THE PSYCHOLOGICAL ASPECTS AND MANAGEMENT OF CHRONIC FATIGUE SYNDROME

A Thesis submitted for the degree of Doctor of Philosophy

by

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ABSTRACT

Chronic fatigue syndrome (CFS) describes a condition characterised by severe fatigue of at least six months' duration. In this thesis, it is argued that the complexity of CFS with respect to other symptoms, the patients' response to their illness and the determinants of emotional distress, has yet to be fully recognised. This may have narrowed the focus of research and limited the range of treatments available.

The first of the three studies investigated CFS from the patients' perspective. The findings challenge some of the generalisations concerning CFS, particularly those relating to the patients' attributions and their choice of coping strategies. They also suggest that the effects of the condition may have been underestimated and that certain influences on emotional distress may have been overlooked.

The second study assessed a number of variables thought to be associated with emotional distress and psychological adjustment. The results show that uncertainty and lack of social support were significantly correlated with anxiety and depression while functional impairment was more closely linked to cognitive difficulties and other illness-related measures.

The third study evaluated a management programme which acknowledges the complexity of CFS. After six months, significant differences between the treated patients and waiting-list controls were found for a number of variables, including fatigue, somatic symptoms, anxiety and perceived self-efficacy. However, many patients continued to record high levels of emotional distress, showing that the programme was not sufficient to deal with all the problems experienced.

The findings suggest that variables such as uncertainty, lack of social support, self-efficacy and illness severity may all play an important role in the psychological adjustment to CFS. Increased awareness of these possible influences may further enhance understanding of this illness and thus improve patient care.

CONTENTS

ABBREVIATIONS

CBV	Coxsackie B virus
CFS	Chronic fatigue syndrome
CFS(ME)	Patients diagnosed with myalgic encephalomyelitis or post-viral fatigue syndrome
CFS(PIFS)	Patients who fulfil criteria for post-infectious fatigue syndrome
DSM-III-R	Diagnostic and Statistical Manual of Mental Disorders Third Edition-Revised
EBV	Epstein-Barr virus
HAD	Hospital Anxiety and Depression Scale
IMQ	Illness Management Questionnnaire
MUIS	Mishel Uncertainty in Illness Scale
PFRS	Profile of Fatigue-Related Symptoms
SCI	Spinal cord injury

CHAPTER 1

Myalgic encephalomyelitis and chronic fatigue syndrome. A brief review of the literature

1.1 Introduction

Chronic fatigue syndrome (CFS) is the term used to describe a number of disorders characterised by disabling, ongoing fatigue. Although the nomenclature for these conditions is comparatively new, the disorders themselves are not. Indeed, references to illnesses closely resembling CFS have been documented in the British literature since 1750 (Bakheit 1993). Some cases of CFS are closely associated with and may represent psychiatric disorders (David 1991, Hickie et al 1995, Wessely 1994). Others have been linked with infections such as glandular fever and Lyme disease (Bruce-Jones et al 1994, Coyle et al 1994) and exposure to toxins (Behan and Haniffah 1994, Chester and Levine 1994). However, much of the attention in recent years has focused on a mysterious illness known as myalgic encephalomyelitis (ME) or post-viral fatigue syndrome (PVFS).

The following review will describe the history and clinical features of ME and discuss some of the research and controversies relating to CFS in general and ME in particular.

1.1.1 Historical background

The first detailed description of the illness ME can be found in a report on the epidemic at the Los Angeles County General Hospital in 1934 (Gilliam 1938). The outbreak, which coincided with an epidemic of poliomyelitis, lasted seven months and affected 198 members of staff. The presenting symptoms included headache, nausea, sensory disturbances e.g. numbness, and a stiff neck or back. Localized muscular weakness developed in 80% of the cases

and paraesthesiae in 42%. Some patients also suffered from sore throat, fever, "easy fatigue" and "emotional instability".

The illness generally lasted between four and eight weeks but a significant proportion of the patients experienced relapses. Some of these were attributed to overexertion, others to cold and damp weather. A few, however, were deemed to be "hysterical" in nature.

Aside from the relapses, the illness had a number of other unusual features. For instance, there was a marked variability in the severity of symptoms and, with regard to muscle weakness, a variability in location. There were also variations in physical signs so that a group of muscles sometimes performed normally on one test but abnormally a week later.

Another unusual characteristic of the disorder was the protracted recovery. In fact, Gilliam observed that 55% of the patients continued to experience symptoms and were still unwell and absent from duty 28 weeks later.

Although the initial symptoms were suggestive of poliomyelitis, this diagnosis was rejected for a number of reasons. For instance, there was no evidence of severe muscle wasting which is a characteristic feature of polio (Ramsay 1988). Moreover, the muscle pain and tenderness in these patients persisted much longer than expected, the cerebrospinal fluid was invariably normal and the mortality rate was far lower than that associated with polio. Nevertheless, Gilliam believed that there was a connection between the illness and the polio virus.

Since then, similar epidemics have been documented throughout the world (Acheson 1959, Jenkins 1991a, Ramsay 1988). Virtually all of these featured symptoms and signs sug-

gesting the involvement of the brain, spinal cord and peripheral nerves. Moreover, most occurred between the months of May and November; they had a bi-phasic onset with a worsening of symptoms after the first week, and they were characterised by marked fluctuations in symptoms and signs. Also noted were high rates of psychological disturbances, and a close link between exacerbations of symptoms and exertion (Acheson 1959, Ramsay 1988).

However, the epidemics were not identical. For example, neurological signs such as cranial nerve lesions were much more common in the outbreaks affecting Los Angeles, Durban, the Royal Free and Middlesex hospitals while the Royal Free Hospital epidemic was unusual in terms of the high incidence of lymphadenopathy (Ramsay 1988).

1.1.2 Sporadic cases

The first person to focus on the sporadic cases was Dr. Melvin Ramsay, who had been the consultant physician at the Royal Free Hospital in London at the time of the outbreak there. In an article published in 1957, he described 34 patients aged from 9 to 45, many of whom were seen after the end of the epidemic and most of whom had had no contact with the hospital cases. However, they were clinically very similar (The Medical staff of the Royal Free Hospital 1957). Ramsay also noted in this report that all the patients experienced sequelae consisting of emotional lability and a proneness to fatigue.

Interestingly, descriptions of sporadic cases seen between 1964 and 1967 (Ramsay 1978, Scott 1970) indicated a slight change in the nature of the illness. Typical symptoms still included headache, myalgia, giddiness, emotional lability, fatiguability and visual disturbances such as diplopia, but there was far less evidence of the cranial nerve palsies which had been observed in the past. In fact, reports of

any type of paralysis became increasingly rare after 1960 (e.g. Calder and Warnock, 1984, Dillon 1970, Innes 1970). Hyde et al (1992, p. 114) has recently suggested that this change may be linked with the introduction of the polio vaccine in the late 1950s. However, this remains a matter for debate.

Ramsay himself explained the differences between the epidemic and sporadic cases, particularly those seen after 1955, in terms of the time when the patient presented to the doctor. While the former were generally seen immediately after the onset of the symptoms, sporadic cases often consulted their general practitioner "after the initial illness was long passed". At this stage, he observed, patients were more likely to present with symptoms such as muscle fatiguability, impairment of memory and an inability to concentrate (Ramsay 1978).

Aside from identifying the different characteristics of the acute and chronic phases of the illness, Ramsay was also the first specialist to emphasize that recovery was often incomplete, and that some showed no recovery at all.

1.1.3 Chronicity

The chronicity of the illness had been observed in a number of the early reports including that by Gilliam (1938). However, most observers were not aware that the condition could last for years. For instance, when Acheson (1954) described the epidemic at the Middlesex Hospital, he noted that "excessive fatigue and intermittent backache were present in several cases for 3 or 4 months after return to nursing duties. As there were no objective signs, the significance of these symptoms was difficult to assess". Meanwhile Pellew, who reviewed the epidemic in Adelaide (1951), wrote that while recurrences did occur, and psychological sequelae were prolonged in some people, the prognosis was "univer-

sally good".

However, a more systematic follow-up revealed a different picture. For instance, Sigurdsson and Gudmundsson (1956) examined 39 of those involved in the epidemic in northern Iceland in 1948 and found that six years later, nearly a half still had muscle tenderness and neurological signs. Indeed, only 13% regarded themselves as completely well.

The chronicity of ME was also observed by Marinacci (Marinacci and Von Hagan 1965). In 1937, he had worked as an intern in the Los Angeles General Hospital and had come into contact with many of the patients from the epidemic in 1934. Some were still hospitalized; others had been re-admitted because of a recurrence of symptoms. Marinacci reported that many carried the label of 'malingerer' and 'compensationitis'. "This attitude often produced a conflict between the patient and the attending staff, and the patients were transferred from clinic to clinic, and from department to department". Between 1948 and 1952, he examined 21 of the patients again. Even though it was more than 14 years after the epidemic, they were still experiencing fatigue, memory problems, muscle spasms and pain.

Given the interest in the epidemics and the limited number of follow-up studies, the extent and the severity of the chronic symptoms were not fully appreciated until Ramsay's report in 1978. This revealed that some patients remained ill for at least ten years (Ramsay 1978, 1988) and began to redirect many researchers' attention towards the chronic phase of the condition.

Ramsay's observations were supported by those of Hyde and Bergmann (1988). They interviewed 10 patients from the Iceland epidemics of 1948 and 1955, and reported that 40 years on, only two (20%) were in good health. The others still experienced fatigue, muscle pain, tenderness and

weakness, all of which had also been documented in the previous report (Sigurdsson and Gudmundsson 1956).

1.1.4 The Mass Hysteria debate

The failure to isolate an infectious agent in most of the epidemics, the low mortality associated with the illness, the high attack rate among women and the presence of symptoms such as emotional lability, led some to suggest that the disorder has an hysterical origin (for a review see Acheson 1959, Jenkins 1991a).

For example, two psychiatrists who reassessed the literature in 1970 argued that, although some of the epidemics could be attributed to "altered medical perception" among medical staff, others were almost certainly the result of anxiety and mass hysteria (McEvedy and Beard 1970a and b).

Taking the Royal Free outbreak as an example of the latter, they noted that most of those affected had been young and socially segregated young females, that the illness had not spread beyond the institutional population and that it had not affected many male members of staff. This, they felt, supported the view that the illness was a result of hysteria. However, they did not speculate as to what may have triggered the anxiety, or what may have led to the lymphadenopathy or the morphological abnormalities seen in circulating lymphocytes (Crowley et al 1957, Medical Staff of the Royal Free Hospital 1957).

The majority of ME specialists rejected the mass hysteria theory, pointing out, for example, the objective evidence of central nervous system involvement, the difference between the emotional disturbances in ME and those associated with hysteria, the chronicity of the former and the fact that in one study, the putative agent had been transferred to monkeys (Editorial 1978, Jenkins 1991a, Leitch 1994, Ramsay

1988). It has also been noted that the nurses were not socially segregated and that a very high proportion of the staff and students at the hospital were female, so that there were relatively few men at risk (Gosling 1970). This analysis was supported by Crowley et al (1957) who examined the attack rates among the staff in residence. They found that the proportion of cases among the men and women was almost the same: 20 versus 19 per 100 respectively. Thus the preponderance of female patients may simply have reflected the large number of women in the population at large. More recently, Goudsmit (1993) argued that if the epidemic had indeed resulted from anxiety in a segregated group of women, then one would have expected a high proportion of cases in the Elizabeth Garrett Anderson Hospital and Maternity Home, both of which were part of the Royal Free Hospital and both of which were run for and by women. However, these two hospitals remained relatively unaffected (Crowley et al 1957).

A further argument against McEvedy and Beard's explanation is that the epidemic showed certain features which are far from typical in mass hysteria. According to Sirois (1983) who reviewed 70 outbreaks of the latter, most last between 10 and 20 days and affect primarily women in the first years of adolescence. Yet, the epidemic at the Royal Free Hospital lasted for four months (from July 13th 1955 until November 24th) and only a minority of those affected were under 20 years old (Crowley et al 1957).

Finally, the existence of sporadic cases which clearly resembled those seen in the epidemics (e.g. Ramsay 1957, Scott 1970), plus the evidence of viral involvement both in the Royal Free patients (Parish 1994, personal communication) and in others (Innes 1970), does not support the view that all cases can be explained in terms of mass hysteria or anxiety.

Three years after writing the review of the Royal Free cases, McEvedy and Beard published a follow-up study of 71 affected patients (1973). This indicated that the latter had higher Neuroticism scores than a comparison group of unaffected nurses; that they had more admissions to hospital and that they had borne fewer children. On the basis of these findings, McEvedy and Beard suggested that the majority of the affected nurses were probably normal women who had behaved hysterically under stress, but they maintained that some must have been "pathological hysterics". An alternative explanation, that their results reflected the sequelae of the illness, was not considered. Likewise, while the higher rate of sick-leave prior to the epidemic may have been an indication of neurosis, it could equally have been a sign of poorer general health and therefore of greater susceptibility to infection.

Although the doctors who attended the patients totally rejected the hysteria explanation, they admitted that a number of the cases at the Hospital were unusual and that some of these might have suffered from anxiety. In fact, this is the reason why these patients were omitted from the main report on the epidemic published in 1957 (Ramsay personal communication).

The articles on the mass hysteria theory proved highly influential. Indeed, it may be argued that McEvedy and Beard's conclusions did much to direct the scientific community's attention away from virological aspects of the illness for well over ten years. More importantly, their 'either mind or body' approach provided the basis for the organic versus psychological debate which continues to this day (Leitch 1994).

1.2 Current terminology and definitions

The condition that became known as ME in 1956 has had a

variety of other names (Acheson 1959, Hyde et al 1992). Some of the older terms reflected the assumed links between the acute illness and poliomyelitis, e.g. abortive polio (Gsell 1949). A few referred to the places where epidemics had taken place, for instance 'Akureyri disease' (Sigurdsson and Gudmundsson 1956) and 'Tapanui Flu' (Snow 1992). American researchers proposed the name 'epidemic neuromyasthenia' (e.g. Henderson and Shekolov 1959) but this implies that the illness is confined to outbreaks and suggests a connection with myasthenia gravis. Since this could be misleading, the name was dropped (Editorial, BMJ 1978).

Other terms, like ME and PVFS, have linked the illness to disease processes and specific aetiological factors. However, since many patients seen today do not show evidence of inflammation of the spinal cord and peripheral nerves (i.e. encephalomyelitis), and given that virtually identical conditions can be triggered by infections other than viral ones, these terms are no longer regarded as accurate. Consequently, many researchers prefer the name chronic fatigue syndrome; a neutral term which focuses on the most common symptom without reference to presumed causes (Holmes et al 1988).

The original working case definition for CFS, proposed in 1988, required the presence of persistent or relapsing, debilitating fatigue or easy fatiguability in someone who had no previous history of similar symptoms. It was further specified that the fatigue should be severe enough to reduce or impair daily activity to less than 50% of the patient's premorbid level; that it should have been present for a period of at least 6 months, and that it should not resolve with bedrest.

An additional requirement was the presence of at least six of the eleven listed symptoms and two or more physical findings. If the latter were not present, patients were

required to have eight of the symptoms listed. Clinicians were also asked to exclude a number of other disorders, including chronic psychiatric conditions such as endogenous depression and anxiety neurosis, and medical disorders such as malignancies.

Four years later, the Centers for Disease Control (CDC) recommended a revision of the criteria for CFS in order to include people with fibromyalgia (if they also fulfilled the original criteria), and people with recurrences of adequately treated Lyme borreliosis, toxoplasmosis and brucellosis (Schluederberg et al 1992). It was also decided that patients suffering from non-psychotic depression (concurrent, one month post onset or six months or more before onset) should be included, as should anyone with somatoform disorders and anxiety disorders. However, it was agreed that these cases should be identified clearly to allow separate analysis of the data.

During the evaluation of the criteria in 1994, it was felt that the inclusion of six or more symptoms was too restrictive. The case definition was amended accordingly (Fukuda et al 1994) and now requires the presence of just four symptoms from a list of eight. For further details, see Appendix I.

In the United Kingdom, criticisms of the original CDC criteria led a British clinicians to formulate their own guidelines for research (Sharpe et al 1991). These 'Oxford criteria', which are still in use today, differentiate CFS from a subtype named post-infectious fatigue syndrome (PIFS).

According to the guidelines, CFS can be diagnosed where the fatigue has a definite onset and is not 'life-long'. Furthermore, the principal symptom, chronic fatigue, has to be severe, disabling and affect physical and mental functioning. A separate category of post-infectious fatigue

syndrome (PIFS) allows researchers to identify those patients whose illness was triggered by a specific infectious agent. To qualify, there has to be definite, laboratory evidence of infection either at onset or presentation. As in the case of the CDC criteria, established medical diseases and psychiatric conditions such as schizophrenia and eating disorders are excluded. However, the number of exclusions are limited, and the 'Oxford criteria' for CFS are probably the broadest of all those currently in use.

Australian researchers have also devised their own research criteria. For instance, those formulated by Lloyd et al (1990a) require the presence of chronic and disabling fatigue which is exacerbated by minor exercise and the presence of neuropsychiatric dysfunction such as new onset short-term memory dysfunction. If alternative diagnoses can explain the symptoms, the case must be excluded.

1.2.1 Disadvantages of the criteria for CFS

Both the CDC and Oxford criteria are widely used but it has recently become clear that they are identifying a number of disorders, not just one (Bock and Whelan 1993, Hickie et al 1992, Hickie et al 1995a, Hyde et al 1992, Klimas and Fletcher 1995, Straus et al 1994).

For example, Hickie (1993a) studied 565 patients with CFS and found that 30% suffered from somatisation disorder, based on the presence of a diverse range of somatic and psychological symptoms, abnormal illness behaviour and concurrent psychological morbidity. This is not surprising, since as Buchwald (1994) has pointed out, eleven of the minor symptoms listed in the original CDC case definitions also contribute to four DSM-III-R diagnoses. Thus patients endorsing symptoms characteristic of CFS are often simultaneously endorsing symptoms of psychiatric illnesses like

major depression and somatisation disorder (see also Hickie 1993b, Katon and Russo 1992, Katon et al 1991).

As far as the Oxford criteria are concerned, these automatically exclude some patients simply because their illness did not have a definite onset (e.g. Shepherd 1992). Moreover, research has indicated that they too may cover a number of disorders. For instance, Wassif et al (1994) studied 10 patients who fulfilled the Oxford criteria for CFS. However, dynamic tests of muscle function and muscle histology revealed that one person suffered from myopathy while a second had polymyositis. Similarly, Lynch et al (1991) completed a follow-up of 42 patients who met the Oxford guidelines and found that after 18 months, 29 (69%) still fulfilled those criteria. In the intervening period, 9 had received a medical diagnosis and 3 had developed a psychiatric illness. Although these disorders could have represented co-morbidity, Lynch and his colleagues decided that the initial complaint of fatigue was probably part of the prodromal phase of the other disorders and that the predictive validity of the criteria was thus quite poor.

Aside from the lack of specificity, there are a number of other problems which limit the value of both the CDC and Oxford criteria. For example, as Straus et al (1994) noted, the earlier American case definitions (Holmes et al 1988, Schluederberg et al 1992) listed the presence of fever twice, once as a minor criterium and once as a physical sign. The presence of sore throats was also listed twice. Secondly, the requirement for a greater than 50% reduction in the level of activity is not only difficult to measure, but the actual amount of activity could be related to socio-economic considerations, not just to the severity of fatigue.

There has also been criticism in relation to the Australian criteria (e.g. Wilson et al 1994a). Indeed, neither they,

nor the CDC and Oxford guidelines are currently recommended for clinical use and none are considered as particularly accurate or valid (Fukuda et al 1994, Wessely 1995).

1.2.2 The diagnosis of ME

There are a number of definitions and guidelines which have been formulated for the diagnosis of ME. For clinical purposes, some specialists use the definition suggested by Ramsay (1988) and Dowsett (Dowsett et al 1990 and Dowsett and Welsby 1992). This recognises both acute onset cases which follow an infection and the cases which develop more gradually.

The cardinal features of ME as described in Dowsett and Welsby (1992) and Macintyre (1992) are considered to be:

1. Generalised or localised muscle fatigue following minimal exertion with prolonged recovery time.

2. Neurological disturbances.

3. Variable involvement of cardiac and other bodily systems.

4. An extended relapsing course with a tendency to chronicity.

5. Marked variability of symptoms in the course of a day.

For research purposes, ME specialists have devised what have become known as the 'London criteria' (National Task Force Report 1994). These require the presence of fatiguability following minor exertion, evidence of central nervous involvement and the marked fluctuation of symptoms. Furthermore, the symptoms should have lasted at least six months and must be ongoing.

The emphasis on both fatiguability and central nervous system involvement means that the criteria for ME are consistent with the guidelines for PVFS formulated by Ho-Yen (1993) and the case definitions of CFS developed by Australian researchers (Lloyd et al 1990a) and Walsh and Cunha (1993). They are also similar to the definitions of PVFS adopted by Behan and his colleagues (Behan and Bakheit 1991) and Weir (1991).

The criteria for ME differ from the American and Oxford definitions for CFS in three ways. Firstly, the latter do not require evidence of central nervous system dysfunction. Secondly, they do not include any references to the fluctuation of symptoms or the close links between symptoms and exertion. Thirdly, the older CDC criteria place a much greater emphasis on infection-related symptoms such as mild fever, sore throat and tender glands compared to the definitions of ME (Hyde et al 1992).

The view that ME may not be identical to all cases covered by the term CFS led the National Task Force on CFS, PVFS and ME, an independent body of experts which was set up to advise the British Department of Health, to describe the various disorders as the "chronic fatigue syndromes". They also chose to distinguish between specific subgroups, for instance, giving the name CFS(ME) to cases of CFS who also met the criteria for ME. In line with their approach and similar suggestions by Wilson et al (1994a) and Schluederberg et al (1992), this classification will also be adopted here to denote cases diagnosed as either ME or PVFS.

1.2.3 CFS(ME) versus other fatigue states

Since the introduction of the term CFS, many researchers have expressed concern about the growing emphasis on the symptom of fatigue (e.g. Hyde et al 1992). For instance, it has been pointed out that tiredness is a common complaint

among the general population (Cathebras et al 1992, Popay 1992), and associated with a variety of disparate causes (Cope et al 1994, Pawlikowska et al 1994). Indeed, as recent studies have shown, most patients who seek help for chronic fatigue do not fulfil the criteria for CFS(ME) or strictly-defined CFS[1] (Wessely et al 1995, Wilson et al 1994a). For example, in one study of 611 people attending their general practitioners, 70 (11.5%) reported experiencing fatigue for three months or more (David et al 1990). Of these, only one person (1.4%) was thought to have CFS(ME).

These results are consistent with those of Elnicki et al (1992) who identified only one case (2%) of CFS among 52 patients with chronic fatigue. Similarly, a study of 135 patients complaining of fatigue for one month revealed that only six (4.4%) met the CDC criteria for CFS (Manu et al 1988a). It is possible therefore, that factors which are of aetiological and therapeutic significance for most patients presenting with unexplained fatigue may not be relevant to people with either CFS or subgroups such as CFS(ME). Until more is known about the differences between chronic fatigue and CFS, generalising findings from one sample to another may lead to an inaccurate interpretation of the data and possibly to inappropriate advice and an exacerbation of symptoms (cf. Wessely et al 1995).

Unfortunately, researchers do not always distinguish between subgroups of patients with chronic fatigue (e.g. Pawlikowska et al 1994). Moreover, where subgroups are identified, it is not always clear whether the diagnosis was made by clinicians using accepted definitions. For instance, MacDonald et al (1993a) noted that 4 (23.5%) of their CFS patients thought that they had CFS(ME). However, these researchers did not state how this diagnosis had been made.

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[1] Strictly-defined CFS refers to cases which fulfil the Australian criteria, or early versions of the CDC criteria (1988, 1992).

A number of features can be used to distinguish CFS(ME) from other fatigue-related disorders. One is the nature of the fatigue. For example, Durndell (1989) reported that a group of students with CFS(ME) were able to differentiate between their fatigue and the normal tiredness which might follow an activity such as a sporting event. According to Durndell: "the latter was described as pulsating, exhilarating and pleasant, whilst the former was described as overwhelmingly negative, draining, like flu and being ill".

A second difference between CFS(ME) and other disorders relates to the marked fluctuations in symptoms and signs (e.g. Durndell 1989, Patarca et al 1993, Ramsay 1988). The presence of the latter can be used to differentiate CFS(ME) from the condition colloquially referred to as 'tired-all-the-time' or TATT (Dowsett and Welsby 1991). A third feature which may distinguish CFS(ME) from other conditions is the characteristic link between exertional and fatigue. Research has shown that this is far less pronounced in psychiatric disorders such as depression (White et al 1995). A diagnosis of depression is further supported by the presence of anhedonia, apathy, reduced feelings of self-worth, suicidal ideation, delusions and psychomotor retardation, all of which are less common in CFS(ME) (Calabrese et al 1992).

Another disorder which may be confused with CFS is hyperventilation or effort syndrome (Nixon 1993). However, while overbreathing has been documented in some patients with CFS, research to date has not found this to be a common problem in the patient group as a whole (Riley et al 1990, Saisch et al 1994).

Since a number of conditions now referred to as CFS are clearly different from the disorder described in 1988 (cf Price et al 1992), some American specialists have referred to the more severe condition as CFS with encephalopathy or chronic fatigue immune dysfunction syndrome (Bell 1991,

Peterson et al 1992, Pross 1992, Suhadolnik et al 1992).

1.2.4 CFS(ME) and fibromyalgia

CFS(ME) patients may report areas of muscle tenderness similar to those documented in fibromyalgia. However, the latter more often has a gradual onset, morning stiffness is a more prominent symptom, fatigue tends to be worse early in the day, and there are generally fewer signs of ongoing infection (Calabrese et al 1992, Yunus 1994). Fibromyalgia is also more common than CFS, affecting an estimated 2-4% of the population at large (Wolfe 1993). While further clarification is clearly required, the consensus of opinion seems to be that the two conditions share certain similarities, but that they are not one and the same (Ho-Yen 1994, Norregaard et al 1993, Wysenbeek et al 1991).

1.3 The clinical picture of CFS(ME)

The illness seen nowadays tends to start as an unremarkable viral infection, with myalgia, lymphadenopathy and in some cases, a gastro-intestinal or respiratory upset (Shepherd 1992). However, instead of recovering, patients begin to experience profound fatigue following activities which were previously completed without difficulty. Also typical is a prolonged delay in the restoration of muscle power (Ramsay 1988).

The fatigue, which some have likened to that reported by people with multiple sclerosis (Behan and Bakheit 1991), is invariably accompanied by other complaints. For instance, many patient report a flu-like malaise, general weakness and neurological symptoms such as disequilibrium and vertigo (Dowsett et al 1990, Murdoch 1987, Shepherd 1992).

The involvement of the autonomic nervous system may lead to frequency of micturition, night sweats, palpitations and

disturbances in thermoregulatory control e.g. feeling weak after a hot bath (Macintyre 1992, Ramsay 1988, Shepherd 1992). Patients may also experience sensory disturbances such as paraesthesia, tinnitus and hyperacusis as well as visual abnormalities such as photophobia (Potaznick and Kozol 1992), sluggish accommodation (Hyde and Jain 1992) and/or increased sensitivity to certain patterns (Smith 1991). At the same time, problems with co-ordination may lead to falls, while clumsiness can make it harder to complete fine motor tasks.

Neuropsychological symptoms associated with CFS(ME) include headaches and cognitive problems such as loss of short-term memory, an inability to concentrate and difficulty in finding the right word (Fleming 1994). In addition, many patients become emotionally labile, and some also begin to experience panic attacks, depression (Macintyre 1992, Shepherd 1992) and sleep disorders (Krupp et al 1993, Whelton et al 1992).

Aside from the fatiguability, the muscle weakness and apparent central nervous system dysfunction, there may also be symptoms associated with impaired circulation. This manifests itself in cold extremities, low temperatures and a sudden facial pallor (Ramsay 1983). Other symptoms commonly reported by patients with CFS(ME) include gastro-intestinal disturbances such as recurring nausea and abdominal pain, and the development of adverse reactions to alcohol, foods and chemicals (Hobbs et al 1989, Innes 1970, Smith 1989).

All these symptoms show a marked diurnal and cyclical variability in their intensity, and although it is not always possible to identify a specific cause for the exacerbations, reports suggest that the condition may worsen as a result of over-exertion, concurrent infections, changes in the weather, and in some cases, by 'stress' (Dowsett et al 1990, Komaroff 1994).

Unfortunately, since the adoption of the term CFS, less attention has been paid to some of the features and symptoms of CFS(ME), e.g. the fluctuations and the presence of neurological complaints. For instance, David and Wessely (1993) summarised the illness as "characterised by a main complaint of fatigue, both mental and physical, with other somatic symptoms and mental phenomena like worry and depression present". This is consistent with other descriptions of CFS and although it is recognised that space often prohibits a fuller discussion, the emphasis on fatigue may have limited many clinicians' understanding of CFS(ME). It may also undermine the diagnostic process, since there is still no laboratory test for CFS(ME), and physicians have little to guide them except their knowledge of symptomatology (Holmes et al 1988, Weir, 1991).

In the interest of clarity, the following sections will include CFS(ME) and CFS(PIFS) under the general heading of CFS. However, where the findings relating to a specific sample has not been documented in any study using the criteria for CFS, and generalisation to the latter might therefore not be valid, the reference to the specified subgroup will be retained.

1.4 Epidemiology

Cases of CFS have been documented around the world, from America to Japan and from Norway to South Africa (Hyde et al 1992, Kawai and Kawai, 1992). Moreover, it has been diagnosed in all age groups, from children of 5 to senior citizens of 76 (e.g. Hilgers and Frank 1992, Hinds and McCluskey 1993).

Estimates of its prevalence tend to vary depending on the definitions used and the presence of local epidemics. However, it does not appear to be a common disorder. For instance, Lloyd et al (1990a) studied a rural population in

Australia on the basis of which they suggested a prevalence of 37 cases per 100,000. In contrast, a British study involving a number of Scottish general practitioners identified an average of 1.3 cases per 1000 patients (Ho-Yen and McNamara 1991).

Both studies revealed that the illness affected all classes. In the Australian sample, only 14% were professionals while in the Scottish sample, the figure was 5%. Thus there appears to be little support for the media's portrayal of the illness as 'Yuppie flu'.

Although some studies have found that CFS affects a greater proportion of females than males (e.g. Bates et al 1994, Hyde et al 1994, Murdoch 1987), this may, in part, reflect gender-based differences in help-seeking behaviour and the patients' access to treatment (Richman et al 1994). Such a view is supported by findings from community surveys which have generally shown the ratio of women to men to be less than 2:1. For example, among the patients assessed by Ho-Yen and McNamara (1991), the ratio was 1.8:1, while Lloyd et al (1990a) reported a ratio of 1.3:1.

Another variable which could have produced an overrepresentation of females in some studies is age. For instance, a retrospective assessment of 393 CFS patients from Northern Ireland indicated that there were proportionately more women among older age-groups than among younger patients (Hinds and McCluskey, 1993). Indeed, among patients under 20 years, the ratio of females to males was 1:1, while for those between 20-39 it was 2.6:1. The ratio fell thereafter to 1.7:1.

Studies using broader definitions are more difficult to interpret. One study of a CFS-like illness lasting at least three months was estimated to affect at least 127 per 100,000 (Murdoch 1987). Another study which used compara-

tively broad criteria estimated the prevalence of CFS to be 560 per 100,000 (Lawrie and Pelosi 1995).

As for prognosis, most of the existing research supports Ramsay's observation that recovery is often very slow. For example, a retrospective survey of 1826 CFS patients who had been ill between 1935 and 1992 revealed that the condition lasted on average for 6.7 years and although 61% noted a partial recovery, only 2% had recovered completely (Hyde et al 1994).

These findings are similar to those of a smaller study by Wilson et al (1994b). This showed that 63% had improved and 6% had made a complete recovery in the three years since their participation in clinical trials. However, Hinds and McCluskey (1993) found that only 18.6% of their patients had recovered fully within 6.5 years. More recently, Clark et al (1995) reported that of the 19 patients they assessed, 37% recovered in the next 2.5 years. The rate was slightly reduced if patients suffered from a concurrent psychiatric condition. The predictors of recovery included a lifetime

Table 1. Course of the illness: the results from two surveys.

	Dowsett et al 1990 (N=420)	Hinds/McCluskey 1993 (N=234)
Fluctuating course (%)	20	41.7
Steady (%)	25	14.4
Improving (%)	31	34.7
Worse (%)	24	5.1
Duration range	<12 mths-60 yrs	6 mths-26 yrs

history of dysthymia, the presence of more than eight unexplained symptoms other than those listed as criteria for CFS, increased length of fatigue, less education and being older than 38.

Table 1 summarises data relating to the progress of the illness. The finding that relatively few patients follow a deteriorating course has also been documented by others (Hyde et al 1994).

1.5 Research into CFS

1.5.1 Research focusing on the role of viral infections

The occurrence of epidemics and the frequent reports that the illness began after an infection have led a number of researchers to suggest that CFS may be a post-viral syndrome. Among the micro-organisms which have been implicated in the pathogenesis of CFS are enteroviruses (Behan and Behan 1993, Dowsett 1988) and the Epstein-Barr virus (Jones 1993).

In Britain, interest has focused primarily on enteroviruses such as Coxsackie B (CBV). Studies in Scotland using the ELISA CBV IgM test found that about 40 per cent of people with CFS had abnormally high levels of IgM antibodies in their blood. In comparison, only 9% of controls were CBV IgM positive (Bell et al 1988). Meanwhile an American study recently identified elevated antibody titres to Coxsackie B1 in 75% of the patients and Coxsackie B4 in 45% of the patients with CFS (Manian 1994). These rates were significantly higher than those found in age and gender matched controls.

Other researchers have identified the viral protein VP1, which is common to all enteroviruses, in both the blood and stools of about half of their patients with CFS (Yousef et

al 1988). However, the presence of VP1 and the elevated levels of antibodies against CBV are also found in other patient groups (Halpin and Wessely 1989) and given this lack of specificity (Miller et al 1991), their value for diagnostic purposes is extremely limited.

Another way of identifying the presence of enteroviruses is by using the polymerase chain reaction (PCR) assay. In a recent study, this test detected enteroviral sequences in 41% of the patients with CFS, compared with 27% of acutely ill people with suspected enteroviral disease, and 2% of healthy controls (Clements et al 1995).

Perhaps more significant is the evidence of enteroviral infection in the central nervous system and muscles (Archard 1988, Gow et al 1991, Innes 1970). For example, enteroviral sequences were detected post mortem in the hypothalamus, brainstem, heart and skeletal muscle of a patient with CFS(ME) (McGarry et al 1994). The virus, which had a 83% similarity to Coxsackie virus B3, was not found in four age and sex-matched patients who died from cardiovascular diseases or in people who committed suicide during severe depression.

Others studies have detected enteroviral sequences in muscle tissue (e.g. Bowles et al 1993). However, while this is considered to be highly pathological, it is not specific for CFS. For instance, Gow et al also detected enteroviral RNA in patients with malignancies of the colon and breast.

There is also other evidence implicating enteroviruses in CFS. For instance, some have noted the clinical similarity between the CFS(ME) and polio (e.g. Gilliam 1938), while others have pointed out that none of the patients from the 1948 outbreak of CFS(ME) in Iceland succumbed to the polio epidemic there in 1955 (e.g. Ramsay 1988). Since this took place before mass vaccination, their apparent immunity to

polio might have resulted from previous exposure to another enterovirus (see also Hyde et al 1992, p. 122). This theory is also consistent with the discovery of a mutant enterovirus by Cunningham et al (1990) and the identification of an apparently novel enterovirus by Galbraith et al (1995).

Critics of the viral persistence theory have noted that enteroviral RNA has been detected in only a proportion of the patients with CFS(ME). While this is true, the findings reported to date can not be taken as proof that the virus was not present in others. For instance, it has been suggested that the infection may be focal and that the lesion site could have been missed on biopsy (Gow and Behan 1991). Moreover, the PCR assays may not have been sensitive enough (Fekety 1994).

A second virus which has been linked with CFS is the Epstein-Barr virus (EBV), (Jones 1993, Straus et al 1985). EBV is a DNA virus which can lie dormant in the cells for years. However, during periods of physical or mental stress, the virus may be reactivated producing symptoms which are virtually identical to those of CFS. About 20% of CFS patients show evidence of reactivated EBV in their blood (Smith 1989) and the virus has also been identified in muscle (Archard et al 1988). However, both Natelson et al (1994) and Woodward and Cox (1992) found antibody levels to EBV unhelpful when trying to distinguish between different patient groups. As a result, they proposed that the presence of antibodies were a general marker of illness rather than an indicator of a specific disease.

This may also be true for enteroviruses. There are studies where several members of a family became ill at the same time but where one had a positive VP1 test and the others had high titres to reactivated EBV. Such findings support the hypothesis that increased levels of antibodies against both viruses may just be a manifestation of a general immune

disturbance, caused by another agent yet to be identified (Smith 1989 and see section 1.5.3 below).

Other viruses which have been implicated in CFS include human herpes virus-6 (Buchwald et al 1992, Levine and Komaroff 1993) and the human T-lymphotropic virus type I and II (DeFreitas et al 1991). The presence of the former is generally regarded as a marker of illness rather than primary infection, while attempts to confirm an aetiological role for the retrovirus have so far been unrewarding (Flugel et al 1992, Gow et al 1992, Khan et al 1993). It is currently considered to be a passenger virus (Prof. R.A. Weiss, personal communication).

More recently, a new infectious agent resembling cytomegalovirus (CMV) was isolated and cultured from the cerebrospinal fluid and blood of a patient with CFS (Martin et al 1994). Repeated culturing of the virus over a three-year period indicated that it had established a clinically persistent infection. Since there were no overt clinical signs of an inflammatory reaction, the researchers have named it a 'stealth' virus.

While it is now generally accepted that CFS is not caused by one particular micro-organism, the presence of viral protein in muscle and brain tissue several years after the acute phase supports the view that many cases represent some kind of chronic infection (Behan et al 1993, Dowsett 1988, Fekety 1994). In these patients, the virus is believed to interfere with the normal specialized functions of the host cell without causing tissue damage (Oldstone 1989).

1.5.2 Evidence of disease in brain and muscle

Aside from the isolation of viral RNA from brain and muscle tissue (McGarry et al 1994), the persistent viral theory is also supported by evidence of pathology and functional

disturbances in the CNS. For instance, MRI scans from patients with CFS have revealed a number of abnormalities including lesions of the frontal lobe and cerebellum (Buchwald et al 1992, Daugherty et al 1991, Fisher Portwood 1988, Hyde et al 1992 p.425, Schwartz et al 1994a).

Pathological changes have also been observed in muscle tissue. For example, an analysis of muscle biopsies identified a number of abnormalities including atrophy of type II fibres and structural changes in the mitochondria (Behan et al 1991). However, the clinical significance of these findings remains unclear (Behan and Behan 1993).

Aside from the reports of ongoing pathology, there is also growing evidence of functional abnormalities in the central nervous system and muscles. For instance, SPET scans have identified marked reductions in the blood flow (hypoperfusion) in a number of areas in the brain (Costa et al 1994, Douli et al 1992, Ichise et al 1992, Simon et al 1991, Troughton et al 1992). One study compared CFS patients with people suffering from major depression and AIDS (Schwartz et al 1994b). Significant hypoperfusion was found in 80% of the patients with CFS, and the pattern of the defects and similarities with the AIDS patients were "consistent with the hypothesis that the chronic fatigue syndrome may be due to a chronic viral encephalitis".

This was echoed by Costa et al (1995) who conducted two studies comparing people with CFS(ME), healthy volunteers and patients with major depression. All the scans of the CFS(ME) patients revealed hypoperfusion in the brainstem. Moreover, the perfusion ratios in this area were significantly lower than those of the depressed patients and the healthy volunteers. Discussing these findings, Costa and colleagues suggested that there could be a relationship between their findings and "direct neuronal damage by a viral agent."

There is also evidence supporting early suggestions of hypothalamic dysfunction (Hill et al 1959, Ramsay 1978). For instance, patients with CFS were found to have significantly higher levels of prolactin when challenged with buspirone than either healthy controls or people suffering from clinical depression (Bakheit et al 1992). This particular finding led them to suggest that some of the symptoms of CFS may be linked to an increased sensitivity of the hypothalamic 5-hydroxytryptamine receptors.

There have also been other signs of central nervous system disturbance in CFS. For instance, researchers have reported a reduction in the levels of growth hormone and abnormalities in the production of the latter when challenged with steroids like dexamethasone (Majeed et al 1995a). Similarly, Bakheit et al (1993) found abnormalities in the secretion of arginine and vasopressin.

Another hormone which has been implicated in CFS is cortisol. For instance, Demitrack et al (1991) found a number of abnormalities relating to cortisol secretion, including a reduction in total plasma cortisol, an elevated basal evening ACTH concentration and an attenuated ACTH response to ovine corticotropin hormone. It was suggested that these results reflected dysfunction of the hypothalamic-pituitary-adrenal axis, but later studies using different samples found no abnormality in cortisol levels (Hilgers and Frank 1992, Richardson 1995). Consequently, the role of cortisol and its relationship with the symptoms of CFS remains unclear.

Other studies assessing CNS function have also produced inconsistent results. For example, Prasher et al (1990) studied patients with CFS(ME) and found abnormalities in the cognitive evoked potentials, particularly the N2 and P3 components, but this was not replicated in patients with CFS (Scheffers et al 1992).

There has been also been research evaluating muscle function in patients with CFS but again, the findings have not been consistent. For instance, electrical recordings made by Jamal and Hansen (1985) using single fiber electromyelography revealed significantly increased jitter in 75% of their CFS patients. However, a more recent study on people with CFS found abnormalities in only 17% of those tested (Roberts and Byrne 1994). Similarly, Wong et al (1992) reported changes in skeletal muscle metabolism and reduced intracellular concentrations of ATP after exercise, but this was subsequently challenged by Kent-Braun et al (1993) and Barnes et al (1993).

Some findings appear to be more robust. For instance, Lane et al (1994) found raised levels of lactate levels at comparatively low work rates while Teahon et al (1988) and Preedy et al (1993) reported a reduction in the muscle RNA content. The latter indicates a fault in the ability to synthesise muscle protein. Furthermore, Japanese researchers have reported finding a deficiency in serum acylcarnitine in 38 patients with CFS (Kuratsune et al 1994). They believe that this may be associated with mitochondrial abnormalities and accordingly, with muscle weakness and fatigue (see also Majeed et al 1995b). Meanwhile, abnormalities in cardiac function have been noted by a number of researchers including Lerner et al (1993) and Montague et al (1989).

In contrast, general measures of muscle strength have been normal (Lloyd et al 1988, Lloyd et al 1991a, Gibson et al 1993, Rutherford and White 1991). Indeed, where abnormalities have been found, they have been restricted to specific muscle groups (Maffulli et al 1993).

Unfortunately, methodological flaws mean that the research on muscle function must be interpreted with care. For instance, some of the tests may have been too demanding for

the more severely affected sufferers and the samples used may therefore not be representative. Similarly, where studies did not include sedentary controls, it is possible that the documented abnormalities reflect a lack of physical fitness, rather than the presence of disease.

1.5.3 Research assessing the immune system.

The fact that a number of different microbial pathogens can produce very similar symptoms has led to the suggestion that the primary cause of these illnesses may lie in the body's response to infection, not the nature of the infectious trigger. This is supported by several studies which have found changes in the immune system consistent with ongoing antigenic stimulation.

Evidence of immune activation include:
1. the increased expression of surface antigens and adhesion molecules, both of which are usually associated with the presence of disease (e.g. Gupta and Vayuvegula 1989, Landay et al 1991),
2. the presence of circulating immune complexes (Bates et al 1995, Behan et al 1985, Buchwald and Komaroff 1991, Komaroff 1994),
3. the shift in the ratio of the CD45RA/CD45RO T cells and increases in the number of 'mature' CD45RO T cells which express surface adhesion markers (Straus et al 1993),
4. raised levels of cytokines (e.g. Chao et al 1990, Cheney et al 1989, Cheney 1992, Landay et al 1991, Lever et al 1988, Linde et al 1992, Lloyd et al 1991b, Patarca et al 1995).

Studies have also found an increased number of CD56 cells, which may have natural killer cell-like functions (Klimas et al 1990, Morrison et al 1991, Tirelli et al 1993), and raised levels of immunoglobulins, including IgE and IgG (Bates et al 1995, Hobbs et al 1989). The increase in IgE

is associated with allergic reactions which are also manifestations of immune hyperactivity (Hobbs et al 1989).

These findings have led some researchers to propose a theory which posits that CFS is the result of a disordered immune response to viral antigens and other factors such as stress (see Figure 1).

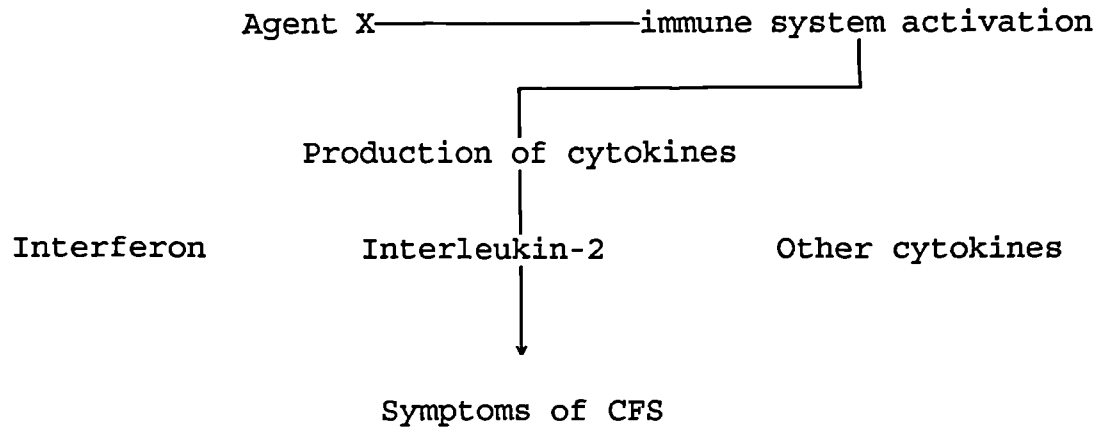

Figure 1. The immune dysfunction theory of CFS (Adapted from Bell 1991).

It has been suggested that a compromised immune system allows the reactivation of latent viruses, and that these contribute to the morbidity of CFS both directly, by damaging certain tissues (e.g. pharyngeal mucosa) and indirectly, by eliciting an ongoing immunological response. The activated immune system produces cytokines like interleukin-2 and interferon, and it is these which are believed to cause many of the symptoms of CFS (Bell 1991, Komaroff 1992, Lloyd et al 1994a). However, it is not clear whether the immune system remains activated because of the continued presence of an infectious agent or because of a failure in the process which should inhibit and suppress the response.

While some of the evidence supports the theory, there are a

substantial number of findings which do not. For instance, the general failure to find either elevated levels of CD-8 T cells or markers such as CD57 which indicate cell activation, suggests that if there is ongoing infection, it is very mild (Strober 1994). Furthermore, studies have also demonstrated *reduced* immune responses, both in vivo (e.g. Lloyd et al 1992) and in vitro (Aoki et al 1993, Caliguiri et al 1987, Gupta and Vayuvegula 1991, Klimas et al 1990). These are difficult to explain in terms of immune activation. The theory is also inconsistent with findings of normal levels of cytokines (e.g. Ho-Yen et al 1988, Straus et al 1989) and reduced levels of immunoglobulin subsets, particularly IgG3 (Lloyd et al 1989, Peterson et al 1990).

Some of the conflicting findings may be due to differences in the samples studied and the infectious triggers thought to be involved. It has also been suggested that immunological markers may change according to the severity and stage of the illness (Ho-Yen et al 1991). This idea is supported by Landay (1991), Cheney (1992) and Ojo-Amaize et al (1994), all of whom found a relationship between immunological status and severity of the disease.

Another factor that may have contributed to the confusion is the variation in techniques and tests employed. For example, a number of researchers have documented normal numbers of certain surface molecules but they did not test for specific subsets. It is therefore possible that evidence of immune activation might have been overlooked (cf. Landay et al 1991).

It has also been pointed out that the failure to detect a clear-cut and reproducible abnormality of lymphokine/cytokine secretion may reflect the insensitivity of the tests used and the rapid clearance of these substances from the circulation (Strober 1994).

Perhaps the most consistent finding to date is that of a significant impairment of natural killer (NK) function (e.g. Caliguiri et al 1987, Klimas et al 1990, Morrison et al 1991, Pross 1992). Indeed, according to Strober (1994), these findings provide the strongest evidence that immune disturbances are primary rather than secondary to chronic infection.

Immunological changes and depression

It has recently been suggested that the changes in immune function are not due to infection but a result of mood disturbance (Straus et al 1993). To support this view, it has been pointed out that immune activation has also been found in patients suffering from major depression (Maes et al 1992, Masuda et al 1994). Moreover, lymphocytes taken from CFS patients show reduced responses to mitogens, a finding which has also been noted in depressed populations (Stein et al 1991). However, findings from controlled trials have shown that most of the abnormalities identified in relation to CFS are different from those seen in depressed patients tested at the same time (Lloyd et al 1992, Landay et al 1991). This suggests that the two conditions are immunologically distinct.

Immune dysfunction versus viral persistence

At the moment, there appears to be slightly more support for the view that immune changes are secondary to, rather than the primary cause of CFS. For instance, two recent studies have investigated the theory of general immune disturbance by measuring the levels of antibody titres to a number of different viruses. In the first, testing was restricted to two herpes viruses, Epstein-Barr virus and human herpes virus-6, both of which have been linked with CFS (Natelson et al 1994). The researchers argued that if the illness was the result of a primary, nonspecific immunological problem

allowing viral reactivation, then the levels of antibodies to both viruses should be elevated. On the other hand, if the illness was due to a specific infection and the immune changes a result of this, then rises in the levels of antibodies to one virus would not necessarily be matched by rises in the antibody titres to the other. The results supported the latter view.

The second study measured antibody titres to 18 common viruses including EBV and CBV (Manian 1994). It was found that 95% of patients with CFS had high titres to either EBV VCA IgG, Coxsackie B1 or Coxsackie B4. However, simultaneously elevated levels of antibodies to the EBV and Coxsackie antibodies were detected in only 20% of cases. Thus in 80% of the patients, the elevation of viral antibody titres was not due to a nonspecific immune response.

Further support for the view that the immune abnormalities are not the primary cause of CFS comes from Griffin (1991). In her review, she mentions that the combined presence of immune activation and immunosuppression has also been documented during a number of acute and chronic viral infections, including those caused by the measles virus and HIV. Thus the inconsistencies noted above are consistent with a viral cause.

On the other hand, it is also possible that viral infection is only one of the factors underlying this disorder. There may be co-factors which, when present, allow the virus to evade destruction by the immune system. In addition, genetic influences could explain why some people recover faster from the same infection than others. In this respect, it is interesting to note that the HLA-DR/DQ haplotype, plus HLA-DQ3 and HLA-DR4 antigens were found to be more prevalent in patients with CFS than in a group of matched, healthy controls (Keller et al 1994).

Some commentators, however, believe that the evidence for a persistent viral infection is very limited, pointing out for instance, the generally subtle abnormalities in the immune system, the variable findings with regard to pathology, the failure to find notable reductions in muscle strength, the comparatively high prevalence among white, middle-aged women, and the similarities between the symptoms of CFS and depression (e.g. Bearn and Wessely 1994, Deale and David 1994, Hotopf and Wessely 1994, Manu et al 1992). This has led a number of researchers to suggest that while infection may trigger many cases of CFS, other factors may play an important role in perpetuating it (see also section 1.5.5).

1.5.4 Miscellaneous theories.

In the past few years, CFS has been attributed to a number of localised lesions. For instance, Chester (1993) hypothesized that some cases of CFS might be related to disorders of the nose. In his view, this explains the high incidence of allergies, the links with weather changes and the symptoms relating to the upper respiratory tract. CFS has also been associated with damage to the spine. For example, Perrin (1993) suggested that abnormalities in the thoracic and upper lumbar region could undermine the functioning of the sympathetic nervous system, resulting in symptoms such as headaches, dizziness, palpitations, and disturbed sleep.

So far, these explanations have received little attention. This may be partly because CFS is regarded as a complex, multi-system disease (e.g. Dowsett et al 1990), and partly because localised lesions can not explain the epidemics, nor abnormalities such as the presence of viral genetic material in the muscle and brain (e.g. Behan and Behan 1993). Nevertheless, disorders in specific parts of the body could be responsible for some cases of unexplained chronic fatigue.

CFS has also been linked to nutritional deficiencies (e.g. Cox et al 1991, Jacobsen et al 1993). For instance, Van Riel et al (1988) found that despite an adequate intake, many CFS patients had less vitamin B_2, B_6 and C than a control group. More recently, McLaren Howard et al (submitted for publication) compared 30 CFS(ME) patients with 11 healthy controls and reported that the former had significantly lower levels of magnesium, potassium, zinc, chromium, selenium, glutathione peroxidase, vitamins B_1, B_2, B_3, and B_6 and ten out of 39 amino acids in plasma. The CFS(ME) group also had higher levels of serum glutathione-S-transferase indicating increased 'toxic stress'. The reductions in both omega-6 and omega-3 series of essential fatty acids were suggestive of a block at the delta-6-desaturase level, a finding consistent with infection (Horrobin and Manku 1990).

Reduced levels of essential fatty acids have also been implicated as a cause of the abnormalities in red cell shape (Simpson et al 1993). Compared with healthy individuals, patients with CFS(ME) were found to have a reduction in the number of cells with cup forms but an increase in the proportion with altered margins. According to Simpson and his colleagues (1993 and personal communication), the changes in the red cell shape reduce the filterability of the blood, thus impairing blood flow and limiting the supply of oxygen and substrates to the tissues. The inadequate delivery of nutrients to the muscles could lead to weakness, fatigue and reduced aerobic work capacity (cf. Riley et al 1990). Similarly, the impaired blood flow to the hypothalamus might result in sleep disturbance, irregular temperature regulation and emotional lability (Simpson 1990). Essential fatty acids act by improving the flexibility of the cells, thereby increasing perfusion of oxygen in the microcirculation and the filterability of the cell membrane (Dutta-Roy 1990, Simpson et al 1993).

Given that similar changes in red cell shape have also been documented in other conditions characterised by fatigue, e.g. pernicious anaemia and MS, the abnormalities cannot be used to confirm the diagnosis. However, Simpson's theory is consistent with the view of CFS(ME) as a multi-system disorder and compatible with recent findings of hypoperfusion in certain areas of the brain (e.g. Costa et al 1995, Goldstein et al 1995, Ichise et al 1992, Schwartz et al 1994b, Simon et al 1993).

Finally, some researchers have implicated the overuse of antibiotics and exposure to neurotoxins as contributory factors in the aetiology of CFS. So far there is little evidence linking antibiotics with the onset of CFS (Hyde et al 1994, Smith 1989). However, a study of patients who developed a CFS-like condition following exposure to organophosphate pesticides showed that toxic chemicals can produce abnormalities of hypothalamic function resembling those seen in people with CFS (Behan and Haniffah 1994). Moreover, the similarity between CFS and Gulf War Syndrome supports the argument that neurotoxins may have effects on the brain and immune system similar to those documented in CFS (NIH Workshop Statement 1995).

Although much remains unclear, the abnormalities which have been found in the blood, muscles and brains of patients all support the view that there is ongoing disease in some cases of CFS (Behan and Behan 1988, Lieberman and Bell 1993, Preedy et al 1993, Ramsay 1988, Simpson et al 1993). Useful information relating the role of infection and immunology may be obtained from further controlled research into CFS, as well as investigations into post-polio syndrome (Bruno et al 1994ab, 1995), fatigue connected to organophosphate poisoning and the CFS-like condition in horses and mice (Behan and Haniffah 1994, Chao et al 1992, Ricketts et al 1992, Shepherd 1993).

1.5.5 Research into the psychological and psychiatric aspects of CFS.

The failure to find consistent evidence of disease in the patient group as a whole has focused attention on the role of mood disorders, maladaptive beliefs and 'stress'. For example, some have suggested that CFS represents an atypical form of depression (e.g. Higgins 1992). This view is supported by a number of studies which have found high rates of major depression in people with CFS (e.g. Lane et al 1991, Manu et al 1988, Taerk et al 1987, Wessely and Powell 1989, see also chapter 4). Moreover, it has been pointed out that depression can also cause profound fatigue, sleep disorders and cognitive disturbances (Buchwald 1994, Komaroff 1994, Pepper et al 1993, Ray 1991).

However, other findings are inconsistent with this view. For example, Hickie et al (1990) found that the pattern of symptoms in CFS patients was significantly different from that seen in patients with non-endogenous depression (cf Jenkins 1991b). Similarly, Pepper et al (1993) reported that CFS patients had a distinct psychiatric profile compared with patients with major depression.

CFS and major depression have also been found to differ in terms of the type of neuropsychological deficits (Sandman et al 1993); the diurnal variation in energy levels (Wood et al 1992); severity of overall disability (Natelson et al 1995); response to exercise (Lane et al 1995); the type of neuroendocrine responses (Bakheit et al 1992, Bearn et al 1995, Bearn and Wessely 1994, Cleare et al 1995, Demitrack et al 1991, Majeed et al 1995); and the pattern of cerebral hypoperfusion (Costa et al 1994, Goldstein et al 1995, Schwartz et al 1994b).

Finally, the concept of CFS as a form of affective disorder conflicts with the general consensus of opinion that this

illness is a heterogeneous entity with a multifactorial aetiology (Bearn and Wessely 1994, Wilson et al 1994a).

A more frequently expressed view is that the presence of mood disorders plus the patient's adherence to maladaptive beliefs and coping strategies play a major role in maintaining the symptoms of CFS (Wessely et al 1989, Sharpe 1993).

According to the cognitive-behavioural model of CFS, the patients' tendency to attribute their symptoms to a physical cause begins a vicious circle of avoidance, frustration, depression and further fatigue. Thus it has been argued that when patients become ill, normal postviral debility is interpreted as a result of continued disease which leads them to extend their period of rest and convalesce (e.g. Butler et al 1991, Wessely et al 1991). This reduces their physical fitness so that symptoms are elicited at increasingly lower levels of activity. The patients' belief that the cause of their predicament is entirely viral leads them to reject alternative explanations and coping behaviours so that their continued inactivity reduces their fitness even more. At the same time, their lack of perceived control compounds the feelings of frustration, helplessness and depression. This in turn adds to the fatigue, leading to even more inactivity, depression and so on.

Some regard the influence of unhelpful cognitions as paramount, since these can influence a number of behaviours, not just inactivity (Sharpe 1994, Surawy et al 1995). For example, Surawy et al (1995) have suggested that a desire to achieve high standards of performance and an extreme need to meet the expectations of others may lead people to 'press on' in the initial stages, when they should rest. At the same time, the patients' need to achieve combined with their perfectionism and an unwillingness to show weakness may trigger episodic attempts to perform at premorbid levels.

When these inevitably fail, patients may feel frustrated and become increasingly preoccupied with the symptoms and invalidism. This will result in increasing disability, demoralization and depression.

The model also posits that patients with CFS tend to attribute their illness to an external cause primarily to avoid a loss of self-esteem (Manu et al 1992a and b). This explanation is supported by Powell et al (1990) who found that people with CFS had higher levels of self-esteem than patients suffering from depression. It is also consistent with the research by Cope et al (1994a) which found that a tendency to somatise was the most important predictor of fatigue, six months after a viral illness. Although the raised somatisation scores could have reflected the presence of physical disease (Robbins and Kirmayer 1991), it is equally possible that the patients' somatising may have led to more important emotional problems being neglected, thus prolonging the fatigue (cf. Sharpe 1994).

Indeed, it has been difficult to evaluate the influence of specific beliefs in the aetiology of CFS. For instance, although it is well documented that most patients tend to attribute their illness primarily to a physical cause (e.g. Powell et al 1990, Ray et al 1992b, Sharpe et al 1992, Ware and Kleinman 1992, Wilson et al 1994), the limited knowledge of the aetiology of CFS makes it impossible to judge the 'correctness' of this attributional style (Powell et al 1990). Thus while a particular belief or view may be closely related to poor outcome (e.g. Wilson et al 1994b), it does not necessarily follow that those attributions are wrong and that a change in attitude would lead to a reduction in fatigue and distress. Moreover, the findings of Wilson et al were not supported by Bonner et al (1994) who found that belief in a physical cause did not predict outcome.

Other aspects of the cognitive-behavioural model of CFS have also been difficult to assess. For instance, since all the pre-1995 definitions for CFS required patients to have reduced their activity levels by at least 50%, some of the sufferers who remained active and whose symptoms can therefore not be attributed to physical deconditioning may have been excluded. This could have produced unrepresentative samples and made it difficult to determine the real influence of inactivity on the symptoms of CFS. Interestingly, where avoidance of activity was assessed, researchers found that it did not predict either fatigue or psychological well-being (Vercoulen et al 1994).

Indeed, there is still comparatively little objective evidence that fatigue is largely the result of physical deconditioning (Gibson et al 1993, Lane et al 1994, Montague et al 1989). Similarly, the findings have also failed to show that CFS is due to a lack of motivation (Lloyd et al 1991a, Rutherford and White 1991) or disuse atrophy (Connolly et al 1993).

While there is as yet little supportive evidence for the cognitive-behavioural model, particularly for the CFS population as a whole, it is possible that the combination of maladaptive beliefs and behaviours could be a major determinant of chronic fatigue in subgroup (Macdonald et al 1993a). It is also likely that concurrent depression may delay recovery from infection (cf. Cluff 1991) and there is evidence that both depression and maladaptive beliefs can increase the patients's perceived disability and actual emotional distress (see Chapter 4).

The role of stress.

A number of researchers have hypothesized that somatisation may be one way of dealing with high levels of 'stress'. For example, Ware and Kleinman (1992) interviewed a number of

patients with CFS and found evidence of distressing experiences and exhausting work schedules prior to the onset of fatigue. As a result, they proposed that some patients may have used illness as a means to escape from their busy lives.

This 'flight-into-illness' theory was subsequently challenged by Mechanic who noted that it conflicts with the presence of ongoing distress and the observation that many patients actively continue to seek treatment (Mechanic 1993, p.79). It has also been pointed out that since the findings were based on retrospective reports, they may have been influenced by memory distortion and 'effort after meaning' (Cope et al 1994a, Hotopf and Wessely 1994).

On the other hand, there is some support for the argument that stress may be implicated in CFS. For example, research has found that stress can lead to a reduction in immune competence (Adler and Matthews 1994). In one study, mobilization stress caused a transient depression of virus-induced interferon production in mice, aggravating the course of the influenza and allowing the virus to penetrate the brain (Chetverikova et al 1987). Experimental stress has also been shown to increase the susceptibility of mice to other viruses, including herpes simplex, poliomyelitis and Coxsackie B (Rasmussen 1969).

In humans, stress has been associated with the development of minor respiratory infections such as colds (Cohen et al 1991, Evans and Edgerton 1991) and with the reactivation of herpes viruses such as EBV (Kiecolt-Glaser and Glaser 1987).

In terms of CFS, the immunosuppressive effect of distress could help to explain why the immune system fails to clear the initial infection and why the illness becomes chronic (cf. Cluff 1991, Adler and Matthews 1994, Stein et al 1991). However, while there are some retrospective and anecdotal

reports linking stress and the onset of CFS, the results of more formal research have been inconsistent. For example, Stricklin et al (1990) found that patients with CFS had more severe stress in the 12 months prior to illness than a group of healthy controls. In contrast, Durndell (1989) reported no difference in the number of life stress events, or in the perception of those events between CFS patients and controls. Others have also failed to demonstrate a significant link between life events and the onset of fatigue lasting at least 6 months (Bruce-Jones et al 1994, Lewis et al 1994).

Approaching the subject from an epidemiological perspective, Hyde et al (1994) estimated that people in high stress jobs who were not in contact with infectious disease or with a recently immunized public had a relatively low risk of developing CFS. However, they added that pre-existing exhaustion due to demands at work etc could compromise the immune system, thereby increasing certain individual's vulnerability to CFS.

It is worth noting, however, that while stress may lead to immune suppression and hence to an increased susceptibility to disease (Adler and Matthews 1994, Hotopf and Wessely 1994), most of the immunological changes which have been associated with stress are different from those reported in patients with CFS. Indeed, evidence of immune activation such as that documented in the literature on CFS is comparatively rare in relation to stress (e.g. Landay et al 1991, Lloyd et al 1994a, Patarca et al 1995). Moreover, stress has been tentatively linked with reduced levels of IgA (Jemmott et al 1983), which tend to be normal in patients with CFS (Behan et al 1985, Gupta and Vayuvegula 1991, Hobbs et al 1989, Lloyd et al 1989).

Thus while it is possible that stress may make people more vulnerable to the infection which may trigger CFS, there is as yet little support for the view that the failure to re-

cover can be attributed to a stress-induced attenuation of the immune response.

A psychodynamic view

Taking a slightly different approach, Taerk et al (1994) hypothesized that CFS may be the end result of early disturbances in the mother-child relationship. They proposed that these lead individuals to turn to others on whom they then become overly dependent. The strained relationship with the mother also undermines their ability to deal with stress, which leads to physiological instability and makes these individuals susceptible to a variety of diseases later in life. When the person loses or is separated from those on whom he/she has become dependent, this triggers changes in the hypothalamic-pituitary-adrenal axis and the immune system, which then provides the basis for CFS. In their view, a psychotherapeutic relationship which allows the patient to internalize new selfregulatory tension-reducing structures can help to stabilize the illness and reduce fatigue.

Research on personality

Others researchers have chosen to focus on the personality of patients with chronic fatigue. For instance, Stricklin et al (1991) used the Minnesota Multiphasic Personality Inventory and found a profile suggesting a neurotic or psychophysiological illness rather than an hysterical one. A second study also identified the 'neurotic triad' but noted a significant amount of heterogeneity (Blakely et al 1991). Indeed, one subgroup appeared to have no psychiatric disorders at all. Meanwhile, a comparison of 40 patients with CFS(ME) and multiple sclerosis revealed almost identical scores for both neuroticism and extraversion (Goudsmit, unpublished).

As yet, studies have not identified a specific personality related to CFS. Although it has been suggested that patients may be motivated by a strong need to achieve (e.g. Sharpe 1994, Surawy et al 1995, Ware and Kleinman 1992), Stricklin et al (1990) found no difference between CFS(ME) patients and healthy controls on a measure assessing this trait.

Researchers have also assessed Type A behaviour which has been linked with an increased incidence of infectious mononucleosis, a condition which can also lead to chronic fatigue (Barton and Hicks 1985). Although Durndell (1989) and Lewis et al (1994) found that CFS patients had similar global Type A personality scores to comparison groups, both reported that some individuals may have pushed themselves prior to the onset of illness. Thus it is possible that a tendency to ignore symptoms and remain active could have predisposed some people to develop CFS. On the other hand, it can not be ruled out that the high activity levels before the onset of fatigue may simply have reflected the patient's personal circumstances, e.g. an inability to take time off and rest (Hyde et al 1994, Shepherd 1992, see also Chapter 3).

To summarise, studies have indicated that personality and stress may increase a person's vulnerability to infection but there is as yet little evidence that these same factors play a major role in perpetuating CFS.

Neuropsychological research.

Findings from studies assessing neuropsychological functioning have been generally supported patients' reports of difficulties related to memory and concentration. For example, sufferers tested by Smith (1992) and Smith et al (1993) had deficits in selective and sustained attention, as well as impaired recognition and recall. These abnorma-

lities persisted over time and were observed in both a well-defined hospital sample and a group taken from the community.

Meanwhile, DeLuca et al (1993) showed that the impairments noted in a group of patients with CFS were similar to those of people with multiple sclerosis. Like Smith et al, they examined the possibility that the cognitive deficits might have resulted from co-existing depression but found the correlation between depressed mood and performance to be weak. Although a later study failed to reproduce some of their earlier findings, they identified abnormalities in information processing speed and once again found that neither depression nor anxiety were related to performance (DeLuca et al 1995).

The influence of mood on cognitive functioning was also assessed by Sandman et al (1993) who tested 39 patients with CFS, 23 patients suffering from major depression and 129 age-matched healthy controls. Their results indicated that the patients with CFS were vulnerable to interference and slow or uncertain in decision making. "Apparently, CFIDS patients made weak memory traces that were easily perturbed". The results also revealed that the cognitive and memory profiles of people with CFS were distinctly different from those of patients with depression. Their findings are in agreement with those of Johnson et al (1994), who suggested that the documented problems may reflect deficiencies in information processing and the encoding of the memory trace.

It should be noted here that many of the abnormalities which have been found are comparatively subtle, and that a number of studies have failed to find differences between CFS patients and controls (e.g. Altay et al 1990, Grafman et al 1993, Macdonald et al 1993b, Ray et al 1993a). According to Ray et al (1993a), the inconsistencies may be related to the

differences in the composition of the samples and the measures used. They also suggested that the performance on cognitive tasks could be influenced by differences in the severity of fatigue, physical malaise and emotional distress. However, since the impairments are often quite specific, it might be argued that these variables do not affect the patient's response on every task (e.g. Grafman et al 1993, Krupp et al 1994, Ray et al 1993a).

In general, the documented neuropsychological impairments associated with CFS appear to be consistent with multifocal cerebral dysfunction (Bastein and Thomas 1988, Smith 1991, Riccio et al 1992, Thompson 1989). Nevertheless, the inconsistencies between the studies mean that the significance of the abnormalities remains difficult to determine.

1.6 Treatment

A large number of treatments have been evaluated for CFS including interferon-α (Brook et al 1993), acyclovir (Straus et al 1988), transfer factor (Lloyd et al 1993), psychotherapy (Taerk and Gnam 1994), diet (Durndell 1989), aminoacids (Bralley and Lord 1994), vitamin B12 (Simpson 1991), kutapressin (Ablashi et al 1994), other nutritional supplements (Aoki et al 1993), osteopathy (Perrin 1993), Chinese medicine (Lee 1992, Jiang and Franks 1994), herbs and homeopathy (Leyton and Pross 1992) and combinations of various therapies (e.g. Anderson 1988, Dowson 1993, Hilgers and Frank 1993). However, relatively few of the treatments have been subjected to double-blind, placebo-controlled clinical trials. Of those that have, only four were found to be superior to a placebo. These are an antiviral and immunoregulatory agent known as Poly(I).Poly(C_{12}U) 'Ampligen', high dose intravenous immunoglobulin, evening primrose oil and IV magnesium sulphate (Behan et al 1990, Cox et al 1991, Lloyd et al 1990b, Strayer et al 1994).

There are still no published reports of a controlled trial of anti-depressants. However, the results of open trials indicate that although certain drugs appear to be helpful in improving general functioning and reducing pain (Goodnick and Sandoval 1993), the effects on the illness as a whole tend to be variable (Behan et al 1994, Hickie and Wilson 1994, Klimas et al 1993, Jenkins 1991, Weir 1991). Moreover, there is anecdotal evidence that CFS patients are sensitive to drugs and as a result may not tolerate the therapeutic dosages required (Abbey 1994, Bell and Concemi 1994, Wilson et al 1994b).

As for cognitive-behavioural therapy, controlled studies have been generally disappointing (Friedberg and Krupp 1994, Lloyd et al 1993). Although an early open trial led to improvements in about 80% of patients with CFS (Bonner et al 1994, Butler et al 1991), these findings have not been replicated by others (e.g. Cox and Findley 1994). Nevertheless, there is some evidence that a cognitive-behavioural approach is helpful for more general fatigue (Sharpe 1994 personal communication), and for individual patients with maladaptive beliefs (Faas 1992). A more detailed discussion of CBT can be found in Chapter 5.

Given the limited number of effective treatments available, some physicians have focused primarily on lifestyle management and symptomatic care (e.g. Bell and Concemi 1994, Ho-Yen 1993, Wilson et al 1994a). Views on exercise continue to vary although most specialists currently advise CFS patients to accept the limitations imposed by the illness, to balance rest and activity and to avoid overexertion and 'stress'. A few also recommend vitamin and mineral supplements, sometimes in combination with a low-sugar, low-yeast diet plus anti-fungal medication (Dawes 1991). So far, anecdotal reports suggest that this regime is beneficial in a proportion of cases, but there has been only one study to test this approach (Hilgers and Frank 1992), and the theory

on which it is based still lacks scientific support.

1.7 Discussion

As noted above, the literature on CFS reveals many inconsistencies and ambiguities. Thus in some cases, the illness follows an infection while in others, the onset is gradual. In some patients, there is evidence of ongoing infection but this has not been found in others. Even in those with post-infectious CFS, a number of micro-organisms have been implicated, suggesting that different infections can result in the same clinical syndrome.

In terms of the psychological aspects of CFS, research seems to indicate that many patients fulfil criteria for psychiatric illness. However, here too, no single disorder has been consistently linked with all patients and the differences between CFS and depression suggest that the fatigue can not be explained in terms of mood disorder alone.

While the controversies about the aetiology of CFS have been generally acknowledged, a number of issues have yet to be fully addressed. For instance, the current emphasis on fatigue may have reduced some clinicians' awareness of the severity and disability associated with other symptoms, particularly those related to cognitive function.

Another issue which must be considered is the diagnosis of CFS. As discussed above, the use of less restrictive definitions means that CFS has become an umbrella term which covers a number of disorders (e.g. Bock and Whelan 1993, Klimas and Fletcher 1995). For instance, recent studies on CFS have described patients with giardiasis, major depression, somatisation disorder, fibromyalgia as well as post-viral syndromes and Lyme disease (Coyle et al 1994, Levine et al 1992, Straus et al 1994). According to Straus et al (1994), the fact that the CDC case definition allows

a number of different conditions to be included under the rubric of CFS has broadened "the scope of the clinical entity to the point at which it is no longer definable".

The heterogeneity of CFS may be one possible explanation for the inconsistent findings which have been documented, for instance, in relation to cognitive impairments (e.g. Smith et al 1993 versus Macdonald et al 1993b), attributions (e.g. Wessely and Powell 1989 versus Ray et al 1992b), the rate of depressive disorders (e.g. Yeomans and Conway 1991 versus Wessely and Powell 1989) and the type of depressive symptoms (e.g. Powell et al 1990 versus Hickie et al 1990). On the other hand, the inconsistencies could also be due to the fluctuation of the disorder (Patarca et al 1993) or the use of different laboratories and measures.

To reduce the 'noise' introduced by the inclusion of different disorders, both the CDC and Australian researchers have recommended that specific subgroups be distinguished and that their results be analysed separately. This will enable scientists to compare different fatigue syndromes, and to identify similarities and differences.

A third issue concerns the research into the psychological and social aspects of CFS. To date, most studies have focused on the prevalence of psychiatric morbidity and the patients' adherence to a set number of maladaptive beliefs and behaviours. As a result, there is a lack of information concerning the impact of CFS and the patients' response to their illness. Indeed, the current knowledge about the nature of the attributions or the types of coping strategies used, may actually be incomplete. Moreover, in contrast to the research on other chronic disorders, little is known about some of the additional factors which may underlie emotional distress (Mayou and Hawton 1986, Rodin et al 1991, Wells et al 1989). For example, there have been few studies assessing the possible influence of social support, and how

this may affect the patient's psychological health.

This lack of knowledge about the nature of the illness, the types of coping strategies used and the possible determinants of emotional distress suggests that the full complexity of CFS may not yet have been recognised. Further research into the psychological and social effects of CFS is therefore required, not only to obtain a fuller understanding of the illness-as-lived, but also to identify more effective ways of alleviating the patients' distress.

CHAPTER 2

Reactions to chronic illness and disability

> "All chronic illnesses represent assaults on multiple areas of functioning, not just the body. Patients ... may face separation from family, friends, and other sources of gratification; loss of key roles; disruption of plans for the future; assault on self-images and self-esteem; uncertain and unpredictable futures; distressing emotions such as anxiety, depression, resentment, and helplessness; as well as such illness-related factors as permanent changes in physical appearance or in bodily functioning."
>
> (Turk 1979)

Although it is generally accepted that CFS has a profound effect on those who suffer from it, there has been comparatively little research into the psycho-social consequences of the illness. To help determine which factors may be involved, and how these might affect adjustment to CFS, it was decided to examine the research on other chronic disorders. Section 1 focuses on the process of adaptation and the assessment and prevalence of emotional distress. This is followed by a review of the various factors which have been associated with adjustment and distress in the chronically-ill and a discussion about the variables which may play a similar role in patients with CFS.

2.1 The process of adjustment

Chronic conditions are rarely stable and for most patients, changes in the illness or disability cause disruption at various times throughout their lives. Adaptation is therefore an ongoing process, not an end state (Dimond 1983, Turk

and Rudy 1986).

In most cases, the patients' reactions to the changes vary according to the demands placed on them and their resources at the time. However, there appears to be general agreement about what patients should aim for. According to Dimond (1983), successful adaptation is achieved when one's way of life "sustains hope, diminishes fear, and preserves a quality of life that takes account of, perhaps transcends, but is not controlled by, the limitations of the illness".

In contrast, Wright (1960) suggested that patients needed to change certain attitudes. For example, she believed that they should subordinate concerns about physique to factors such as personality and effort and that they had to contain the effects of their disability so that the latter would not affect how they perceived every aspect of their life. She also felt that patients should learn to value their own assets and strengths instead of basing judgements on a comparison with others.

A completely different approach to the study of adjustment has been to assess the reactions to specific events. For instance, Moos and Tsu (1977) identified 7 major 'adaptive tasks' which most patients encounter while they are ill. These include:

1. Dealing with symptoms and incapacitation, learning to control symptoms and prevent exacerbations.
2. Dealing with the hospital environment, treatments and procedures.
3. Developing adequate relationships with professional staff and dealing with problems that may occur.
4. Preserving a reasonable emotional balance, for instance, by managing upsetting feelings, by dealing with anxiety and apprehension if the outcome of the illness is uncertain, and most importantly, maintaining some hope, even when its scope is sharply limited by circumstances.

5. Preserving a satisfactory self-image, and maintaining a sense of competence and mastery (which often necessitate a change of personal values as discussed by Wright, above).
6. Preserving relationships with family and friends, dealing with feelings of isolation and alienation.
7. Preparing for an uncertain future, the loss of functions such as sight, speech etc., and in some cases, death.

According to Moos and Tsu, people's reactions to these tasks are generally aimed at re-establishing their sense of social and psychological equilibrium. Responses which achieve a "new balance" and which promote maturation and personal growth are regarded as adaptive. Conversely, those which lead to psychological deterioration and decline are maladaptive.

The adaptation to chronic disorders can also be studied using the cognitive-motivational-relational theory developed by Lazarus (1991, 1993, Cohen and Lazarus 1979). This focuses on the ways in which individuals appraise changes in their relationship with the environment and how they react to those appraisals. For instance, it posits that if a person judges new demands to be manageable, these will be interpreted in a positive way, as a challenge and an opportunity for growth. Similarly, where outcomes relating to change are judged to be either irrelevant to the person's well-being or goals and unlikely to lead to harm or loss, they may be ignored. Thus, changes in these situations are unlikely to produce significant distress. Indeed, it is only when a particular demand *threatens* the individual's well-being, values or goals, and the person does not feel able to master or control that demand, that he or she may experience psychological distress. Moreover, if the individual fails to solve the problem or to regulate their emotional distress, it may adversely effect their health, their daily functioning and their morale.

Whether a situation is judged to be threatening depends on individual characteristics, e.g. a person's knowledge, beliefs, goals, values and personality; and environmental variables, e.g. the imminence of harm and the resources which are available to deal with the harm. Similarly, how a patient *responds* to the threat will be determined by a variety of factors such as the individual's options for coping, the expected outcome, and whether the person will be blamed or not if things go wrong.

Another influence on both appraisal and response is the state of a person's health (Nerenz and Leventhal 1983). For instance, it has been pointed out that fatigue and dysphoria may both make demands seem more overwhelming and distressing than they are, and undermine people's confidence about their ability to cope (Lazarus 1991, Fontana and Palfai 1994). Likewise, cognitive deficits might interfere with the processing of information, and the performance of any coping strategies which require a high level of concentration (Earll 1989). Similarly, pain or feeling ill may disrupt focused thinking (Trieschmann 1989), leading to maladaptive responses and possibly, to emotional distress (Donoghue and Siegel 1993, Harkapaa 1991).

In summary, psychological disturbances can be perceived both as stressors which threaten well-being and undermine coping, and as the effect of the persons's inability to cope with other 'stressors' (Lazarus et al 1985).

The following section will outline the various types of psychological disturbances associated with chronic illness and disability and discuss the prevalence of the more serious psychiatric disorders such as depression.

2.1.1 The psychological response to illness and disability

Psychological disturbances associated with illness or disa-

bility may involve one emotion or a combination. They can also vary in intensity, from mild to severe. If psychological symptoms occur within three months of the onset of physical illness or trauma and they cause significant impairment, they are classed as 'adjustment disorders' (Razavi and Stiefel 1994). The latter differ both from acute stress reactions, which are transient and last for a shorter period of time, and from post-traumatic stress disorder, which is dominated by the reliving of the trauma in memories and dreams, and the avoidance of activities and situations which remind the person of the stressful event.

Another disorder related to illness is organic mood syndrome (Lloyd 1991). This is diagnosed when psychological disturbances appear to be aetiologically related to specific organic factors such as infection, a degenerative process in the brain, or the effects of certain drugs. Also recognised are generalized psychological distress and the more specific and severe psychiatric diagnoses.

According to Mayou and Hawton (1986), the most common form of affective disorder among the medically-ill is an undifferentiated neurotic syndrome and only a small, but important minority suffer from more specific psychiatric conditions[1]. The latter are best diagnosed using specific, standardized measures or structured interviews (Rodin et al 1991). However, the former may be assessed using general measures of emotional distress such as the General Health Questionnaire (GHQ).

Information about the patient's psychological state may also be obtained indirectly, through data about self-care and the

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[1] Although the presence of psychiatric disorder is a clear indication of psychological suffering, it is generally accepted that it does not capture the experience in its entirety (Breslau and Davis 1986, Rodin et al 1991).

prevalence of suicides, drug-abuse and divorce (Craig et al 1990, Krause and Crewe 1987, Long 1989, Wilkinson 1989).

Other researchers have combined measures of the patient's psychological well-being with information about physical and social functioning to give an indication of the person's quality of life and/or satisfaction with life (Anson et al 1993, Fallowfield 1990, Fuhrer et al 1992, Schulz and Decker 1985). However, some of these measures have been criticised for relying on the value judgements of professionals or the general public as to what actually constitutes a good or poor quality of life (Wilkinson 1989).

Additional assessments of the patient's response to their condition have focused on self-concept (Matson and Brooks 1977); acceptance of disability (Woodrich and Patterson 1983); behaviour (Walford et al 1993); participation in various activities (MacDonald et al 1987, Terry 1992, Stenager et al 1991); and functional impairment (Rosenstiel and Keefe 1983). Indeed, since chronic conditions can affect so many aspects of daily life, many studies now use multiple indicators, notably measures of emotional well-being, physical impairment and functional capacity, to give a more complete view of adjustment and distress (cf. Goodenow et al 1990).

The prevalence of psychological distress and psychiatric morbidity

Studies have shown that psychiatric disorders as well as subclinical distress tend to be more common among the medically ill and disabled than in the population at large (Rodin et al 1991, Robins et al 1984, Weissman and Myers 1978). For instance, Cavanaugh (1984) studied 335 hospitalized patients and found that 61% had a GHQ-30 score above 5, indicating the presence of emotional distress. The rates of depression (Beck Depression Inventory score >13) varied

according to the illness, from 55.6% in people with gastro-intestinal conditions to 14.3% in people with endocrine diseases.

Similarly, Derogatis et al (1983) assessed 215 cancer patients and assigned a psychiatric diagnosis in 47% of them. Six per cent were judged to have depression or dysthymia, while just 2% fulfilled the criteria for clinical anxiety. Likewise, a study of patients with long-standing diabetes mellitus (type 1) revealed that 51% were suffering from one or more psychiatric disorder (Popkin et al 1988). Major depression was diagnosed in 10.7% and phobic disorder in 20%.

The estimated prevalence of affective disorders doesn't just vary from group to group but also within groups. Taking depression as an example, the estimates in patients with diabetes have ranged from 8.5% to 60% (Lustman et al 1992). This does not include a study of patients suffering from additional complications, which revealed 74% to be depressed (Leedom et al 1991). With regard to multiple sclerosis, estimates of affective disorders have varied from 14% (Joffe et al. 1987) to 90% (Dalos et al 1983). The latter was identified in patients with a progressive course; in those with a more stable condition, the rate of emotional disturbance was much lower (39%). Similarly, Cummings (1992) in his review of the research on depression in Parkinson's disease listed rates from 9 to 81%, with a mean of 40%. Some of the recent estimates of depression in other medically-ill populations are shown in Table 1.

There are a number of reasons why the research relating to the prevalence of psychiatric disorders should be interpreted with caution (Rodin et al 1991, Rodin and Voshart 1986). For example, estimates may vary depending on:

a. the type of measure used (e.g. self-report measures

Table 1. Depression in patients suffering from medical conditions.

Depression %	Measure	Group	Authors
7	Interview	AIDS	Atkinson et al. 88
11	Interview	S.L.E.	Hay et al. 92
12.5	Interview	Muscle Dis.	Wood et al. 91
17	Interview	R.A.	Frank et al. 88
19	Interview	M.I.	Forrester et al.92
20	Interview	SCI	Judd et al. 89
25	HAD	Rectal cancer	MacDonald 1988
38	BDI	Chronic pain	Blakely et al. 91
60	Interview	Tinnitus	Sullivan et al. 88

Key

BDI	Beck Depression Inventory
HAD	Hospital Anxiety and Depression Scale
R.A.	Rheumatoid arthritis
S.L.E.	Systemic lupus erythematosus
M.I.	Myocardial Infarction
Muscle Dis.	Muscle diseases
M.S.	Multiple sclerosis

focus on depressive symptoms whereas standardised interviews can identify depressive illness),
b. the suitability of the measure for a particular sample,
c. the use of different cut-off points,
d. the heterogeneity of the sample (e.g. general medical population or sub-divided into groups according to diagnosis, stability, severity).
e. the unwillingness of some patients to admit to symptoms of depression.

The effect of different types of criteria and measures was clearly demonstrated by Bukberg et al (1984). They studied 90 patients with various types of cancer and found that 42%

met the standard DSM-III criteria for major depression. However, using the BDI, 33% were classified as suffering from depression while on the Hamilton Rating Scale, only 17% had scores above the accepted cut-off point.

Some of the discrepancies documented above can be attributed to the inclusion of somatic complaints. According to Cavanaugh (1991), the core symptoms of depression in the medically-ill are the same as those reported by psychiatric patients e.g. anhedonia, frequent crying, severe indecisiveness and a loss of interest in people. Similarly, a sense of failure and sense of punishment are signs of depression which are generally not confounded by the presence of physical illness (Clark et al 1983). However, there are a number of symptoms which are common to both medical and psychiatric illness and whose inclusion can produce an artificially high estimate of psychiatric morbidity. These include fatigue, insomnia, loss of appetite, psychomotor retardation and difficulties with concentration (Bukberg 1984, Cavanaugh 1991, Clark et al 1983, Frank et al 1988, Krupp et al 1988, Minden 1986, Starkstein et al 1990).

Interest in co-existing psychiatric disorders and emotional distress has been fuelled by an increased awareness of their effects. For instance, it has been found that medically-ill patients with anxiety and depression do much worse in terms of physical, role and social functioning than patients who are psychologically well (e.g. Devellis 1993, Wells et al 1989). More specifically, depression has been shown to increase the risk of angina and emotional instability in people recovering from myocardial infarction (Ladwig et al 1994) and it was associated with marked psycho-social and behavioural dysfunction in patients with rheumatoid arthritis (Beckham et al 1992). In multiple sclerosis, depression has been linked to immune disregulation (Foley et al 1992) while a study on cancer patients found that improvements in affect was related to positive changes in

immune status (Fawzy et al 1990a).

Psychiatric morbidity and distress have also been linked to less effective coping (Zautra and Wrabetz 1991) and may interfere with clinical management and rehabilitation (Malec and Neimeyer 1983, Mayou and Hawton 1986, Rodin and Voshart 1986, Rodin et al 1991).

2.2 Factors affecting psychological adjustment

2.2.1 The effects of social and economic problems

When considering the possible sources of emotional disturbance, it is important to take into account the social as well as the financial consequences of chronic illness and disability. For instance, a variety of surveys over the years have revealed that people with disabilities have a higher rate of unemployment and lower income compared to the non-disabled (Lonsdale 1990). Furthermore, fewer disabled people of working age own their own home, and financial help to enable disabled people to live independently is not always adequate to meet the costs involved (Disability Rights Bulletin, Summer 1993).

Some of these limitations reflect political and judicial systems. For example, the social security benefits for people who became disabled as a result of war or industrial injury are much higher than those for people whose disability resulted from illness, even though the degree of impairment may be same (Disability Rights Handbook 1994).

Other constraints on people with chronic conditions often reflect attitudes among the general population. Thus people with chronic disorders may be subjected to discrimination, both when applying for work and in terms of their salary (Nelson 1992). Lastly, the inaccessibility of certain buildings means that some disabled people are effectively

barred from particular training courses, schools, jobs and recreational activities.

2.2.2 The problem of stigma

Some conditions carry a social stigma (e.g. psoriasis, cancer, AIDS) and this may lead patients to conceal their illness or disability if possible (Goffman 1963, Lonsdale 1990). For instance, they may 'cover up' and 'keep up' in order to appear normal and avoid the negative reactions from others (Locker 1983, Robinson 1988, Wiener 1984). If that is not possible, for example, if the effects of the condition are difficult to hide, then the person may be made to feel inferior and deviant.

Stigma may lead others to infer additional negative attributes, for example, it may be assumed that an individual with a physical disability is also emotionally or intellectually impaired. This further reinforces the inferior-status position of the disabled, and accordingly, their ability to influence decisions which concern them (Thoresen and Kerr 1978).

The media sometimes reinforce the stereotypes of stigmatized groups by providing selective information and presenting composite portraits of the people involved (Schur 1980). However, since stigmatization often lowers people's self-confidence and self-esteem, the 'victims' are generally not inclined to challenge erroneous information about themselves.

Stigma can also affect a patient's medical and emotional health. For example, feelings of stigma attached to rectal cancer was found to be related to an increased risk of poor sleep, fatigue and complications after surgery; with greater use of tranquillisers and analgesics, and with increased rate of clinical depression and anxiety (Macdonald 1988).

Finally, like other social constraints on people with chronic disorders, stigma can undermine psychological health simply by reducing the number of resources which patients can draw on to deal with the consequences of their disease (Dimond 1983, Locker 1983).

2.2.3 The effects of demographic and illness-related variables

2.2.3.1 Demographic variables

a. Age

Since previous experience of illness and coping might help in the process of adaptation, it has been suggested that older patients may react to certain conditions with more confidence and maturity than younger ones. On the other hand, age-related disorders which cause brain damage and/or cognitive disorganization might impair coping capacity and undermine adjustment (Lipowski 1970).

The different possibilities are reflected in the research. Thus studies have linked increased emotional distress both with youth (e.g. Tate et al 1994, Mishel et al 1984, Noyes et al 1990, Viney and Westbrook 1981), and with maturity (e.g. Carroll et al 1993, Cassileth et al 1984, Gilchrist and Creed 1994, McIvor et al 1984).

b. Gender

To date, the research has failed to find a consistent difference in the way men and women react to chronic illness and disability. For instance, while Tate et al (1994) found that men with spinal injuries reported more distress than women, Coyle and Roberge (1992) reported that female patients with a variety of disorders had higher scores on a depression scale than males. Similarly, Forrester et al (1992) found that major depression was more common among women than men following myocardial infarction but Woodrich and Patterson (1983) showed that women were more likely to

accept their disability than men.

c. Education

Higher levels of education have been associated both with lower levels of psychological distress (Christman et al 1988, Moser et al 1993, Viney and Westbrook 1981) and with acceptance of disability (Woodrich and Patterson 1983). This is consistent with the view of Ben Sira (1983), that education is an important resource which enables patients to find and use effective coping strategies.

d. Socio-economic status

Low income has been correlated with high depression scores (e.g. Coyle and Roberge 1992, Tate et al 1994, McIvor et al 1984), as has low social class (Nielsen and Williams 1980).

2.2.3.2 Illness-related variables

a. Onset and course

It has been noted that conditions with a gradual onset may provide more time for patients to adjust to diminishing body function than those with a sudden onset. Likewise, it may be easier to cope with relatively stable, predictable disorders than with diseases which lead to sudden and unexpected problems such as seizures, loss of bowel control, loss of recent memory or severe pain (Dimond 1983).

It has also been suggested that chronic and unstable conditions such as multiple sclerosis can encourage the development of patterns of somatisation. According to Pavlou and Stefoski (1983), the long-term uncertainty and changing nature of the symptoms, plus their severity, raises the likelihood that people will become increasingly vigilant and involved in their bodies. Furthermore, since each new symptom may signal a deterioration, patients may demonstrate heightened responses to minor physical changes (cf Trigwell et al 1995).

b. Severity of symptoms and disability

A number of studies have shown that depression in the medically-ill is at least partly related to the severity of the symptoms and degree of impairment. For example, Stewart et al (1965) examined patients using a standardized interview and found that 20% of the severely affected were clinically depressed. In contrast, only 3% of those with milder illnesses were suffering from depression.

More recently, Cassileth et al (1984) studied a group of 758 out-patients and reported that patients with cancer who were capable of normal activity had significantly lower scores for psychological distress than people who were experiencing more symptoms or who were bedridden. Their findings are consistent with those of Bukberg et al (1984), Craig et al (1994), Folkman et al (1993), Forrester et al (1992), Littlefield et al (1990), McIvor et al (1984), Moffic and Paykel (1975), Noyes et al (1990), Skevington (1986), Tate et al (1994), Viney and Westbrook (1981) and Wineman (1990). However, some studies have failed to find any significant link between level of disability and emotional distress (e.g. Christman et al 1988, Coyle and Roberge 1992, Dalos et al 1983, Gilchrist and Creed 1994, Hay et al 1992, Maybury and Brewin 1984, Ron and Logsdail 1989, Moller et al 1994).

c. Site and extent of disease

Levels of psychological distress have also been associated with the site and/or extent of the lesion. For example, Fleminger (1991) examined 30 patients with Parkinson's disease and found that individuals whose symptoms were worse on the left side were more likely to become depressed and anxious than patients whose symptoms were worse on the right side. This supports the hypothesis that depression in these patients may, at least in part, be due to striatal dopamine depletion in the right cerebral hemisphere.

Likewise, a number of studies on people with spinal cord

injuries have found higher rates of depression among quadriplegics, who have lesions in the neck region, compared to paraplegics, who have lesions lower down (e.g. Judd and Brown 1992, MacDonald et al 1987). Furthermore, Carroll et al (1993) reported higher depression scores in cancer patients with active disease and metastases.

Sometimes a particular condition can alter the emotional repertoire of the patient. For instance, people may become more emotionally labile following a stroke (Gregg et al 1989).

d. Illness intrusiveness

Illness intrusiveness refers to the effects of physical impairment on valued activities and interests. According to Devins et al (1992), this results in patients having less access to positive and rewarding experiences and it compromises their personal control over important outcomes.

Research on this subject is in its infancy but studies have already shown that multiple sclerosis is more intrusive than either rheumatoid arthritis or end-stage renal disease (Devins et al 1993a). Furthermore, illness intrusiveness was found to be correlated with depression in all three patient groups, even after controlling for relevant variables such as the severity of symptoms (Devins et al 1992, 1993b). The measure used to assess illness intrusiveness is currently being validated for British samples.

e. Duration

Increased psychological distress has been documented both in recently diagnosed patients (e.g. Cassileth et al 1984, Richards 1986, Shadish et al 1981) and in those who have been ill or disabled longer (e.g. McIvor et al 1984). Time has also been linked with increased acceptance of disability (Woodrich and Patterson 1983).

f. Visibility of the condition

A number of researchers have associated the visibility of the condition with the risk of emotional distress. For instance, Andreasen and Norris (1977) identified psychological problems in 30% of patients who had been severely burned up to five years previously. They observed that the burns had led to an altered self-image and in some cases, to an identity crisis. In their opinion, those who had adjusted well had redefined their self-image in terms of non-physical and intangible attributes such as courage, perseverence and living for others (cf. Wright 1960). Disfigurement as a result of diseases like cancer has also been linked with increased distress, particularly among women (Noyes et al 1990).

However, having a visible disability can have certain advantages. For example, Viemero (1991) studied patients with muscular dystrophy and found that the visibly disabled reported less depression than those whose disease was not yet visible. The latter had more difficulties in forming new friendships and they felt more ashamed when they had to ask for help. These difficulties were not related to the disability per se, but to the fear of negative reactions to disability.

g. Past history of illness

Previous experience of illness can be both a help and a hindrance. For instance, a past history of angina and mood disorder has been linked with depression following myocardial infarction (Forrester et al 1992). On the other hand, previous stressful experiences in relation to illness and hospitalization were found to help children with cancer withstand the subsequent stressors of a bone marrow transplant (Pot-Mees 1989).

A history of depressive episodes does not appear to have such a protective effect. Indeed, they have been linked

with subsequent emotional distress in both general medical patients (Moffic and Paykel 1975), and the chronically-ill (Minden et al 1987, Lustman et al 1988).

h. Information

Information from health care professionals affects not only how patients perceive their illness but also how they cope (Marteau 1989, Nerenz and Leventhal 1983). It can reduce the ambiguity, anxiety and fear associated with certain conditions, and increase perceived control (Counte et al 1983, Dimond 1983). It is especially important in chronic disorders such as rheumatoid arthritis, where the patient not only has to manage a variety of distressing symptoms but also has to learn to adapt to new and more limited lifestyles (Locker 1983).

Where patients do not receive adequate information from medical staff, they or their families may 'shop around' and seek knowledge from others with experience of the disease (Comaroff and Maquire 1981, Davis 1963). However, if those people lack the necessary expertise, their advice could be harmful and cause further distress (Shepherd 1992).

2.2.4 Psychological factors

2.2.4.1 Personality

A number of personality dimensions have been associated with psychological adjustment. These include resilience (Visotsky et al 1961), sense of coherence (Antonovsky 1987), learned resourcefulness (Rosenbaum 1988) and a number of characteristics discussed below. Conversely, the lack of these traits have been linked with emotional distress and psychiatric morbidity.

a. Hardiness

Hardiness is a composite of three dimensions: commitment, control and challenge. It has been suggested that hardy

people feel able to control or influence the events in their lives, and that they are deeply involved and committed to certain activities. They also tend to perceive change as an exciting challenge to further development (*Kobasa* 1979).

Research to date has indicated that hardiness may also moderate the negative effects of stress on health (e.g. Hills and Norvell 1991, Kobasa et al 1982). Moreover, a study on patients with systemic sclerosis revealed that hardiness was positively correlated with psychological adjustment (Moser et al 1993).

However, the construct of hardiness has been criticised because it overlaps to some extent with neuroticism. This should be taken into account when interpreting the research (Williams et al 1992).

b. Optimism versus pessimism

Optimism has been defined as "an inclination to ... anticipate the best possible outcome" (Scheier and Carver 1987). It's a disposition or orientation, and as such, tends to be fairly stable over time.

In terms of outcomes, optimism has been linked with fewer reports of symptoms, with less depression and with a faster rate of recovery from heart surgery (Scheier and Carver 1987). It has also been associated with less uncertainty and psychological distress in gynaecological cancer patients (Mishel et al 1984); with emotional well-being in people with asthma (Maes and Schlosser 1988) and with good coping in newly diagnosed cancer patients (Weisman and Worden 1976).

In contrast, pessimists appear to be more inclined to dwell on negative experiences, and will use more denial and distancing than optimists (Carver et al 1989, Scheier and Carver 1987). Research has suggested that pessimists also

have poorer immune function (Kamen and Seligman 1989) and that they suffer from more ill-health in the long-term (Peterson et al 1988). However, as in the case of hardiness, there is evidence that neuroticism may have confounded some of the findings relating to pessimism (Smith 1989).

c. Self-esteem

Self-esteem has been defined as "pride in oneself in which one becomes aware of and accepting of one's imperfections while cherishing one's inherent strengths and positive qualities" (Lazarus 1991, p. 441). It may be undermined by illness, especially if an individual's sense of self is rather fragile, or it is closely tied to the person's physical integrity or bodily appearance (Rodin et al 1991).

Low self-esteem has been linked with increased psychological distress three months after a heart attack (Terry et al 1992) and with reduced psychological well-being in patients with rheumatoid arthritis (Krol et al 1994). It has also been associated with feelings of vulnerability in patients with cancer (Weisman and Worden 1976) and with negative beliefs about pain (Williams and Thorn 1989). However, it was not related to the risk of developing a cold (Cohen et al 1991).

2.2.4.2 Attribution

Although the patients' views of what caused their illness may not be accurate, having a causal explanation for symptoms is associated with more positive outcomes than not having an attribution at all (Turnquist et al 1988). However, it has been difficult to identify which types of belief are associated with successful adjustment. For instance, certain attributions may be linked with a positive outcome at one stage of illness, but with poor outcome in another (Van den Bout 1988). Nevertheless, there is some evidence that explanations which attribute symptoms to ex-

ternal sources tend to be related to poor adjustment (Turnquist et al 1988). More specifically, it has been suggested that blaming another agent may reduce some patients' sense of control which in turn might promote feelings of helplessness and hence increase emotional distress (Butler et al 1991, Cope et al 1994b, Lawrie and Pelosi 1994).

Recent research into the relationship between certain beliefs and adjustment has focused in particular, on the influence of perceived control.

a. Locus of control

People's attributions about the onset of their illness may be independent of the perceived controllability of future outcomes. Thus people might blame their fatigue on an external agent such as a virus, yet still see the outcome as something they can control through diet, relaxation, and adjuvant medical interventions.

Much of the research into the effects of perceived control has been stimulated by the concept of locus of control (Rotter 1975). This divides people, dispositionally, into those with an internal locus and those with an external locus. People with an internal locus of control perceive that outcomes are determined by their own actions or their own "relatively permanent characteristics". In contrast, people with an external locus perceive outcomes to be the result of luck, chance, fate or the result of powerful others.

A number of studies have linked an internal locus of control with successful adaptation and adjustment to illness and disability. For instance, both Schulz and Decker (1985) and Devins et al (1993c) found that greater perceived control was correlated with increased psychological well-being. Similarly, Partridge and Johnston (1989) showed that an

internal locus was associated with faster recovery from strokes and fractured wrists, while Frank et al (1987) reported that people with spinal injuries who were high on internal locus of control appeared to be better adjusted. Conversely, Craig et al (1994) found that feeling out of control was a predictor of long term depression in people with spinal cord injuries. Moreover, Shadish et al (1981) noted that external locus of control was related to increased psychological distress in their disabled patients (Shadish et al 1981).

It has recently been suggested that the actual experience of illness or disability may shift perceptions of control, increasing attributions to chance and powerful others (Nagy and Wolfe 1983). This is supported by a study on patients with back pain which found that those with the severest symptoms reported lower levels of internal locus of control but stronger beliefs in others (Harkapaa 1991).

While one might deduce from the literature that attempts to stimulate the patients' internal locus may be adaptive, it is important to note that people may perceive control over one aspect of their well-being but not over another. For instance, Affleck et al (1987a) reported that patients with rheumatoid arthritis tended to perceive more control over their symptoms than over the course of their disease, which they saw as being controlled by others. In this study, greater perceived control over symptoms and treatment was related to positive mood and to psychosocial adjustment, while greater personal control over the course of the disease was associated with increased mood disturbance and poorer adjustment. According to Affleck and colleagues, perceived personal control over symptoms aids adaptation, but a belief in personal control where there is little, may be counterproductive. Thus any attempt to increase a patient's confidence about controlling outcomes where this is actually unrealistic, is likely to lead to frustration as

well as demoralization.

Other studies have also cast doubts on the view that an internal locus of control is invariably related to positive adjustment. For instance, perceived control was not associated with depression in patients with HIV (Folkman et al 1993), nor with psychological distress three months after a heart attack (Terry 1992). Moreover, it was not related to effective coping with health problems (Zautra and Wrabetz 1991), and did not predict health status 10 months after myocardial infarction (Affleck et al 1987b).

It has been argued that perceived control may sometimes have an indirect effect on well-being, for instance, through its influence on coping (Folkman et al 1993). According to Folkman (1984), people who feel in control of their lives may be more likely to appraise demands in terms of challenge; they therefore experience fewer negative emotions and will therefore be in a position to engage in more efficient problem-focused coping. Conversely, people who feel less control may appraise the same demand as a threat, thus experiencing more negative emotion which in turn impedes problem-focused coping.

This view is supported by studies showing that people with an internal and external locus do tend to use different types of strategies (Harkapaa 1991), and that those used by externals may be less effective (Frank et al 1987). However, whether changing patients' attributions will automatically lead to the use of more adaptive coping strategies remains unclear. Indeed, it may not even be helpful in the management of chronic illnesses where shared control with others in the family, and with health professionals, might be more appropriate (Earll 1989).

Thus further research is required to establish if changing people's locus of control is of actual clinical value or

whether it may be more useful to concentrate on specific beliefs about the symptoms and their ability to cope. Until then, attempts to amend patient's beliefs about the control over their condition as a whole may be premature and counterproductive.

b. Self-efficacy

In contrast to the locus of control which focuses on the person's perceived influence on *outcomes or reinforcement*, self-efficacy describes the patients's belief or feeling that they can exercise some control over specific *behaviours and tasks* (Bandura et al 1988, Bandura 1989)[2]. It is therefore a narrower concept than locus of control.

It has been argued that if people are confident that they can do what is required to achieve a certain outcome, this will increase their motivation and their perseverance in the face of adversity (Holman and Lorig 1992). It has also been proposed that self-efficacy influences the self-enhancing or self-hindering nature of people's thoughts and therefore their vulnerability to depression and stress .

Although levels of self-efficacy are dependent, in part, on the person's previous experiences, it is not a generalised trait and can be modified through learning and practising certain activities and techniques. For instance, a course which taught self-management techniques to patients with chronic arthritis raised self-efficacy scores and significantly reduced those for depression and pain (Lorig et al 1989). Moreover, there was a positive correlation between self-efficacy and improvements in health.

Other studies support the view that self-efficacy is associ-

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[2] In theory, people can believe that they have control over certain behaviours (self-efficacy) but regard an outcome or reinforcement as being outside their control (outcome expectancy), and vice versa.

ated with positive outcomes. Thus Terry (1992) found that high levels were correlated with low scores for trait anxiety, as well as low psychological distress, three months after myocardial infarction.

c. Illusion

Some patients have a tendency to evaluate themselves and their degree of control or mastery in an overly positive way and they may be unrealistically optimistic in the face of adversity (Rodin et al 1991). However, this is not always maladaptive. In a few situations, adhering to illusions may help reduce feelings of helplessness and distress (Langer 1976). For instance, believing that one has influence over the cause of illness or relapses, can help patients to cope with the fear and uncertainty of conditions like cancer (Taylor 1983).

2.2.4.3 Uncertainty

Uncertainty is a perceptual state which occurs when internal or external stimuli are vague or unclear. In terms of illness, lack of clarity can make it difficult to interpret the meaning or significance of changes. There may also be ambiguity concerning diagnosis, prognosis, symptoms, treatment, and/or relationships with others (Moser et al 1990).

One condition surrounded by a great deal of uncertainty is AIDS. Although much is known about the disease, it is still difficult to predict the course that the illness will take, the type and severity of the symptoms which patients will experience and the effects of any treatments tried. Sufferers also face the prospect of an undignified death (Weitz 1989).

In other illnesses, uncertainty may arise as a result of marked fluctuations in the severity of symptoms, as in multiple sclerosis (Robinson 1988) and rheumatoid arthritis (Wiener 1975). In these cases, the changeability in symptoms

means that the patient's appraisal about being able to manage is constantly challenged (Robinson 1988).

The Uncertainty in illness theory

The Uncertainty in illness theory (Mishel 1988) explains how patients cognitively process illness-related stimuli and construct meaning in these events.

According to Mishel, how much uncertainty is perceived depends on the salience and pattern of the symptoms, familiarity with illness-related events and situations, the degree of congruence between the expected and the unexpected, the patients' information-processing abilities (cognitive capacity), their confidence in a credible authority such as doctors, their own education and the availability of social support (see Fig.1).

Mishel (1988) proposed that those with an external locus of control may perceive uncertainty as threatening, people with a disposition towards internal control may appraise it as an opportunity and see it as a sign that there is still hope. However, although there is some evidence that lack of certainty can be perceived in a positive way (e.g. Taylor 1983), it is more often regarded as a source of distress (Davis 1960-1). Indeed, studies on a number of different conditions have linked increased uncertainty with pessimism (Mishel et al 1984), emotional distress (Christman et al 1988, Wineman 1990) and poor adjustment (Mishel and Braden 1987, Moser et al 1993, Wineman 1990). It has also been associated with a reduction in self-help (Braden 1990), a reduced sense of mastery (Mishel et al 1991, Mishel 1990), a lack of motivation, poor expectations about the future (Mishel et al 1984) and a reduction in active behaviours (Mishel et al 1984, Christman et al 1988). These findings indicate that uncertainty may undermine the psychological well-being of the medically-ill and as such, should be taken

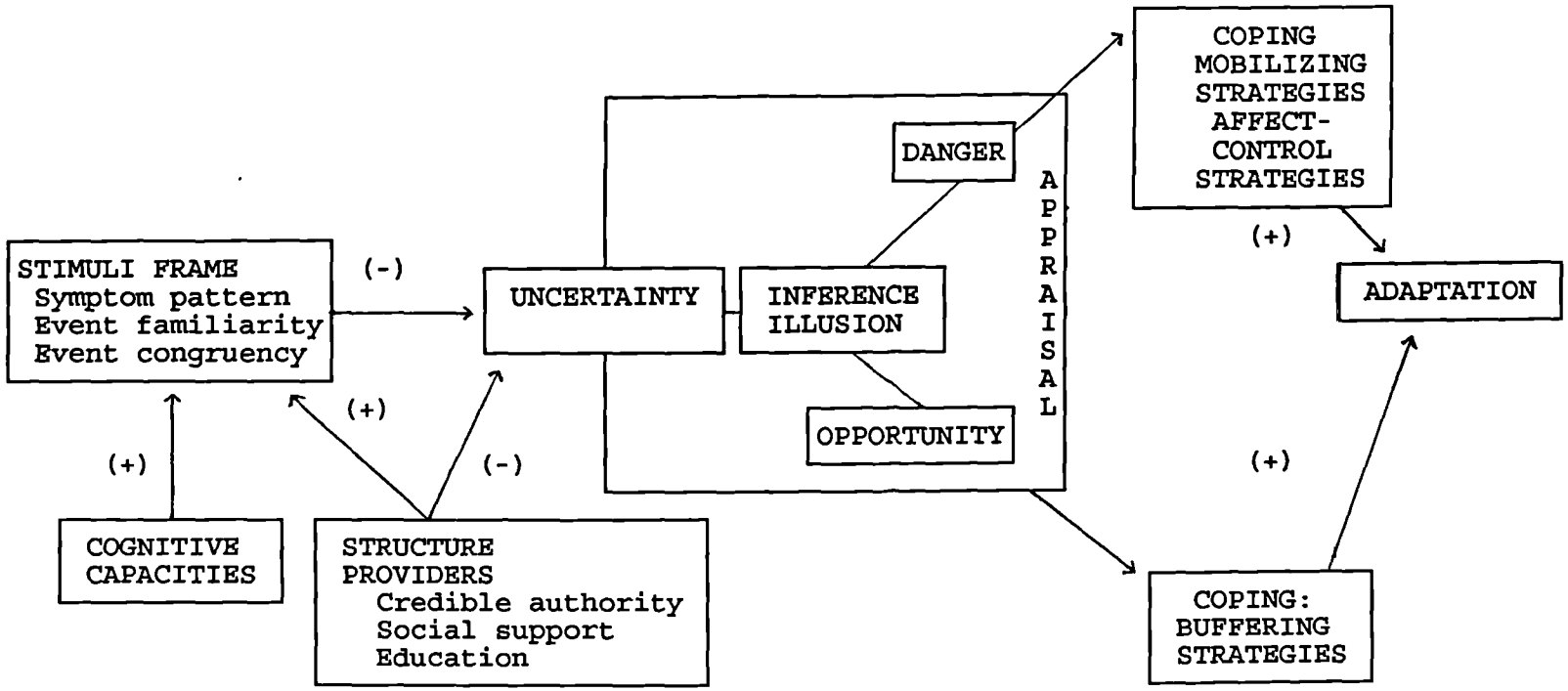

Figure 1. Model of perceived uncertainty in illness (Mishel 1988)

into account when considering the variables associated with emotional distress.

2.2.4.4 The role of life events and stress

Distressing life events can have a direct as well as indirect effect on psychological well-being (Andrews et al 1978, Folkman et al 1993). For instance, they may influence immune function (Adler and Matthews 1994, Brown and Harris 1989) and increase the susceptibility to relatively minor disorders such as colds (Cohen et al 1991).

The relationship between stress or 'daily hassles' on the one hand, and adjustment on the other (Kanner et al 1981) may be mediated by variables such as perception of control and the choice of coping strategy. The literature on the links between coping and emotional distress will be discussed in more detail below.

2.2.5 The relationship between coping and psychological adjustment

Lazarus (1991) defined coping as 'cognitive and behavioural efforts which are used to manage specific or internal demands that are appraised as taxing or exceeding the resources of the person'. The strategies which people chose are often divided into two categories, namely problem-focused ones, which are directed at managing the difficulty and changing the actual situation for the better, and emotion-focused ones, which are primarily aimed at regulating emotions, including distress.

Commonly used problem-focused coping strategies include:

1. information-seeking. This involves searching for knowledge which will enable a person to learn more about the problem and what can be done to deal with it. It provides a basis for action and rationalisation.
2. direct actions. These include concrete acts like taking

medicines, arguing with opponents and running away (escape-avoidance).
3. inhibition of action. This includes the holding back of impulses that may increase the probability of negative outcomes.

Commonly used emotion-focused strategies include:
1. intrapsychic strategies. These involve ways of reappraising the situation and redirecting attention, and include defence-mechanisms such as denial and intellectualization, wishful thinking, minimizing the threat, and ignoring or withdrawal of attention (distancing).
2. turning to others for help and/or support. (However, where support is needed for direct actions, this may also be regarded as a problem-focused strategy).

Which approach is chosen depends on a number of factors. For example, the choice of strategy may vary according to the severity or stage of illness (Bracken and Shepard 1980, Buddeberg et al 1991, Cohen and Lazarus 1979, Davis 1963, Heim et al 1993, Matson and Brooks 1977, Shapiro et al 1994, Viney and Westbrook 1982, Visotsky et al 1961). Other influences include the number of medical problems (Ehmann et al 1990), the physical environment i.e. whether one is in hospital or at home (Christman et al 1988, Heim et al 1993) and the site of the lesion (Krantz and Deckel 1983).

Situational variables may also play a role. For instance, research has shown that distancing and escape-avoidance are used more often in situations which are regarded by the person as outside their control (Folkman et al 1993). Conversely, problem-focused strategies are more common in encounters appraised as controllable (Folkman et al 1986, Carver et al 1989).

It is important to underline that coping strategies may change over time because what is attended to, and the

threats themselves, also change. Moreover, the complexity of most stressful encounters means that people will tend to use a variety of strategies, changing from one to another on the basis of feedback (Lazarus 1991).

In addition to illness and situation-related variables, coping may also be influenced by a personality factors such as optimism (Carver et al 1989) and by the level of education (Lacroix 1991).

Recent research on coping has also looked at the way in which patients use information. For instance, Miller et al (1988) proposed that some people, referred to as 'monitors', might be more inclined to attend to symptoms and to seek out information about illness and treatments than others. In contrast, 'blunters' may prefer to avoid and distract themselves from threat-relevant information.

This view is supported by a study of hospital attenders which showed that high monitors (information-seekers) demanded more tests, more details about their health problems and more counselling from their doctors than the low monitors, who typically ignored information. Interestingly, the former preferred to play a less active role in their own care than their low-monitoring counterparts. Thus it appears that the high monitors sought information, not so much to control their illness but to reduce uncertainty and concomitant arousal.

In terms of outcome, blunting has been associated with reporting of psychological symptoms and with complaints related to infections such as colds and flu (Davey et al 1993).

The actual act of seeking information has also been associated with positive psychological adjustment (Felton and Revenson 1984). However, it has been suggested that

this may be partly due to its value as an attention-diverting strategy, helping patients to focus away from pessimistic thoughts towards seemingly more useful matters.

Another variable which may affect the choice of coping strategy is mood. For example, depression has been associated with a greater use of emotion-focused coping such as wishful thinking, with increases in self-blame (Beckham and Adams 1984) and with the seeking out of social support (Coyne et al 1981, Vitaliano et al 1989, 1990). These strategies appear to be influenced by the actual level of depression, rather than constitutional differences or a general vulnerability (Parker and Brown 1982).

Anxiety too may affect coping. Fisher (1986) hypothesized that anxiety would produce "ragged, disorganised, unplanned" behaviours, leading to a reduction in efficiency and competence. Nevertheless, moderate levels of certain emotions may be adaptive (Dirks et al 1978).

In contrast to the illness-related variables and the effects of mood noted above, there is little evidence that either income or gender influence coping in the chronically-ill (Ehmann et al 1990, Viney and Westbrook 1982).

Coping and adjustment

In terms of outcome, the effectiveness of most strategies must be judged in context (Lazarus 1991). Thus an approach which is helpful in one situation may be far less useful in another. For example, although denial prior to diagnosis of breast cancer temporarily reduces emotional distress, it also delays diagnosis and treatment and therefore increases the risk of metastases and poor outcome. At this stage, the efforts at coping do not meet the requirements of the environmental conditions being faced, and they are therefore regarded as maladaptive. On the other hand, the same stra-

tegy used after diagnosis might help the person to deal with the threat of death and may even lengthen survival time (Greer 1991, Taylor 1983).

While it is therefore important to consider the context, there is evidence to suggest that efforts to enhance coping skills may lead to a overall reduction of psychological distress (Fawzy et al 1990b, Cunningham et al 1993). In terms of the efficacy of specific strategies, active behavioural approaches appear to be useful in reducing depression for some patient groups, while more emotion-focused strategies, e.g. distancing and passive-avoidance seem to have the opposite effect (Ehmann et al 1990, Fawzy et al 1990, Felton and Revenson 1984, Folkman et al 1993).

This does not mean that problem-focused strategies are always adaptive. For instance, Terry (1992) studied patients recovering from myocardial infarction and found that problem-focused strategies were not related to any measures of favourable adaptation.

Finally, it is worth noting that the influence of coping efforts may be more limited than is sometimes claimed. For instance, Macrae and Costa (1986) suggested that coping strategies might be used particularly as a means of maintaining good spirits despite adversity, and they observed that in some situations, coping behaviours had only modest effects on wellbeing. This view is supported by Felton and Revenson (1984) whose research showed that the effects of coping on adjustment were approximately equal in strength to the effects of adjustment on coping. On the basis of these findings, they concluded that coping appears to have a relatively modest role.

Coping with uncertainty

To deal with uncertainty, patients have used a number of

strategies, for instance, information seeking (Comaroff and Maguire 1981, Weitz 1989) and pacing of activities (Wiener 1975). They have also engaged in affect management through denial, avoidance, optimism and by comparing themselves with other patients (Mishel 1988, Comaroff and Maguire 1981). Furthermore, some have coped by choosing to believe that they have control over their illness, even though in reality, this is very limited (Taylor 1983).

2.2.6 The role of culture

Cultural norms determine how a society reacts towards the disabled, whether they are accepted or isolated, pitied or censured (Cassel 1982). Another way in which culture may influence health care was described by Lopez (1989). He argued that doctors who do not have a great deal of contact with a certain group of patients may have a more homogeneous view of these individuals and as a result, may only consider a relatively limited range of diagnoses. In his view, stereotypic beliefs concerning certain races, women or older people can interfere with the process of gathering evidence to test diagnostic impressions and hypotheses. In this way, hypotheses may be prematurely accepted as valid, leading to diagnostic errors, inappropriate management and increased distress for the patient concerned.

Culture may also influence the actual labelling of symptoms. For example, neurasthenia is a relatively common diagnosis in some countries and rare in others (Ware and Kleinman 1992) while low blood pressure is regarded as a disorder in Germany but not in the UK (Wessely et al 1990). This is important because the actual diagnosis not only determines the treatment offered by physicians but also the attitude of the general population and the coping strategies adopted by the patient. This will be discussed in more detail in the section on support from physicians.

2.2.7 The role of social support

Another factor which is thought to play a major role in the psychological adjustment to chronic illness and disability is social support. This has been defined in various ways (Wortman and Dunkel-Schetter 1987), but generally refers to the presence of others or the resources provided by them, prior to, during or following a stressful event (Ganster and Victor 1988).

Social support may include any or all of the following components:
1. help to clarify or further one's understanding of problems and possible solutions (cf informational support),
2. help with fears, making people feel valued and loved, caring, sympathy, understanding and reassurance,
3. provision of tangible assistance (practical help) with chores and tasks,
4. provision of feedback on how one is doing,
5. provision of physical comfort,
6. access to material resources (this is sometimes referred to as instrumental support),
7. provision of companionship, reducing people's sense of isolation and strengthening their sense of identity in times of uncertainty (Bloom 1982, Cohen and Wills 1985, Ganster and Victor 1988, Fiori et al 1986).

It has been suggested that the most important aspect of support is the recipient's perception of affection, acceptance and the affirmation of personal worth (Justice 1994, Sarason et al 1988). However, this generalisation does not take into account the changing needs of patients with chronic conditions.

Direct and indirect effects

Social support has both direct and indirect effects. Direct

effects encompass the general positive influences of social support, irrespective of whether a person is under stress. For instance, relationships may reduce or prevent illness by providing people with regular positive experiences, and a sense of predictability and stability during periods of rapid change (Cohen and Wills 1985, Stout et al 1964). Social support may also enhance well-being by facilitating health-promoting activities such as proper sleep, exercise and the appropriate use of alcohol and drugs (House 1988).

Research on the direct effects of social support has linked it with successful coping with various crises (Andrews et al 1978), while a lack of support has been associated with destructive behaviours (Brennan and Moos 1990) and an increased risk of illness and mortality (Bloom 1982, Joseph and Syme 1982, Rosengren et al 1993).

As well as exerting a direct effect on well-being, social support may also help to reduce illness indirectly by acting as a buffer between the individual and the negative psychological consequences of stress. According to Cohen and Wills (1985), contact with others may prevent a stressful event from being appraised as harmful and bolster one's perceived ability to cope. Social support can also influence later appraisals and reactions, for instance, by facilitating an adaptive counter response or by moderating physiological processes (e.g. Henry 1986).

It has been suggested that the buffering effects of social support may help to reduce people's general vulnerability to distress and disease (e.g. Brown and Harris 1989) and increase their resistance and resilience (e.g. Cassel 1976).

Structural and functional characteristics

In addition to the direct and indirect effects, it is also

useful to distinguish between the structural and functional characteristics of support. Structural characteristics include the size of the social network, the frequency of contacts, the stability of the support over time and the source of support, e.g. whether it comes from family and friends, or from more informal sources e.g. club members, church fellowship or a support group. Functional characteristics include the perceived quality of the available support, i.e. whether people feel that there is someone they can turn to if need be, and the satisfaction with the support received.

The distinction is important since there is evidence that structural measures tend to be associated with main (direct) effects whereas functional measures are more frequently associated with interactive or buffering effects (Cohen and Wills 1985). Furthermore, it has been found that the quality of support is generally a stronger predictor of health outcome and psychosocial dysfunction than the quantity of the support (Broadhead et al 1983, Fiori et al 1986, Fitzpatrick et al 1991).

The effects of social support are also influenced by a number of other variables. In terms of dealing with chronic illness and disability, research has shown that social support may lead to different outcomes depending on:
a. the type and stage of illness i.e. whether it is serious or trivial, treatable or manageable (Ell et al 1992, Elliott et al 1992, Neuling and Winefield 1988, Revenson 1993),
b. the environment, e.g. whether one is at home or in hospital (Broadhead et al 1991),
c. the specific needs of the patient, e.g. whether they desire information, reassurance, tangible assistance, economic help etc (Dakof and Taylor 1990, Neuling and Winefield 1988),
d. the nature of the support provided, and whether it matches the needs of the patient (Broadhead and Kaplan 1991,

Revenson 1993),
e. the psychological state of the patient, e.g. depression may limit the amount of support offered or perceived (Billings et al 1983, Fitzpatrick et al 1991),
f. the source of the support e.g. partner, friend, relative, health-care professional (Elliott et al 1992, Neuling and Winefield 1988),
g. the extent of the support, e.g. sufficient, inadequate (e.g. Stewart and Sullivan 1982, Faucett and Levine 1990),
h. the health and skills of the support provider, e.g. whether they have the patience required, the knowledge to give the right advice etc, the strength to assist with needs relating to self-care (e.g. Strauss et al 1984).

One study which examined the effects of different types of social support given to cancer patients found that emotional support (presence, concern, affection) from family and friends was regarded as most helpful (Dakof and Taylor 1990). Most unhelpful was a lack of emotional support, avoidance of contact and misguided support. In terms of help from other patients and physicians, informational support was most helpful and misguided or absent informational support was considered as unhelpful. About 40% complained about not receiving enough emotional support from these quarters and many were upset when other patients acted in a self-destructive or foolish way. These findings, plus those of Neuling and Winefield (1988) and Elliott et al (1992), underline the importance of distinguishing the sources and types of the support since they may have different effects on outcome.

Finally, the presence of social support may affect the actual choice of coping strategies. For instance, Moos and Moos (1990) found that those adults who received support from their spouse and friends relied more on approach coping (e.g. seeking guidance and support) when dealing with stressors and were less likely to use avoidance strategies, espe-

cially cognitive avoidance and emotional discharge compared to those who received less support.

Positive and negative effects of social support in the chronically-ill

Social support has been shown to have beneficial effects on the well-being of patients with various chronic conditions (e.g. Bowden et al 1980, Fitzpatrick et al 1991, Goodenow et al 1990, Lloyd and Cawley 1983, Ray 1992, Schulz and Decker 1985, Terry 1992, Wineman 1990). More specifically, it has been found that support can help people to accept their illness (Martin 1982) and enhance their ability to cope and comply with difficult treatment regimes (Ganster and Victor 1988, Gregg et al 1989). It has also been linked with reductions in psychological distress (Goodenow et al 1990, Littlefield et al 1990, Revenson et al 1991) and with higher levels of functioning (Goodenow et al 1990, Ward and Leigh 1993). Moreover, it was a significant predictor of higher natural killer cell activity in women with breast cancer (Levy et al 1990).

Conversely, lack of support has been correlated with increased depression (e.g. Revenson et al 1991, Wineman 1990) and anxiety (Whalley Hammell 1994), while limited attachments and "loneliness" have been associated with reductions in immune function (Kiecolt-Glaser et al 1984, Theorell et al 1995).

However, support can hinder as well as help patients to cope. This was summarised by Suls (1982) as follows:

	Positive effects	Negative effects
Prevention	reduce uncertainty/worry	increase uncertainty and worry
	set good example	set bad example

	Positive effects	Negative effects
	share problems	create new problems
	distract	distract
		contact with germs
Coping	label beneficial	label negative
	provide sympathy	subject to irritation and resentment
	give helpful information	give misleading information
Recovery	maintain regimen	discourage regimen
	contrast with health (incentive)	contrast with health (depressant)
	create desire to stop being a nuisance	create power/dependence need

For example, while being part of a social network allows people to share their problems, this can both reduce fear and create more uncertainty and anxiety. Furthermore, the presence of others can make people feel embarrassed thus increasing their distress.

Research has also shown that people may actually withhold support. For instance, when confronted by the suffering of others, some individuals may try to protect themselves from the fear of illness and feelings of vulnerability by convincing themselves that the patient was to blame (Lerner and Simons 1966). Negative views may co-exist with positive ones, or they may dominate. For example, if an illness can be linked to the patient's behaviour, e.g. lung cancer to cigarette smoking, significant others may signal their irritation and resentment to the person concerned. It has also been observed that where conditions have no clearly identifiable organic cause, some people find it hard to accept the

patients' suffering and this could result in conflict and increased distress (Donoghue and Siegel 1993, Faucett and Levine 1990).

In the case of diseases like cancer, others may feel helpless or find it difficult to hide their pessimism. These factors, and the strain of certain conditions, can cause people to turn away from those who are ill and therefore reduce their social support (Buunk and Hoorens 1992, Mitchell and Moos 1984, Wortman and Dunkel-Schetter 1979).

Social support and uncertainty

Support from others may help to reduce uncertainty and as a result, limit the level of emotional distress (Mishel 1988). For instance, on the basis of their study on women with gynaecological cancer, Mishel and Braden (1987) claimed that the awareness that help was available reduced the uncertainty about the future. This allowed the women to invest in their present relationships and activities, thus improving psychosocial adjustment.

The support from the physician.

It has been suggested that the ability of practitioners to communicate with and support their patients is of enormous influence (Davis 1963, Lloyd 1991, Macdonald 1988). This is true both in the prediagnostic phase and following diagnosis (Stewart and Sullivan 1982).

One factor which may undermine the communication between physicians and patients and thus limit support is that both groups approach illness from different perspectives. As Toombs (1992) has pointed out, physicians approach illness from a scientific perspective; their training leads them to focus on signs and symptoms and on identifying a particular disease state. In a sense, they reclassify the patient's

experience of illness in terms of the findings of the basic sciences, with the body as scientific object.

Patients have a totally different view of illness. To them, it is a disruption to everyday life; a subjective and unique experience which limits their ability to engage the world in habitual ways. Illness represents a loss of total body integrity, certainty, control and freedom to act. It reduces choices, and the suffering is related to all these factors, not just the biological malfunctioning of one or more organs.

It has been proposed that the physicians' emphasis on the biological body leaves them with an incomplete knowledge of the illness and the suffering that it causes (Baron 1985). The resulting lack of understanding may lead to inappropriate treatments and consequently, to the suboptimal management of the illness.

A more specific problem associated with the biomedical approach relates to the patients who do not meet the criteria for disease. If clinicians cannot find evidence of abnormalities on 'objective' tests or if the patient's complaints do not correlate with demonstrable pathoanatomical and pathophysiological findings, they may well conclude that the patient does not have a bona fide disease. Consequently, patients may be told that "there is nothing wrong", or that the illness "is all in your head"; two assessments which not only contradict actual experience but imply that the distress is not legitimate. In such cases, the physician's failure to construe their illness as a 'disease state' is an additional source of suffering which the patient must cope with.

According to Rippere (1992a), the inaccurate labelling of disorders as psychogenic may have a negative effect on social interactions with significant others. This view is

consistent with that of Stewart and Sullivan (1982) who studied a number of patients with multiple sclerosis and found that the prediagnostic uncertainties, plus the doctors' refusal to legitimize the adoption of the sick role during this period, led to negative reactions from family and friends and to emotional conflicts and tension. All these factors combined to cause what they refer to as "a type of iatrogenic disease". In fact, they found that "feelings of frustration, worry and intermittent periods of depression were nearly universal. Over half of the patients also reported experiencing more severe psychological problems". In their opinion, the latter was directly attributable to the stress of the doctor-patient relationship.

Support from groups

During his survey of people with arthritis, Locker (1983) discovered that a major source of support for his patients were fellow sufferers. Apparently, those not affected could not understand the pain associated with the disease, and consequently tended to minimize the distress. The shortage of support led many patients to join self-help groups.

Support groups have also been found to play an increasingly important role for patients with other chronic disorders, such as motor neurone disease and multiple sclerosis. Robinson (1988) summed up the benefits of the British organisations for people with MS as follows: "they provide a means of focusing hope, anger, desperation, needs for companionship in a common situation, a wish to help others, a search for practical advice, and many other concerns which cannot easily be met in the intimacy of family life, or in the colder world of professional medicine".

In conclusion, it appears that the many benefits of social support will be optimised when it fits and satisfies the

needs of the individual (Broadhead et al 1983). Greater awareness of the patient's wishes and the importance of support may help improve psychological well-being and reduce the rate of psychiatric morbidity in the chronically-ill.

2.3 Discussion

The research shows that different illnesses and conditions exert different demands on the person. Therefore, it is important when assessing psychological adjustment to take into account the severity and stability of the symptoms, not just the nature of the pathology and the duration of impairment (cf Walford et al 1993). This is particularly relevant for conditions like CFS, which are characterised by a number of different and fluctuating symptoms. Indeed, the changeability of CFS means that variables which are linked to adjustment in cross-correlational studies should also be examined using longitudinal designs (cf. Kobasa 1985, Patarca et al 1993).

Given the differences between the various conditions, any comparison of psychological morbidity among the chronically-ill must take into account such variables as the severity of symptoms and the level of overall disability in each of the patient group. Moreover, if assessing the influence of a medical disorder on emotional distress, note should be made of specific influences such as the attitude of the general population towards that condition, and the availability of treatment. For instance, it is possíble that disorders like CFS, which are surrounded by controversy and for which there is limited treatment, may provide more challenges for people than conditions which are acknowledged as genuine sources of disability and for which there are several treatments to reduce or control the most distressing symptoms.

Further consideration is also required when assessing the influence of personality on adaptation. For instance, there

is evidence that measures of personality dimensions such as hardiness and pessimism may be confounded by neuroticism (e.g. Smith 1989). Indeed, it has been suggested that the instruments used to measure *constructs such as hardiness*, optimism, self-esteem and locus on control actually form a single major dimension which Marshall et al (1994) call 'optimistic control'. Thus the relationships between those constructs and adjustment may be partly due to fact that the measures may be tapping the same underlying domain. If these issues are not addressed, it will be difficult to evaluate their role in the psychological adjustment to chronic illness and disability.

In terms of examining the influence of attributions and perceived control, research seems to indicate that confidence about being able to manage certain aspects of the illness may be associated with successful adjustment. However, when considering conditions like CFS, two problems need to be addressed. Firstly, given the lack of clarity regarding the aetiology of the syndrome, one cannot judge whether a viral attribution is correct or incorrect. Moreover, a somatic attribution cannot be regarded as maladaptive, simply on the basis that it is statistically associated with ongoing ill-health (Powell et al 1990). Since there is also a positive relationship between somatic attributions and poor outcome in patients with known 'organic' diseases, the significance of an external versus internal attributions has yet to be established (Salkovskis, personal communication, Trigwell et al 1995).

The same argument applies to the research into perceived control. The complexity of many conditions means that patients may feel they can control specific symptoms but not the course or outcome of disease. Furthermore, some of their beliefs may be realistic, others may be regarded as illusions. Again, lack of knowledge about the mechanisms underlying CFS makes it difficult to assess which aspects of

the disorder are controllable and to what extent. Given these uncertainties and the literature on other conditions, it may be more useful to focus on self-efficacy, i.e. perceived control over certain behaviours, and how this relates to coping and adjustment (cf. Holman and Lorig 1992).

Another variable which has received limited attention in relation to CFS is uncertainty. Given the fluctuating nature of CFS and the difficulties relating to its diagnosis, this variable should be taken into account when considering the psychological effects of this condition (e.g. Wineman 1990).

Research on the effects of CFS might also focus on the nature of coping. Studies on other chronic disorders suggest that patients may use a variety of strategies, depending on the nature and severity of the symptoms, the stage of the illness, concurrent mood and the success of the strategies (Davis 1963, Fisher 1986, Lazarus 1991, Matson and Brooks 1977). However, the range of strategies used by patients with CFS has yet to be determined. Moreover, since a specific strategy may have different effects at different times and on different aspects of functioning, research on the relationship between coping and adjustment should include a number of different outcome measures, and if possible, study their effects over time (Lazarus 1993).

There is also limited knowledge about the extent of social support given to patients with CFS and its relationship with emotional distress. The literature on other disorders suggests that social support can be of enormous benefit, but it remains unclear how support exerts its effects. For instance, information provided by others may lead to an increased perception of control, so that threats are more likely to be seen as challenges and they arouse less distress. Equally, it could make people aware of adaptive responses and therefore affect coping more than primary

responses and therefore affect coping more than primary appraisal. At the same time, information could enhance mood (e.g. optimism) and thereby influence the neuroendocrine pathways. Unfortunately, these questions require comprehensive studies which, given the health of many people with CFS, may not be practical. Other issues which might be addressed include the influence of the uncertain aetiology on support and the effects of support provided by different sources. Moreover, studies should distinguish between the quality and quantity of the support (e.g. Cohen and Wills 1985, Faucett and Levine 1990).

Finally, given the importance of the doctor-patient relationship, it may be useful to assess the satisfaction with medical support and advice in studies relating to adjustment and treatment of CFS.

The table below summarises some of the variables which are thought to influence psychological adjustment in the chronically-ill and disabled.

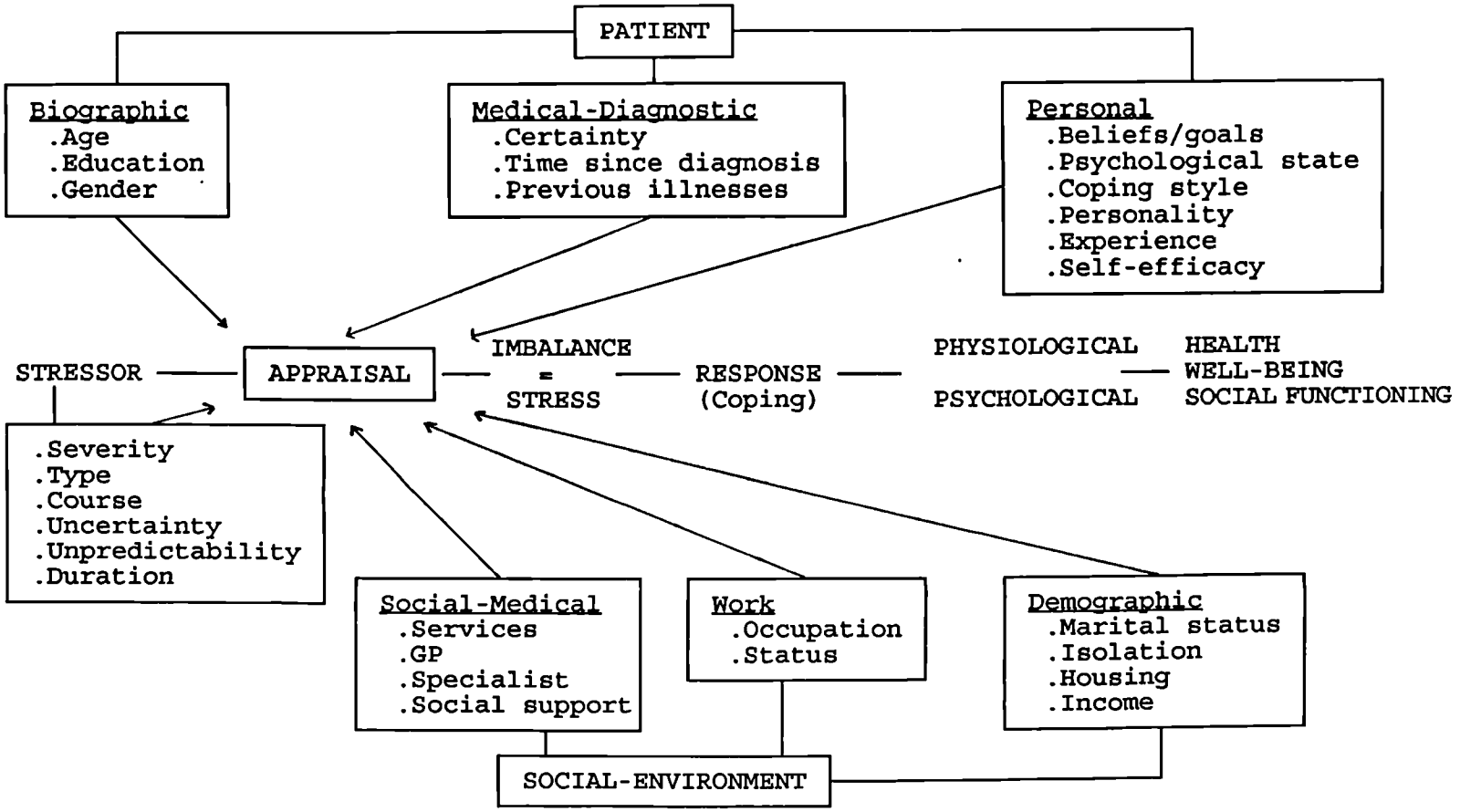

Figure 2. Possible influences on adjustment to illness (Adapted from Lazarus 1993).

CHAPTER 3

Study into the experiences of patients with chronic fatigue syndrome

> "It was unfair to have an illness without a name, without recognition, without outer signs, without looking ill, without knowing what to do to get better" (2.34).

3.1 Introduction

Accounts of the psychological aspects of CFS suggest that most patients respond to their symptoms in similar ways (e.g. Sharpe 1994, Taerk and Gnam 1994). A recurring theme is that sufferers adhere to fairly straightforward aetiological models, that they are reluctant to consider the possible role of emotional problems (Lawrie and Pelosi 1994, Surawy et al 1995) and that they use ineffective coping strategies (Lewis et al 1994).

One view which many patients appear to share is that CFS is a result of ongoing infection (Shepherd 1992). According to Wessely et al (1989, 1991), this belief "conveys certain advantages, irrespective of its validity. It is simple, frequent and easily accepted". He also notes that it removes self-blame and guilt and avoids the stigma of mental illness. However, the disadvantages are that it also takes away some control over the symptoms and that it may lead patients to reject potentially effective treatments.

The patients' tendency to blame a physical cause has been documented by several studies. For example, both Wessely and Powell (1989) and Manu et al (1993) found that the vast majority of their patients attributed their condition to non-psychological factors. However, other studies have shown that the adherence to external attributions may not be

as widespread as some have suggested (Ray et al 1992b, Yeomans and Conway 1991, Ware and Kleinman 1992).

In terms of behaviour, it is often assumed that patients with CFS tend to adopt a fairly passive approach to their illness. As Wessely et al (1991) put it, the reaction of many is to "rest and to wait either for remission or a medical cure".

This view supported by Blakely et al (1991) who found that patients with CFS tended to use more escape/avoidance and distancing than people with chronic pain. However, other researchers have shown that people with CFS also use problem-focused strategies (Lewis et al 1994). These will be discussed in more detail in Chapter 5.

The portrayal of patients with CFS as passive 'victims' who do little other than rest also conflicts with studies carried out by self-help groups. For instance, a survey conducted by the British patient association Action for ME revealed that rest, hydrotherapy, relaxation techniques, dietary changes, massage, anti-candida treatment, healing and aromatherapy were all assessed as useful by at least 50% of respondents (Interaction 1991, 8). Least helpful were antibiotics, steroids, fasting, tranquillisers and graded exercise. In all, 71% of those who improved attributed this to some or all of the therapies they had tried.

In the past, modern medicine has tended to ignore and discount subjective experience in favour of the hard, objective, quantitative data of laboratory tests, x-rays and so forth. As a result, few scientists have been aware of the patient's lived experience and what that experience means to the person concerned (Baron 1985). However, as Cassell (1982) pointed out, the physician can not begin to address the patients' suffering unless attention is paid to such meaning. Indeed, he believes that "failure to understand

the nature of suffering can result in medical intervention that (though technically adequate) not only fails to relieve suffering but becomes a source of suffering itself".

Toombs (1992) has therefore advocated that doctors should take more account of the patients' own story, the 'clinical narrative', to find out what is significant to them, what their values are, and how they would like their illness to be treated. This approach may be particularly relevant to CFS, where the emphasis on fatigue may have limited the recognition of the strain associated with this condition. Moreover, the simplified accounts of the illness and the patient's experiences may have resulted in an underestimation of the real psychological sequelae of CFS (Dutton 1992).

To summarise, there has been relatively little research on the psychological and social aspects of CFS as viewed by the patients. Moreover, the interest in fatigue and the paucity of information about other sources of disability and distress may have led many researchers to regard psychological disturbances as a cause rather than a response to the illness.

3.2 Research aims

Given the lack of research, it was decided to conduct an exploratory study into the illness from the patients' perspective, focusing in particular on the difficulties which they face and the type of coping strategies they use.

3.3 Method

Information was obtained from interviews and questionnaires, providing both qualitative and quantitative data.

3.3.1 The interviews (Group 1)

Six local practices, giving access to at least 15 general practitioners, were approached for permission to interview patients with CFS (then known as ME or PVFS). Of those contacted, one doctor replied that he had no ME patients and one practice did not reply. However the others were happy to co-operate with the study and to send on letters asking for volunteers. A few also provided names directly.

When approached, one patient felt too ill to take part at that time and one had recovered to the extent that he thought that he was unsuitable. Since it is not known how many letters were sent out by the general practitioners, it is not possible to calculate the exact response rate.

Seven of the interviews took place in the patients' homes and the responses were taped for later transcription. However, due to circumstances beyond the researcher's control, the other interviews were conducted via the telephone and the researcher attempted to take down the responses as fully as she could.

The patients attended a total of 11 different doctors, with the majority coming from practices in Twickenham and Teddington.

Although it was hoped that the general community sample would be representative of the patient population as a whole, it was found that all the 17 people in this group were or had been members of either the ME Association or Action for ME. This may have coloured the people's views of the illness and their approach to treatment. Nevertheless, the interviews provided an opportunity to explore the personal experiences of patients in much more detail than the questionnaires.

The choice of questions reflected the aims of the study, i.e. to learn more about the illness from the patients' perspective. These are described in more detail in section 3.3.3.

No one was paid for their participation.

3.3.2 The questionnaires (Group 2)

One hundred people, chosen at random from the membership list of the ME Association, were sent a letter asking for volunteers. In total, 66 replied that they were willing to participate. Of the ten who did not return their questionnaires, one withdrew, one had recovered and did not feel she could be useful, and five had moved or did not wish to be contacted further. Three copies were apparently lost in the post. The remaining three completed the questionnaires but these could not be used because the respondents had not been formally diagnosed. This left a total of 53 questionnaires, giving a response rate of 53% for the sample as a whole and 80% among those who initially agreed to participate.

3.3.3 Design

The first part of the questionnaire requested demographic information (age, gender, marital status, work status, education, housing and income). This was followed by questions about the illness, for instance, whether the onset was acute and triggered by a specific infection or gradual; the duration and course of the illness; the main symptoms; how the diagnosis was made and who had made the diagnosis (people who had not been diagnosed by a physician were not included in the statistical analysis).

In the second part, patients were requested to list the factors which they saw as causes of their own illness. They were also asked which factors they regarded as general

causes of CFS and which factors they saw as irrelevant or unlikely causes. This would help to establish if, as has been suggested, patients have a tendency to blame external factors such as viruses.

The rest of the questionnaire focused on the effects of the illness and the strategies which patients had used to cope with them. Again, it was felt that open-ended questions would be most appropriate. To guide patients, there were three separate questions relating to the consequences of the illness. The first asked about the effect of the condition on activities, career and so on; the second focused specifically on relationships and the third enquired about the consequences of their illness on their personality, feelings and attitudes.

The section on coping strategies asked what patients had done to deal both with day-to-day symptoms and with the illness in general. A question enquiring about advice they would give to newly diagnosed sufferers was added to give an indication which strategies they regarded as the most helpful. Finally, two more questions asked the patients to sum up the experience of CFS and to assess their future. A copy of the questionnaire can be found in Appendix 1.

The interviews were structured in a similar way to the questionnaires, but using additional questions to explore specific issues of interest, such as the nature of fatigue, the history of psychiatric disorders and the choice of certain coping strategies.

3.4 Results

3.4.1 Analysis

Replies from both groups were divided into categories in order to provide quantitative information about the symp-

toms, attributions, extent of the illness and the types of coping strategies used. The categories were selected using information from the literature on CFS and other chronic illnesses. Following a preliminary analysis of the data, further categories were added to the list in order to cover responses which had not been anticipated.

Since the questions asked during the interviews were virtually identical to those in the questionnaire, it was decided to analyse the data from both samples in the same way, although results will be reported separately for the two groups. Where patients have been quoted, their group and identification number are noted in brackets.

3.4.2 Demographic information

The demographic information is presented in Table 1. The mean age of both groups was 41 years and just over three-quarters of the subjects were female. Of those who had completed their education, over a half had been to college or university and approximately a quarter had received some kind of professional training. Three people in both groups were either still at school or at college.

At the time of the interview, 6 of the subjects in Group 1 (35%) were working full-time and 7 (41%) had either retired or left their job. However, during the whole course of the illness, 15 people (88% of the group) had either left their job and retired, or changed their job or working hours because of illness. In Group 2, nearly 38% of the respondents were working full or part-time, and 45% had either retired or were unemployed. Nineteen reported that they had actually lost their jobs because of their illness. Sixty per cent had stopped or changed their job or working hours.

In terms of socio-economic status, the majority in both groups were classed as professional or manual skilled. Two

Table 1. Demographic information for Groups 1 and 2.

	Group 1	Group 2
Age (years)	No. (SD)	No. (SD)
Mean	41 (11.5)	41 (15.09)
Range	14-62	11-74
Gender	No. (%)	No. (%)
Male	4 (23.5)	11 (21)
Female	13 (76.5)	42 (79)
Education	No. (%)	No. (%)
Finished school	3 (20)*	9 (19.6)*
College/university	8 (53.3)*	26 (56.5)*
Professional training	4 (26.7)*	11 (23.9)*
Total who completed secondary education	15	46
Still at school	1	3
Still at college	2**	
Did not finish school or missing data		4
Mean age on completion of education	Years 22.9	Years 20.9

* Percentage of those who completed secondary education, including those presently retraining.

**Includes one person now retraining

Table 1 cont.

	Group 1	Group 2
Current occupation	No. (%)	No. (%)
Student	3 (17.6)	4 (7.5)
Housewife	0	5 (9.4)
Unemployed	1 (5.9)	12 (22.6)
Employed part-time	0	7 (13.2)
Employed full-time	6 (35.3)	13 (24.5)
Retired	7 (41.2)†	12 (22.6)
*Change of occupation**	No. (%)	No. (%)
Left job, retired or Reduced hours at work	11 (64.7)	25 (59.5)
Reduced/stopped study	2 (11.8)	6 (14.3)
Changed job only	4 (23.5)	1 (2.4)
None		10 (23.8)
No information/occupation		11
*Classification of occupation***		
Professional	6 (37.5)	15 (30.6)
Managerial	1 (6.2)	9 (18.4)
Manual skilled	5 (31.3)	16 (32.7)
Manual unskilled	3 (18.8)	3 (6.1)
Student	1 (6.2)	6 (2.2)
Not classified/at school or missing information	1	4

† all for medical reasons.
One also ticked 'housewife'.

* Group 2 n=42.

** Group 1 n=16, Group 2 n=49

Table 1 cont.

	Group 1	Group 2
Marital status	No. (%)	No. (%)
Single	6 (35.3)	17 (32.1)
Married	6 (35.3)	26 (49)
Cohabiting	1 (5.9)	6 (11.3)
Separated	0	1 (1.9)
Divorced	3 (17.6)	3 (5.7)
Change in marital status due to illness	0	1
Children	No. (%)	No. (%)
Yes	7 (41)	28 (53)
No	10 (59)	25 (47)
*Income per week**	No. (%)	No. (%)
<£70	4 (26.7)	15 (30)
£70-200	7 (46)	24 (48)
>£200	4 (26.7)	11 (22)
Information missing/no income	2	3
Housing	No. (%)	No. (%)
Owner/occupier	13 (71.7)	38 (71.7)
Private tenant	2 (11.8)	4 (7.5)
Council tenant	1 (5.9)	5 (9.4)
None of the above	1 (5.9)	6 (11.3)
Live alone**	No. (%)	No. (%)
Yes	6 (37.5)	9 (17)
No	10 (62.5)	44 (83)
Missing information	1	

* Group 1 n=15, Group 2 n=50.

** Group 1 n=16, Group 2 n=53.

of the three students in Group 1 had worked before their illness and were classified according to that work.

About one-third of the patients were single and around a half were either married or cohabiting. Only one person had changed marital status during the illness.

The income of the majority was comparatively low and often consisted of state benefits. However, most (71%) owned their own home.

3.4.3 Data on the nature of the illness

As shown in Table 2, nearly one-half of the patients in both groups developed CFS suddenly and many were able to link the onset with a specific viral illness, e.g. influenza or glandular fever. In Group 1, blood tests were used in 70% of patients both to exclude other conditions and to support the diagnosis of CFS(ME) (e.g. the VP1). In contrast, investigative tests were used less frequently in Group 2. The greater use of tests in the former may be partly due to the some of the local doctors' special interest in the illness.

Patients had been ill for a mean of 8.3 years (Group 1) and 5.2 years (Group 2). More specifically, 53% of the interviewees and 32% of the questionnaire sample had been ill for more than 5 years. In the majority, the illness followed a fluctuating course. Indeed, only 2 people reported that their illness had been stable.

3.4.4. Main symptoms

As shown in Table 3, the most frequently reported symptom was fatigue. In fact, all the subjects in Group 1 and 94% of subjects in Group 2 mentioned either fatigue or exhaustion as their main symptom. The three respondents who didn't, noted the effects of tiredness in later sections, so

Table 2. Details of illness-related variables.

	Group 1	Group 2
Onset	No. (%)	No. (%)
Sudden	3 (17.6)	13 (24.5)
Sudden following specific viral infection	6 (35.3)	13 (24.5)
Gradual	8 (47.1)	27 (51)
Diagnostic tests	No. (%)	No. (%)
Elisa IgM (CBV)	1 (5.9)	2 (3.8)
VP1	6 (35.3)	6 (11.5)
Blood tests to exclude other conditions	12 (70.6)	10 (19.2)
Brain scan	1 (5.9)	1 (2)
At least two of above	7 (47.1)	12 (23.1)
None	2 (11.8)	21 (40.4)
Duration of illness		
Mean years (SD)	8.26 (8.9)	5.2 (6.8)
Range	19 mths-40 yrs	7 mths-41 yrs
Progress of illness	No. (%)	No. (%)
Same	1 (5.9)	1 (2)
Fluctuating	10 (58.8)	21 (39.6)
Generally deteriorating	1 (5.9)	11 (20.7)
Generally improving	2 (11.8)	3 (5.7)
Fluctuating, now improving	3 (17.6)	17 (32.1)

they may simply have forgotten to include this symptom in their list.

Other symptoms, in order of prevalence, were muscle aches and memory impairment (53 and 51% respectively) and concentration problems (over 80% in both groups). A significant

Table 3. Main symptoms reported by Groups 1 and 2.

Symptom	Group 1 No (%)	Group 2 No (%)
Fatigue/exhaustion	17 (100)	50 (94)
Muscle pain, tenderness	9 (53)	27 (51)
Memory problems	9 (53)	16 (30)
Concentration problems	6 (35)	27 (51)
Weakness	6 (35)	18 (34)
Headaches	5 (29)	24 (45)
Malaise	5 (29)	9 (17)
Nausea	5 (29)	7 (13)
Pain	5 (29)	20 (38)
Visual disturbances	5 (29)	21 (40)
Walking difficulties	4 (24)	12 (23)
Dizziness, fainting	3 (18)	13 (25)
Moodiness	3 (18)	14 (26)
Sensitivity to temperature	3 (18)	11 (21)
Allergies	2 (12)	5 (9)
Depression	2 (12)	11 (21)
Sleep disorders	2 (12)	12 (23)
Panic attacks	2 (12)	4 (8)
Digestive disturbances	1 (6)	11 (21)
Neurological problems	1 (6)	11 (21)
Confusion	1 (6)	7 (13)
Sore throats	1 (6)	7 (13)
Swollen, tender glands	1 (6)	3 (6)
Slowing down	1 (6)	1 (2)
Palpitations	0	10 (19)
Speech disturbances	0	2 (4)
Thrush	0	1 (2)
Hysteria	0	1 (2)
Loss of libido	0	1 (2)

number of patients also listed pain, headaches, visual disturbances, dizziness, walking difficulties, nausea and a flu-like malaise. Up to a quarter of patients reported suffering from moodiness and/or depression.

The least common symptoms in both groups were swollen glands, speech disturbances, hysteria, thrush and slowness.

Five people in Group 2 reported muscle twitches as a main symptom and two admitted to "apathy". One also noted swallowing difficulties while one interviewee, an accomplished pianist, related how her illness prevented her from playing the piano. Although she had no problem sight-reading, she said that her brain wouldn't translate the information on the sheet to her fingers. She described this as a type of "musical dyslexia".

Several of the interviewees mentioned difficulties with sex, including loss of libido and being too tired. However, these were not regarded as main symptoms. Other problems which were reported included intolerance to alcohol and ear symptoms.

3.4.5 Attributions regarding the illness

a. Attributions about their own illness

As the first columns in Tables 4a and b show, the main factors which were considered responsible for the patient's own illness included viruses, a pressured and busy lifestyle, stress, and a reduced resistance to infection.

A recurring theme was that of working through a viral infection and taking insufficient time off to convalesce. For instance, one respondent attributed her illness to the following factors:

> "Catching what I thought was a cold and not giving in to it. Carrying on going to work. Trying to cope with a full-time job, housework and children and everything else. Perhaps putting a strain on me making me low and susceptible to any virus that comes along. Trying to do too much in effect" (2.6).

From the interviews, it became clear that the term 'stress' was often used to refer to "overdoing it", pressure at work and at home, as well as the presence of emotional problems. A number of patients saw a link between the 'stress' in their lives and their illness. For instance, one respondent noted that she'd had to deal with her own illness (glandular fever), getting married, moving house and "builders wrecking the house", all within one year. In addition, she worked 15 hours a day, 7 days a week until as she put it "I came to a grinding halt". In the year before the current illness, she suffered from colds every month. In her view:

> "The body could no longer cope. But you don't realise how ill you are until it's too late" (2.3).

Others led less busy lives, but the reaction was generally the same. Thus instead of resting at the start of what appeared to be a normal respiratory infection, they continued to focus on their emotional problems, using strategies such as exercise to deal with them.

When patients attributed their ill-health to other factors, these too were generally not judged as requiring them to convalesce. For instance, one person thought her symptoms were due to the change of life and that's why she did not take time off to rest.

It is important to note, however, that not everyone per-

Table 4a. Attributions regarding aetiology: Group 1

Causes	Own No. (%)	General No. (%)	Unlikely No. (%)
Infection	15 (88)	13 (77)	0
Immunological factors	1 (6)	4 (24)	0
Lowered resistance	7 (41)	6 (35)	0
Pressure	7 (41)	2 (12)	0
Stress	8 (47)	2 (12)	3 (18)
Fatigue	4 (24)	2 (12)	0
Genetics	4 (18)	1 (6)	1 (6)
Illness/surgery	4 (24)	0	0
Life Events	4 (24)	0	0
Drugs	1 (6)	1 (6)	2 (12)
Allergies	0	0	1 (6)
Candida	0	0	7 (42)
Pollution	1 (6)	3 (18)	2 (12)
Chemicals	1 (6)	0	0
Vaccination	1 (6)	0	0
Diet	0	0	0
Change of house	0	0	0
Change of job	1 (6)	0	0
Relationship problems	1 (6)	0	0
Change marital status	0	0	0
Personality factors	1 (6)	4 (24)	1 (6)
Psychological factors*	0	1 (6)	13 (77)
Accidents	0	0	0
Not sure	0	1 (6)	0
Combination	1 (6)	0	0
Others	4 (24)	2 (12)	3 (18)

*Includes depression/fear of activity and hypochondria

Table 4b. Attributions regarding aetiology: Group 2

Causes	Own No. (%)	General No. (%)	Unlikely No. (%)
Infection	37 (70)	37 (70)	0
Immunological factors	7 (13)	15 (28)	0
Lowered resistance	18 (34)	17 (32)	0
Pressure	30 (57)	12 (23)	0
Stress	21 (40)	16 (30)	8 (15)
Fatigue	12 (23)	9 (17)	0
Genetics	1 (2)	4 (8)	2 (4)
Illness/surgery	10 (19)	1 (2)	0
Life Events .	11 (21)	4 (8)	0
Drugs	5 (9)	7 (13)	2 (4)
Allergies	4 (8)	4 (8)	5 (9)
Candida	1 (2)	2 (4)	5 (9)
Pollution	1 (2)	7 (13)	2 (4)
Chemicals	1 (2)	4 (8)	0
Vaccination	2 (4)	1 (2)	0
Diet	3 (6)	6 (11)	4 (8)
Change of house	5 (9)	0	0
Change of job	2 (4)	0	0
Relationship problems	6 (11)	1 (2)	0
Change marital status	1 (2)	0	0
Personality factors	1 (2)	5 (9)	1 (2)
Psychological factors*	3 (6)	1 (2)	23 (43)
Accidents	1 (2)	0	0
Not sure	0	2 (4)	7 (13)
Combination	1 (2)	5 (9)	0
Others	11 (21)	6 (11)	6 (11)

*Includes depression/fear of activity and hypochondria.

ceived their busy lives in a negative way. For instance, one lady related that she had:

> "always worked hard and played hard ... my life was full, fast and wonderful, the happiest time I have ever had, and I was very strong, both mentally and physically" (2.34).

Nevertheless, like others, she attributed her illnesses to going beyond her limits as a result of which her body lost its ability to fight off infections.

Some of the factors which were reported less often as causes included diet, the use of antibiotics, thrush, personality factors, operations, lighting and pollution.

b. Beliefs regarding the illness in general

The types of attributions about the illness in general are shown in the second column of Tables 4a and b. As before, the majority of patients mentioned viral infections as a major cause.

While most of the subjects reiterated the view that infection and lowered resistance played an important role, far fewer mentioned stress or pressure this time and many more identified personality factors. Several of the interviewees observed that the illness appeared to be more common in very busy people who take on too much and go beyond their limits. One of them, who had counselled many patients observed that:

> "ME people are generally very active and very busy people - on the go, motivated people ... conscientious, perfectionist" (1.7).

A less common view was that CFS tended to affect mainly "sensitive people" (1.11).

Pollution was also regarded as more important this time. For instance, a number linked immune function with pesticides in food, the overuse of antibiotics, the Pill etc. One also associated environmental factors with a perceived increase in infectious agents:

> ... There seem to be more viruses around ... As a child, I didn't get all these strange viruses. You got flu and you got a cold, but not these strange things" (1.1).

Other factors which were mentioned by the interviewees as possible causes included additives and mercury fillings.

c. Factors regarded as unlikely causes

When asked about the unlikely causes of CFS, a large number in both groups rejected the view that the illness was purely psychological or 'all in the mind' (see third column, Tables 4a and b). Also mentioned in this regard were drugs, hypochondria, laziness and boredom, malingering, lack of motivation, phobias, "being a yuppie", amalgam dental fillings and wanting attention. One respondent wrote:

> "If I 'needed' an illness I'd certainly pick something else" (2.44).

It was possible during the interviews to ask about unlikely causes in more detail. For instance, while less than half of Group 2 believed that psychological factors like hyperventilation and clinical depression did not play a major role in CFS, 77% of the interviewees rejected these as main causes.

In response to the question about specific theories, most dismissed the idea that CFS was due to inactivity and/or depression. One interviewee said:

"Initially perhaps I did overrest and I got rather unfit. Then it was striking the balance and getting yourself fit enough to carry on and walk and do things like that and not ... relapsing. Doing a little bit but not too much ... I try to do something everyday to keep my system going ..." (1.3).

There was no evidence that patients in this study had become afraid of exercise to the extent that they avoided all activity. Although some exercised until they relapsed, they tried again, often changing the nature of the exercise e.g. from swimming to yoga. Moreover, they were not put off by symptoms. For instance, one person who tried yoga found that she felt rotten two days later. However, she persevered and now feels that it really helped her.

Those who had experienced clinical depression in the past noted how different this was from CFS. In their view, the former was associated with more general tiredness, more apathy, anhedonia and the loss of hope. One interviewee who had suffered from depression from the age of 16 said:

"...you do get depressed with this illness, but it lifts the minute the physical symptoms lift ... I know it's an entirely different feeling, the feeling of longterm depression. When you're depressed, when the world's at an end, you haven't any energy, you don't want to do this and you can't see any future. Throughout the 8 years of ME I've always been able to see a future. There's always been hope".

Q: Were you apathetic, like not wanting to get out of a chair, whereas in ME you want to, but you can't?"

A: "Absolutely" (1.3).

The differences between CFS and clinical depression were also outlined by another interviewee. She had experienced a period of depression earlier in her life and said that she remembered:

> "enormous apathy, you burst into tears without knowing why you're crying. It's like a sort of grey cloud that descends on you. You feel impossibly tired, you drag yourself about ..."

She also noted that the energy levels did not change so quickly when she was depressed, and that her mood did not fluctuate quite so much as it does now she has ME.

> "... The depression in ME is different. Although it's very real and very acute at times, it lifts, it passes and you have days when at least mentally you feel quite normal. That doesn't happen in depression. It really is a relentless thing. This is why it's so horrible to go through. There isn't any hope in the way that you have hope in between your spasms of depression in ME" (1.8).

Nevertheless, psycho-social factors were regarded as influential later on. For instance, a number identified factors such as 'stress' as something which exacerbated their condition, while support and care from others had the opposite effect.

3.4.6 The effects of CFS

Many patients reported that the illness had had a profound effect on their lives (see Table 5). All except three subjects mentioned that they'd had to reduce activities, particularly walking, travelling and driving and hobbies. Many

Table 5. Main effects of the illness

Effect	Group 1 No. (%)	Group 2 No. (%)
Activities	15 (88)	52 (98)
Job	14 (82)	25 (47)
Social life	13 (76)	25 (47)
Housework	9 (53)	17 (32)
Can't make plans	7 (41)	13 (25)
Change plans for future	2 (12)	2 (4)
Slowed down	2 (12)	11 (21)
School/higher education	2 (12)	16 (30)
Finances	1 (6)	8 (15)
Change in environment	1 (6)	0
Diet	0	8 (15)

patients had had to give up or change their jobs, and the women had been forced to limit the amount of shopping and housework. Some patients also had problems reading, talking and writing, and a minority had changed their diet to reduce nausea or 'allergies'. One respondent summed her life up as follows:

> "My whole life has been curtailed. I have to plan everything around the illness. I can't walk or run, or decorate, or play energetically with my children. All my strength seems to have gone (and I was quite strong physically.) Mentally I am much less 'intelligent', my long and short term memory have been affected as my ability to concentrate, study, learn and write. My judgement and reasoning are not as good (and I was very bright). My brain is permanently "out to lunch" (2.44).

The limitations, both physical and mental led one interviewee to sum up her situation as "a sheltered life" (1.12) while a respondent wrote that she felt that she was "merely existing, as opposed to living in a creative way" (2.50). Another, now recovering, described how the ME had led to a reduction of choice and control in the sense that she could not always shop, go out, drive or even have a bath when she wanted to or needed to.

The responses indicate that although patients were able to do less overall, the extent of their impairment varied quite considerably and they could identify good periods as well as bad ones. The unpredictability of the illness also meant that many had difficulties making plans. This was noted by a number of people in Group 2 but specific questioning of Group 1 revealed that it was a much more common experience than their responses had indicated.

One described the situation as follows:

> Physical activity has to be considered almost daily - and it is difficult to make plans ahead. At times I can do an almost normal amount - and it is then ... necessary to beware of 'overdoing it'. Some days it is only possible to do the barest essentials - and to abandon plans for e.g. gardening, decorating, walking, even shopping. At worst, there is no alternative to bed rest, for at least part of the day. This, of course, is very puzzling for one's family and friends, and often precludes long term projects" (2.62).

A number of patients noted that the fluctuating nature of the illness also restricted their activities outside the home because of the fear that others would be unsympathetic if they began to feel unwell and that they might not be able to find the help they need. One interviewee summed it up by

saying:

> "you are afraid of getting into a situation you can't cope with" (1.15).

One rather surprising effect described by a number of the patients was that they were no longer getting different viral infections. Instead, their reactions tended to be an exacerbation of ME symptoms:

> "If I'm with people who've got germs, I tend to in the days subsequent, feel very ill. I have all the symptoms of ME, I feel very tired, my glands tend to swell up but I don't actually get it. I dose myself up with vitamin C and it's the fighting off that makes one feel ill" (1.3).

3.4.6.1 The effect on relationships

As shown in Table 6, many patients reported that CFS put a great strain on their relationships. Indeed, over 80% from both groups noted problems in this regard.

Table 6. The effect of the illness on relationships.

Effect	Group 1 No. (%)	Group 2 No. (%)
Relationships strained	14 (82)	43 (81)
Lost contact with friends	7 (41)	17 (32)
Relationships improved	4 (24)	12 (23)
Marriage closer	1 (6)	6 (11)
Marriage strained	0	7 (13)
None	1 (6)	1 (2)

Many revealed that they had lost contact with friends and

about 12% admitted to feelings of isolation and loneliness. Relationships were also affected by difficulties in following and therefore having conversations. However, about a quarter considered that CFS had had a positive effect on their relationships. The following describe some of the most frequently reported experiences.

a. The lack of understanding

Many mentioned a lack of understanding and sympathy and most attributed this to their friends and family's limited knowledge of the illness.

Most reactions fell into three categories:
1. scepticism regarding the status of the illness, i.e. the suggestion that it did not exist in a any form, and that the person was therefore not sick,
2. belief that the illness had a psychological basis, e.g. that it was a type of nervous breakdown or just hypochondria, and consequently, that help was available if they wanted it or that they could pull themselves out if it,
3. acceptance that the patient was ill and that they required help.

Many patients commented on the difficulty they had experienced trying to persuade their family and friends that they were genuinely unwell and not malingering or exaggerating; for instance, that they really had something which can not be cured with a brisk walk or a holiday. For instance, one person wrote that:

> "some members of my family were sceptical about the illness and my degree of suffering. With friends I found myself having to justify what I was doing and explain the nature and type of illness which I was suffering from" (2.13).

Another noted:

> "I think that some friends regard me as a bit of a hypochondriac as I am always feeling ill ... It makes me angry inside at times to think that I have suffered and conquered so many stages of ill-health in my life and no-one believes me" (2.29).

Not surprisingly, a number of patients lost friends as a result of their illness, either because the contact was broken or because of conflicts. One person felt that people interpreted the fatigue personally, that is, they thought that the patient was tired of them. Others believed that friends avoided contact because they felt embarrassed, that they didn't know what to say.

An interviewee summed up the problem as follows:

> "The illness is so contrary to anything that anybody else has experienced before - they just don't understand" (1.7).

This was echoed by a respondent who identified a problem common to many whose disability is invisible:

> "It would have helped if there had been some viable symptom of the illness (apart from appearing very tired and lazy!) which other people could understand and relate to more easily" (2.13).

There was also disbelief among doctors. One nurse wrote that:

> "Even on the neurological ward the doctors, consultants did not believe in its existence and I was continually having to defend the disease" (2.17).

The fluctuations in the illness was identified as a complicating factor by a number of patients. As one respondent commented, when others see you looking better, they:

> "do not realize that each day is different, and you go back to being ill again" (2.9).

A number of respondents stated that they coped with the distress associated with relationships by avoiding certain people or pretending they were fine. For instance, one person wrote that she had stopped seeing a close friend because the latter couldn't accept her illness and was insensitive.

> "She kept on saying things that upset me and I felt ... that I'd been putting all the energy into maintaining the friendship ... I felt very hurt and ... the only option was to 'get out'" (2.39).

To stop others worrying, or to avoid difficult questions, some lied about their condition when they were feeling unwell. Others sometimes avoided the topic of CFS altogether:

> "A lot of people are either patronising or totally non-understanding. Because there is such doubt surrounding ME I feel disinclined to keep on explaining why I can not commit myself or be sure that I'll be O.K. on a certain day" (2.47).

b. <u>The effect on children and family life</u>

The strain of the illness also appears to have had a major effect on people's relationship with their partners and children.

> "It's been devastating for my husband and son. Over the years it's slowly destroyed our lives

> until the point came where I was so ill we thought I would die. My husband became mentally and physically worn out washing me, feeding me and looking after my son and eventually had 3 months off work, which still left him with everything else as we had no family to help and our two requests for home help were refused. It broke my heart to see my son's unhappiness. He used to cry a lot and became very introvert. Although it wasn't my fault, I still felt very guilty. I couldn't be a wife or a mother or hold a proper conversation, as I couldn't remember the day, time or people's names" (2.21).

Again the problem of having an invisible illness came to the fore:

> "The children ... cannot relate to mum being ill when I don't look particularly ill. If I had a broken arm or something 'visible' I think try could have coped better. Now they are older they understand more ... I feel that I have failed my children in not being able to do things with them, and for them" (2.63).

On a more positive note, one mother observed that her illness meant that she had more time to sit and talk with her children and another felt that it was good for her children to have more responsibility. Many also referred to having become closer to their families, and a number described how their relatives and friends had been prepared to learn about the illness and support the patient.

Finally, the illness prevented a few patients from actually building up relationships. As one admitted:

> "I never had a steady boyfriend, never had steady

relationships with people of the opposite sex. I was too ill and too tired to give people the time they need" (1.6).

3.4.6.2 The effect of CFS on the person

The illness seemed to affect people in different ways (see Table 7). The majority noted feelings and emotions such as frustration, anger and unhappiness. Many also thought that it had changed their personality, their attitudes and their sense of being in control.

Table 7. Effect of the illness on the person.

Effect	Group 1 No. (%)	Group 2 No. (%)
Emotional distress	12 (71)	39 (74)
Changes in personality (esp. self-confidence)	9 (53)	26 (49)
Changes in attitude	9 (53)	20 (38)
Learned from it	4 (24)	8 (15)
Accept situation	2 (12)	7 (13)
Limited life	2 (12)	2 (4)
More positive outlook	2 (12)	9 (17)
Isolation/loneliness	2 (12)	10 (19)
Lost control over life	2 (12)	3 (6)
Change in appearance	1 (6)	3 (6)
Gained respect for body	1 (6)	1 (2)
Guilt	0	5 (9)
Why me?	0	1 (2)

a. Emotional distress

Over 70% of the patients mentioned experiencing some form of emotional distress as a result of their illness. Many were frustrated by the limitations in their lives, including the

inability to do trivial things like washing their hair, lifting the washing out of the washing machine etc. Others were upset by the lack of understanding about CFS (see above).

Some linked their emotional state to the changes in their symptoms. For instance, one person who described herself as generally "cheerful and carefree" became "irritable and depressed" when her symptoms worsened. Others also related how exacerbations upset and depressed them, although some managed to cope better than others. As one respondent put it:

> "I am grateful for a good day and look forward to the next day if it is a bad day. If I have had a few good days and feel I am going into remission I do get upset if I am ill again, but I have learnt to live with it" (2.40).

However, another summarised the experience as follows:

> I never realised that one could feel so dreadful and still stay alive" (1.5).

b. Effects on personality

About half the patients noted that their personality had changed. Some felt that the illness had made them more patient, understanding, tolerant, serious and mature. Others had become more introspective, shy and reserved. One optimist had changed into a pessimist, finding that events which used to be challenges were now more likely to be evaluated as problems. Other negative effects which were mentioned included becoming more short-tempered and self-centred, being less out-going and spontaneous, and loss of confidence, particularly about being able to complete tasks. The limitations imposed by the illness also affected people's perception about themselves in another way. As one

interviewee said:

> "It's bad for your self-esteem because you suddenly find yourself unable to cope with situations you didn't even know were situations that required coping with" (1.5).

c) Changes in attitudes and other aspects of self

A number of interviewees mentioned how the experience of illness had led them to change their attitudes to life, to other people, to their diet, the environment and to themselves. People described how they had become more tolerant and patient, less materialistic, less critical of others, and that they obtained more pleasure from simple things. Some noted having become more aware of spiritual matters and the importance of good health. One person also mentioned that she'd become more assertive in her relationships, and a few had changed their political affiliation. One interviewee said:

> "It's altered my values and I appreciate different qualities in people and put more importance on qualities in myself, like having time for people, having the strength to carry on. I have a high opinion of anyone who's had to cope with a setback in his life" (1.13).

The fact that more people in this group reported changes in their attitudes may have been due to being asked to elaborate on this point.

Some people noted the lack of control, particularly the inability to fight the illness, ignore it or work through it. As one person put it:

> "I used to feel I had more control over myself, body and the world. I now feel things are more

> complicated, beyond my control to a greater extent - that I have to meander a path through what I want to do and what I can do - the gap is far greater than it used to be" (2.55).

A few felt that the illness had also led to a change in their appearance.

> "I also look much older - the eyes have been a sure sign of my state of health" (2.26).

Finally, several patients said that they had learned from the experience, for instance, that life is more than work, education and achieving. One summed it up as follow:

> "I have learned not to become flustered, to take life as it comes, and enjoy whatever small pleasures I may find. Learned to 'stop and smell the roses' in fact" (2.16).

3.4.7 Coping

People were asked two questions about coping. The first focused on the ways they dealt with symptoms on a day-to-day basis; the second asked about the ways they coped with the illness as a whole.

a. Coping with symptoms

As shown in Tables 8a and b, rest was the main coping strategy used to deal with the symptoms. Indeed, it was mentioned by 88% of the patients in Group 1 and 74% of those in Group 2. This was often combined with the pacing of activities and relaxation (for instance, with yoga and meditation). A significant number also tried drugs, particularly antidepressants. The following section describes the strategies in more detail.

Table 8a. Main coping strategies employed by Group 1.

Ways of coping	With symptoms No. (%)	With illness No. (%)
Activities*	4 (24)	2 (12)
Accept illness	0	1 (6)
Allergy treatment	2 (12)	0
Alternative medicine	15 (88)	0
Avoid stress	2 (12)	0
Keep problems to oneself	0	0
Diet	12 (71)	0
Distancing from illness	0	0
Diversion of attention	3 (18)	1 (6)
Drugs	7 (41)	0
Find good doctor	1 (6)	0
Hobbies	3 (18)	0
Holidays, outings	1 (6)	0
Humour	1 (6)	1 (6)
Seek information	0	1 (6)
Pacing, live within limits	10 (59)	5 (29)
Plan activities,	2 (12)	1 (6)
Positive outlook	2 (12)	8 (47)
Religion, faith	1 (6)	1 (6)
Relaxation (e.g. yoga)	6 (35)	3 (18)
Rest	15 (88)	4 (24)
Sleep more	2 (12)	1 (6)
Seek social/emotional support	6 (35)	4 (24)
Vitamin, mineral supplements	7 (41)	0

* e.g. includes gentle exercise (such as walking), gardening, voluntary work, listening to music.

Table 8b. Main coping strategies employed by Group 2.

Ways of coping	With symptoms No. (%)	With illness No. (%)
Activities*	18 (34)	18+ (34)
		1- (2)
Accept illness	0	7 (13)
Allergy treatment	4 (8)	1 (2)
Alternative medicine	16 (30)	3 (6)
Avoid stress	8 (15)	3 (6)
Diet	24 (45)	4 (8)
Distancing from illness	1 (2)	0
Diversion of attention	5 (9)	3 (6)
Drugs	12 (23)	5 (9)
Find good doctor	2 (4)	4 (8)
Hobbies	3 (6)	4 (8)
Holidays, outings	1 (2)	6 (15)
Humour	4 (8)	6 (11)
Keep problems to oneself	1 (2)	1 (2)
Pacing, live within limits	26 (49)	11 (21)
Plan activities	7 (13)	3 (6)
Positive outlook	9 (17)	15 (28)
Religion, faith	3 (6)	9 (17)
Relaxation	16 (30)	10 (19)
Rest	39 (74)	10 (19)
Focus on self	1 (2)	1 (2)
Seek information	6 (11)	3 (6)
Sleep more	6 (11)	4 (8)
Social life	2 (4)	4 (8)
Seek social/emotional support	7 (13)	28 (53)
Vitamin, mineral supplements	17 (32)	0

+ added activities - reduced activities

Rest and conserving energy

Resting for certain periods was clearly the most common way patients tried to deal with their symptoms. However, no one mentioned staying in bed all day or for long periods of time. Nor did many people report an increase in time spent asleep.

Resting was often combined with a process referred to as pacing. Eleven (65%) of the interviewees and 32 (60%) of the respondents noted that they used pacing either to control symptoms and or the illness. It involves working out how much one can do without triggering a relapse. Most patients had adopted this approach through trial and error and from experience. However, it often amounted to making an educated guess. Others referred to listening to their body in order to estimate what they could do.

Pacing was not easy. For instance, one interviewee said:

> "When I do start picking up, I have to resist the temptation to start running around and doing things. If I do have energy, I start using it really quickly ... too much. I always end up feeling much worse a few days later" (1.2).

However, those who used this strategy generally found that they were able to do more as a result. Indeed, one or two reported that it allowed them to live a reasonably normal life.

Aside from limiting activities, a few mentioned that they could reduce exacerbations by controlling other known trigger factors, e.g. certain foods. Someone who suffered from food sensitivities commented on the effects as follows:

> "Quite importantly, every outing has to be planned with military precision if we want to eat out.

> Due to food allergy there can be no spontaneity or surprise meals out" (2.56).

Complementary medicine

A very large percentage of Group 1 reported that they had tried alternative medicine and vitamin and mineral supplements. Many had also experimented with their diet. The use of supplements was also reported by patients in Group 2 but other forms of self-help, such as dietary changes and alternative therapies, were less common. The interest in alternative medicine did not stop patients trying more orthodox treatments as well. In fact, far more people in Group 1 used drugs such as anti-depressants.

Practical help

Some of the coping strategies were practical. One person moved to a house which would be easier to keep clean and another asked her employer for a transfer so she did not have to travel so far to get to work. People also used aids such as crutches to enable them to walk, and made lists to compensate for their poor memory. Others planned their activities. As one patient explained:

> "I bring downstairs everything I'll need during the day, so that I don't have to go upstairs again until bedtime (I'm fortunate to have a downstairs toilet). Throwing the washing downstairs ahead of me - this is a bit untidy I know, but I found it easier to collect it at the bottom than to carry it down. Keeping the things I am going to use most of the day on a level with my arms, to avoid bending or stretching up too often. This tends to take up room on the worktops, but again, is worth the saving on effort" (2.16).

Emotion-focused strategies

Seeking emotional support, either from professional counsel-

lors or from other patients was mentioned by a third of patients in Group 1 and 13% of those in Group 2. Others related how they coped by diverting attention away from thoughts about their illness and by keeping themselves occupied. One person kept a diary, and had embarked on an Open University course primarily to distract her attention. Trying to keep positive and humour were mentioned by only a few.

Exercise

Some patients made a determined effort to keep active, for instance, by taking short walks, playing the piano, doing woodwork, painting and gardening. One interviewee discovered weight-training when recovering; others limited themselves to gentle physiotherapy and massage. One patient wrote:

> "I do try to keep fairly active, (that doesn't mean running around the block!) and tend to set myself small targets to reach. It is important to feel that one is achieving something albeit small" (2.30).

Less common coping strategies included avoiding alcohol, taking warm baths and Christian healing, all of which were regarded as useful. One interviewee tried a graded exercise regime but found it unhelpful.

b. Coping with the illness in general

In response to the question what people did to stop being overwhelmed by the illness in general, there was a slight change of emphasis. In contrast to the largely practical approach towards symptoms, e.g. resting, dietary changes and drugs, people used more emotion-focused strategies to deal with the illness as a whole. These included adopting a positive outlook, for instance, telling oneself that they would recover and concentrating on what they could do,

rather than what they couldn't. In addition, many sought and appreciated support from family and friends. As one person revealed:

> "Above all, I think positive thinking and belief in myself has helped me most. Also talking through my fears and problems either with a friend or other ME sufferers and letting go of emotions and negative feelings and having a good cry now and then or expressing anger if I feel like it ..." (2.5).

Others valued the help from professionals such as general practitioners and counsellors. A few also mentioned the support from fellow patients.

In addition to the above, some also mentioned living within one's limits and accepting the fluctuating nature of the disorder as a way of dealing with the illness in general. Again, there was a tendency to use a number of different strategies. One person summed up her approach as follows:

> "Gentle acceptance, relaxation and keeping within my limitations, which of course become wider as time goes on, these are the things which help me deal with the illness at this time. Coupled with a sense of humour! When you drop glasses on the kitchen floor, when you were sure they were going on to the draining board, or realise that you can't see the ball the men are kicking around the pitch while you are watching the World Cup Series, if you can see the funny side, it eases the strain" (2.16).

However, some admitted that they were occasionally over-whelmed. One noted that at such times:

> "Support from my husband and friends was invaluable. When the doctors tell you there's nothing they can do to help, you feel very lonely" (2.3).

Finally, a number of patients were clearly helped by having a religious faith while others compared their situation with those who were worse off than themselves in order to keep a sense of proportion and feel better about their lives. As one person told herself: "if other people can survive this (and worse) then so can I" (2.63).

3.4.8 Advice to others

As Table 9 shows, the advice which most patients would give to new sufferers is to get sufficient rest. However, no one recommended total bedrest except for a short period in the early stages. During the chronic phase, the general advice was to 'listen to your body' in order to assess how much rest was needed, and to remain as active as possible within one's own limits. Some also referred the value of balancing rest with energy output, while a few recommended gentle exercise as part of the approach.

A significant number mentioned the need to remain positive and about one-third recommended patient groups, both for information on the illness and as a source of support. As one respondent explained:

> "They will understand how you are feeling and sympathise, offer advice, and make you realise that you are not alone and that there is a future for you. If ever there was a time when a 'trouble shared is a trouble halved' becomes true, it is for the ME sufferer" (2.16).

One of the interviewees felt that coming to terms with the illness was perhaps the most difficult part of recovery. He

Table 9. Advice which would be offered to a new patient.

Advice	Group 1 No. (%)	Group 2 No. (%)
Rest	14 (82)	32 (60)
Be positive	7 (41)	24 (45)
Join group	7 (41)	16 (30)
Pace yourself	5 (29)	16 (30)
Accept the illness/yourself	5 (29)	12 (23)
Alternative medicine	5 (29)	9 (17)
Seek emotional support	4 (24)	14 (26)
Seek information	4 (24)	12 (23)
Listen to body	3 (18)	9 (17)
Change diet	2 (12)	9 (17)
Find a good G.P.	2 (12)	8 (15)
Keep sense of humour	1 (6)	3 (6)
Take exercise	1 (6)	3 (6)
Enjoy activities	0	8 (15)
Keep diary	0	7 (13)
Accept help, benefits	0	6 (11)
Change lifestyle	0	6 (11)
Sleep	0	3 (6)
Have a hobby	0	2 (4)
Take vitamin supplements	0	2 (4)
Drugs	0	1 (2)

also said that newly diagnosed sufferers should be reminded that life has a lot to offer even if you have ME. Others concurred with this.

Although many had tried alternative medicine, particularly in Group 1, surprisingly few would recommend it to new sufferers. When it was, the approach was generally guarded:

"There is no miracle cure but don't be dissuaded

> by anyone from trying alternative therapies. They could ease your suffering but be sure you can cope financially and chose a qualified practitioner" (2.21).

There was also advice on dealing with sceptics. Some thought that patients should ignore, and not worry about people who don't understand. One wrote:

> "Don't let people (including doctors) make you feel guilty for being ill. You are sick, not mad" (2.44).

Other advice included: finding a knowledgeable GP, avoiding stress, being patient, remaining aware of the spiritual side of life, getting fresh air, avoiding alcohol and anaesthetics, keeping a lively mind and a sense of humour. Several people also advocated certain changes in lifestyle, such as eating sensibly and learning to relax. In contrast, almost no one recommended extra sleep or taking particular drugs.

3.4.9 What having CFS meant to the patients

This question asked respondents to sum up the experience of CFS in their lives. Not surprisingly, the answers sometimes overlapped with those given in the section dealing with the effects in more general terms.

The replies suggest that the illness had caused a great deal of emotional distress (see Table 10). For example, many mentioned feeling despair, pain, sadness and utter frustration. It had been a "trauma" (1.14), a "disaster" (1.7), "hell on earth" (2.3) and something that "totally destroyed my life" (1.15). Most notably, it was seen as a lonely fight, a battle and struggle against a lack of understanding. One summed up how she felt as follows:

"It has meant that two years of my life have been taken away from me ... feeling a victim ... feeling 'persecuted' and imprisoned in a bedroom for a crime I didn't commit! It means misunderstanding - a continual battle - struggling against doctors, psychologists, specialists, dentists, friends etc. to be taken seriously. It means loneliness and isolation. It means depression and sadness. It means boredom" (2.39).

Table 10. The meaning of illness

Experience	Group 1 No. (%)	Group 2 No. (%)
Emotional distress (eg frustration, anger)	12 (71)	30 (60)
Change in lifestyle	5 (29)	14 (26)
Change of view of person, priorities	4 (24)	10 (19)
Learning experience	3 (18)	8 (15)
Change in relationships	2 (12)	12 (23)
Change of job/career	2 (12)	7 (13)
Change of personality	2 (12)	6 (11)
Slowing down	2 (12)	5 (9)
Loss of control over life	1 (6)	1 (2)
Change in attitudes	1 (6)	9 (17)
Change of housing	1 (6)	1 (2)
Loneliness	0	7 (13)

To another, CFS meant:

"A devastated life ... I know this ailment is not fatal and therefore we should thank our lucky stars but the disruption it brings to a normal life is unbelievable" (2.38).

The enforced change in lifestyle, and in particular, the amount of time spent resting was generally seen in negative terms. One said:

> "I begrudge the time that's been wasted over the last 7 years because I'm not going to get them back again" (1.8).

People also noted a sense of loss in relation to themselves as people. One described this as "almost like a bereavement" (2.44).

As mentioned above, the illness also affected relationships and work. Even though some returned to their jobs after a time, the illness often meant that they had moved down on the career and promotion ladder. Ambitions had to be given up. One, who had to "jettison" her ambitions, described the illness as follows:

> "It's been a kind of crossroads in a way ... it pushes you in a direction - you don't make the decision - it makes it for you" (2.8).

Someone who had started to improve wrote about the continued care which had to be taken:

> "I dislike the feeling of being a bit unwell or tired now and again ... and of having to be conscious of this instead of just doing what I want to do. It's a nuisance, a bore or irritation" (2.35).

However, some saw positive aspects, the illness having changed their priorities and outlook on life. Another looked forward saying:

> "... I do feel very fortunate to have had the

> experience. The rest of my life is going to be more fulfilling, rewarding and worthwhile than if this hadn't happened" (2.3).

Several referred to suffering caused by the lack of understanding. One summed up her experience as follows:

> "I am labelled neurotic and I could write a book about the soul destroying things that doctors have said to me... Deep inside, I have feelings of anger and resentment bottled up about this. Occasionally it bubbles up to the surface if anyone dares to suggest 'it's all in the mind.'" (2.29).

Another person summed up the confrontation with disbelief as follows:

> "It has hurt to be called a malingerer" (2.41).

Finally, one respondent noted that ME had led to "a great cynicism towards the medical profession in general, and a questioning of our drug-orientated medical treatments". However, it had also resulted in "a reawakening of my Christian faith, and a desire to grow spiritually" (2.1).

3.4.10 Views of the future

While the assessment of the experience was overwhelmingly negative, 15 (88.2%) of interviewées saw their future in generally positive terms. However, there was less optimism in Group 2 where just 31 (58.5%) felt that their future would be better. Nevertheless, the majority were cautiously optimistic, hopeful that things would get better and realistically, predicting a slow recovery and having more control. For instance, one person wrote that she intended to live life to the full, but qualified this by adding that:

> "I shall adapt it to my abilities, but I intend to enjoy everything I am able to ... I am determined that although I must live with ME it will be kept in its place!!!" (2.44).

Another felt more ambivalent:

> "Well, it can't be any worse (or can it)" (2.7).

Others replied that they took one day at a time and preferred not to think too far ahead.

> "If I think of the future, I become depressed, so the most sensible thing to me is to take things day by day" (2.22).

There was little sign of a general hopelessness; only a tiny minority in both groups responded in clearly negative terms and expressed fear of what might happen to them. As one admitted:

> "I ... feel despair that things may never change. The possibility of being really ill (confined to bed, unable to do anything) haunts me" (2.63).

Finally, one expressed the hope that in the future:

> "doctors - especially GPs - would learn to respect ME as a "proper" illness, so that ME sufferers of the future, do not have to fight to convince her/him, that they are ill - I find this is quite inhuman" (2.34).

3.5 Discussion

The main findings from this exploratory study suggest that CFS caused a great deal of emotional distress and that many patients lacked support from their friends, families and physicians. The results also indicate that the illness caused a wide range of symptoms in addition to fatigue and that patients used a combination of coping strategies to deal with the physical and the emotional aspects of their illness.

Before discussing the data in more detail, it should be pointed out that neither Group 1 or Group 2 may have been representative of the patient population as a whole. Group 2 consisted of members from a national self-help association and although an attempt was made to obtain a more representative sample through general practitioners, all the interviewees turned out to be members of that organisation's local branch. Since that particular group was very active, it is possible that their views may have coloured those of the participants in this research.

Compared with other patients, both Group 1 and 2 contained a larger proportion of women, more professionals and more chronic cases than the community sample described by Lloyd et al (1990). In fact, the subjects in the present study were probably more like the clinic attenders studied by Dowsett et al (1990) and Ray et al (1992b).

3.5.1 Symptoms

As expected, fatigue and exhaustion were reported by almost everyone. Also common were muscle pain and tenderness, memory and concentration problems, visual disturbances (e.g. photophobia) and weakness. The pattern of symptoms provided by patients was consistent with the London criteria for CFS(ME) as well as the descriptions of CFS(ME) by specia-

lists such as Ramsay (1988) and Dowsett et al (1990). They were also compatible with their doctors' diagnosis of 'ME' or 'post-viral fatigue syndrome'.

Interestingly, neither Group 1 nor Group 2 reported the high levels of depression, allergies and sore throat/pharyngitis which have been documented in some of the other studies on CFS (e.g. Hilgers and Frank 1992, Tirelli et al 1993, Straus et al 1985). More specifically, while most of the patients admitted to periods of unhappiness and frustration (see section on the experience of CFS), the interviewees did not feel they were clinically depressed. Indeed, a number of the patients revealed that they had suffered from serious depression in the past, and were therefore able to compare and contrast this with their present condition.

The low prevalence of allergies and sore throats was unexpected, given descriptions in the literature and the inclusion of pharyngitis in the CDC case definition of CFS (Bell 1991, Holmes et al 1988, Straus et al 1985). It is possible that these symptoms were present but not regarded as troublesome or that the symptoms had been treated and were no longer a problem. Certainly, many of the interviewees had made changes to their diet specifically to reduce allergy-linked reactions. As far as sore throats are concerned, it has been suggested that these are less common and severe among chronic cases (Jessop 1990). This could explain the low incidence among the subjects in this study; 88% of Group 1 and 81% of patients in Group 2 had been ill for at least two years and further inspection of the data revealed that sore throats were less common in those who had been ill longer.

The presence of symptoms like photophobia, which can not be explained solely in terms of fatigue, demonstrate that the illness is more complex than is sometimes implied in the medical press (e.g. Thomas 1993).

3.5.2 Beliefs about causation

In this study, only three subjects in Group 2 (6%) and two interviewees (12%) believed that infection was the sole source of their ill-health. The majority attributed their illness to a combination of external and internal factors, for instance, by referring to an increased susceptibility to infection due to lifestyle or 'stress'.

The tendency to attribute the illness to external and internal sources has also been reported by other researchers. For instance, Ray et al (1992b) noted that 43% of their patients believed that their condition was due to physical and psychological factors. Likewise, Yeomans and Conway (1991) found that 33% of their subjects felt the main cause to be an infection, although 67% accepted that psychological factors might have played a contributory role.

The results of this study are also consistent with those of Ware and Kleinman (1992). They reported that while 8% of patients attributed their illness to "stress only", a large proportion cited stress either as the single probable cause or as a contributory factor. As in the present study, stress was often invoked as the instigating factor in a chain reaction leading to hypothesized immune dysfunction and subsequent viral infection - a kind of "biopsychosocial" aetiological model. Furthermore, their subjects also referred to psychosocial factors such as overwork, to having led "lives of intense activity and involvement before their illness began" and to having been "always on the go".

The identification of psychosocial influences as contributory factors was also reported by Cathebras et al (1992) who noted that "most fatigue patients willingly acknowledged psycho-social causes for their symptoms". Moreover, Wood et al (1991) stated that 62% of their patients believed psychological factors played a part in their illness. This

contrasts with Wessely and Powell (1989), who found that only 3 out of 47 subjects (6%) attributed their condition to psychological factors.

The fact that many of the people in this study accepted that their own actions (e.g. overdoing it) may have predisposed them to ill-health does not support the view that most patients with CFS adhere solely to external attributions in order to avoid blame and responsibility (Abbey 1993).

When asked to speculate about the likely causes of CFS in general, the vast majority of patients continued to implicate infection. In contrast, fewer regarded pressure and a busy lifestyle as an important cause, and in Group 2, only two respondents mentioned stress. On the other hand, pollution and immunological factors (e.g. immune dysfunction) were considered to be more influential as general causes and amongst the interviewees, so were personality factors such as perfectionism.

It is unclear why patients felt that psychological factors were less important as a cause of illness in general, but the results do indicate a willingness to accept a multifactorial aetiology. One possible explanation for the discrepancy is that when considering general causes, the patients may have been influenced by the scientific literature. For example, the reviews of the research in the ME Association's newsletter have tended to emphasise studies implicating viral and immunological factors and have generally been highly critical of theories focusing on a psychological causes. This could be one reason why the subjects rejected explanations implicating a lack of activity, helplessness and hyperventilation. This does not mean that the scientific literature was accepted at face value; some of the interviewees also mentioned that the persisting infection theory seemed to fit their experience of the illness better than the psychiatric theories.

The patient organisation's critical stance towards alternative medicine combined with their own experiences may also explain the dismissal of candida as a likely cause. At the time of the study, many health magazines had discussed the problem of candida and a significant number of subjects had tried an anti-candida diet. However, while not rejecting the possibility that a yeast infection might play a role in perpetuating or exacerbating symptoms, most felt that it was unlikely cause of the illness itself.

3.5.3 The effects of CFS

The patients' accounts revealed that the illness had affected almost every aspect of their lives and that it had been a major source of disruption and distress. For instance, many had had to reduce the number of hours they worked, or take sick leave. This often meant a change in income and indeed, just under a third had become dependent entirely or almost entirely on benefits. Women noted problems doing housework and looking after their children and a significant proportion had had to give up their social life.

The illness had also led to marked changes in their personality and attitudes. For instance, many felt that being ill had made them more introspective and reserved, but also more patient and less critical of others. Some also mentioned that it had reduced their confidence about what they were able to do and achieve. Moreover, a significant proportion noted a change in values and priorities. For example, some reported having become more spiritual, less materialistic and obtaining more pleasure from simple things. Although not all the effects on self-concept and values were positive, most of the changes were consistent with those described by Wright (1960) as requirements for successful adjustment.

In terms of relationships, over 80% mentioned that the illness had led to strained communications and conflicts.

Some of these appear to have emanated from people's doubts about the aetiology of the illness and the severity of the symptoms. As a result, a significant number of patients stated that they had lost friends and that they felt misunderstood and unsupported.

One factor which may have influenced both sufferers and significant others is the fluctuating nature of the disease. Some patients referred to experiencing 'good' days and 'bad' days; others mentioned not being able to plan their lives and having periods when they did not know how they would feel from one day to the next. This led to a reduced sense of control and immense frustration.

The changeability of the symptoms may also have contributed to some of the conflicts between the patients and their families and friends. For example, the fact that a person is physically and mentally capable of going shopping one day, but not at the same time the following week is hard to comprehend, especially when there are no visible signs of impairment (cf. Viemero 1991). As a result, many outsiders may have questioned whether the patient was genuinely ill.

A further problem which might have contributed to the lack of understanding and sympathy towards patients is the actual nature of the symptoms. Like those of early multiple sclerosis, they may be perceived as nothing more than "exaggerated versions of conditions experienced by many people in the rough and tumble of everyday life" (Robinson 1988). Since most people regard complaints like fatigue, dizziness and headaches not only as transient but also as trivial and eminently manageable, it is not surprising that the general population's perception of the disability associated with CFS may differ from that of the patients.

People's doubts about the severity of CFS may have been reinforced by the media's description of the disorder as

'Yuppie flu', as well as some commentators' portrayal of patients as overworked people who wish to escape their busy lives (cf. Ware and Kleinman 1992).

The findings of this study suggest that where the lay concept of the illness was inaccurate, friends and relatives often questioned the effects of the disorder and accused some patients of exaggerating. Others simply did not wish to accept a change in their relationships, for example, fearing a loss of help and support from the person who was ill as a result of which they refused to legitimise the patient's complaints (cf. Wiener 1975).

While it is possible that criticism might encourage some patients to be more active, most of subjects in this study reported that it simply led to tension, conflict and rejection (cf. Faucett and Levine 1990, Ware 1992). Nevertheless, about a quarter of the subjects indicated that certain relationships had actually improved. In these cases, the other people involved had clearly accepted the patients' assessments of their condition as being valid and they were helpful and supportive.

The illness has also led to distressing experiences with doctors. Some problems stemmed from uncertainties about the aetiology of the illness while others were related to the doctor's emphasis on the influence of psychiatric disorders and their refusal to acknowledge the extent of the disability. Similar difficulties in the doctor-patient relationship were also documented by Denz-Penhey and Murdoch (1993) who concluded that both parties "were operating on different levels of knowledge". In their view, many doctors' had an "inadequate and impoverished view" of the illness. This 'abnormal illness perception' had led to adherence to models which denied the ill person's experience and was unhelpful in assisting either the doctor-patient relationship or the healing process. Similar difficulties

in connection with medical advisors have also reported in other studies on CFS (Woodward et al 1995) and in accounts on people suffering from MS (Stewart and Sullivan 1982).

One effect of the illness which is more difficult to explain is the change in the experience of other infections, particularly the inability to sustain a raised temperature. This phenomenon was mentioned by several interviewees and has also been noted by others (e.g. Shepherd 1989), but why it occurs remains unclear.

The responses to the question asking patients to sum up the experience of CFS underlined both the disabling nature of the condition and the emotional distress associated with it. Some referred to the impact of the symptoms, the inability to lead a normal life and the problem of obtaining practical help and treatment. Others focused on the lack of understanding and the immense difficulties to obtain recognition of their disability (see also Denz-Penhey and Murdoch 1993). The combination of factors, physical, psychological and social, meant that many regarded the illness not only in terms of loss and limitations, but also as a traumatic and devastating experience, an assault (cf. Turk 1979), a battle, or struggle. Some felt it was a waste of time, that they were merely existing rather than living. Nevertheless, some also identified a positive side, for instance, that they had learned valuable lessons from it.

In contrast to the largely negative views of the experience, the majority saw their future as being better. The apparent optimism was not mere wishful thinking; nearly 40% in both groups had already noted signs of improvement. They may also have been heartened by the steadily growing increase in research and awareness of the disease among doctors, and by the more sympathetic images of the disabled in the media. Nevertheless, a few did admit to fears about the future, and some preferred to take each day at a time.

In general, the finding that many were fairly optimistic and had a positive attitude towards their disability conflicts with the portrayal of patients as hopeless, helpless and depressed (Butler et al 1991, Sharpe 1994).

3.5.4 Coping with symptoms

The principal coping strategy used by people with CFS in this study appeared to be that of rest. However, none of the patients advocated resting all day or 'total bed rest'. Instead, people increased the amount of rest and this was often related to the level of activity.

A number of patients used a strategy referred to as pacing. This is also used by other patient groups, e.g. those suffering from rheumatoid arthritis (Wiener 1975) and post-polio syndrome (Bruno 1994b). It involves the identification of activities one is able to do, how often, and under what circumstances. Thus it is essentially a matter of balancing disability on the one hand and available resources on the other. Pinder (1988) observed that when "making such calculations, various options are negotiated - or 'traded off' - in search of the most viable outcome at any point in time".

The use of this strategy contrasts with reports of patients with CFS who consistently avoid all activity or who occasionally attempt activities at premorbid levels because of a personal desire to maintain high standards (Butler et al 1991, Surawy et al 1995, Wessely et al 1991).

Social and emotional support from relatives and friends was another source of help. So was the additional advice and understanding provided by fellow sufferers, counsellors and therapists (cf Locker 1983, Robinson 1988). In terms of medical help, many interviewees found their GP to be understanding and supportive, but they were offered little other

than symptomatic treatment and lifestyle advice. Nevertheless, those who were prescribed drugs like antidepressants did try them. Two were also accepted on a rehabilitation programme. One interviewee tried graded exercise, which she found unhelpful; another was taught to do gentle exercise within the limits of his capabilities and found this beneficial.

Given the shortage of orthodox medical treatment, many ended up exploring complementary therapies; in this study, 88% of the interviewees had tried at least one type, and more than one-third in both groups took vitamin and/or mineral supplements.

Another common strategy was to experiment with diet. Some tried the anti-candida diet, avoiding sugar, yeasts and molds. Others eliminated certain foodstuffs from their diet, and/or introduced more organic food. The greater use of complimentary medicine in Group 1 may have reflected the fact that the local group leader is the daughter of a homeopath and has a specific interest in alternative approaches to health.

In trying to deal with the illness as a whole, people relied more on positive thinking and other emotion-focused strategies. Many referred to remaining optimistic, concentrating on what they had and hoping for new treatments, for more understanding, and for a cure. It is of interest, here, that optimism has been linked with effective coping in other conditions (e.g. Scheier and Carver 1987, Weisman and Worden 1976).

Few patients used strategies such as normalisation (Wiener 1975). It is possible that most of the patients in this study did not cover up the fact that they were unwell because they were too ill to do so. Indeed, only two people mentioned that they preferred to keep symptoms to themselves

and in both cases, it was because they did not want to burden friends and/or cope with possible conflicts.

Similarly, only a few people mentioned that they tended to compare themselves with others who were worse off than themselves. In their opinion, this helped them to put their own problems in perspective and deal with self-pity. This approach is also used by other patients groups e.g. Smith (1979) and Taylor (1983).

The variety of coping strategies which were reported is inconsistent with the notion that people with CFS are generally passive, and that they do little other than rest and wait for a cure. Moreover, the findings do not support the view that these patients are rigid in their response to fatigue and indeed, the references to changing values and priorities indicates that many did adapt to changing circumstances (cf. Surawy et al 1995).

Restraints on coping

As noted above, CFS is characterised by marked fluctuations in the severity of symptoms. Although over-exertion was identified as one of the commonest reasons for exacerbations, the coping strategies used gave only a certain amount of control. Consequently, many patients had only a limited idea of which symptoms they would have to deal with at a given time, how severe they would be and how long any flare-ups would last.

Research on other disorders has suggested that the changeability of symptoms requires frequent reassessments and modifications to coping strategies and that it makes it more difficult for people to evaluate the effectiveness of the latter (Mishel and Braden 1988, Strauss et al 1984). This should be taken into account, not just when considering the sources of emotional distress but also when assessing the

outcomes associated with specific strategies and treatments (Sibley 1988).

A second constraint on coping were the symptoms themselves. Muscle weakness and fatigue limited activities, including ones which might be therapeutic such as exercise. Similarly, nausea sometimes caused difficulties with eating and taking medicines. In terms of Lazarus's theory, this means that CFS was not only an ongoing source of change in the person-environment relationship but also a factor which reduced the choice of available coping strategies (Lazarus 1991).

The effectiveness of coping

The effectiveness of the strategies mentioned by the patients with CFS was difficult to assess from the available data. Some patients admitted to being overwhelmed and feeling despair at times. However, many people had tried a number of different approaches, and some of these, such as rest and pacing, were felt to be helpful. Indeed, the patients believed that with careful planning or pacing, they were able to conserve sufficient energy to complete certain tasks every day. In the long-term, they felt that pacing allowed them to limit their dependence on others and in many cases, that it reduced the number of relapses. However, while it appeared to help them to retain a degree of control over their lives, it may also have restricted their activities and in some cases, increased their isolation (cf. Ray et al 1995).

As far as social support is concerned, a significant number mentioned this both to deal with the symptoms and with the illness as a whole. However, as noted above, many reported having been confronted with a lack of support which caused them great distress. According to Bloom (1982), support from others aids adaptation to illness because it can

decrease a patient's sense of vulnerability, reduce feelings of isolation, maintain the person's sense of identity, increase access to useful information and provide a source of affection and acceptance. On this basis, it might be useful to study the effects of support on both emotional distress and the level of symptoms and disability in more detail.

The limited use of coping strategies such as avoidance and wishful thinking is noteworthy since these have been linked to poor adjustment in people with other conditions (e.g. Frank et al 1987, Felton and Revenson 1984, Vitaliano et al 1989). Instead, many CFS patients appeared to have approached their illness in ways which has been associated with good coping (e.g. Weisman and Sobel 1979). These include staying optimistic, being practical about the kind of solutions that are feasible, flexibility in not insisting upon a rigid approach to any problem and resourcefulness in finding support or additional information that helps implement behaviour.

It is also interesting to note the similarities between the strategies used by the CFS patients and those employed by people with multiple sclerosis and rheumatoid arthritis (see for instance, Locker 1983, Robinson 1988).

Finally, some indication of the perceived value of various coping strategies can be obtained from the advice which patients would give to new sufferers. The most frequently cited coping strategy was rest. Moreover, about 40 per cent advocated that people should adopt a positive attitude and slightly fewer advised that they pace their activities and join a patient group (for support and information).

The recommendations were generally consistent with the strategies which patients used themselves. However, one anomaly was the cautious approach to alternative medicine,

particularly by Group 1. Although many felt it was useful, the expense and the limited effectiveness of some of the therapies may have reduced the extent of their enthusiasm.

3.6 Questions arising from the research

The findings above suggest that both the severity of symptoms and a lack of social support may be important sources of psychological distress. Further research is required to clarify the influence of these variables in more detail, for instance, by distinguishing between types of symptoms and sources of support. Secondly, given the uncertainty and unpredictability associated with this illness and the difficulties this apparently caused in terms of planning activities, it may be useful to assess in a more formal way if these variables are correlated to functional impairment and psychological well-being. Lastly, research is needed to determine if the patient's strategies of rest and pacing are associated with a positive outcome, or whether, as suggested by the cognitive-behavioural model, these will perpetuate the fatigue and undermine recovery.

3.7 Summary

This study showed that the illness caused considerable disruption and distress. Some of this could be due to the controversy relating to the status of the illness since the outside world did not always recognise CFS as a genuine disease (Pepper et al 1993).

The results also revealed that most patients engaged in fairly complex reasoning far beyond that described by the cognitive-behavioural models (e.g. Butler et al 1991, Sharpe 1992, 1994, Surawy et al 1995). Indeed, many patients attributed their illness to a combination of internal and external factors, and did not reject a role for psychological variables as has been suggested.

The finding that patients used a variety of coping strategies and that they did not reject psychological help when offered contrasts with the view that many adopt a passive approach to the illness and that their response to their symptoms is largely limited to rest.

Finally, this study suggests that the extent and impact of the social and psychological consequences of CFS may have been underestimated.

CHAPTER 4

Study into the psycho-social sequelae of chronic fatigue syndrome (myalgic encephalomyelitis)

4.1 Introduction

The presence of emotional distress in the chronically-ill is now acknowledged as a serious problem which can limit the individual's quality of life and exacerbate symptoms, undermine adaptation and even prevent recovery (Billings et al 1983, Butler et al 1991, Sharpe 1994, Wells et al 1989). However, little is known about the variables which may cause or exacerbate emotional distress in patients with CFS.

Although a number of studies have investigated the psychological aspects of this illness, most of the attention has focused on the nature and prevalence of psychopathology (e.g. Blakely et al 1991, Kruesi et al 1989, Krupp et al 1994, Millon et al 1989, Taerk et al 1987, Wessely and Powell 1989, Wood et al 1991). Using a variety of criteria and measures, it has been estimated that between 13% and 79% of patients suffer from current major depression (Yeomans and Conway 1991, Millon et al 1989). However, one study failed to find any patient who fulfilled the criteria for psychiatric illness (Peterson et al 1991).

The lack of agreement concerning the actual prevalence of clinical depression can be attributed, at least in part, to the differences in instruments and cut-off points (see Table 1a). Another potential source of error is the inclusion of symptoms like fatigue, insomnia and cognitive deficits, which are part of the clinical picture of CFS as well as depression (Dutton 1992, Millon et al 1989, Ray 1991, Thase 1991). As a result, a number of studies have omitted one or more of these complaints from their assessment of psychiatric morbidity (e.g. Katon et al 1991, Kruesi et al 1989,

Macdonald et al 1993a, Wessely and Powell 1989, Wood et al 1991). However, others have not (Buchwald et al 1994, Hickie et al 1990, Krupp et al 1994, Lane et al 1991, Manu et al 1988, Pepper et al 1993, Walford et al 1993).

In some cases, correcting the data for overlapping symptoms made no significant difference to the results (e.g. Wood et al 1991). On the other hand, Katon et al (1991) found that omitting fatigue from the criteria reduced the rates of current major depression from 15.3% to 10.2%.

Certain disability-related items included in questionnaires may also inflate prevalence rates. For example, Yeomans and Conway (1991) noted that 33% of their patients had a score of 11 or more on the depression subscale of the Hospital Anxiety and Depression Scale (HAD). However, when the item 'I feel as if I am slowed down' was omitted, this reduced the scores to such a degree that no one reached the cut-off point for caseness.

In spite of the difficulties in estimating the prevalence of mood disorders, it is generally accepted that depression is more common in patients with CFS than in the population at large (Fukuda et al 1994, Hickie et al 1990). To assess whether the higher rates could be attributed to the degree of disability, some researchers have compared CFS with other medically-ill groups. For instance, Krupp et al (1994) and Pepper et al (1993) found that patients with CFS had higher levels of self-reported depressive symptoms and a greater frequency of major depression than people with MS.

Similar studies have compared CFS patients with people suffering from rheumatoid arthritis (Katon et al 1991), cystic fibrosis (Walford et al 1993) and neuromuscular and muscle disorders such as myasthenia gravis, myopathies and muscular dystrophy (Wessely and Powell 1989, Wood et al 1991). However, in the majority of studies, the comparison

groups were not matched for the degree of impairment or severity of symptoms. For example, where fatigue was measured, the levels recorded by these medically-ill samples were significantly lower than those of the patients with CFS. Where fatigue was not assessed, other illness-related variables showed that the comparison group had fewer symptoms than the patients with CFS (Katon et al 1991). Thus although CFS has been associated with a greater frequency of psychiatric disorders than other disabled groups, the failure to take into account their higher levels of symptomatology and their greater functional impairment means that the relationship between illness-related factors and psychopathology is still far from clear.

Another variable which may influence psychological distress and psychiatric morbidity is the intrusiveness of the disease (Devins et al 1993ab, Schubert and Foliart 1993). For instance, one can not assume that the impact of fatigue or nausea on a person's life will be identical to that caused by weakness in one's legs. Other illness-related variables which should be considered when assessing emotional distress include:

1. the nature of the disease, i.e. whether it affects primarily physical functioning or also 'mental' activities; whether the symptoms are localised or systemic,
2. the course of the disease, i.e. whether it is stable, fluctuating, or progressive and likely to lead to premature death,
3. whether symptoms can largely be controlled,
4. whether there are additional complications or concurrent disorders (Dalos et al 1983, Leedom et al 1981, Paradis et al 1993, Rodin et al 1991).

Thus in terms of identifying the degree of emotional disturbance attributable to illness and disability, it is important not to limit the assessment to one symptom or to one measure of disability or impairment. This is particularly

important in the case of multi-system disorders like CFS, where some symptoms may be overlooked or simply misunderstood (cf. English 1991, Fleming 1994, and Study 1). For instance, Ray et al (1992a) found that cognitive difficulties and somatic symptoms were significantly related to a number of measures of emotional distress, yet the possibility that these complaints may undermine psychological well-being is rarely considered.

Instead, the tendency has been to regard the presence of mood disorders among patients with CFS as reflecting primary psychiatric illness and little else (e.g. David 1991, Hotopf and Wessely 1994). Although it is acknowledged that psychological disturbances may occur as a reaction to disability, most researchers have conceptualized the latter in fairly narrow terms (David 1991). As a result, the role played by variables such as social support in the adjustment to CFS remains unclear.

4.1.1 The role of psycho-social factors in CFS

In one of the few longitudinal studies which included emotional distress as an outcome variable, the only predictor was found to be the score on the affective inhibition subscale of the Illness Behaviour Questionnaire (Wilson et al 1994b). Variables which were not related to psychological adjustment included duration of illness, premorbid psychiatric diagnoses, neuroticism, belief in a somatic cause or denial. However, the use of measures assessing illness behaviour has recently been challenged. According to Trigwell et al (1995) the similarity of the results from patients with CFS and MS suggests that the findings may reflect the nature of a condition rather than the presence of psychopathology.

There is also a lack of clarity about the relationship between somatic attributions and emotional distress (Bonner

et al 1994, Vercoulen et al 1994 and see Chapter 3. Nevertheless, there is evidence that negative thinking about the illness, e.g. denial and self-blame, may lead to maladaptive coping, an increase the perceived disability and disruption of social relationships (Antoni et al 1994). Similarly, maladaptive thoughts might result in greater functional impairment and more severe fatigue (Petrie et al 1995). A link between dysfunctional cognitions and disability has also been documented in other patient groups (e.g. Rodin et al 1991), and has stimulated interest in treatments such as cognitive-behaviour therapy (see Chapter 5). As for the disruption in relationships, one study found that negative social support was significantly correlated with both depression and anxiety (Ray 1992). Conversely, positive support was associated with reduced anxiety scores.

The levels of support for people with CFS may have been undermined by the generally unsympathetic publicity in the media. During the past few years, the press have trivialised this illness, dismissing it as 'yuppie flu' (e.g. Hodgkinson 1991), while medical articles have accentuated the role of psychiatric factors (Thomas 1993, Lawrie and Pelosi 1994) and portrayed sufferers in an exceedingly negative light (cf. Read 1993). It is possible that this may have reduced the available support not only from friends and family but also from health care professionals. Indeed, Fleming (1994) noted that in her dealings with doctors, she "encountered undermining attitudes and hurtful words" and that she only obtained the help and advice she needed by going outside the NHS.

It has also been suggested that doubts about the origins of an illness may lead some to withdraw or reduce their support (Woodward et al 1995). For instance, Faucett and Levine (1990) surveyed patients suffering from chronic pain and found that where the cause was an organic lesion, people were much more sympathetic than where the cause was unknown.

They also reported that the patients who were denied social support were more likely to suffer from depression. If Faucett and Levine are correct, people with CFS would receive less support and lose more friends than people whose condition has a known, organic cause e.g. spinal cord injury.

As well as increasing distress directly, lack of support may undermine the patient's well-being indirectly. For instance, Lewis et al (1994) proposed that low levels of perceived support might lead to immunological changes which could predispose individuals to depression as well as CFS.

Another possible source of emotional distress which has received comparatively little attention so far is perceived uncertainty. According to Mishel (1988), feelings of uncertainty can result from ambiguities concerning the illness itself, a lack of information about the diagnosis or seriousness of the illness, and the unpredictability of the course and outcome.

With regard to CFS, the lack of a specific diagnostic test means that there may be continuing doubts about what is wrong. The illness also has a highly variable course and in many cases, an unknown outcome. Likewise, the presence of cognitive dysfunction may undermine the patient's ability to process information, thus further increasing uncertainty (Mishel 1988). However, although this aspect of the illness has been implicated as one factor undermining successful coping (e.g. Ray 1991), there has been little research to assess its role in more detail.

Research into other disorders has identified a wide range of variables associated with emotional distress and psychiatric morbidity (see Chapter 2). For instance, studies on patients with spinal injuries have identified a number of possible influences, including demographic variables such as

age (Tate et al 1993) and gender (Tate et al 1994), disability-related variables such as level and completeness of lesion (Fullerton et al 1981, Judd and Brown 1992, MacDonald et al 1987), level of handicap (Tate et al 1993, 1994), duration (Richards 1986) and the severity of pain (Craig et al 1994). Additional predictors (see Tables 1b and 1c) include economic difficulties (Tate et al 1994) and social factors such as isolation (Tate et al 1994).

Research on other disorders has also underlined the complexity of the relationship between social support and emotional distress. To summarise, it has been found that the effects of support may vary according to the provider (e.g. partner or friend), the time one has been ill or disabled (Dakof and Taylor 1990, Elliott et al 1992, Neuling and Winefield 1988) and the severity of disease (Ell et al 1992). Studies have also shown that it is important to distinguish between the quality or adequacy of support and the availability of support (e.g. Fitzpatrick et al 1991, Goodenow et al 1990), with the former often having a greater influence on emotional well-being than the latter. Clearly, these factors should also be taken into account when assessing patients with CFS.

Other aspects of adjustment

To date, few studies on CFS have examined the variables associated with functional impairment. However, as in the case of research on emotional distress, a number of different factors may be involved. For instance, Vercoulen et al (1994) found that problems with work, housekeeping and general level of activity were related to difficulties with mobility, impaired concentration, strained relationships as well as psychological variables such as anxiety and depression.

According to the cognitive behavioural model, there should

be a strong and direct relationship between depression and impairment and more specifically, between depression and fatigue. However, while studies support an association between the latter and depression (McDonald et al 1993, Ray et al 1992a), the correlations have generally been modest. Moreover, not all the findings have been consistent. Given the paucity of research, further studies are required to clarify the association between the symptoms of CFS, including cognitive difficulties, and functional impairment.

Research aims

Given the present lack of knowledge about the possible sources of emotional distress and disability in patients with CFS, it was decided to study the key variables which have been implicated in research on other disorders, and to compare the results with those of people with spinal cord injuries (SCI). The latter were chosen because in contrast to CFS, it has a known cause, the course and outcome are more predictable and the condition is accepted as a genuine source of disability. As a result, it was felt that they would provide useful information regarding the psychological consequences of having a chronic and disabling disorder, without many of the uncertainties which surround CFS.

The main aims of the study were:
1. to determine the degree of adjustment, social support and uncertainty in patients with CFS and to compare the results with those of people with a spinal cord injury. In this study, adjustment was assessed using three variables: anxiety, depression and functional impairment.

2. to determine the relationship between adjustment on the one hand and illness or disability-related variables, social support and uncertainty on the other.

Table 1a. Recent research assessing psychological and psychiatric morbidity in CFS

Authors	Sample type (number)	Diagnostic Criteria	Measures and main findings
Blakely et al. 1991	1. CFS (58) 2. Chronic pain (81) 3. Well controls (104)	McKenzie	GHQ-28 >5 59%/54%/13.5% BDI-13 ≥8 55%/38%/11%
Buchwald et al. 1994	1. CFS (287) subdivided into female/male	CDC	DIS Current MD 22%/22% .. anxiety 12%/16% All cur. diag. 33%/44% Lifetime MD 74%/64% .. anxiety 28%/31% All lifet. diag. 81%/82%
Demitrack et al. 1991	1. CFS (30) 2. Well controls (72)	CDC	DIS[1] Lifetime MD 20%/NA Lifetime anxiety 23.3%/NA All lifet. diag. 53% BDI ≥19 (N=19) 16% HAM-D ≥16 (N=19) 16%

Table 1a cont.

Authors	Sample type (number)	Diagnostic Criteria	Measures and main findings	
Friedberg and Krupp 1994	1. CFS (pre-therapy 22) 2. CFS (no therapy 22) 3. Depression (20)	CDC	SCID All diagn. MD	77%/50%/100% 45%/27%/85%
Hickie et al. 1990	1. CFS (48) 2. Depressed patients (48) *N=33 before treatment	LLoyd	SCID-P premorbid MD current MD Anxiety All MD during CFS GHQ-30 >5 Zung >48 HAM-D m.	12.5%/62% 21%*/58% 0%/NA 24.5%/100% 46%/- 45%*/NA 24%*/77% 10.6/19.0
Katon et al. 1991	1. CF (79) 2. CFS (19) 3. Rheumatoid arthritis (31)	CDC	DIS[1] Current MD CF/RA Lifetime MD Current anxiety Lifetime anxiety	10%/3% 67%/39% 17%/3% 31%/16%

Table 1a cont.

Authors	Sample type (number)	Diagnostic Criteria	Measures and main findings		
Katon et al. cont.				All cur. diag.	45%/6%
				All lifet. diag.	86%/48%
				GHQ≥ 11 CF/RA	53%/12.5%
Kruesi et al. 1889	1. CFS (28)	CDC	DIS	Lifetime MD	46%
				All diagnoses	75%
Krupp et al. 1994	1. CFS (20)	CDC	SCI	All diagnoses	25%/NA/NA
	2. MS (20)			Current MD	15%/6%/NA
	3. Well controls (20)			Atypical depr.	5%/NA/NA
				Dysthymia	5%/NA/NA
Lynch et al. 1992	1. CFS (34)	Oxford	SCID[1]-Depr.		41%/100%
	2. Depression (34)			dysthymia	29%
				anxiety	9%/NA
				All diagnoses	79%
			MADRS m.		21.1/21.6

Table 1a cont.

Authors	Sample type (number)	Diagnostic Criteria	Measures and main findings	
Lynch et al. cont.			GHQ-30 ≥8 HAS m.	50%/59% 15.9/20.8
Millon et al. 1989	1. Allergy clinic attenders with CFS (24)	CDC	HAM-D >20 MCMI-II>75 MD -anxiety MCMI-II>BR75 MD	79% 17% 18.8 54%
Pepper et al. 1993	1. CFS (69) 2. MS (65) 3. Depr. (20)	CDC mod.	SCID Current diagn. Lifetime diag. Diag. after illness onset	23%/7.5%/all 51%/32%/NA 56%/11%
Peterson et al. 1991	1. CFS (135)	CDC	Interview (for major psychiatric disorders)	0

Table 1a cont.

Authors	Sample type (number)	Diagnostic Criteria	Measures and main findings	
Schweitzer et al. 1994	1. CFS (40) 2. GP attenders (150) 3. Psychiatric pts (250) 4. Cancer pts (47) 5. Hypochondria (100)	Lloyd	BDI ≥ 21 CFS only GHQ-28 ≥5 CFS patients had higher scores on IBQ hypochondriasis scale than groups 2 and 3.	20.5% 67%
Shanks and Ho-Yen 1995	1. CFS (64)	Oxford	SADS[1] No psychiatric disorders Psych. disorder with or after onset CFS Psych. disorder also prior to onset CFS	55% 34% 11%
Sharpe et al. 1992	1. CF (177)	Own	HAD total >11 HAD -1 item	66% 56%

Table 1a cont.

Authors	Sample type (number)	Diagnostic Criteria	Measures and main findings	
Wessely and Powell 1989	1. CFS (47) 2. Neuromuscular disorders (33) 3. Depressed in-patients (26)	Oxford[1]	SADS[1] MD anxiety All diagnoses Past history GHQ-12 m. HAD total	47%/NA 2%/NA 72%/36%/NA 43%/30%/64% 7/2/10.5 16/11/27
Wood et al. 1991	1. CFS (34) 2. Muscular Dystrophy (24)	Oxford	PSE[1]- MD anxiety all history HAD depression m. anxiety m. STAI state anxiety m. trait anxiety m.	23.5%/12.5% 6%/0 41%/12.5% 26.5%/0 8.3/5.3 8.6/7.0 39.1/34.8 36.5/38.3

Table 1a cont.

Authors	Sample type (number)	Diagnostic Criteria	Measures and main findings
Wood et al. 1992	1. CFS unwell (23) 2. CFS recovered (14) 3. Well students (20)	Oxford	HAD N=19/N=9 Dep. >8 32%/0/NA >10 5%/11%/NA anxiety >8 37%/0/NA >10 16%/11%/NA Negative affect was not correlated with either physical or mental energy.
Yeomans and Conway 1991	1. CFS (15)	Oxford CDC (60%)	MADRS (depression) 93% HAD .. 33% PSE .. 13% HAD (anxiety) 27%

[1] Fatigue not included.

† equivalent of

Key to abbreviations

BDI - Beck Depression Inventory

CDC - Working case definition devised by Holmes et al, Centers for Disease Control

CES-D - Center for Epidemiologic Studies Depression Scale

CF - Chronic fatigue

CFS - Chronic fatigue syndrome

DIS - Diagnostic Interview Schedule

GHQ - General Health Questionnaire

HAD - Hospital Anxiety and Depression Scale

HAM-D - Hamilton Rating Scale for Depression

HAS - Hamilton Rating Scale for Anxiety

IBQ - Illness Behavior Questionnaire

PSE - Present State Examination

MADRS - Montgomery-Asberg Depression Rating Scale

MCMI-II - Millon Clinical Multiaxial Inventory II.

NA - Information not available

SADS - Schedule for Affective Disorder and Depression and Schizophrenia

SCI - Structured Clinical Interview for DSM III-R

SCID-P - Standardised Psychiatric Interview Schedule

STAI - State and Trait Anxiety Inventory

Zung - Zung Self Rating Scale for Depression

Anx. - Anxiety
Dep. - Depression
Diag. - Diagnosis
Lifet. - Lifetime
M. - Mean
MD - Major depression
Mod. - modified

Table 1b. Research assessing psychiatric morbidity in patients with spinal cord injuries

Authors	Sample type (number)	Measures used	Main findings
Frank et al. 1985.	1. SCI (32) Community sample	Interview	MD 38% All depr. disorders 44% Lifetime depr. 45%
Fullerton et al. 1981	1. SCI (30) rehabilitation unit patients	SADS	MD 30% All diagnoses 50% Depr. was more common in those with complete lesions.
Gerhart et al. 1992	1. SCI (121) "minimally disabled"	Medical records Interviews	*Depression* documented in 10%
Judd et al. 1989	1. SCI (71) acute patients	SCID BDI ≥14	MD 19% 38% "understandable dysphoria" 18%

Table 1b cont.

Authors	Sample type (number)	Measures used	Main findings
Judd et al.		HAM-D m.	24.4 Rates of depr. were similar for paraplegics and quadriplegics (17% vs. 23%)
Judd and Brown 1992	1. SCI (227) admissions to SCI unit	Semi-structured interviews	MD 14% Anxiety/depr. mood 3% All disorders 21% Past history 6% *Depr. more common in* quadriplegics (18% vs 11%)
MacDonald et al. 1987	1. SCI (53) community sample av. 7.3 yrs post-injury	BDI >10 CDM clin. depr. API	45% 13% Of these, 86% were quadriplegic, 14% were paraplegic. Depression was associated with fewer personal care activities.

Table 1b cont.

Authors	Sample type (number)	Measures used	Main findings
Malec and Neimeyer 1983	1. SCI (28) inpatients (rehab. programme)	BDI ≥10-20.	25%
		>21.	18%

Table 1c. Research assessing factors relating to adjustment of people with spinal cord injuries

Authors	Sample type (number)	Measures used	Main findings
Anson et al. 1993	1. SCI (125) injured>1 year	QOLI RSSS	People who received and give support perceived themselves as better adjusted and had fewer heath problems than those with less support.
Fuhrer et al. 1992	1. SCI (140) Community sample	LSIA FIM CHART Soc.Sup. scale	Life satisfaction scores were associated with health, soc. support, mobility, social integration and perceived control.
Krause and Crewe 1987	1. SCI (179) 2. SCI deceased (46)	LSQ	Survivors had better psychosocial and vocational adjustment and were socially more active than persons who had died.

Table 1c cont.

Authors	Sample type (number)	Measures used	Main findings
Schulz and Decker 1985	1. SCI (100) 40 yrs.or more SCI of 20 years+	Interview LSIA IPWB m. CES-D >16	70% highly satisfied soc. sup. Depends on disability 4.4 (just below gen. pop.) 22% Those with high levels of social support and perceived control reported high level of well-being.
Tate et al 1993	1. SCI (119) 1 year post injury follow-up	BSI Psych. distress	14% at admission and one year after injury. Predictors included distress at admission, incomplete lesions, occupational status and less insurance coverage. Younger patients reported more distress than older ones.

Table 1c cont.

Authors	Sample type (number)	Measures used	Main findings
Tate et al 1994	1. SCI (163) outpatients Av. 4.6 yrs post-injury	BSI Psych. distress Zung	Predictors of distress included level of handicap, lack of social integration, gender. Predictors of depression included handicap, lack of social integration, economic problems.
Woodrich and Patterson 1983	1. SCI (251) Min. duration 6 months.	ADS	Self-rated acceptance of disability was higher in women, younger patients, those who had been injured longer and in people with higher levels of education.

Key to additional abbreviations

ADS - Acceptance of Disability Scale

API - Activity Pattern Indicators

BSI - Brief Symptom Inventory

CHART - Craig Handicap Assessment and Reporting Technique

CDM - Clinical Depression Measure

FIM - Functional Independence Measure

IPWB - Index of Psychological Wellbeing

LSIA - Life Satisfaction Index-A

LSQ - Life Situation questionnaire

RSSS - Reciprocal Social Support Scale

QOLI - Quality of Life scale

4.3 Method

4.3.1 Sample characteristics and procedure

The CFS group consisted of consecutive patients attending the Neurocare Support Centre at Romford Hospital and the Microbiology clinic at St. Andrew's Hospital, Billericay. All had been diagnosed by an experienced specialist as suffering from ME using the criteria described by Dowsett et al (1990) and Ramsay (1992)[1]. People whose diagnosis was in doubt, or who suffered from another disorder characterised by fatigue, were not included in this research. The sample was therefore relatively homogenous and could be designated as CFS(ME). Most of the patients were enrolled during clinic visits. However, a few were sent a letter by the consultant asking for volunteers.

Since a small number of questionnaires for the CFS(ME) group were lost when the consultant moved to another hospital, it was not possible to establish exactly how many were given out. However, it has been estimated that the number lies between 70 and 90, and given that 67 were completed and returned, the response rate is at least 74% and may be as high as 96%.

Thirty-six per cent of the patients reported having another condition in addition to CFS(ME). The illnesses mentioned included asthma, epilepsy, eczema, hypothyroidism, hypertension, migraines and psoriasis. Some were transient, some were controlled with medication and others were not causing problems at the time of testing. However, seven respondents reported having a second chronic disorder which was also associated with significant levels of fatigue and/or emotional distress. They included pernicious anaemia, seasonal affective disorder, major depression and arthritis. One person

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[1] See Appendix 1.

who suffered from a number of conditions also noted that a recent bereavement was causing additional "exhaustion". Because of the confounding influence of these particular disorders in evaluating the adjustment to CFS(ME), particularly in relation to the levels of fatigue and emotional distress, and in line with advice from the American Centers for Disease Control (Schleuderberg et al 1992) and Australian researchers (Wilson et al 1994a), the consultant agreed that these seven patients should be excluded from the main sample. Further details relating to this 'CFS-plus' group can be found in Appendix II.

Two other cases were later excluded from the main sample, one because the respondent denied having symptoms which had been observed by both the consultant and her assistant, and the other because the patient was undergoing tests for suspected post-polio syndrome.

The comparison group consisted of people with spinal cord injuries (SCI). These were approached by the psychologist attached to the Regional Spinal Injuries Centre at Southport General Infirmary. One hundred individuals who attended the Centre and who had lesions from C6 to L5 were sent a letter which described the study and asked for volunteers. Since the vast majority had lesions above T10, it could be assumed that they would have marked difficulties with physical functions, particularly walking (Grundy et al 1986). Unfortunately, it was not possible to match for gender since the number of women with SCI tends to be relatively small.

Thirty people responded to the letter and were sent questionnaires. Three did not return them, and one refused to participate because of the nature of the study. Another respondent had recovered from his injury and therefore felt unable to participate. The response rate among the volunteers was therefore 83%.

Four (16%) of the respondents in the SCI group reported having a concomitant disorder unrelated to their injury. However, in none of the cases were these associated with significant chronic fatigue or emotional distress and they were therefore not likely to confound the evaluation of the latter.

Every participant was provided with a stamped addressed envelope to return the completed questionnaires. None were paid.

4.3.2 Measures

All the participants were asked to fill in the following questionnaires:

The Profile of Fatigue-Related Symptoms (PFRS)

This 54-item measure developed by Ray et al (1992) assesses the pattern and severity of a number of symptoms commonly reported by patients with CFS. Respondents rate the extent to which they experienced symptoms during the past week on a 7-point scale from 0 ('not at all') to 6 ('extremely'). The score is the mean of the items for each subscale.

A principal components analysis of the original data yielded 4 scales: emotional distress, cognitive difficulty, fatigue and somatic symptoms. The internal consistency of each of the scales was high, ranging from .88 to .96, while the correlations between the scales were lower (range .44-.64). The test-retest reliability was high (range .86-.97) and the subscales correlated well with the relevant mood and symptom measures.

The Medical Outcomes Study (MOS): physical functioning subscale

Originally adapted from the Rand Health Insurance Experiment, the MOS Short-Form consists of 20 items focusing on

physical and role functioning, mental health, health perception and pain (Stewart et al 1988). The subscale evaluating physical functioning contains 6 items which assess the extent to which health interferes with a variety of activities, e.g. walking, sport, climbing stairs etc. Since this is the only MOS subscale used in this study, the answers were scored from 1-3, following Katon et al (1991), rather than 1-100 as originally specified. The values were summed to produce a single variable with higher scores indicating better functioning.

This measure is both short and reliable (α = .86), and has been used to assess a number of patient groups, including people with hypertension, coronary heart disease, diabetes, myocardial infarction and depression. It was also one of the measures used by Katon et al (1991) in their research into CFS (see Table 1a).

A further question to determine the subject's degree of impairment was devised by the researcher. It was named 'help required' and addressed the patient's need for assistance when engaging in 8 basic activities of daily living (e.g. getting in and out of bed, cooking, eating etc). Positive responses were summed and analysed separately from the data above.

Mishel Uncertainty in Illness Scale - Community Form (MUIS-C).
This 23 item Likert-format scale is the only currently available, validated measure assessing the uncertainty related to illness (Mishel and Epstein 1990). It was developed from the original four-factor MUIS scale for use by patients who are not hospitalized.

The items in the community form focus on 4 different aspects of uncertainty: the ambiguity of cues related to illness and treatment, the complexity of these cues, the inconsistency

of information and the unpredictability of symptoms and outcome. The responses are scored from 1 to 5 with the highest score representing greater uncertainty. If an item is not applicable, it is scored zero. The total for the MUIS-C can range from 0 to 115, and represents one factor.

To make some of the questions applicable to the samples being studied, all items referring to 'pain' were changed to 'symptoms' and all references to illness were changed to 'condition'. Likewise, since many of the patients with CFS were not having treatment as such, items focusing on the latter were amended to include advice, and people were instructed to take the last consultation with a doctor as their reference.

Since the unpredictability of the illness might be an important influence with regard to CFS, and given that the MUIS-C includes only one item which addresses this aspect of the condition, it was decided obtain a more detailed assessment of this variable by adding a further four items. These were all taken from the original MUIS (Mishel and Epstein 1990), and the total scores for this complete subscale range from 0-25.

Normative data are available for patients with epilepsy, myocardial infarction, lupus, multiple sclerosis and cancer. Moreover, De Groot recently used the MUIS-C in a study involving CFS patients (personal communication).

The alpha co-efficients for the MUIS-C have been found to be in the moderate to high range (α=.75 -.90).

Life Stressors and Social Resources Inventory (LISRES): subscales for interpersonal resources and stressors

This measure, devised by Moos and Moos (1988), focuses on stressors and resources in a number of areas including physical health, finances, interpersonal relations, as well as

positive and negative life events. The subscales addressing interpersonal resources and stressors were chosen to provide both quantitative and qualitative information about social support, or lack of it, in an outpatient sample.

Three of the subscales assessed the quality of support provided by the spouse/partner, friends and relatives. These measures consist of 11 items each, five relating to stressors and six relating to resources. All the answers are rated on a 5-point scale from 1 ('never') to 5 ('often'). The score for the subscale is the sum of the scores divided by the number of items.

Two additional subscales enquired about the subject's contact with friends and relatives. The items were taken from the LISRES friends resources subscale but scored separately from those above to give a measure of the number of close contacts as opposed to the quality of support. Each subscale consisted of two questions which were scored from 1 signifying no contact to 5 referring to contact with four or more friends or relatives, or to contact several times a week. The score for each subscale, called 'contact with friends' and 'contact with relatives', consisted of the total for the two questions.

A further question asking if respondents had lost friends as a result of the illness/disability was added to those from the LISRES. This item was named 'loss of friends' and the scores ranged from 1 ('none') to 3 ('most').

To reduce the load on the subjects, it was decided to omit the additional subscales for male and female parents and children. For the same reason, and because of the overlap with questions for functional impairment, it was also decided to omit the items asking about membership of clubs and attendance at religious services. The exclusion of these questions means that it was not be possible to compare all

the subscales with the normative data available.

The complete subscales from the LISRES were found to have high reliability and good construct and concurrent validity. Internal consistencies (Cronbach's alpha) for resources from spouse, partner, children, family and friends ranged from .82 to .91; those for stressors from .77 to .86.

Research using the LISRES has been limited to patients with rheumatoid arthritis and people suffering depression and alcoholism.

Background Information

This questionnaire asked for demographic information including the subject's gender, age, years of education beyond 16, occupation, marital status and house ownership. It also included an item to determine whether income consists entirely of social security benefits ('Low Income').

General Information CFS(ME)

This questionnaire was completed by the CFS(ME) patients only, and asked for information about the main symptoms experienced, the onset of the illness (i.e. acute/gradual), the duration and course of the illness, the use of drugs, treatments tried and the presence of other medical conditions which might colour the responses. A further question, asking whether the subject had been told that the condition would improve, was added to provide a separate indication of uncertainty regarding the future.

The effect of the illness was assessed by two questions taken from Ray et al (1992b). The first, 'level of activity', measured what the patient was able to do compared to the past. Answers consisted of five ranked alternatives ranging from 'I can do hardly anything compared with before' (scored 1) to 'I can do most of what I could do before' (scored 5). The second question focused on the frequency of

symptoms and asked if patients experienced symptoms 'all of the time' (scored 1), 'most of the time', 'some of the time' or 'rarely' (scored 4). For these variables, higher scores therefore indicated higher levels of activity and less frequent symptoms.

General Information SCI

In this questionnaire, people with SCI were asked about the cause of the injury, the site of the lesion, the main symptoms associated with the injury and the presence of other medical conditions. Items also enquired about the course of the disability, the level of activity, frequency of symptoms and the use of drugs. Since it can be assumed that in most cases, the lesion is permanent and that significant improvements were unlikely, the question to assess uncertainty about prognosis was rephrased to determine if doctors had given any indication about possible improvement.

Measures assessing adjustment

Hospital Anxiety and Depression Scale (HAD)

This is a 14-item self-rating scale specifically designed to assess anxiety and depression in people with medical conditions (Zigmond and Snaith 1983). It has two subscales: anxiety and depression, each with 7 items rated from 0 to 3. To reduce false positive ratings due to presence of symptoms common to medical conditions and affective disorders, items referring to somatic symptoms have been excluded. Nevertheless, one item: "I feel as if I am slowed down" may still be unduly influenced by the presence of disease and disability. Since this particular item may not be an independent indicator of emotional disturbance (Yeomans and Conway 1991), a corrected Depression score was calculated omitting this question.

The HAD is a reliable measure with a Cronbach's alpha of .93 for the anxiety scale and .90 for the depression scale

(Moorey et al 1991). It is widely used and has been compared with both clinical rating scales and standardised interviews (Lewis and Wessely 1990, Snaith and Zigmond 1994). It has also been used in a number of studies on CFS, including those by Yeomans and Conway (1991), Wessely and Powell (1989) and Wood et al (1992).

The total subscale scores range from 0 to 21. Scores from 0-7 indicate normal levels of anxiety and depression, scores of 8 or 9 are regarded as indicating possible (borderline) cases of clinical disorder while scores of 11 or above are considered to reflect probable cases of morbidity (Snaith and Zigmond 1994).

Functional Impairment Scale

This measure consists of four visual analogue scales covering the ability to work and manage the home, as well as the ability to take part in social and private leisure activities (Marks 1986). Participants are asked how much their condition has affected each of the designated areas, with ratings ranging from 0 ('not at all') to 8 ('very severe'). The scores are summed and treated as a single variable.

This scale was used by Butler et al (1991) in their research on patients with CFS.

Copies of all the questionnaires can be found in Appendix 1.

4.4 Results

4.4.1 Initial analysis

The alpha co-efficients were computed for the three complete measures used, i.e. the PFRS, the MUIS and the Functional Impairment Scale.

Table 2. Summary of Alpha coefficients for reliability

Variable	CFS(ME) group	SCI group
PFRS		
Fatigue	.93	.95
Emotional distress	.95	.94
Cognitive difficulty	.95	.93
Somatic symptoms	.91	.81
MUIS-C		
Uncertainty	.86	.94
Functional impairment		
All subscales	.79	.81

The reliability of all the above scales was satisfactory for both groups, with co-efficients ranging from .79 to .95.

4.4.2 The CFS(ME) group

Demographic information

The mean age of the CFS(ME) patients was 37.3 years (SD 13.4). Forty-two (72%) of the subjects were female and 16 (28%) were male. Just over half the group were either married or cohabiting while 41% were single. All except three patients had completed secondary school and 31% had also completed college or university.

At the time of the study, 26% were engaged in full or part-time work while a further 26% were unemployed or on sick-leave. In total, 72% reported that they had changed their jobs or reduced their hours because of their illness and nearly one-third currently had an income consisting solely of social security benefits. For more detailed information,

see Appendix II.

Illness-related variables

The median duration of the illness was 3.5 years (the distribution of the latter being positively skewed). Ten patients had been ill for 10 years or more and 4 people had been unwell for at least twenty years.

Fifty-two per cent of the CFS(ME) group described their illness as having begun suddenly and 60% felt able to identify an infectious condition which occurred at the start. Thus in some cases, the illness followed an apparent infection but developed only slowly after that. Forty-seven per cent of the patients thought that the diagnosis was made solely on the basis of their clinical history. Indeed, where tests were done, this was usually to exclude other conditions, not to support the diagnosis of CFS(ME).

At the time of the assessment, 17% of the patients considered their illness to be fluctuating, 41% were stable, 29% were improving and only 12% felt that they were getting worse. In terms of their disability, 91% reported having symptoms most or all of the time and about 60% were able to do less than 50% compared to what they did before. Just 16% were able to do three-quarters or more of the activities they used to do.

As regards medication, 66% of the CFS(ME) group were taking some type of drug and in at least 18 cases (31%), these included antidepressants. For more detailed information, see Appendix II.

4.4.3 The SCI group

Demographic information

The mean age of the SCI group was 36.5 (SD 7.26) and 20 (80%) of the respondents were male. Just over half the group were married or cohabiting while 40% were single. At the time of the study, 28% were unemployed or on sick leave. Sixty-eight per cent, however, had retired or reduced their hours because of their disability and 52% had an income consisting solely of social security benefits. For more detailed information, see Appendix II.

Disability-related variables

The injuries had occurred a median of 11.00 years (SD 8.80) previously and 20% had been disabled for twenty years or more. In approximately half the group, the injury had resulted from a road traffic accident but in three cases, it had been caused by illness.

Sixty-four per cent of the group had lesions from C5 to T10, and a further 4 respondents (16%) had a lesion at the level of T12. The injuries of the remaining subjects were between L1 and L5. In 13 cases (52%), the lesions were complete.

Seventy-two per cent of the group made use of a wheelchair, and only one person (with a lesion at L5) was able to walk without any mobility aids. The condition was classed as stable by 64%, while 16% reported that it was fluctuating. Only three people (12%) felt their health was getting worse.

Injury-related symptoms such as pain, muscle spasms, numbness and infections were common, with 88% reporting at least one of these. Less than 30% experienced symptoms most or all of the time, while 48% were able to do a half (or less) compared with their previous level of activity.

The commonest drugs used by this group were pain killers and muscle relaxants. Some subjects were also on antibiotics. For further details of disability-related factors, please consult Appendix II.

4.4.4 Comparison between groups

The following sections compare the samples in terms of demographic and illness-related variables, as well as the measures of adjustment, uncertainty and social support.

Where variables were scored using an ordinal and interval scale, differences between the groups were assessed using the two-tailed Mann-Whitney U test (Siegel 1956). The latter was considered to be more appropriate than the parametric T-test because of the difference in sample size, the skewness of some of the data (e.g. functional impairment, duration, support from friends) and the lack of homogeneity of variance (Hays 1994). Categorical data were analysed using the Chi-square test. To reduce the possibility of Type I errors, alpha was set at 0.01.

Demographic variables

The results indicated that the CFS(ME) and SCI groups were similar in terms of age, marital status and years spent in education from the age of 16. However, the CFS(ME) group contained a greater percentage of women (χ =19.5, $p<.001$), and more people who had completed college, university or some kind of professional training.

In terms of their housing, fewer CFS(ME) patients lived alone compared to the people with SCI. On the other hand, there were more people among the SCI group who owned their own home.

Financially, a greater percentage of people with SCI were

dependent on social security benefits (51% versus 29%).

Illness and disability-related variables

The SCI group had been disabled longer (p=.002) and they were more impaired in terms of physical functions like walking than the people with CFS(ME) (p=.0009). They also required help with a greater number of activities (p=.003). However, the CFS(ME) patients reported experiencing symptoms more frequently (p<.0001). There was no difference between the groups in terms of their reported overall level of activity compared to the past.

Specific symptoms were measured by the PFRS. As Table 3 shows, the CFS(ME) patients reported significantly more fatigue and cognitive difficulties than the SCI group (p=.0003, p<.0001 respectively). There was also a trend towards more severe somatic symptoms in the CFS(ME) group (p<0.02).

Table 3. Mean scores (SD) for the PFRS subscales

	CFS(ME) group		SCI group	
	Mean	SD	Mean	SD
Fatigue	3.77	1.23	2.22	1.75**
Cognitive difficulty	3.34	1.45	1.29	1.37**
Somatic symptoms	2.40	1.29	1.63	.94

** p<0.001

Uncertainty

The CFS(ME) group (N=57) reported significantly more uncertainty than the people with SCI (mean 66.26 versus 56.71, p<0.01). They also had higher scores on the subscale measuring unpredictability (mean 17.18 versus 14.45, p<0.01).

It should be noted, however, that there were a number of subjects who felt that many of the items on the MUIS did not apply to them. Since inclusion of large numbers of missing answers would have made it difficult to interpret low scores, it was decided to use only those questionnaires where more than 80% of the items had been completed. This affected the SCI group in particular, reducing the sample size to 21 for uncertainty and 22 for unpredictability.

In answer to the question whether a doctor had told them that their condition would improve, 43 (74%) of the CFS(ME) group answered yes, compared to 5 (20%) of the people with SCI. This difference was highly significant (χ= 18.8, p<.0001).

Social support

The results of the various measures assessing social support are shown in Table 4. No significant differences between the groups were found.

Adjustment

In this study, adjustment was assessed using the scores on the HAD and the Functional Impairment Scale (see Table 5). Due to missing data, the number of cases varied per subscale. Given the strong relationship between the emotional distress subscale of the PFRS and the scores on the HAD, it was decided not to include the former in the analysis.

There was no significant difference between the CFS(ME) and SCI groups with respect to the severity of anxiety. However, the uncorrected depression scores of the CFS(ME) group were significantly higher than those of the SCI group (p=.002) and there was a trend towards significance when one fatigue-related item was omitted (p=.03).

Further analysis revealed that there were more people in the CFS(ME) group who scored above the cut-off points indicating clinical anxiety and depression (see Table 6). However, the difference between the groups did not reach significance. It should also be noted that correcting the depression subscale reduced the number of cases of mild and more severe depression in both groups.

In terms of functional impairment, the CFS(ME) group was found to be significantly more disabled than the people with SCI ($p=0.01$).

Table 4. Means and standard deviations for the social support variables

Variable	CFS(ME) group Mean	SD	N	SCI group Mean	SD	N
Friends resources	14.79	4.40	56	16.54	5.11	24
Spouse resources	19.54	3.51	39	18.53	5.30	15
Relatives resources	15.93	4.37	58	14.80	4.69	25
Friends stressors	5.75	2.89	56	5.83	2.87	24
Spouse stressors	6.08	2.69	39	8.07	4.23	15
Relative stressors	7.57	3.50	58	7.96	3.90	25
Friends contact	6.98	2.27	58	7.64	2.46	25
Relatives contact	6.52	1.98	58	7.28	2.46	25
Friends loss	1.83	.80	58	1.88	.78	25

Table 5. Means (and SD) for anxiety, depression and functional impairment

	CFS(ME) group		SCI group	
	Mean	SD	Mean	SD
Anxiety	9.65	4.03	8.32	5.51
Depression	8.24	3.03	5.61	3.55*
Depression corrected	5.72	2.98	4.13	2.86
Functional impairment	22.57	5.07	17.64	8.91*

* $p \leq .01$

Table 6. Prevalence of possible and probable cases of anxiety and depression

		Anxiety		Depression		Depression corrected	
Scores		CFS(ME)	SCI	CFS(ME)	SCI	CFS(ME)	SCI
≥8	No.	39/57	12/25	38/58	8/23	16/58	3/24
	%	68	48	55	35	27.5	13
≥11	No.	27/57	8/25	13/58	2/23	4/58	0/24
	%	47	32	22	7	7	0

4.4.5 Relationships between variables

The following report focuses primarily on the relationship between key variables and adjustment in each group. The results concerning the other variables are summarised in the text but more detailed information can be found in Appendix

II.

Some of variables were found to be skewed, with Z exceeding the critical value for $p<.01$. They included functional impairment and duration (CFS(ME) group only), and support from friends (SCI group only). To allow comparisons to be made, both within and between the groups, it was decided to analyse all the data using Spearman's rho (two-tailed). However, if the data met the assumptions for parametric tests, the adjustment variables were also analysed using Pearson's r (for details, see Appendix II). Significant relationships using Pearson's r will be noted in the Tables where appropriate.

The CFS(ME) group

None of the demographic variables in this study were significantly correlated with other measures except for age and relative stressors ($r_s= -.37$, $p<.01$).

The analysis of the illness-related variables revealed a number of inter-correlations. For example, duration was related to the number of years a person had been diagnosed ($r_s=.55$, $p\leq.001$), while the level of activity correlated both with fatigue ($r_s=-.44$, $p<.001$) and cognitive difficulty rs = $-.35$, $p<.01$). For more detailed information, see Appendix II.

The relationship of demographic and illness-related variables with adjustment

None of the demographic variables, and none of the general illness-related measures were associated with anxiety or depression (see Table 7). However, there were significant correlations between fatigue and somatic symptoms on the one hand and anxiety on the other. Moreover, when the symptom scores were analysed using Pearson's product moment corre-

lation, the coefficient between cognitive difficulty and depression just reached significance (r=.34, p<.01).

There were also significant correlations between illness-related variables and functional impairment. For instance, the latter correlated with physical functioning as measured by the MOS subscale, showing that difficulties related to work, looking after the home etc were strongly linked with the ability to walk, carry etc. Functional impairment also correlated significantly with cognitive difficulties, but not with fatigue or somatic symptoms.

The correlation co-efficients between the outcome measures themselves were very weak, ranging from -.04 (anxiety and functional impairment) to .25 (functional impairment and depression).

Table 7. Correlations (Spearman's Rho) between illness variables and adjustment: the CFS(ME) group

	Anxiety	Depression	Depression corrected	Functional impairment
Duration	.10	.13	.13	.23
Years diagnosed	-.04	.07	.08	.40*
Physical functioning	.11	.10	.15	-.38*
Fatigue	.34*[a]	.23	.17	.33
Cognitive difficulty	.32	.32[b]	.26	.37*
Somatic symptoms	.52**	.14	.17	-.01

* p≤.01 ** p≤.001

[a] r = .32 p=.016 [b] r = .34*

The relationship between uncertainty and other variables

Neither uncertainty nor unpredictability were related to demographic variables or any of the illness-related measures. However, as shown in Table 8, uncertainty showed a strong association with levels of emotional distress, particularly anxiety ($r=.56$, $p<.001$). In contrast, there was only a weak relationship with functional impairment. There were no significant relationships between unpredictability and any of the key variables.

Table 8. Correlations (Spearman's Rho) between uncertainty, social support and adjustment: the CFS(ME) group

	Anxiety	Depression	Depression corrected	Functional impairment
Uncertainty	.56**	.42*	.48**	.12
Unpredictability	.12	.14	.17	-.02
Interpersonal resources				
Friends	-.20	-.34	-.39*[a]	-.08
Spouse/partners	-.09	-.25	-.25	-.07
Relatives	-.14	-.33	-.35*[b]	-.08
Interpersonal stressors				
Friends	.17	.16	.17	-.12
Spouse/partners	.40	.37	.43*	-.19
Relatives	.31	.29	.28	.01
Contact				
Friends	-.33*	-.34*	-.37*	-.11
Relatives	-.01	.04	.07	-.19
Loss of friends	.24	.26	.24	.39*

* $p \leq .01$ ** $p \leq .001$

a $r = -.33$ $p = .012$ [b] $r = -.31$ $p = .018$

Table 9a. Relationships between uncertainty, interpersonal resources and illness-related variables: the CFS(ME) group

	Uncertainty	Friends resources	Relatives resources	Spouse resources	Contact with friends	Contact with relatives
Duration	-.04	-.11	.02	.00	-.05	.05
Years diagnosed	-.12	.05	-.08	-.17	.07	.10
Physical functioning	.26	-.10	-.37*	-.23	.02	.11
Fatigue	.11	.18	.23	.17	.02	.14
Cognitive difficulty	.20	.22	.11	.04	.10	-.04
Somatic symptoms	.27	.02	.20	.03	.07	.19
Uncertainty	1.00	-.21	-.37*[a]	-.16	-.27	-.10
Unpredictability	.11	-.21	.00	-.24	-.31	-.20

* $p \leq .01$ ** $p \leq .001$ [a] $r = -.32$ $p = .014$

TABLE 9b. Correlations between interpersonal stressors, uncertainty and illness-related variables: the CFS(ME) group

	Friends stress	Relative stress	Spouse stress	Loss of friends
Duration	.33	.14	.06	.32
Years diagnosed	.23	.18	.21	.34*
Physical functioning	.28	.09	.31	-.10
Fatigue	-.12	.04	-.18	.17
Cognitive difficulty	.10	.06	-.19	.15
Somatic symptoms	.07	.09	.13	.05
Uncertainty	.13	.26	.12	.36*
Unpredictability	-.08	-.10	.24	-.18

* $p \leq .01$ ** $p \leq .001$

The correlations between uncertainty and social support are shown in Table 9a. Uncertainty was found to be correlated with two social support measures: the loss of friends (r_s = .36, P<.01) and a lack of support from relatives (r_s = -.37, p<.01).

Interpersonal relationships

Like the illness-related variables, some of the measures of interpersonal resources and stressors also correlated with one another (see Appendix II). For instance, contact with friends was significantly related to resources from friends (r_s=.61, p<.001).

There were few significant associations between interpersonal relationships and illness. For example, the loss of friends correlated significantly with years since diagnosis, and resources from the family was negatively associated with physical functioning. However, there were no significant relationships between either interpersonal resources or stressors and the number of symptoms, their frequency, the duration of illness and the requirement for help. More detailed information on the relationship between support and illness-related variables can be found in Table 8 and Appendix II.

As far as the association with emotional distress is concerned, contact with friends was negatively correlated with both anxiety and depression. An inverse relationship was also found between resources from friends and the corrected depression score but this was not significant when the analysis was repeated using Pearson's r. Likewise, the correlation between resources from relatives and depression reached significance using Spearman's rho, but not with Pearson's r. Lastly, there was a significant relationship between stress with partners and depression when the score was corrected. However, the correlation with anxiety just failed to reach significance.

None of the social support measures were related to functional impairment, except for the loss of friends ($r=.39$, $p<.01$).

Multiple regression analysis

The data from the CFS(ME) group were analysed further using hierarchical multiple regression. This method was preferred over alternatives because it gives researchers more control over variables of theoretical interest and because stepwise regression requires a larger cases-to-IV ratio (Tabachnick and Fidell 1983). The variables of particular interest were

those relating to the symptoms, measures of disability, social support and uncertainty.

The three dependent variables were depression[2], anxiety and functional impairment.

In the two equations relating to emotional distress, fatigue was the first independent variable to be entered. This symptom is generally regarded as the main complaint associated with CFS(ME) and early entry would give an indication of the influence of the remaining variables independent of fatigue.

The second variable of interest was social support. The correlation coefficients were examined to determine which of the support variables were likely contributors to emotional distress and this was entered next. Initial analysis had identified contact with friends, resources from friends, lack of support from the spouse and uncertainty as possible predictors. Contact with friends was selected since this was most consistently related to both anxiety and depression. Also, since not every subject was married, inclusion of spouse-related stressors would have significantly reduced the number of cases used and the results may not have reflected the experience of the sample as a whole. Resources from friends was rejected because of the discrepancy between parametric and non-parametric correlation co-efficients.

Uncertainty was the final variable to be entered to give an indication of its influence on depression and anxiety after the aforementioned variables had been accounted for.

Examination of the correlation coefficients relating to

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[2] Since the two depression scores appeared to be strongly correlated ($r = 0.98$), it was decided to use only the corrected scores in the regression analysis.

functional impairment revealed that it was not closely associated with either uncertainty, social support or with fatigue. Instead, it appeared more strongly associated with three illness-related variables, namely the number of years patients had been diagnosed, cognitive difficulty and physical functioning. To assess their relative influence on functional impairment, years diagnosed was entered first while the MOS subscale measuring physical functioning was entered last.

Because the scores for functional impairment and years diagnosed were not distributed normally, the values were transformed by calculating the square root (in the former's case after reflexing them first).

All analyses were performed using SPSS/PC Regression.

Results of the multiple regression analysis

Table 10 gives the results of the analysis using anxiety as the dependent variable.

The results indicated that fatigue, contact with friends and uncertainty accounted for 41% of the variance in anxiety. Both contact with friends and uncertainty significantly added to the prediction but uncertainty had a stronger effect.

Table 11 displays the results of the analysis where the corrected depression score was the dependent variable.

It was found that lack of contact with friends contributed to the prediction of depression, as did uncertainty. However, the three variables explained just 27% of the variance. As before, uncertainty was the strongest predictor, adding significantly to the variance in depression even after the influence of two other variables had been accoun-

ted for.

Table 12 gives the results of the analysis with functional impairment (transformed) as the dependent variable.

The findings revealed that physical functioning was the strongest predictor in this equation. The three variables explained 38% of the variance in functional impairment, and each variable made a unique contribution to the equation.

Given the modest number of subjects, it is difficult to draw firm conclusions. However, in terms of both depression and anxiety, the data suggest that uncertainty was a significant predictor of emotional distress. Noteworthy is also the limited effect of fatigue, particular in relation to depression. Finally, the patient's physical limitations as measured by the MOS subscale, were found to be the strongest predictor of functional impairment.

Table 10. Results of a hierarchical multiple regression analysis with the anxiety score as the dependent variable

Variable	R^2	R^2 change	Mult. r	F change	ßeta	T
Fatigue	.09	.09	.31	5.61	.26	2.47
Contact with friends	.21	.11	.46	7.67*	-.23	-2.08
Uncertainty	.41	.20	.64	17.76**	.46	4.21**

* $p \leq .01$ ** $p \leq .001$

Table 11. Results of a hierarchical multiple regression analysis with the corrected depression score as the dependent variable

Variable	R^2	R^2 change	Mult. r	F change	ßeta	T
Fatigue	.03	.03	.18	1.84	.15	1.28
Contact with friends	.17	.14	.41	8.92*	-.29	-2.43
Uncertainty	.27	.10	.52	7.23*	.33	2.69*

* $p \leq .01$

Table 12. Results of a hierarchical multiple regression analysis with functional impairment as the dependent variable

Variable	R^2	R^2 change	Mult. r	F change	ßeta	T
Years diagnosed	.12	.12	.34	7.01*	-.25	-.22
Cognitive difficulties	.27	.15	.52	10.34*	-.27	-2.2
Physical Functioning	.38	.11	.62	9.17*	.36	3.0*

* $p \leq .01$

Relationships between variables: SCI group

As in the CFS(ME) group, there were no significant relationships between demographic variables and the other measures, except for age which was correlated with duration (r_s = .63, p<.001).

There were also a number of inter-correlations between the various disability-related variables. For instance, better physical functioning was significantly related to greater activity (r_s =.64, p <.001), lower lesions (r_s = .58, p<.001) and a lesser requirement for help (r_s = -.76, p<.001). For more detailed information, see Appendix IIB.

Of the PFRS subscales, fatigue was significantly correlated with the number of injury-related symptoms (r_s = .62 p<.001). Similarly, somatic symptoms correlated with impaired physical functioning (r_s = -.58 p<.01) and with a greater frequency of symptoms (r_s= -.65, p<.001).

The relationship between disability-related variables and adjustment

The relationship between various measures of disability, symptoms and adjustment are depicted in Table 7b.

It was found that fatigue correlated significantly with depression while cognitive difficulty correlated significantly with anxiety. Neither of these symptom subscales, however, were significantly related to functional impairment. Indeed, only somatic symptoms showed a significant correlation with this adjustment measure.

There were a number of other variables which were also strongly associated with functional impairment. For example, it was linked both to higher lesions and lower physical functioning.

Table 7b. Correlations (Spearman's Rho) between disability-related variables and adjustment: the SCI group

	Anxiety	Depression	Depression corrected	Functional impairment
Duration	-.03	-.36	-.45	.18
Lesion	.05	-.13	-.07	-.59*
Physical functioning	.08	-.29	-.14	-.77**
Fatigue	.35	.57*	.42	.37
Cognitive difficulty	.60*	.50[a]	.44[b]	.34
Somatic symptoms	.39	.41	.25	.61*

* $p \leq .01$ ** $p \leq .001$ [a] $r = .51$ [b] $r = .53$*

The relationship between uncertainty and other variables

Uncertainty and unpredictability were not related to any of the demographic variables. Moreover, there was no association between unpredictability and any of the other measures. However, there were significant correlations between uncertainty and disability-related measures e.g. the frequency of symptoms ($r_s = -.58$, $p<.01$). For further details, see Appendix IIB. The degree of uncertainty was also strongly correlated with the severity of fatigue and more modestly, with somatic symptoms.

In terms of interpersonal relationships, there was a significant positive correlation between uncertainty and support from relatives although this was not replicated when the scores were analysed using Pearson's r.

Table 8b. Correlations (Spearman's Rho) between uncertainty, social support and adjustment: the SCI group

	Anxiety	Depression	Depression corrected	Functional impairment
Uncertainty	.21	.43	.42	.22
Unpredictability	.15	.17	.19	.09
Interpersonal resources				
Friends	-.27	-.26	-.36	-.26
Spouse/partners	-.56	-.23	-.28	.06
Relatives	-.27	-.11	-.12	.04
Interpersonal stressors				
Friends	-.03	-.26	-.26	-.35
Spouse/partners	.13	-.09	-.19	-.16
Relatives	.01	-.47	-.38	-.20
Contact				
Friends	-.23	-.22	-.17	-.22
Relatives	-.28	.03	-.09	.04
Loss of friends	-.01	-.19	-.17	.52*

** $p \leq .01$ ** $p \leq .001$

Neither uncertainty nor unpredictability were related to any of the adjustment measures.

Social support

The results of the LISRES subscales revealed a number of inter-correlations between variables, showing for instance, that if patients were supported by their relatives, they were also likely to receive support from friends (see Appendix IIB for details).

As Tables 8b, 9c and 9d show, few other relationships reached significance. There were no strong correlations between the social support variables and adjustment, except for loss of friends and functional impairment. Loss of friends was also positively correlated with higher lesions and reduced physical functioning. However, in the latter case, only the nonparametric correlation reached significance.

Table 9c. Relationships between uncertainty, interpersonal resources and disability-related variables: the SCI group

	Uncertainty	Friends resources	Relatives resources	Spouse resources	Contact with friends	Contact with relatives
Duration	.19	.07	.03	.23	.04	-.21
Lesion	-.14	.01	-.18	-.08	.07	-.38
Physical functioning	-.12	-.12	-.34	-.43	.08	-.23
Fatigue	.75**	.18	.28	-.03	.13	.31
Cognitive difficulty	.51	.11	.15	-.11	.19	.12
Somatic symptoms	.64*[a]	.15	.35	.22	.10	.08
Uncertainty	1.00	.26	.58*[b]	.01	.36	.27
Unpredictability	.23	-.18	-.21	.11	-.03	.05

* $p \leq .01$ ** $p \leq .001$ [a] $r = .53$ $p = .014$ [b] $r = .49$ $p = .02$

Table 9d. Relationships between interpersonal stressors, uncertainty and disability-related variables: the SCI group

	Friends stress	Relative stress	Spouse stress	Loss of friends
Duration	.34	.50	.23	.37
Lesion	.18	.12	-.13	-.58*
Physical functioning	.15	.30	.27	-.53*[a]
Fatigue	.25	-.03	-.17	-.03
Cognitive difficulty	-.03	-.09	-.27	.08
Somatic symptoms	.10	.05	-.18	.33
Uncertainty	.27	.22	-.18	.02
Unpredictability	-.07	.14	.10	.06

[a] r = .50 p=.014

4.5 Discussion

The findings of this study revealed that the CFS(ME) patients were significantly more depressed and functionally impaired than the people with spinal cord injuries. They also reported higher levels of fatigue, more problems with memory and concentration and greater uncertainty. However, there were no significant differences between the groups in terms of anxiety and perceived social support.

Further analysis showed that lack of contact with friends, uncertainty and the severity of symptoms were significantly related to emotional distress in the patients with CFS(ME). This was not the case in the comparison group, where emo-

tional distress was more strongly associated with the severity of symptoms and level of disability.

Variables which were associated with functional impairment in the CFS(ME) group included physical functioning, cognitive difficulties and the number of years a person had been diagnosed. In the SCI group, functional impairment was also linked with disability-related variables, particularly physical functioning and somatic symptoms.

4.5.1 Adjustment to CFS(ME) and SCI

The finding of higher depression scores in the CFS(ME) patients compared to the people with spinal injuries is difficult to explain. While the two groups were matched in terms of their level of current activity, the greater severity of the fatigue and cognitive difficulties in the CFS(ME) group may have added to their depressive mood. On the other hand, the higher scores could be an indication of more extensive psychiatric morbidity among the patients with CFS(ME).

The latter is supported by the finding that there were more cases of possible clinical depression among the CFS(ME) patients than in the comparison group (55% versus 35%). A similar trend was also found for probable depression (22% versus 7%), and this was replicated, even when the fatigue-related item 'I feel as if I'm slowed down' had been removed (7% versus 0%).

Cases of possible and probable anxiety were also more common among the patients with CFS(ME). Indeed, using a cut-off point of 8, the number of cases of possible clinical disorder among this group was 68%, which not only exceeded the estimate for the people with SCI (48%) but also all previously reported estimates for CFS (e.g. Buchwald et al 1994, Lloyd et al 1990, Lynch et al 1992, Katon et al 1991,

Millon et al 1989, Pepper et al 1993, Wood et al 1994). The number of cases of probable clinical anxiety was slightly lower: 47% of the patients with CFS(ME) had a score of 11 or more on the HAD, compared with 32% of the people with SCI.

It should be noted here that the HAD is a screening tool and that a formal psychiatric interview is required to confirm the diagnosis (Thase 1991). However, comparing the results with those of studies which also used the HAD shows that the findings reported above were generally similar to the rates documented in certain medically-ill groups. For example, a study of two groups of cancer patients revealed that 40% and 30% respectively suffered from possible clinical depression. In addition, 59% and 68% respectively scored on or above the cut-off point of 8 suggesting possible clinical anxiety (Greer et al 1992). Other researchers have reported slightly lower rates among their patients (e.g. Carroll et al 1993) but this may reflect differences in disease activity and impairment, and it is therefore difficult to draw firm conclusions (Rodin et al 1991).

With regard to the patients with SCI, the levels of depression among the SCI group were similar to those of other community samples (e.g. Frank et al 1985, Malec and Neimeyer 1983, MacDonald et al 1987). However, the number of cases of possible clinical anxiety was rather higher than expected (cf. Table 1b). The raised scores may reflect more severe psychopathology or it may be due to other factors, such as the use of a rating scale (e.g. Judd et al 1989, Rodin et al 1991).

As far as functional impairment is concerned, the CFS(ME) group had significantly higher scores than the people with SCI. This suggests that CFS(ME) had a more global effect on patient's lives than SCI. It is also consistent with the reports from patients documented in Study 1.

4.5.2 Variables associated with adjustment

Emotional distress

The results from this research revealed a number of significant relationships between symptoms and emotional distress. For instance, both fatigue and somatic symptoms were significantly correlated with anxiety while cognitive difficulty was significantly related to depression. However, the latter was found only for uncorrected scores when assessed using Pearson's r (see Appendix II).

Since 75% of the CFS(ME) patients reported moderate or severe levels of fatigue on the PFRS, this symptom was not only the most common but also the most disabling complaint. However, when it was entered in the multiple regression analysis, it did not contribute significantly to the variance in either anxiety or depression. Thus while it may influence the emotional distress associated with CFS(ME) to some degree, its role may be more limited than has been suggested previously (Butler et al 1991).

Less common than fatigue were cognitive difficulties. Nevertheless, over 60% of the CFS(ME) patients rated these as moderate or severe on the PFRS. The raised scores may have resulted from co-existing depression (e.g. Deale and David 1994, Macdonald et al 1993b, Krupp et al 1994). However, studies on other samples have revealed notable differences in the cognitive deficits associated with CFS and affective disorders (Sandman et al 1993, Smith et al 1993).

While it was not possible to determine the direction of the association between cognitive difficulties and depressive mood in this study, the possibility that the former could have contributed to the latter deserves further attention. For example, problems with memory and concentration may undermine the appraisal of stressors and the planning,

selection and monitoring of appropriate responses (cf. Earll 1989, Nerenz and Leventhal 1983). Cognitive dysfunction might also lead to misunderstandings in interpersonal relationships, including those with medical professionals. Thus by interfering with the patient's problem-solving capacities, cognitive difficulties could increase emotional distress and adversely affect adjustment (Davidoff et al 1992). The significant relationship between cognitive difficulty and both anxiety and depression in the patients with SCI is consistent with this view.

Other illness-related variables which were associated with emotional distress were somatic symptoms which correlated with anxiety in the CFS(ME) group, and fatigue, which correlated with depression in the people with SCI. These findings are in line with the research on other disorders although in many studies, the reported relationship between depression and disability is somewhat stronger (e.g. Bukberg et al 1984, Carroll et al 1993, Craig et al 1994, Dalos et al 1983, Fleminger et al 1991, Stewart et al 1965).

Functional impairment

Illness and disability-related variables were also associated with the second adjustment measure: functional impairment. Indeed, it was significantly related to physical functioning in both groups. In the CFS(ME) group, it also correlated with the number of years which a patient had been diagnosed. Thus the longer this period, the more impaired the patient was in terms of ability to work etc. Likewise, in the SCI group, functional impairment was related to the level of lesion and the severity of somatic symptoms.

The comparatively weak relationship between functional impairment and fatigue in patients with CFS(ME) was unexpected. It conflicts with the cognitive behavioural model which posits that this symptom is strongly associated with

ongoing disability and distress (Butler et al 1991). Interestingly, the correlation between fatigue and functional impairment in the SCI group also failed to reach significance.

In contrast, the finding that cognitive difficulty was significantly correlated with functional impairment in the CFS(ME) group supports the view that problems with concentration and memory may have a marked impact on these particular patients' lives.

Finally, the lack of association between the HAD subscales and functional impairment in both groups underlines the independence of these measures and provides some support for the view expressed by Trieschmann (1988) that: "sadness, anger, anxiety, and hopelessness may all be apparent in varying degrees for varying periods of time but usually will not interfere with daily function to a major degree".

Uncertainty and its relationship with adjustment

So far, the role of uncertainty has not received a great deal of attention in relation to CFS(ME). However, two findings indicate that it may be a significant influence on the level of emotional distress. Firstly, the mean scores of the CFS(ME) group (66.3) exceeded those documented in many other conditions. Thus it was not only higher than that of people with spinal injuries (56.7), but it also exceeded those of patients with myocardial infarction (mean = 49), multiple sclerosis (mean = 63) and various cancers (mean = 41) (Mishel and Epstein 1990). Secondly, uncertainty was significantly related to anxiety and depression, and was a significant predictor of both when assessed using multiple regression.

Given the lack of knowledge about the aetiology and prognosis of CFS(ME), the apparent connection between uncer-

tainty and emotional distress is not surprising. However, it was interesting to note that the degree of uncertainty was not influenced by the duration of illness; patients who had been unwell for less than three years had only slightly higher uncertainty scores than those who had been ill for more than 7 years (mean 64.9 versus 63.7). This contrasts with the report by Moser et al (1993) which revealed significantly higher uncertainty scores in newly diagnosed patients with systemic lupus erythematosis (SLE).

Two other findings are of interest in relation to CFS(ME). Firstly, uncertainty was not related to functional impairment, so it did not affect the patients' ability to work or engage in leisure pursuits. Secondly, unpredictability was not linked with either anxiety or depression, suggesting that the fluctuations in the condition did not contribute to these patients' emotional distress.

Given that the majority of people with SCI had rated their condition as stable, their uncertainty scores were expected to be relatively low. However, while the mean was significantly lower than that of the CFS(ME) group, it exceeded those of a number of other disorders. The extent to which uncertainty influenced adjustment in the people with SCI is unclear. For instance, in contrast to the CFS(ME) group, there was no significant relationship between uncertainty and emotional distress.

According to Mishel's theory (1988), uncertainty may be due, in part, to the presence of fatigue and cognitive dysfunction which weaken the accuracy of appraisals. If this was the case, there should be a significant relationship between these symptoms and the MUIS score. However, while fatigue and uncertainty were related in the SCI group, the correlation co-efficients in the patients with CFS(ME) were comparatively weak. Thus the high levels of uncertainty among the latter were probably not the result of their

symptoms.

Mishel also argued that lack of familiarity with health-related events, e.g. the novelty of investigations, treatments etc may play an important role. However, since investigations of CFS(ME) patients tend to be restricted to blood tests to exclude other disorders and the number of treatments are limited, this factor is probably not the reason for the high degree of uncertainty in this patient group.

Finally, it has been suggested that uncertainty may be connected with the inadequacy of structure providers e.g. a lack of social support. The finding of a significant correlation between uncertainty and both the loss of friends and the lack of resources from relatives is consistent with this view, although of course, the direction of the relationship cannot be determined.

The complexity of the subject is further illustrated by the finding among the SCI group that higher levels of uncertainty were associated with increased support from relatives. This may reflect the paradox of social support, namely, that in some cases, help from others has negative effects. For instance, as Buunk and Hoorens (1992) have noted, support can sometimes undermine feelings of competence and control, particularly where this conflicts with values of self-reliance and independence. Alternatively, as Dakof and Taylor (1990) found, relatives may be more of a hindrance than a help if they express too much worry or pessimism.

Social support and its relationship with adjustment

One of the most interesting findings in this study was that the levels of support from partners, relatives and friends in the CFS(ME) group were similar to those recorded by people with SCI. This contrasts with the research of

Faucett and Levine (1990) which showed that patients whose illness had an uncertain aetiology received less support than those whose condition had a known organic cause. However, it may be that the severity of the illness in the CFS(ME) group influenced the support providers to a greater degree than the doubts about the origins of the disease (Skelton 1991).

Comparison with the data for other disorders is difficult, since the LISRES scale used in this study is relatively new and published reports which have included this measure are limited. Moreover, most of the studies to date have combined the scores for several variables e.g. resources from relatives have included data relating to the mother and father. Only the scores for spouse/partner resources have been reported separately and can therefore be compared to the findings reported above.

With regard to the latter, the scores of the CFS(ME) patients were found to be similar to those of community controls and patients with arthritis, but higher than those for alcoholics and psychiatric patients (Moos et al 1989). The similarity between the scores from the arthritis and CFS(ME) group is another indication that having a disorder with an ambiguous aetiology does not necessarily lead to reduced support.

On the other hand, it could be argued that the extra demands on people with CFS(ME) results in a greater need for support and that they did not receive the additional help they desired. For instance, as the findings above have shown, patients have to cope with severe problems related to fatigue and concentration as well as the uncertainty about the progress and outcome of their disease. Moreover, treatment options are limited (see Chapter 1) and there are still relatively few specialised clinics where patients can go for help and advice. Unfortunately, since the LISRES scale does

not assess the adequacy of support, it is not possible to determine whether it actually matched these patients' needs.

In terms of the relationship between the levels of social support and emotional distress, the results of the CFS(ME) group were consistent with research on other disorders, showing that greater support was associated with lower depression scores and that a lack of support was related to increased emotional distress (e.g. Littlefield et al 1990, Ray 1992, Revenson et al 1991, Rohde et al 1990, Wineman 1990)

The role of social support as a predictor of emotional distress in patients with CFS(ME) was examined in the multiple regression analysis, alongside uncertainty. It was found that although lack of contact with friends contributed to the variance in anxiety and depression over and above the influence of fatigue, it was not a unique predictor of either. Thus although having a number of close friends with whom one has regular contact may have helped to reduce emotional distress, its effect was limited. An alternative explanation, that the presence of distress led patients to avoid social contact also deserves consideration. However, if this were the case, one might have expected a similar relationship to exist in the SCI group, or between distress and contact with relatives, and this was not observed.

In addition to the research relating to the quality of support, this study also assessed the different sources of support. One finding was that the resources from spouses and partners exceeded that from friends and relatives in both groups. Since partners are often the primary providers of social support, and the levels in this case were relatively high, it was hypothesized that married patients with CFS(ME) might be less distressed than single ones. However, inspection of the data revealed that this was not the case. The mean depression scores for married and single patients

were 8.89 and 7.46 respectively. Similarly, the means for anxiety were 10.22 and 8.96 respectively. Thus the single patients reported slightly less distress than the married ones.

Nevertheless, the source of support does appear to play a role in terms of adjustment to CFS(ME). For example, contact with friends was negatively and significantly correlated with depression but contact with relatives was not. Indeed, the strength of the correlation between contact with relatives and emotional distress was extremely low.

A separate issue is whether the quality of support has a stronger relationship with mood that the quantity of support (Cohen and Wills 1985, Fitzpatrick et al 1991, Goodenow et al 1990). Unfortunately, since this study only included three measures assessing the 'quantity' of support, it is difficult to draw firm conclusions. However, it is noticeable that the correlation between the resources from relatives and mood exceeded that between contact with relatives and mood. The discrepancy was far less marked for these variables where it concerned friends, showing once more that the connections between social support and either depression or anxiety depends in part on the source of the support and that a simple comparison between structural and functional measures could be misleading. The reason for the lack of association between the various social support variables and emotional adjustment in the SCI group is unknown.

Aside from its links with emotional distress, social support was also associated with the second adjustment measure: functional impairment. However, due to the design of the study, it is not possible to determine whether the loss of friends contributed to functional impairment or whether the latter led to the loss of friends.

Finally, the loss of friends was related to certain illness

and disability-related variables. In the patients with CFS(ME), it was significantly correlated with the years since diagnosis, while in the SCI group, loss of friends was associated with the level of the lesion and degree of physical functioning. Thus the more severe the disability, the greater the loss of friends.

In summary, the findings suggest that variables such as cognitive difficulties, uncertainty and lack of social support may contribute to the emotional distress associated with CFS(ME). Consequently, any analysis of the patients' experience which does not take these possible influences into account, may be incomplete.

Further research is required, not only to examine the effects of variables such as uncertainty, support and cognitive dysfunction in more detail, but also to assess the value of counselling in which these problems are acknowledged and addressed.

4.5.3 Methodological issues

The results of this study should be interpreted with caution for a number of reasons. First of all, the symptoms of CFS(ME) tend to fluctuate markedly, from hour to hour and from week to week (e.g. Gilliam 1938, Patarca et al 1993). Thus a patient may feel reasonably well one day but severely ill 24 hours later. In this sample, only 17% described their condition as stable, compared with 14.4% of the patients surveyed by Hinds and McCluskey (1993) and 25% of those examined by Dowsett et al (1990). This contrasts with the results from the people with SCI, of whom 64% rated their condition as stable. Although the changeability of CFS(ME) makes it difficult to obtain a truly representative assessment of disability, it is noteworthy that the scores for all the symptom subscales were similar to those of another group with CFS, and that as expected, they exceeded those from a

student comparison group (Ray et al 1992b).

A second point which should be taken into account when evaluating the data is the limited number of variables assessed. Since most people with CFS(ME) are very ill and they find it difficult to concentrate for long periods of time, it was decided to restrict the number of questions asked. However, this meant that factors which may have been a major influence on the mood and functioning of the patients could have been excluded. For instance, possible mediators of emotional distress which were not assessed in this study include coping strategies (e.g. Blakely et al 1991, Ehmann et al 1990, Lazarus 1991, Weisman and Worden 1976, Ray et al 1993), personality variables such as self-efficacy (e.g. Terry 1992, Lorig et al 1989), satisfaction with medical advice and care (Dakof and Taylor 1990, Stewart and Sullivan 1982, Toombs 1992), and membership of a support group (Dimond 1983, Robinson 1988). Any of these factors could have influenced the severity of anxiety and depression, as well the experience of the illness as a whole, and this deserves further research.

A third problem which should be considered when interpreting the results is that some of the symptoms reported as troublesome by the CFS(ME) patients were not included in the PFRS. One of these was nausea which was a main symptom in 19% of the group. It is therefore possible that the lack of association between somatic symptoms and functional impairment could have been due, in part, to the fact that important complaints were not assessed. It would also have been useful to obtain an outsider's rating for symptoms and level of disability, but this would have put an extra burden on the patients and was therefore not pursued.

Unfortunately, the size of the SCI group in the current study limits the conclusions which can be drawn in relation to spinal injuries. For instance, it was not always pos-

sible to compare variables, since in some cases (e.g. gender), there were too few people in one of the groups to make such a comparison meaningful. However, it should be pointed out that in terms of demographic and disability-related characteristics, the sample was not atypical. For instance, as in other studies, about a half the patients were injured in road traffic accidents, men greatly outnumbered women and most were not employed (cf. Grundy et al 1986, Oliver et al 1988, Trieschmann 1989). On the other hand, this was a community sample and unlike many of the previous studies assessing the emotional distress associated with SCI, the patients were not in a hospital environment which might have protected them from experiencing higher levels of uncertainty and anxiety.

4.5.4 Questions arising from the research

Although uncertainty appears to be a significant problem in CFS(ME), it is not clear whether it has a direct relationship with anxiety and depression or whether it is mediated through its effects on coping. For example, it has been suggested that patients can deal with the lack of certainty associated with disease by obtaining relevant information and by finding medical help or advice. Although all the patients in this study had access to a consultant and information, there was no assessment of the amount of contact with the practitioner, or their use of information.

A further source of knowledge, contact with others who have faced the same problems, was not assessed either. Given the lack of understanding among the general population (see Study 1), and the inadequacies of the services provided by the medical profession (Denz-Penhey and Murdoch 1993), advice and support from patient groups may do much to reduce the levels of uncertainty and emotional distress (Dimond 1983). Consequently, the role of patient groups deserves further attention.

Aside from the lack of advice and support, people with CFS(ME) may also be disadvantaged by other problems. For example, their attempts at coping could be undermined by high levels of fatigue or cognitive dysfunction (Lazarus 1991). In this study, fatigue was common and often severe but this symptom did not have as close a relationship with functional impairment as cognitive difficulties did. However, this does not rule out the possibility that this symptom could interfere with coping and other aspects of adjustment.

Also of interest in this respect are the high levels of anxiety. Although the latter may be a result of the somatic symptoms and the degree of uncertainty, it is also possible that the reverse is true, i.e. that the symptoms and the uncertainty are actually caused by the patients' anxiety. If that is the case, it follows that emotional distress could play a major role in perpetuating chronic fatigue syndrome (Wessely et al 1991, Sharpe 1994).

Unfortunately, the cross-sectional design of this study means that the direction of the relationship between variables can not be determined. However, since just over thirty per cent of the patients did not have high anxiety scores, and given that most studies have not reported similar rates of anxiety disorder in patients with CFS, it is more likely that the present sample was unusual, representing perhaps, individuals who had not yet received the help they required, and were not able to manage their illness in an effective way. It would be useful, therefore, to repeat the study in a second sample of patients with CFS using a longitudinal design to allow a more detailed evaluation of the relationship between fatigue, cognitive dysfunction, uncertainty and emotional distress.

A longitudinal design would also help to determine if and how symptoms change as a result of regular contact with and

support from a medical practitioner, and whether specific coping strategies have a positive effect on emotional distress and functional impairment.

A further question which should be addressed is the relationship between fatigue and depression. In this study, the correlation between these variables was relatively low in the CFS(ME) group but modest positive correlations between fatigue and psychiatric symptoms have been found in other groups of CFS patients (McDonald et al 1993, Ray et al 1992b), as well as people with Parkinson's disease (Friedman and Friedman (1993) and systemic lupus erythematosus (Krupp et al 1990). However, fatigue was not correlated with depression in patients with multiple sclerosis (Krupp et al 1988, Moller et al 1994) or post-polio fatigue (Bruno et al 1994b), and did not contribute to the depression reported by patients with rheumatoid arthritis (Belza et al 1993). Given these contradictory findings, further studies are required to clarify the situation.

4.6 Summary

In this study, patients with CFS(ME) reported more severe fatigue and cognitive difficulty, and they were more impaired in terms of work and other activities than people with spinal cord injuries. The CFS(ME) group also recorded higher levels of depression, although when one fatigue-related item was omitted, the difference between the groups was no longer significant.

The results also suggest that uncertainty may play an important role in both anxiety and depression, and this possibility should be taken into account both in research and in clinical practice.

In contrast, most of the social support measures showed only a limited relationship with emotional distress. However,

contact with friends was significantly associated with lower anxiety and depression scores.

Overall, these findings indicate that factors such as the severity of symptoms, uncertainty and the lack of support may add to the patients' emotional distress and therefore undermine both their adjustment and recovery.

CHAPTER 5

Learning to cope with post-infectious fatigue syndrome A follow-up study

5.1 Introduction

The previous studies have shown that many patients with CFS are severely impaired and that many experience high levels of emotional distress. However, while drugs can help to alleviate specific symptoms such as depression and pain, there is still no treatment which has led to consistent improvements in the illness as a whole.

Given the limited value of prescribable drugs, some doctors have begun to explore other ways of managing the disability and distress associated with CFS. The approach which has received most attention in this respect is cognitive-behavioural therapy (CBT).

CBT is based on the assumption that a person's thoughts and beliefs influence their behaviour as well as their emotional and physiological state. Even in disorders with an underlying organic cause, the presence of faulty or irrational thoughts may undermine effective coping, impede recovery and significantly increase emotional distress. The main task in CBT is therefore to identify and challenge maladaptive cognitions such as distortions, overgeneralisations and all-or-nothing thinking (Rimm and Masters 1979). Patients are also encouraged to engage in adaptive tasks and activities which were previously avoided.

In dealing with CFS, therapists try to challenge any dysfunctional attributions which may have led patients to avoid exertion or to adopt other strategies which might have perpetuated their distress (see Chapter 1). At the same time, patients are instructed to gradually increase their

activities and not to be afraid of the fatigue which may ensue (Butler et al 1991, Sharpe 1993).

It is claimed that programmes featuring 'graded exercise' improve the individual's physical fitness and accordingly, their fatigue. Moreover, by engaging in planned activities, it is hoped that patients will regain their sense of mastery and thus overcome their feelings of helplessness and hopelessness (Sharpe 1994).

There are currently five published accounts of studies which have assessed the effectiveness of CBT in CFS. Two were uncontrolled trials (Butler et al 1991, Cox and Findley 1994) and three were controlled (Lloyd et al 1993, Friedberg and Krupp 1994, Sharpe et al 1996).

As shown in Table 1, the results of the uncontrolled trials were encouraging. However, the findings from the controlled trial have been less consistent. For instance, one of the controlled trials which combined CBT and planned, graded exercise found that this combination was not superior to clinic attendance or a placebo drug (Lloyd et al 1994). In a second study, the treated patients showed significantly greater improvements than the controls on the Karnofsky scale but there were no significant differences between the groups on the other measures (Sharpe et al 1996).

The third controlled trial differed from the two above in that the programme involved a combination of CBT, group therapy, relaxation training and "shared coping" (Friedberg and Krupp 1994). Furthermore, the researchers advocated that patients should stay within the limits imposed by the illness rather than increase their activity according a schedule. They rejected graduated increases in exercise because they found no evidence of phobic avoidance among their patients and many were thought to be performing near or at their activity ceiling already. While the results

revealed no significant differences between the treated patients and controls, improvements were noted in the subgroup with above average depression scores.

Table 1. Studies assessing psychological therapies for CFS

Treatments (number of subjects) Reference	Criteria	Results
1. *Uncontrolled trials*		
CBT, graded exercise plus anti-depressants where appropriate (N=50). Butler et al 1991.	Own	27 completed treatment. Significant improvement noted in fatigue, functional impairment, depression, somatic symp - toms. 70% of those with some treatment felt better or much better.
As above. Follow-up at four years (N=46). Bonner et al 1994.		87% who completed treat ment (total 23) had improved. 13% of those who did not complete treatment also improved.
CBT, graded exercise plus anti-depressants (N=28). Cox and Findley 1994.	CDC	57% had increased activity levels six months after discharge. 14.3% noted reduced symptoms, 35.7% were unchanged, 7% were worse.

Table 1 cont.

Treatments (number of subjects). Reference.	Criteria	Results
2. *Controlled trials*		
CBT, graded exercise, immune therapy (IT) (N=20) versus CBT, graded exercise, placebo (N=21), versus IT clinic attendance (N=26) versus placebo, clinic attendance (N=23). Lloyd et al 1993.	Aus*	No sign. difference between groups. Trend towards greater improvement in quality-of-life scores among the CBT+IT group but no improvement in their POMS or activity scores.
CBT, graded exercise (N=30) versus diagnosis and advice to increase activity (N=30). Sharpe et al 1996.	Oxford	No significant differences between the groups after completion of trial but treated patients showed greater improvements (e.g. in functioning) at follow-up.
CBT, gentle exercise, relaxation and shared coping. CFS (N=22) versus untreated CFS (N=22) versus depressed group (N=20). Friedberg and Krupp 1994.	CDC	No significant improvement in CFS groups. However, CFS patients with higher depression reported less fatigue, stress and depression after the trial.

* Aus = Australian

Since the number of studies evaluating CBT is limited, it is difficult to ascertain its effectiveness for CFS at the present time. However, there is evidence that this treatment may be beneficial for specific subgroups, for instance, patients with concurrent clinical depression (Friedberg and Krupp 1994, Butler et al 1991). Indeed, the fact that 20 of the 32 CFS patients (63%) tested by Butler et al were diagnosed with major depression may partly explain the discrepancy between their results and some of the other trials. Moreover, the finding that CBT appears to be more effective in patients with co-existing depression is consistent with the results from other medically-ill populations (e.g. Larcombe and Wilson 1984).

CBT and graded exercise may also be helpful in patients who are overly anxious about activity and whose fatigue may be attributed almost entirely to physical deconditioning, demoralization and low mood (Butler et al 1991). However, it is still unclear just how many patients with CFS actually fit this stereotype. For instance, Faas (1992) has challenged the descriptions of people with CFS as fearful of activity and passive. Like Friedberg and Krupp (1994), her patients had a tendency to do too much rather than too little. She also noted that those individuals who had been diagnosed early and who had followed advice to rest had generally improved quickly, and had been able to return to something resembling their old levels of activity within a relatively short period of time.

Given the equivocal results of the controlled trials, it could be argued that the current emphasis on challenging somatic attributions and reducing the levels of deconditioning may not address all the sources of disability and distress in this patient group. For example, the model which most therapists use as a basis for CBT assumes that there is no underlying disease process which can cause a recrudescence or exacerbation of symptoms. Indeed, few

accounts have considered the possibility that some complaints may not be attributable to deconditioning and depression, and that therefore the current advice to increase activity irrespective of one's state of health may not always be appropriate.

Another possible limitation of CBT is that the therapists' views regarding the aetiology of the symptoms and the role of psychiatric morbidity have tended to conflict with the beliefs and experiences of many people with CFS. The lack of agreement may undermine the patient-therapist relationship and lead to high attrition rates (Hickie et al 1995b).

An alternative approach resembles the broad-based programmes which have been devised for patients with medical conditions such as cancer. These do not aim to treat the underlying causes of the illness, but try to enhance the patients' psychological well-being and reduce their disability. Patients can be treated individually or in groups but all the programmes tend to include the following components:
- education about the condition,
- stress management,
- enhancement of coping skills to deal with the disorder and its effects,
- provision of emotional support.

For example, Fawzy et al (1990b) devised a 6-week, structured, group-based programme for postsurgical patients with malignant melanoma. It contained a number of elements including information (e.g. about cancer prevention); psychological support; stress-management (e.g. relaxation training) as well as a discussion of effective coping strategies to deal with personal difficulties such as isolation, fear, change of body image and general mood.

After 6-months, the treated patients recorded significantly lower scores for depression, fatigue and total mood distur-

bance than the no-treatment controls. The strategies which were associated with successful outcome included talking to others for information and support, and distraction (e.g. going out, doing something for oneself). Coping modes which correlated with increased psychological distress included passive resignation, taking drugs or avoiding others.

Other broad-based programmes offering support and information to patients with chronic disorders have also been found to improve mood and quality-of-life (e.g. Cunningham et al 1993). Indeed, one increased the survival rate of patients with cancer (Spiegel et al 1989).

5.1.1 Are broad-based programmes helpful for patients with CFS?

There is evidence that people with CFS would welcome programmes such as those described above. For instance, patients surveyed by Denz-Penhey and Murdoch (1993) identified a number of areas which "needed to be worked on" e.g. recognition of factors which trigger relapses, problems with managing stress, difficulties in relationships and the development of communication skills. While one-to-one counselling was rejected on the basis of cost and the perceived judgmental attitudes of some counsellors, the patients indicated that affordable, safe and effective counselling would be very helpful.

Whether such an approach might be effective in terms of reducing symptomatology is difficult to predict. However, from a theoretical point of view, a broad-based management programme could help to alleviate the psychological distress and possibly limit the disability associated with CFS as follows:

1. information about the nature of the illness should increase the patient's understanding of their condition, en-

abling them to interpret symptoms correctly and to identify factors which trigger exacerbations. This in turn could help to reduce the uncertainty and unpredictability associated CFS. Since uncertainty has been linked to both anxiety and depression in patients with CFS(ME) (see Chapter 4), this approach might reduce the severity of psychological distress. Information may also counter the helplessness and demoralization which undermine effective coping (Braden 1990) and predispose some patients to depression (Butler et al 1991).

2. Knowledge about effective coping strategies and available resources could help the individual to increase their sense of mastery and personal control. Lack of control has been linked with increased emotional distress in people with multiple sclerosis and spinal cord injuries (Devins et al 1993c, Schulz and Decker 1985).

3. Physicians can provide much needed emotional support as well as advice (Mishel 1988, Mishel and Braden 1988). Indeed, help and guidance from physicians has been previously associated with reduced psychological distress (Elliott et al 1992).

One of the few broad-based treatment programmes which is available to CFS patients in the UK is that devised by Dr. Ho-Yen (1990, 1993). His approach acknowledges the possibility of ongoing disease and the fluctuating nature of the symptoms. It also recognises the importance of information in order to gain control and the distress caused by factors such as the lack of understanding.

His 5-step management programme includes:

1. Advice aimed at limiting and preventing psychological problems. Antidepressants are used in low-doses where appropriate.

1. Information. Patients are encouraged to learn more about their illness so that they can understand their disorder and identify the triggers of relapses. To help them, he has written a book about post-viral syndromes (Ho-Yen 1993). Patients are also encouraged to learn about themselves e.g. what might have predisposed them to become ill, and to examine their reactions to the illness e.g. to recognise and confront fears etc.

3. Regular assessment of the illness and the patient's feelings using a daily diary. This lists the hours of relaxation and the patient's activities, quality of sleep, problems and mood and is used by the patient and consultant to identify variables associated with relapse and improvement.

4. Advice about energy and exercise. For instance, using the diary, patients can estimate how much activity can be carried out without causing symptoms. This strategy of identifying limits and adjusting activity according to available energy levels is sometimes known as pacing. Dr. Ho-Yen advocates gentle exercise (which does not result in an exacerbation of symptoms), as opposed to graded exercise (where activity is increased regardless of the consequences). Sleep is encouraged, as is relaxation, to increase the patient's available energy reserves. Advice regarding energy also includes a discussion of ways to deal with mental exertion, stress and difficult relationships.

5. Advice regarding food and diet. This includes a discussion of the Hay system, identification of allergies (which he regards as very rare) and limiting alcohol intake. Specific advice about diet is usually not given until 6 months after the first consultation.

Patients are seen every two months to discuss their progress and any problems which arise.

Given the emphasis on specific self-management practices e.g. keeping a diary, pacing of activity, avoiding stressful relationships etc, the effectiveness of the programme may be influenced by the patient's perceived self-efficacy, i.e. their belief that they can do the tasks recommended for them. For example, in a trial of patients with arthritis, increased levels of self-efficacy was associated with reduced pain and depression (Lorig et al 1989). However, there has been little research into the self-efficacy of patients with CFS and it has not been assessed in the trials of CBT.

It may also be of interest to compare the different approaches to exercise. Dr. Ho-Yen's advice to conserve energy is consistent with the coping strategy referred to as 'accommodating to the illness' (Ray et al 1993). In a recent study on patients with CFS, this strategy was associated with lower scores for anxiety but with greater functional impairment. In contrast, the strategy of 'maintaining activity', which Dr. Ho-Yen's programme discourages, was linked with increased anxiety but with less functional impairment (Ray et al 1993). These findings suggest that advice to keep within one's limits may be enhance emotional well-being, while other approaches such as graded activity may be more helpful to improve general functioning (cf. Sharpe et al 1996).

Finally, it has been suggested that different strategies may have different effects depending on the duration of illness. For instance, Ray et al (1995) found that 'accommodating to the illness' was related to increased fatigue in those who had been ill longer but not in those who had been ill for a shorter period. If this is correct, then Dr. Ho-Yen's programme may be more helpful for those who have not been ill for very long.

5.2 Research aims

1. The main aim of the study was to ascertain the effectiveness of Dr. Ho-Yen's programme in the management of patients with post-infectious CFS. Outcome measures were the symptom scores, anxiety, depression and functional impairment.

2. The data were also examined to assess the effect of the programme on uncertainty, perceived self-efficacy and type of coping strategies.

3. A further analysis was performed to establish whether the programme had differential effects in particular subgroups, for instance, patients with high scores for depression, anxiety and fatigue, or people who had been ill for a shorter period of time.

4. The baseline scores for all participants were examined in order to assess whether the relationships between symptoms, uncertainty, emotional distress and functional impairment, which were documented in patients with CFS(ME) in Study 2, were replicated in this sample. An additional variable, membership of a self-help group, was included to ascertain its association with emotional distress, disability and coping.

5.3 Method

5.3.1 Sample characteristics and procedure

The patients were recruited from the waiting list of Dr. Ho-Yen, a consultant microbiologist at the Raigmore Hospital in Inverness. They were contacted personally by Dr. Ho-Yen to ask if they would participate in the research. Those who were willing in principle were then sent a letter explaining the trial and asking for their consent. Patients were subse-

quently assigned to one of two groups depending on their position on the waiting list.

The treatment group comprised patients who had been on the waiting list for one to six months. They were sent their first set of questionnaires two weeks prior to their first consultation and the second set between five and six months later, prior to their fourth appointment.

The control group comprised patients who had been on the waiting list for one month or less. They were sent the first set of questionnaires immediately following receipt of the consent form and they completed the second set just prior to their first consultation, approximately six months later.

This design was chosen because at the time of the study, the delay between acceptance on the waiting list and the first consultation was about 7 months. This was sufficient to carry out two assessments with a time interval equivalent to that of the treatment group. Thus the waiting list controls provided an estimate of the effect of time, while the assessments of the treatment group allowed for a before/after treatment comparison.

An alternative design using randomised groups was rejected because this would have required half the patients to spend a further 6 months without diagnosis and treatment, which was considered inappropriate given the severity of the symptoms.

At the first consultation, Dr. Ho-Yen checked the patient's diagnosis and explained his programme (see description above). This lasted about one hour. The patients returned every two months for a further 5-10 minutes in which their progress could be checked and specific problems discussed.

	Waiting list	Before Time 1	Before Time 2
Months since joining waiting list (approx.)	0	6	12
Group 1 (treatment)		T1	T2
Group 2 (waiting list controls)	T1	T2	

T1-2 Time 1 and 2 (assessments)

Figure 1. Summary of study design

Post-infectious fatigue syndrome (PIFS) was diagnosed using the criteria formulated by Dr. Ho-Yen (1990, see Appendix 1). Most patients were thought to have a post-viral syndrome but a few patients whose fatigue followed bacterial and parasitic infections e.g. Lyme disease, were also included. Accordingly, the disorder will henceforth be referred to as CFS(PIFS).

Twenty-five patients were initially entered into treatment group and 27 became waiting list controls. In total, eight patients were excluded from the analysis; three did not have CFS(PIFS), two did not wish to continue treatment and the questionnaire from one patient was lost in the post. Another patient was excluded because she stopped taking oral contraceptives which led to a marked improvement in all symptoms while on the waiting list. This suggests that the severity

of her complaints at baseline may have been influenced by the use of this drug and that the change in scores at Time 2 cannot be interpreted as reflecting the natural course of CFS(PIFS). Finally, one patient was followed-up after only three months and this was not considered to be long enough to evaluate the effect of the treatment satisfactorily. His data were therefore also excluded from analysis. As a result, there were 22 patients in each of the groups.

Only one person refused to participate in this study. Moreover, once the trial had begun, the drop-out rate was extremely low.

None of the participants had a concurrent condition which could have had a significant influence on the assessment of outcome. However, 8 (36.4%) patients in the treatment group and 15 (68%) of the waiting list controls reported having an additional illness. These included asthma, epilepsy, localised arthritis, ulcers, diverticulitis, hiatus hernia, recurrent sinusitis and kidney infections. It was assumed that the patients were taking the medications prescribed for these conditions and that at the time of testing, their presence was not considered a major influence on the symptoms reported.

Between the first and second assessments, 15 (68%) of the treatment group changed their diet or began new therapies for their CFS-related symptoms and 14 considered these as helpful. Two began taking antidepressants. Similarly, 12 (55%) of the controls changed their diet or began a new treatment and 2 were prescribed antidepressants. The new treatments were considered helpful by 7. The change towards a healthier diet and use of anti-depressants for CFS etc is consistent with Dr. Ho-Yen's programme, although in many cases, the 'treatments' were prescribed by another advisor (acupuncturist, GP). Since the number in each group who were taking antidepressants at some stage during the trial

was similar (9 of the treated patients versus 6 of the controls), the treatment group's scores at Time 2 can still be interpreted as reflecting the general recommendations incorporated in the programme.

None of the patients were paid for their participation.

5.3.2 Details of the questionnaires

First assessment

Each set of questionnaires comprised the Profile of Fatigue-Related Symptoms (Ray et al 1992a), the Functional Impairment Scale (Marks 1986), the Hospital Anxiety and Depression Scale (Zigmond and Snaith 1983) and the 23-item Mishel Uncertainty in Illness Scale-Community Form (Mishel and Epstein 1990). Details relating to these can be found in Chapter 4.

In addition, patients were asked to complete the following questionnaires:

Background Information

This questionnaire asked for demographic information including gender, age, years of education beyond 16, occupation, marital status and housing. It also included an item to determine whether income consisted entirely of social security benefits ('Low Income').

General Information

This questionnaire asked patients about the main symptoms experienced, the duration and onset of the illness (i.e. acute/gradual), the use of drugs in the present and past, membership of self-help groups and presence of other medical conditions. A question was also included to assess whether patients had already read Dr. Ho-Yen's book and followed his advice, as this could affect baseline measures for both

groups.

Two further items assessed the level of activity and frequency of symptoms. These are described in more detail in Chapter 4. However, in contrast to the previous study, the scoring was reversed so that high scores now reflected less favourable health status. A third question asked patients about the course of their illness during the past 6 months. The choice of answers ranged in order from 'worsened a lot' (scored 1) to 'improved a lot' (scored 5).

The Illness Management Questionnaire (IMQ)

This questionnaire assessed problem-focused coping strategies and was specifically designed for use by people with CFS (Ray et al 1993). Patients were asked to describe their approach to the illness in the last six months on a scale from 1 ('never') to 6 ('always'). The scores reported consist of the means for each subscale.

The scale comprises four factors which have been interpreted as follows:

1. Maintaining activity: attempting to ignore symptoms; carrying on even though unwell and disregarding possible adverse effects of activity.
2. Accommodating to the illness: organizing and planning one's life in order to avoid over-exertion and control stress. Accepting limitations.
3. Focusing on symptoms: a preoccupation with symptoms, linked with an appraisal of helplessness and of one's life as dominated by the illness.
4. Information-seeking: seeking relevant information and a readiness to try remedies.

Ray et al (1993) reported that alpha reliabilities for these scales were high, ranging from .85 to .93.

Self-efficacy Scale

This measure comprised a modified version of the Self-

Efficacy Other Symptoms subscale (Lorig et al 1989). Patients were asked to rate their confidence regarding their ability to control their illness on a scale ranging from 10 ('very uncertain') to 100 ('very certain'). The questions included: 'How certain are you that you can control your fatigue' and 'How certain are you that you can deal with the frustration of your illness'. The score was the mean for all six items.

Since this measure was originally devised for patients with arthritis, references to 'arthritis' were changed to 'fatigue', 'illness' or 'PVFS', depending on the context. (PVFS is the name used by Dr. Ho-Yen in his clinical practice.) Furthermore, in one question, a reference to 'feeling blue' was changed to 'feeling down'.

The alpha co-efficient for internal reliability in the original study was found to be .87 and the test-retest reliability was satisfactory (Lorig et al 1989).

Patients were informed by letter that their answers would be confidential. Indeed, the questionnaires were coded by number and access to the data was limited to the researcher and consultant.

Second assessment

At the second assessment, patients were again asked to fill in the PFRS, IMQ, Self-Efficacy Other Symptoms subscale, HAD, Functional Impairment Scale and MUIS-Form C. In addition, there were questions to determine if the patients had begun any new treatments since the last assessment, and if there had been any change in their state of health. Answers to the latter ranged from 'much better' (scored 1) to 'much worse than before' (scored 5).

Further items assessed the level of activity and frequency

of symptoms. There was also an open-ended question asking patients who had improved to identify the reasons for this. A final question asked participants to rate the medical care and support which they had received for this illness from doctors other than Dr. Ho-Yen. Scores ranged from 1 ('very poor') to 5 ('very good').

Copies of all the questionnaires can be found in Appendix I.

5.4 Results

The results will be presented in two parts. The first section focuses on the differences between the groups and the changes associated with the treatment in relation to the outcome variables, plus uncertainty and self-efficacy. This will be followed in part 2 by an analysis of the relationships between these variables using the Time 1 scores from both groups.

Due to the overlap between the PFRS subscale for emotional distress and the two HAD subscales, only the latter was included in the analyses.

5.4.1 Statistical analysis

Analysis of the MUIS Scale revealed that many patients had found it difficult to complete all the items. Since low scores in these cases might be wrongly interpreted as indicating low levels of uncertainty, questionnaires with more than four missing items were discarded. As a result, Time 1 data was available for only 13 patients in the treatment group and 16 controls.

Categorical data from the two groups were compared using the Chi-square test with Yates correction for two-by-two comparisons (Siegel 1956). Data on an ordinal scale, and interval data whose scores deviated from the normal distribution,

were analysed using the Mann-Whitney U test (Cramer 1994). Other group comparisons were evaluated using a T-test for independent samples.

The effect of the treatment on the outcome variables was assessed using analysis of covariance (ANCOVA). This was considered more appropriate than a test of change scores (Time 1 minus Time 2) or MANOVA with repeated measures because the participants were not randomly selected and it was therefore not possible to control for potential bias in the composition of these groups such as differences in baseline scores. The results were checked to insure that they satisfied the conditions necessary for use of ANCOVAs.

Initial analysis of the data from the treatment group revealed the presence of outliers among the Time 2 scores for anxiety, depression, self-efficacy and uncertainty. Since these may lead to errors when calculating ANCOVAs, it was decided to reduce their effect by giving them a value one unit larger than the next most extreme score as recommended by Tabachnick and Fidell (1983). This strategy preserves the deviancy of a case but reduces the chance of misleading results. The transformation made little change to the results relating to self-efficacy so the original data were retained.

Where the group differences using transformed data reached significance, the analysis was repeated but omitting the outlier altogether. Details of these calculations can be found in Table 3, Appendix III.

Failure of linearity in the fatigue scores led to a square root transformation of the values. However, given the similarity of the results (see Table 3, Appendix III), the original data were retained.

All equations were computed using the ANOVA command in

SPSS.PC. The MANOVA command was used to obtain F values and to check for homogeneity of variance and regression. The alpha level was set at 0.05 and all tests were two-tailed.

The internal consistency of the measures which had not been used in earlier studies was assessed using Cronbach's alpha. The results, shown below, indicate that the self-efficacy scale and all four IMQ subscales had an acceptable level of internal reliability.

Table 1. Summary of Alpha coefficients for reliability

Variable	Treatment group	Control group
IMQ		
Maintaining activity	.92	.92
Accommodating to the illness	.90	.89
Focusing on symptoms	.79	.90
Seeking information	.83	.84
Self-efficacy		
Six items	.83	.86

5.4.2 Demographic information

The mean age of the treatment group was 39.6 years (SD 13.40), the youngest participant being 15. Seventy-three per cent of the patients were female, 59% were married and 50% had completed secondary school. At entry to the study,

9% were still studying, 27% were on sick leave, 5% had retired and 36% were doing either part-time or full-time work. Eighty-six per cent of those who responded to the question reported that they had changed their job or reduced their hours because of their illness. Only 14% were totally dependent on social security benefits. For more detailed information, see Appendix III.

The mean age of the waiting list controls was 37.7 years, the youngest being 14. In this group, 59% were female and 50% were married. At the time of the study, 59% had completed secondary school, 14% were still studying, 41% were on sick leave, 9% had retired and just 18% were in part-time or full-time work.

There were no major differences between the groups in terms of age, marital status, years spent in education after the age of 16, house ownership and dependence on benefits. Furthermore, the proportion of patients classified as professional or semiprofessional (e.g. teachers, nurses) were exactly the same. However, the control group contained more people in unskilled manual jobs ($p>.05$). It also included a slightly greater number of men and more patients who were on sick leave.

To determine the possible effects of the difference in gender, the scores for the key variables at Time 1 were compared using the Mann-Whitney U Test. This showed that men were significantly more depressed than women ($p<.05$).

5.4.3 Information about the illness variables

The findings relating to the illness variables are summarised below. None of the differences between the groups reached significance.

The median duration of illness among the treatment group was

5 years (SD 3.69) with a range from 6 months to 14 years. The controls had generally been ill for a shorter time (median 2.1 years, SD 3.34, range 8 months to 15 years, p=.06).

In 40% of the patients from treatment group, the symptoms had begun suddenly following an infectious illness such as glandular fever or influenza. During the six months prior to the study, 36% had been getting worse, 23% were stable and 41% had been improving. Among the controls, 63% reported a sudden onset and in 74% of the cases, the trigger was an infectious condition. Fifty per cent had been getting worse in the 6 months prior to the study, 18% felt the same and 32% noted some improvement.

Both groups were severely impaired. In terms of activities, just 4.5% of the treatment group and none of the controls were able to do more than a half of what they could do prior to their illness. Indeed, 86% of the former and 95.5% of the controls experienced symptoms most or all of the time.

Ten (45.5%) patients in the treatment group and 12 (54.5%) of the controls were taking drugs at entry to the study. In some cases, these formed part of the management for other disorders. However, up to a third were using drugs often prescribed for CFS.

The majority of the patients in both groups had not joined any patient group. However, 41% of the treatment group and 50% of the controls had read Dr. Ho-Yen's book on PVFS. Moreover, 55% of the former and 36% of the latter were following his advice most or all of the time. This difference was not statistically significant. For further details relating to the illness, see Appendix III.

Finally, patients were asked what they thought of the medical care and support from doctors other than Dr. Ho-Yen.

Twenty-seven per cent of the treatment group rated it as poor or very poor, 23% felt that it was adequate but 50% classed it as good or very good. Among the controls, 18% felt that the treatment to date had been poor, 27% viewed it as adequate but 55% rated it as good or very good.

Results for the key variables at Time 1

The baseline scores for the somatic symptoms, anxiety, impairment, self-efficacy, uncertainty and coping revealed no significant differences between the groups (see Tables 2, 3 and 4). However, the controls reported slightly more severe fatigue, cognitive difficulty and depression at Time 1 than the treatment group.

5.4.4 Changes associated with treatment

Asked about the changes in their condition during the six months between Time 1 and Time 2, 82% of the treatment group rated themselves as better or much better, 9% regarded themselves as unchanged and 9% felt worse. There were also increases in the level of activity, with 55% able to do half or more compared with the past. At Time 2, five patients (23%) had improved to such an extent that further treatment was thought unnecessary.

In contrast, 50% of the controls felt better overall, 32% perceived no change and 18% were worse or much worse than before. However, 41% could do half or more compared to what they used to be able to do. Although the improvements were greater in the treatment group, analysis using the Mann-Whitney test showed that their ratings of change were not significantly greater than those reported by the controls.

Illness-related variables

The differences associated with treatment relating to the

three symptom subscales is shown in Table 2. Analysis of covariance, controlling for baseline scores, revealed that there were no major group differences in the scores for cognitive difficulties at Time 2. However, there was a significant difference for both fatigue, F (1,40) = 5.13, p=.03 and somatic symptoms, F (1,40) = 4.66, p=.04.

Table 2. Means (and SD) of illness-related measures

	Treatment group		Control group		
	Time 1	Time 2	Time 1	Time 2	F
Symptom subscales					
Fatigue					
Mean	3.50	2.68	4.20	3.84	5.13*
SD	1.61	1.41	1.14	1.40	
Cognitive difficulty					
Mean	2.53	2.28	3.06	2.96	
SD	1.33	1.42	1.44	1.51	1.17
Somatic symptoms					
Mean	1.94	1.54	2.29	2.26	4.66*
SD	1.34	1.15	1.04	1.06	

* $p < .05$.

Uncertainty, self-efficacy and coping

Inspection of the data for uncertainty suggested marked reductions in the scores for both groups (see Table 3). Unfortunately, a lack of homogeneity of variance and the differences in the size of the groups reduced the robustness of ANCOVA, so the effect of the treatment was assessed using a T-test on the change in scores. This indicated no signi-

ficant difference between the groups.

Table 3. Means (and SD) for uncertainty, self-efficacy and the IMQ subscales

	Treatment group		Control group		
	Time 1	Time 2	Time 1	Time 2	F
Uncertainty					
Mean	64.77	54.30	70.19	62.71	na
SD	7.88	12.14	15.87	14.05	
Self-efficacy					
Mean	47.05	62.14	47.22	50.20	6.79**
SD	17.97	14.55	16.20	17.87	
Coping subscales					
Maintaining activity					
Mean	3.22	2.59	3.42	3.13	na
SD	.85	.79	.83	.87	
Accommodating to the illness					
Mean	4.00	4.45	4.17	4.34	1.57
SD	.88	.86	.83	.91	
Focusing on symptoms					
Mean	3.60	3.46	3.67	3.59	.20
SD	.83	1.05	1.08	1.03	
Seeking information					
Mean	3.21	3.46	3.29	3.22	na
SD	.91	.86	1.11	1.21	

* p=.01

na Ancova not computed.

The scores for self-efficacy increased during the six months of the study, with the more marked changes in the treatment group. Analysis of covariance revealed that the difference between the groups was significant, F (1,38) = 6.79, p=.013.

The effects of the treatment on coping strategies were assessed using ANCOVAs for the scores for accommodating to the illness and focusing on symptoms. Failure to meet the requirements for ANCOVA meant that the data for the two remaining strategies were analysed using a two way ANOVA for the change in scores with the two factors being the baseline score (high/low) and group. There was no main effect for group for either maintaining activity or seeking information. However, there was a significant interaction between group and maintaining activity (F=4.4 [1,40], p=.042).

When asked whether they had been able to put Dr. Ho-Yen's advice into practice, some patients admitted that they had not. Reasons included feeling depressed and isolated as a result of the reduction in activity, and financial constraints.

Adjustment variables

In this study, adjustment was assessed using the scores on the HAD and the Functional Impairment Scale (see Table 4). Due to missing data, the number of cases varied per subscale.

Analysis of covariance on the original data revealed no significant differences between the groups, although there was a trend towards significance for anxiety (F 3.77, p=.059). However, since one case had unusually high scores on the HAD, the values were transformed as described above and the analysis was repeated. This revealed significant group differences for both depression, F (1,41) = 4.52, p=.04 and anxiety, F (1,41) =4.62, p=.04.

There were no differences in the Time 2 scores for functional impairment, $F(1,39) = 1.03$, $p=.32$, or for the corrected depression scores, $F(1,41) = 2.80$, $p=.10$.

Table 4. Means (and SD) for anxiety, depression and functional impairment scores

	Treatment group		Control group		
	Time 1	Time 2	Time 1	Time 2	F
Anxiety					
Mean	8.77	7.14	8.81	8.73	4.62*α
SD	4.90	3.86	4.00	3.93	
Depression					
Mean	7.95	6.59	9.59	9.05	4.52*α
SD	3.84	4.12	4.04	3.62	
Depression corrected					
Mean	5.82	4.91	6.86	6.59	2.80α
SD	3.26	3.58	3.89	3.43	
Functional Impairment					
Mean	22.81	20.86	22.91	22.73	1.03
SD	4.74	6.24	4.73	5.71	

* $p<.05$

α variable where outlier was transformed.

The prevalence rates for possible and probable cases of clinical anxiety and depression are shown in Table 5. The estimates for possible anxiety and depression at Time 1 were similar in both groups. Indeed, about 50% of the subjects had scores suggestive of clinical disorder, with rates of depression being the highest among the waiting list controls. However, the difference did not reach significance.

Table 5. Prevalence of possible and probable cases of anxiety and depression

		Treatment group		Control group	
		Time 1	Time 2	Time 1	Time 2
Anxiety					
≥ 8	No.	12/22	9/22	12/21	11/22
	%	55	41	57	50
≥ 11	No.	8/22	3/22	7/21	9/22
	%	36	14	33	41
Depression					
≥ 8	No.	13/22	11/22	14/22	14/22
	%	59	50	64	64
≥ 11	No.	5/22	3/22	11/21	11/22
	%	23	14	50	50
Depression corrected					
> 8	No.	8/22	4/22	11/22	11/22
	%	36	18	50	50
≥ 11	No.	1/22	1/22	4/22	2/22
	%	4.5	4.5	18	9

After 6 months, there was a slight reduction in the number of cases of possible clinical disorders among the treatment group but not among the controls. In contrast, there was a much more marked fall in the number of probable disorders among the treated patients, and for depression, the difference between the groups reached significance ($\chi^2 = 5.1$, $p=.02$).

Omitting the item 'I feel as if I am slowed down' from the calculation for depression reduced the estimates for both groups. For example, the Time 1 rate for probable clinical depression fell from 23% to 4.5% among the treatment group and from 50% to 18% among the controls. This particular item therefore had a marked influence on the estimates of cases. However, since there is no generally agreed position on cut-off points for corrected scores, further analysis relating to this variable was not considered meaningful.

5.4.4.1. The influence of self-efficacy on outcome

To determine if self-efficacy had influenced the success of the programme, the analysis was repeated with the scores for self-efficacy at Time 2 as an additional covariate.

The results showed that after adjusting for self-efficacy at Time 2, there were no significant differences between the groups on any of the measures. Thus the patients' self-efficacy had mediated the outcomes noted at Time 2.

5.4.4.2 The role of duration, fatigue, functional impairment and emotional distress on outcome

To ascertain if the programme benefited a specific subset, the Time 1 scores from the treated patients were split at the median, producing two samples defined in terms of high or low scores on specific variables. The latter comprised: duration, fatigue, functional impairment, anxiety and depression. Since the data did not satisfy the requirements for ANCOVA, the comparison of the newly created groups focused on the change in their scores for the outcome measures using T-tests for independent samples or where inappropriate, the Mann-Whitney test. Alpha was set at .05.

The results indicated that people who had been ill for a shorter period of time did not show greater changes in

scores compared to patients who had been ill longer. There were also no differences in outcome when patients were defined according to the degree of functional impairment and emotional distress.

Fatigue had no effect on the change in outcome, except in terms of perceived self-efficacy. In this case, those who reported more initial fatigue showed greater changes in self-efficacy scores ($t=2.34$, df 10.55, $p=.04$).

5.4.4.3 Patients' assessment of improvement

When asked to what they attributed their improvement during the previous 6 months, both treated patients and controls mentioned increased rest and relaxation. A few also noted the value of supplements and alternative therapies such as acupuncture and homoeopathy.

5.4.5 The relationships between variables

The relationships between variables were examined to determine if the correlations found in Study 2 were replicated in the patients studied here. Pearson's product-moment correlation coefficients were calculated using the pooled scores for Time 1. Where the data were not distributed normally, i.e. education, duration of illness and years diagnosed, Spearman's rank correlation coefficient was used instead.

The key variables in this part of the analysis were fatigue, anxiety, depression, functional impairment and uncertainty. The relationship between membership of a self-help group and these variables was assessed using the Mann-Whitney test for independent samples and for categorical data, with the χ^2 test.

Given the number of comparisons, the alpha level was set at 0.01 to reduce the risk of Type 1 errors.

Relationships between demographic data and key variables

There were no significant correlations between the demographic data and any of the key variables. The exception was the years of education after the age of 16 which was significantly related to functional impairment (r_s = .56, P<.001).

The relationships between illness and key variables

The analysis of the symptom scores showed that fatigue, cognitive difficulty and somatic symptoms were all significantly related to anxiety and depression (see Table 6). Furthermore, fatigue and cognitive difficulty both correlated with functional impairment.

Table 6. Relationships between illness and key variables at Time 1

	Anxiety	Depression	Depression corrected	Functional impairment
Fatigue	.50**	.58**	.53**	.42*
Cognitive difficulty	.43*	.47*	.46*	.51**
Somatic symptoms	.59**	.39*	.42*	.22
Functional Impairment	.32	.53**	.53**	1.00**

* p ≤.01 * p ≤.001

The relationship between uncertainty, self-efficacy, coping strategies and key variables

It should be noted that many of the patients had not been

formally diagnosed at Time 1, and some therefore found it difficult to complete the uncertainty questionnaire, with its questions about diagnosis, advice and treatment. This reduced the sample size to 29.

The results indicated modest correlations between uncertainty and the key variables. However, as shown in Table 7, the relationships with depression and with the corrected depression score reached significance. Furthermore, the correlation between uncertainty and anxiety showed a trend towards significance (r=.36, p=.05).

Self-efficacy was negatively related with fatigue (r = -.46, p<.01) and with both measures of depression (see Table 7). In contrast, there was only one significant correlation

Table 7. Relationships between uncertainty, self-efficacy, coping and key variables at Time 1

	Anxiety	Depression	Depression corrected	Functional impairment
Uncertainty	.36	.57*	.59**	.26
Self-efficacy	-.36	-.46*	-.45*	-.06
Coping:				
Maintaining activity	.31	.21	.18	-.17
Accommodating to illness	-.13	-.05	-.12	.28
Focusing on symptoms	.18	.22	.23	.39*
Seeking information	-.07	-.21	-.18	.12

* p ≤.01 * p ≤.001

between coping and the key variables. This was the relationship between focusing on symptoms and functional impairment (see below).

Membership of a patient group and support from doctors

Patients who were members of the local or the national self-help group did not report more fatigue or higher levels of emotional distress compared to people who had not joined such a group. Nor was membership related to amount of activity, frequency of symptoms or whether patients had taken sick leave. However, members did report greater functional impairment than non-members ($z=-2.8$, $p=.006$). More detailed analysis revealed that this relationship was limited to activities connected with work ($z=-2.7$, $p=.007$) and that there were no group differences for household duties and leisure pursuits.

There was also a link between membership of a patient association and two coping strategies. More specifically, people who had joined a self-help group were more likely to focus on symptoms than the others, and less likely to maintain activities. However, the differences between the two groups just failed to reach significance ($z=-2.27$ and $z=-2.36$, $p=.02$ respectively).

The patient's opinion of the support given by their doctors was not associated with any key variable.

5.5 Discussion

5.5.1 The effects of the treatment

The main aim of this study was to evaluate the effectiveness of a broad-based treatment programme for CFS. After six months, the results revealed significant differences between the treated patients and the waiting list controls on a

number of variables, including fatigue, somatic symptoms, self-efficacy and anxiety. There were also significant differences in the number of patients who scored above the cut-off point for probable clinical depression. However, no group differences were found for the number of cases of clinical anxiety, the severity of cognitive symptoms, the degree of uncertainty and the level of functional impairment. Moreover, although patients changed their coping strategies in the expected direction, the differences between the groups failed to reach significance.

The programme provides an alternative approach to most of the other treatments currently available for CFS. For instance, in contrast to CBT, it does not attempt to treat the causes of the illness, nor does it assume that external attributions will adversely affect coping and outcome (cf. Ray et al 1995). Indeed, Dr Ho-Yen's own views regarding the aetiology of CFS generally match those held by patients (cf. Sharpe 1994). This may have prevented the type of conflicts which have been documented in relation to CBT (cf. Hickie et al 1995) and limited the attrition rate.

However, the main difference between this management programme and CBT is the former's emphasis on rest. Thus patients are given advice about ways to conserve energy, for instance by giving up sport, getting sufficient sleep, and by increasing relaxation (Ho-Yen 1993). In contrast to CBT, they do not have to engage in certain activities when they were tired, nor do they have to meet targets set by the therapist. Indeed, this may have been one reason why there was only a limited reduction in functional impairment scores.

A more detailed comparison with other controlled trials is difficult given the differences in samples, design and assessment of outcome. However, it is noteworthy that the patients on Dr. Ho-Yen's programme were seen less often and

for a shorter period of time than the participants in the other trials. Consequently, some may not have received the additional counselling and emotional support which they required to deal with their individual problems and this may be why their anxiety and depression scores remained comparatively high.

Despite the variable results, 82% of the treated patients rated themselves as better or much better at Time 2. Moreover, five (23%) recovered to such an extent that further treatment was deemed unnecessary. However, the reasons for the improvement noted here, and in relation to the key measures, are unclear. For example, the significant differences between the groups at Time 2 were all mediated by increases in perceived self-efficacy. Thus the programme boosted the patients' confidence about being able to cope, and this in turn, influenced outcome (Holman and Lorig 1992).

At the same time, there was only a limited shift in the actual strategies used. For example, given the programme's emphasis on the need to balance rest and energy, one might have predicted a significant increase in the strategy of accommodating to the illness. Yet, while there was a change in the expected direction, the results suggest that patients may have found it difficult to pace their activities and avoid overexertion. Indeed, some patients admitted not being able to follow the advice to rest. Moreover, one or two reported that if they spent more time resting, they felt isolated and became depressed. Others mentioned difficulties with finances as a result of which they could not take further time off work. Unfortunately, since none of the questionnaires included an item focusing specifically on rest, it was not possible assess how many patients had actually reduced their activities and whether this particular strategy was related to outcome or not.

It is also possible that the remaining symptoms undermined the modification of the patient's behaviour. According to the self-help model proposed by Braden (1990), severe illness is positively associated with disruptive factors, and these can interfere with the patients' ability to engage in self-help. One of the problems in this respect may have been the continuing difficulties with concentration and memory. These may have undermined both the appraisal of stressors and the selection of appropriate responses (Earll 1989, Nerenz and Leventhal 1983).

Adherence to the programme did result in lower scores for maintaining activity. However, a similar, albeit smaller change was also found among the controls. The latter is consistent with anecdotal evidence that most patients cut down on activity through trial and error, or as a result of experience (English 1991, Fleming 1991 and see Study 1).

Another variable which was expected to show marked changes was the degree of uncertainty. The results at Time 1 had revealed very high scores in both groups. Indeed, the mean for the controls exceeded that for most other patient groups tested to date (Mishel and Epstein 1990). However, while the scores of the treated patients fell during the trial, the change was not significantly different from that recorded by the controls.

To obtain a more complete picture of the variables associated with successful outcome, patients were asked to what they personally attributed the improvements in their health. Most of those who answered this question valued the advice to increase rest. However, a few also listed other treatments such as vitamin injections, acupuncture and supplements. Only one person attributed the improvement in symptoms to antidepressants.

The value of the programme for specific subgroups

One of the additional aims of the study was to ascertain whether the more severely affected patients had a different outcome from those who were less severely ill. The results showed, however, that treated patients who were initially more anxious or depressed could not be distinguished from those with lower scores. This contrasts with the findings of Friedberg and Krupp (1994) who reported that patients with high scores for depression benefited more from their treatment than those who were less depressed.

The data were also analysed to determine the value of the programme for the more chronically-ill. This followed a suggestion by Ray et al (1995) that the advice to rest and pace activities might be helpful only for those who have been unwell for a relatively short period of time. The treated patients were therefore subdivided into two groups according to the duration of their symptoms, but no differences were found in relation to outcome. Coversely, there was a link between fatigue and self-efficacy. Thus patients with more severe fatigue at Time 1 showed significantly greater changes in self-efficacy than people with less severe fatigue. This finding may indicate the former's motivation to follow Dr. Ho-Yen's advice.

In summary, the results suggest that a broad-based programme providing information, support and specific advice on coping may help to alleviate some of the symptoms and the distress associated with CFS. However, there was only a limited improvement in terms of cognitive difficulties and functional impairment, and the levels of anxiety and depression remained comparatively high. Thus it is possible that some individuals might have benefited from more extensive counselling and advice.

Improvements in the controls

It is noteworthy that 50% of the controls felt better when reassessed after six months. Indeed, only 18% felt worse than before. Part of the improvements may have been due to a change in therapy prescribed by their doctor or alternative practitioner. However, only 7 of the 12 patients who began new treatments found them helpful. When patients were asked to what they attributed their improvement during the previous 6 months, some mentioned rest and relaxation, others referred to changes in diet or taking supplements. Interestingly, no-one listed the use of drugs.

5.5.2 The possible variables underlying emotional distress and functional impairment

To assess which factors were associated with emotional distress and functional impairment, the relationships between the key variables were analysed using pooled scores from Time 1. This part of the study also allowed a comparison to be made between the patients with CFS(PIFS) and the people with CFS(ME) investigated previously (see Chapter 4).

The findings from this study revealed that there was a significant relationship between the severity of symptoms and emotional distress. For instance, as in the case of patients with CFS(ME), somatic symptoms correlated with anxiety and cognitive difficulty correlated with depression. However, in contrast to the previous study, there was a much stronger and significant correlation between fatigue and depression.

The apparent link between symptoms and emotional distress found above is consistent with research on other patient groups (e.g. Friedman and Friedman 1993, Krupp et al 1990, see also Chapter 3).

Another finding which was replicated was the association between cognitive difficulty and functional impairment. As was argued in relation to CFS(ME), this variable has often been overlooked as a potential source of disability and distress.

A further relationship which could be compared was that between uncertainty and emotional distress. It was found that uncertainty significantly correlated with depression, replicating the finding documented in patients with CFS(ME). However, in contrast to the earlier study, the correlation between uncertainty and anxiety was more modest and just failed to reach significance.

The link between uncertainty and depression, which has also been reported in other patient groups (Christman et al 1988, Mishel and Braden 1987, Wineman 1990), supports the view that the former may undermine the psychological well-being of patients with chronic fatigue, and that it should be taken into account when considering the origins and treatment of co-existing psychiatric morbidity. However, further studies using larger samples are required to clarify the relationship between uncertainty and the severity of symptoms and to ascertain its specific role in the aetiology of clinical depression and anxiety.

A variable which had not been assessed before was self-efficacy. The results of the scores obtained prior to the programme revealed a significant correlation between self-efficacy and depression. This is consistent with other research (e.g. Lorig et al 1989, Terry 1992) and indicates that lack of self-efficacy is an additional variable which should be taken into account when considering the psychological status of the chronically-ill.

In terms of the relationship between coping and adjustment, the only strategy which was significantly associated with a

key variable was focusing on symptoms. The correlation between this strategy and functional impairment has also been documented by Ray et al (1993).

It is possible that focusing on symptoms may have encouraged patients to become more vigilant and introspective, thus increasing their awareness of the illness's impact on their lives (Mechanic 1993, Pavlou and Stefoski 1983). Indeed, this could have exacerbated perceived disability and it may have led to greater emotional distress (Hansell and Mechanic 1986, Ray et al 1993).

The lack of a relationship between either maintaining activity or accommodating to the illness and the HAD scores was also reported by Ray et al (1995). Thus whether patients ignore symptoms and keep going or plan their activities and pace themselves does not appear to have major consequences as far as anxiety or depression are concerned. On the other hand, Ray et al (1993) found a significant correlation between accommodating to the illness and functional impairment, suggesting that for some patients at least, pacing may reduce what patients are able, or feel able to do.

In general, however, it is not possible to draw firm conclusions about the role of specific strategies in the adjustment to CFS. More research is required to examine the relationship between coping and outcome in larger samples, and to determine the influence of rest and emotion-focused strategies which were not assessed here.

Two other issues which were addressed in this study related to the influence from doctors and self-help groups. Using scores from Time 1, it was found that support from doctors did not play a significant role in terms of adjustment. Neither did membership of a patient support group, except for the relationship with impairment at work. However, there was no evidence that members of a patient group were more

likely to give up work and take sick leave. Indeed, the converse may be true, i.e. that problems relating to employment may have led the patients to join a support group.

5.5.3 Methodological issues

At the time of testing, the length of the waiting list was approximately seven months. Given the severity of the illness and the fact that randomization would have increased the waiting time for many of the patients, it was decided to opt for the quasi-experimental design described above. This avoided additional delays, but may have created artifactual differences between the groups.

In order to reduce the influence of selection bias and non-random errors, the response to treatment was assessed after adjusting for baseline rates. Nevertheless, there were a number of differences between the groups which may have affected the results. For example, in the months prior to the study, 41% of the treatment group had already begun to improve, compared with 32% of the controls. While the greater tendency towards improvement among the treated patients may have had a positive effect on outcome, it should be noted that at Time 1, there were no significant differences between the groups on any of the key variables.

The groups also differed in terms of the length of illness; the treated patients having been ill longer than the controls. However, analysis at Time 1 and Time 2 revealed that there was no relationship between duration and outcome.

Another difference between the groups concerned the proportion of patients taking antidepressants. Indeed, 7 of the patients in the treatment group were taking such drugs at the start of the trial, compared to 4 of the controls. If their use had a positive influence on the course of the

illness, it would have affected the treatment group more than the controls. Indeed, it could be one reason why the treatment group had slightly lower baseline scores for fatigue, cognitive difficulty, and depression than the patients on the waiting list. However, only one or two people attributed their improvement during the programme to the drugs.

An additional factor which should be considered when interpreting the data is the small sample size. This precluded a more comprehensive analysis of the results, for instance, the use of multiple regression to identify predictors of outcome.

It should also be noted that the management programme was conducted by a single consultant with limited resources. In his view, short, bimonthly appointments seemed to be satisfactory for most of the patients. However, the limited time available was probably not sufficient to deal with all the problems experienced and this may be one reason for the continuing anxiety and cognitive difficulties reported by some of the patients in the trial.

Caution is also required in the interpretation of the findings relating to fatigue, anxiety and depression. With regard to the fatigue, transformation of the scores had only a limited effect. Other procedures were also attempted but these were considered unsatisfactory as they may have increased the risk of a Type II error. Since the lack of linearity for fatigue affected only one group and following expert advice, the data was assessed using analysis of covariance. Nevertheless, given the difficulties relating to the data, it is possible that the group differences for this variable may be more modest than the results suggest.

This is also the case for the data relating to anxiety and depression. The problem here was an outlier which had a

marked effect on the results. Since the values concerned came from the same source and conflicted with the individual's reports of improvement, and because inclusion may have increased the risk of a Type 1 error, it was decided to transform that person's scores. The analysis was also repeated without the outlier, but as the results were similar to those using the transformed score, the latter were retained.

Finally, there were specific problems associated with two of the measures. Firstly, the IMQ did not include specific questions addressing the amount of rest. Since anecdotal reports identified increased rest as the most important reason for improvement, further research to assess this strategy in a more formal way may be useful.

Secondly, the MUIS Community Form which was used to assess uncertainty assumes that patients have been diagnosed and are being treated. This means that patients who were still waiting for a diagnosis found it difficult to complete the whole questionnaire. Because low scores might have been wrongly interpreted as reflecting lack of uncertainty rather than lack of diagnosis, all questionnaires with more than 4 missing items were discarded. This not only led to a significant loss of information, particularly among the controls, but also made it difficult to interpret the data on uncertainty at Time 1.

While the results suggest that uncertainty should be included in future studies into the psychological well-being of patients with CFS, amendments to the MUIS are required to increase its reliability and accuracy.

5.6 Summary

The results of this study showed that a programme focusing on increased rest and relaxation led to marked improvements

in fatigue, somatic symptoms and perceived self-efficacy. Indeed, at six months, the differences between the treated patients and waiting list control on these measures reached significance. There were also differences between the groups for anxiety and depression, although the scores for some of the patients remained high. Moreover, there were limited changes in the severity of cognitive difficulties and the degree of functional impairment. Thus while the treatment programme helped many people, a significant number might have benefited from more extensive counselling, advice and support.

When asked for the reasons for improvement, most patients mentioned increased rest. This, however, was not directly reflected in the measures used to assess coping.

Finally, the analysis of the relationships between variables at Time 1 suggested that the severity of the illness played an important role both in terms of emotional distress and functional impairment. Moreover, uncertainty was significantly related to depression as was a lack of self-efficacy. Some of these findings replicated the results documented in Study 2.

Overall, the results suggest that this type of programme may provide a useful basis for the clinical management of patients with post infectious chronic fatigue syndrome.

CHAPTER 6

Summary, conclusions and recommendations for future research

The studies described in this thesis examined the psychological effects of CFS and the patients' response to their illness. Information was also obtained about the nature, extent and possible origins of the psychological problems experienced by people with CFS. Finally, a 5-step treatment programme was evaluated to ascertain if information, support and practical advice could improve symptoms and alleviate distress.

Previous studies on CFS had tended to focus on the prevalence of psychiatric morbidity and the association between certain beliefs and ongoing fatigue (David 1991, Sharpe 1994). Their findings formed the basis both of the cognitive-behavioural model and the cognitive-behavioural rehabilitation programmes (Butler et al 1991, Sharpe et al 1996). However, given the relative paucity of information about the illness and its impact on the patients' lives, the thesis here is that some sources of emotional distress may not have been recognised.

The results of the first two studies showed that CFS is far more complex, both in terms of the type of symptoms and its effects than the cognitive-behavioural model suggests. The third study supported some of these findings and also revealed that patients on a broad-based programme had less fatigue, fewer somatic symptoms and less emotional distress than the waiting list controls. The following sections will review the main results in more detail.

6.1 CFS and its effects: the patients' perspective

The first study assessed a number of different aspects of

the illness. Information was collected from 53 members of the ME Association and 17 patients who were recruited from local general practices. However, since this part of the research was conducted before the introduction of the UK criteria for CFS (Sharpe et al 1992), all the patients had been diagnosed as having either ME or PVFS.

Fatigue was the most frequently reported symptom but many patients also described symptoms such as muscle pain, difficulties with memory and concentration, weakness and malaise. Other prominent complaints included visual disturbances, nausea and sensitivity to temperature changes, all of which are often overlooked (e.g. Lapp and Cheney 1995, Thomas 1993, Sharpe 1993).

A question which asked the patients' views on the aetiology of CFS revealed that many attributed their illness to infection. However, this was rarely regarded as the sole cause, and many implicated a busy and stressful life as a contributory factor. The latter conflicts with suggestions that most patients adhere exclusively to external attributions and that they refuse to consider psychological ones (Lawrie and Pelosi 1994, Surawy et al 1995, Woods and Goldberg 1991). Nevertheless, when asked about unlikely causes, a number mentioned factors such as lack of activity and clinical depression. This indicates that many patients rejected the main psychiatric explanations for CFS.

A question about their perception of the future revealed that the majority of the patients were generally optimistic, predicting a slow recovery and gaining more control. This is inconsistent with the suggestions in the literature that CFS is frequently associated with helplessness, hopelessness and demoralization (Butler et al 1991, Sharpe 1994).

Another finding was that the illness had a profound effect on almost every aspect of the sufferers' lives. Aside from

the practical problems related to the symptoms e.g. mobility, self-care and household management, many patients faced occupational difficulties and a loss of income (cf Locker 1983).

The illness also led to a perceived loss of control and a restricted life. For instance, limited energy levels meant that patients often had to reorganise their priorities, and drop or reduce the time spent on previously valued leisure pursuits. Others were so disabled that they described their lives as an "existence". On a personal level, it often changed many people's personalities, reducing their self-confidence, and in some cases, their self-esteem. The condition also led to strained relationships with family and friends, to a loss of contact with significant others and hence to social isolation and loneliness.

Further indications of the disabling nature of the illness were obtained from comments on the meaning of CFS. Some people listed emotions such as anger, frustration, sadness and despair. Others noted that the condition had led to a complete change in their lives, to "devastation", "trauma" and a "hell on earth". Likewise, a number described their illness in terms of a constant "fight", "battle" and "struggle".

These answers underline the severity of the illness as well as the many challenges which patients faced. However, it may be argued that neither have been fully recognised in the cognitive-behavioural model, with its emphasis on physical and mental fatigue. The results also suggest that the nature of the symptoms and the effects of the disability may have contributed, at least in part, to the emotional distress which many experienced.

6.2 The emotional distress associated with CFS

Study 2 assessed the levels of emotional distress in a more structured way and tried to clarify the influence of specific variables such as the severity of symptoms and the quality and quantity of social support. The sample comprised 58 patients with CFS(ME) recruited from a hospital clinic in Essex and the results were compared with those 25 people with spinal injuries (SCI).

The findings showed that the level of anxiety among patients with CFS(ME) was not significantly different from that recorded by the comparison group. However, there were many CFS(ME) patients whose scores suggested the presence of a clinical disorder. In fact, the percentage of patients with severe anxiety was higher than expected and contrasted with estimates reported in the literature (Buchwald et al 1994, Hickie et al 1990, Katon et al 1991, Pepper et al 1993). This indicates that the presence of anxiety may be an additional source of suffering in many patients with CFS and that it deserves more attention, both in the research on psychopathology and in clinical practice (Butler et al 1991, Lynch et al 1992, Sharpe 1994, Wessely and Powell 1989).

The other measure of emotional distress assessed depression. It was found that the level of depressive symptoms was markedly higher than that of the comparison group. However, when one fatigue-related item was omitted, the difference between the groups was no longer significant. This is important because the inclusion of that item could have given a misleading view of the prevalence of depressive mood in CFS. For example, the number of cases of *possible* clinical depression among people with CFS(ME) was estimated to be 55%, but removing this item reduced this figure to just 28%.

As far as the estimate of *probable* clinical depression is concerned, the rate of 22% is lower than most of the figures

reported to date (e.g. Lane et al 1991, Wessely and Powell 1989, Yeomans and Conway 1991). The discrepancy between the various estimates could be interpreted in a number of ways. First and foremost, it may be the result of the type of measure used. The HAD, used in studies 2 and 3, is a screening tool rather than a means of making accurate diagnoses (Rodin et al 1991). Even so, the estimate above was also lower than that found in another study which used the HAD (Yeomans and Conway 1991).

Secondly, the higher rates of psychiatric illnesses reported by other researchers could reflect sample differences (David 1991, Katon et al 1991, Wessely and Powell 1989, Lane et al 1991). For instance, the use of different diagnostic criteria may have led some to include patients with other fatigue syndromes such as fibromyalgia. There may also have been inter-group variations in the severity of illness and level of physical and functional impairment.

Study 3, which assessed 44 patients with CFS(PIFS) on two occasions, provided further evidence that the condition is associated with significant emotional distress. Indeed, more than a half of those tested at Time 1 had scores suggesting the possible presence of clinical disorders.

6.3 The relationship between illness and adjustment

The results of Study 2 revealed that emotional distress was not correlated with physical functioning as measured by MOS Short Form. This was true for both the CFS(ME) group and the people with SCI, indicating that the difficulties with walking and other physical functions did not contribute significantly to depressed mood. Conversely, there was a significant relationship between the severity of symptoms and psychological distress, and many of the findings reported below were replicated in Study 3.

For instance, fatigue correlated with anxiety in both studies, which supports the suggestion by Lazarus (1991) that "when we are fatigued, demands that might have been challenging and exhilarating are now too much to handle, leading to a sense of threat and anxiety feelings". On the other hand, the possibility that anxiety may have perpetuated ongoing fatigue can not be ruled out and this should be taken into account, not only when interpreting the research but also when determining appropriate treatments.

Study 2 also examined the relationship between cognitive difficulty and emotional distress. It was found that the former correlated with depression and this was again replicated in Study 3. Cognitive difficulty also correlated with anxiety, but only in Study 3.

While it is possible that the documented problems with memory and concentration were the result of concomitant affective disorders, research reported elsewhere suggests that the performance on certain psychoneurological tests is not affected by mood (DeLuca et al 1995, DeLuca et al 1993, Smith et al 1993). Moreover, studies have found significant differences in terms of the degree of cognitive impairment recorded by patients with CFS and depression (e.g. Sandman et al 1993). Thus mood disorders probably do not account for all the cognitive deficits associated with CFS.

Further studies are needed not only to investigate the relationship between cognitive functioning and emotional distress in more detail, but also to consider its role in adjustment to CFS. For instance, cognitive difficulties could interfere with the appraisal of stressors and the selection and evaluation of appropriate coping strategies (Earll 1989). In the case of patients with CFS, problems in information processing may be especially maladaptive because of the uncertainty which surrounds this disorder. Indeed, failure to comprehend information about the illness may

increase anxiety and fear and reduce the perception of control (Dimond 1983). Lastly, problems with memory and concentration should be recognised and treated because they may lead patients to misinterpret medical advice and misunderstand the views of significant others.

The findings relating to the number and severity of somatic symptoms are less easy to interpret. The symptom scores were significantly related to anxiety in both studies 2 and 3, and with depression in Study 3. Again, it is difficult to establish whether the symptoms contributed to the patient's distress or whether the reverse is true.

Aside from their links with emotional distress, the symptoms of CFS were also associated with functional impairment. Although the strongest predictor of this adjustment measure was found to be physical functioning, cognitive difficulty made a significant contribution to the variance in functional impairment scores in Study 2 and correlated significantly with the latter in Study 3. Fatigue was also associated with functional impairment, but only in Study 3. Thus while fatigue may have had a disruptive effect in the lives of some patients with CFS (Fisk et al 1994, Monks 1989), problems with concentration and memory appeared to have a greater influence with regard to their ability to work and pursue leisure activities.

Interestingly, there was no association between somatic symptoms and functional impairment. Thus complaints such as dizziness, sore throats, pain or digestive disorders did not influence the patients' ability to work or vice versa. However, one potentially important symptom, nausea, was not assessed and therefore the findings relating to somatic symptoms and impairment may not be reliable.

The lack of information in the medical press concerning the severity and possible influence of symptoms other than

fatigue might help to explain the apparent discrepancy between the patients and doctors' conceptualization of CFS (eg Toombs 1992). It may also have added to the latter's 'abnormal illness perception' as described by Denz-Penhey and Murdoch (1993). There is therefore a need to increase awareness of these symptoms, not only to further the clinicians' understanding of the illness-experience but also help them assess the requirements for vocational rehabilitation, and to develop more effective treatments.

6.4. The role of uncertainty

The findings from both studies 2 and 3 suggest that emotional distress was related, at least in part, to the uncertainty of the illness. However, the association with anxiety was stronger in Study 2 than amongst the patients from Study 3. The inconsistencies between samples may reflect differences in the way in which patients appraise their illness (Mishel et al 1991). For instance, many of the patients in Study 3 were tested before they had received a diagnosis. However, because they knew this problem would soon be resolved, they might have been less anxious than the CFS(ME) group who had already been diagnosed and who may have been troubled by more challenging questions, e.g. the reasons for the fluctuations or their failure to improve.

It is also possible that the weaker links between uncertainty and anxiety in Study 3 may have been influenced by the slightly lower symptom scores. In fact, the findings support the view that variables such as uncertainty might have a greater effect on emotional well-being in those who are more severely ill. This could also explain why uncertainty was more closely linked to psychological distress in the patients with CFS(ME) than in the people with spinal injuries.

Although the results are consistent with Mishel's uncer-

tainty in illness theory (Mishel 1988, 1990) and the research on other patient groups (Mishel et al 1991, Wineman 1990), it is possible that the high uncertainty scores among the CFS patients were the result of emotional disturbances, rather than a cause. Indeed, there may be a vicious circle, whereby uncertainties about the illness exacerbate the patients' fears and anxieties, which in turn increase perceived uncertainty and so on.

Further longitudinal research is required to clarify the influence of perceived uncertainty on the psychological well-being of people with CFS. In the meantime, this variable should not be overlooked when considering the possible determinants of emotional distress and the more severe clinical disorders in this patient group.

6.5 The role of social support

Following the reports of patients in Study 1, the role of social support was examined *in more detail in Study 2. It* was found that stressful relationships with partners were significantly related to increased depression while resources from friends were associated with lower depression (corrected score). The findings also showed that the relationship between social support and emotional distress varied according to the source of the former. For instance, although contact with friends was significantly related to lower depression scores, this was not the case for contact with relatives.

It is of course possible that the presence of depression led to a reduction in contact with friends and an increase in stressful relationship with partners (Billings et al 1983, Fitzpatrick et al 1991). However, if mood disorder was the primary reason for the reduced social support, one might have expected similar correlations for contact with relatives. Moreover, information obtained from the inter-

views in Study 1 suggests that the uncertainty about the aetiology of the illness was a major source of conflict and distress.

As Donoghue and Siegel (1992, p42) noted "fatigue without an evident cause makes us suspicious; we treat the person with 'unsubstantiated fatigue' with distrust". In their view, illnesses with a degree of social unacceptability, including CFS, can have "an extraordinary impact on the psychological well-being of a person... If a disease is deemed unacceptable, overtly or covertly, by society, its victims suffer the added burden of isolation and shame".

Similar experiences have also been reported in relation to other conditions which have no clearly identifiable organic cause (Faucett and Levine 1990). In contrast, the uncontroversial diagnosis and social acceptability of spinal injuries may have prevented the type of disputes reported by patients with CFS, as a result of which there was no clear association between lack of social support and emotional distress among the people with SCI.

Unfortunately, the importance of social support for people with CFS has received comparatively little attention so far. Yet, as research on other chronic conditions has shown, it may help patients and care-givers to understand the problems they face, it may reduce uncertainty and ambiguity and also enable them to find and use effective coping strategies (Mishel and Braden 1987). It may also increase their motivation to take certain actions and accordingly reduce emotional distress (Revenson 1993). Aside from its links with adjustment (Brown 1988, Radley 1988), lack of support might also have a direct effect on the disease process. For instance, research has found that loneliness and isolation can undermine immune function, thus increasing the risk of further ill-health (Laudenslager 1987, Levy 1990).

Since problematic relationships can cause considerable suffering, they need to be taken into account both when considering the origins of psychological distress and in terms of patient care. The partners may also benefit from support, particularly in the early stages when they may not yet have learnt how to deal with the sudden relapses and the limited availability of medical advice. Later on as the patient's needs change, care-givers may value additional information and in some cases, more tangible forms of assistance at home.

6.6 Coping with CFS

The types of strategies used to cope with CFS were investigated in studies 1 and 3. The results from Study 1 showed that patients did not respond to their illness primarily by resting and 'waiting for a cure' (cf. Wessely et al 1991). Indeed, a variety of strategies were reported, including emotion-focused ones like positive thinking and diversion, as well as problem-focused strategies such as pacing of activities, seeking information and support, a change in diet, and the use of complementary therapies. Some patients also mentioned relaxation and counselling and a significant number took or had taken psychotropic drugs. These findings conflict with suggestions that patients with CFS adopt a relatively passive approach to their condition, and they indicate that the descriptions of CFS which focus primarily on avoidance behaviours may be incomplete (cf. Surawy et al 1995, Wessely et al 1991).

Study 3 investigated coping further by assessing four different type of strategies. The results showed that neither accommodating to the illness nor maintaining activity was significantly related to emotional adjustment. Indeed, the only strategy to be linked with outcome was focusing on symptoms. This suggests that any treatment requiring patients to pay close attention to how they are feeling may

encourage introspectiveness and lead to greater perceived disability (Mechanic 1993). Alternatively, the relationship between functional impairment and focusing on symptoms might be mediated by fatigue (cf. Ray et al 1995).

The value of pacing as a specific strategy remains unclear. It was used by some patients assessed in Study 1 and it has been reported as helpful by other patient groups (e.g. Wiener 1984, Monks 1983). However, the lack of an association between accommodating to the illness, which covers this strategy, and the various outcome measures suggests that any benefits are probably limited. It is also possible that this strategy may only be of value in specific subgroups (Ray et al 1995, see also section 6.7).

When asked for their personal opinion, patients in Study 3 considered that resting was the most helpful of all strategies. This finding is consistent with the experience of CFS specialists (e.g. Macintyre 1992, Shepherd 1992), with the literature (e.g. Dawes 1991, Denz-Penhey and Murdoch 1993, Fleming 1994) and with the views of patients from Study 1. Unfortunately, the measures used in Study 3 did not assess rest on its own and it was therefore not possible to clarify the relationship between rest and outcome in a more formal way.

Further studies using larger samples are required to evaluate the effectiveness not only of rest but also of strategies such as pacing and relaxation. These should be assessed at different stages of the illness, to ascertain if they are helpful (or unhelpful) *throughout* or *only during a* certain phase (Ray et al 1995). Moreover, it may be useful to study different degrees of rest to determine if there is an optimum level which promotes recovery.

6.7 The effects of treatment

The management programme which was examined in Study 3 focused on the importance of rest and relaxation and thus constitutes an alternative to CBT, with its emphasis on increasing activity (Butler et al 1991). The results showed that after adjusting for the baseline scores, there were significant differences between the treated group and controls for fatigue, somatic symptoms, anxiety, depression and self-efficacy. Further analysis revealed that the benefits were not restricted to a particular subset of patients, e.g. those with high levels of depression (cf. Friedberg and Krupp 1994). Although the sample was small and the results for fatigue and depression should be interpreted with caution, the findings above suggest that the programme may be a useful, basic approach towards the management of CFS (cf. Friedberg and Krupp 1994, Lloyd et al 1993).

It should be noted, however, that the rates of emotional distress after 6 months of treatment remained comparatively high. This indicates that for some patients, the programme did not meet all their needs. The results also underline the importance of further research into the determinants of anxiety and depression, and the value of fostering skills which will enable patients to cope with their emotions more effectively. The latter could involve giving additional information about the illness, increasing the time spent on identifying triggers of relapses and teaching patients specific relaxation techniques.

Further consideration should also be given to the treatment of cognitive difficulties, the scores for which showed little change at Time 2. In contrast, there was a marked increase in the patients' self-confidence about being able to control the illness. This proved to be an important variable since self-efficacy mediated the outcomes described above. The results also revealed an inverse relationship

between self-efficacy and depression at Time 1, showing that confidence about controlling the illness may help to reduce emotional distress. It is also consistent with research on other disorders (e.g. Holman and Lorig 1992, Terry 1992) and suggests that self-efficacy should be included not only in assessment of psychological morbidity but also in the evaluation of all psychological interventions for CFS.

Lastly, although the programme did not include ongoing counselling for carers, there is evidence from the literature on other chronic conditions that this could improve interpersonal relationships. For example, health care professionals could teach patients and their carers how to develop and maintain family ties. In addition, they could show partners how to assess the patient's support needs and inform them how to recognize and accept help and emotional encouragement provided by others (Revenson 1993).

To summarise, the programme appeared to be a promising form of treatment for many patients with CFS(PIFS), although there is evidence that some individuals might have benefited from additional counselling and advice. Whether the programme is as effective for other fatigue syndromes e.g. fibromyalgia, has yet to be established.

6.8 The role of the health-care profession

Although about a half of the patients in Study 3 were happy with the medical advice and support they had received from doctors other than Dr. Ho-Yen, a significant number were not. Together with the anecdotal reports from patients in Study 1, this provides some evidence that a sizeable proportion of health care professionals remain unsympathetic towards people with CFS (cf. Dalrymple 1992, English 1991, Hartnell 1987).

Although unsatisfactory medical support was not related to

outcome in Study 3, there have been suggestions that a lack of diagnosis and conflicts with health care professionals may affect the course of the illness' as well as the patients' emotional state. For example, without a diagnosis which fits their experience, patients may find it hard to interpret their symptoms and to determine effective coping responses (Mishel 1988, Nerenz and Leventhal 1983, Stewart and Sullivan 1982). Furthermore, failure to legitimize the person's ill-health could encourage the person to 'carry on' as normal beyond the time when they are capable, thus placing unnecessary demands upon their adaptive capacities (Rippere 1992b).

The lack of a diagnosis might also damage the patient in other ways. For instance, a survey of 50 people with CFS revealed that the absence of an acceptable diagnosis led to fear, anxiety, confusion self-doubt and bitterness. Some patients also lost their sense of identity and purpose (Woodward et al 1995). Indeed, Woodward et al (1995) found that receiving a diagnosis gave meaning to the patients' suffering and allowed them to create a linguistic distinction between themselves and their illness. They could begin to say "I am not crazy; it's this illness that is crazy". It also eased their distress and instead of contributing to chronicity as doctors feared, early diagnosis avoided some of the harmful social and psychological consequences associated with uncertainty. Thus patients tended to see their illness as less traumatic, and they felt less despair and helplessness.

Adverse reactions have also been noted in relation to inaccurate diagnoses. For example, Rippere (1991, 1992b) has described how receipt of an inappropriate psychological diagnosis can undermine feelings of self-efficacy and self-confidence, leading in some cases to an emotional state resembling post-traumatic stress syndrome.

At the same time, doubts about the origins of the illness may initiate negative reactions from family and friends. This is particularly likely if others adhere to the view that psychological disorders are 'all in the mind' and that the person could sort themselves out 'if only they wanted to'. According to Denz-Penhey and Murdoch (1993), this can lead to "a fault, guilt, blame, shame game" which is extremely unhelpful for those who are ill.

The support and guidance from health care professionals is also important following diagnosis, especially for those conditions where the treatment options are limited (Elliott et al 1992). Aside from symptomatic treatment, they can help patients adjust to disorders like CFS by giving them information and teaching them skills so they can:
1. interpret and manage new or changing symptoms; minimize physical disability (e.g. by avoiding deconditioning or nutritional deficiencies),
2. establish realistic expectations and emotional responses to the vicissitudes of the illness,
3. become adept at ways to solve problems as they arise and
4. use the available resources in the community to advantage (Holman and Lorig 1992).

Above all, doctors should try to offer patients support, reassurance and hope (cf. Woodward et al 1995). Moreover, given the nature of the illness, a multi-disciplinary approach involving physicians, counsellors and nutritionists may be more appropriate than the traditional biomedical or psychiatric management of patients.

6.10 Methodological issues

There were certain limitations in methodology and design which should be taken into account when evaluating the results. Some of these have already been discussed in relation to the individual studies. Others will be briefly

described below.

For example, because there is no diagnostic test for CFS, it is possible that some of the participants may have suffered from another, unrecognised condition. In fact, Australian researchers found that even the use of strict criteria did not prevent 20% of their patients being wrongly classified (Wilson et al 1994b). Given that the diagnosis of CFS remains a clinical one, it was hoped that the consultants' extensive experience would reduce the error rate in studies 2 and 3 to an absolute minimum.

There are also other reasons why the symptoms scores must be interpreted with care. For example, the fluctuating nature of CFS means that the scores collected at one particular time may not accurately reflect the severity of the illness in general. While the reliability of the data could have been improved by asking the patients to complete questionnaires once a week for a month or two, this would have placed too great a burden on the participants so this was not pursued. The results of both studies therefore represent just one estimation of the severity of the symptoms experienced during the previous week.

A further problem which has to be considered is that the responses regarding symptoms could have been influenced by 'disease prototypes'. According to Bishop (1991), people have schemata of diseases and when they experience symptoms, they try to match them with the available prototypes in their memory. When reporting symptoms, they are more likely to recall complaints which fit in with a certain prototype and to ignore or downplay symptoms which don't. In other words, the way in which information about symptoms are represented in one's memory can produce a bias when recalling, interpreting and describing those symptoms. Since the media has focused on the problem of fatigue, recall bias may have influenced the patients' reports related to this symptom.

However, most lay articles on CFS have not discussed cognitive impairment in any detail, so these symptoms probably do not figure very strongly in most patients' prototype of CFS. Consequently, the recall bias for cognitive problems was probably limited.

Another factor which may have influenced the symptom reporting is what Watson and Pennebaker (1992) refer to as negative affectivity (NA). This is described as a pervasive personality trait which is more or less synonymous with several other dispositional constructs, including neuroticism, trait anxiety, pessimism and general maladjustment. Negative affectivity is therefore a dimension which reflects negative mood and self-concept.

Watson and Pennebaker have suggested that high-NA subjects are more likely to complain about their internal physical sensations and to exaggerate or magnify their actual health problems. Thus one might expect high-NA patients to overreport the symptoms associated with CFS. As these studies did not include any personality measures, it is not possible to evaluate the possible role that negative affectivity may have played. However, it is interesting to note that the patients did not rate all the symptoms equally highly. Indeed, it was not uncommon to find patients with high levels on some subscales and very low scores on others. This is not consistent with a general tendency to overreport all health complaints.

Finally, it should be noted that some patients may have been influenced by the stigma associated with mental illness. As a result, they might have suppressed feelings of sadness and despair, either consciously or unconsciously, and focused on physical symptoms such as headaches, myalgia and pain instead. This could have affected the scores for anxiety and depression, and might lead to the misdiagnosis of psychiatric disorders in clinical practice (Komaroff 1994, Lane et

al 1991).

6.10 An alternative model of CFS

On the basis of the research presented above, it may be argued that the cognitive-behavioural model of CFS does not acknowledge the complexity of CFS. More specifically, the findings suggest that it underestimates the disabling effects of symptoms other than fatigue. Moreover, its analysis of the emotional distress associated with CFS does not appear to consider the influence of non-medical variables such as uncertainty and social support and most of all, it does not recognise the consequences of disbelief and controversy.

The model may also have oversimplified the patients' response to their illness. For example, it seems to suggest that most patients react in a similar way, i.e. by blaming external causes, by avoiding activity and by rejecting psychological treatments which might help them to cope (Butler et al 1991, Sharpe 1993, Surawy et al 1995, Wessely et al 1991, Wessely 1993). This conflicts with the results from Study 1 which revealed that most patients used a variety of strategies and that many accepted that psychological factors may have played a contributory role in the onset of their disease. Moreover, the fact that about a third of the patients with CFS were taking antidepressants is inconsistent with the view that these individuals refuse all psychiatric help (Wessely 1993). Likewise, the interest in self-help and the largely optimistic views about the future are difficult to reconcile with the model which predicts that most patients will inevitably become helpless and demoralized (Butler et al 1991).

The model below acknowledges that many different factors may increase emotional distress and undermine adjustment to CFS. They include the type, severity and impact of the symptoms;

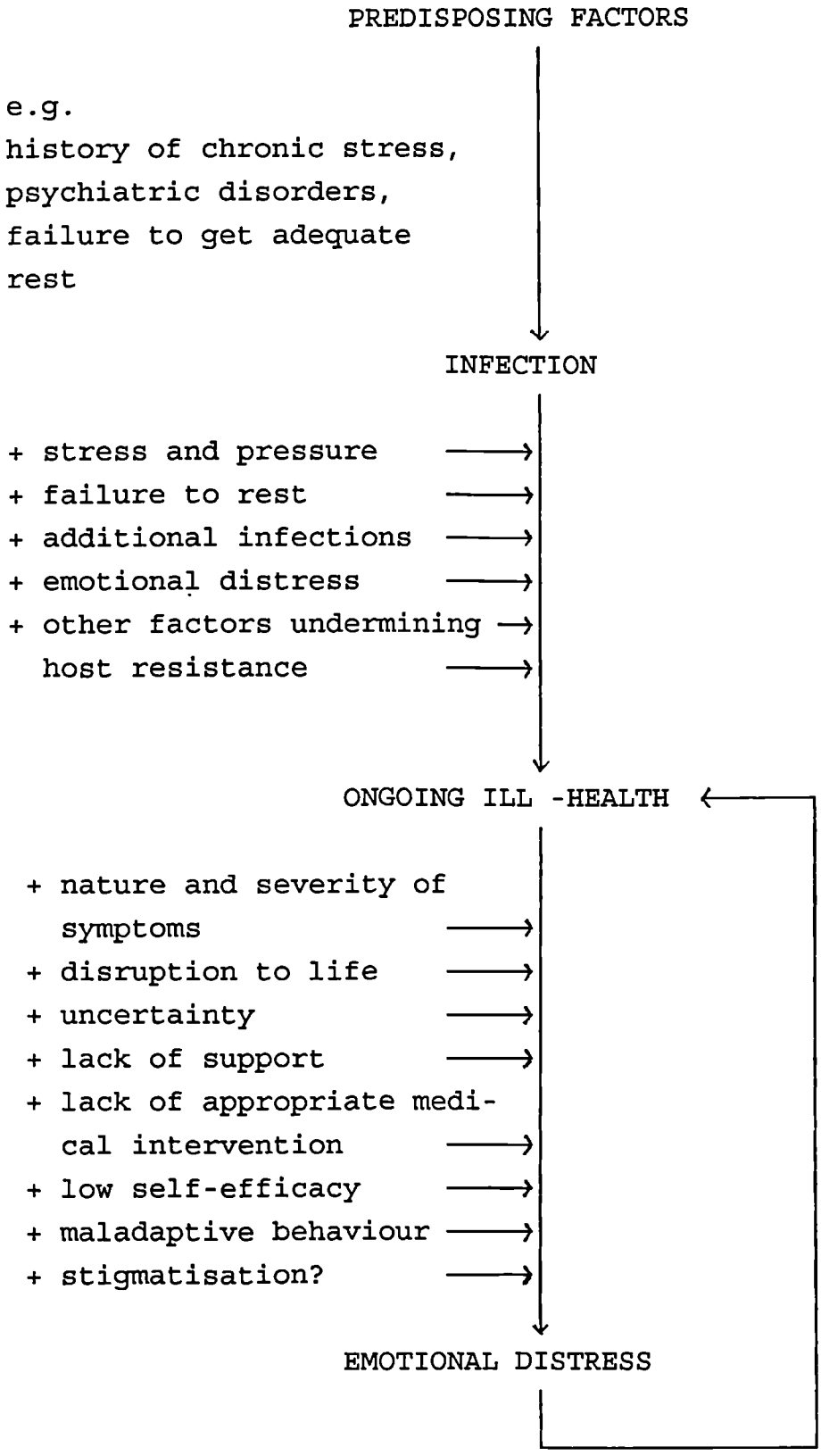

Figure 1. Model showing the main variables associated with emotional distress in CFS

the uncertainty which surrounds CFS, the unsatisfactory social support as well as the lack of medical help and advice. It is proposed that the failure to acknowledge such variables will reduce our understanding of the 'dis-ease' and consequently undermine both treatment and recovery.

It should be noted, however, that neither the attributions about aetiology nor the avoidance of activity are regarded as a major sources of ongoing disability and distress. Admittedly, these factors may be relevant in specific individuals, where certain beliefs may lead them adopt maladaptive behaviours, such as total bedrest, withdrawal from significant others, continued overexertion and the refusal of appropriate help (cf. Antoni et al 1994). In such cases, CBT must continue to be the treatment of choice (Goldenberg et al 1994, Larcombe and Wilson 1984).

Further research is required to assess the validity of the model, not just for patients with CFS but also for syndromes associated with the exposure to toxins and adverse reactions to vaccinations. However, it is important that studies should continue to separate the various subgroups. Without the careful delineation of samples, it will not be possible to identify any differences in aetiology or response to treatment.

6.11 The stigmatisation of CFS

One problem associated with CFS which was not formally studied here but which may affect the welfare of patients and which therefore deserves more attention is the stigmatisation of the illness.

For instance, patients have been consistently stereotyped in a negative way, those who support and represent them have been discredited and the illness itself has been trivialised as comprising little more than tiredness and malaise. More

specifically, patients have been portrayed as poor problem solvers who refuse to consider and deal with their emotional conflicts in order to avoid the stigma of mental illness.

For example, Cluff (1991) wrote that:

> "When we say 'chronic fatigue syndrome' (CFS), we are describing an experience of people who have symptoms that ... all of us experience many times during our lives - lassitude, fatiguability and tiredness. Generally, as I have gotten older, I do not enjoy getting up early in the morning as much as I did when I was younger. We could assume I am acquiring CFS as a result of advancing age".

In addition, Silver (1994) claimed that:

> "The extreme reluctance of ME sufferers to admit that their condition is of psychological origin demonstrates that the much-vaunted acceptance by our society of psychiatric conditions is bogus. If the condition is viral in origin, it is "real" and therefore beyond the individual's control; but if on the other hand it has a psychological cause ... it is only an elaborate form of malingering, more a character defect than a bone fide illness."

These comments are not just implying that patients are exaggerating their distress but also that they do not require some of the resources available to the physically ill (wheelchairs, home help etc). Moreover, the portraits are composites of misguided individuals. In the first quote, the suggestion is that patients have comparatively trivial symptoms which everyone else can cope with (i.e. tiredness). In the second, the insinuation is that CFS is a psychological disorder but that (all) patients have a prejudiced view of psychiatry and refuse to acknowledge the real source of

their distress.

Other commentators have gone further by claiming that sufferers themselves are largely responsible for their ongoing ill-health with the implication that if they would accept the real origins of their fatigue (i.e. psychological problems), they would get well (Lawrie and Pelosi 1994).

In line with the depictions of other stigmatized groups, there are no positive attributes associated with either the patients or their illness. Moreover, highly selective reporting has reinforced the negative stereotype (e.g. Read 1993), supporting the inferior identity of the CFS patient and in some cases, portraying them as a threat. For example, Silver (1994) claimed that any doctor or researcher who suggests that ME has a psychological component can expect intimidation and persecution. He added:

> "Medical journalists of my acquaintance will not touch the subject because they fear the response... One television producer was so intimidated by the response to a programme he made about ME that he vowed never to return to the subject. He had his family to consider and was not prepared to risk their well-being over something which is, after all, only of marginal importance to most people".

In short, the current image of the ME patient is of people who are simply tired but don't want to face up to their psychiatric problems, and who don't tolerate any other opinions. It is the image of an inferior, deviant person who is a threat to others. Clearly, this portrayal makes it difficult for those affected to maintain a positive view of themselves. The negative stereotype may also shape public attitudes and undermine social support (Schur 1980). Indeed, the general social disvaluation could encourage the avoidance of social contact and lower the patient's self-

esteem.

Some of the participants in Study 1 referred to the lack of understanding among significant others and the problems of not being believed. However, whether it also contributed to their self-concept and emotional distress as has been noted in patients with rectal cancer (Macdonald 1988), remains a matter for debate.

The stigmatisation of CFS may certainly help to explain the hostility shown by organisations and institutions involved in the financial support for the chronically-ill. For instance, Lloyd and Pender (1994) reported that:

> "The label of CFS stimulates widely varied responses from both public and private sector agencies. These responses range from overt persecution of the patient (presumably with the aim of having a claim withdrawn) to aggressive antagonism of the validity of the label via medicolegal avenues. Rarely, support and acceptance of the disorder and its associated disability is provided".

The views cited above support the argument that the existence of stigma should be considered alongside the occupational, educational, environmental, psychological and medical constraints associated with illness and disability.

6.12 Final remarks

The results of the studies above, plus the literature on CFS have shown that a narrow, medicalized view of suffering is clearly inadequate (Charmaz 1983). In order to obtain a more complete and accurate picture of the condition, researchers should take a greater account of subjective information relating to CFS (cf. Toombs 1992). Such a shift-

in-focus is essential, not only to obtain a fuller understanding of the illness-as-lived but also to improve patient care.

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APPENDICES

APPENDIX I

Definition of ME(CFS) and ME(PIFS)

Summary of revised CDC criteria (1994)

Letters and questionnaires for Study 1, 2 and 3

Definition of chronic fatigue syndrome (myalgic encephalomyelitis) used in Study 2

Source: Dowsett et al (1990) and Ramsay (1992).

A syndrome commonly initiated by respiratory infection and/or gastro-intestinal infection but a gradual or more dramatic onset following neurological, cardiac or endocrine disability is recognized.

The cardinal features are:
a complaint of general or local muscular fatigue following minimal exertion with prolonged recovery time; neurological disturbance, especially of cognitive, autonomic and sensory functions; variable involvement of cardiac and other systems; a prolonged relapsing course. There is also a marked variability of symptoms both within and between episodes.

Definition of post-viral/infectious fatigue syndrome used in Study 3

Source: Ho-Yen (1990).

The patient with post-viral fatigue syndrome:
1. Has had generalized, relapsing fatigue exacerbated by minor exercise causing disruption of usual daily activities for at least three months.
2. Complaints of prominent disturbance of concentration and/or short term memory impairment.
3. Has no other obvious, organic causes for a similar syndrome.

Supportive evidence (at least four items from section A, B and C)

A. History

Patient well before illness

An initiating viral* illness (clinical description/viral serology)
Myalgia
Gastrointestinal disturbance
Headaches
Depression
Tinnitus
Paraesthesiae
Sleep disturbance
Cardiovascular complaints
Adverse effect of alcohol
Adverse effect of heat

B. Clinical
Lymphadenopathy
Localized muscle tenderness
Pharyngitis

C. Laboratory
Evidence of viral* infection
Abnormalities in immune function

* Patients with evidence of bacterial or parasitic infections would be diagnosed as having post-infectious fatigue syndrome.

Revised CDC Criteria

Source Fukuda et al 1994.

The revised CDC criteria for CFS require the presence of self-reported persistent or relapsing fatigue lasting 6 or more consecutive months, plus 4 or more of the following symptoms occurring at the same time and post-dating the onset of fatigue:
1. selfreported impairment in short-term memory or concentration severe enough to cause substantial reduction

in previous levels of occupational, educational, social or personal activities

2. sore throat
3. tender cervical or axillary lymph nodes
4. muscle pain
4. multijoint pain without joint swelling or redness
6. headaches of a new type, pattern or severity
7. unrefreshing sleep
8. post-exertional malaise lasting more than 24 hours.

STUDY 1

Letter asking for volunteers.
Letter sent with questionnaires

Copy of questionnaire

Brunel
THE UNIVERSITY OF WEST LONDON

Department of Human Sciences
Head of Department:
John T E Richardson DPhil CPsychol FBPsS

Uxbridge, Middlesex UB8 3PH
United Kingdom
Telephone Uxbridge (0895) 56461
Telex 261173 G Fax: (0895) 32806

20th November 1989

Dear Sir/Madam

I am a postgraduate student at Brunel University interested in the different ways people with Myalgic Encephalomyelitis cope with the effects of this illness.

To this end, I am hoping to talk to eighty sufferers about their experience of this most debilitating condition. If you agree to participate, I would arrange to meet you at a place and time that suits you. The first interview will take about an hour and, to supplement the information you give me, I may ask you to fill in a number of short questionnaires. Then after six months or so, I will meet you again, to see if your condition and views have changed.

If you would like to help this research, please contact me, either by telephoning 01-977-2386 or by returning the slip attached to this letter. The interviews will be taped so I have a detailed record of what you tell me. However for reasons of confidentiality, I will at no time divulge your identity to anyone else.

Thanking you in anticipation

Yours faithfully

Ellen M. Goudsmit
Chartered Psychologist

Name .....

Address .....

.....

.....

Tel. No .....

I would like to help with this research on coping with Myalgic Encephalomyelitis.

Please return to E. Goudsmit, 23 Melbourne Rd, Teddington, Middx TW11 9QX

Brunel
THE UNIVERSITY OF WEST LONDON

Department of Human Sciences
Head of Department:
John T E Richardson DPhil CPsychol FBPsS

Uxbridge, Middlesex UB8 3PH
United Kingdom
Telephone Uxbridge (0895) 56461
Telex 261173 G Fax: (0895) 32806

M.E. QUESTIONNAIRE

Dear

Thank you for volunteering to help me with this research. The enclosed questionnaire may look long and complicated, but please don't be disheartened. It begins by asking for background information about you and your illness. This is followed by some questions asking you to describe, in your own words, your views on its causes and effects. That is what this study is about: M.E. as seen from the sufferers' point of view.

Being a sufferer myself, I know that you may find it difficult to concentrate for long periods of time. I therefore suggest that you do not try to complete the questionnaire in one go but that you complete one or two sections, read the next section, then take a break. If you still have the energy, fill in another section, otherwise leave it for a day or so. As far as the study is concerned, the more you can tell me, the better, so take as much time as you feel you need.

When you've gone through the whole questionnaire, you might like to keep it for a few days before returning it to me. That will give you the chance to reconsider some of the responses or to add to them. As I said before, I am only interested in your views and opinions and, as such, there are no right or wrong answers. All your ideas are relevant. Needless to say, if I quote you in my final report, I will not divulge your identity to anyone.

If you have any queries, please do not hesitate to contact me. If you wish to phone, I am usually around from 2.00 to 8.00pm.

I look forward to hearing from you,

Yours sincerely

ELLEN GOUDSMIT

PLEASE RETURN THE COMPLETED QUESTIONNAIRES TO E. GOUDSMIT, 23 MELBOURNE RD., TEDDINGTON, MIDDX., TW11 9QX IN THE ENCLOSED PREPAID ENVELOPE.

BACKGROUND QUESTIONNAIRE

To help me with my research, I would be grateful if you could answer the following questions.

Name .....

Address.....

.....

Phone No .....

Age.....

Sex.....

Work

Please tick which best describes the work that you do:

Student	Employed part-time
Housewife	Employed full-time
Unemployed	Retired

If you have a job, what is it .....

If you are a woman and you are not employed, what is your husband or partner's job .....

Have you changed your job, or given up your job, as a result of the illness? Please describe .....

Education

Did you complete (please tick)

Secondary education?

College/university?

Other professional training?

Age when you left?

Marital status (Please tick where appropriate)

Single

Married

Living with a partner

Separated

Divorced

Widowed

If your marital status has changed since becoming ill, please describe.....

Children (please state number, sex and age) .....

Housing Are you: (Please tick appropriate answer)

Owner occupier?

Private tenant?

Council tenant?

Do you live alone?

Income

What is your weekly disposable income? Please tick the appropriate answer.

Less than £70

Between £70 and £200

More than £200

GENERAL INFORMATION

1. How long have you had M.E.? .....
2. Was the onset sudden or gradual? .....
3. If acute, did it start as a specific infection such as glandular fever, hepatitis, flu? .....
4. When were you diagnosed? Month.....Year.....
5. Who diagnosed you? .....
6. Which tests were used to diagnose you? (Please tick where appropriate)

   Elisa IgM Coxsackie

   VP1 (Prof. Mowbray's test)

   Other blood test (please specify)

   Muscle biopsy

   Brain scan (if yes, please state which scanner was used)

7. If no laboratory tests were used, how were you diagnosed? (e.g. clinical history, VEGA test etc.)

.....

8. Has the illness (please tick your answer)

   Stayed the same since the beginning?

   Fluctuated, with relapses followed by remissions?

   Generally/steadily deteriorated?

   Improved?

9. What are your main symptoms? .....

.....

.....

.....

M.E. QUESTIONNAIRE PART 2

To help me with my research, I would be grateful if you could answer the following questions as fully as you can. (If you need more space, please use the back of the sheet).

A. CAUSES

1. At the moment, no one knows what causes M.E. In your opinion, which factors played a part in making you ill? Please list as many as you can in the space below.

2. Taking everything you have read and heard about M.E. into account, which factors do you think are likely causes?

3. Taking everything you have read and heard about M.E. into account, which factors do you think are unlikely causes?

B THE EFFECT OF THE ILLNESS

Illness affects people's lives in many ways. I would like to know to what extent and in which ways M.E. has affected those who suffer from it.

1. What can you not do now that you used to be able to do? In this section, I'm particularly interested in the ways M.E. has affected your work and daily life, (e.g. shopping, hobbies).

2. How has the illness affected other people in your life and your relationships with others?

3. How has it affected you as a person, your feelings and attitudes in general; your feelings about yourself?

C. COPING

1. What things have you done, or do you do, to get better and to deal with day-to-day symptoms?

2. What things have helped you deal with the illness in general, so it does not overwhelm you?

D. Can you sum up what having M.E. has meant to you? Please write down the first things that come to mind.

E. How do you see your future?

F. Finally, what advice would you give to a newly diagnosed sufferer?

Thank you very much for your time and energy.

STUDY 2

Letter sent with questionnaires - CFS(ME) group.

Letter asking for volunteers - SCI group.
Letter sent with questionnaires.

Part I. Background questionnaire - CFS(ME) group.
General questionnaire - CFS(ME) group.
Background questionnaire - SCI group.
General questionnaire - SCI group.
Profile of Fatigue-Related Symptoms (PFRS).

Part II. Functional Impairment Scale.
Physical Functioning Scale (from MOS Short-Form).
Question assessing need for help with activities.

Part III. Questionnaire assessing interpersonal resources and stressors (from LISRES).

Part IV. Hospital Anxiety and Depression Scale (HAD).
Mishel Uncertainty in Illness Scale-Form C (MUIS-C) plus additional items assessing unpredictability.

CFS(ME) Group

23 Melbourne Road
TEDDINGTON
Middx.
TW11 9QX

081-977-2386

November 1992

REF: EMG/M.E.

Dear Sir/Madam,

Thank you for volunteering to help me with my research. I know the enclosed package of questionnaires looks daunting but most of the items are short and require little more than a tick. If you can spare some time, I would be most grateful.

The first section begins by asking for background information about you and your illness. This is followed by some questions about the effects which M.E. has had on your social and personal life.

Being a sufferer myself, I know that you may find it difficult to concentrate for long periods of time. I therefore suggest that you do not try and complete all the questionnaires in one go but that you complete one or two, then take a break. If you still have energy left, fill in another one, otherwise leave it for a day or two. Needless to say, all your answers will be strictly confidential.

I hope to obtain data on several illnesses and disabilities because different problems will affect people in different ways.

If you have any queries, please do not hesitate to contact me. If you wish to phone, I'm usually around from 2.00 to 8.00 pm.

Thank you for your time. I look forward to hearing from you,

Yours faithfully,

Ellen M. Goudsmit

Encs. Questionnaires and SAE.

SCI Group

23 Melbourne Road
TEDDINGTON
Middx.
TW11 9QX

October 1992

Dear Sir\Madam,

I am a postgraduate student at Brunel University, researching the effects of having a chronic illness or disability.

Having had to cope with disability myself, I would like to document how different conditions affect people's lives in Britain today. If you would like to help me, I will send you a set of questionnaires which will ask about your disability, daily life, relationships and how you feel about yourself. Most of the questionnaires are quite short and shouldn't take too long to fill in.

I hope to obtain data on several illnesses and disabilities, so that I can compare and contrast the experiences of different groups.

Needless to say, all your answers will be strictly confidential.

If you have any questions, please do not hesitate to call me. I'm usually at home between 2 and 8 pm (081-977-2386).

Thank you for your time. I hope that you can help me and look forward to hearing from you.

Yours faithfully,

Ellen M. Goudsmit

- -

If you are willing to help with my research, please complete this slip and send it to me at the address above, or give me a call.

Name: .....

Address .....

.....

Phone number .....

SCI Group

23 Melbourne Road
TEDDINGTON
Middx.
TW11 9QX

081-977-2386

February 1993

REF: EMG/SCI

Dear

Thank you for volunteering to help me with my research. I know the enclosed package of questionnaires looks daunting but most of the items are short and require little more than a tick. If you can spare some time, I would be most grateful.

The first section begins by asking for background information about you and your disability. This is followed by questions which enquire about the effects of your injury on your social and personal life.

If you find it difficult to concentrate for long periods of time, please don't try and complete all the questionnaires in one go. One of the reasons why I've used differently coloured paper for the sections is that I thought it may help you, should you decide to divide the work over several days. Needless to say, all your answers will be strictly confidential.

I hope to obtain data on several illnesses and disabilities because different problems will affect people in different ways.

If you have any queries, please do not hesitate to contact me. If you wish to phone, I'm usually around from 2.00 to 8.00 pm.

Thank you for your time. I look forward to hearing from you,

Yours sincerely

Ellen M. Goudsmit

Encs. Questionnaires and SAE.

PART I

BACKGROUND QUESTIONNAIRE (M.E.)

SOME QUESTIONS ABOUT YOU AND YOUR ILLNESS

To help me with my research, I would be grateful if you could answer the following questions.

Name .....

Address .....

Phone no. .....

Age .....

Sex .....

Work: Please tick which of the following best describes the work you do:

Student	Employed part-time
Housewife	Employed full-time
Unemployed	Retired
Unemployed - on sick leave	

If you have a job, what is it? .....

If you are not currently employed, what, if any, was your previous job?

.....

.....

Have you changed your job, or have you had to retire because of your illness?

.....

.....

Education:

Did you complete (please tick)

Secondary education?
College/university?
Other professional training?

How many years did you spend studying or training after the age of 16? (Please circle your answer).

Years 0 1 2 3 4 5 6 7 8 9 10 11 12

Marital status (Please tick)

Single

Married

Living with a partner

Separated

Divorced

Widowed

If your marital status has changed since becoming ill, please describe

.....

Children (please state number, age and sex) .....

.....

Housing Are you: (Please tick)

Owner occupier?

Private tenant?

Council/housing association tenant?

Do you live alone? .....

Income

Does your entire income consist of social security benefits? (Please tick)

Yes
No

GENERAL INFORMATION

1. How long have you had M.E.? .....

2. Was the onset sudden or gradual? .....

3. If acute, did it start as a specific infection such as glandular fever, hepatitis, flu? .....

.....

4. When were you diagnosed? 19 ..

5. Who diagnosed you? .....

6. Which tests, if any, were used to diagnose you? (Please tick)

Elisa IgM Coxsackie

VP1 (Prof. Mowbray's test)

Muscle biopsy

Brain scan (if yes, please state which scanner was used)

Other laboratory tests (please specify) .....

7. If no laboratory tests were used, how were you diagnosed? (e.g. clinical history, VEGA test etc.)

.....

8. What are your main symptoms?

.....

.....

.....

9. In the past 6 months, has the illness (please tick the most appropriate answer)

Stayed the same?

Fluctuated, with relapses followed by remissions?

Generally/steadily deteriorated?

Improved?

10. Are you having any treatment/taking any drugs at present, either for M.E. or for another condition?

Yes/No

If yes, please give details .....

.....

11. Which, if any, treatments have you had for M.E. in the past? Please also indicate how many months ago any treatment ended.

Antibiotics	yes/no	..... months ago
Antiviral drugs (eg acyclovir)	yes/no	..... months ago
Antifungal drugs (eg nystatin)	yes/no	..... months ago
Antidepressants	yes/no	..... months ago
Sleeping pills/tranquillisers	yes/no	..... months ago
Homeopathy	yes/no	..... months ago
Acupuncture	yes/no	..... months ago
Allergy treatment	yes/no	..... months ago
Magnesium injections	yes/no	..... months ago
Vitamin/mineral supplements	yes/no	..... months ago
Psychotherapy/counselling	yes/no	..... months ago
Dietary changes	yes/no	..... months ago
Graded exercise/cognitive therapy	yes/no	..... months ago
Inpatient rehabilitation programme (eg Findley, Weir)	yes/no	..... months ago
Any other?	yes*/no	..... months ago

*If yes, please give details.....

.....

12. How much does your illness now affect your everyday life? (please tick)

I can do hardly anything compared with before

I can do about a quarter of what I could do before

I can do about half of what I could do before

I can do about three-quarters of what I could do before

I can do most of what I could do before

13. Do you now have symptoms

all the time?

most of the time?

some of the time?

rarely?

14. Have you been told that your condition will improve?

Yes/No

15. Do you have any other medical problems, aside from M.E.? If yes, please describe.

.....

.....

SCI Group Questionnaire no.

PART I

SOME QUESTIONS ABOUT YOU AND YOUR DISABILITY

To help me with my research, I would be grateful if you could answer the following questions.

Age .....

Sex .....

Work: *Which of the following best describes the work you do* (please tick):

Student	Employed part-time
Housewife	Employed full-time
Unemployed	Retired
Unemployed - on sick leave	

If you have a job, what is it? .....

If you are not currently employed, what, if any, was your previous job?

.....

.....

Have you changed your job, or have you had to retire because of your disability?

.....

.....

Education:

Did you complete (please tick)

Secondary education?

College/university?

Other professional training?

How many years did you spend studying or training after the age of 16? (Please circle your answer).

Years 0 1 2 3 4 5 6 7 8 9 10 11 12

Marital status (Please tick)

Single

Married

Living with a partner

Separated

Divorced

Widowed

If your marital status has changed since you were injured, please describe

.....

Children (please state number, age and sex) .....

.....

Housing Are you: (Please tick)

Owner occupier?

Private tenant?

Council/housing association tenant?

Do you live alone? .....

Income

Does your entire income consist of social security benefits? (Please tick)

Yes
No

GENERAL INFORMATION

1. How long have you been injured? .....

2. What caused the SCI? .....

3. Where exactly is the injury? .....

4. Is your lesion (please tick)

   complete?
   incomplete?

5. Do you mostly use (please tick)

   a wheelchair?
   crutches or other aids?
   both?

6. Do you have injury-related symptoms such as pain, muscle spasms, numbness, regular infections etc? If yes, please list the main ones.

.....

.....

.....

.....

7. Do you have any other medical problems, not due to your injury? If yes, please describe.

.....

.....

.....

8. In the past 6 months, has your condition (please tick the most appropriate answer)

   Stayed the same?
   Fluctuated?
   Generally/steadily deteriorated?
   Improved?

9. How much does your disability now affect your everyday life? (please tick)

   I can do hardly anything compared with before
   I can do about a quarter of what I could do before
   I can do about half of what I could do before
   I can do about three-quarters of what I could do before
   I can do most of what I could do before

10. Apart from problems with mobility and movement, do you experience symptoms (please tick)

all the time?
most of the time?
some of the time?
rarely?

11. Do you take any drugs to manage symptoms like pain, spasms etc? (For instance Baclofen).

Yes
No

If yes, please give details .....

.....

12. Have you been told that your condition could improve? (Please tick)

Yes
No

YOUR SYMPTOMS IN THE PAST WEEK

Below is a list of problems which may or may not apply to you. For each problem, please say to what extent you have experienced this during the **PAST WEEK** (including today). Do not think for too long before answering but give your immediate reaction. Please be careful not to miss out any of the items. Remember, we are talking about the past week and not your condition in general. Give your answer by circling any number from 0 to 6, to the right of the item, where

0 = not at all

3 = moderately

6 = extremely

Feeling physically tired even when taking things easy	0	1	2	3	4	5	6
Your limbs feeling heavy	0	1	2	3	4	5	6
Getting easily upset by things	0	1	2	3	4	5	6
Difficulty concentrating	0	1	2	3	4	5	6
Stomach pain	0	1	2	3	4	5	6
Not having the physical energy to do anything	0	1	2	3	4	5	6
Difficulty standing for long	0	1	2	3	4	5	6
Losing your temper easily	0	1	2	3	4	5	6
Difficulty remembering things	0	1	2	3	4	5	6
Muscles feeling weak even after resting	0	1	2	3	4	5	6
Feeling depressed	0	1	2	3	4	5	6
Muscles tender to the touch	0	1	2	3	4	5	6
Slowness of thought	0	1	2	3	4	5	6
Tremor or twitching	0	1	2	3	4	5	6
The slightest exercise making you physically tired	0	1	2	3	4	5	6
Being irritable	0	1	2	3	4	5	6

	0 = not at all 3 = moderately 6 = extremely						
Difficulty reasoning things out	0	1	2	3	4	5	6
Burning, tingling or crawling sensations	0	1	2	3	4	5	6
Numbness in some part of your body	0	1	2	3	4	5	6
Back pain	0	1	2	3	4	5	6
Feeling anxious	0	1	2	3	4	5	6
A feeling of confusion ("mental fog")	0	1	2	3	4	5	6
Bouts of sweating (day or night)	0	1	2	3	4	5	6
Feeling physically drained	0	1	2	3	4	5	6
Dizziness or giddiness	0	1	2	3	4	5	6
Absent-mindedness	0	1	2	3	4	5	6
Worrying about things that do not matter	0	1	2	3	4	5	6
Feeling physically tired even after a good night's sleep	0	1	2	3	4	5	6
Difficulty understanding e.g. what someone was saying to you	0	1	2	3	4	5	6
Feeling pessimistic about the future	0	1	2	3	4	5	6
Cold hands or feet	0	1	2	3	4	5	6
Having to stop doing something, that was easy in itself, because it made you tired	0	1	2	3	4	5	6
Muscles feeling weak after slight exercise	0	1	2	3	4	5	6
Difficulty following things e.g. a simple plot on TV	0	1	2	3	4	5	6
Hot or cold spells	0	1	2	3	4	5	6

	0 = not at all 3 = moderately 6 = extremely						
Feeling tense	0	1	2	3	4	5	6
Feeling faint	0	1	2	3	4	5	6
Difficulty finding the right word	0	1	2	3	4	5	6
Feeling chilled or shivery	0	1	2	3	4	5	6
Tearfulness	0	1	2	3	4	5	6
Irregular or rapid heart beats	0	1	2	3	4	5	6
Feeling worthless	0	1	2	3	4	5	6
Forgetting what you were trying to say	0	1	2	3	4	5	6
Being easily angered when things went wrong	0	1	2	3	4	5	6
Feeling mentally tired even after a good night's sleep	0	1	2	3	4	5	6
Diarrhoea or constipation	0	1	2	3	4	5	6
Feeling nervous	0	1	2	3	4	5	6
Feeling sad	0	1	2	3	4	5	6
The slightest effort making you mentally tired	0	1	2	3	4	5	6
Feeling like you had a temperature	0	1	2	3	4	5	6
Other people annoying you	0	1	2	3	4	5	6
A sore throat	0	1	2	3	4	5	6
Feelings of resentment	0	1	2	3	4	5	6
Being slow to react	0	1	2	3	4	5	6

PART II

THE EFFECTS OF YOUR ILLNESS/DISABILITY

A. GENERAL FUNCTIONING

Please circle a number for the items on this page to indicate how your illness/disability has affected different areas of your life.

1. **WORK**

Because of my illness/disability, my ability to work/go to school is impaired:

0	1	2	3	4	5	6	7	8
not at all		slightly		definitely		markedly		very severely I cannot work

2. **HOME MANAGEMENT**
(cleaning, tidying, shopping, cooking, looking after home or children, paying bills)

Because of my illness/disability, my ability to look after my home is impaired:

0	1	2	3	4	5	6	7	8
not at all		slightly		definitely		markedly		very severely I cannot do it

3. **SOCIAL LEISURE ACTIVITIES**
(<u>with other people</u>, e.g. going to parties, pubs, clubs, outings, visits, dating, home entertainments)

Because of my illness/disability, my social life is impaired:

0	1	2	3	4	5	6	7	8
not at all		slightly		definitely		markedly		very severely I never do these

4. **PRIVATE LEISURE ACTIVITIES**
(<u>done alone</u>, e.g. reading, gardening, collecting, sewing)

Because of my illness/disability, my private leisure activities have been affected:

0	1	2	3	4	5	6	7	8
not at all		slightly		definitely		markedly		very severely I never do these

B. SPECIFIC PHYSICAL ACTIVITIES

How, if at all, has your condition limited you in each of the following activities?
(Please tick one box on each line)

	Severely limited	Somewhat limited	Not limited at all
1. The kinds or amounts of vigorous activities you can do, like lifting heavy objects, running or participating in strenuous sports	☐	☐	☐
2. The kinds or amounts of moderate activities you can do, like moving a table, carrying groceries or bowling	☐	☐	☐
3. Walking uphill or climbing a few flights of stairs.	☐	☐	☐
4. Bending, lifting or stooping	☐	☐	☐
5. Walking to the end of the street (without aids)	☐	☐	☐
6. Eating, dressing, bathing, or using the toilet	☐	☐	☐

C. HELP WITH ACTIVITIES

Which of the following activities are so difficult for you that in most instances, you either enlist help from another person or you use a specialised piece of equipment/aid to assist you? (please tick).

Getting in/out of bed	Cooking
Eating	Getting dressed
Toileting	Driving
Writing	Working

PART III

THE PEOPLE IN YOUR LIFE

FRIENDS

Here are some questions about your friends. (Please do not include your relatives, or spouse/partner as friends when answering these questions.)

1. How many close friends do you have, people you feel at ease with and can talk to about personal matters?

[]None [] One [] Two [] Three [] Four or more

2. How often are you in touch with the friend or friends to whom you feel closest?

[]Never [] Less than once a month [] Once or twice a month [] Once a week [] Several times a week

3. Did you lose friends as a result of your illness/disability?

[]None [] Some [] Most

4. Here are some more questions about your friends. Please indicate how often these things happen with your friends.

How often:	Never	Seldom	Some-times	Fairly Often	Often
Can you count on your friends to help you when you need it?	[]	[]	[]	[]	[]
Do your friends cheer you up when you are sad or worried?	[]	[]	[]	[]	[]
Do you confide in any of your friends?	[]	[]	[]	[]	[]
Do you share mutual interests or activities with your friends?	[]	[]	[]	[]	[]
Do any of your friends disagree with you about important things?	[]	[]	[]	[]	[]
Are any of your friends critical or disapproving of you?	[]	[]	[]	[]	[]
Do your friends really understand how you feel about things?	[]	[]	[]	[]	[]
Do any of your friends get on your nerves?	[]	[]	[]	[]	[]
Do any of your friends get angry or lose their temper with you?	[]	[]	[]	[]	[]
Do your friends respect your opinion?	[]	[]	[]	[]	[]
Do any of your friends expect too much of you?	[]	[]	[]	[]	[]

SPOUSE/PARTNER

5. The following questions concern your current relationship with your spouse/partner. For each question, please indicate how often these things happen with your spouse/partner.

If you do not have a spouse or partner, please move on to the next section and mark this page with the letters NA.

How often:	Never	Seldom	Some-times	Fairly Often	Often
Can you count on him or her to help you when you need it?	[]	[]	[]	[]	[]
Does he or she cheer you up when you are sad or worried?	[]	[]	[]	[]	[]
Do you confide in him or her?	[]	[]	[]	[]	[]
Do you share mutual interests or activities with him or her?	[]	[]	[]	[]	[]
Does he or she disagree with you about important things?	[]	[]	[]	[]	[]
Is he or she critical or disapproving of you?	[]	[]	[]	[]	[]
Does he or she really understand how you feel about things?	[]	[]	[]	[]	[]
Does he or she get on your nerves?	[]	[]	[]	[]	[]
Does he or she get angry or lose their temper with you?	[]	[]	[]	[]	[]
Does he or she respect your opinion?	[]	[]	[]	[]	[]
Does he or she expect too much of you?	[]	[]	[]	[]	[]

RELATIVES

Here are some questions about your close relatives, other than your spouse/partner.

6. During the last month, how often did you get together with one or more of your relatives (those that do not live with you)?

Not at	Less	Once or	Once a	Several
[]all	than	[]twice	[]week	times a
	[]once			[]week

7. How many relatives do you feel close to - that is, relatives you feel at ease with and can talk to about personal matters?

				Four or
[]None	[]One	[]Two	[]Three	[]more

8. When you spend time with your relatives, including those living with you, how often do these things happen?

How often:	Never	Seldom	Some-times	Fairly Often	Often
Can you count on any of your relatives to help you when you need it?	[]	[]	[]	[]	[]
Do any of your relatives cheer you up when you are sad or worried?	[]	[]	[]	[]	[]
Do you confide in any of your relatives?	[]	[]	[]	[]	[]
Do you share mutual interests or activities with any of your relatives?	[]	[]	[]	[]	[]
Do any of your relatives disagree with you about important things?	[]	[]	[]	[]	[]
Are any of your relatives critical or disapproving of you?	[]	[]	[]	[]	[]
Do any of your relatives really understand how you feel about things?	[]	[]	[]	[]	[]
Do any of your relatives get on your nerves?	[]	[]	[]	[]	[]
Do any of your relatives get angry or lose their temper with you?	[]	[]	[]	[]	[]
Do any of your relatives respect your opinion?	[]	[]	[]	[]	[]
Do any of your relatives expect too much of you?	[]	[]	[]	[]	[]

PART IV

YOUR MOODS

The following questions will give me an indication of how you have been feeling recently. Please read each item and put a tick beside the response which comes closest to how you have felt in the last few days.

Don't take too long over your replies; your immediate reaction will probably be more accurate than a long thought out response.

I feel tense or 'wound up'
- Most of the time
- A lot of the time
- From time to time, occasionally
- Not at all

I still enjoy the things I used to enjoy
- Definitely as much
- Not quite so much
- Only a little
- Hardly at all

I get a sort of frightened feeling as if something awful is about to happen
- Very definitely and quite badly
- Yes, but not too badly
- A little, but it doesn't worry me
- Not at all

I can laugh and see the funny side of things
- As much as I always could
- Not quite so much now
- Definitely not so much now
- Not at all

Worrying thoughts go through my mind
- A great deal of the time
- A lot of the time
- From time to time but not too often
- Only occasionally

I feel cheerful
- Not at all
- Not often
- Sometimes
- Most of the time

I can sit at ease and feel relaxed
- Definitely
- Usually
- Not often
- Not at all

I feel as if I am slowed down
- Nearly all the time
- Very often
- Sometimes
- Not at all

I get a sort of frightened feeling like 'butterflies' in the stomach
- Not at all
- Occasionally
- Quite often
- Very often

I have lost interest in my appearance
- Definitely
- I don't take so much care as I should
- I may not take quite as much care
- I take just as much care as ever

I feel restless as if I have to be on the move
- Very much indeed
- Quite a lot
- Not very much
- Not at all

I look forward with enjoyment to things
- As much as ever I did
- Rather less than I used to
- Definitely less than I used to
- Hardly at all

I get sudden feelings of panic
- Very often indeed
- Quite often
- Not very often
- Not at all

I can enjoy a good book or radio or TV programme
- Often
- Sometimes
- Not often
- Very seldom

THE UNCERTAINTY ASSOCIATED WITH ILLNESS AND DISABILITY

Instructions: Please read each statement, taking your time. Then place a tick under the description that most closely matches how you are feeling TODAY. For instance, if you agree with the statement, tick under "Strongly agree" or "Agree". If you disagree with a statement, then tick under either "Strongly disagree" or "Disagree". If you are undecided about how you feel, then tick under "Undecided".

This questionnaire was devised to cover a range of conditions. Please respond to every statement, relating it as best you can to your own condition.

Where a question refers to treatment or medical advice and you are no longer being treated for your condition, please take your last consultations as an example. If you cannot recall these, just mark that statement with the letters NA (not applicable).

1. I don't know what's wrong with me.

Strongly agree	Agree	Undecided	Disagree	Strongly Disagree
-----	-----	-----	-----	-----

2. I have a lot of questions without answers.

Strongly agree	Agree	Undecided	Disagree	Strongly Disagree
-----	-----	-----	-----	-----

3. I am unsure if my condition is getting better *or worse*.

Strongly agree	Agree	Undecided	Disagree	Strongly Disagree
-----	-----	-----	-----	-----

4. It is unclear how bad my main symptoms might be.

Strongly agree	Agree	Undecided	Disagree	Strongly Disagree
-----	-----	-----	-----	-----

5. The explanations they give about my condition seem hazy to me.

Strongly agree	Agree	Undecided	Disagree	Strongly Disagree
-----	-----	-----	-----	-----

6. The purpose of each treatment/piece of advice is clear to me.

Strongly agree	Agree	Undecided	Disagree	Strongly Disagree
-----	-----	-----	-----	-----

7. My symptoms continue to change unpredictably.

Strongly agree	Agree	Undecided	Disagree	Strongly Disagree
-----	-----	-----	-----	-----

8. I understand everything explained to me.

Strongly agree	Agree	Undecided	Disagree	Strongly Disagree
-----	-----	-----	-----	-----

9. The doctors say things to me that could have many meanings.

Strongly agree	Agree	Undecided	Disagree	Strongly Disagree
-----	-----	-----	-----	-----

10. My treatment/the advice I'm given is too complex to figure out.

Strongly agree	Agree	Undecided	Disagree	Strongly Disagree
-----	-----	-----	-----	-----

11. It is difficult to know if the treatment/advice I am getting is helping.

Strongly agree	Agree	Undecided	Disagree	Strongly Disagree
-----	-----	-----	-----	-----

12. Because of the unpredictability of my condition, I cannot plan for the future.

Strongly agree	Agree	Undecided	Disagree	Strongly Disagree
-----	-----	-----	-----	-----

13. My symptoms keep changing. I have good and bad days.

Strongly agree	Agree	Undecided	Disagree	Strongly Disagree
-----	-----	-----	-----	-----

14. I have been given many different opinions about what is wrong with me.

Strongly agree	Agree	Undecided	Disagree	Strongly Disagree
-----	-----	-----	-----	-----

15. It is not clear what is going to happen to me.

Strongly agree	Agree	Undecided	Disagree	Strongly Disagree
-----	-----	-----	-----	-----

16. The results of my tests are inconsistent.

Strongly agree	Agree	Undecided	Disagree	Strongly Disagree
-----	-----	-----	-----	-----

17. The effectiveness of the treatment/advice is undetermined.

Strongly agree	Agree	Undecided	Disagree	Strongly Disagree
-----	-----	-----	-----	-----

18. Because of the treatment/advice, what I can do and cannot do keeps changing.

Strongly agree	Agree	Undecided	Disagree	Strongly Disagree
-----	-----	-----	-----	-----

19. I'm certain they will not find anything else wrong with me.

Strongly agree	Agree	Undecided	Disagree	Strongly Disagree
-----	-----	-----	-----	-----

20. The treatment/advice I am receiving has a known probability of success.

Strongly agree	Agree	Undecided	Disagree	Strongly Disagree
-----	-----	-----	-----	-----

21. They have not given me a specific diagnosis.

Strongly agree	Agree	Undecided	Disagree	Strongly Disagree
-----	-----	-----	-----	-----

22. The seriousness of my condition has been determined.

Strongly agree	Agree	Undecided	Disagree	Strongly Disagree
-----	-----	-----	-----	-----

23. The doctors and nurses use everyday language so I can understand what they are saying.

Strongly agree	Agree	Undecided	Disagree	Strongly Disagree
-----	-----	-----	-----	-----

24. I can predict how long my condition will last.

Strongly agree	Agree	Undecided	Disagree	Strongly Disagree
-----	-----	-----	-----	-----

25. I usually know if I am going to have a good or bad day.

Strongly agree	Agree	Undecided	Disagree	Strongly Disagree
-----	-----	-----	-----	-----

26. I can generally predict the course of my condition.

Strongly agree	Agree	Undecided	Disagree	Strongly Disagree
-----	-----	-----	-----	-----

27. My physical distress is predictable; I know when it is going to get better or worse.

Strongly agree	Agree	Undecided	Disagree	Strongly Disagree
-----	-----	-----	-----	-----

Many thanks for your time and energy.

- -

Most of these questions have asked about problems which might have arisen as a result of your condition. If there are any difficulties that you have experienced but which have not been mentioned, you might like to indicate these briefly in the space below. I would also be interested to learn of any positive aspects of your illness or disability.

STUDY 3

Letter asking for volunteers (Patient Information Sheet). Consent Form.

Time 1

Letter sent with questionnaires.

Part I. Background questionnaire (sent with PFRS).

Part II. General assessment (course of illness, level of activity, frequency of symptoms).
Also sent: Functional Impairment Scale.

Part III. Illness Management Questionnaire (IMQ).
Self-Efficacy Scale.

Part IV. Mishel Uncertainty in Illness Scale Form C (MUIS-C). Also sent: HAD.

Time 2

Letter sent with questionnaires (wording for treatment group was similar *except when referring to date of next* consultation).

Follow-up questionnaire (new treatments, level of activity, frequency of symptoms, reasons for improvement and support from medical profession). Also sent: PFRS, Functional Impairment Scale, IMQ, Self-Efficacy Scale, HAD, MUIS-C.

Raigmore Hospital NHS Trust
Perth Road
Inverness IV2 3UJ

Tel: 0463 704000 Fax: 0463 711322

Raigmore Hospital NHS TRUST

Microbiology Department ext 4206/7

Patient Information Sheet

Dear

You have been referred by your doctor to the Post Viral Fatigue Syndrome clinic in the Microbiology Department at Raigmore Hospital. As you may know, Dr Ho-Yen is involved in several aspects of research into Post Viral Fatigue Syndrome, mainly in the management and the laboratory investigations that may be useful in diagnosing such patients. At the moment, he is involved in a study with Ms Ellen Goudsmit, a postgraduate student at Brunel University into how patients cope with their illness.

Dr Ho-Yen and Ms Goudsmit would be very grateful for your help in this study. The study is composed of several questionnaires for you to answer at various times. Please find enclosed the questionnaires. We recognise that this will take some time and we are very grateful for your cooperation. You may find it easier not to attempt to do all of the questionnaires at the same time. However, we would be very grateful if you would make the effort to complete the questionnaires over the next week. Please return the questionnaire in the enclosed envelope. Over the next year, you will be given a further two sets of questionnaires so that we can establish the progress that you have made.

You are under no obligation to take part in this study, and you are completely free to withdraw at any time you wish and this will not affect your continuing medical management in any way.

Needless to say, all you answers will be strictly confidential.

Dr Ho-Yen will telephone you over the next few days so the he can answer any questions that you may have.

Your cooperation is greatly valued.

With best wishes,

Yours sincerely,

ELLEN GOUDSMIT DR DARREL HO-YEN

CONSENT FORM

CONSENT BY PATIENT/VOLUNTEER TO PARTICIPATE IN .....

.....

Name of Patient/Volunteer : .....

Name of Study : .....

Principal Investigator : .....

I have read the patient/volunteer information sheet on the above study and have had the opportunity to discuss the details with ..... and ask questions. The doctor has explained to me the nature and purpose of the tests to be undertaken. I understand to my satisfaction what is proposed to be done and that I am under no obligation to take part in this study.

I have agreed to take part in the study as it has been outlined to me, but I understand that I am completely free to withdraw from the study at any time I wish and that this will not affect my continuing medical treatment in any way.

I understand that these trials are part of a research project designed to promote medical knowledge, which has been approved by the Ethics Committee, and may be of no benefit to me personally.

I also understand that where appropriate my General Practitioner will be informed that I have taken part in this study.

I readily and freely consent to participate in the study which has been satisfactorily explained to me.

Signature of Patient/Volunteer : .....

Date : .....

I confirm that I have explained to the patient/volunteer named above, the nature and purpose of the tests to be undertaken.

Signature of Investigator : .....

Date : .....

23 Melbourne Road
TEDDINGTON
Middx.
TW11 9QX

081-977-2386

February 1994

REF: EMG/HY.

Dear Sir/Madam,

Thank you for volunteering to help Dr. Ho-Yen and myself with our research. I know the enclosed package of questionnaires looks daunting but most of the items are short and require little more than a tick. If you can spare some time and complete these questionnaires within the next week or so, we would be most grateful.

The first section begins by asking for background information about you and your illness. This is followed by some questions about the ways your condition has affected your daily life and the extent to which you can control your symptoms. Finally, there are some questions which will tell us how you have tried to cope with your illness so far, if the advice and explanations you have been given are clear, and whether the symptoms are predictable or not.

We know that you may find it difficult to concentrate for long periods of time. We therefore suggest that you do not try and complete all the questionnaires in one go but that you complete one or two, then take a break. If you still have energy left, fill in another one, otherwise leave it for a day or two. After you've finished, please check that you've filled in every page. All your answers will be strictly confidential.

We are aware that some of the questions appear similar to others. However, please bear with us! It's important that you respond to every item on each of the questionnaires; otherwise we will not be able to analyse the results.

If you have any queries, please do not hesitate to contact me. If you wish to phone, I'm usually around from 2.00 to 8.00 pm.

Thank you for your time. I look forward to hearing from you,

Yours faithfully,

Ellen M. Goudsmit

Encs. Questionnaires and SAE.

Questionnaire No.

PART I

BACKGROUND QUESTIONNAIRE

SOME QUESTIONS ABOUT YOU AND YOUR ILLNESS

Age .....

Sex .....

Work: Please tick which of the following best describes the work you do:

Student	Employed part-time
Housewife	Employed full-time
Unemployed	Retired
Unemployed - on sick leave	

If you have a job, what is it? .....

If you are not currently employed, what, if any, was your previous job?

.....

.....

Have you changed your job, reduced your hours or have you had to retire because of your illness? (Please tick)

Yes

No

Education:

Did you complete (please tick)

Secondary education?	yes	no
College/university?	yes	no
Other professional training?	yes	no

How many years, if any, did you spend studying or training from the age of 16? (Please circle your answer)

Years 0 1 2 3 4 5 6 7 8 9 10 11 12

Marital status (please tick)

Single

Married

Living with a partner

Separated or divorced

Widowed

Has your marital status changed as a result of your illness? (Please tick)

Yes

No

Children (please state number and age) .....

.....

Housing Are you: (please tick)

Owner occupier?

Private tenant?

Council/housing association tenant?

Do you live alone? .....

Income

Does your entire income consist of social security benefits? (Please tick)

Yes

No

GENERAL INFORMATION

1. How long have you been ill? .....

2. Was the onset sudden or gradual? .....

3. If sudden, did it follow a specific infection such as glandular fever, hepatitis, flu? .....

.....

4. What are your main symptoms?

.....

.....

.....

5. Have you already been given a diagnosis? (Please tick)

Yes

No

6. If yes, what is the diagnosis? .....

7. When were you diagnosed? month .... year ....

If you have not yet received a diagnosis, please tick ...

8. Who made the diagnosis? .....

If not applicable, please tick ...

9. Have any laboratory tests been used to diagnose your illness?

Yes

No

10. Have you read Dr. Ho-Yen's book 'Better Recovery from Viral Illness'?

Yes

No

11. If yes, have you been following his advice concerning rest, exercise, and avoiding stress etc? (Please tick the most appropriate answer)

Not yet

Sometimes

Most of the time

All the time

12. Are you a member of a self-help/support group for people with PVFS/M.E.?

Yes

No

13. Are you having any treatment/taking any drugs <u>at present</u>?

Yes

No

If yes, please give details .....

14. Which, if any, treatments have you had for your illness <u>in the past?</u> Please also indicate how many months ago any treatment ended.

Antibiotics	yes/no	..... months ago
Antiviral drugs (eg acyclovir)	yes/no	..... months ago
Antifungal drugs (eg nystatin)	yes/no	..... months ago
Antidepressants	yes/no	..... months ago
Sleeping pills/tranquillisers	yes/no	..... months ago
Homeopathy	yes/no	..... months ago
Allergy treatment	yes/no	..... months ago
Vitamin/mineral supplements	yes/no	..... months ago
Dietary changes	yes/no	..... months ago
Psychotherapy/counselling	yes/no	..... months ago
Graded exercise/cognitive therapy	yes/no	..... months ago
Any other?	yes*/no	..... months ago

*If yes, please give details .....

15. Have you been told that your present condition will improve? (Please tick)

Yes

No

16. Do you have any other medical problems, aside from your present illness?

Yes

No

If yes, please describe.

.....

.....

PART II

GENERAL ASSESSMENT

1. In the past 6 months, has the illness (please tick the most appropriate answer)

   improved a lot?

   improved a little?

   stayed the same?

   worsened a little?

   worsened a lot?

2. Do you now have symptoms

   rarely?

   some of the time?

   most of the time?

   all the time?

3. How much does your illness now affect your everyday life?

   I can do most of what I could do before

   I can do about three-quarters of what I could do before

   I can do about half of what I could do before

   I can do about a quarter of what I could do before

   I can do hardly anything compared with before

PART III

DEALING WITH YOUR ILLNESS

Listed below are a number of ways in which people may deal with their illness. These include things that they feel and tell themselves, and things that they do or avoid doing. Think of your own experience of being ill, and say to what extent you respond to your illness in the way described by each of the statements below.

There are no right or wrong answers: say what is TRUE FOR YOU. Your feelings and the things you do may vary from time to time. In your answers, describe your overall response to your illness DURING THE PAST MONTH.

For each statement, circle a number where

1 = never
2 = almost never
3 = sometimes
4 = quite often
5 = very often
6 = always

I make myself carry on, despite how I feel	1	2	3	4	5	6
I try to do the things I did before my illness	1	2	3	4	5	6
I avoid emotionally stressful situations	1	2	3	4	5	6
I plan my activities carefully, to take account of my limitations	1	2	3	4	5	6
I deliberately break "the rules" to give my spirits a lift	1	2	3	4	5	6
I tell myself I can overcome the fatigue	1	2	3	4	5	6
I tell myself I don't feel too bad	1	2	3	4	5	6
You have to realise you are helpless in the face of this illness	1	2	3	4	5	6
I organise my life to avoid overdoing things	1	2	3	4	5	6
I push myself to stay active.	1	2	3	4	5	6
I tell myself I can't let my symptoms stand in the way of what I want to do	1	2	3	4	5	6

1 = never	**4 = quite often**
2 = almost never	**5 = very often**
3 = sometimes	**6 = always**

I try not to think about my illness	1	2	3	4	5	6
I think a great deal about my symptoms	1	2	3	4	5	6
I try to strike a balance between resting and being active	1	2	3	4	5	6
You have to realise that your life is ruled by the illness	1	2	3	4	5	6
I try not to pay attention to my symptoms	1	2	3	4	5	6
I find out as much as I can about this illness	1	2	3	4	5	6
From my own experience, I feel I know what is best for me to do	1	2	3	4	5	6
You have to give up trying to lead a normal life	1	2	3	4	5	6
My symptoms are always at the back of my mind	1	2	3	4	5	6
I manage my time so that I don't have to do too much in one day	1	2	3	4	5	6
I do something regardless of how it affects my symptoms	1	2	3	4	5	6
I look for new information about this illness	1	2	3	4	5	6
I try to pretend my symptoms aren't there	1	2	3	4	5	6
I try to make my life stress-free	1	2	3	4	5	6
I push myself until I can do no more.	1	2	3	4	5	6
I plan my day so that there are times when I am active and times when I can rest	1	2	3	4	5	6
I control my negative feelings	1	2	3	4	5	6

1 = never	4 = quite often
2 = almost never	5 = very often
3 = sometimes	6 = always

I look up medical information	1	2	3	4	5	6
Even if I feel ill, I just keep going	1	2	3	4	5	6
I try to ignore my symptoms	1	2	3	4	5	6
My illness is the most significant thing in my life	1	2	3	4	5	6
I am constantly aware of how I am feeling	1	2	3	4	5	6
Even though unwell, I just go on as if I was feeling OK	1	2	3	4	5	6
I do something I want to do, even though I know I will feel worse after	1	2	3	4	5	6
I make sure that I don't overdo things	1	2	3	4	5	6
I follow the advice of others	1	2	3	4	5	6
I select an approach to my illness and persevere with that	1	2	3	4	5	6
I pay close attention to how well or badly I am feeling	1	2	3	4	5	6
I read books and articles about my illness	1	2	3	4	5	6
I spend a lot of time thinking about my illness	1	2	3	4	5	6
I try to control how much stress there is in my life	1	2	3	4	5	6
I try to keep some energy "in reserve" in case I need it	1	2	3	4	5	6
I take a chance and do something, even though I may feel worse later	1	2	3	4	5	6
I try anything I hear of that might help me to get better	1	2	3	4	5	6

CONTROLLING THE ILLNESS

In the following questions, we'd like to know how you feel about your ability to control your illness. For each of the following questions, please circle the number which corresponds to the certainty that you can now manage your symptoms and perform the following activities or tasks.

1. How certain are you that you can control your fatigue?

10	20	30	40	50	60	70	80	90	100
very uncertain				moderately uncertain					very certain

2. How certain are you that you can regulate your activity, so as to be active without aggravating your symptoms?

10	20	30	40	50	60	70	80	90	100
very uncertain				moderately uncertain					very certain

3. How certain are you that you can do something to help yourself feel better if you are feeling down?

10	20	30	40	50	60	70	80	90	100
very uncertain				moderately uncertain					very certain

4. As compared with other people with PVFS, how certain are you that you can manage the fatigue during your daily activities?

10	20	30	40	50	60	70	80	90	100
very uncertain				moderately uncertain					very certain

5. How certain are you that you can manage all your main symptoms so that you can do the things you enjoy doing?

10	20	30	40	50	60	70	80	90	100
very uncertain				moderately uncertain					very certain

6. How certain are you that you can deal with the frustration of your illness?

10	20	30	40	50	60	70	80	90	100
very uncertain				moderately uncertain					very certain

PART IV

THE UNCERTAINTY ASSOCIATED WITH ILLNESS AND DISABILITY

Instructions: Please read each statement, taking your time. Then place a tick under the description that most closely matches how you are feeling TODAY. For instance, if you agree with the statement, tick under "Strongly agree" or "Agree". If you disagree with a statement, then tick under either "Strongly disagree" or "Disagree". If you are undecided about how you feel, then tick under "Undecided".

This questionnaire was devised to cover a range of conditions. Please respond to every statement, relating it as best you can to your own illness.

If you have not yet received any treatment or medical advice for your illness, just mark statements relating to treatment with the letters NA (not applicable).

1. I don't know what is wrong with me.

Strongly agree	Agree	Undecided	Disagree	Strongly Disagree
-----	-----	-----	-----	-----

2. I have a lot of questions without answers.

Strongly agree	Agree	Undecided	Disagree	Strongly Disagree
-----	-----	-----	-----	-----

3. I am unsure if my illness is getting better or worse.

Strongly agree	Agree	Undecided	Disagree	Strongly Disagree
-----	-----	-----	-----	-----

4. It is unclear how bad my main symptoms will be.

Strongly agree	Agree	Undecided	Disagree	Strongly Disagree
-----	-----	-----	-----	-----

5. The explanations they give about my condition seem hazy to me.

Strongly agree	Agree	Undecided	Disagree	Strongly Disagree
-----	-----	-----	-----	-----

6. The purpose of each treatment/piece of advice is clear to me.

Strongly agree	Agree	Undecided	Disagree	Strongly Disagree
-----	-----	-----	-----	-----

7. My symptoms continue to change unpredictably.

Strongly agree	Agree	Undecided	Disagree	Strongly Disagree
-----	-----	-----	-----	-----

8. I understand everything explained to me.

Strongly agree	Agree	Undecided	Disagree	Strongly Disagree
-----	-----	-----	-----	-----

9. The doctors say things to me that could have many meanings.

Strongly agree	Agree	Undecided	Disagree	Strongly Disagree
-----	-----	-----	-----	-----

10. My treatment/the advice I'm given is too complex to figure out.

Strongly agree	Agree	Undecided	Disagree	Strongly Disagree
-----	-----	-----	-----	-----

11. It is difficult to know if the treatment/advice I am getting is helping.

Strongly agree	Agree	Undecided	Disagree	Strongly Disagree
-----	-----	-----	-----	-----

12. Because of the unpredictability of my illness, I cannot plan for the future.

Strongly agree	Agree	Undecided	Disagree	Strongly Disagree
-----	-----	-----	-----	-----

13. The course of my illness keeps changing. I have good and bad days.

Strongly agree	Agree	Undecided	Disagree	Strongly Disagree
-----	-----	-----	-----	-----

14. I have been given many different opinions about what is wrong with me.

Strongly agree	Agree	Undecided	Disagree	Strongly Disagree
-----	-----	-----	-----	-----

15. It is not clear what is going to happen to me.

Strongly agree	Agree	Undecided	Disagree	Strongly Disagree
-----	-----	-----	-----	-----

16. The results of my tests are inconsistent.

Strongly agree	Agree	Undecided	Disagree	Strongly Disagree
-----	-----	-----	-----	-----

17. The effectiveness of the treatment/advice is undetermined.

Strongly agree	Agree	Undecided	Disagree	Strongly Disagree
-----	-----	-----	-----	-----

18. Because of the treatment/advice, what I can do and cannot do keeps changing.

Strongly agree	Agree	Undecided	Disagree	Strongly Disagree
-----	-----	-----	-----	-----

19. I'm certain they will not find anything else wrong with me.

Strongly agree	Agree	Undecided	Disagree	Strongly Disagree
-----	-----	-----	-----	-----

20. The treatment/advice I am receiving has a known probability of success.

Strongly agree	Agree	Undecided	Disagree	Strongly Disagree
-----	-----	-----	-----	-----

21. They have not given me a specific diagnosis.

Strongly agree	Agree	Undecided	Disagree	Strongly Disagree
-----	-----	-----	-----	-----

22. The seriousness of my illness has been determined.

Strongly agree	Agree	Undecided	Disagree	Strongly Disagree
-----	-----	-----	-----	-----

23. The doctors and nurses use everyday language so I can understand what they are saying.

Strongly agree	Agree	Undecided	Disagree	Strongly Disagree
-----	-----	-----	-----	-----

Many thanks for your time and energy.

- -

If you have any comments to make on any of the questionnaires, or if you would like to add information which you think might be relevant, please do so here.

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Raigmore Hospital NHS TRUST

Microbiology Department ext 4206/7

DHY\LW\Q2

Dear

You agreed to take part in the research project on how patients cope with their illness. It is hoped that the results of this study will enable us to determine how to best manage patients.

You were sent a questionnaire when you joined the waiting list. You are about to have your first appointment and it is very helpful for us to look at how you have progressed over the period.

Please find enclosed a second questionnaire which we would like you to fill up. Please send the questionnaire back to Dr Ho-Yen, Microbiology Department, Raigmore Hospital. If there are any problems, please do not hesitate to contact me.

Yours sincerely,

LORNA WYCHERLEY
Secretary to Dr Ho-Yen

No:

FOLLOW-UP QUESTIONNAIRE

PART I

In the last five to six months, have you felt (please tick the most appropriate answer)

much better?

better?

the same?

worse than before?

much worse than before?

In the last 5 or 6 months, have you begun any of the following treatments or remedies? (Please tick appropriate answer)

	Yes	No
supplements		
anti-candida or allergy diet		
reflexology		
acupuncture		
homeopathy		
antidepressants (please name)		
sleeping pills (please name)		
tranquillisers (please name)		
antifungal medication		
antibiotics		
other (please describe)		

.....

If you have tried any of the above, please indicate if you found it (them) helpful.

Treatment	helpful	not helpful
.....		
.....		
.....		

GENERAL ASSESSMENT

1. Do you now have symptoms

   rarely?

   some of the time?

   most of the time?

   all the time?

2. How much does your illness now affect your everyday life?

   I can do most of what I could do before

   I can do about three-quarters of what I could do before

   I can do about half of what I could do before

   I can do about a quarter of what I could do before

   I can do hardly anything compared with before

3. If you have improved in the last five or six months, what do you feel are the most important reasons for this?

.....

.....

.....

.....

4. Excluding Dr. Ho-Yen, how would you rate the medical care and support you received for this illness from your doctor(s)? (Please circle the number which most closely corresponds with your assessment).

1	2	3	4	5
Very poor	Poor	Adequate	Good	Very good

APPENDIX II

Results from Study 2

Results from the 'CFS-plus' group

Demographic information and details of illness-related variables

CFS(ME) group

SCI group

Relationships between variables

CFS(ME) group

SCI group

Results of the 'CFS-plus' group

These CFS patients were generally similar to the main group in terms of age, gender, education and other demographic variables. However, as far as their illness was concerned, there were a number of differences. For instance, all except one of the patients in this group reported having a gradual onset and a greater proportion felt that their condition was deteriorating. They were also more severely impaired in terms of physical functioning and they had very high scores for fatigue and cognitive difficulty.

Meanwhile, the findings of the social support measures revealed that they had less contact and received fewer resources from relatives than the main group. They also reported a greater number of interpersonal stressors both from friends and relatives. However, they had similar scores for contact with and support from friends.

The 'CFS-plus' group had very high anxiety and depression scores. Indeed, all the patients scored above the cut-off point suggesting possible clinical anxiety or depression, and all but one scored above the cut-off point for probable clinical anxiety or depression.

At the time of the assessment, three of the 'CFS-plus' group were on anti-depressants, and one had been prescribed tranquillisers.

Table 1. Demographic information for the 'CFS-plus' group

Variable		
Age		
Median (SD)	35 (10.3)	
Range	19-50	
Gender	No.	%
Men	1	14
Women	6	86
Marital status	No.	%
Single	5	71
Married	2	29
Marital status changed during illness	2	29
Education completed	No.	%
Secondary school	4	57
College/University	1	14
Professional training	1	14
Did not complete	1	14
Mean years (SD) spent in education from the age of 16	2.1 (2.2)	
Range	0-5	
Employment status	No.	%
Housewife	1	14
Unemployed	1	14
On sick leave	4	57
Parttime work	0	
Fulltime work	0	
Retired	1	14

Table 1 cont.

	No.	%
Classification of work		
Professional	1	14
Managerial	1	14
Manual skilled	2	29
Manual unskilled	2	29
Have never worked	1	14
Changed job or reduced hours due to disability	6	86
Housing		
Owner-occupier	1	14
Council tenant/housing Association	6	86
Living alone	4	57
Income		
Dependent on social security benefits	3	43

Table 2. Details of illness-related variables for 'CFS-plus' group

Variable	No.	%
Onset		
Sudden	1	14
Gradual	6	86
Known infectious trigger		
Yes	3	43
No	4	57
Tests used in diagnosis of illness		
IgM Coxsackie	0	
VP1	1	14
Muscle biopsy	0	
Brain scan	1	14
Other laboratory tests	2	29
More than two of the above	2	29
Clinical history only	1	14
Course of condition		
Fluctuating	1	14
Deteriorating	5	71
Improving	0	
Unknown	1	14
Current use of drugs		
Yes	6	86
No	1	14
Presence of other medical conditions		
Yes	7	100
No	0	

Table 3. Means (SD) relating to independent variables for the 'CFS-plus' group

Variable	Mean	SD	N
Symptoms			
Fatigue	5.10	1.21	7
Emotional distress	3.93	1.91	7
Cognitive difficulty	4.91	.95	7
Somatic symptoms	3.39	1.45	7
Interpersonal resources and stressors			
Friends resources	13.43	5.16	7
Spouse resources	15.67	7.37	3
Relatives resources	9.33	4.27	6
Friends stressors	10.14	4.98	7
Spouse stressors	9.33	3.06	3
Relatives stressors	13.50	3.67	6
Friends contact	7.14	1.68	7
Relatives contact	4.00	1.53	7
Friends loss	2.00	.58	7
MUIS scores			
Uncertainty	78.86	11.89	7
Unpredictability	18.57	3.69	7
Illness-related variables			
Duration	14.00	11.14	6
Years diagnosed	4.00	2.38	7
Number of symptoms	5.14	1.35	7
MOS Physical functioning	8.50	2.07	6
Help required	2.14	2.73	7
HAD			
Anxiety	14.57	4.65	7
Depression	13.43	3.51	7
Depression corrected	10.71	3.64	7

Table 3 cont.

Variable	Mean	SD	N
Functional impairment			
Total	29.29	2.36	7

Table 1. Demographic information

Variable	CFS(ME) group		SCI group	
Age				
Median (SD)	36 (13.4)		35 (7.3)	
Range	16-70		26-50	
Gender	No.	%	No.	%
Men	16	28	20	80
Women	42	72	5	20
Marital status	No.	%	No.	%
Single	24	41	10	40
Married	27	46	12	48
Cohabiting	3	5	1	4
Separated/divorced	4	7	2	8
Marital status changed during illness	1	2	3	12

Table 1 cont.

Variable	CFS(ME) group		SCI group	
Education completed	No.	%	No.	%
Secondary school	54	93	23	92
College/University	18	31	6	24
Professional training	19	33	3	12
Did not complete	1	3	2	8
Mean years (SD) spent in education from the age of 16	3.05 (3)		2.68 (3.2)	
Range	0-12		0-13	
Employment status	No.	%	No.	%
Student	8	14	1	4
Housewife	10	17	3	12
Unemployed	4	7	3	12
On sick leave	11	19	4	16
Parttime work	6	10	1	4
Fulltime work	9	16	6	24

Table 1 cont.

Variable	CFS(ME) group		SCI group	
	No.	%	No.	%
Employment status				
Retired	10	17	7	28
Classification of work				
Professional	8	14	4	16
Managerial	2	3	2	8
Computer/technical	5	9	4	16
Administration, skilled	4	7	0	
Manual skilled	12	21	5	20
Manual unskilled	14	24	8	32
H.M. Forces/police	3	5	1	4
Other job	3	5	0	
Have never worked	7	12	1	4
Changed job or reduced hours due to disability	42	72	17	68

Table 1 cont.

Variable	CFS(ME) group		SCI group	
	No.	%	No.	%
Housing				
Owner-occupier	34	59	20	80
Private tenant	5	9		
Council tenant/housing Association	5	9	3	12
Living with parents	12	21	2	8
Unknown	2	3	0	
Living alone	6	10	6	24
Income				
Dependent on social security benefits	17	29	13	52

Table 2. Details about the illness of the CFS(ME) group

Variable	No.	%
Tests used to diagnose illness		
IgM Coxsackie	0	
VP1	2	3
Muscle biopsy	0	
Brain scan	2	3
Other laboratory tests	12	21
More than two of the above	12	21
None/unknown	3	5
Clinical history only	27	47
Current use of drugs		
Yes	38	66
No	20	34
Level of activity compared with past (n=57)		
Can do little	10	18
Can do a quarter	24	41
Can do a half	13	23
Can do three-quarters	9	16
Can do most	1	2
Frequency of symptoms (n=57)		
All the time	21	37
Most of the time	31	54
Some of the time	5	9

Table 2b. Details about the disability of the SCI group

	No.	%
Lesion (n=21)		
Complete	13	61.9%
Incomplete	8	38.1%
Cause		
Road traffic accident	12	52%
Other accident	8	35%
Illness	3	13%
Use of mobility aids		
Wheelchair	18	72%
Crutches	3	12%
Both	3	12%
None	1	4%
Current use of drugs		
Yes	15	60%
No	10	40%
Level of activity compared with past		
Can do little	8	32
Can do a quarter	4	16
Can do a half	3	12
Can do three-quarters	5	20
Can do most	1	4
Can not assess	1	4
Frequency of symptoms (n=24)		
All the time	1	4
Most of the time	6	25
Some of the time	9	38
Rarely	7	29
Never	5	4

Relationships between variables

The relationships between the PFRS subscale scores of the CFS(ME) group

The relationships between the four PFRS subscales were assessed using Pearson's r. As shown in Table 3, all the correlation co-efficients were significant, the strongest relationship being between fatigue and cognitive difficulty. Given the strong relationship and the theoretical overlap between the emotional distress subscale and the HAD, it was decided not to include the former in the main analysis.

Table 3. Intercorrelations (Pearson's r) among PFRS subscales for the CFS(ME) group

	Fatigue	Emotional distress	Cognitive difficulty	Somatic symptoms
Fatigue	1.00**	.43**	.74**	.60**
Emotional distress		1.00**	.46**	.55**
Cognitive difficulty			1.00**	.49**
Somatic symptoms				1.00**

** $p \leq .001$

Table 4. Correlations (Pearson's r) between main variables and HAD subscales for the CFS(ME) group

	Anxiety	Depression	Depression corrected
Age	-.23	.11	.11
Education	-.05	-.14	-.13
Physical functioning	.12	-.01	.03
Help required	-.09	.05	.03
Functional impairment	-.10	.26	.20
Duration	-.10	.11	.08
Years diagnosed	-.02	.02	.04
No. of symptoms	.15	.09	.10
Level of activity	.09	-.15	-.11
Frequency of symptoms	.08	.04	.06
Uncertainty	.54**	.35*	.41*
Unpredictability	.15	.19	.22
Friends resources	-.18	-.27	-.33
Spouse resources	-.11	-.14	-.16
Relatives resources	-.12	-.29	-.31
Friends stress	.15	.19	.19
Spouse stress	.37	.36	.42*
Relatives stress	.33	.29	.28
Contact with friends	-.33	-.34*	-.37*
Contact with relatives	.02	.02	.04
Loss of friends	.22	.24	.25
Anxiety	1.00**	.40*	.46**
Depression	.40*	1.00**	.98**
Depression corrected	.46**	.98**	1.00**

* $p<.01$ ** $p\leq.001$

Table 5. Correlations (Spearman's Rho) between main variables and PFRS subscales for the CFS(ME) group

	Fatigue	Cognitive difficulty	Somatic symptoms
Age	-.02	.04	-.00
Education	-.16	-.05	-.14
Physical functioning	-.44**	-.22	-.24
Help required	.02	.14	.12
Duration	.29	.28	.21
Years diagnosed	.17	.13	.09
Number of symptoms	.19	.27	.07
Level of activity	-.44**	-.35*	-.13
Frequency of symptoms	-.36*	-.26	-.19

* p<.01 ** p<.001

Table 6. Correlations (Spearman's Rho) between main variables and interpersonal resources for the CFS(ME) group

	Friends resources	Relatives resources	Spouse resources	Contact with friends	Contact with relatives
Age	-.17	-.03	-.11	.08	.15
Education	.04	-.02	.11	.08	-.08
Help required	-.09	.03	.01	-.06	-.19
No. symptoms	-.16	.05	.27	-.25	.07
Level of activity	-.02	-.16	-.17	.12	-.01
Frequency of symptoms	.01	-.13	-.03	-.06	.02
Friends resources	1.00**	.46**	.19	.61**	.05
Spouse resources	.19	.17	1.00**	.01	-.10
Relatives resources	.46**	1.00**	.17	.20	.17

Table 6 cont.

	Friends resources	Relatives resources	Spouse resources	Contact with friends	Contact with relatives
Friends stress	-.26	-.33	-.12	-.10	-.10
Spouse stress	-.31	-.14	-.44*	-.14	.17
Relatives stress	-.07	-.35*	-.25	-.10	-.02
Contact friends	.60**	.20	.01	1.00**	.33
Contact relatives	.06	.17	-.10	.33	1.00**
Loss of friends	- 17	-.15	-.04	-.37*	-.13

* $P \leq .01$ ** $P \leq .001$

Table 7. Correlations (Spearman's Rho) between main variables and interpersonal stressors for the CFS(ME) group

	Friends stress	Relative stress	Spouse stress	Loss of friends
Age	-.08	-.37*	.16	-.27
Education	.12	.09	.03	.20
Help required	.14	-.03	.07	.14
No. symptoms	.03	-.09	-.23	.13
Level of activity	.02	-.01	.18	-.38*
Frequency of symptoms	-.03	-.19	.24	-.27
Fatigue	-.12	.04	-.18	.17
Cognitive difficulty	.10	.06	-.19	.15
Somatic symptoms	.07	.09	.13	.05
Friends resources	-.26	-.08	-.32	-.19
Spouse resources	-.12	-.25	-.44*	-.04
Relatives resources	-.33	-.36*	-.14	-.16
Friends stress	1.00*	.48**	.43*	.36*
Spouse stress	.44*	.34	1.00**	.09
Relatives stress	.47*	1.00**	.34	.37*
Contact friends	-.12	-.10	-.14	-.37*
Contact relatives	-.10	-.02	.17	-.13
Loss of friends	.36*	.37*	.09	1.00**

* $p \leq .01$ ** $p \leq .001$

Table 8. Correlations (Spearman's Rho) between variables and functional impairment subscales for the CFS(ME) group

	Work	Home	Social leisure	Private leisure	Functional impairment total
Age	.02	-.12	-.27	-.02	-.12
Education	-.09	-.18	.03	-.15	-.18
Work	1.00**	.60**	.43**	.26	.78**
Home	.60**	1.00**	.52**	.50**	.88**
Social	.43**	.52**	1.00**	.32	.70**
Private	.26	.50**	.32	1.00**	.58**
Physical functioning	-.31	-.35*	-.23	-.21	-.38*
Help required	.37*	.40*	.29	.17	.41*
Duration	.23	.22	.10	.15	.23
Years diagnosed	.30	.43**	.23	.18	.40*
No. symptoms	.02	.26	.16	.06	.23

Table 8 cont.

	Work	Home	Social leisure	Private leisure	Functional impairment total
Level of activity	-.51**	-.67**	-.38*	-.30	-.61*
Frequency of symptoms	-.18	-.34	-.13	-.30	-.31
Fatigue	.43**	.32	.24	.23	.33
Cognitive difficulty	.32	.35*	.24	.35*	.37*
Somatic symptoms	.12	.04	.03	.17	-.01
Uncertainty	.05	.04	.29	.26	.12
Unpredictability	-.10	.11	.02	-.03	-.02
Friends resources	.02	.00	-.09	.07	-.08
Spouse resources	.06	-.20	-.04	-.17	-.07
Relatives resources	-.00	-.07	-.20	.03	-.08

Table 8 cont.

	Work	Home	Social leisure	Private leisure	Functional impairment total
Friend stress	-.24	.02	.04	-.07	-.12
Spouse stress	-.19	.01	-.34	.03	-.19
Relatives stress	-.06	.06	.21	.14	.01
Contact friends	.02	-.07	-.15	-.10	-.11
Contact relatives	.07	-.18	-.15	-.25	-.19
Loss of friends	.18	.37*	.50**	.20	.39*
Anxiety	.01	-.02	.02	.05	-.04
Depression	.14	.24	.30	.25	.25
Depression corrected	.07	.19	.25	.24	.19

* $p \leq .01$ ** $p \leq .001$

Table 9. Correlations (Spearman's Rho) between demographic and illness-related variables for the CFS(ME) group

	Physical functioning	Help required	Duration	Level of activity	Frequency of symptoms
Age	.01	.08	.30	.23	.15
Education	.14	-.12	-.07	.22	.03
Physical functioning	1.00**	-.28	-.25	.44**	.49**
Duration	-.25	.29	1.00**	-.17	-.35*
Years diagnosed	-.16	.31	.55**	-.25	-.15
Number of symptoms	-.10	.07	.09	-.34	-.12
Level of activity	.44**	-.30	-.17	1.00**	.48**
Frequency of symptoms	.49**	-.25	-.35*	.48**	1.00**
Help required	-.28	1.00**	.29	-.30	-.25
Uncertainty	.26	-.06	-.04	.05	.05
Unpredictability	-.12	.26	.07	.10	.03

* $p \leq .01$ ** $p \leq .001$

Appendix IIB

Relationships between variables: SCI group

Table 3b. Intercorrelations (Pearson's r) among PFRS subscales for SCI group

	Fatigue	Emotional distress	Cognitive difficulties	Somatic symptoms
Fatigue	1.00**	.66**	.60*	.62**
Emotional distress		1.00**	.71**	.46
Cognitive difficulty			1.00**	.45
Somatic symptoms				1.00**

** p<.001

Table 4b. Correlations (Pearson's r) between the main variables, HAD subscales and functional impairment for the SCI group

	Anxiety	Depression	Depression corrected	Functional impairment
Age	-.11	-.10	-.14	.03
Education	-.44	-.33	-.33	-.28
Physical functioning	.04	-.25	-.17	-.77**
Help required	.00	.24	.19	.64**
Functional impairment	.30	.49	.43	1.00**
Duration	-.04	-.39	-.46	.15
Lesion	.09	-.11	-.09	-.58*
Number of symptoms	.16	.31	.25	.50
Level of activity	-.01	-.62*	-.48	-.70**
Frequency of symptoms	-.07	-.29	-.22	-.67**
Fatigue	.36	.58*	.43	.43
Cognitive difficulty	.55*	.51*	.53*	.35
Somatic symptoms	.40	.31	.20	.61*
Uncertainty	.17	.25	.29	.22
Unpredictability	.09	.13	.14	.06
Friends resources	-.31	-.35	-.42	-.29
Spouse resources	-.62	-.25	-.27	-.08
Relatives resources	-.25	-.15	-.14	-.02
Friends stress	-.12	-.40	-.36	-.32
Spouse stress	.24	.01	-.07	-.18
Relatives stress	-.05	-.42	-.40	-.19
Contact with friends	-.34	-.28	-.22	-.18
Contact relatives	-.17	.06	-.10	-.01
Loss of friends	-.03	-.22	-.17	.52*

Table 4b cont.

	Anxiety	Depression	Depression corrected	Functional impairment
Anxiety	1.00**	.46	.43	.30
Depression	.46	1.00**	.96**	.49
Depression corrected	.43	.96**	1.00**	.43

* $p \leq .01$ ** $p \leq .001$

Table 5b. Correlations (Spearman's Rho) between demographics, disease-related variables, uncertainty and PFRS subscales for the SCI group

	Fatigue	Cognitive difficulty	Somatic symptoms
Age	-.01	-.17	.29
Education	-.03	-.43	-.15
Physical functioning	.34	-.31	-.58*
Help required	.20	.41	.53*
Duration	.09	-.16	.45
Lesion	-.29	-.24	-.26
No. symptoms	.62**	.41	.74**
Level of activity	-.22	-.18	-.47
Frequency of symptoms	-.40	-.11	-.65**
Uncertainty	.75**	.51	.64*
Unpredictability	.14	.04	.04

* $p \leq .01$. ** $p \leq .001$

Table 6b. Correlations (Spearman's Rho) between main variables and interpersonal resources for the SCI group

	Friends resources	Relatives resources	Spouse resources	Contact with friends	Contact with relatives
Age	-.07	-.14	-.25	.17	-.40
Education	.24	.04	-.18	.05	.05
Help required	.14	.32	.19	.10	.33
No. of symptoms	.24	.37	.17	.29	.17
Level of activity	.06	-.11	-.23	.13	-.28
Frequency of symptoms	.16	-.11	-.31	.27	.04
Friends resources	1.00**	.67**	.34	.42	.56*
Spouse resources	.34	.59	1.00**	.15	.34
Relatives res.	.67**	1.00**	.59	.44	.55*
Relatives stress	.26	.01	-.15	.29	-.22
Friends stress	-.02	-.05	-.14	.19	-.05
Spouse stress	-.07	-.15	-.42	.01	-.19
Contact friends	.42	.44	.15	1.00**	.22
Contact relatives	.56*	.55*	.34	.22	1.00**
Loss of friends	-.25	.04	.32	-.16	.02

* $p \leq .01$ ** $p \leq .001$

Table 7b. Correlations (Spearman's Rho) between main variables and interpersonal stressors for the SCI group

	Friends stress	Relatives stress	Spouse stress	Loss of friends
Age	.32	.20	.65*	-.01
Education	.32	.26	.42	-.24
Help required	-.27	-.19	-.26	.48
No. of symptoms	.06	.09	-.19	.12
Level of activity	.37	.38	.31	-.27
Frequency of symptoms	.09	-.09	.23	-.38
Uncertainty	.27	.22	-.18	.02
Unpredictability	-.07	.14	.10	.06
Friends resources	-.02	.31	-.20	-.28
Spouse resources	-.14	-.10	-.56	.32
Relatives resources	-.05	.02	-.22	.04
Relatives stress	.27	1.00**	-.04	.11
Friends stress	1.00**	.28	.02	.02
Spouse stress	.03	-.04	1.00**	-.33
Contact friends	.19	.29	.01	-.16
Contact relatives	-.05	-.22	-.19	.02
Loss of friends	.03	.11	-.33	1.00**

$p \leq .01$. ** $p \leq .001$

Table 8b. Correlations (Spearman's Rho) between main variables and functional impairment subscales for the SCI group

	Work	Home	Social leisure	Private leisure	Functional impairment total
Age	.14	.24	-.15	.02	.03
Education	-.35	-.16	-.30	-.19	-.27
Work	1.00**	.47	.43	.67**	.77**
Home	.47	1.00**	.45	.47	.73**
Social leisure	.43	.45	1.00**	.54*	.77**
Private leisure	.67**	.47	.54*	1.00**	.84**
Physical functioning	-.57*	-.73**	-.52*	-.47	-.77**
Help required	.49	.63**	.40	.37	.60*
Duration	.08	.36	.05	-.03	.18
Lesion	-.40	-.52*	-.54*	-.32	-.59*
No. of symptoms	.61*	.42	.21	.42	.50

Table 8b cont.

	Work	Home	Social leisure	Private leisure	Functional impairment total
Level of activity	-.64**	-.46	-.47	-.70**	-.71**
Frequency of symptoms	-.61*	-.50	-.32	-.66**	-.63**
Fatigue	.33	.25	.11	.45	.37
Cognitive difficulty	.35	.33	.08	.35	.34
Somatic symptoms	.60*	.54*	.23	.58*	.61*
Uncertainty	.43	.12	-.08	.39	.22
Unpredictability	.48	-.10	-.10	.17	.09
Friends resources	-.04	-.11	-.53*	-.25	-.26
Spouse resources	-.02	-.09	-.29	.07	.06
Relatives resources	.16	-.00	-.17	.06	.04

Table 8b cont.

	Work	Home	Social leisure	Private leisure	Functional impairment total
Friends stress	-.31	-.21	-.27	-.35	-.35
Spouse stress	-.01	.12	-.27	-.20	-.16
Relatives stress	-.12	-.13	-.15	-.34	-.20
Contact friends	-.07	-.25	-.34	-.21	-.22
Contact relatives	.14	-.13	-.15	.07	.04
Loss of friends	.40	.34	.52*	.23	.52*
Anxiety	.21	.26	.21	.27	.25
Depression	.48	.38	.34	.74**	.54*
Dep. corrected	.36	.26	.40	.58*	.41

* $p \leq .01$ ** $p \leq .001$

Table 9b. Correlations (Spearman's Rho) between demographics, disability-related variables and uncertainty for the SCI group

	Physical functioning	Help required	Lesion	Number of symptoms	Level of activity	Frequency of symptoms
Age	-.06	.06	.13	.26	-.07	-.33
Education	.12	-.35	.17	-.19	.37	-.03
Help required	-.76**	1.00**	-.62**	.38	-.70**	-.31
Physical functioning	1.00**	-.76**	.58*	-.51	.64**	.38
Duration	.14	.14	-.14	.34	.02	-.49
Level of lesion	-.58**	.62**	1.00**	-.18	.39	.24
No. symptoms	-.51	.38	-.18	1.00**	-.41	-.50
Level of activity	.64**	-.70**	.39	-.41	1.00**	.49
Frequency of symptoms	.38	-.31	.24	-.50	.49	1.00**
Uncertainty	-.12	.17	-.14	.64*	-.15	-.58*
Unpredictability	.21	-.28	-.05	.21	.01	-.34

* $p \leq .01$ ** $p \leq .001$

APPENDIX III

Results from Study 3

Demographic information

Details about illness-related variables

Results of Ancovas using transformed and original scores

Table 1. Demographic information relating to the samples

Variable	Treatment group	Control group
Age		
Mean (SD)	39.6 (13.40)	37.7 (14.35)
Range	15-64	14-66
Gender	No. %	No. %
Men	6 27	9 41
Women	16 73	13 59
Marital status	No. %	No. %
Single	5 23	7 32
Married	13 59	11 50
Cohabiting	2 9	2 9
Separated/divorced	2 9	2 9
Marital status changed during illness	1 5	1 5

Table 1 cont.

Variable	Treatment group		Control group	
Education completed	No.	%	No.	%
Secondary school	11	50	13	59
College/University	2	9	2	9
Professional training	5	23	2	9
Did not complete	4	18	5	23
Mean (SD) years spent in education from the age of 16	3.0 (3.3)		2.0 (3.4)	
Range	0-10		0-10	
Employment status	No.	%	No.	%
Student	2	9	3	14
Housewife	4	18	3	14
Unemployed	1	5	3	14
On sick leave	6	27	9	41
Parttime work	6	27	2	9
Fulltime work	2	9	0	
Retired	1	5	2	9

Table 1 cont.

Variable	Treatment group		Control group	
Classification of work	No.	%	No.	%
Professional	2	9	2	9
Managerial	2	9	4	18
Manual skilled	7	32	2	9
Manual unskilled	2	9	6	27
Other job	5	23	3	14
Have never worked	4	18	5	23
Changed job or reduced hours due to disability	18	86	18	95
	n=21		n=19	
Housing	No.	%	No.	%
Owner-occupier	13	59	12	55
Private tenant	1	5	2	9
Council tenant/housing Association	4	18	2	9
Living with parents	4	18	4	18

Table 1 cont.

Variable	Treatment group		Control group	
Housing	No.	%	No.	%
Unknown			2	9
Living alone	0		5	23
Income				
Dependent on	No.	%	No.	%
social security benefits	3	14	5	24
			n=21	

Table 2. Details about the illness and illness-related experiences

	Treatment group		Control group	
Onset	No.	%	No.	%
Sudden	8	40	12	63
Gradual	12	60	7	32
Known infectious trigger	No.	%	No.	%
Yes	8	40	14	74
No	12	60	5	26
Tests used to diagnose illness?	15	68	17	85
	n=19		n=20	
Followed advice in Dr. Ho-Yen's book?	No.	%	No.	%
Not yet	1	11	2	18
Some of the time	3	33	5	45
Most of the time	3	33	4	36
All of the time	2	22	0	
	n=9		n=11	

Table 2 cont.

	Treatment group				Control group			
	Time 1		Time 2		Time 1		Time 2	
	No.	%	No.	%	No.	%	No.	%
Level of activity compared with past								
Can do little	6	27	5	23	9	41	8	36
Can do a quarter	10	45	5	23	10	45	5	23
Can do a half	5	23	6	27	2	9	7	32
Can do three-quarters	1	5	6	27	1	5	2	9
Can do most	0		0		0		0	
Frequency of symptoms								
All the time	9	41	8	36	10	45	11	50
Most of the time	10	45	6	27	11	50	7	32
Some of the time	3	14	7	32	1	5	4	18
Rarely	0		1	5	0		0	