke wrote her thesis (1989, unpublished) on chronic sorrow in mothers of school-age children with a myelo-meningocele disability. Together with her colleagues Lindgren, Hainsworth and Eakes, she developed this concept into a model also encompassing chronically ill adults and those caring for them (Burke, Hainsworth, Eakes, & Lindgren, 1992). In addition
they developed the instrument Burke/NCRCS, a semi-structured interview guide for chronically ill individuals with a caregiver version (Burke, Hainsworth, Eakes, & Lindgren, 1992). The questions are designed to determine the occurrence of chronic sorrow, milestone events in which it recurs, factors that are helpful or not helpful in coping with this type of emotional distress, advice for others affected by recurrent losses and directives for nurses and other professionals caring for individuals and families experiencing chronic or life-threatening conditions (ibid). Since the 1990s chronic sorrow has been recognized in patients with chronic diseases such as multiple sclerosis (Eakes, Burke, Hainsworth, & Lindgren, 1993; Hainsworth, 1994), cancer (Eakes, 1993) and Parkinson’s (Lindgren, 1996), also in spouses of patients with multiple sclerosis (Hainsworth, 1996) and Parkinson’s (Lindgren, 1996) and furthermore in wives of chronically mentally disabled men (Hainsworth, Busch, Eakes, & Burke, 1995). One of the researchers in this group also did a study on parents with chronically mentally ill children (Eakes, 1995). The research team of Burke, Hainsworth, Eakes and Lindgren continued their work and developed in the end a middle-range theory of chronic sorrow (Eakes, Burke, & Hainsworth, 1998; Schreier & Droes, 2006).

Definition of chronic sorrow

Chronic sorrow can be defined as a pervasive sadness that is permanent, periodically intense and progressive in nature. The individual experiences ongoing loss, due to disability, chronic illness or a relative’s illness (Burke, Hainsworth, Eakes, & Lindgren, 1992). The defining critical attributes of chronic sorrow as explicitly described both in the concept development and in the presentation of the middle-range theory are the following: a perception of sadness or sorrow over time in a situation with no predictable end; sadness or sorrow that is cyclic or recurrent; sadness or sorrow that is triggered internally or externally and brings to mind a person’s losses, disappointments or fears; sadness or sorrow that is progressive and can intensify (Burke, Hainsworth, Eakes, & Lindgren, 1992; Eakes, Burke, & Hainsworth, 1998)

Additionally, Susan Roos (2002) offers a thorough discussion of chronic sorrow. Her definition is in agreement with that of Burke and colleagues, with the addition that chronic sorrow includes the loss of self-image, involving the loss not only of perceived reality but also of what was dreamt of. The loss is ongoing since the
source of the loss continues to be present. The loss is a living loss — it is a question of living with the realization that the loss cannot be removed and requires continuing re-adaptation.

Chronic sorrow is not depression. Feelings of depression can accompany chronic sorrow but there are distinctive differences (Burke, Hainsworth, Eakes, & Lindgren, 1992; Mayer, 2001). Chronic sorrow is a natural reaction as a response to ongoing loss or multiple losses over time. It does not interfere with daily function and can be permanent or periodic. Depression is a pathological state, with psychological and psychosomatic components. It is a mood disturbance and can lead to decreased initiative, reduced concentration or change in sleep pattern (Burke, Hainsworth, Eakes, & Lindgren, 1992; Siegert & Abernethy, 2005). Chronic sorrow is distinguished from prolonged grief, which is a long-term reaction to a single event (Burke, Hainsworth, Eakes, & Lindgren, 1992).

The Middle-Range Theory of Chronic Sorrow

The theory covers loss experiences, ongoing or single event, and how the perceived disparity brings feelings of sorrow (Figure 1). The sorrow is pervasive and permanent and the sadness recurs periodically and is potentially progressive in nature (Schreier & Droes, 2006). The sadness can be triggered by remembering the loss. In developing the theory of chronic sorrow the research group incorporated Lazarus and Folkman’s work on stress and coping (Lazarus & Folkman, 1984). They chose to name the coping strategies used as management methods which are more or less successful (Eakes, Burke, & Hainsworth, 1998). In this theory, attempts by the individual to cope are termed internal methods of managing the situation, while different forms of help and support from health-care professionals are termed external (Figure 1) (Eakes, Burke, & Hainsworth, 1998). Effective management leads to increased comfort and ineffective management leads to discomfort or increases the chronic sorrow. Effective internal management includes maintaining activities and interests, seeking information, adopting a positive attitude and taking one day at a time (ibid). Effective external management includes the recognition of the patient’s sorrow and the offering of support and encouragement, also empathy (Eakes, Burke, & Hainsworth, 1998; Schreier & Droes, 2006). Coping in terms of a process is not further discussed in this
middle-range theory except for an implicit interpretation of the successful management of chronic sorrow as being a part of the individual’s experience of comfort. For those individuals experiencing loss and sorrow, managing efficiently could be a vital part of the process of coping with chronic illness.

Figure 1. The Middle-Range Theory of Chronic Sorrow
(Eakes, Burke, & Hainsworth, 1998)
[The figure is reprinted with the permission of Blackwell Publishing Ltd and G. G. Eakes.]
Managing chronic illness

Persons who are exposed to stress because of disease (or of course other unwelcome circumstances) have to cope with it in some way to regain well-being. Coping is an effort to change cognitive and behavioural strategies to better manage the stress of being ill, which challenges or exceeds the person’s resources (Lazarus & Folkman, 1984). The coping strategies are often referred to as either problem-focused or emotion-focused. In problem-focused coping strategies, problem-solving is one part; the other part comprises internal cognitive strategies such as learning new behaviour or new skills. Forms of emotion-focused coping include reduction of the emotional distress by avoiding, distancing or minimizing, or by positive comparison (ibid). The two types of strategy are independent but can also work together, supplementing each other in the coping process (Lazarus, 2000). Cognitive ability is used, consciously or unconsciously, and personality influences the adaptation efforts (Somerfield & McCrae, 2000). Health-care staff should acknowledge the patients’ unique experiences of distress and provide support when needed. Coping research has focused on coping strategies and how successful or not these are assumed to be. In recent literature not only ineffective coping has been discussed but also findings of positive coping results. Through the effort to infuse a positive meaning into the situation of being exposed to the adverse experiences of illness, knowledge and insight has been gained (Folkman & Moskowitz, 2004). However, Lazarus (2000) cautions that these positive coping outcomes should not be interpreted in a simplistic, inspirational or quasi-religious way.

There is an additional feature in coping with the effects of MS in comparison with most other chronic diseases. Patients with MS have to manage to live with their illness not knowing when the next bout or progression will come, which will increase their worrying over possible future illness trajectories. Consequently this uncertainty fosters emotional and psychological distress in the form of anxiety, depression and loneliness (Gulick, 2001; Mullins et al., 2001). This added emotional stress may explain why coping strategies have been found to be more important as predictors of mood than health-related variables (McCabe & Battista, 2004). In comparing coping between MS patients and a normal population, McCabe and Battista found that the MS patients used a detached style of coping and were less likely to adopt problem-focused strategies (ibid). The result was not to be explained by poor health or lack of work in the MS group. Living with illnesses like MS may in several
ways tax the ability to handle the normal, everyday demands and require special effort on the part of the patients. Developing educational programmes that include strategies which are more problem-focused can be of value in enabling MS patients to adjust more readily to their impairments (McCabe, McKern & McDonald, 2004). Nurses and other health-care staff should provide good psychological support for patients with MS so that they can learn to manage the difficulties and adjust to their illness (Williamson, 2000). Furthermore, the psychological effects of having MS have been shown to be very distressing, and patients are coping by accepting responsibility and escape/avoidance (Mohr et al., 1999). In the same study beneficial ways to cope in the form of cognitive adaptation, seeking social support and positive reappraisal were detected. Although the patients were experiencing benefits when coping with MS, anxiety and anger were also found. The conclusion is that the psychological impact of MS does not always take the same form, and individual differences have to be acknowledged. Health-care staff should recognize the need of knowledge of the most appropriate coping support in helping patients manage to live with such a complex disease as MS.

To sum up this review of MS, its detection, treatment and management, the literature reveals several problematic aspects of contracting and having to live with MS. Since the mid 90s it has become more important to diagnose MS early because of new treatments and their potential to delay the deteriorating effects of the disease. It is now also possible to diagnose MS early, thanks to new techniques such as MRI. MRI is able to show the inflammatory processes in the central nervous system which may give rise to clinical bouts of MS, in addition to the visual evidence of the reduction of inflammatory effects in the central nervous system by means of immunological treatments (Compston & Coles, 2002). It is important to investigate how these treatments affect quality of life. When it comes to developing good health care for MS patients, their experiences of living with the disease, including emotional distress, constitute vital knowledge (Mohr & Cox, 2001). One of the forms of emotional distress described is the recurrent episode of sorrow. The phenomenon and theory of chronic sorrow in connection with losses caused by MS have been studied only in a small group of MS patients (Hainsworth, 1994), and should therefore be studied in a larger sample to evaluate their applicability to empirical data.
Aims

The overall aim of the research project was to increase our understanding and knowledge of the patients’ experiences of living with MS. The project is presented in four studies with the following specific aims:

I: to describe patients’ conceptions of MS before they were diagnosed as well as their illness experiences connected with their initial symptoms and diagnosis.

II: to describe the quality of life of patients with MS on, and of those not on, immunological treatment and to investigate the relationship between impairment and quality of life for these patients.

III: to describe the presence of chronic sorrow and depression in patients with MS and to explore the meaning of chronic sorrow in this study group.

IV: to describe the way the MS patients were managing chronic sorrow and to relate this to the theoretical model of chronic sorrow.
Material and Methods

Design

This research project had a mixed-methods design (Tashakkori & Teddue, 2003) including descriptive, comparative and correlative designs (Table 1).

Mixed-methods design means using quantitative and qualitative methods for different but co-ordinated purposes within the same project (Morgan, 1998). In each of the four studies, there was an awareness of the aim and the methodological assumptions behind each method and its deductive or inductive approach (Tashakkori & Teddue, 2003). Data collection was conducted in such a way as to achieve the quality of the data that each method and analysis requires (Morgan, 1998).

Table 1. Overview of the studies in this thesis

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<th>Design</th>
<th>Method</th>
<th>Analysis</th>
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<td>I</td>
<td>Descriptive</td>
<td>Interview with MS patients regarding initial illness experiences (n=61)</td>
<td>Qualitative inductive content analysis.</td>
</tr>
<tr>
<td>II</td>
<td>Descriptive</td>
<td>Three self-assessment questionnaires concerning, respectively, impairment (SIC), health-related quality of life (SF-36) and subjective quality of life (SQoL) (n=58).</td>
<td>Statistical analysis: Wilcoxon’s matched pair rank sum was used in the between-group comparison. Spearman’s non-parametric correlations.</td>
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<tr>
<td>III</td>
<td>Descriptive</td>
<td>Interview with MS patients on chronic sorrow experiences; interview guide NCRCS and rating of depression in MADRS (n=61).</td>
<td>Deductive assessment of chronic sorrow in the 61 interviews in accordance with criteria from the literature. Qualitative inductive content analysis of 38 interviews with chronic sorrow. Descriptive statistics when comparing depression in chronic sorrow group with depression in non-chronic-sorrow group.</td>
</tr>
<tr>
<td>IV</td>
<td>Descriptive</td>
<td>Interview with MS patients on the experiences of managing chronic sorrow; interview guide NCRCS (n=38).</td>
<td>Qualitative inductive content analysis. The Middle-Range Theory of Chronic Sorrow was used in sorting the interview text in the analysis.</td>
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</tbody>
</table>
The selection of subjects was in line with both comparative and descriptive qualitative design. The group was limited in size in order to be able to manage qualitative interviews but still retained sufficient statistical power in the matched pair analyses (O’Brien & Muller, 1993). This mixed-method design allowed the employment of different techniques. Quantitative differences between a treatment group and matched controls were investigated in Study II while at the same time the demands of a qualitative study design were also satisfied. It was deemed feasible to interview all the 61 participating patients and they were interviewed twice. Subsequently the interviews were subjected to content analysis and the large volume of interviews provided a rich variation in the qualitative data in Studies I, III and IV.

Participants
The patients were selected from a local MS register at the hospital. Inclusion criteria were the following: a clinically definite and/or laboratory-supported definite multiple sclerosis (Poser et al., 1983); age within the range 18–64 years; time since diagnosis at least 6 months; and cognitive ability, as assessed in the medical record, to complete questionnaires. Forty-four patients in the age-range 18-64 years were currently being treated with immunological therapy. In order to get an even age distribution in the sample, 31 of the 44 were randomly selected from two age strata, 18-39 and 40-64 (Table 2). In order to obtain a distribution of men/women in the proportion of 1:2 (reflecting the gender distribution of MS patients), gender was another stratifying variable. Four patients receiving immunological treatment declined to participate and four of the remaining patients receiving such treatment replaced these drop-outs. Twenty of the patients participating in this study were being treated with Interferon-β1b, seven with Interferon-β1a, three with Copolymer-1 and one with Mitoxantrone. They had been on treatment between seven years and six months; the mean treatment time was two years (median one year). The 31 participating patients were matched with patients who were not receiving immunological treatment, serving as a control group. Matching criteria were applied in the following order: gender, disability score on EDSS (Kurtzke, 1983), time since diagnosis and age. Eleven of the matched controls were not willing to participate. Another patient had moved abroad for a year and therefore could not participate. These patients were replaced with another twelve untreated patients from the local register. In addition there was one drop-out from the
Table 2. Selection procedure regarding the 61 participants.

<table>
<thead>
<tr>
<th></th>
<th>Random selection</th>
<th>Matching</th>
</tr>
</thead>
<tbody>
<tr>
<td>Eligible subjects in MS register (n=151)</td>
<td>MS patients on immunological treatment (n=44)</td>
<td>MS patients not on immunological treatment (n=107)</td>
</tr>
<tr>
<td>Planned number of subjects in the project</td>
<td>Random selection (n=31)</td>
<td>Matched control (n=31)</td>
</tr>
<tr>
<td>Withdrawal</td>
<td>Declined to participate (n=4)</td>
<td>Declined to participate (n=11) Moved abroad (n=1)</td>
</tr>
<tr>
<td>Complementation</td>
<td>Random selection (n=4)</td>
<td>Matched control (n=12)</td>
</tr>
<tr>
<td>Drop-out</td>
<td></td>
<td>(n=1)</td>
</tr>
<tr>
<td>Participants in the data collection</td>
<td>31 MS patients on immunological treatment</td>
<td>30 MS patients not on immunological treatment</td>
</tr>
</tbody>
</table>

control group. Consequently the study group consisted of 61 patients, 31 receiving immunological treatment and 30 not (Table 2).

Table 3 shows the demographics of the whole group of 61 patients and of the separate groups of patients receiving immunological treatment and those not receiving such treatment.

In Study I all 61 patients were interviewed about their initial illness experiences and diagnosis. In Study II the active treatment and control groups were compared. The analysis was conducted on 29 matched pairs since one matched pair were excluded because one patient ended the treatment and the other pair were lost because of attrition. In Study III all 61 participating patients were interviewed regarding the presence of chronic sorrow. Assessment of the occurrence of chronic sorrow criteria in the interviews revealed that 38 patients were experiencing chronic sorrow. Consequently the content analysis of chronic sorrow experiences and their management in Study IV was conducted on these patients (Table 4).
Table 3. Demographic and diagnostic characteristics of the study group.

<table>
<thead>
<tr>
<th></th>
<th>Total sample n=61</th>
<th>Immunological treatment n=31</th>
<th>Controls n=30</th>
</tr>
</thead>
<tbody>
<tr>
<td>Women, n (%)</td>
<td>39 (64)</td>
<td>20 (65)</td>
<td>19 (63)</td>
</tr>
<tr>
<td>Men, n (%)</td>
<td>22 (36)</td>
<td>11 (35)</td>
<td>11 (37)</td>
</tr>
<tr>
<td>Age in years, mean (SD)</td>
<td>42.4 (10.7)</td>
<td>39 (9.6)</td>
<td>46 (10.8)</td>
</tr>
<tr>
<td></td>
<td></td>
<td>43 (20-70)</td>
<td>47 (25-70)</td>
</tr>
<tr>
<td>Years since diagnosis, mean (SD)</td>
<td>5.0 (3.4)</td>
<td>5.2 (3.6)</td>
<td>4.9 (3.0)</td>
</tr>
<tr>
<td></td>
<td></td>
<td>4 (1-15)</td>
<td>4 (1-11)</td>
</tr>
<tr>
<td>Disease pattern, n (%)</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Relapsing-remitting</td>
<td>37 (61)</td>
<td>17 (55)</td>
<td>20 (67)</td>
</tr>
<tr>
<td>Secondary progressive</td>
<td>17 (28)</td>
<td>13 (42)</td>
<td>4 (13)</td>
</tr>
<tr>
<td>Primary progressive</td>
<td>7 (11)</td>
<td>1 (3)</td>
<td>6 (20)</td>
</tr>
<tr>
<td>EDSS scores at inclusion, n (%)</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>EDSS 0 – 2.5</td>
<td>26 (43)</td>
<td>12 (39)</td>
<td>14 (47)</td>
</tr>
<tr>
<td>EDSS 3.0 – 5.5</td>
<td>23 (38)</td>
<td>11 (35)</td>
<td>12 (40)</td>
</tr>
<tr>
<td>EDSS 6.0 - &lt; *</td>
<td>12 (20)</td>
<td>8 (26)</td>
<td>4 (13)</td>
</tr>
<tr>
<td>EDSS group mean (SD)</td>
<td>3.4 (1.9)</td>
<td>3.5 (2.0)</td>
<td>3.3 (1.7)</td>
</tr>
<tr>
<td>Civil status, n (%)</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Living alone</td>
<td>24 (39)</td>
<td>13 (42)</td>
<td>11 (37)</td>
</tr>
<tr>
<td>Married/cohabitor partner</td>
<td>37 (61)</td>
<td>18 (58)</td>
<td>19 (63)</td>
</tr>
<tr>
<td>Working capacity, n (%)</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>No influence on working capacity</td>
<td>21 (34)</td>
<td>13 (42)</td>
<td>8 (27)</td>
</tr>
<tr>
<td>MS influences working capacity to some extent</td>
<td>18 (30)</td>
<td>10 (32)</td>
<td>8 (27)</td>
</tr>
<tr>
<td>Unable to work because of MS</td>
<td>20 (33)</td>
<td>8 (26)</td>
<td>12 (40)</td>
</tr>
<tr>
<td>Old age pensioner</td>
<td>2 (3)</td>
<td>0</td>
<td>2 (7)</td>
</tr>
</tbody>
</table>

Note: * highest was 6.5
Table 4. Participation and internal drop-out in Studies I–IV.

<table>
<thead>
<tr>
<th>Study</th>
<th>Study I</th>
<th>Study II</th>
<th>Study III</th>
<th>Study IV</th>
</tr>
</thead>
<tbody>
<tr>
<td>Participants in the data collection</td>
<td>61 MS patients</td>
<td>31 MS patients on immunological treatment</td>
<td>61 MS patients</td>
<td>38 MS patients experiencing chronic sorrow</td>
</tr>
<tr>
<td>Remaining patients in data analysis</td>
<td>(n=61)</td>
<td>Change of treatment (n=1)</td>
<td>Deductive assessment of chronic sorrow and MADRS</td>
<td>(n=38)</td>
</tr>
<tr>
<td></td>
<td></td>
<td>Did not fill in the SF-36 (n=1)</td>
<td>(n=61)</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>Change of treatment in between data collections, attrition in SQoL (n=2)</td>
<td>Content analysis (n=38)</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>29 matched pairs remained in the study (n=58)</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

**Data collection**

*Interview guide regarding illness experiences*

The interview guide concerning the perception of MS before diagnosis, of initial symptoms and of being informed of the diagnosis was was adapted to the aim of Study I. The guide contained three main open-ended questions and the patients’ narratives could be followed up with clarifying questions such as: “Could you elucidate that, give an example?”. The patients were asked to describe what their conception of MS was before they had fallen ill or received any official information about it. They were also asked to describe their initial symptoms and what they had felt and thought about it and how they had managed it at the time. Finally they were asked to say how they had received information of the diagnosis, what they had felt and thought about this information and how they had managed the situation at the time (Study I).
Interview-guide of the Nursing Consortium for Research on Chronic Sorrow (NCRCS)

The NCRCS is a semi-structured open-ended interview guide used in Studies III and IV. It concerns the occurrence of chronic sorrow with particular reference to events associated with the recurrence of losses (Burke, Hainsworth, Eakes, & Lindgren, 1992). The sixteen open-ended interview questions and the follow-up questions are about experiences of loss and managing feelings associated with loss in connection with chronic illness. The interview-guide starts with questions to capture feelings associated with the initial loss experiences, formulated as recall of feelings connected with diagnosis, including whether those feelings have been recurring. There are then questions about other loss experiences, about whether thinking of the illness brings feelings of sorrow to the surface, and about whether such feelings get more intense over time. The interview-guide continues with questions about the patients’ experiences of managing their losses and sorrow. Are there circumstances or persons that have been helpful or not helpful in adjusting to the losses? Finally, there are also questions concerning managing the illness in connection with the behavior of health-care staff. The NCRCS interview-guide has been used in a number of research studies concerning chronically ill adult patients and/or their relatives. Face validity of the instrument was established by two experts in the field and its content validity was established by a panel of seven experts (Burke, Hainsworth, Eakes, & Lindgren, 1992). Inter-rater reliability was tested by comparing coding of the themes of a sample of 10 interviews and tested with kappa statistics, resulting in a good-to-excellent range of agreement (ibid). The Swedish version of the NCRCS was used in the study, permission having been given for translation of the guide into Swedish (Ahlström, 2006).

Self-reported Impairment Check-list (SIC)

The Self-reported Impairment Check-list is a structured guide for the screening of 15 signs of impairment associated with MS. The patients reported in Study II whether they had had the symptoms and whether the impairment was moderate or severe, also whether the symptoms were constant or fluctuating. SIC concerns problems to do with balance/vertigo, walking, hand/finger function, sensitivity, spasticity, co-ordination, pain, fatigue, speech, bladder function, eyesight, sexual function, concentration/poor memory, heat sensitivity and paroxysmal symptoms. The instrument has been used in an earlier study concerning stroke patients (Widar & Ahlström, 2002). In the present
case it was modified for MS symptoms on the basis of the literature and clinical experience (Olek & Dawsson, 2000).

**The 36-Item Short-Form health survey questionnaire (SF-36)**

SF-36, used in Study II, is an instrument for self-assessment with regard to health-related quality of life (Ware & Sherbourne, 1992). It captures health problems for MS patients and has been used on several occasions (Nortvedt, Riise, Myhr, & Nyland, 1999). There are 36 items, 35 of which are grouped into eight scales: Physical Function, Role limitations due to physical problems, Bodily pain, General health, Vitality, Social function, Role limitations due to emotional problems and Mental health. The items were coded and summed following the standards in the SF-36 manual (Sullivan & Karlsson, 1998; Sullivan, Karlsson, & Ware, 1994). The other item, “Health change,” concerns health during the past year. The English version of SF-36 has been used in several studies and has been well validated (Freeman, Hobart, Langdon, & Thompson, 2000; Nortvedt, Riise, Myhr, & Nyland, 1999; Sullivan, Karlsson, & Ware, 1995). The instrument has been translated and undergone psychometric testing with satisfactory results for use in many countries, including Sweden (Sullivan, Karlsson, & Ware, 1995). Inter-correlation between the SF-36 dimensions showed that each dimension measured a related but distinct construct (Freeman, Hobart, Langdon, & Thompson, 2000). The Swedish version was used in this research and reliability for the eight dimensions and all items was high, with Chronbach’s alpha ranging between 0.74 and 0.93.

**Subjective estimation of Quality of Life questionnaire (SQoL)**

The instrument SQoL, used in Study II, reflects an interactionistic perspective on quality of life and includes social, psychological and interpersonal aspects (Kajandi, 1994; Naess, 1987). SQoL is intended both for healthy persons and for different patient groups. The instrument involves three socio-economic variables, five on relations, nine on personal feelings and one on quality of life as a whole. The respondent evaluates the degree of satisfaction in his or her life along each of the 18 variables. There is a Likert scale 1-5 for the answers, including half steps. Each variable is represented by one item, except for three variables having two items, which are mutually exclusive. For example, if the person has rated the quality of the
relationship with his or her partner, they should not answer the next item, since that involves rating the situation of having no partner. SQoL has been used in several studies and has shown acceptable validity (Hugosson et al., 1997; Kajandi, 1994). Chronbach’s alpha in this study was 0.90 for all items.

Montgomery–Asberg Depression Rating Scale (MADRS)
MADRS, used in Study III, is a self-report screening questionnaire for depression. The patients estimate how they have been feeling the last three days. The questionnaire has 9 items concerning discomfort, ranging from 0 = absence of discomfort to 6 = maximum of discomfort. The items concern Reported sadness, Inner tension, Reduced sleep, Reduced appetite, Concentration difficulties, Lassitude, Inability to feel, Pessimistic thoughts and Suicidal thoughts/Zest for life (Montgomery & Asberg, 1979). The Swedish Medical Products Agency suggests four classification levels: 0–18 = no depression, 19–27 = mild depression, 28–36 = moderate depression, 37–54 = severe depression (Medical Products Agency, 2004). MADRS has been demonstrated to be reliable and valid as well as a sensitive measure of depression in psychiatric populations (Montgomery & Asberg, 1979) and reliable for self-reporting in the case of individuals with depression (Mundt et al., 2005).

Procedure
The selected patients received a letter containing information about the research project where it was indicated how the data would be collected and that participation was voluntary. After three to five days I phoned them to give more information about the study and ask if they were willing to participate. If the patient was willing to participate an appointment for the first interview was booked. The interviews were mostly conducted in the patients’ homes, except in eight cases where the patient preferred to come to a secluded room in the hospital. The interviews were tape-recorded. In the first data collection the interview concerned their conception of MS before diagnosis, their initial symptoms and experiences of receiving their diagnosis. After the interview the patients filled in the Self reported impairment Check-list. They were then given SF-36 (Sullivan, Karlsson, & Ware, 1995) and a stamped addressed envelope, the questionnaire to be completed and returned within two weeks.
Approximately four months later, I phoned to ask if they were willing to continue participating in the project, which would involve being interviewed a second time where the interview was tape-recorded. All patients stayed in the project and took part in the second data collection. First they were interviewed with the aid of the semi-structured interview guide concerning chronic sorrow, NCRSC (Burke, Hainsworth, Eakes, & Lindgren, 1992), then they completed the SQoL (Kajandi, 1994) and MADRS (Montgomery & Asberg, 1979) questionnaires. All interviews were transcribed verbatim, including expressions of emotion.

Analysis
In three of the studies content analysis was applied to the interviews. The patients were interviewed twice during the data collection, each interview about one to two and a half hours.

Content analysis in Study I
In Study I, narrative methodology was used in the analysis, with a categorical-content perspective (Lieblich, Tuval-Mashiach, & Zilber, 1998). First the interviews were read to get a sense of the whole. Then NVivo Software, a computer-based program for coding qualitative data, was used in coding and sorting the material (Richards, 2002). Five randomly selected interviews were analysed and content areas related to the interview questions were developed. These content areas were then used as an analytical framework in sorting the rest of the interview texts (Graneheim & Lundman, 2004). In the next stage of the analysis preliminary categories were developed. The analysis was conducted in respect of both the whole interview and parts of it, in a circular procedure (Lieblich, Tuval-Mashiach, & Zilber, 1998). The meaning units and preliminary categories were pasted in a regular Word document and further analysed and condensed, then labelled in accordance with the core content. Then comprehensive themes were developed. The various steps in the analysis, together with the development of categories and themes, were scrutinized by the co-author and discussed with her, this in order to establish trustworthiness (Graneheim & Lundman, 2004).
Content analysis in Studies III and IV

The interviews guided by the NCRCS were listened to in order to identify the phenomenon of chronic sorrow in the patients’ narratives. The assessment of chronic sorrow for each patient was performed in accordance with the following criteria from the literature: (1) The experience of significant loss of personal meaning, (2) Ongoing loss or several loss experiences, (3) Pervasive sadness or grief, (4) Permanent sadness or grief, (5) Periodic sadness or grief, (6) Potentially progressive sadness or grief and (7) Periods can be triggered by occurrence events (Eakes, Burke, & Hainsworth, 1998). The seven criteria were marked on a protocol by means of a plus (present) or minus (absent). The patient was regarded as experiencing chronic sorrow if at least four out of seven criteria were fulfilled: 1, 2, 3 or 4, 5 or 7 (Ahlström, 2006). To ensure that the criteria were fulfilled and that credibility was guaranteed, a second researcher (GA) listened to uncertain cases and helped decide (n = 5).

The interviews revealing chronic sorrow were selected for inductive content analysis to interpret the essence of the text. The texts were read several times to get a sense of the whole and then meaning units were marked for further analysis (Graneheim & Lundman, 2004). In Study III the criteria of chronic sorrow were treated as predetermined content areas, and meaning units from the interviews were sorted into them. In Study IV meaning units related to helpful or not helpful events and managing or not managing the situation were identified.

The analysis continued with the condensation of the text, preserving its essence and keeping it as close as possible to the original wording. Thereafter the condensed units of meaning were analysed, with an interpretation of the underlying meaning expressed in terms of codes. The essences of the codes were further abstracted into sub-themes. Themes were in turn developed from the sub-themes, whereby in the case of Study III the focus was on losses and in the case of Study IV on management of chronic sorrow and the patients’ experiences of discomfort or comfort.

I wrote the condensations and developed the codes, critically reviewed and discussed with a second researcher (GA). We independently read and reflected on the statements as well as discussed the interpretations in the analysis, this in order to establish trustworthiness. Themes were developed through agreement concerning key contents (Graneheim & Lundman, 2004).
Statistical analysis

Statistical analysis was used in Studies II and III. A result at the p<0.05 level was considered statistically significant. In Study II the non-parametric Wilcoxon’s matched pair rank sum was used to compare the group receiving immunological treatment with the group receiving no such treatment (Altman, 1991). To investigate the relationship between impairment and quality of life Spearman’s non-parametric correlation (r) was used (Altman, 1991). Significant correlations were considered as follows: little or none (≤0.25), fair (0.25-0.50), moderate to good (0.50-0.75) and excellent (≥0.75) (Colton, 1974). Correlations were also investigated between quality of life and MADRS.

The screening of depression through MADRS in Study III was subjected to non-parametric statistical tests. The ratings of discomfort on MADRS of patients with chronic sorrow were compared with the ratings of those without chronic sorrow by means of a Mann-Whitney U-test (Altman, 1991). The same test was used to investigate whether there was any group difference in quality of life as between the group with chronic sorrow and the group without.

Demographic variables were also tested to control for group differences regarding quality of life between the immunologically treated and the untreated patient groups and between the chronic sorrow group and those not experiencing chronic sorrow. Examined variables were investigated by means of Mann-Whitney U-test (Altman, 1991).
Ethical considerations

The research project was approved by the Research Ethics Committee, Örebro University Hospital, Sweden, (§ 500:16 188/01) on 8 February 2001.

All patients were informed about confidentiality with regard to personal information and research data and gave their informed consent regarding their participation in the project. They were informed of the possibility of withdrawing at any time without having to say why. One patient withdrew after inclusion, all the others remained and participated in both interviews.

The research project was not initiated or administered by the local neurological clinic. The clinic was, however, supportive of the project in terms of attending to the needs of patients which were revealed by the data collected. The doctors at the clinic had no information as to which patients were contacted by the researchers or which of them decided to participate. If the patient expressed a need for psychological counselling or other support, the researcher could contact the social worker or the nurse at the clinic and set up a meeting for the patient. On some occasions patients did express such needs and the appropriate support from health-care staff was arranged. We also thought it was important to inform the patients about autonomy, assuring them that their treatment or other health-care interventions would not be affected by their participating or not.

In studies where patients are interviewed about negative emotional experiences there is always a risk in that it can cause distress to recall and talk about such experiences. In this research project several types of such experiences were discussed, such as distress related to the diagnosis, depression and sorrow. In the interview situation, time was initially spent to introduce the researcher and make the patient feel comfortable in the interview situation. Throughout the interview and after it, the patient’s emotions were taken into consideration. After the interview consideration was taken in regard to how emotionally the interview experience had been. Some patients cried during the interview when remembering their initial illness experiences and diagnosis. In the talk afterwards they expressed a wish to share their experience in order to help others and contribute to an improved health care, and that was a strong motive for participating in the research. Patients must always be considered as an exposed group and it is of highest importance to respect them and not
threaten their integrity. It is at the same time vital to listen to their illness experiences and life stories as a way to learn from them and thereby promote the development of good care.

In the screening of depression patients with symptoms of low mood or reduced zest for life, a contact with a counsellor after the screening was offered. Even so, the questionnaire or interview could have awakened thoughts afterwards of which the researcher had no knowledge. On the other hand many of the patients expressed an insight into what they were going through and experienced a feeling of courage when talking about it. They gave suffering a voice but they were not weak individuals — it was probably more of a combination of being exposed and becoming stronger through their illness experience.

Confidentiality with regard to patients’ identities in the presentation of the results has received top priority, not least when quotations have been used to illustrate the patients’ experiences.
**Summary of results**

The results from the retrospective interview concerning initial symptoms and diagnosis are presented first (Study I) and thereafter the results of the studies on quality of life and chronic sorrow (Studies II, III and IV).

**Illness experiences of MS, from symptom to diagnosis, Study I**

*Early perception of MS*

The narratives about the perception of MS before falling ill and prior to any official information about MS commonly concerned disablement and death. The patients spoke of having a negative picture of disabled MS patients in wheelchairs or of patients who had died of MS. The wheelchair was a symbol of lost capacity as a threat to life and was a trademark element in the narratives. There were also more nuanced descriptions of MS: it was a severe disease but there also was the possibility of having it in a milder form, not disturbing life and health. In sum, the terms in which the patients perceived MS before falling ill ranged from disablement to good health.

*Initial illness experiences*

Falling ill with MS was emotionally distressing for most of the patients (Study I). The initial symptoms were commonly sensory changes or dysfunction in the legs (Table 5). The degree of seriousness of the symptoms varied from mild (where there was no seeking of medical assistance) to severe (with ensuing hospitalization). With regard to how the patients conceived of the cause of their symptoms, a variety of factors emerged: a neural defect, a brain tumour, a virus and being overworked. Becoming vulnerable was a theme of the experiences of the initial symptoms. The patients had felt emotional distress initially. Anxiety was described by almost all patients, in terms of worrying over the symptoms and what caused them. They were frightened at the prospect of having a brain tumour or other life-threatening disease. Sadness at lost functions was described, and sadness at not being believed. Some had experienced shame regarding their symptoms when the body did not function as before, and falling in public was especially described as being very shameful. They had sought medical assistance in different health-care settings, and some were told, by health-care staff without neurological expertise, that their symptoms were imaginary or psychiatric in
Table 5. Patients’ descriptions of initial symptom of MS

<table>
<thead>
<tr>
<th>Symptom</th>
<th>Number</th>
</tr>
</thead>
<tbody>
<tr>
<td>Sensory dysfunction</td>
<td>21</td>
</tr>
<tr>
<td>Physical impairment</td>
<td>18</td>
</tr>
<tr>
<td>Physical loss, legs did not work as usual (n=16)</td>
<td></td>
</tr>
<tr>
<td>One side of the body did not function (n=2)</td>
<td></td>
</tr>
<tr>
<td>Eye-sight problems</td>
<td>10</td>
</tr>
<tr>
<td>Vertigo</td>
<td>4</td>
</tr>
<tr>
<td>Fatigue</td>
<td>3</td>
</tr>
<tr>
<td>Pain</td>
<td>1</td>
</tr>
<tr>
<td>Paroxysmal symptoms</td>
<td>1</td>
</tr>
<tr>
<td>Bladder problems</td>
<td>1</td>
</tr>
<tr>
<td>Problems with swallowing/neck feeling</td>
<td>1</td>
</tr>
</tbody>
</table>

One missing, n=60

nature. None had received a psychiatric consultation, which is surprising. They were angry at not being taken seriously by health-care staff with regard to their symptoms and they were angry at not getting any information about their condition.

However, some also described a feeling of trust in connection with going to the clinic on account of the initial illness incident. Experiences of friendly treatment, support and consolation were described. The patients had been seen, listened to and acknowledged in connection with their symptoms and felt that they had been respected.

Initially many of the patients repressed their symptoms and the thoughts of them. They tried to minimize them and deliberately avoided thinking about them, hoping it would all go away. The patients tried to understand the symptoms and searched for a physical cause. Eventually they sought help. Some had to go to see several health-care providers in different settings with the resulting mistrust and misunderstandings on the way, others experienced good emotional support before being referred to a neurologist for a more thorough diagnostic investigation.
Diagnosis of MS

The occasion when the patient was informed of the diagnosis of MS was described, and it included strong emotional reactions and a sense of having to cope with the situation. This was the beginning of a long journey, stretching from their first sense of vulnerability to the acquirement of the requisite strength to successfully manage emotional distress (see below).

Receiving the diagnosis had been a shock even for the patients with whom the possibility of MS had been discussed during the medical investigation. It was a terrifying diagnosis. They felt anxiety and fear related to thoughts of developing severe disability as a result of the progress of MS. The uncertainty did not decrease. Even though they now knew the cause of their symptoms the future was still unpredictable as they were now living with a progressive chronic illness. Some spoke of how they lost hope: they were doomed and the diagnosis was a threat to their life.

Earlier perception of MS in terms of disablement and death, as well as seeing other disabled patients during the medical investigation, increased their despair. Sorrow was a common reaction to the diagnosis and crying was described. Some cried at the interview as they remembered the occasion when they had been informed: the sadness returned through their talking about it and it still remained a strong emotional experience. Anger at being informed of the diagnosis over the phone was spoken of, also anger of having the disease. They lost their identity as healthy persons but were reluctant to identify themselves as ill or disabled.

Those that did not experience a sufficient follow-up after their diagnosis felt abandoned by the health-care staff. A feeling of being abandoned was also described in connection with lack of support from family and friends. It was often the patient who had to be strong and comforting when the family was shocked and devastated by the diagnosis. In some cases the diagnosis was actually a relief: finally the patient had an explanation of the symptoms and the reduced physical capacity. In other cases the confirmation was not a positive experience but a confirmation of patient’s worst fears. In contrast, some patients were confirmed in a positive way through satisfactory medical check-ups with supportive health-care staff and the appropriate information.

In the beginning the diagnosis was too threatening to handle so they repressed it. Some also doubted the diagnosis; maybe it was wrong. Existential
brooding was described, involving asking oneself “Why me?” or feeling that one had wrongly sentenced (by fate). Seeking social support from family, friends and healthcare staff helped the patients initially. Accepting the disease, acknowledging this new fact of life, took time, some of the patients reflected. They were fighting for as normal a life as possible, not giving up or pitying themselves. They saw ordinary things with new eyes; new values became important. They also took more care of themselves and were more content with their bodies. Hope was expressed, as in the hope of having a mild form of MS, of being able to recover from a bout or of a new treatment’s being developed. Complementary treatment also provided a sense of hope.

In sum, the illness experience was an inner journey of becoming vulnerable and continuing in that vulnerability after diagnosis. The patients were vulnerable until they could manage their emotional distress and the disease in such a way that they could reappraise their values and again see a hopeful future. It was for many of the patients a journey wrought with anger, sadness and other emotional distress, and they had to make a great existential effort in order to cope and to move beyond despair to a new perspective on life, which involved acknowledging MS but not being defeated by it. This prolonged process of acquiring the needed strength to live with MS will be further gone into below.

Consequences of living with MS: measures of impairment, quality of life, chronic sorrow and symptoms of depression

*Impairment and quality of life, Study II*

In Study II 29 MS patients receiving immunological treatment were compared in respect of quality of life with 29 matched controls not receiving such treatment. Relationships between impairment and quality of life in all these patients were also investigated. The treated group were younger, the mean age being seven years lower than in the control group. No other demographics differed between the groups to the level of statistical significance (a table of demographics is to be found in the paper presenting Study II).
The Self-reported Impairment Check-list captured many of the problems caused by MS. In the overall group of 61 MS patients, mostly with mild to moderate EDSS, the most frequent signs of impairment were disturbance in balance and problems with walking, reported by 82% of the patients. Fatigue was also a common problem among the patients, experienced by 77%. Problems with bladder function were reported by 67%, and muscular spasticity by 59%. The patients reported that most problems were of constant duration. There was no statistically significant difference in the matched pair analyses between the 29 treated and 29 untreated patients with respect to the impairment check-list.

In health-related quality of life as measured by SF-36, the most negative influence of MS, taking all patients into account, was detected on Physical role, Vitality, General health and Physical function (Table 6). There was no statistically significant difference between the treated and untreated patient groups with regard to health-related quality of life.

Table 6. SF-36 in MS patients (n=60)

<table>
<thead>
<tr>
<th>SF-36 scales</th>
<th>Median</th>
<th>IQR*</th>
<th>Mean (SD)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Physical functioning</td>
<td>55</td>
<td>30-89</td>
<td>57 (29)</td>
</tr>
<tr>
<td>Physical role limitation</td>
<td>50</td>
<td>0-100</td>
<td>45 (43)</td>
</tr>
<tr>
<td>Bodily pain</td>
<td>68</td>
<td>41-100</td>
<td>69 (28)</td>
</tr>
<tr>
<td>General health</td>
<td>50</td>
<td>35-70</td>
<td>53 (21)</td>
</tr>
<tr>
<td>Vitality</td>
<td>47</td>
<td>31-64</td>
<td>48 (24)</td>
</tr>
<tr>
<td>Social function</td>
<td>75</td>
<td>50-100</td>
<td>70 (28)</td>
</tr>
<tr>
<td>Emotional role limitation</td>
<td>67</td>
<td>33-100</td>
<td>63 (40)</td>
</tr>
<tr>
<td>Mental health</td>
<td>76</td>
<td>56-87</td>
<td>70 (21)</td>
</tr>
</tbody>
</table>

** Health change item  3  2-4  3 (1)

Low score indicates more physical, social and psychological influence on health and quality of life. One questionnaire missing in the study, n=60

* IQR = interquartile range (25th–75th percentile)

** based on the single item Health change, not included in the index above. Low item score indicates better health change.
In the measurements of subjective quality of life in SQoL, patients rated their self-satisfaction with regard to social features, relations and psychological well-being. The group as a whole were most satisfied with their relation to their own children, frequently assigned the highest score (median 5.0) (Table 7). Housing quality and emotional experiences were also assessed positively (both 4.5). Having no meaningful work, having no partner, having no children and having insufficient energy were the most negatively experienced (all 3.0). However, there were no statistically significant differences between the matched pair groups in SQoL. In checking for influences of negative and positive events in the time between the two data collections, still no significant differences were found.

With regard to possible existing relationships between impairment and health-related quality of life in MS patients, pain and problems with walking showed the strongest association. Bodily pain in SF-36 and the impairment Pain had the highest correlation (-.78). Even though Physical function in SF-36 had a high correlation with the impairment Walking (-.71), it was Vitality in SF-36 that significantly correlated with most signs of impairment. Vitality in SF-36 is concerned with feelings of alertness, being strong and filled with energy, or the opposite, feeling worn-out and tired. Lack of vitality was moderately associated with the impairments of Fatigue (-.52), Concentration/poor memory (-.53) and Pain (-.54) (all correlations p<.01). (All correlations are expected to be negative since scores in the instrument were in opposite directions.)
Table 7. Subjective quality of life in SQoL of all 61 MS patients.

<table>
<thead>
<tr>
<th>Items</th>
<th>n</th>
<th>Median</th>
<th>IQR</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. Housing quality</td>
<td>61</td>
<td>4.5</td>
<td>4.0-5.0</td>
</tr>
<tr>
<td>2. a. Have work, meaningful occupation</td>
<td>42</td>
<td>4.0</td>
<td>3.5-4.5</td>
</tr>
<tr>
<td>b. Have no work, meaningful occupation</td>
<td>19</td>
<td>3.0</td>
<td>1.5-4.0</td>
</tr>
<tr>
<td>3. Quality of personal economy</td>
<td>60*</td>
<td>4.0</td>
<td>3.0-4.5</td>
</tr>
<tr>
<td>4. a. Relationship to partner</td>
<td>41</td>
<td>4.0</td>
<td>4.0-5.0</td>
</tr>
<tr>
<td>b. Have no partner</td>
<td>20</td>
<td>3.0</td>
<td>2.0-3.0</td>
</tr>
<tr>
<td>5. Relationship to friends</td>
<td>60*</td>
<td>4.25</td>
<td>3.5-5.0</td>
</tr>
<tr>
<td>6. Relationship to mother</td>
<td>60*</td>
<td>4.25</td>
<td>3.6-5.0</td>
</tr>
<tr>
<td>7. Relationship to father</td>
<td>60*</td>
<td>4.0</td>
<td>3.6-5.0</td>
</tr>
<tr>
<td>8.a. Relationship to own children</td>
<td>42</td>
<td>5.0</td>
<td>4.0-5.0</td>
</tr>
<tr>
<td>b. Have no children</td>
<td>19</td>
<td>3.0</td>
<td>3.0-5.0</td>
</tr>
<tr>
<td>9. Involvement</td>
<td>61</td>
<td>4.0</td>
<td>3.5-4.5</td>
</tr>
<tr>
<td>10. Energy</td>
<td>61</td>
<td>3.0</td>
<td>2.5-4.0</td>
</tr>
<tr>
<td>11. Self-actualization</td>
<td>61</td>
<td>3.5</td>
<td>3.0-4.0</td>
</tr>
<tr>
<td>12. Freedom</td>
<td>61</td>
<td>4.0</td>
<td>2.75-4.25</td>
</tr>
<tr>
<td>13. Self-assuredness</td>
<td>61</td>
<td>4.0</td>
<td>3.0-4.0</td>
</tr>
<tr>
<td>14. Self-acceptance</td>
<td>61</td>
<td>4.0</td>
<td>3.0-4.5</td>
</tr>
<tr>
<td>15. Emotional experiences</td>
<td>61</td>
<td>4.5</td>
<td>4.0-5.0</td>
</tr>
<tr>
<td>16. Security</td>
<td>61</td>
<td>4.0</td>
<td>3.0-4.5</td>
</tr>
<tr>
<td>17. General mood</td>
<td>61</td>
<td>4.0</td>
<td>3.5-4.5</td>
</tr>
<tr>
<td>18. Quality of life as a whole</td>
<td>61</td>
<td>4.0</td>
<td>3.5-4.5</td>
</tr>
</tbody>
</table>

Low score indicates more negative influence on subjective quality of life.
IQR = interquartile range (25th-75th percentile)

* one missing data point.

Presence of chronic sorrow and symptoms of depression, Study III

Criteria of chronic sorrow from the literature were used to identify chronic sorrow in the total group of 61 patients. There were 38 patients (62%) with chronic sorrow experiences.

In screening for the presence of depression in the whole study group of 61 patients, 57 were found to have no depression. The remaining four patients had mild depression, two scoring 20 and two 26 out of 54 points. There was a higher occurrence
of depressive symptoms in the group with chronic sorrow than among those having no chronic sorrow, but none was clinically depressed according to the guidelines of the Swedish Medical Products Agency with reference to MADRS (Table 8).

Demographics, quality of life and impairment in relation to chronic sorrow and mood
The chronic sorrow group experienced significantly lower Mental health in SF-36 (p=.012) than those not experiencing chronic sorrow. There was also a tendency towards more Pain (p=.051) and lower General health (p=.054).

Disability in EDSS had no association with chronic sorrow or MADRS. SIC revealed that the group experiencing chronic sorrow reported more problems with Fatigue (p=.041).

Lower Physical functioning was not associated with lower mood in this group of patients. No associations between SF-36 and MADRS were shown on Physical functioning or Physical role limitations. All other SF-36 scales were associated with depressive symptoms: Pain -.35 (p=.01), General health -.52 (p=.00), Vitality -.44

Table 8. Comparison of the scores on MADRS when screening for depression (n=61).

<table>
<thead>
<tr>
<th>Item score</th>
<th>Chronic Sorrow</th>
<th>No Chronic Sorrow</th>
<th>P-value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Item score mean (SD) median (IQR)</td>
<td>n=38</td>
<td>n=23</td>
<td></td>
</tr>
<tr>
<td>Reported sadness</td>
<td>0.42 (1.10)</td>
<td>0.17 (0.58)</td>
<td>n.s</td>
</tr>
<tr>
<td>Inner tension</td>
<td>1.42 (1.22)</td>
<td>0.35 (0.76)</td>
<td>p&lt;0.01</td>
</tr>
<tr>
<td>Reduced sleep</td>
<td>1.58 (1.75)</td>
<td>1.04 (1.46)</td>
<td>n.s</td>
</tr>
<tr>
<td>Reduced appetite</td>
<td>0.26 (0.69)</td>
<td>0.09 (0.42)</td>
<td>n.s</td>
</tr>
<tr>
<td>Concentration difficulties</td>
<td>1.00 (1.21)</td>
<td>0.52 (0.90)</td>
<td>n.s</td>
</tr>
<tr>
<td>Lassitude</td>
<td>1.05 (1.38)</td>
<td>0.70 (1.30)</td>
<td>n.s</td>
</tr>
<tr>
<td>Inability to feel</td>
<td>0.79 (1.5)</td>
<td>0.17 (0.58)</td>
<td>n.s</td>
</tr>
<tr>
<td>Pessimistic thoughts</td>
<td>1.32 (1.63)</td>
<td>0.09 (0.43)</td>
<td>p&lt;0.001</td>
</tr>
<tr>
<td>Suicidal thoughts/Zest for life</td>
<td>1.26 (1.35)</td>
<td>0.26 (0.69)</td>
<td>p&lt;0.002</td>
</tr>
<tr>
<td>Total score</td>
<td>9.11 (7.24)</td>
<td>3.39 (3.97)</td>
<td>p&lt;0.001</td>
</tr>
</tbody>
</table>
(p=.00), Social function -.46 (p=.00), Emotional role limitation -.30 (p=.02) and Mental health -.53 (p=.00). (As before, the instruments go in opposite directions and so the correlations are negative.)

There were no statistically significant demographic differences between the groups with and without chronic sorrow, except that married persons and cohabitor partners experienced chronic sorrow more frequently (p=.034).

Lower satisfaction with subjective quality of life in SQoL was reported in the chronic sorrow group with regard to having no work (p=.038). Lower degrees of Energy (p=.049), Freedom (p=.026), Self-assuredness (p=.013) and Self-acceptance (p=.010) were also reported by those who were experiencing chronic sorrow than by those who were not.

MADRS was moderately associated with the SQoL items Self-assuredness (-.58; p=.000), Self-acceptance (-.52; p=.000) and Total quality of life (-.59; p=.000).

Disease pattern was not a matching variable and it differed with respect to gender in the whole study group of 61 patients. Men were experiencing a progressive form much more often than women. No other demographic variables differed between genders.

Problems with sexual function were significantly more reported by men in SIC (p=.008), but no other gender differences were found regarding self-reported impairments. In health-related quality of life in SF-36 there were no statistically significant differences between the men and the women. In SQoL the women rated their relations to partner (p=.046) and to friends (p=.023) higher then the men. In addition, the women rated emotional experiences significantly higher (p=.044) than the men, but there were no gender differences in the overall quality of life. The presence of chronic sorrow or depressive symptoms in MADRS did not differ with regard to gender.

In sum, MS deeply influenced the patients’ lives: they reported several impairments that were the result of the disease process and they subsequently had a decreased health-related quality of life. The subjective estimation of quality of life was not reduced overall, and quality of life as a whole was estimated as good (4 out of 5). There were no statistically significant differences in impairment or quality of life between the 29 patients receiving immunological treatment and the 29 untreated patients. Thirty-eight
(62%) of the MS patients experienced chronic sorrow with its emotional distress, although only four reported symptoms of being mildly depressed. The chronic sorrow group experienced a decreased state of mental health, with increased fatigue and a lower subjective quality of life. The men had more problems with sexual function, while the women had better social relationships.

**Qualitative findings in the light of the theory of chronic sorrow**

*Meaning of chronic sorrow, Study III*

The analysis of the interview material was guided by the middle-range theory of chronic sorrow (Studies III and IV). In Figure 2 the 38 MS patients’ experiences are shown (Studies III and IV), this figure being similar to the original figure of the theory of chronic sorrow (Figure 1). The purpose of Figure 2 is to present an overview of the qualitative results.

The content analysis of the interview text revealed that many expressions of emotion emerged in the description of the MS patients’ sorrow (Study III). The patients had become less stable in their mood and experienced emotional ups and downs. Frustration and sadness was constantly present or periodically emotionally overwhelming. There were feelings of sorrow, fear, and anxiety and they had also become more pessimistic. Losses and restrictions had become a part of their lives and they were living with the sorrow caused by that. The essence of the losses that caused them sorrow was developed in seven themes (Figure 2), which will be highlighted in the text by italicization.

A prominent part of the patients’ experiences was the *Loss of hope*, expressed by 35 of the 38 patients (92%). They had lost hope and saw disease progression and lost independence in the future. They had lost confidence in life, lost their sense of safety and security, regarding both the present and the future, and they were afraid to make plans. They grieved as they expected that the course of the disease would take a negative turn and saw no hope of a long life. Death was coming closer. *Loss of control over the body* was also frequently experienced, described by 33 of 38 patients (87%). The body did not function as before and life had been reduced since the body’s malfunction hindered them. Reduced energy was a large problem, both bodily and mentally, leading to reduced capacity. They grieved at the loss of their vitality and strength. Sorrow was also spoken of in connection with the *Loss of*
integrity and dignity (53%). They expressed feelings of being worthless and of being met with disrespect. When symptoms revealed the illness to others, the patients felt exposed. They were not the same person as before because of the Loss of a healthy identity (50%). They had lost their previous self-image, including both their inner self and appearance. Similarly the identity of being an active and alert parent disappeared. Loss of faith that life was fair was described and they saw the disease as unfair punishment (34%). They had lived their lives as responsible persons and still been stricken with illness. The Loss of social relations, both private and with fellow-workers, (26%) and the Loss of freedom (21%) were also hard to bear. Since the physical effect of MS interfered with the performance of different activities they now always had to make plans for their daily activity.

MS patients managing chronic sorrow, Study IV
Study IV describes the experiences of the study group of 38 MS patients having to manage chronic sorrow in the form of themes (in italics) (Figure 2). External methods of managing included the efforts of family and friends, as well as the support received from health-care staff. Internal methods of managing were constituted by the patient’s own efforts to handle the situation. The 38 patients’ utilization of both types of methods in an effective way increased their comfort, while discomfort resulted when their illness situation was not properly handled.

The analysis showed that ineffective internal management left the patients Struggling with vulnerability. They were caught up in worries and uncertainty with regard to the progression of the disease, which was a recurrent subject when talked about their sorrow. They did not want to be reminded of the disease, avoided talking about it or were in disease denial when they were with other people. Ineffective external management manifested itself as not being understood or receiving Deficient affirmation in situations where there was a considerable need for social support. Lack of understanding in the family was a question of not grasping the need of support or being over-protective. Friends’ lack of understanding and knowledge of MS caused friendship to fade away. Uncaring health-care staff were not sensitive to patients’ needs or were disrespectful in the encounter with the patient. Some of the patients had also been treated with disrespect in other encounters, such as at work.
These experiences of discomfort are reflected in the theme of *Sorrow from vulnerability* (Figure 2). Recurrence of losses made the patients feel sad and unbalanced in their mood. Their everyday life was ruled by the symptoms of MS and they had not yet found a way to adjust. The patients expressed a lack of personal knowledge of MS but were at the same time scared about what they might learn, which hindered successful management of the recurring losses. Being in a vulnerable situation, as well as being met with disrespect, was experienced as a trespass on their integrity. The patients described ways in which they managed their chronic sorrow ineffectively, which resulted in discomfort, but there were also effective ways that increased their comfort.

Effective internal management was characterized by the theme of *Mastering with realistic awareness* (Figure 2). Patients created distraction, for example, by distancing themselves from their emotions or doing pleasant thing that made them feel better. They allowed themselves to experience sadness and they could have a good cry and then go on with what they had been doing. Accepting MS and adjusting to losses was a long process and required effort. In order to feel better they adopted a “can do” attitude to fight impairment and do things in spite of symptoms. Thinking positively and constructively helped, together with the appreciation of family or friends and setting up realistic goals. The patients were creating hope, hope that they would not get any worse and hope in the progress of MS research. By talking to others they were articulating and reflecting upon their worries. Finally, they were also managing internally by taking care of themselves for the purpose of maintaining health, both emotionally and physically. The theme of *Endorsing management* described effective external management. Caring families were described by the patients, and these families were helpful emotionally as well as with practical tasks in the home. Helpful was also friends’ respectful acknowledgement of their losses and sorrow. In addition, the patients had encountered confirmation from health-care staff, for instance in the form of consolation and friendly treatment.

*Appreciation of life and trust in the future* was developed as a theme that reflected increased comfort. The patients had acquired an insight into the nature of having MS and being ill and their knowledge made them more understanding. Personal confidence was developed with knowledge of their ability to adjust to their illness situation, leading to calmness and increased self-esteem. Hope of a normal life in spite
of MS was described despite their experiences of losses. Successful management could also lead to a feeling of regaining and keeping the initiative in their life, with a sense of being in control of their own destiny.

To sum up: seven themes of loss described the meaning of these MS patients’ sorrow. Large emotional distress was connected to their perceived and experienced losses due to MS. Chronic sorrow was expressed as a pattern of losses where loss of hope was central, together with loss of control of the body. In Study IV the theoretical model was used for sorting the data associated with the management of chronic sorrow. The patients were managing in varying ways (Study IV). Ineffective internal and external management increased the patients’ vulnerability and discomfort. Effective internal managing was described in many ways, with good examples of personal efforts made to enhance their own well-being, which together with external managing increased their comfort in life, which included enhanced personal growth, confidence and trust in life.
Figure 2. Chronic Sorrow in Multiple Sclerosis
Discussion

Discussion of results

_Initial illness experiences and diagnosis – Study I_

The experiences of falling ill and receiving the diagnosis of MS exposed the patient to severe emotional stress, giving rise to a feeling of vulnerability. The patients still remembered their experiences vividly, and it is known that the occasion when a person is informed of having a chronic disease is never forgotten (Ahlström & Sjödén, 1996; Christianson, 1989). The patient’s perception of how the diagnosis was communicated to him or her can therefore leave a lasting impression and long influence the relationship with health-care staff (Johnson, 2003). The patients had been worried, sad and scared about the initial symptoms and the psychological distress was great, also among those with no or mild physical impairment at the time of diagnosis. In sharing their experience of being frightened of the new, unexplained symptoms and the emotional upheaval connected with diagnosis, the patients indicated several important issues. They were met with mistrust and lack of support from health-care staff when presenting diffuse symptoms and complaints. The possibility of a person's having a neurological disease that is difficult to diagnose such as MS should be carefully considered even outside neurological departments, accompanied in the appropriate case by improved psychological support and prompt referral to a neurologist. Another issue was the intensity of negative feelings experienced in connection with falling ill and being diagnosed. The patients described a range of emotional distress and not much of it was reduced by the diagnosis (Johnson, 2003). Struggling with the emotional adjustment was as difficult for the patient as accepting the disease (Lewis, 1998). Anxiety, fear and sorrow remained, leaving the person exposed and vulnerable. This was recognized by health-care staff in some cases but the majority of patients did not receive the needed help and support. The last, though by no means least, important consideration was the uncovering of patients’ ability to manage their vulnerability.

Patients managed their initial shock and devastation in several ways in order to finally be able to accept their illness, which eventually led to the acquisition of the strength needed to face the consequences of having a chronic and progressive disease. One example is the described existential brooding in the context of struggling to find a meaning to their illness, illustrated by statements such as “Why me?” Recent
research has found that MS patients who successfully managed to transform their illness experience into something that made sense or provided a meaning in their life had a significant higher quality of life and life satisfaction (Russell, White, & White, 2006). It should be acknowledged that MS is a disease that evokes existential issues, and nurses should communicate understanding and support to this patient group. This may better enable patients to engage in the aforementioned search for meaning (Folkman & Moskowitz, 2004; Russell, White, & White, 2006). It is equally important to acknowledge that is not fruitful for all patients to discuss such issues related to their diagnosis, since it may cause too much emotional stress. However, for some of the patients the initial illness experience was not perceived as threatening and they did manage to accept the diagnosis with the support and confirmation of family and health-care staff. This diverse range of experiences calls for the discretion of the nurses in handling each patient’s individual needs. In order to provide the patient with the optimal level of support, the family should also be invited to participate in counseling and information-giving sessions. To sum up the findings of Study I, it is important that health-care staff listen to the patients and learn from the patients’ own experiences and ways of successfully managing stress associated with MS, for this will facilitate the giving of the best support in the form of, to take but one example, helping to sustain positive reappraisal (Folkman & Moskowitz, 2004; Livneh & Antonak, 1997). Managing will be further discussed in connection with Studies III and IV.

**Impairment and quality of life in MS patients – Study II**

Living with MS can include both emotional distress and the prospect of having to live with impairments of different kinds. In the self-rating of impairments, 82% of patients noted difficulties with walking and balance in addition to fatigue. No significant differences were found in quality of life between the immunologically treated group and those not receiving such treatment. This result is similar to what has been found in other studies, although some studies did report improvement because of treatment received (Arnoldus et al., 2000; Mitchell, Benito-Leon, Gonzalez, & Rivera-Navarro, 2005; Nortvedt & Riise, 2003). Health-related quality of life was, in any case, reduced in the overall group of patients as compared to the general population, which is in agreement with earlier research (Nortvedt & Riise, 2003). Health-related quality of life is negatively affected by the many medical complications and impairments caused by
MS (Forbes, While, Mathes, & Griffiths, 2006). Disability ratings in EDSS are particularly correlated with health-related quality of life, but the strength of the association varies between studies (Benito-Leon, Morales, Rivera-Navarro, & Mitchell, 2003). Study II revealed that EDSS correlated significantly with six variables of SF-36, most evident with respect to physical function. EDSS is the strongest predictor of the costs of illness of MS, and all types of costs increased with worsening disease (Kobelt, Berg, Lindgren, Fredriksson, & Jonsson, 2006). Immunological treatment that slows down disease progression early on and prevents or delays progression to a more severe level, where patients are unable to work and become dependent on help from healthcare staff or family, will thus provide large cost-benefits to society (Kobelt, Berg, Lindgren, Fredriksson, & Jonsson, 2006). However, personal suffering in connection with bodily deterioration is, of course, the largest cost for the individual and the family. With the progress of medical research there is hope of better treatments in the future, even though it is important not to be overly optimistic and to keep patients’ expectations at a realistic level. Support with regard to psychological and medical issues, as well as rehabilitation, will continue to be vital elements in the long-term care of MS patients (Barker, 2002).

Even if health-related quality of life decreased, the patients’ subjective well-being as shown by the SQoL scores was quite satisfactory in many respects. Despite the infliction of MS, the patients did not estimate their well-being to be reduced overall. Muscular dystrophy patients had, in comparison, lower subjective well-being in a recent study, which may be explained by greater disease impairments and longer disease duration (Boström, 2005). There were also some interesting findings with regard to well-being and quality of life from a gender perspective. Women rated their relation to partner and friends as more satisfying than did men in SQoL. This result may be due to social roles and to the fact that women nurture their relations more than men do (Jung Suh, Moskowitz, Fournier, & Zuroff, 2004). Another gender difference was found in the case of sexual function, which has been shown to be impaired in both men and women with MS (Schmidt, Hofmann, Niederwieser, Kapfhammer, & Bonelli, 2005). However, in this study problems with sexual function as measured by the impairment check-list were more often reported by men, a finding that deserves closer scrutiny in future studies.
To sum up the results of Study II, living with MS constituted a complex situation. It could mean living with impairment and disability that would reduce the health-related quality of life. The patients had to cope with decreased mobility and fatigue, for example. However, their well-being was not generally reduced and many of them clearly were able to manage the problems resulting from MS and adjusted well. The subjective quality of life has to do with the more immaterial values and on the ability to adjust to life as it is, even when one has a severe chronic illness (Kajandi, 2006; Mohr & Cox, 2001). In the sections on Studies III and IV this line of inquiry will continue in the form of qualitative interview studies exploring both illness adjustment and the loss and sorrow caused by MS.

Presence of chronic sorrow in MS patients – Study III

In Study III all 61 MS patients were interviewed with the NCRCS interview-guide constructed to detect chronic sorrow experiences, in addition to investigating how the patients were actually managing such experiences (Study IV). Earlier research has used smaller study groups and reported the presence of chronic sorrow in 80% of the MS patients (Hainsworth, 1994). In the present analysis most with chronic sorrow were easily distinguished from those who had no such sorrow. Thirty-eight (62%) of the participating patients were found to experience chronic sorrow. In comparison, almost all 61 MS patients frequently spoke of the experience of frustration interwoven with anxiety and sadness in connection with the diagnosis (reported in Study I). After managing the initial shock and the devastating feelings related to having a chronic and progressive disease, 23 of the patients came to cope with MS and its accompanying losses without the recurrence of sorrow (recurrence being the characteristic of chronic sorrow). This finding was independent of the level of disability since EDSS did not differ between the groups with and without chronic sorrow. Only one out of 15 impairments on the self-reported impairment check-list differed between these groups, the patients with chronic sorrow having more problems with Fatigue. Physical function and earlier bodily losses were not directly connected with the experiences of chronic sorrow.

With regard to subjective quality of life (see above, Study II), the group with chronic sorrow did have lower well-being, for example a reduced energy level and low self-acceptance. There was an interesting correspondence in the quantitative and
qualitative data of the chronic sorrow group. These patients described losses in terms of loss of social relations and fellow-workers, loss of bodily energy, loss of freedom, loss of hope and confidence (feeling safe and secure) and loss of a healthy identity. This corresponded to the SQoL findings in the chronic sorrow group indicating the patients as having no work, reduced energy, reduced freedom, reduced self-assuredness and reduced self-acceptance (Kajandi, 2006). With regard to health-related quality of life, the chronic sorrow group only experienced a reduction in General health.

Another psychological factor, symptoms of depression, likewise had a direct connection with chronic sorrow. Depressive symptoms as measured by MADRS were more frequently reported in the group with chronic sorrow than among those not experiencing recurrent sorrow, but only four had symptoms of being mildly depressed (Medical Products Agency, 2004). Clinical depression is a complex mixture of emotional and mood disturbances in concert with psychosomatic symptoms such as change in sleep and appetite (Siegert & Abernethy, 2005; Lazarus, 2000). It is known that chronic sorrow indeed can be a risk condition for depression (Lindgren, Burke, Hainsworth, & Eakes, 1992) and that depression is quite common in MS patients (Siegert & Abernethy, 2005; Wallin, Wilken, Turner, Williams, & Kane, 2006). Depression in MS patients is treatable but has been reported to be under-recognized and therefore under-treated (Wallin, Wilken, Turner, Williams, & Kane, 2006). For this reason it is important to recognize symptoms of depression in MS patients, at the same time clearly distinguishing chronic sorrow from clinical depression in order to give the appropriate support in each case (ibid). Nurses and other health-care staff should be aware of these different types of emotional and psychological distress encountered in MS. Furthermore, depressive symptoms were not associated with physical incapacity as rated by EDSS or the physical scales in SF-36.

Characteristics of chronic sorrow in MS – Study III

When interviewing the patients with the NCRCS interview guide, it became clear that many of them had recurring sorrow experiences in connection with the losses caused by MS (Schreier & Droes, 2006). Chronic sorrow is one way of explaining the findings of persistent sadness in their narratives. The patients lost control over the body, over its physical functions, and suffered a loss of energy. This pattern of sadness-related feelings in connection with their losses, and the many physical problems encountered
as shown by the self-reported impairment check-list, can fittingly be described in terms of having chronic sorrow. In a previous study with more severely physically disabled patients, chronic sorrow was frequently described in terms of loss of bodily function (Ahlström, 2006).

Loss of hope was frequently expressed as a consequence of receiving the MS diagnosis and of the MS-related losses. The patients lost hope and confidence, lost the expectation of having a long life. The sense of hopelessness has been described in earlier research in connection with receiving the diagnosis of MS, as well as different feelings of loss (Miller, 1997). Gradually learning to cope with having received the diagnosis did not put an end to the patient’s emotional distress; new losses brought back sadness and vulnerability. The patients with rather low levels of impairment, some of these patients quite young, had lost hope just the same. They were deprived of a hopeful future, of dreams and personal possibilities, of education and of building a family — all this contributing to the development of chronic sorrow (Roos, 2002). Chronic sorrow experiences were also described in terms of loss of integrity and dignity. Not being treated and seen as a whole person (Widäng & Fridlund, 2003) led to the loss of the sense of human dignity. To sum up the results of Study III, the patients in the chronic sorrow group clearly experienced emotional distress related to repeated incidences of sorrow leading to chronic sorrow. It is important to understand that the essence of the sorrow could derive from losses either physical, social or psychological in nature. Nurses and other health-care staff must provide support and intervention to alleviate the suffering caused by chronic sorrow.

**Managing chronic sorrow, Study IV**

The patients’ ways of managing chronic sorrow were investigated in Study IV. Personal efforts in management are named *internal* management in the theory of chronic sorrow (Eakes, Burke & Hainsworth, 1998). Ineffective internal management had only two sub-themes emerging in the qualitative analysis, Caught in worries and Living in denial, which may reflect two obstacles to the successful management of chronic sorrow. Denial in itself is neither good nor bad; it depends upon the context, and denial can be part of a coping process where the intrusion of reality is temporarily blocked off, giving some respite from overwhelming stress (Cramer & College, 2000; Lazarus & Folkman, 1984). With respect to anxiety and worrying, each patient
naturally felt great concern about the progressive nature of MS and its accompanying impairments. Anxiety is known to be high after the initial MS diagnosis, both amongst patients and their partners, and the patient is exposed and vulnerable (Janssens, van Doorn, de Boer, van der Meche et al., 2003). Lack of knowledge and understanding of patients’ needs on the part of both health-care staff and family members characterizes ineffective external managing of emotional distress and sorrow (Burke, Eakes, & Hainsworth, 1999). Therefore it is vital to include the family when giving information about MS, the experienced loss of function as the disease progresses and the phenomenon of chronic sorrow. It is, of course, a main responsibility of health-care staff to update their knowledge of chronic sorrow, as well as their ability to support the management of losses and emotional distress in connection with MS (Hainsworth, Burke, Lindgren, & Eakes, 1993; Schreier & Droes, 2006; Williamson, 2000).

Several ways of successful internal management of chronic sorrow were described in Study IV, from distraction to taking care of oneself. Where the appropriate external management existed in the form of caring family and confirming health-care staff, patients increased their internal managing skills for handling distress. The results of Study IV concerning the outcome of the successful management of chronic sorrow can be summarized as follows: the gain of personal insight, personal confidence and hope, which show that the patients had experienced personal growth. This is similar to Mohr’s finding of more positive than negative statements when interviewing MS patients about their experiences of the disease (Mohr et al., 1999). The experiences of positive benefits have also been found to be in the majority in earlier research (Pakenham, 2005). This beneficial outcome of managing severe illness should be supported and acknowledged (Somerfield & McCrae, 2000).

**Critical reflections on the theory of chronic sorrow**

The research in Study III and Study IV was based upon and drew inspiration from the theory of chronic sorrow, and the findings confirm the theory’s premises (Eakes, Burke, & Hainsworth, 1998; Schreier & Droes, 2006). However, the presence of chronic sorrow was not as frequent in this group of patients as has been reported in earlier research on chronically ill individuals (Eakes, 1993; Hainsworth, 1994; Lindgren, 1996). In those studies convenience samples of patients were recruited from support groups, MS societies, or recruited by a nurse at a cancer clinic. In this study all
61 participating patients were interviewed regardless of earlier expressions of sorrow experiences. Even so, in 62% of the patients experiences of chronic sorrow were detected, which is in line with a more recent study on chronic sorrow on patients with chronic diseases (Ahlström, 2006). This emotional aspect of loss experiences should therefore be considered in the encounter with MS patients.

One criticism of the theory is with regard to the trigger factor of sorrow, which was difficult to separate from the periodic episodes of sorrow resulting from the burden of continuing physical loss experiences in this study (Figure 1) (Eakes, Burke, & Hainsworth, 1998). In the patients’ descriptions of their recurrent sorrow experiences, those trigger factors were in some cases easy to distinguish, in others not. For instance, if seeing another person in a wheelchair triggered sorrow, this would be in line with the trigger factor in the theory. However, when the patient said that sorrow was triggered by a bout of MS it was instead estimated to be a case of periodic or cyclical sorrow since occurrence events initiate periods of sorrow in this theory (Schreier & Droes, 2006). Furthermore, the experience of being interviewed about one’s losses and sorrows may in itself be a trigger. It was therefore imperative that the study participants had access to skilled health-care staff that could provide professional help if needed.

Another concern is the theory’s proposal that chronic sorrow management is either effective or ineffective (Eakes, Burke, & Hainsworth, 1998), and the authors of the theory refer to Lazarus and Folkman’s theories of stress and coping (1984). However, judgment of the efficacy of certain ways of coping or managing is a strategic mistake according to Lazarus (2000). The overall process of managing stress and sorrows includes many different strategies, which will vary in their effectiveness depending upon the specific situations encountered (Lazarus & Folkman, 1984). It is rather the combined effect of management that, in the end, will enable the patient to adequately handle the distress of having MS. Therefore the theory may improve as an explanatory model with the development of a more process-adapted way of describing the management of chronic sorrow (Folkman & Moskowitz, 2004). In the next section the inherent duality in the process of managing chronic sorrow will be further explored.
The loss and creation of hope

Loss of hope was identified as one category in Study I from the way the patients were experiencing the initial symptoms and diagnosis. In managing the diagnosis, one important element was the effort to sustain hope. Similarly, patients described loss of hope as an essential element of chronic sorrow in Studies III and IV. With nothing to hope for, there is just despair. Without hope for a better life, feelings of hopelessness will arise. Helplessness, hopelessness and despair overlap in their definitions (Lazarus, 1999), and in the present study patients expressed despair in connection with the loss of hope for a life in good health. These experiences do not imply that the persons felt helpless in the sense of feeling powerless, since they did not view themselves as powerless victims in circumstances not directly related to MS.

It is interesting to explore how the patients were managing the diagnosis and chronic sorrow experiences where initially loss of hope was described. They evidently managed by again creating hope in their lives. They were feeling deprived of hope, but they managed to regain it. Hope is a personal belief in the possibility of a positive outcome and it is also an emotional response to having a goal (Lazarus, 1999). Hope can stimulate efforts to seek ways out of an unsatisfactory situation. In despair and depression hope is not perceived, often leading to a dysfunctional state (ibid.). Almost all of the patients with chronic sorrow expressed loss of hope, but interestingly enough they had few symptoms of severe depression. In addition, many of them spoke of regaining their hope through the advances of science in an effort to manage their feelings of hopelessness. Even if we believe we are unable to radically influence the outcome, we can hope for a favorable intervention — from God, from a scientific breakthrough, from some other person or agency, or simply through good luck (Lazarus, 1999). If an illness or bodily function worsens, what we hope for must change in order to be in line with the shifting of reality, whilst if there is an improvement new hope can be entertained; and in this study the sense of hope varied in accordance with the illness trajectory and forthcoming losses. The way of coping should also change with the efforts that must be made to facilitate managing and to reduce stress (Folkman & Moskowitz, 2000; Folkman & Moskowitz, 2004). This is quite like the results of Study IV, where the growth of hope and comfort was a result of successfully managing sorrow. However, even after adequately managing one loss the patient could still be feeling sorrow and experience renewed loss of hope when a
new loss occurred. Successful management does not protect from the impact of new losses in the form of bouts of MS, increasing impairment, loss of work and so on, which may draw the patient back into sorrow. Likewise, the cycle of gain and loss of hope will continue in such a chronic and progressive disease as MS.

**Vulnerability contra strength**

As shown in Study IV, managing the adverse effects of a disease like MS is difficult and demands a lot of effort on the part of the patient. At the same time it may also bring the benefits of personal growth (Folkman & Moskowitz, 2004). In mapping out how the patients were managing chronic sorrow from the perspective of the theory of chronic sorrow, both vulnerability and strength were detected. Falling ill with MS was described in this study as a process of becoming vulnerable and staying so until sufficient strength has been acquired to manage the situation. This involved accepting and adjusting to MS and the consequences of new losses in the form of both physical deficits and psychological distress — a time-consuming process (Chesla, 2005).

Managing difficulties successfully thus required strength, at the same time as the patient would gain strength from the achievement of handling distress well (Reynolds & Prior, 2003). Chronic sorrow was connected with the entire illness trajectory as described above, starting with the diagnosis and continuing throughout life, demanding an unusual measure of strength in this patient group, involving the management of their sadness and unstable mood.

Discomfort was described in connection with the unsuccessful managing of chronic sorrow. Some of the patients lived in a continual circle of loss and sorrow (Schreier & Droes, 2006). After gaining some level of strength, once again these patients were experiencing vulnerability and distress when they were confronted with new losses. Lack of support, from family, friends and health-care staff, was spoken of, and patients were very exposed to the impact of MS on all levels of life. Strength and comfort were mentioned when patients were speaking of successfully managing chronic sorrow. In some cases, patients’ accounts of their handling of loss and sorrow became an extended description of a process of maturation and personal growth in the form of increased insight, confidence and hope. This long and tortuous process often included finding a meaning in their experiences and the subsequent emergence of newly founded strength. Finding a meaning did not imply a passive attitude: it was not a question of
waiting for good things to happen, but a question of making an effort to bring them about. It could also include taking an ordinary event that in itself was affectively neutral and infusing it with positive meaning, a form of positive reappraisal (Folkman & Moskowitz, 2004). Patients in this study described how they managed to feel better by listening to the birds singing in the spring, by going for an inspiring walk in the forest or by planning enjoyable activities. The role of positive emotions in the process of managing stress should therefore not be underestimated while at the same time recognizing that positive and negative emotions co-occur throughout this process (Folkman & Moskowitz, 2004). Also spoken of was the perception of the actual benefits of the illness, such as closer relationships with family and friends, the reprioritization of goals and greater appreciation of life. All the management skills provided the patients with increased mental and emotional strength. Such benefits and personal changes have been called stress-related growth and benefit findings (Folkman & Moskowitz, 2000; Pakenham, 2005), which this patient group exhibited to an unusual degree considering the nature of MS.
Methodological considerations

**Mixed-methods design**

A mixed-methods design is suitable when investigating several aspects of a research topic and combining qualitative and quantitative approaches in the same project. The overall purpose of this thesis was to gain new knowledge of MS patients, including their experiences of illness and their everyday life as influenced by MS. Each study and each specific aim has been carefully considered with respect to the qualitative or quantitative approaches employed (Tashakkori & Tedde, 2003). The four specific studies in the thesis are complete investigations in themselves but are also part of the overall project. Randomization and having a large sample size is of great concern in quantitative studies whereas a smaller, purposeful sample is usually considered sufficient in qualitative studies. Combining these different approaches in the same project is often impractical (Morgan, 1998), but it was decided to pursue a mixed-methods design in this thesis in order to gain the maximum amount of data. In the quantitative part of the project a random selection of a number of patients receiving immunological treatment was used in conjunction with a matched-pairs design to achieve acceptable validity and statistical power (Altman, 1991). In the qualitative part of the project it was decided to include all recruited patients and the interviews generated were subsequently subjected to content analysis. The main researcher has been working as an MS nurse and has met this patient population in her work at the neurology department. The research team involved in the thesis has been aware of that and recognizes the importance of separating the nurse’s clinical role from her role as a researcher. Nevertheless, the researcher’s clinical experience and knowledge in the area may also have been beneficial for the unfolding of knowledge in the research process.

**Patient selection**

The reason for choosing patients that had already received immunological treatment was both pragmatic and ethical in nature since this kind of treatment is already in clinical use. Furthermore, the focus of the thesis was not on treatment effects but on quality of life. Quality of life is of interest in ongoing clinical treatment and can be more thoroughly investigated with the use of a matched, untreated control group. Studies have investigated the effect of immunological treatment on quality of life, some finding a beneficial effect and others not (Mitchell, Benito-Leon, Gonzalez, & Rivera-
Navarro, 2005; Nortvedt & Riise, 2003). Patients participating in this thesis project were selected from a local MS register including all patients in a county in the middle of Sweden (with both urban and rural population). Time since diagnosis had to be at least 6 months to avoid including patients in acute crisis. The upper limit was set at 15 years since diagnosis so the patient would have no difficulty in remembering the initial illness experience in the interviews (Study I). In the final study group only three patients had longer time than ten years since diagnosis and all patients remembered their diagnosis experiences vividly. Only patients that, according to their medical records, were cognitively able to complete questionnaires were included. The distribution of gender was controlled in Study I in order to have the same prevalence as in the population, i.e. twice as many women as men (Compston & Coles, 2002). In this study none of the patients experienced symptoms of moderate to severe depression. Possibly patients with lower mood chose not to participate in a study about living with MS with two interviews and the filling in of several questionnaires.

**Matching participants and attrition**

The fact that the design of Study II was comparative and correlative further governed the selection of participants. The aim in Study II was to study group differences of quality of life and impairment between patients receiving immunological treatment and patients not receiving such treatment. From the county register, 31 treated patients were chosen at random and then matched with untreated patients. The matched-pair design minimizes the differences between groups (Altman, 1991). It was difficult to fulfil all the four matching criteria even though the group of untreated patients was quite large — 107. There were four patients from the treated group that declined participation, and four new registered and treated patients replaced them. Twelve out of 31 matched, untreated patients had to be replaced since 11 declined to participate and one moved abroad. The matching criterion age became slightly higher when replacing drop-outs with new subjects, the untreated group being on average seven years older. The difference in age may have influenced the results since age can be a determining factor in health-related quality of life — the older the person, the more reduced the person’s health-related quality of life (Sullivan, Karlsson, & Ware, 1994). However, gender, EDSS and time since diagnosis did not differ between the groups. In addition, disease pattern did show a more progressive pattern in men than in women,
as well as in treated versus untreated patients. However, since disability rating did not differ between gender or study groups, it is unlikely that disease pattern influenced the results of this study. After the inclusion phase was completed, one untreated patient dropped out, the study group thus comprising 61 patients at the end. In Study II there was further internal drop-out. A patient in one matched pair had ended the treatment without informing the doctor; another pair had to be excluded since one patient did not return the SF-36 questionnaire, and during the period between the data collections another matched pair was excluded because of treatment change. Validity can be threatened in the case of attrition and if power is reduced (Kazdin, 2003). However, the remaining sample was sufficiently large to conduct statistical analysis with acceptable power (Altman, 1991; O’Brien & Muller, 1993).

Questionnaires used in this project
The SF-36, SQoL and MADRS used in the research project were validated questionnaires and had undergone psychometric testing with satisfactory results (Kajandi, 1994; Montgomery & Asberg, 1979; Sullivan & Karlsson, 1998). The Self-reported Impairment Check-list had been used in a previous study with stroke patients (Widar & Ahlström, 2002) and was adapted in this project for use on MS patients and their manifestation of impairment. For example, MS patients may find that some symptoms fluctuate while other symptoms are more permanent in nature, therefore the duration of problems was added to the scale. Mostly the impairments were reported as being constant. The self-rating scale can be criticized in that it lacks an item regarding problems with the bowel, and such an item should be added to the scale since earlier research reported problems with the bowel in 12% of the MS patients (Ford, Gerry, Johnson, & Tennat, 2001). The impairment list was easy to assess and could be a valuable complement to disability rating in EDSS for the purpose of exploring difficulties arising from the disease.

When performing many correlational analyses between the subscales of the impairment scale and SF-36 in Study II, there is always a risk of false positives with so many comparisons between different variables. SF-36 has been used in many studies on MS despite criticism of the floor effect in patients with severe disability (Benito-Leon, Morales, Rivera-Navarro, & Mitchell, 2003; Nortvedt & Riise, 2003). In this study patients mostly with mild to moderate disability participated and therefore the
risk of floor effect was reduced. One advantage in using SF-36 is that results from different studies can be compared.

The time interval between the interviews was approximately four months. Notes were taken at the second interview of life events that could have affected the results of the research. The summer season was in between the two interviews and most of the patients had enjoyed the summer holiday. Data analyses of SQoL were conducted in such a way as to investigate if patients with positive or negative life event experiences changed the between-group comparisons, but that was not the case. The analysis was conducted first by excluding the patients with negative life event experiences (n=4) and then patients with positive ones (n=8) and it showed the same pattern, i.e. no differences were found between the groups.

Trustworthiness of the qualitative analysis
In the content analysis in Study I the computer software N Vivo was used in sorting the text of the 61 interviews (Richards, 2002). The program is useful in that the researcher can move back and forth from the entirety of the text to the meaning units deriving from the analysis (Lieblich, Tuval-Mashiach, & Zilber, 1998). In this study this program was used as a practical way of selecting meaning units for further analysis and sorting them in the context of the large amount of interview text comprising what was said by all 61 patients. The inductive analysis was continued through the condensation of the text into Word files and these were printed out to be critically reviewed by a second researcher, the co-author in all four studies. The various steps in the analysis were scrutinized by both the participating researchers and discussed, this in order to achieve a high level of trustworthiness (Graneheim & Lundman, 2004). To promote continuity in the qualitative process, the analysis was directed towards both the whole emerging from the text and its constituent parts (Downe-Wambolt, 1992). The knowledge gained can be of use to nurses and other health-care staff in contact with patients who have the initial symptoms of what could develop as MS as well as when it comes to having to inform the patient of the final diagnosis.

The theory-guided qualitative analysis
Studies III and IV demonstrate the value and potential of using the theoretical model of chronic sorrow when analysing empirical data. The assessment of the identification of
chronic sorrow in the 61 interviews by means of the interview-guide NCRCS in Study III was done deductively in accordance with the predetermined criteria from the literature (Eakes, Burke, & Hainsworth, 1998). The second researcher evaluated uncertain cases to ensure credibility. The group of 38 patients with chronic sorrow offered a large variation in the data when it came to qualitative content analysis of the narratives. The text was read and inductive analysis was performed in several steps and the both researchers (main author and co-author) critically reviewed the process in order to establish stability (Downe-Wambolt, 1992). The study was in line with the theory of chronic sorrow (Eakes, Burke, & Hainsworth, 1998) and previous research (Hainsworth, Burke, Lindgren, & Eakes, 1993), with the extended aim of discovering the meaning of the losses that caused the sorrow reaction among the patients, this through inductive analysis (Ahlström, 2006). The analysis in Study IV continued to explore the way these MS patients were managing their chronic sorrow by use of the theory of chronic sorrow in sorting the text and then inductively analysing the contents (Graneheim & Lundman, 2004). In this study, factors ensuring trustworthiness were also investigated by critically scrutinizing the analytical process and the emergence of results.

In Study IV the theoretical model indicates that ineffective management of chronic sorrow leads to discomfort, effective management to comfort, and this makes intuitive sense (Schreier & Droes, 2006), but it is important to emphasize that this study was not designed to investigate or verify a causal connection a specific kind of managing and a particular outcome. Underlying factors that are not studied may also be involved. In following the model the study describes patterns of managing on the part of the patients and how they experienced their situation. Using the theory of chronic sorrow deductively in the empirical data showed that valuable knowledge and understanding could be gained concerning the chronic sorrow experiences of patients with MS. Other researchers interested in the area are invited to scrutinize and evaluate the findings and thereby promote further development of the theory of chronic sorrow with respect to MS and other chronic and progressive diseases.
Conclusions

This study has deepened our understanding and knowledge of patients with MS in relation to how they experienced falling ill and how they come to terms with living with the disease, including the influence of impairment and chronic sorrow on quality of life. The initial MS experience gave the patient a sense of having become vulnerable. This psychological distress can be high regardless of level of physical impairment at the time of diagnosis. The patients felt exposed and vulnerable until they could manage their emotional distress and the disease in such a way that they could reappraise their situation and again feel confidence in their lives and be hopeful about the future.

No differences in health-related quality of life or subjective estimation of quality of life were found between an immunologically treated patient group and a matched control group. MS reduced health-related quality of life but not overall well-being. In the subjective estimation of quality of life, lack of energy, not having work, not having a partner and not having children were the source of greatest dissatisfaction in this group of MS patients. The self-reported impairment check-list captured the patients’ various problems and complements EDSS. The scale showed association with health-related quality of life, most evident in physical function such as walking and pain.

Presence of chronic sorrow in the study group was 62% and characterized by emotional distress, but only four in the group were mildly depressed. The theory of chronic sorrow increases the understanding of the recurrent sadness and low mood reported in chronic illness populations and is new knowledge relevant to the understanding of the consequences of MS. The essence of chronic sorrow in the present case was a pattern of losses where the loss of hope was central, together with loss of control over the body, loss of integrity and loss of identity. The sorrow could be psychological, physical or social in nature, which it is important to keep in mind when it comes to providing suitable support for such patients.

Patients were caught in discomfort by the ineffective management of chronic sorrow and spoke of recurrent sorrow deriving from vulnerability due to the unsuccessful managing. Some also experienced a lack of support from family, friends and health-care staff. Effective management frequently meant increased comfort instead. Personal growth in the form of insight, confidence and hope was a positive
finding with regard to the patients’ experience of managing chronic sorrow. A caring family, friends and supportive health-care staff sustained successful managing of the illness experiences.

The theoretical model of chronic sorrow was useful in sorting interview data when performing the analysis, which also elucidated the connection between theory and empirical data, and the results indicated the model’s empirical trustworthiness. The results especially highlighted the patients’ twofold experience of being afflicted with MS. The patients were initially suffering from emotional distress but they also developed their innate capacity and resources in the course of managing this distress. It is important to identify vulnerable and exposed patients suffering from losses and emotional distress in addition to lacking the needed support from their families and friends. This sub-group of patients should be the main concern for the MS team. In addition, this study reports evidence of positive outcomes deriving from the successful management of illness experiences and chronic sorrow. The patients described ways of managing the emotional distress in connection with the diagnosis and chronic sorrow in order to be able to develop hope, personal growth, appreciation for life and trust in the future.
Implications

• With increased knowledge and understanding of the individual experiences of patients, nurses and other health-care staff can improve the care of MS patients.

• It is important to identify patients exposed and vulnerable during the initial illness experience and directly after diagnosis, this in order to provide sufficient follow-up with the appropriate confirmation of the patient’s needs and to prevent the patient’s feeling abandoned by health-care staff.

• Establishing contact with the MS nurse directly after diagnosis is one way to provide further follow-up. The patient can then easily get in contact with other members of the MS team, for instance the physiotherapist, occupational therapist, social worker and doctor.

• Nursing should be conducted through a comprehensive approach involving the patient’s family if this is welcomed by the patient. It can mean including the family in counselling and information-giving sessions.

• Nurses and other members of the MS team should closely follow how the patients manage the problems of impairment and disability. The self-reported impairment check-list can be used for screening for problems when patients come to the clinic. Information about the disease and about available rehabilitation and medical treatment for impairment can help patients feel confident and in control of their situation. Patient education should involve the families to increase the understanding of MS and promote illness management within the family. Nursing interventions should include recognition and affirmation of the patient's and the family's resources as a rewarding means of managing effectively.

• Some patients need support in adjusting to disabilities brought about by MS. After adjusting to such disabilities, patients usually manage to accept their situation and report an acceptable level of well-being.
• Chronic sorrow is a consequence of losses due to MS that needs to be considered in health care. It is important to be aware of recurrent sorrow and to be able to distinguish chronic sorrow from clinical depression. Depression is frequently experienced by MS patients and should be treated with therapy or medication.

• The theoretical model of chronic sorrow provides a frame of reference within which the phenomena of recurrent sadness connected with losses brought about by MS can be explored. The model highlights the nature of the distress experienced and facilitates communication within the MS team with regard to the individual patient’s problems. This plays a vital part in the identification of patients with chronic sorrow.

• Knowledge of the meaning of chronic sorrow can contribute to the nurse’s ability to provide psychological support and promote a dialogue with the MS patient concerning the sorrow. Chronic sorrow should be acknowledged as a normal reaction to the tragic losses caused by MS. Management of losses and the accompanying sorrow should involve both the patient and the family, which will contribute towards solving illness-related problems and strengthen illness management in the family.

• Nurses should give psychological care by communicating their understanding of distress, vulnerability and chronic sorrow when listening to the individual patient’s needs. Health problems and impairment can be closely related to the emotional experiences, the vulnerability and feeling of being exposed. Nurses should be promoting a sense of hope and control, encouraging a positive self-esteem and supplying relevant information to patient and family. In addition, nurses should improve their recognition of the positive effects of managing chronic sorrow, as well as their recognition of the distress arising from the experience of chronic sorrow in relation to losses caused by MS.

• Finally, this study presents implications for further research. There must be continuing attention to medical, nursing and rehabilitation efforts in improving care for MS patients. The progressive nature of MS gives rise to deficits of a physical and psychological nature whereby the patient’s vulnerability may increase. There should be further investigation of the positive outcomes deriving from the successful management
of MS, followed by recommendations as to how nurses and other health-care staff can sustain such management. An intervention of chronic sorrow support should be tested and evaluated to provide evidence-based care in MS in the case of the presence of this emotional distress.
Swedish summery/Svensk sammanfattning

Kronisk sorg och livskvalitet bland patienter med multipel skleros

Bakgrund
Multipel skleros (MS) är en kronisk neurologisk sjukdom med episoder av inflammation i det centrala nervsystemet, vilket leder till olika symptom för patienten. Dessa inflammatoriska processer kan variera i lokalisation och svårighetsgrad. Initialt läker symptom oftast ut efter en sjukdomsperiod, medan de kan kvarstå som funktionsnedsättning senare i sjukdomsförloppet. Patienter med MS måste anpassa sig och leva med många olika sjukdomsbesvär såsom gångproblem, känselstörningar och trötthet bland andra. Sjukdomssymtom och kvarstående funktionsnedsättning kan inverka negativt på familjeliv, arbete och möjligheter till sociala relationer.

Immunmodulerande läkemedel med en uppbromsande effekt på sjukdomsutvecklingen finns sedan några år; det är dock ingen kurativ behandling. Läkemedelsbehandling av besvärande symtom som till exempel smärta är av stor vikt för patienternas välbefinnande och även rehabiliteringsåtgärder efter perioder av försämring.


Syfte
Det övergripande syftet med avhandlingen är att öka förståelsen för och kunskapen om hur det är att leva med MS. Forskningsprojektet har fyra delstudier med följande syften:
I Att beskriva patienternas uppfattning av MS före diagnos samt deras upplevelse av initiala symtom och diagnos.
II Att beskriva livskvalitet hos patienter som behandlats med immunmodulerande behandling jämfört med en liknande patientgrupp utan sådan behandling samt att undersöka förhållandet mellan upplevda sjukdomsbesvär och livskvalitet för dessa patienter.

III Att beskriva förekomst av kronisk sorg och depression i patientgruppen samt undersöka innebörden av kronisk sorg i denna studiegrupp.

IV Att beskriva på vilket sätt MS-patienterna hanterade kronisk sorg och relatera detta till den teoretiska modellen av kronisk sorg.

Material och metod

Detta forskningsprojekt har en design med mixade metoder som inkluderar deskriptiv, komparativ och korrelativ design, Tabell 1. Deltagare valdes ut genom att 31 patienter med immunomodulerande behandling lottades till deltagande, varefter de matchades mot 31 patienter utan sådan behandling vad gäller kön, funktionsnedsättning, tid sedan diagnos och ålder. En patient avbröt deltagandet varefter 61 patienter deltog i studien.

Datainsamlingen genomfördes vid två tillfällen, båda gångerna med intervju och frågeformulär. De senare undersökte förekomsten av sjukdomsbesvär, hälsorelaterad livskvalitet, subjektiv livskvalitet (välbefinnande) samt depression. Intervjuerna berörde initiala sjukdomsbesvär och diagnos vid första tillfället och upplevelserna av kronisk sorg vid det andra. Intervjuerna analyserades med innehållsanalys och frågeformulärens svar med statistisk beräkning. Den teoretiska modellen om kronisk sorg gav struktur i analysen av delstudie IV.
Tabell 1. Delstudier i avhandlingen

<table>
<thead>
<tr>
<th>Design</th>
<th>Metod</th>
<th>Analys</th>
</tr>
</thead>
<tbody>
<tr>
<td>I</td>
<td>Deskriptiv Kvalitativ</td>
<td>Intervju med MS-patienter om initiala sjukdomsupplevelser (n=61).</td>
</tr>
<tr>
<td>II</td>
<td>Deskriptiv Komparativ Korrelativ</td>
<td>Tre frågeformulär: Självskattade sjukdomsbesvär, Hälsorelaterad livskvalitet (SF-36) och Subjektiv livskvalitet (SQoL) (n=58).</td>
</tr>
<tr>
<td>III</td>
<td>Deskriptiv Kvalitativ</td>
<td>Intervju med MS-patienter om kronisk sorg, intervju guide (NCRCS) och självskattning av depression (MADRS) (n=61).</td>
</tr>
<tr>
<td>IV</td>
<td>Deskriptiv Kvalitativ</td>
<td>Intervjuer med MS-patienter om erfarenheter av att hantera kronisk sorg, intervju guide (NCRCS) (n=38).</td>
</tr>
</tbody>
</table>

**Resultat**

Insjuknandet och diagnosen orsakade stor känslomässig påverkan för de 61 patienterna, delstudie I. De hade upplevt sig utsatta och sårbara när de fick symtom och blev oroliga för deras orsak. Rädsla för att det kunde vara livshotande, men även måttlig oro, fanns beskrivet. Patienter hade mött misstro för sina symtom, speciellt om neurologisk specialistkompetens saknades. Tiden runt diagnosen hade varit känslomässigt svår för de flesta, och diagnosen minskade inte oron eller ovissheten. Patienterna hade upplevt att hoppet och tryggheten inför framtiden försvann. Genom personliga ansträngningar att hantera att de blivit sjuka kunde de till slut acceptera sjukdomen samt uppskatta nya värden i livet och finna nytt hopp.
Det fanns ingen statistisk skillnad i MS-besvär eller livskvalitet mellan patientgruppen som behandlades med immunomodulerande behandling jämfört med den utan sådan behandling, delstudie II. Hela gruppen hade många MS-relaterade besvär i varierande grad; 82 % upplevde besvär med gångförmågan och balans/yrsel, och även trötthet var vanligt. Den hälsorelaterade livskvaliteten var reducerad i gruppen, medan den subjektiva livskvaliteten inte var nedsatt, utan patienterna hade i stor utsträckning ett gott välbefinnande.

Kronisk sorg fanns hos 62 % av patientgruppen, delstudie III. Innebörden för dessa MS-patienter som led av kronisk sorg var förlust av hopp och förlust av kontroll över sin kropp i stor utsträckning. De hade förlorat tillförsikten inför framtiden och istället såg de sjukdomsutveckling och restriktioner. Funktionsnedsättningar och trötthet hade förändrat kroppen, och de kunde inte kontrollera den som tidigare. Även förlust av integritet, värdighet och identitet beskrevs av patienterna.


Sammanfattning

Denna studie har fördjupat förståelsen för och kunskapen om hur det är att leva med MS i relation till erfarenheter vid insjuknandet, om aspekter av sjukdomsbesvär och livskvalitet samt om erfarenheter av kronisk sorg och hur patienterna hanterade denna känslomässiga påfrestning.

Insjuknande och diagnos var känslomässigt påfrestande för patienterna. De hanterade denna stress, och med tiden återfann de tillförsikt i livet. Patienterna hade många besvär på grund av sin sjukdom, men deras välbefinnande var inte starkt nedsatt. Erfarenheter av kronisk sorg fanns hos 62 % med periodvis sorg och frustration, men endast fyra i gruppen hade symtom av mild depression. Innebörden av förlusterna som orsakade kronisk sorg visade sig vara olika i denna grupp av MS-patienter. Mest frekvent beskrevs dessa förluster i form av förlorat hopp och förlust av kontroll över sin kropp. Innebörden av kronisk sorg kan vara förluster av psykologisk, fysisk eller social natur. Den negativa stressen återkom eller fanns kvar då de inte klarade av att hantera sorgen över återkommande förluster på grund av brist på stöd eller därfor att den egna oron var för stark. Andra patienter hanterade sin sorg mer lyckosamt samt erhöll stöd, vilket ökade deras välbefinnande. Genom att hantera den känslomässiga påfrestning som diagnosen och den känslomässiga belastningen vid kronisk sorg innebar kunde patienterna finna insikt och styrka.

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